

## Systematic Review

# A Comprehensive Study of Pericardial Hydatid Cyst: Systematic Review and Meta-Data Presentation

Hiwa O. Abdullah<sup>1,2</sup>, Berun A. Abdalla<sup>1,2</sup>, Dana H. Mohammed-Saeed<sup>1,3,4</sup>, Soran H. Tahir<sup>1,3</sup>, Fattah H. Fattah<sup>1,3</sup>, Sabah Jalal Hasan<sup>1</sup>, Hussein M. Hamasalih<sup>1</sup>, Bnar J. Hama Amin<sup>1</sup>, Abdulwahid M. Salih<sup>1,3</sup>, Savo Sh. Noori<sup>5</sup>, Fahmi H. Kakamad<sup>1,2,3\*</sup>, Shvan H. Mohammed<sup>2</sup>

1. Smart Health Tower, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq
2. Kscien Organization, Hamdi St., Azadi Mall, Sulaimani, Kurdistan, Iraq
3. College of Medicine, University of Sulaimani, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq
4. Sulaimani Center for Heart Disease, François Mitterrand Street, Sulaimani, Kurdistan, Iraq
5. Ministry of Health, Sulaimani, Kurdistan, Iraq

\* Corresponding author: [fahmi.hussein@univsul.edu.iq](mailto: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

Pericardial hydatid cysts constitute 7% of all cases of cardiac hydatidosis, yet their occurrence is often associated with several life-threatening complications. This study presents a systematic review of reported cases of pericardial hydatid cysts.

### Methods

A systematic review of published studies on pericardial hydatid cysts was conducted. The included studies meeting the following criteria: (1) Confirmation of pericardium infection through diagnostic modalities, surgical findings, or histopathology; (2) Presentation of case details within the study; (3) Presence of cyst(s) originally located or adhered to the pericardium without rupture from adjacent cardiac structures or organs.

### Results

In total, 106 studies met the inclusion criteria. The majority of cases (29.72%) were reported in Turkey, followed by India (18.24%). No gender predilection was observed, and patients' ages ranged from 5 to 80 years. The most common symptoms reported were chest pain (43%) and dyspnea (36%). Hydatid cysts were exclusively located in the pericardium in 56% of cases, while 44% involved multiple locations. Surgery was the preferred treatment choice (87.8%), with cystectomy (72.3%) being the primary technique for cyst removal. Only three cases (2%) experienced recurrences, with a significant correlation between recurrence and a history of hydatidosis. The mortality rate was 2.7%.

### Conclusion

Pericardial hydatid disease is more prevalent in subtropical regions. The definitive treatment for pericardial hydatid cysts is primarily surgical, typically performed through a median sternotomy. A history of hydatidosis increases the likelihood of recurrence.

## 1. Introduction

Hydatidosis is a well-known zoonotic disease caused by the larval form of the tapeworm *Echinococcus granulosus*. Humans

typically serve as intermediate hosts for this parasite, becoming infected through direct contact with the primary hosts such as sheep, goats, cattle, dogs, and other canines, or by ingesting

contaminated food and water containing the parasite's eggs [1]. This disease poses a global threat as a parasitic infection, primarily afflicting areas dedicated to farming and domestic animal husbandry. It maintains endemic status across various geographical regions, including the Middle East, the Mediterranean, the Americas, South Africa, and Australia [2, 3]. Hydatid disease most commonly targets the liver, followed by the lungs. While cardiac hydatid cysts (HC) are a rare occurrence, they can still manifest with fatal consequences [4, 5]. The systemic circulation of HC larvae is believed to underlie the development of cardiac hydatid disease [6]. Diagnosis often presents challenges due to the multitude of clinical presentations and nonspecific symptoms [7]. The clinical manifestation of hydatidosis depends on the size and location of the cyst. At times, the disease remains silent or asymptomatic for several years, only becoming apparent when the cyst reaches a size that triggers compression and associated symptoms [5, 8, 9]. Nearly 90% of cardiac HC cases are asymptomatic, although some constitutional symptoms like dyspnea, atypical chest pain, and cough may be associated with them [2, 10]. Pericardial HC accounts for 7% of all cases of cardiac hydatid diseases, yet its occurrence is linked to several life-threatening complications, including arrhythmia, pericardial effusion, cardiac compression, and anaphylactic shock resulting from cyst rupture, among others [2, 11]. While the literature contains various reviews focusing on specific aspects of cardiac hydatid disease, there is a dearth of systematic reviews with comprehensive meta-data presentation [12, 13, 14].

This study aims to fill this gap by providing a systematic review and meta-data presentation of all reported studies concerning pericardial hydatid disease [1, 2, 4-11, 15-110]

## 2. Methods

### 2.1. Study design

The present systematic review adhered to the preferred reporting items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.

### 2.2. Data sources and search strategy

A systematic review of all the published studies of pericardial HC was conducted using Google scholar, PubMed/MEDLINE, Cochrane Library, Science Direct, CINAHL, Web of Science, and EMBASE databases. The keywords that were used in the search included: (pericardial OR pericardium OR epicardium OR epicardial OR heart OR cardiac) AND (hydatid OR echinococcoses OR echinococcosis OR echinococcal OR echinococcus).

### 2.3. Eligibility criteria

Studies of non-English-language and those unrelated to humans were directly excluded before or during the initial screening, respectively. All studies of cardiac HCs that had the following properties were included: 1) The pericardium infection had been confirmed by diagnostic modalities, surgical findings, or histopathology. 2) The case presentation was provided in the study. 3) The cyst(s) was originally located or adhered to the

pericardium and did not rupture into it from the other adjacent cardiac structures or organs. Studies had been published in predatory journals (inappropriately peer-reviewed) [111], and all those that were not compatible with the inclusion criteria were excluded.

### 2.4. Study selection and data extraction

The titles and abstracts of identified studies were initially screened before an intensive full-text screening for eligibility. Multiple data were recorded from the included studies such as study design, country of study, age, gender, resident, symptoms, medical history of HC, serology test, diagnosis, cyst's location, management, follow-up, and recurrence.

### 2.5. Statistical analyses

The data were initially used in qualitative synthesis and then quantitatively re-analyzed by the Chi-square test and Fisher T-test using Statistical Package for Social Sciences (SPSS) 25.0 software. The statistical level of significance was determined at 0.05.

## 3. Results

In total, 750 studies were obtained from the resources, 146 of which were directly removed before any screening due to duplication and non-English language. On the initial screening, the titles and abstracts of 321 studies did not match the inclusion criteria, and they were excluded. Overall, 283 studies underwent full-text screening and 119 of them were assessed for eligibility. Finally, 106 studies (148 cases) were compatible with the inclusion criteria (Figure 1). Out of the included studies, 93 (87.7%) were case reports, and the remaining (13, 12.3%) were case series (Table 1). Most of the cases (29.72%) were reported in Turkey, followed by India (18.24%), Spain (8.87%), and China (6.76%) (Table 2). There was no gender predilection and both genders were affected almost equally. The age of patients was distributed between 5 and 80 years old, with a mean of 38.36 years. Most of the cases (85.8%) were affected by the disease during the first to sixth decades of their lives (Table 3). The history of hydatid disease was positive in 17.6% of the affected cases (Table 4). The most commonly presented symptoms were chest pain (43%), dyspnea (36%), followed by cough (9%), and palpitation (9%) (Table 3). There were 24 cases in rural areas and three cases (2%) in urban areas. The residency of the remaining 117 cases was not reported.

Serology had been done for 88 cases (59.9%) and it was positive in 42%. Echocardiography was the most commonly used diagnostic imaging tool (62.8%), followed by computed tomography (CT) scan (52%), and chest X-ray (43.9%). HCs were only found in the pericardium in 56% of cases and multilocation in 44% (Table 5). Surgery was the treatment of choice (87.8%). The type of surgical approach was not defined in 41% of the cases. Median sternotomy (25.7%) and thoracotomy (19%) were the main surgical approaches in the remaining cases, and cystectomy (72.3%) was the major technique of cyst removal.

Conservative management was performed in 13 cases (8.8%). Five cases (3.4%) underwent no management, and three of them died before any intervention. Among those cases that had a follow-up, its period was mostly between 1 month and 1 year (19%). Albendazole was administered in 40.5% of the cases (Table 5). The total recurrence was 3 cases (2%), and two of them (66.66%) had been managed with only conservative treatment. The third recurrent case had not been administered albendazole during the follow-up period after surgical intervention. There was a significant correlation between recurrence and the history of HC disease (P-value <0.05).

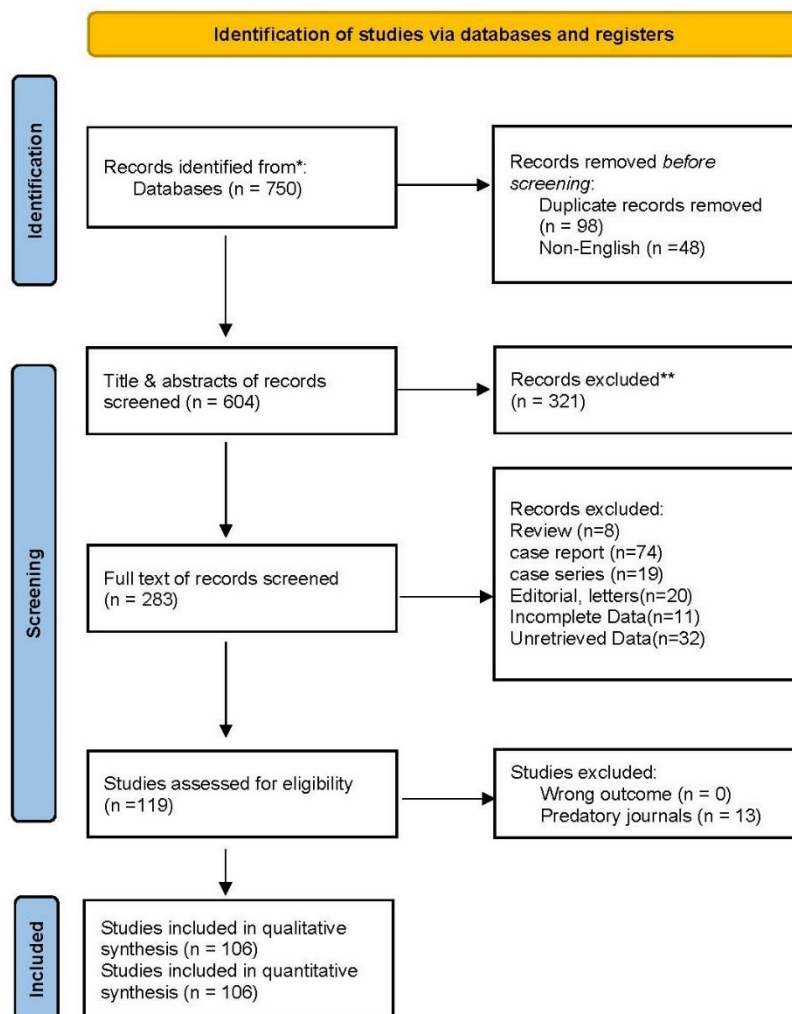
However, there was no correlation between recurrence and location of the cysts (P-value > 0.05) (Table 4). The mortality rate pre/post-intervention was 2.7% (Table 5).

#### 4. Discussion

Since Hippocrates' time, hydatid disease has been known as a parasitic infection, and it has remained endemic in many geographical regions like the Middle East, Asia, Africa, Australia, America, and southern Europe [18,41,67]. This

disease most commonly occurs in countries that have large numbers of livestock farming areas. However, it has recently become a serious global health problem due to immigration and increasing travel [17]. Hydatidosis was thought to be only caused by the larval stage of *Echinococcus granulosus*, but it has lately been reported that a mixture of five species with ten distinct genotypes (G1-G10), including two bovid strains (G3/G5), two pig strains (G7 and G9), two sheep strains (G1 and G2), two Cervidae strains (G8 and G10), a horse strain (G4), and a camel strain (G6) can cause this disease. The main five species are *E. oligarthrus* (G5), *E. equinus* (G4), *E. granulosus sensu stricto* (G1-G3), *E. canadensis* (G6-G10), and *E. felidis*. Among these species, *E. granulosus sensu stricto*, *E. granulosus sensu lato*, and *E. canadensis* are more frequent in humans [17].

Cardiac HC is a very rare form of echinococcosis, comprising about 0.5% to 2% of all cases, and it was first described in 1836. The incidence of cardiac HC has been reported by a study to be 0.1% in 577 cases of hydatid disease [17,85]. For the migration of echinococcus larvae to the heart, several pathways have been proposed: coronary circulation, thoracic duct, the superior-inferior vena cavae, intestinal lymphatics, pulmonary veins, and hemorrhoidal veins. The common sites of the heart to be affected



**Figure 1.** Study selection PRISMA flow chart.

**Table 1.** The characteristics of the included studies

| First Author              | Study design | No. of included case(s) | First Author              | Study design | No. of included case(s) | First Author               | Study design | No. of included cases | Reference |
|---------------------------|--------------|-------------------------|---------------------------|--------------|-------------------------|----------------------------|--------------|-----------------------|-----------|
| Goksel <sup>1</sup>       | *            | 1                       | Erol <sup>41</sup>        | *            | 1                       | Mughal <sup>77</sup>       | *            | 1                     | 77        |
| Charfeddine <sup>2</sup>  | *            | 1                       | Exadactylos <sup>42</sup> | *            | 1                       | Naoui <sup>78</sup>        | *            | 1                     | 78        |
| Bogdanovic <sup>4</sup>   | *            | 1                       | Feng <sup>43</sup>        | **           | 8                       | Narin <sup>79</sup>        | *            | 1                     | 79        |
| Ghareep <sup>5</sup>      | *            | 1                       | Fertin <sup>44</sup>      | *            | 1                       | Nasri <sup>80</sup>        | *            | 1                     | 80        |
| Shakil <sup>6</sup>       | *            | 1                       | Franquet <sup>45</sup>    | *            | 1                       | Nawaiseh <sup>81</sup>     | *            | 1                     | 81        |
| Bakirci <sup>7</sup>      | *            | 1                       | Fortunato <sup>46</sup>   | *            | 1                       | Nemes <sup>82</sup>        | *            | 1                     | 82        |
| Dwivedi <sup>8</sup>      | *            | 1                       | Geber <sup>47</sup>       | *            | 1                       | Noah <sup>83</sup>         | *            | 1                     | 83        |
| Gormus <sup>9</sup>       | *            | 1                       | Girit <sup>48</sup>       | *            | 1                       | Oliver <sup>84</sup>       | **           | 3                     | 84        |
| Kothari <sup>10</sup>     | *            | 1                       | Gibson <sup>49</sup>      | *            | 1                       | Oneri <sup>85</sup>        | *            | 1                     | 85        |
| Salati <sup>11</sup>      | **           | 11                      | Gomez <sup>50</sup>       | **           | 2                       | Onursal <sup>86</sup>      | **           | 5                     | 86        |
| Abou-Bekr <sup>15</sup>   | *            | 1                       | Guha <sup>51</sup>        | *            | 1                       | Oraha <sup>87</sup>        | **           | 1                     | 87        |
| Akkus <sup>16</sup>       | *            | 1                       | Gurlek <sup>52</sup>      | *            | 1                       | Ozates <sup>88</sup>       | *            | 1                     | 88        |
| Akpinar <sup>17</sup>     | *            | 1                       | Hammel <sup>53</sup>      | *            | 1                       | Panayotov <sup>89</sup>    | *            | 1                     | 89        |
| Alloubi <sup>18</sup>     | *            | 1                       | Herrero <sup>54</sup>     | *            | 1                       | Papo <sup>90</sup>         | **           | 7                     | 90        |
| Allouch <sup>19</sup>     | *            | 1                       | Heye <sup>55</sup>        | *            | 1                       | Parihar <sup>91</sup>      | *            | 1                     | 91        |
| Alonso <sup>20</sup>      | *            | 1                       | Ileri <sup>56</sup>       | *            | 1                       | Ramakrishnan <sup>92</sup> | *            | 1                     | 92        |
| Antonovic <sup>21</sup>   | *            | 1                       | Ilyas <sup>57</sup>       | *            | 1                       | Rey <sup>93</sup>          | **           | 1                     | 93        |
| Atilgan <sup>22</sup>     | *            | 1                       | Inzirillo <sup>58</sup>   | *            | 1                       | Sachdeva <sup>94</sup>     | *            | 1                     | 94        |
| Barbetseas <sup>23</sup>  | *            | 1                       | Jamil <sup>59</sup>       | *            | 1                       | Sakarya <sup>95</sup>      | *            | 1                     | 95        |
| Behzadnia <sup>24</sup>   | *            | 1                       | Karadede <sup>60</sup>    | *            | 1                       | Salih <sup>96</sup>        | *            | 3                     | 96        |
| Bennis <sup>25</sup>      | *            | 1                       | Karangelis <sup>61</sup>  | *            | 1                       | Sahin <sup>97</sup>        | *            | 1                     | 97        |
| Bernardo <sup>26</sup>    | *            | 1                       | Kardaras <sup>62</sup>    | **           | 1                       | Simonsen <sup>98</sup>     | *            | 1                     | 98        |
| Blanco <sup>27</sup>      | *            | 1                       | Kocogullan <sup>63</sup>  | *            | 1                       | Singhal <sup>99</sup>      | *            | 1                     | 99        |
| Boussaadani <sup>28</sup> | *            | 1                       | Kosar <sup>64</sup>       | *            | 1                       | Tellez <sup>100</sup>      | *            | 2                     | 100       |
| Bzikha <sup>29</sup>      | *            | 1                       | Kosecik <sup>65</sup>     | *            | 1                       | Thapaliya <sup>101</sup>   | *            | 1                     | 101       |
| Cakici <sup>30</sup>      | *            | 1                       | Kotoulas <sup>66</sup>    | *            | 1                       | Tükek <sup>102</sup>       | *            | 1                     | 102       |
| Cakir <sup>31</sup>       | *            | 1                       | Kumar <sup>67</sup>       | *            | 1                       | Tufekcioglu <sup>103</sup> | **           | 7                     | 103       |
| Ceyran <sup>32</sup>      | *            | 1                       | Kumar <sup>68</sup>       | *            | 1                       | Uygun <sup>104</sup>       | *            | 1                     | 104       |
| Cheng <sup>33</sup>       | *            | 1                       | Lahiri <sup>69</sup>      | *            | 1                       | Vural <sup>105</sup>       | *            | 1                     | 105       |
| Cimpoesu <sup>34</sup>    | *            | 1                       | Maffeis <sup>70</sup>     | *            | 1                       | Vurdem <sup>106</sup>      | *            | 1                     | 106       |
| Dasbaksi <sup>35</sup>    | *            | 1                       | Marci <sup>71</sup>       | *            | 1                       | Wadhawa <sup>107</sup>     | **           | 2                     | 107       |
| Delgado <sup>36</sup>     | *            | 1                       | Marouf <sup>72</sup>      | *            | 1                       | Ward <sup>108</sup>        | *            | 1                     | 108       |
| Deutsch <sup>37</sup>     | *            | 1                       | Menendez <sup>73</sup>    | **           | 1                       | Yaliniz <sup>109</sup>     | **           | 2                     | 109       |
| Dodek <sup>38</sup>       | *            | 1                       | Mestres <sup>74</sup>     | *            | 1                       | Yimamu <sup>110</sup>      | *            | 1                     | 110       |
| Dogra <sup>39</sup>       | *            | 2                       | Moorthy <sup>75</sup>     | *            | 1                       |                            |              |                       |           |
| Elbeyli <sup>40</sup>     | *            | 1                       | Mouhsine <sup>76</sup>    | *            | 1                       |                            |              |                       |           |

Study design: \* Case report, \*\* Case series

by HCs include the ventricles (70%), the pericardium (7%), the pulmonary artery (6%), the left atrium (6%), and the interventricular septum (4%). The left ventricle is the most common site of cardiac HCs owing to the rich blood supply and its thickness. An HC usually grows in the direction of the weaker side of the ventricular wall [2].

Pericardial HC is a rare type of cardiac hydatidosis. There are two possibilities for its involvement; the first is known as

hematogenous spread; the pericardium can be affected directly by the artery that supplies it. The second possibility is either due to cardiac HC perforation in the pericardial cavity or rupturing an HC that affected other visceral and neighboring organs in the pericardial cavity. The first possibility has been reported to be rarer than the second one [2,85]. Moreover, several studies have found that cardiac hydatid disease is frequently associated with hepatic or pulmonary HCs, with pericardial involvement occurring most commonly in multifocal cardiac hydatidosis. A

**Table 2.** The distribution of the reported cases among countries

| Country      | No. of cases | Percentage % |
|--------------|--------------|--------------|
| Turkey       | 44           | 29.72%       |
| India        | 27           | 18.24%       |
| Spain        | 13           | 8.78%        |
| China        | 10           | 6.76%        |
| Morocco      | 8            | 5.40%        |
| Serbia       | 8            | 5.40%        |
| Greece       | 5            | 3.38%        |
| US           | 4            | 2.70%        |
| Iraq         | 3            | 2%           |
| Brazil       | 2            | 1.35%        |
| Egypt        | 2            | 1.35%        |
| Germany      | 2            | 1.35%        |
| Italy        | 2            | 1.35%        |
| Pakistan     | 2            | 1.35%        |
| Tunisia      | 2            | 1.35%        |
| Algeria      | 1            | 0.68%        |
| Argentina    | 1            | 0.68%        |
| Australia    | 1            | 0.68%        |
| Belgium      | 1            | 0.68%        |
| Bulgaria     | 1            | 0.68%        |
| Denmark      | 1            | 0.68%        |
| France       | 1            | 0.68%        |
| Iran         | 1            | 0.68%        |
| Jordan       | 1            | 0.68%        |
| Libya        | 1            | 0.68%        |
| Qatar        | 1            | 0.68%        |
| Romania      | 1            | 0.68%        |
| Saudi Arabia | 1            | 0.68%        |
| Unknown      | 1            | 0.68%        |
| Total        | 148          | 100 %        |

solitary pericardial HC has been reported as a rare phenomenon. [10,41,51]. In this systematic review, an intensive review was conducted of all the studies of pericardial HCs. The results revealed that Turkey is the most susceptible country to the occurrence of pericardial hydatidosis in which most of the reported cases (29.72%) belonged to this country. Following Turkey, India, Spain, and China were among the first countries that reported a high incidence of this disease. Although five species with ten genotypes have been proposed to cause echinococcosis, among all of the studies that were reviewed in this systematic review, none of them identified a different species than *E. granulosus*. In contrast to the previous studies that mentioned the occurrence of cardiac HC mostly in combination with lung and liver HCs, only 44% of the cases in this study were concomitant with other locations' HCs. Meanwhile, the findings of this review disagree with the assumption that regarded solitary pericardial HC was a rare entity, in which 56% of reviewed cases were solitary infections. In addition, these findings support the hematogenous spread and

cardiac HC perforation in the pericardial cavity as the major causes of pericardial hydatidosis rather than the rupturing of HCs of other locations into the pericardium.

A cardiac HC grows gradually and may remain asymptomatic for years until it reaches a size that can compress and invade the surrounding and neighboring structures [66]. The symptoms are generally related to the location and size of the cyst, compression, age, and the involvement of the surrounding structures. The presentation of a pericardial HC can be varied, from asymptomatic to sudden death. The most frequent symptoms include dyspnea, palpitation, angina, and chest pain owing to pericardial stretch. Many patients can be asymptomatic with only intermittent fever or weakness. Perilous conditions can arise due to cyst growth, such as arrhythmia, circulatory collapse, heart failure, cardiac tamponade, and anaphylactic shock [4,15,20,24]. In this study, the major symptoms were chest pain (43%) and dyspnea (36%). Furthermore, 3.4% of the cases experienced an anaphylactic reaction.

Raising awareness for timely diagnosis of cardiac HCs is crucial, as a delay in diagnosis could lead to fatal consequences [4,15,26]. Several serological tests have been reported for the diagnosis of HC, but there are a lot of controversies about the accuracy of these tests. Some studies support them as having a high degree of specificity [17,20,27,41]. On the other hand, many other studies have found that serologic tests can produce a false-negative result and their sensitivity is not enough to confirm the pericardial HC [4,15,24,25,38,51]. It has been reported that the diagnosis of pericardial HC mainly depends on the imaging modalities. Transthoracic echocardiography (TTE) is the major imaging tool to diagnose pericardial masses because of its high sensitivity, simplicity, and noninvasiveness [4,25,27,28,53]. Other imaging techniques like computed tomography (CT) and magnetic resonance imaging (MRI) are useful in the diagnosis of myocardial and pericardial HCs [16]. The CT scan has good properties in the detection of cardiac localization and multiorgan involvement. Besides, MRI is much more reliable for localization and visualization [15,29,53]. The TTE can often be inaccurate in expressing the relationship of the cyst with the cardiac chambers and surrounding structures. Therefore, CT and MRI would be sufficient to determine this feature and also to differentiate solid tumors like fibromas and myxomas from any cystic mass and intracavitary thrombosis [4]. Angiocardiography is another diagnostic technique that has been mentioned to diagnose cardiac masses and provide proper information about the nature and location of a suspicious mass. Cardiovascular neoplasms, pericardial cysts, mediastinal tumors, and ventricular aneurysms can all be distinguished using this technique [21]. Despite all of these, TTE remains the first-choice diagnostic tool for cardiac hydatid disease [17,20,27,41]. Regarding the findings of this systematic review, a serology test had been performed in 59.5% of the studies, and it was positive in 42% of the cases. Although this study could not statistically confirm the exact role of serology in the detection of pericardial HC, based on the data presented, we suggest that serology cannot be depended on alone in the diagnosis of cardiac HC. Concerning the imaging modalities, echocardiography was the most commonly used tool (62.8%), followed by a CT scan (52%).

**Table 3.** The baseline characteristics of the patients

| Variables                                    | Frequency/<br>Percentage |
|--|--------------------------|
| Demographics                                 |                          |
| Gender                                       |                          |
| Male   | 75 (50.7%)               |
| Female                                       | 72 (48.6%)               |
| Non-Identified                               | 1 (0.7%)                 |
| Patients' age (Years)                        |                          |
| 5-10   | 3 (2%)                   |
| 12-20  | 22 (15%)                 |
| 21-30  | 30 (20.3%)               |
| 32-39  | 31 (21%)                 |
| 41-49  | 24 (16%)                 |
| 51-60  | 20 (13.5%)               |
| 61-70  | 11 (7.4%)                |
| 71-80  | 5 (3.4%)                 |
| >80  | 1 (0.7%)                 |
| Unknown                                      | 1 (0.7%)                 |
| Mean age                                     | 38.36                    |
| Clinical presentations                       |                          |
| Chest pain                                   | 64 (43%)                 |
| Dyspnea                                      | 53 (36%)                 |
| Asymptomatic                                 | 14 (10%)                 |
| Cough  | 13(9%)                   |
| Palpitation                                  | 13 (9%)                  |
| Fever  | 12 (8%)                  |
| Syncope/Seizures                             | 8 (5.4%)                 |
| Anaphylactic Reaction                        | 5 (3.4%)                 |
| Fatigue                                      | 5 (3.4%)                 |
| History of hydatid cyst                      |                          |
| Cardiac Hydatid cyst<br>(Intra/pericardial)  | 9 (34.6%)                |
| Liver Hydatid cyst                           | 5 (19.2%)                |
| Lung Hydatid cyst                            | 4 (15.4%)                |
| Mediastinal Hydatid cyst                     | 4 (15.4%)                |
| Lung and liver Hydatid cyst<br>(Combination) | 2 (7.7%)                 |
| Intracerebral hydatid cyst                   | 1 (3.85%)                |
| Systemic Hydatidosis                         | 1 (3.85%)                |

The management of cardiac and pericardial HCs is generally divided into surgical intervention and medication. Surgical intervention usually includes a median sternotomy with a cardiopulmonary bypass. Further dissection should be done to reach the location of the cyst to do puncture, enucleation, and aspiration using hypertonic saline solution. Resection is regarded as an important technique to prevent a recurrence, and if required, pericardiectomy is supposed to be done carefully to avoid injury to the phrenic nerves [15,24,28,]. Abou-Bakir et al., and some other studies revealed that using medical treatment such as albendazole following surgical intervention can lead to a better outcome and lessen the risk of recurrence [15,101, 110]. Endoscopic surgery is a minimally invasive technique, but due

**Table 4.** The correlation of hydatidosis history and cyst location with recurrence

| Variables              | Recurrence       |             | P-value        |       |
|------------------------|------------------|-------------|----------------|-------|
|                        | Yes              | No          |                |       |
| History of Hydatidosis | Yes              | 3<br>(100%) | 23<br>(15.9%)  | 0.005 |
|                        | No               | 0<br>(0%)   | 122<br>(84.1%) |       |
| Location of cyst       | Only pericardium | 2<br>(2.4%) | 81<br>(97.6%)  | 0.59  |
|                        | Multi-location   | 1<br>(1.5%) | 64<br>(98.5%)  |       |

to the risk of anaphylactic shock, few surgeons may perform this approach. However, it has been reported that laparoscopic surgeries had satisfactory outcomes in treating hydatid disease of abdominal organs like the liver and omentum. Concerning this vision, Akkus et al., managed a case of pericardial HC using video-assisted thoracoscopic surgery (VATS) and the outcome was content. They proposed that this technique may be associated with incomplete cyst removal and recurrence. Therefore, determining the accuracy and safety of this technique in the management of pericardial HC requires further investigations and studies with large sample sizes. Furthermore, it may be challenging for surgeons to use endoscopic techniques at a time of active heart beating. The drawback of these techniques is that they can only be used whenever there is a single unruptured cyst without the involvement of other structures like the myocardium, great arterial, or venous systems [16]. Some studies have managed their cases with only medication (albendazole) without surgical intervention, and they reported acceptable outcomes [7,26,34,41]. Surgery has been reported as the definitive treatment of cardiac HC due to life-threatening complications, and postoperative medication such as albendazole and mebendazole has been suggested to prevent recurrence [2,17,29]. A follow-up of two years with medical therapy has been recommended [51]. In the present review, the major treatment was surgical intervention in most of the cases (87.8%). The surgical approaches were commonly median sternotomy (25.7%) and thoracotomy (19%). Cystectomy was the major technique of cyst removal (72.3%), while pericardiectomy was conducted in only 6 cases (4%). Thirteen cases (8.8%) had been managed with conservative treatment, and recurrence occurred in two of them. For this reason, this study recommends surgical intervention over conservative treatment. The results of this review statistically revealed a significant correlation between the history of hydatidosis with the recurrence, and this indicates that any case of the previous HC must be under intensive follow-up due to this risk. In addition, the recurrence was uncorrelated to the HC location. The follow-up period was mostly ranged from one month to one year. In total, the mortality rate was 2.7%, and three of them died prior to any intervention.

**Table 5.** Diagnostic modalities and management outcomes.

| Variables   | Frequency/<br>Percentage |
|---|--------------------------|
| <b>Diagnostic modalities</b>                      |                          |
| Serology  |                          |
| Positive  | 62 (42%)                 |
| Negative  | 26 (17.5%)               |
| Unavailable                                       | 60 (40.5%)               |
| Echocardiography                                  | 93 (62.8%)               |
| Computed Tomography (CT) scan                     | 77 (52%)                 |
| Chest X-ray                                       | 65 (43.9%)               |
| Magnetic resonance imaging (MRI)                  | 45 (30.4%)               |
| Angiography                                       | 5(3.38%)                 |
| Ultrasound  | 3 (2%)                   |
| Fluoroscopy                                       | 1(0.7%)                  |
| Ventriculography                                  | 1(0.7%)                  |
| <b>Location of the Hydatid cyst(s)</b>            |                          |
| Pericardium                                       | 83 (56%)                 |
| Multiple location                                 | 65 (44%)                 |
| 2   | 40 (61.5%)               |
| Pericardium + Ventricle                           | 15 (23.07 %)             |
| Pericardium + Myocardium                          | 11 (16.92%)              |
| Pericardium + liver                               | 8 (12.3%)                |
| Pericardium + Mediastinum                         | 4 (6.15%)                |
| Pericardium + Retrocardiac                        | 1 (1.538%)               |
| Pericardium + lung                                | 1 (1.538%)               |
| 3   | 19 (29.23 %)             |
| Pericardium +Liver/ Ventricle                     | 4 (6.15%)                |
| Pericardium +Lung/Liver                           | 3 (4.62%)                |
| Pericardium +Ventricle/Atrium                     | 3 (4.62%)                |
| Pericardium                                       | 3 (4.62%)                |
| +Myocardium/Ventricle                             | 3 (4.62%)                |
| Pericardium +Myocardium/Lung                      | 3 (4.62%)                |
| Pericardium +Lung/Ventricle                       | 2 (3.07%)                |
| Pericardium +Myocardium/ Liver                    | 1 (1.538%)               |
| 4   | 4 (6.2 %)                |
| Pericardium +Lung/Liver/Breast                    | 1 (1.538%)               |
| Pericardium                                       | 1 (1.538%)               |
| +Liver/Ventricle/Atrium                           | 1 (1.538%)               |
| Pericardium                                       | 1 (1.538%)               |
| +Lung/Myocardium/Ventricle                        | 1 (1.538%)               |
| Pericardium                                       | 1 (1.538%)               |
| +Ventricle/Atrium/Mitral leaflet                  | 1 (1.538%)               |
| 5   | 2 (3.07%)                |
| Pericardium                                       | 1 (1.538%)               |
| +Liver/Kidney/Atrium/<br>Intraperitoneal          | 1 (1.538%)               |
| Pericardium+Liver/Myocardium/<br>Ventricle/Atrium | 1 (1.538%)               |
| <b>Management</b>                                 |                          |
| Surgery (Undefined type)                          | 61 (41%)                 |
| Median Sternotomy                                 | 38 (25.7%)               |
| Thoracotomy                                       | 28 (19%)                 |
| Conservative                                      | 13 (8.8%)                |
| None  | 5 (3.4%)                 |

|  |                 |
|--|-----------------|
| Video-assisted thoracoscopic surgery (VATS)            | <b>1 (0.7%)</b> |
| Thoracoabdominal approach                              | 1 (0.7%)        |
| Transdiaphragmatic approach                            | 1 (0.7%)        |
| <b>Major technique of cyst removal</b>                 |                 |
| Cystectomy   | 107 (72.3%)     |
| Unknown  | 17 (11.5%)      |
| Conservative + None                                    | 18 (12.2%)      |
| Pericardiectomy  | 6 (4%)          |
| <b>Follow up</b>                                       |                 |
| Less than one month                                    | 6 (4%)          |
| 1 month - 1 years                                      | 28 (19%)        |
| >1 year - 2 years                                      | 15 (10.1%)      |
| > 2 years - 3 years                                    | 16 (10.8%)      |
| 4 years - 6 years                                      | 4 (2.7%)        |
| 7 years - 10 years                                     | 5 (3.4%)        |
| > 10 years   | 2 (1.4%)        |
| Unknown  | 72 (48.6%)      |
| <b>Medication (During the six months of follow up)</b> |                 |
| Albendazole  | 60 (40.5%)      |
| Mebendazole  | 2 (1.4%)        |
| None   | 86 (58.1%)      |
| <b>Mortality and Causes</b>                            |                 |
| Four cases died  | (2.7%)          |
| Heart Failure  | 1 (25%)         |
| Staphylococcus Sepsis Infection                        | 1 (25%)         |
| Anaphylactic shock                                     | 1 (25%)         |
| Unknown  | 1 (25%)         |

## 5. Conclusion

Pericardial HC is more common in subtropical regions. The major diagnostic modality of this disease is echocardiography, and serological tests cannot be relied on alone. The definitive treatment of a pericardial HC is surgery, mainly through a median sternotomy. A history of hydatidosis increases the likelihood of recurrence, and an extensive follow-up is required.

## 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.

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manuscript; FHK and AMS confirmed the authenticity of the data; all authors approved the final version of the manuscript.

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