https://doi.org/10.1016/j.hpb.2024.12.001 HPB

ORIGINAL ARTICLE

Hepato-thoracic cystic echinococcosis transit. Clinical features, postoperative complications and hospital mortality. A systematic review

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Abstract

Background: Hepato-thoracic hydatid transit (HTT) is an evolutionary complication of hepatic cystic echinococcosis. This study aimed to report the available evidence regarding postoperative complications (POC) and hospital mortality (HM).

Methods: Systematic review. Studies related to HTT were included. Searches were performed in Trip Database, SciELO, BIREME-BVS, WoS, PubMed, EMBASE and SCOPUS. Primary outcomes: POC and HM. Secondary outcomes: publication date, origin and designs, number of patients, cyst type, hospital stance, treatments; and methodological quality (MQ) of studies applying MInCir-T and MInCir-Pr_2 scales. Descriptive statistics, weighted means (WM) and their comparison using least squares logistic regression, and meta-analysis of prevalence of POC and HM were applied.

Results: 604 studies were retrieved (101 met selection criteria, representing 1020 patients). WM age: 42.6 years, 58.3 % male. Reports are mainly from Spain (19.8 %) and Turkey (17.8 %). With a WM of 18.3 days of hospital stance, it was verified 28.9 % of POC, 12.6 % needed re-interventions, and 9.7 % died. MQ of studies: 9.1 ± 1.9 (MInCir-T) and 13.2 ± 2.9 (MInCir-Pr_2). Comparing the behavior of variables in two periods (1983–2002 vs. 2003–2024), statistically significant differences were observed in POC, HM, and reinterventions.

Conclusion: HTT is associated with high POC, and significant HM, despite the passage of time.

Received 24 October 2024; accepted 2 December 2024

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Introduction

Echinococcus granulosus (*E. granulosus*) with their distinct genotypes, primarily G1-G3 and G6-G7, is the etiological agent responsible for hepatic cystic echinococcosis (HCE).[1] The infection becomes evident approximately three weeks after the ingestion of the eggs giving rise to the formation of hepatic cysts with a diameter ranging from 2 to 3 cm, three months after the infection.[1,2]

As the cyst develops, the pressure it generates results in atrophy and fibrosis of the hepatic parenchyma. Consequently, continued growth may lead to the adherence of the cyst to the diaphragm, subsequent thinning, and progression into the pleural cavity. Adhesion to the pulmonary parenchyma and bronchial tree follows, leading to the development of a cysto-bronchial communication, associated with the phenomenon known as "hydatid cough" (the expulsion through the mouth, following a coughing episode, of hydatid fluid with or without daughter cysts or fragments of the germinal membrane). An alternative manifestation in this pathogenic process involves the concurrent development of cysto-biliary communications,[3–6] giving rise to "biliptysis" when the transition into the thoracic cavity concludes with the opening to the airway,[7,8] resulting in the expectoration of sputum containing bile. This entire process is referred to as hepatothoracic transit (HTT).[9,10]

The rupture of HCE from the abdomen into the thorax is an uncommon complication, occurring in approximately 0.6 %–16.0 % of cases of HCE[8,11–13]; with a cumulative estimated incidence of 0.02–0.05 cases over 5 years,[10,14,16] associated with postoperative complication (POC) rates up to 42.5 %,[16] and hospital mortality (HM) rates of up to 23.6 %.[17,18] Fig. 1.

Presentation (for original articles only). This paper is not based on a previous communication to a society or meeting.

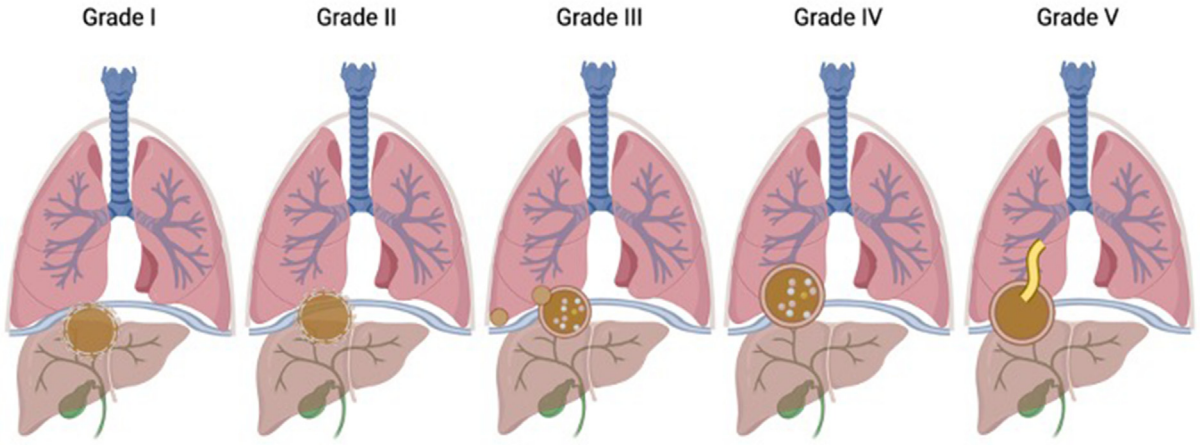

Figure 1 Diagram of the etiopathogenesis of HTT. Figure created with BIORENDER.COM, accessed on May 29, 2023

Thus, HTT can progressively involve the diaphragm, the pleural cavity, and eventually the bronchial tree, as described by Gómez et al., in their proposed classification of HTT, in which it is noted regarding pleural involvement: "diaphragmatic perforation with growth of the cyst within the pleural cavity, called pleurothoracic vesiculation".[12]

One of the earliest reports of HTT was made by Peacock in 1850, who reported the findings and clinical course of two patients (a 20-year-old woman and a 31-year-old man). However, Peacock himself commented in his report that the first case was described by Collet in Medical Transactions in 1772.[19]

The aim of this study was to describe the available evidence regarding the clinical characteristics of HTT, as well as the associated POC and HM resulting from its treatment.

Materials and methods

This manuscript has been reported in line with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) and AMSTAR (Assessing the methodological quality of systematic reviews) Guidelines.

Study protocol

Registered in PROSPERO (International prospective register of systematic reviews, NIHR).

Study design

Systematic review (SR) of prognostic scenarios.

Eligibility criteria

Primary studies related to HTT were included; without language restriction or year of publication. Editorials, letters to the editor, narrative reviews (narratives and systematics), consensus documents and discussions, and articles contaminated with patients infected with *Echinococcus multilocularis*, *Echinococcus vogelii,* or other parasites were excluded. Articles regarding BBF following hepatic surgery for HCE, BBF of traumatic, oncological, or inflammatory origin (subphrenic abscess, biliary tract lithiasis, etc.) were also excluded.

Information sources

The following metasearch engines, libraries, and databases were reviewed: Trip Database, SciELO, BIREME-BVS, Web of Science (WoS), PubMed, EMBASE, and SCOPUS. The search and recruitment of articles closed on May 30, 2024.

Search criteria

This was carried out using the PECO components (population study [P: patients with HCE], exposure [E: HTT development], comparator [C: None], and result [O: postoperative complications and hospital mortality]). Sensitive searches were carried out using MeSH and DeCS terms (Echinococcosis, Hepatic; Echinococcosis, Hepatothoracic transit, hepatic pulmonary hydatidosis), free words (hepatic hydatid cyst; liver hepatic cyst; hepatic hydatidosis; liver hydatidosis; hepatic cystic echinococcosis; hepato-thoracic transit), and boolean connectors (AND and OR), with strategies adapted to each database (Table 1). A manual and cross-reference search was also carried out.

Study selection

Identified documents in each information source were filtered by duplication between databases. They were subsequently examined by title and abstract, applying eligibility criteria. The articles were then extensively analyzed by the reviewers (CM, JR, TO and CR-P), all experienced in searching and analyzing biomedical studies. Discrepancy situations were resolved by consensus.

Data collection

Critical review of each selected article, as well as the data extraction and its subsequent verification, was carried out by two researchers (CM and JR). Then, data was collected in an Excel

Table 1 Search strategies and results obtained for each source of information (N = 564)

Sources of information	Search strategies
BVS-BIREME (n = 113)	(Echinococcosis OR "cystic echinococcosis" OR "hydatidos*" OR "echinococcus granulosus infection*" OR "infection echinococcus granulosus" OR "hepatic hydatid cyst" OR "liver hydatid cyst" OR "quiste hidatídico" OR "equinococosis quística" OR "equinococosis hepática") AND (biliptisis OR (hepatothoracic transit) OR (hepatopulmonary hydatidosis) OR "thoracic involvement" OR "Bronchobiliary fistula*" OR "hepatopulmonary fistula" OR "hydatid vomica" OR (transito hepatotoracico) OR (hidatidosis hepatopulmonar) OR "fístula bronquealveolar")
EMBASE (n = 142)	('Liver hydatid cyst'/exp OR 'cystic echinococcosis' OR 'echinococcal cyst*' OR 'echinococcal disease' OR 'echinococcal infection*' OR 'echinococcal infestation' OR 'echinococcus cyst' OR 'echinococcus disease' OR 'echinococcus granulosus cyst' OR 'echinococcus granulosus infection' OR 'echinococcus infection*' OR 'echinococcus infestation' OR 'hydatic cyst' OR 'hydatid cyst' OR 'hydatid disease' OR 'hydatidosis' OR 'infection by echinococcus granulosus' OR 'infection of echinococcus granulosus') AND (biliptisis OR (hepatothoracic AND transit) OR (hepatopulmonary AND hydatidosis) OR 'thoracic involvement' OR 'bronchobiliary fistula*' OR 'bronchobiliary fistula'/syn OR 'hepatopulmonary fistula' OR 'hydatid vomica')
PubMed (n = 104)	("Echinococcosis"[MeSH Terms] OR "Echinococcosis" [all fields] OR "cystic Echinococcosis" [all fields] OR "hydatidos*" [all fields] OR "echinococcus granulosus infection*" [all fields] OR "infection echinococcus granulosus" [all fields] OR "hepatic hydatid cyst" [all fields] OR "liver hydatid cyst" [all fields] OR "Liver Hydatidosis" [all fields] OR "Hepatic hydatidosis" [all fields]) AND ("biliptisis"[all fields] OR ("Hepatothoracic" [all fields] AND ("transit" [all fields] OR "transited" [all fields] OR "transiting" [all fields] OR "transition" [all fields] OR "transitional" [all fields] OR "transitionals" [all fields] OR "transitioned" [all fields] OR "transitioning" [all fields] OR "transitions" [all fields] OR "transits" [all fields])) OR ("hepatopulmonary" [all fields] AND ("echinococcosis" [MeSH terms] OR "Echinococcosis" [all fields] OR "hydatidosis" [all fields])) OR "thoracic involvement" [all fields] OR "bronchobiliary fistula*" [all fields] OR "hepatopulmonary fistula" [all fields] OR "hydatid vomica" [all fields])
SciELO (n = 4)	(((Echinococcosis OR "cystic echinococcosis" OR "hydatidos*" OR "echinococcus granulosus infection*" OR "infection echinococcus granulosus" OR "hepatic hydatid cyst" OR "liver hydatid cyst" OR "quiste hidatídico" OR "Equinococosis quística" OR "Equinococosis Hepática"))) AND (((biliptisis OR (hepatothoracic transit) OR (hepatopulmonary hydatidosis) OR "thoracic involvement" OR "bronchobiliary fistula*" OR "Hepatopulmonary fistula" OR "hydatid vomica" OR (Transito hepatotoracico) OR (hidatidosis hepatopulmonar) OR "fístula bronqueobiliar")))
Scopus (n = 138)	TITLE-ABS-KEY ((echinococcosis OR "cystic echinococcosis" OR "hydatidos*" OR "echinococcus granulosus infection*" OR "infection echinococcus granulosus" OR "hepatic hydatid cyst" OR "liver hydatid cyst" OR "echinococcal cyst*" OR "echinococcal disease" OR "hydatid cyst" OR "hydatid disease") AND (biliptisis OR (hepatothoracic AND transit) OR (hepatopulmonary AND hydatidosis) OR "Thoracic Involvement" OR "bronchobiliary fistula*" OR "hepatopulmonary fistula" OR "hydatid vomica"))
Trip Database (n = 4)	(Echinococcosis OR "cystic echinococcosis" OR "hydatidos*" OR "echinococcus granulosus infection*" OR "infection echinococcus granulosus" OR "hepatic hydatid cyst" OR "liver hydatid cyst") AND (biliptisis OR (hepatothoracic transit) OR (hepatopulmonary hydatidosis) OR "thoracic involvement" OR "Bronchobiliary fistula*" OR "Hepatopulmonary fistula" OR "hydatid vomica")
WoS (n = 59)	TS=((Echinococcosis OR "cystic echinococcosis" OR "hydatidos*" OR "echinococcus granulosus infection*" OR "infection echinococcus granulosus" OR "hepatic hydatid cyst" OR "liver hydatid cyst") AND (biliptisis OR (Hepatothoracic transit) OR (Hepatopulmonary hydatidosis) OR "thoracic involvement" OR "bronchobiliary fistula*" OR "hepatopulmonary fistula" OR "hydatid vomica"))

spreadsheet (Mac Excel, version 15.24; 2016 Microsoft Corporation®).

Data items and outcomes

Primary outcome variables were "POC or postoperative morbidity", defined as any pathologic processes that affect patients after a surgical procedure in patients with HCE with HTT (they may or may not be related to the disease for which the surgery was done, and they may or may not be direct results of the surgery); measured dichotomously (present or absent) and applying the Clavien & Dindo classification.[20] The other primary outcome was HM, defined as a vital statistic measuring or recording the rate of death from any cause in hospitalized patients after HCE with HTT; measured dichotomously (yes or no). Secondary outcome variables were year of primary articles (descriptive in percentages), geographical origin of the studies, designs of primary studies, number of cases considered in each study, main clinical manifestation (descriptive in percentages), anatomical location and type of cyst, type of HTT according to Gómez proposal (Table 2),[12] surgical procedure performed and other type of associated therapeutics (descriptive in percentages), hospital stay (in days), and need for reoperations.

Statistics

Descriptive statics was applied with the calculation of percentages, averages and weighted averages.

Additional analyses

Meta-analysis was performed comparing the behavior of the variables under study in two periods of time (1983–2002, period A vs. 2003–2024, period B), applying a weighted least squares regression model (the weights were the number of patients of

Table 2 Type of HTT adapted from Gómez et al.[12] (Fig. 1)

Grade	Name	Definition
I	Adhered cyst	Presence of adhesions between the cyst wall and the diaphragm, but no perforation.
II	HCE transit	Diaphragmatic perforation with little invasion of the cyst into the thoracic cavity. The defect requires repair.
III	Pleurothoracic vesiculation (cyst in "sandglass")	Diaphragmatic perforation with growth of the cyst within the cavity, or presence of daughter vesicles.
IV	Disease of the pulmonary parenchyma	Connection between the cyst and the bronchial branches, or compression and atelectasis of the lung parenchyma.
V	Chronic bronchial fistula	Complication associated with chronic sequelae or evolution of HCE.

each scientific article). Dichotomization was decided because during the exploratory analysis of the data, we verified that this point divided the sample under study into similar parts.

In addition, meta-analyses of proportions were performed on the variable's postoperative complications and mortality, considering primary articles with more than 10 individuals. A random-effects model was employed using the Hartung-Knapp-adjusted inverse variance method and the Freeman-Tukey method was used for transformation of proportions. Heterogeneity was determined using I^2 and publication bias was assessed qualitatively and by Funel Plot. The R Studio Version 4.4.0 package was used.

Methodological quality (MQ) of primary studies was determined applying MInCir scales for therapeutic procedures (MInCir-T) and prognosis (MInCir-Pr_2).[21,22] Both used scales for determine MQ, are valid (face and content validity, and construct validity for extreme groups) and reliable (interobserver reliability). MInCir-T scale, composed of 3 domains and 6 items: the first related to the study design; the second to the population sample size, and the third related to the methodology used; according to which, a score which represents the sum of the 3 domains is generated, with a final score that can vary between 6 and 36 points (6 points being the worst MQ study and 36 being the best), with a cut-off point to define the construct of 18 points.[21] And MInCir- Pr_2 scale, composed of 4 domains and 11 items (study design, population sample size, methodology used, analysis and conclusions); according to which, a score which represents the sum of the 4 domains is generated, with a final score that can vary between 7 and 60 points (7 points being the worst MQ study and 60 being the best), with a cut-off point to define the construct of 33 points.[22] Then data were entered to a spreadsheet and calculation of weighted means was applied, and its comparison applying a weighted least squares regression model.

The behavior of the study variables was determined by the weighted averages of each one, which is the sum of the product of the study variable by the MQ score of where the article came from, divided by the sum of methodological quality scores of all the study articles. For this purpose, the following formula will be applied ($\boldsymbol{WA} = \frac{\Sigma \mathbf{Xi} * \mathbf{ei}}{\Sigma \mathbf{ei}}$), in which WA is weighted average; Xi, the value achieved for the outcome in the study ***i*** (for all outcomes); ***ei***, represents the methodological quality score obtained in study ***I***; and $\boldsymbol{\Sigma ei}$, correspond to the sum of the scores obtained in all the studies.[21,22]

Risk of bias in individual studies: The likelihood of inaccuracy in the estimate of causal effect in primary studies was assess applying user's guides, checklists, and MQ scales: MInCir-T and MInCir-Pr_2.[21,22] This allowed us to a better organization of the information synthesis.

Ethics

To reduce possible biases in the selection and analysis, masking of authors and study centers were implemented, coding and masking the primary articles, and deleting the names of authors and centers.

Results

Study selection

564 studies were retrieved from the search, and 42 were obtained from cross-searches. Of these, 340 were eliminated due to duplication of information sources. Therefore, 265 articles were examined by title and abstract, 148 of which were discarded because they were considered "not related" to the research; leaving 118 articles to evaluate eligibility by reading the full text (12 were not retrieved). An in-depth analysis of the selected studies was then performed, excluding 7 studies based on the review criteria. Finally, 101 studies correspond to the study sample of this review[7–18,23–45,46–65,66–90,91–111] (Fig. 2).

Study characteristics

Included studies were case reports (77 studies, 76.2 %), case series (23 studies, 22.8 %), and retrospective cohort (1 studies, 1.0 %); representing 1020 patients and 1036 cysts, with a weighted mean age of 42.6 years, 58.3 % male.

Synthesis of results

27.7 % of the included articles were published in the last decade, even though it still has 7 months remaining for potential new publications (Table 3). Spain had the highest number of publications (20 studies, 19.8 %; Table 4).

Weighted means of cyst diameter was 12.9 cm (other clinical characteristics can be verified in Table 5). Clinical manifestations

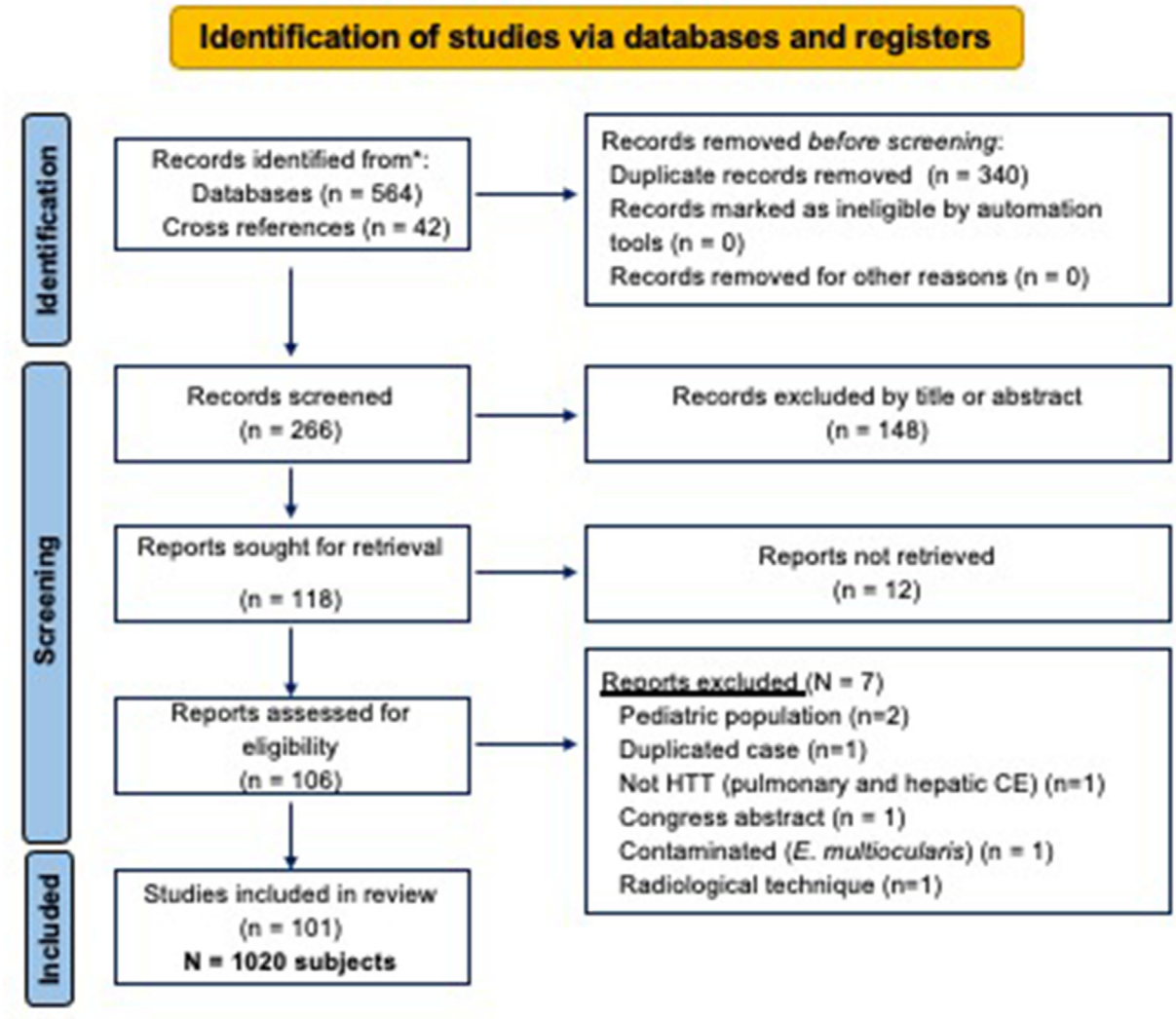

Figure 2 Flow chart of the analyzed studies

Table 3 Year of publication of the included studies (N = 101)

Time period (years)	Nº of studies	%
2020–2024	12	11.8
2015–2019	16	15.8
2010–2014	9	8.9
2005–2009	21	20.7
2000–2004	13	12.8
1995–1999	4	4.0
1990–1994	7	7.0
1985–1989	7	7.0
1980–1984	4	4.0
1975–1979	4	4.0
1974 or less	4	4.0
TOTAL	**101**	**100**

and therapeutic options observed can be found in Table 6. The majority of cysts were classified as grades IIIa, IIIb, and IV according to the WHO-IGWE classification (84.8 %). Seventeen patients (1.7 %) also had concomitant pulmonary echinococcosis.

With a weighted mean of 18.3 days of hospital stance, it was verified a weighted mean of 28.9 % of POC (77.7 % Clavien & Dindo III and IV), 12.6 % needed re-interventions, and 9.7 % died (Table 5). The etiology and Clavien & Dindo distribution of postoperative complications is summarized in Table 7.

Meta-analysis identified that postoperative complications in HHT patients have a prevalence of 0.27 (95 % CI: 0.21–0.33), with moderate heterogeneity (I^2: 66 %) (Fig. 3a and b); similarly, HM has a prevalence of 0.07 (95 % CI: 0.04–0.11) with moderate heterogeneity (I^2: 66 %) (Fig. 3c and d).

MQ of primary studies was 9.1 ± 1.9 applying MInCir-Therapy scale (minimum 6 and maximum 36 points. Only 1

Table 4 Origen of included studies (N = 101)

Study origin	Nº of studies	%
Spain	20	19.8
Turkey	18	17.8
Morocco	14	13.8
Italy	6	5.9
Tunisia	6	5.9
Chile	4	4.0
Greece	4	4.0
India	4	4.0
Iran	3	3.0
UK	3	3.0
Australia	2	2.0
Russia	2	2.0
Serbia	2	2.0
Others[a]	13	12.8
Total	**101**	**100**

[a] Brunei, Bulgaria, Canada, Czechia, Colombia, Croatia, France, Lebanon, New Zealand, Peru, Romania, Saudi Arabia, and Uruguay (one each).

Table 5 Clinical characteristics of the patients (N = 962)

Variable	Weighted mean	Min.	Max.
Age (years)	42.6	12	80
Cyst diameter (cm)	12.9	4	19
Total leukocytes ($10e^3$/uL)	12,940	15	41000
Total bilirubin (mg/dL)	1.2	0.6	6.0
Direct bilirubin (mg/dL)	1.6	0.7	5.0
Gamma-glutamyl transferase (U/L)	624	110	718
Alkaline phosphatase (U/L)	376	520	1448
ASAT (U/L)	64	23	120
ALAT (U/L)	71	21	163
Postoperative complications (%)	28.9	0	100
Re-interventions (%)	12.6	0	100
Hospital stance (days)	18.3	8	56
Hospital mortality (%)	9.7	0	40
Follow-up (months)	64.9	1	102

ASAT: aspartate aminotransferase.
ALAT: alanine aminotransferase.

article achieved score above the cutoff point that defines the construct), and 13.2 ± 2.9 applying MInCir-Prognosis scale (minimum 7 and maximum 60 points. No article obtained a score above the cutoff point that defines the construct). So, the likelihood of inaccuracy in the estimate of causal effect in primary studies is high. When measuring MQ in the time periods under study, no statistical differences were observed (Table 8).

Finally, when comparing different in study variables in two different time-periods (before vs. after 2002), it was observed statistical differences in age, POC, HM, re-interventions, total leukocytes, total bilirubin, Alkaline phosphatase and hospital stance (Table 8).

Table 6 Characteristics of the included studies and its patients

Variables	Nº cases	%
Most frequent clinical presentation (n = 996)[a,b]		
Fever	732	73.5
Cough	686	68.9
Biliptysis	259	26.0
Hemoptysis	207	20.8
Hydatid vomiting	213	21.4
Type of cyst, WHO classification (n = 782) [a]		
CE1	29	3.7
CE2	76	9.7
CE3A	159	20.3
CE3B	121	15.5
CE4	383	49.0
CE5	14	1.8
Type of HTT according to Gómez proposal (n = 892) [a]		
Grade I	86	9.6
Grade II	169	18.9
Grade III	371	41.6
Grade IV (Fig. 4)	162	18.2
Grade V	104	11.7
HTT treatment (n = 706) [a]		
Phrenoplasty[c] + pleural drainage (Fig. 5)	457	64.7
Phrenoplasty[c] + pulmonary resection	115	16.9
Phrenoplasty[c] + pulmonary decortication	73	10.3
Pleural drainage alone	32	4.5
Endoscopic biliary drainage	29	4.1
HCE treatment (n = 824) [a]		
Partial cystectomy	439	53.3
Pericystectomy (Fig. 5)	208	25.2
Liver resection	76	9.2
Cystostomy	47	5.7
Watch and wait approach	13	1.6
Other procedures reported [d]	41	5.0
Associated pharmacotherapy (n = 82) [a]		
Albendazole	32	39.0
Antibiotics	21	25.6
Antibiotics + Albendazole	5	6.1
Mebendazole	6	7.3
Antibiotics + Mebendazole	2	2.4
None	16	19.5

(*continued on next page*)

Table 6 (*continued*)

Variables	Nº cases	%
POC - Clavien & Dindo (n = 269) [a]		
Grade I	38	14.1
Grade II	22	8.2
Grade IIIa	60	22.3
Grade IIIb	69	25.7
Grade IVa	59	21.9
Grade IVb	21	7.8

[a] The values represent the reporting of the variable in only some of the primary studies.
[b] In some cases, more than one was reported.
[c] Phrenoplasty with sutures (n = 509) and with mesh use (n = 136).
[d] Choledocostomy with Kehr tube (n = 25), endoscopic papillotomy (n = 13), and partial cystectomy via laparoscopy (3). For the treatment of residual cyst cavities, the use of simple drainage (n = 193), omentoplasty (n = 103), capitonage (n = 94), and nothing (n = 63) was reported.

Possible biases in the review process

Additional information was requested from the authors to expand or verify some study data, unfortunately, with no response. Therefore, missing data may introduce bias in this review. The risk of missing studies was reduced by searching for cross references.

Table 7 Postoperative complications (n = 269)[a]

Etiology	Nº	%
Surgical wound infection	44	16.4
Pleural empyema	30	11.2
Sepsis	23	8.6
Atelectasis	22	8.2
Bronchopleural fistula	20	7.4
Hemothorax	19	7.1
Pneumothorax	18	6.7
Pneumonia	16	5.9
Pleural effusion	13	4.8
Pulmonary hemorrhage	11	4.1
Acute respiratory failure	10	3.7
Deep vein thrombosis	8	2.9
Bronchobiliary fistula	8	2.9
Pulmonary thromboembolism	6	2.2
Infected residual cavity	5	1.9
Cholangitis	5	1.9
Subphrenic abscess	4	1.5
Acute heart failure	2	0.7
Others[b]	5	1.9
TOTAL	**269**	**100**

[a] This variable was not reported in all articles.
[b] Anaphylaxis, renal failure, arrythmia, biliary fistula, and angiocolitis one each.

Risk of bias between studies

There may be publication bias, given the concentration of studies from a few countries, and the paucity of studies from other geographic areas, places, and countries (Table 4). Finally, is important to resalt the fact that only 8 articles obtained scores above the cut-off point defining the construct MQ for therapy studies.

Discussion

Summary of the evidence

This is the first SR that resumes the available evidence about HTT. The information in this SR was recovered from systematic searches carried out in 7 information sources (metasearch engines, libraries, and databases), including more than 50 years of experience in the diagnosis and treatment of THT worldwide. It corresponds to a compilation of 101 primary articles, which represent 1020 patients treated in different countries, but most frequently in Spain, Turkey, and Morocco.

The first drawback of the selection of MeSH terms and free words, to define the search strategies, was to show that HTT concept is apparently unclear, or is not very widespread, since the number of publications found in whose title or abstract appears HTT is very reduced. Most of the primary articles with which we worked had other types of denominations, such as "hepato-thoracic hydatid transit, hepatic echinococcosis with thoracic involvement, thoracic extension of hydatid cysts of the liver, thoracobilia; bronchobiliary, pleurobiliary, biliobronchial, biliary-bronchial, biliopulmonary, hepatobronchial and broncho-hepatic-cystic fistula; pleurobilia, bilithorax, bilio-tracheal fistula", etc. Regarding it, is important to note that all these concepts only define the existence of a communication between a HCE (with diaphragmatic involvement) and the thoracic cavity, with or without communication with the respiratory tree. On the other hand, the use of different definitions for the same concept, may depict a source of selection and information biases.

There are at least 5 proposals for the classification of HTT,[11–13,68,94] highlighting the proposal of Mestiri (which consists of 4 lesion types and 9 subgroups[68]); the classification of Gómez (5 evolutionary grades[12]); and the categorization of El Hammoumi (with 2 types of bilio-pleural fistula and 5 subgroups[11]). So, we chose the proposal of Gómez,[12] because it seems to be the one that best approximates the clinical-surgical reality.

On the other hand, the anatomical access option may be an issue of preference and conditions but must allow us to treat the entire problem (treatment of the liver, diaphragm, and lung) in a single step. So, there are different strategies: exclusive thoracotomy, thoraco-phreno-laparotomy, and laparotomy alone. Independently of the access option, the liver treatment could be done by partial or total pericystectomy with or without omentoplasty; resection of the protruding dome and closure of fistulas; and

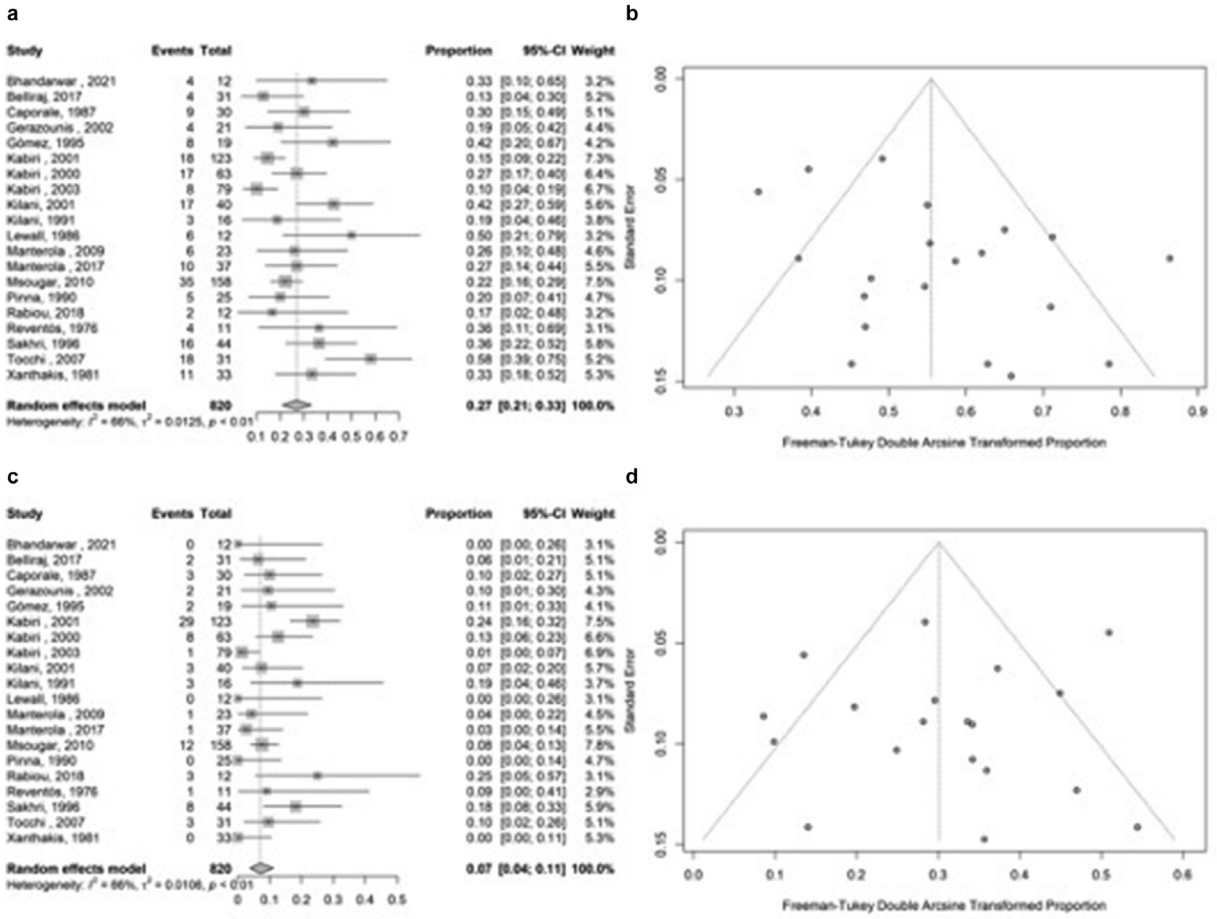

Figure 3 Meta-analyses of prevalences:
a) Forrest plot of POC
b) Funnel plot of POC
c) Forrest plot of HM
d) Funnel plot of HM

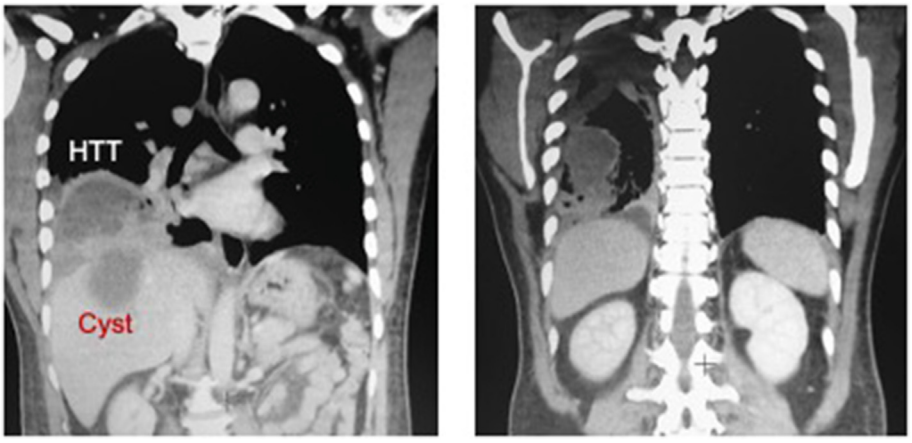

Figure 4 Tomographic image of a HCE connected to the right pleural space. Grade 4 of Gómez proposal

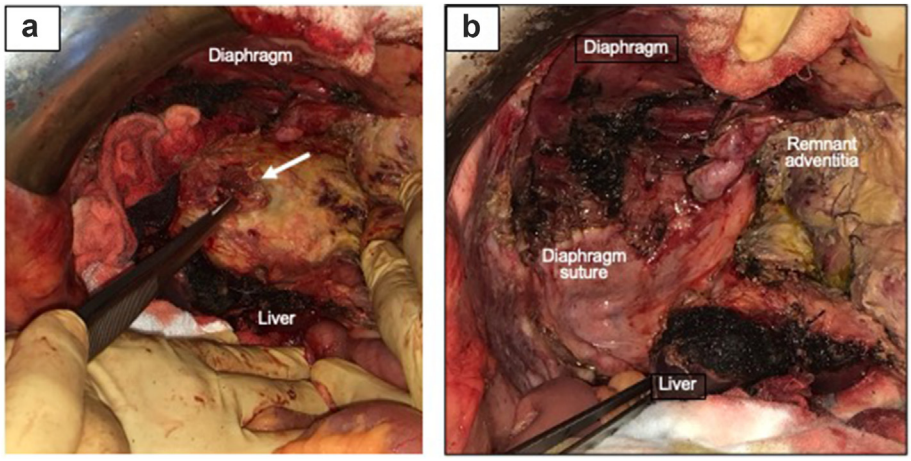

Figure 5 Operative views of a in J laparotomy. The liver has been completely mobilized and freed from its ligaments. The cyst has been resected and disconnected from the diaphragm

a) A bile-pleural fistula indicated by the forceps, can be visualized after phrenopulmonary disconnection

b) The adventitia adhered to the diaphragm was completely resected. The fistula closed, and the diaphragm repaired with continuous suture of absorbable material. The hepatic remnant and adhered adventitia to the liver can be appreciated

Table 8 Clinical characteristics of patients and studies, and behavior of some variables in the two evaluated periods of time

Variables (Weighted means)	T O T A L (Nº studies = 101) (Nº cases = 1020)	1973–2002 (Nº studies = 39) (Nº cases = 550)	2003–2023 (Nº studies = 62) (Nº cases = 470)	*p*
Age (years)	42.6 ± 14.1	45.6 ± 12.9	41.5 ± 14.9	0.180
Postoperative complications (%)	28.9 ± 41.6	31.7 ± 36.6	25.9 ± 11.6	**0.021**
Hospital mortality (%)	9.7 ± 9.8	13.9 ± 13.1	5.4 ± 5.8	**0.001**
Re-interventions (%)	12.6 ± 12.8	16.9 ± 13.4	8.0 ± 6.8	**0.012**
Total leukocytes ($10e^3$/uL)	12,941 ± 6711	13,327 ± 5483	10,866 ± 2353	**0.016**
Total bilirubin (mg/dL)	1.2 ± 2.1	3.8 ± 3.6	1.1 ± 0.8	0.053
Gamma-glutamyl transferase (U/L)	624 ± 256	597 ± 166	624 ± 256	0.062
Alkaline phosphatase (U/L)	376 ± 159	111 ± 34	386 ± 168	**0.041**
ASAT (U/L)	64 ± 39	65 ± 35	64 ± 41	0.785
ALAT (U/L)	71 ± 85	96 ± 46	70 ± 12	0.587
Hospital stance (days)	18.3 ± 10.9	20.7 ± 17.3	15.8 ± 25.2	0.071
Cyst diameter (cm)	12.9 ± 3.5	13.2 ± 2.9	12.6 ± 25.6	0.413
Follow-up (months)	64.9 ± 27.9	67.8 ± 33.4	63.1 ± 27.2	0.512
MQ MInCir-T scale (points)	9.1 ± 1.9	9.6 ± 2.1	8.7 ± 1.8	0.861
MQ MInCir-Pr_2 scale (points)	13.2 ± 2.9	13.5 ± 2.4	12.7 ± 1.9	0.812

ASAT: aspartate aminotransferase.
ALAT: alanine aminotransferase.
MQ: Methodological quality.
In bold, statistically significant values are highlighted.

cysto-biliary disconnection. After hepatic treatment the diaphragmatic fistula will be closed by stitches, and a phrenoplasty has to be performed even if the wound is large, with or without use of mesh. Finally, lung treatment includes releasing it from the diaphragm to access and apply simple sutures to bronchial fistulas. In cases of chronic inflammation of tissues by bile, pulmonary resections could be realized.[11]

Nevertheless, minimally invasive techniques such as video-assisted thoracoscopic surgery (VATS) can also be used. It is a safe option with excellent results for thoracic cystic echinococcosis, among many other indications.[112,113]

The frequency of POC is very high (27 %), and the etiology of these complications is predominantly severe (grades III and IV of Clavien & Dindo). However, it was significantly reduced in the

second study period (31.7 % vs. 25.9 %), although it remains at a high value (one out of every four patients will develop complications).

A similar pattern was observed with the frequency of HM, which was found to be very high (7 %). However, a drastic and significant reduction was observed between the first and second study periods (13.9 % vs. 5.4 %), reaching a still considerable value, but acceptable for a pathology as complex and severe as HTT.

It is striking that despite variations in sample size, research designs, MQ, and epidemiological characteristics (geographical location, etc.) of the primary articles, the confidence intervals described in the meta-analyses of POC and HM are similarly narrow, with moderate heterogeneity assessed by I^2.

Limitations

The available evidence on HTT was obtained only from level 4 studies (case reports, retrospective case series and historical cohorts, with reduced casuistry). On the other hand, methodological quality of primary studies was low: 9.1 ± 1.9 points for MInCir-Therapy and 13.2 ± 2.9 points for MInCir-Prognosis scales, which cutoff-points are 18 and 33 respectively.[21,22] Similarly, there is a risk of publication bias associated with the outcome of funnel plots, where an asymmetric distribution is evident in meta-analyses of POC and HM.

Conclusions

The available evidence is indicative that HTT is an evolutionary complication of HCE, which prognosis remains unfavorable, despite the advancements in surgical and resuscitative techniques, because HTT is associated with persistently high hospital stance, severe POC and HM rates, which underscores the significance of early diagnosis and timely treatment for HCE.

Assistance with the study

None.

Patient consent

Not apply.

Disclosure statement

The authors declare no conflicts of interest related to this article.

Authorship

CM: Research design, wrote the manuscript, data collection, analysis and interpretation.

JR: Interpretation, data collection, intellectual content, review accuracy and integrity of the work.

CRP: Data collection, analysis and interpretation.

TO: Data collection, analysis and interpretation.

Funding

This study was partially financed by the following grants: DIUFRO DI23-0020, Universidad de La Frontera. DIUFRO DI23-0073, Universidad de La Frontera. ANID-Subdirección de Capital Humano/Doctorado Nacional/2024-21242396.

Conflicts of interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Carlos Manterola reports financial support was provided by Universidad de La Frontera. Carlos Manterola reports a relationship with Universidad de La Frontera that includes: employment. If there are other authors, they declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.hpb.2024.12.001.