

# World Journal of *Meta-Analysis*

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**ABOUT COVER**

Dr. Rakhshan is an editorial board member of *World Journal of Meta-Analysis*, and a former lecturer in the Dental School of Islamic Azad University, Tehran, Iran. He graduated from the same university in 2004, with a DDS thesis in which he designed and implemented an AI computer vision program that could extract radiographic landmarks from lateral cephalographs. Since then, besides clinical practice, he has taught dental anatomy and morphology, and has published about 140 peer-reviewed articles on different dentistry topics. He has also peer reviewed more than 500 articles during these years, and has been the lead guest editor of the journals *Pain Research and Management*, *Computational Intelligence and Neuroscience*, and *International Journal of Dentistry*, and an associate editor of *Frontiers in Oral Health*. He is currently a PhD candidate of cognitive neuroscience at the Institute for Cognitive Science Studies, Tehran, Iran

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## Hydatidosis and the duodenum: A systematic review of the literature

Víctor de la Fuente-Aguilar, Patricia Beneitez-Mascaraque, Sergio Bergua-Arroyo, María Fernández-Riesgo, Irene Camón-García, Iris Cruza-Aguilera, Kennia Ugarte-Yáñez, José M Ramia

**ORCID number:** Víctor de la Fuente-Aguilar 0000-0001-9472-8219; Patricia Beneitez-Mascaraque 0000-0002-4394-7347; Sergio Bergua-Arroyo 0000-0001-9746-2671; María Fernández-Riesgo 0000-0003-0026-8245; Irene Camón-García 0000-0001-8366-1688; Iris Cruza-Aguilera 0000-0003-2269-7155; Kennia Ugarte-Yáñez 0000-0001-8024-7182; José M Ramia 0000-0003-1186-953X.

**Author contributions:** Ramia JM designed the research; de la Fuente-Aguilar V, Beneitez-Mascaraque P, Bergua-Arroyo S, Fernández-Riesgo M, Camón-García I, Cruza-Aguilera I, Ugarte-Yáñez K performed the research; de la Fuente-Aguilar V, Beneitez-Mascaraque P, Bergua-Arroyo S, Fernández-Riesgo M, Camón-García I, Cruza-Aguilera I, Ugarte-Yáñez K and Ramia JM analyzed the data; and de la Fuente-Aguilar V, Beneitez-Mascaraque P, Bergua-Arroyo S, Fernández-Riesgo M, Camón-García I, Cruza-Aguilera I, Ugarte-Yáñez K and Ramia JM wrote the paper.

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Víctor de la Fuente-Aguilar, Patricia Beneitez-Mascaraque, Sergio Bergua-Arroyo, María Fernández-Riesgo, Irene Camón-García, Iris Cruza-Aguilera, Kennia Ugarte-Yáñez, José M Ramia, Faculty of Medicine, Universidad of Alcalá, Alcalá de Henares 28801, Spain

José M Ramia, Servicio de Cirugía General y del Aparato Digestivo, Hospital General Universitario de Alicante and Institute for Health and Biomedical Research (ISABIAL) Alicante 03010, Spain

**Corresponding author:** José M Ramia, BSc, FACS, FRCS (Gen Surg), MD, PhD, Head, Department of Surgery, Hospital General Universitario de Alicante. Calle Pintor Baeza 11, Alicante, Spain. [jose\\_ramia@hotmail.com](mailto:jose_ramia@hotmail.com)

### Abstract

#### BACKGROUND

Injury to the duodenum (fistula formation, compression, or other complications) by a hydatid cyst (HC) is an exceptional complication.

#### AIM

To perform a systematic review of the literature on the fistulization of HC in the duodenum.

#### METHODS

Following PRISMA guidelines, a search for HC with duodenal involvement was carried out in the databases of PubMed, SCielo and EMBASE without time limits.

#### RESULTS

Fourteen patients were identified, seven men and seven women, with a mean age of 53.14 years (SD = 17.65, range: 28-78). Three out of the 14 (21%) had HC relapse. The most frequent clinical manifestations were abdominal pain and nausea and/or vomiting. Various imaging studies were performed in almost all cases, the most commonly used being abdominal computed tomography (10/14, 71%). A range of surgical techniques were reported, most frequently HC drainage (41%) and enucleations (16%). Ten of the 14 patients had no complications and one patient died. The follow-up period and recurrences could not be determined.

#### CONCLUSION

The most frequent symptoms were abdominal pain, nausea and vomiting. Computed tomography was the most used diagnostic imaging technique, and HC drainage and fistula closure *via* laparotomy was the most frequent treatment.



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However, all diagnostic and therapeutic options for HC fistulizing the duodenum had a low level of evidence.

**Key words:** Echinococcosis; Hydatidosis; Duodenum; Surgery; Review

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**Core tip:** Hydatidosis is a global zoonosis. The most frequent organs affected are the liver (50%-70%) and the lungs (20%-30%). The duodenum is an organ that is very rarely affected by hydatidosis, either primarily or secondarily. Possible forms of secondary duodenal involvement are compression, or, more frequently, fistulization from hydatid cysts located in neighboring organs. The hydatid cysts that most often fistulize the duodenum are those located in the liver (0.15% of instances), with cases also described from the pancreas and kidney. Its low prevalence and the few existing records mean that the choice of the best therapeutic management is a challenge. A systematic review let readers know all published cases and best management.

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## INTRODUCTION

Hydatidosis is a zoonosis caused by the larval form of the genus *Equinococcus* spp, and *Equinococcus granulosus* (*E. granulosus*) causes the formation of hydatid cysts (HC). The life cycle of *E. granulosus* includes different herbivores (lambs, cows, pigs, deer) and carnivores (dogs, foxes, wolves); the dog is the most frequent definitive host<sup>[1]</sup>. The distribution of *E. granulosus* is associated with livestock exploitation and temperate climate zones such as southern Europe, Latin America, Australia and New Zealand, Africa, and the Middle East. A prevalence of HC of 2%-6% is estimated in the population in endemic areas<sup>[2]</sup>.

The human being is an intermediate host, as it is infected orally with food contaminated by eggs from the feces of the definitive host. The eggs hatch in the intestine, releasing oncospheres that penetrate the intestinal wall and invade different sites *via* the bloodstream, the most frequent being the liver (50%-70%) and the lungs (20%-30%). The oncospheres evolve into HC, which may remain asymptomatic until their growth causes complications derived from the mass effect, depending on their location. Rupture of the HC can spread its contents throughout the body and trigger anaphylactic shock<sup>[3]</sup>.

The duodenum is an organ that is very rarely affected by HC, either primarily or secondarily. Possible forms of secondary duodenal involvement are compression, or, more frequently, fistulization from HC located in neighboring organs<sup>[4]</sup>. The HC that most often fistulize the duodenum are those located in the liver (0.15% of instances), with cases also described from the pancreas and kidney. Its low prevalence and the few existing records mean that the choice of the best therapeutic management is a challenge<sup>[4]</sup>.

Here we present a systematic literature review of cases of hydatidosis with duodenal involvement published in the scientific literature.

## MATERIALS AND METHODS

### Literature search

Following the guidelines of the PRISMA methodology for systematic reviews, a search was performed in the PubMed, SCielo and EMBASE databases<sup>[5]</sup>, unlimited by language or time. The terms used for the search were: (duodenum) AND [(echinococcosis, hepatic) OR (echinococcosis) OR (echinococcosis, hepatic) OR (hepatic echinococcosis) OR (hepatic echinococcosis) OR (echinococcosis, hepatic

alveolar) hepa (alveolar) hepato) OR (alveolar echinococcosis, hepatic) OR (echinococcosis, hepatic alveolar) OR (hepatic alveolar echinococcosis) OR (hepatic alveolar echinococcosis) OR (hydatidosis, hepatic) OR (hepatic hydatidosis) OR (hepatic hydatidosis) OR (hepatic hydatidosis) hepa (alveolar echinococcus, hepatic) OR (echinococcus, hepatic alveolar) OR (hepatic alveolarechinococcus) OR (hydatid cyst, hepatic) OR (cyst, hepatic hydatid) OR (cysts, hepatic hydatid) OR (hepatic hydatid cyst) OR (hepatic hydatid cyst) OR (hepatic hydatid ) OR (hydatid cysts, hepatic) OR (echinococcosis) OR (echinococcus infection) OR (echinococcus infections) OR (infection, echinococcus) OR (cystic echinococcosis) OR (cysticechinococcosis) OR (echinococcosis, cystic) OR (echinoco cystic) OR (hydatidosis) OR (hydatidosis) OR (cysts, hydatid) OR (cyst, hydatid) OR (hydatid cysts) OR (hydatid cyst) OR (hydatid disease) OR (hydatid diseases) OR (echinococcus granulosus infection) OR (echinococcus granulosus infections) OR (granulosus infection, echinococcus) OR (granulosus infections, echinococcus) OR (infection, echinococcusgranulosus) OR (infections, echinococcus granulosus)].

A case was defined as any patient diagnosed with primary duodenal hydatidosis or hydatidosis in any location with secondary duodenal involvement.

A total of 83 articles were obtained with the search strategy described, and all articles that included patients with the characteristics mentioned were selected. Case reports were not excluded. Fourteen articles were chosen, but only 12 could eventually be studied; the other two could not be accessed even though the corresponding publishers were contacted.

The available data on the patients were extracted and included in Tables 1 to 3, for later discussion. They were classified into three groups: Clinical data, including age, sex, main symptoms and signs and personal history; analytical and radiological data, including laboratory data and the main findings reported in the imaging and endoscopic tests performed; and intra and postoperative findings that include the surgeries performed and the complications detected in the postoperative course and follow-up.

## RESULTS

The twelve articles selected were all case reports, describing 14 cases. The countries where the 14 patients were treated were: Spain (6), Tunisia (4), India (2), France (1), and Turkey (1)<sup>[4,6-16]</sup>.

### Clinical data

Seven patients were men and seven women, with a mean age of  $53.14 \pm 17.65$  years (range: 28-78) (Table 1). Nine of the patients (64%) had abdominal pain (3/9 located in the right hypochondrium, 2/9 in the epigastrium and in the remaining 4/9 not specified). As for the rest of the clinical data, nine patients (64%) presented nausea and vomiting, two of them with hematemesis and one with hydatidemesis. Six patients (42%) had fever  $> 38^{\circ}\text{C}$ , three did not, and in five this information was not specified. Abdominal palpation was painful in ten cases (71%), and in seven patients masses were noted. Other symptoms and signs described included jaundice, melena, hydatidenteria, anorexia and weight loss. Three patients had previously undergone surgery for liver HC.

### Analytical, radiological and endoscopic studies

Analytical values were only available in nine patients (Table 2). In six of these (66%) leukocytosis was observed, in three neutrophilia, and in one eosinophilia. Blood analysis was only reported in five patients: In one it was found to be normal, and the remaining four presented low levels of hemoglobin and hematocrit. Serological results of hydatidosis (Casoni test or others) were provided in only six cases (42%: Four positive and two negative); in the other eight, no serological studies were mentioned.

As for radiological studies, only abdominal X-ray was reported in six patients (42%) and thorax in three (21%); in one case both tests were discussed. No simple radiology information was available in six cases (the last six published). Abdominal radiography found masses in four patients, two of which presented calcification. The chest radiograph revealed an over-elevation of the right hemidiaphragm in one patient, the presence of intralesional gas in two, calcification of the wall in one. Abdominal ultrasound was reported in only four patients (28%), three of whom presented cystic lesions.

Computed tomography (CT) was performed in ten patients (71%) (in two of them

Table 1 Clinical data

Ref.	Sex	Age (yr)	Abdominal pain	Nausea and vomiting	Fever	Abdominal exploration	Past Medical history	Other signs and symptoms
Gilmas Y Moco-roa <sup>[16]</sup> , 1963	Male	32	No	Yes (hematemesis)	38.5-39°C	Six scars from previous surgeries. Slight collateral circulation. Soft splenomegaly with increased percussion. Epigastric pain on palpation	Dyspeptic vagal syndrome. Heartburn. Operated six times for hydatidosis	Melena. Daytime sleepiness and nocturnal delirium. Signs of hypoventilation and hypophonesis of right thorax
Perrotin <i>et al</i> <sup>[15]</sup> , 1978	Male	37	Yes (RH)	Yes	38-39°C	Guarding in the right hypochondrium. Right subcostal mass poorly limited and painful	NA	
Cosme <i>et al</i> <sup>[13]</sup> , 1987	Male	55	Yes (E)	NA	38°C	Painful abdominal mass (12 cm × 15 cm) located in the epigastric region	Pleural effusion, recurrent episodes of bronchitis	Asthenia, anorexia and weight loss of 3 kg
Robbana <i>et al</i> <sup>[12]</sup> , 1991	Female	64	NA	Yes (hematemesis)	No	Tender mass in right hypochon-drium	NA	Dysuria, diarrhea
Noguera <i>et al</i> <sup>[11]</sup> , 1993	Female	60	Yes	NA	NA	Large tender epigastric mass	NA	Weakness, anorexia and joint pain
Thomas <i>et al</i> <sup>[14]</sup> , 1993	Male	31	Yes	Yes (hydatidemeses)	39°C	Ill-defined mass in the LH. Non-tender mass in the left iliac fossa. Smooth and firm hepatomegaly (6 cm). Rectal examination: Soft cystic swelling in the rectovesical pouch	Fully excised intraabdominal cysts at age 9 and 21 years	Hydatidenteria
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	Female	68	Yes (E)	Yes	NA	A tender mass in the epigastrium and right hypochon-drium	NA	
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	Female	84	Yes	Yes	Yes	Tenderness and muscle guarding on the right side and a palpable mass	NA	
Patankar <i>et al</i> <sup>[10]</sup> , 1998	Male	35	Yes (RH)	No	No	Non-tender hepatomegaly	NA	
Muinel-lo Lorenzo <i>et al</i> <sup>[8]</sup> , 2012	Male	78	No	No	NA	NA	Osteoarthritis and benign prostatic hypertrophy. Laparoscopy cholecystectomy	
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	Female	28	Yes (RH)	Yes	NA	Tenderness in right hypochon-drium and right lumbar fossa with lumbar contact	Liver hydatid surgery and recurrent hydatid cysts 21 and 17 years previously	
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	Female	63	Yes (EH)	NA	38.6°C	Abdominal involuntary guarding in the right hypochon-drium	NA	Anaphylactic reaction with diarrhea Chills and jaundice: Bilirubin: 10 mg/dl
Jarrar <i>et al</i> <sup>[7]</sup> , 2015	Male	63	NA	Yes	NA	No palpable abdominal mass. Gastric distension	NA	Sensation of weight in RH
Akbulut <i>et al</i> <sup>[6]</sup> , 2018	Female	46	No	Yes	No	Painful epigastric palpation	NA	Intra-abdominal aggressive fibromatosis

E: Epigastric; RH: Right hypochondrium; LH: Left hypochondrium; NA: Not available

using contrast). In all of them cystic lesions were discovered (two with calcification) a

Table 2 Analytical, radiological and endoscopic studies

Ref.	Leukocytosis	Red blood cell count	Hydatid serology	X-ray	Ultrasound	CT	Contrast studies	Endoscopy
Gilmas Y Moco-roa <sup>[16]</sup> , 1963	16000 (84% neutrophils)	Erythrocytes: 3.286.000; Ht: 30% ESR: Katz Index 109	Casoni's test: Positive	Chest: Right hemidia-phragm raised	NA	NA	Barium study: Stomach not filled due to huge cysts compressing the stomach body	NA
Perrotin <i>et al</i> <sup>[15]</sup> , 1978	14300/ $\mu$ L	NA	Positive	Abdominal: No findings	NA	NA	Barium study: Heterogeneous opacity from the first portion of the duodenum, rounded cavity of 10 cm in the liver Cholangiography: Slightly dilated bile duct without signs of obstruction	NA
Cosme <i>et al</i> <sup>[13]</sup> , 1987	12200/ $\mu$ L	Ht: 34%	NA	Abdominal: Supramesocolic mass of 15 cm deforming the stomach	Inconclusive	Cystic lesion with air-fluid level within the pancreas	Barium study: Fistula in the duodenum filling a pancreatic cyst with gas bubbles	Fistulous opening of 5 cm in diameter, in the duodenal bulb
Robbana <i>et al</i> <sup>[12]</sup> , 1991	No	Hgb 11.1 g/dL	Positive	Abdominal: 10 cm right mass by L2-L3	14 cm cyst in right hypochon-drium	NA	X-ray with iodinated intravenous contrast: Right kidney delayed excretion and tumoral syndrome in lower lobe. Barium swallow test: Static stomach, megaduodenum with fistulae to mass	Esophagitis type 1-2, chronic erosive gastritis. No access to duodenum
Noguera <i>et al</i> <sup>[11]</sup> , 1993	NA	NA	NA	Chest and abdominal: Atypical gas bubble in the epigastric region with peripheral calcifications	NA	Cavity in the left hepatic lobe with partially calcified walls with communication with the duodenum. Another cystic multiloculated lesion in the peritoneal cavity lateral to the hepatic flexure of the colon	Iodinated oral contrast passing into the hepatic cavity confirmed the presence of a fistulous communication between the duodenal bulb and the cystic cavity	NA
Thomas <i>et al</i> <sup>[14]</sup> , 1993	No	Hgb 11 g/dL	Casoni's test: Strongly positive	NA	Three cysts: Left and right hypochon-drium, pelvis	Multiple intra-abdominal hydatid cysts. One large cyst in the left hypochon-drium communicating with the stomach and the second part of the duodenum	NA	NA
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	No	Normal	Negative	Abdominal: Calcified circular line in the upper abdomen	NA	Calcified cystic mass in the left hepatic lobe and in continuity with the digestive tube	Barium swallow study: Cavitated mass communicating with the duodenum near the pylorus	Fistula in the pyloric region with features of an echinococcal cyst
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	18000/ $\mu$ L (85% neutrophils)	NA	NA	Abdominal: Calcified mass in right hypochon-drium	NA	Pneumoperitoneum and a liver cyst with an air fluid level	Barium swallow X-ray showed the presence of a cyst opening into de first duodenal knee	Cyst opening into duodenum
Patankar <i>et al</i> <sup>[10]</sup> , 1998	NA	NA	NA	Chest: Crescentic gas shadow under the right dome of the diaphragm	NA	TC Dynamic contrast: Two cystic lesions in right liver lobe. Air in one of the cysts, tracking to the region of the first part of the duodenum. Another multiseptated cyst between urinary bladder and the rectum. CT oral contrast		NA



						showed a megaduodenum and a fistula to the mass		
Muinelo Lorenzo <i>et al</i> <sup>[8]</sup> , 2012	NA	NA	NA	NA	NA	3.5 cm hepatic hydatid cyst in segment IV	NA	Fistulous communication with calcified liver mass
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	11300/μL (18% eosinophils)	NA	NA	NA	NA	One multilocular hydatid cyst in the posterior part of the lateral sector of the right lobe of the liver, extended into retroperitoneum (until right kidney). Second hydatid cyst in segments I and V of the liver compressing the duodenum with a distended stomach	Barium swallow X-ray: Opacification of the hydatid cavity through a duodenal fistula near the pylorus. Preoperative cholangiogram: Retrograde opacification of the cyst through the duodenal fistula	NA
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	13700/μL (92% neutrophils)	NA	NA	NA	Two multivesicular hydatid cysts in segments IV and VI of the liver (5 and 6 cm respectively)	NA	NA	NA
Jarrar <i>et al</i> <sup>[7]</sup> , 2015	NA	NA	NA	NA	NA	Upper gastrointestinal stenosis due to a hydatid cyst located in segment VI of the liver attached to the duodenum compressing it extrinsically	NA	Gastric stasis due to extrinsic compression of the second portion of the duodenum
Akbulut <i>et al</i> <sup>[6]</sup> , 2018	NA	NA	Negative (postoperative)	NA	NA	CT scan with contrast 100 mm x 80 mm lesion originated by the body of pancreas		NA

CT: Computed tomography; NA: Not available; HT: Hematocryte; HGB: Hemoglobin

The location of the HC causing the duodenal fistula, confirmed by imaging techniques, was the liver in 11 cases (78%), and kidney, pancreas and paraduodenal area in one case each.

### ***Intraoperative findings, postoperative period and follow-up***

Twelve patients presented fistula between the HC and the duodenum (Table 3). One patient presented a paraduodenal HC diagnosed incidentally during surgery for abdominal fibromatosis and another had large-scale duodenal compression due to a liver HC, without a true fistula.

Twelve patients (85%) received surgical treatment. Of the other two, one received only pharmacological medical treatment with antiparasitic drugs and in the other the treatment was not specified. The type of laparotomy was reported in seven patients: Three midline, two bilateral subcostal, and two right; in one, the type was not specified.

The techniques performed to treat HC in the twelve patients operated on were conservative in nine [partial cyst resection (3), enucleation (2), evacuation/drainage (2), Lagrot (1) and ablation (1)] and radical in three (all total cystectomies). The procedures performed on the duodenal fistula in the ten patients with this condition

Table 3 Intraoperative findings, postoperative period and follow-up

Ref.	Intraoperative findings	Surgical procedure	Postoperative period morbidity	Follow-up
Gilmas Y Moco-roa <sup>[16]</sup> , 1963	Multiple adhesences from past surgeries. Stomach filled with blood clots. Multiple cysts (> 10) across peritoneum, liver, and spleen; duodenal fistula connecting with 15 cm cavity in left hepatic lobe	Ablation of cysts. Gastrostomy. Pylorotomy. Suture of duodenal fistula with surgical drains placed in cavity	Evisceration with massive hemorrhage. Postoperative death	NA
Perrotin <i>et al</i> <sup>[15]</sup> , 1978	Adherences are found in the right hepatic lobe. Visualization of liver mass with purulent liquid and food remains. Intraoperative cholangiography: Communication of 1-1.5 cm diameter between the cyst and the duodenum	Fistula closed, cyst drained with a gastric aspiration probe and placement of a cholecystostomy	NA	Day 3: Probe is removed The drainage of the cavity after being washed with lactic acid is removed after 18 d. Control cholangiography and duodenal transit are normal. Follow up in clinics
Cosme <i>et al</i> <sup>[13]</sup> , 1987	Infected and multivesicular hydatid cyst in the head of the pancreas closely attached to and communicating with the duodenum	Partial removal of the cyst with two catheters inserted into the cavity	NA	8th week: Injection of contrast through the drainage tubes demonstrating progressive closure of the remaining cavity. Asymptomatic 4 mo after surgery
Robbana <i>et al</i> <sup>[12]</sup> , 1991	Calcified hydatid cyst in anterior kidney area. Fistula connected the kidney mass to the duodenum	Evacuation, intralaminar pericystectomy, and reduction of fistula. Vagotomy	NA	Discharged on 17 <sup>th</sup> postoperative day. 7-mo postoperative ultrasound and urography were normal
Noguera <i>et al</i> <sup>[11]</sup> , 1993	NA	Enucleation	NA	NA
Thomas <i>et al</i> <sup>[14]</sup> , 1993	NA	Medical treatment: Albendazole, Ciprofloxacin, Crystalline penicillin and Chloroquine + US guided aspiration	NA	NA
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	Segment IV of the liver a 5 cm diameter mass adherent to the pylorus	Total resection of the cyst, including a piece of the duodenal wall, a Heinecke Mikulicz pyloroplasty, cholecystectomy and truncal bilateral vagotomy	Postoperative course was uneventful	Discharged on the 7 <sup>th</sup> day
Diez Valladares <i>et al</i> <sup>[9]</sup> , 1998	15 cm multiloculated hydatid cyst in right hepatic lobe in contact with the duodenum. Two hydatid cysts in the greater omentum with purulent fluid	Total cystectomy with resection of the duodenal sinus, excision of the omental cyst. Closure of the bile fistula and bile drainage	NA	Discharged on 15 <sup>th</sup> day
Patankar <i>et al</i> <sup>[10]</sup> , 1998	NA	Enucleation	NA	NA
Muinelo Lorenzo <i>et al</i> <sup>[8]</sup> , 2012	NA	NA	NA	NA
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	Single multilobular hydatid cyst in the posterior part of the lateral sector of the right lobe in close contact with the duodenum. Duodenal fistula affecting the posterior wall of the duodenal knee	Cholecystectomy. Large resection of the prominent cystic dome. Duodenostomy associated with gastrostomy and jejunostomy to treat duodenal fistula	NA	After 6 wk the patient was discharged
Daldoul <i>et al</i> <sup>[4]</sup> , 2013	Two liver cysts: Segment V (5 cm) and in the underside of the segment IV (8 cm) in close contact with the first duodenum knee. Exploration of the cystic cavity: Wide communication with the first duodenum and a large fistula with the confluence of the hepatic biliary ducts	Resection of the dome of the 5 cm cyst, duodenal diverticulization and external drainage	NA	Discharged after 3 wk
Jarrar <i>et al</i> <sup>[7]</sup> , 2015	Multivesicular hydatid cyst measuring 6 cm, at the right lateral sector, with extraparenchymal development, adhering to the duodenum. Exo-	Lagrot's procedure, a partial cystectomy and epiploplasty filling the residual cavity	Postoperative course was uneventful	NA

	vesiculation of 2 cm, communicating with the cyst, compressing the duodenal wall without fistula		
Akbulut <i>et al</i> <sup>[6]</sup> , 2018	The diameter of the fibromatous mass was 120 mm × 100 mm, originated in the pancreatic body and creating adherences to adjacent tissues forming a conglomerate with the fourth portion of the duodenum, jejunal loops and prepyloric stomach antrum	The fourth portion of the duodenum, the jejunum, the distal pancreas and the spleen were removed en bloc. Anastomosis between the third part of the duodenum and the proximal jejunum	Postoperative course was NA uneventful

NA: Not available; US: Ultrasound.

operated upon were: (1) Closure of the duodenal fistula in five patients, in one case associated with vagotomy, and in another with gastrostomy plus pylorotomy; (2) Partial duodenal resection plus pyloroplasty and bilateral truncated vagotomy (one patient); (3) Duodenostomy, gastrostomy and jejunostomy (one patient); (4) Duodenal diverticulization (one patient); (5) Not specified (one patient); and (6) Intracavitary drainage without action on the fistula (one patient).

Postoperative morbidity and mortality was not described in detail in the articles consulted. In three patients the postoperative period was reported to be uncomplicated, and one patient (7%) died due to hemoperitoneum following massive hemorrhage. In the five cases in which mean hospital admission was mentioned, it was  $20.4 \pm 11.72$  d (range: 7-42). The follow-up was not described in detail, though it was usually very short.

## DISCUSSION

Hydatidosis is a zoonosis caused by *Echinococcus spp* associated with livestock activity, predominantly in geographical areas with temperate climates such as Southern Europe, Latin America, Australia, New Zealand, Africa and the Middle East<sup>[2]</sup>. Primary or secondary duodenal involvement due to HC *via* fistulization or compression from other locations is extremely rare and the number of reported cases is very low<sup>[4,6-16]</sup>. The main objective of our study was to evaluate the clinical, diagnostic and therapeutic data of duodenal involvement recorded in the literature and to assess whether a diagnostic and therapeutic algorithm could be obtained.

The articles included do not describe any etiological factors or pathogenesis related to duodenal injury caused by HC. HC located in segments IVB, V and VI, and those located in the kidney or pancreas may affect the duodenum, but this is exceptional. Our (unprovable) opinion regarding this low frequency is that a series of conditions must exist for a fistula to appear: The cyst must be large, complicated, and long-standing, since the duodenal wall is sufficiently thick to allow compression without fistulization. If these conditions are met, after very prolonged inflammatory processes the fistula eventually forms.

The epidemiological data of the 14 patients studied do not indicate any differentiation according to sex (the ratio was 1:1) or age (the range was large). Duodenal injury caused by HC is mostly secondary. There was only one case of a paraduodenal cyst; in the cases reported the duodenum was secondarily affected by hepatic (11/13: 85%), renal (1/13) and pancreatic (1/13) HC<sup>[4,6-16]</sup>. In 90% of patients the injury is in the form of a fistula between the HC and the duodenum. The descriptions in the articles do not indicate which duodenal segment is the most affected. Three patients had previously undergone liver hydatidosis.

The most frequent clinical manifestations in the patients studied were abdominal pain, nausea and vomiting. The rest of the clinical data were very heterogeneous. Two cases presented hydatidemia and/or hydatidenteria<sup>[14]</sup>, but the condition may also be asymptomatic. Therefore, there is no typical clinical picture of HC affecting the duodenum.

The diagnosis of duodenal fistula due to HC is not always easy. Computed tomography was the most used diagnostic tool in the patients reviewed; it allows accurate location of the HC and its relationship with the digestive tract, but in our study only achieved preoperative diagnosis of fistula in four patients (40%). It should be noted that some reports do not specify whether oral contrast was used in the CT, and some of the older cases the CT techniques used are not comparable to the ones used today<sup>[4,6-16]</sup>. The use of various radiological tests with contrast, especially in the first cases, allowed the diagnosis of fistula, although barium contrast studies are not currently used. Upper gastrointestinal endoscopy was used in six patients with a diagnostic capacity of 66% (4/6)<sup>[4,6-16]</sup>. Therefore, we are unable to propose a definitive diagnostic algorithm, but the combination of CT with contrast and upper gastrointestinal endoscopy seems to be the most effective.

In the literature, there is no consensus on which treatment that should be applied. Surgery was performed in practically all patients (12/13), using a wide variety of procedures to treat both HC and duodenal fistula, adapting to intraoperative findings. Conservative techniques were used in 75% of HC, while in the duodenum the most frequent technique (50%) was the closure of the fistula associated with various types of intestinal bypass; partial duodenal resection was performed in only one case. The operative morbidity rate was 7% (1/14), and postoperative mortality 7% (1/14). We believe that the estimation of morbidity is excessively low, because in many articles it is not reported or it is simply stated that the postoperative period was uneventful; in addition, duodenal surgery usually presents a much higher complication rate than that reported in these patients. Furthermore, in the cases in which data on postoperative stays are given, these stays are much longer than one would expect in uncomplicated patients. The follow-up carried out in patients is not usually specified, so we cannot draw any conclusions about disease recurrence or long-term evolution.

Our intention in carrying out this systematic review was to provide a therapeutic algorithm for hydatidosis that affects the duodenum, but especially the surgical strategy to be performed. As we have summarized, no conclusion can be drawn from the literature. Our group thinks that radical surgery is the best approach in hydatidosis, but we also believe that surgical procedures should be tailored to every patient. Our non Evidence Based Medicine supported recommendation is: If the surgeon thinks that radical surgery is technically feasible, should be performed in the liver and duodenum. But if an unacceptable risk is to be assumed to carry it out, conservative procedures can be useful in the most bizarre cases. Duodenal surgery is not simple, nor is it free of complications, and techniques can range from a simple closure, duodenal exclusion to duodenectomies. The decision of which technique to perform in the duodenum will depend on the amount of circumference affected, the duodenal segment to be treated, and anatomical relationship with the Vater's ampulla.

The following conclusions can be drawn from this systematic review. The reports of duodenal damage by HC are based on isolated clinical cases with very low scientific evidence; the most common clinical findings are abdominal pain, nausea and vomiting; the combination of computed tomography plus endoscopy seems to be the best diagnostic option; finally, the most frequently recommended treatment is surgery to treat the HC and to close the fistula.

## ARTICLE HIGHLIGHTS

### Research background

The duodenum is an organ that is very rarely affected by hydatid cysts, either primarily or secondarily.

### Research motivation

Duodenal involvement by hydatidosis is an exceptional finding and this study aimed to characterize epidemiological, clinical and diagnostic features and options of treatments.

### Research objectives

Our aim is to characterize epidemiological, clinical and pathological features and options of treatment in hydatidosis with duodenal involvement.

### Research methods

We perform a system review including all patients with hydatidosis and duodenal involvement following PRISMA guidelines.

### Research results

This review shows that the most frequent symptoms were abdominal pain, nausea and vomiting. Computed tomography was the most used diagnostic imaging technique, and hydatid cyst drainage and fistula closure *via* laparotomy was the most frequent treatment.

### Research conclusions

Literature on hydatidosis with duodenal involvement is scarce, only case reports, so the evidence is low.

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