Case report

A rare revelation mode of hepatic hydatid cyst: cysto-gastric fistula. A Case Report and Literature Review.

Abstract :

Hydatid disease is a zoonosis caused by *Echinococcus granulosus*, most commonly affecting the liver. While biliary rupture represents the most frequent complication, direct rupture into the stomach is an exceptional occurrence. Cysto-gastric fistula constitutes a rare and unusual presentation, often discovered intraoperatively or on advanced imaging. This review synthesizes recently reported cases and radiological reviews to analyze the mechanisms, diagnostic features, and surgical management of hepatic hydatid cysts complicated by cysto-gastric communication.. Computed tomography with oral contrast remains the cornerstone of diagnosis. Radical surgical excision with gastric repair and postoperative albendazole therapy yields excellent outcomes. Recognition of this rare complication is critical for timely diagnosis and definitive treatment.

Keywords : cysto-gastric fistula, Hydatid disease, hepatic hydatid cyst, laparotomy, resection of the protruding dome, Lagrot procedure.

INTRODUCTION

Cystic echinococcosis is a parasitic infestation caused by the larval stage of *Echinococcus granulosus*, a tapeworm prevalent in Mediterranean, Middle Eastern, and South American regions. The liver is involved in approximately 70–80% of human infections. [1,2]

Although hydatid cysts may remain silent for years, they can enlarge, become infected, or rupture into adjacent structures. The commonest form of rupture occurs into the biliary tree; however, communication with the gastrointestinal (GI) tract, particularly the stomach, is exceedingly rare—reported in fewer than 0.5% of cases. [3]

Spontaneous cysto-gastric fistula represents an exceptional clinical entity first described decades ago and remains sparsely documented. Recent case reports from Spain, Turkey, and Morocco have renewed attention to this presentation. [3-5]

CASE REPORT

We report a 34-year-old patient, with no prior medical or surgical history, who presented to the emergency department with a month history of epigastric pain treated with proton-pump inhibitors with no improvement though.

Four days prior to his consultation, the patient reported a worsening of his symptoms manifesting in the expulsion of white membranes in a vomiting effort and fever.

A detailed anamnesis reported no sign of intestinal transit disorders, no hematemesis or melena, normally colored urine and stools and no respiratory symptoms either such as chronic cough, chest pain or shortness of breath.

Physical examination found a conscious patient, hemodynamically stable, febrile at 38°C and a conjunctival jaundice. Abdominal examination found a distended abdomen, epigastric

and right hypochondrium tenderness with a palpable mass in this region. The rest of the physical examination was normal.

Laboratory analysis found a high white blood cell count at 21850/mm^3 increased at the expense of the neutrophils which are at 15860/mm^3. The hepatic biochemical panel was slightly abnormal, ALP-AMP at 205,18 U/L (0-115), g-GT at 127,41 U/L (0-55), GPT at 72,4 U/L (0-40), GOT at 96,72 U/L (0-40), bilirubin direct at 7,04 mg/L (0-4) and bilirubin total at 11,93 mg/L (0-10).

An abdominal ultrasound was first performed revealing a lesion that presents as a heterogeneous isoechoic tissue echotexture containing multiple anechoic fluid pockets, associated with some vascular spots on the color doppler ultrasound. It is located beneath the left hepatic lobe displacing it superiorly. It measures approximatively 170*115mm. The gallbladder contains partially heterogeneous fluid.

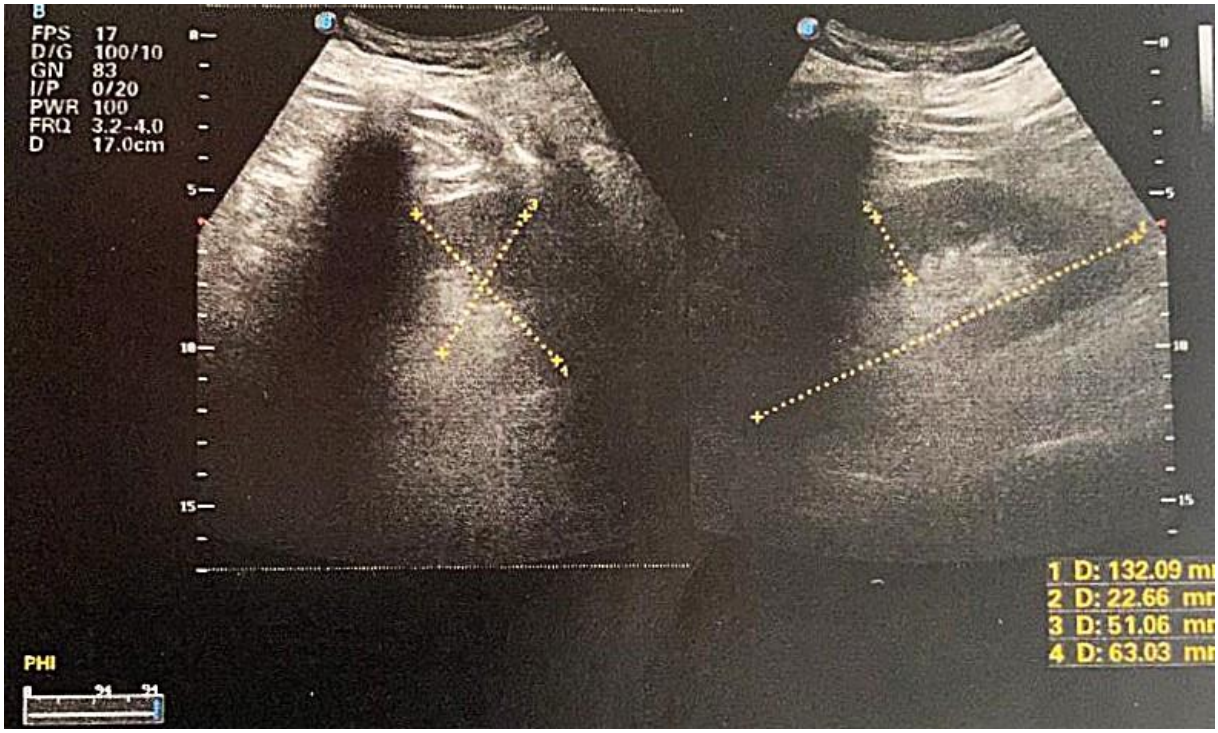

Figure 1 : Abdominal ultrasound showing an epigastric tumoral process: possible gastric GIST, and Gallbladder sludge.

We completed by an abdomen CT-scan that showed a large cystic mass with a thickened wall containing a hydro-aeric level occupying the entire left lobe of the liver and measuring 81*129*184 mm. Medially this lesion reaches the left diaphragm dome with a persistent thin layer of fat separating it. It comes into close contact with the stomach with loss of the fat plane. The lesion demonstrates a very thin and discontinuous wall in its superior-internal portion with extra-parenchymal bulging into the peritoneum, without localized or distant peritoneal effusion. Laterally, the collection shows a fistulous tract in its internal portion establishing communication with the intrahepatic bile ducts with presence of aerobilia. Inferiorly, it closely abuts the duodenal bulb with a fistulous tract communicating with the duodenal lumen. In total, this mass was compatible with a hepatic hydatid cyst fistulized into the intrahepatic bile ducts and into the gastric and duodenal lumen.

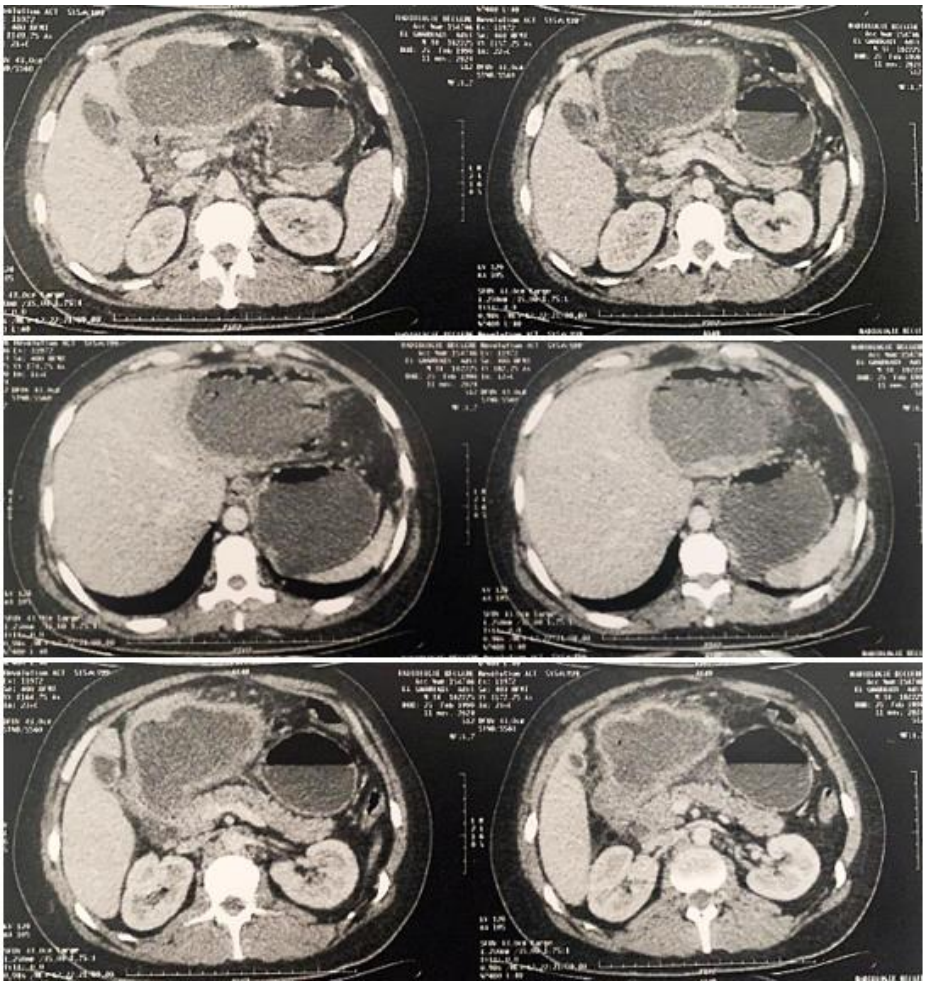

Figure 2 : CT scan axial view showing large superinfected cystic lesion of the left hepatic lobe, fistulised into the intrahepatic bile ducts (with aerobilia) and into the duodenal bulb, with marked thinning of its inner wall and peritoneal bulging, without intra-peritoneal collection or free fluid. Pre-rupture stage.

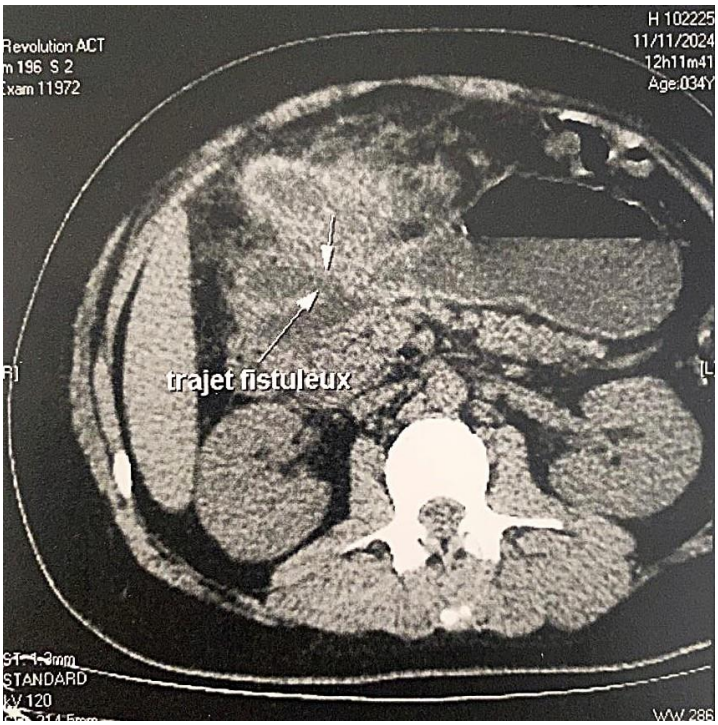

Figure 3 : Ct-scan axial view showing a cysto-gastric fistula.

The patient was taken to the operating theatre and underwent an exploratory laparotomy. After gastric decompression via a nasogastric tube, a midline laparotomy incision extending across the umbilicus was performed.

Exploration of the abdominal cavity revealed a gigantic hydatid cyst involving the entire left lobe of the liver, closely adherent to the lesser curvature of the stomach extending to the duodenal bulb. Resection of the protruding dome was performed after protecting the abdominal cavity with fields soaked in diluted hydrogen peroxide solution, and the hydatid material was aspirated.

The inspection of the residual cavity has indeed revealed the presence of two cysto-digestive fistulas. The first was a cysto-gastric fistula, and the second was a cysto-bulbar fistula.

The surgical therapeutic approach was a resection of the protruding dome (or Lagrot procedure) and to preserve the residual cavity without resecting the wall of the hydatid cyst adherent to the stomach and the duodenal bulb and perform a blind closure of the fistulous

tracts through the residual cavity using sutures with slowly absorbable threads. Finally, a surgical drain was placed in the residual cavity and another one in the subhepatic area.

A Fundus-first cholecystectomy was also performed.

The patient additionally received adjuvant medical therapy for hydatid disease with albendazole (400 mg twice daily for three months).

The patient was discharged on postoperative day ten with satisfying results, and showed no complications in the two-week and three month follow ups.

DISCUSSION

Hydatid disease remains endemic in several regions of the world, including the Mediterranean basin, the Middle East, and parts of Africa and South America. Despite advances in prevention and imaging, it continues to pose a surgical challenge. The liver, being the first filter for the portal venous system, is the most common site, with left-lobe involvement observed in approximately 20–25% of hepatic cases. [1,2]

Several authors, including Onka *et al.* (2021) and Dogukan *et al.* (2023), identified two key mechanisms: (1) infection-induced adhesion and inflammatory thickening of adjacent cyst and gastric walls, and (2) progressive pressure necrosis due to cyst expansion. [3,5]

The cyst gradually erodes through the hepatic capsule and the gastric wall, producing a direct tract, as confirmed intraoperatively in both reports. Ortiz de Guzmán *et al.* (2022) observed a similar process in a patient with a large left-lobe cyst compressing and fistulizing into the gastric corpus. [4]

The rarity of this complication may be attributed to the stomach’s thick muscular wall, which generally resists penetration , howver any left-lobe hydatid cyst exhibiting air within the cavity should prompt suspicion of a gastric fistula , as the left lobe’s anatomical proximity to the stomach explains the occasional occurrence of cysto-gastric communication. [3,5]

The clinical manifestations are nonspecific and may mimic more common complications such as cyst infection or biliary communication. Abdominal pain, fever, and vomiting predominate. A pathognomonic but infrequent symptom is the vomiting of hydatid membranes (*hydatidemesis*), described by Onka *et al.* (2021). Dogukan *et al.* (2023) reported right upper-quadrant pain and fever without jaundice, while Ortiz de Guzmán *et al.* (2022) described intermittent febrile episodes and dyspeptic symptoms. The absence of jaundice distinguishes cysto-gastric communication from the more common cysto-biliary rupture. [3-6]

Radiological imaging is essential for diagnosis. Ultrasonography often shows a multiseptated lesion (Gharbi type III) with irregular walls and internal debris. Computed tomography (CT) remains the gold standard, demonstrating a hypodense lesion with air-fluid levels and, in characteristic cases, passage of oral contrast from the gastric lumen into the cyst cavity . [3,5]

Magnetic resonance imaging (MRI) and MRCP may be used to exclude concomitant biliary communication. Upper endoscopy, when performed, can reveal a mucosal defect or protrusion of cyst membranes into the stomach, as noted by Dogukan *et al.* (2023). The presence of intralesional gas in an unoperated cyst should always raise suspicion of either infection or a fistulous tract. [1,2]

In the case series by Shaikh *et al.* (2021), a hepatic hydatid cyst with a cystoduodenal fistula was identified using CT and confirmed endoscopically, reinforcing the value of multimodal imaging in detecting rare enteric fistulas. [6]

The reviewed literature underscores several diagnostic and management lessons , however surgical management remains the definitive treatment for cysto-gastric fistula. The principal objective is complete cyst excision, closure of the gastric defect, and prevention of peritoneal contamination. Reported procedures include partial or total pericystectomy with direct two-layer gastric repair, sometimes reinforced by an omental patch. Dogukan *et al.* (2023) reported successful management through partial pericystectomy and primary closure of an 8 mm gastric orifice, while Ortiz de Guzmán *et al.* (2022) described a complete cystopericystectomy with closure of the gastric tract, both followed by uneventful recovery.

Intraoperative methylene blue or propofol injection tests can help identify occult cystobiliary communications, as recommended by Shaikh *et al.* (2021). [3-6]

Postoperative anthelminthic therapy with albendazole (10–15 mg/kg/day for 3 months) remains standard to prevent recurrence. In all reviewed reports, no postoperative complications or recurrences were observed at follow-up. [4-6]

REFERENCES

1. *Taheri MS, Pourghorban R, et al. Not-to-tap cystic lesions: spectrum of hydatid disease. Emergency Radiology. 2013;20:553-561.*
2. *Amin MU, Mahmood R, Shafique M, et al. Imaging features of unusual patterns and complications of hydatid disease. Journal of Radiology Case Reports. 2009;3(12):1-24.*
3. *Onka B, Benmoula FZ, Traoré WYM, Mohamed DA, et al. Spontaneous gastric fistula from a hydatid cyst of the liver. BJR Case Reports. 2021;7:20210087.*
4. *Ortiz de Guzmán Aragón S, Perfecto Valero A, Ventoso Castiñeira A, Valdivieso López A. Hydatid cyst with gastric fistula. Cirugía Española (English Edition). 2022;100(12):786-788.*
5. *Doğrul A, Dinçer HA, Çiftci TT, Akıncı D, Parlak E, Doğrul AB. Diagnosis and surgical treatment of cysto-gastric fistula out of an hepatic hydatid cyst: Unusual case. Annali Italiani di Chirurgia. 2023 (May 8 E-pub).*
6. *Shaikh O, Gaur NK, Vijayakumar C, Kumbhar U, Kalayarasan R. Hepatic hydatid cyst with cystobiliary communication and cystoduodenal fistula. Cureus. 2021; 13(8): e17473.*