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(CASE REPORT)

Hydatid cyst of the liver: When fistulization reaches the portal trunk

Oumaima El Abd [1, 2, *], Soukaina Allali [1, 2], Mohamed Idrissi [1, 2], Brahim Boukatta [1, 2], Karim Ibn Majdoub [1, 2], Sidi Adil Ibrahimi [1, 2] and Hakima Abid [1, 2]

[1] *Department of Hepato-Gastroenterology, Hassan II University Hospital, Fez, Morocco.*
[2] *Faculty of Medicine, Pharmacy and Dental Medicine, Sidi Mohamed Ben Abdellah University, Fez, Morocco*

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Abstract

Hydatidosis is a major health problem in Morocco, mainly affecting rural regions due to traditional livestock breeding practices. Complications of hepatic hydatid cyst include fistulization into the biliary tract, with a rare complication of fistulization into the portal vein. We present the case of a 71-year-old patient with a hydatid cyst fistulized into the right branch of the portal vein. Diagnosis is based on biological and imaging examinations. Surgical management is complex due to the hemorrhagic risk. Clinical and radiological monitoring is sometimes preferred. The management of such situations remains a clinical challenge requiring multidisciplinary expertise to ensure optimal outcomes.

Keywords: Hydatid cyst; Liver; Fistulization; Portal trunk; Hepatectomy

1. Introduction

Hydatidosis, a parasitic zoonosis of worldwide scale, poses a major epidemiological challenge in Morocco. This disease, caused by the parasite *Echinococcus granulosus*, is frequent due to traditional livestock breeding practices and geographical proximity to endemic areas, mainly affecting Moroccan rural regions.

This disease can affect various organs of the body, but it has a particular predilection for the liver and lungs. Symptoms are not specific; it may be asymptomatic or manifest with pain, a sensation of heaviness, or complications, particularly superinfection, compression of neighboring structures, fistulization into the biliary tract, and exceptionally fistulization into the portal trunk.

We report here a clinical case of a hydatid cyst fistulized into the right branch of the portal vein.

2. Observation

A 71-year-old patient, followed for diabetes treated with oral antidiabetic drugs (OAD), arterial hypertension (HTA) treated with triple therapy, and pulmonary embolism (PE) treated with anticoagulants, presented with a clinical picture of angiocholitis, febrile jaundice with abdominal tenderness.

Biological parameters showed:

total bilirubin of 68 mol/L predominantly direct, cytolysis dependent on GPT, and cholestasis. The infectious workup was disturbed with a CRP of 199, and the prothrombin rate was 39%.

* Corresponding author: Oumaima El Abd

Imaging examinations (Fig 1) revealed a hydatid cyst of the liver (HCL) measuring 68 mm, dilation of the intrahepatic biliary tract (IHBT) suggesting cystic fistulization, subtotal obstruction of the right portal branch, as well as gallbladder abnormalities.

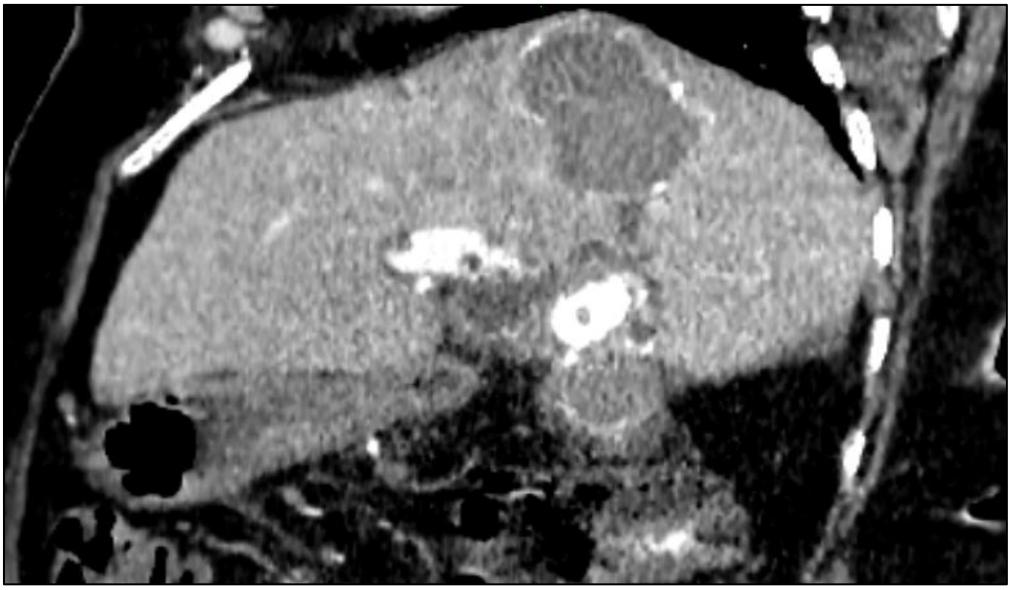

Figure 1 Initial oblique CT scan showing a 68-mm hepatic hydatid cyst fistulized into the right branch of the portal vein

An endoscopic retrograde cholangiopancreatography (ERCP) was performed, with extraction of hydatid membranes and placement of a 10 Fr plastic stent. A sphincterotomy was not performed due to the hemorrhagic risk with a low prothrombin level.

The evolution was marked by the onset of abdominal distension, with clinical examination showing diffuse dullness, which led to imaging (Fig 2), showing a liver of normal size, containing at the level of segment VIII an infected HCL fistulized into the IHBT.

- Partial opacification defect of the portal trunk extending to its right branch.
- Identification of a plastic stent in place at the level of the common bile duct.
- Distended gallbladder, thin-walled, without stones, containing an air bubble.

Large intraperitoneal effusion of liquid density associated with diffuse thickening and enhancement of the peritoneal layers.

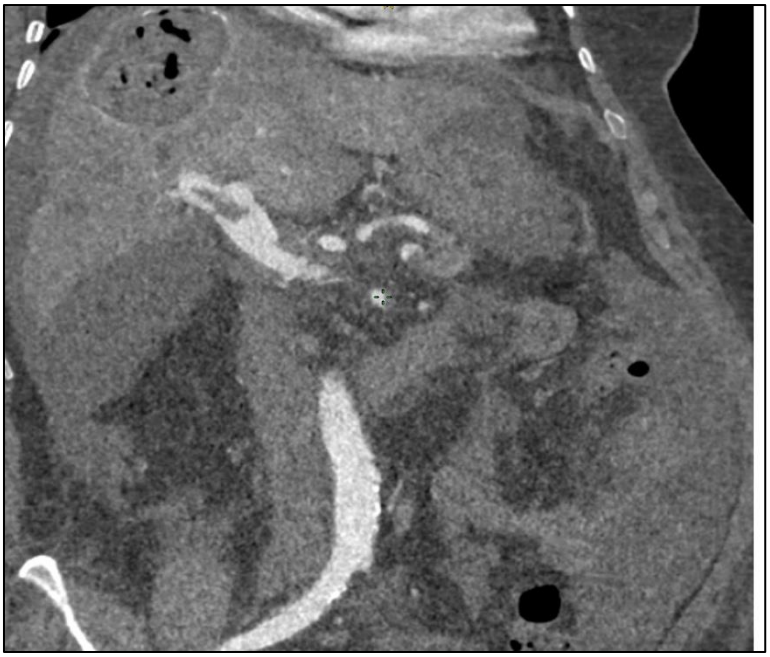

Figure 2 Coronal oblique CT scan showing an infected hepatic hydatid cyst located in segment VIII, fistulized into the intrahepatic bile ducts, with a partial opacification defect of the portal vein extending to its right branch

Ascitic fluid puncture revealed a protein level of 9 g/dl, with an albumin gradient of 25 in favor of portal hypertension. At the end of this assessment, our attitude was to respect the patient, since surgical drainage is at high hemorrhagic risk, as well as percutaneous drainage, and we adopted clinical and radiological monitoring until atrophy of the right liver and hypertrophy of the left liver, for the performance of a hepatectomy.

3. Discussion

Spontaneous rupture of a hepatic hydatid cyst into the portal vein or its branches is an unusual complication. To date, only 5 cases have been reported (Fig 3), all characterized by proximity of the cysts to the hepatic hilum.

This complication is one of the most serious associated with hepatic hydatid cysts, potentially leading to various problems such as portal hypertension (1) and anaphylactic shock due to the release of antigenic material from the cysts. Therefore, it is imperative to rapidly establish the diagnosis of cysto-vascular fistulas and treat this condition as a surgical emergency.

The imaging methods of choice for diagnosing hepatic hydatid cysts and their complications are abdominal ultrasound and computed tomography (CT). In case of rupture into the portal vein, color Doppler ultrasound may show venous thrombosis accompanied by signs of portal hypertension. However, CT with contrast injection is the method of choice to evaluate cysts and their relationship with surrounding structures, as well as to detect daughter vesicles

.Treatment of hydatid disease involves both surgical and medical therapeutic modalities aimed at treating both the hydatid cyst and portal hypertension. However, in some cases, surgery may be delicate due to increased risk of hemorrhage.

In the reported cases, 4 patients were operated on (2,4,5,7).

Two patients underwent hepatectomy, one right (4) and the other left (7), depending on the site of fistulization. The other two underwent cystectomy: one partial with evacuation of debris and closure of the portal vein defect (2), and the second cystectomy with ligation of the portal trunk and evacuation of vesicles from the portal branch (7).

In the fifth case, the decision was surgical abstention (3) and clinical monitoring.

Three cases of operated patients were complicated by hemorrhage during the procedure which was controlled. The evolution of the patients was characterized by the appearance of cavernomatous portal hypertension in 2 patients (5,7).

In our patient, our attitude is to plan a right hepatectomy with ligation of the portal trunk and the left branch of the portal trunk, then evacuation of the right branch, after a good preoperative evaluation, with initiation of medical treatment based on albendazole.

Table 1 Different cases reported in the literature of hepatic hydatid cyst fistulized into the portal vein and the different management approaches

	Nurset Ayurek et Al	**Leire Zubiaurre et Al**	**N Haoues et Al**	**Berkane et Al**	**Houssem et Al**	**Notre cas**
Year	2000	2006	2014	2020	2023	2023
Country	Turkey	Spain	Tunisia	Algeria	Tunisia	Morocco
Age	60	45	49	46	70	71
Sex	M	M	F	M	F	F
Radiology /location	SegV, VIII	Seg IV	SegVII	SegVII	SegV,VI,VII	SegVIII
Treatement	Abz + Cystectomy	Abz + Left hepatectomy	Abz	Abz+Cystectomy	Abz+Hepatectomie droite	Abz + Right hepatectomy
Outcome	Intraoperative hemorrhage / good	Cavernomatous PHT	Stationary	Intraoperative hemorrhage / PHT cavernomatous	Intraoperative hemorrhage / Good	-

4. Conclusion

It is essential not to underestimate vascular complications of hepatic hydatid cysts, even if they are rare. Cysts located near blood vessels require thorough examinations to detect any potential vascular communication.

In case of suspicion of fistulization, a multidisciplinary team must discuss management with great caution to avoid complications.

Managing such situations remains a major clinical challenge requiring expertise and meticulous planning for optimal outcomes.

Compliance with ethical standards

Disclosure of conflict of interest

The authors declare that they have no conflict of interest.

Statement of informed consent

Written informed consent was obtained from the patient for publication of this case report.

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