Portal invasion: An exceptional complication of hepatic hydatid disease

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Dear editor,

Cystic echinococcosis (CE) is a worldwide chronic parasitic zoonosis caused by Echinococcus granulosus. Humans act as accidental hosts as a result of ingesting the parasite’s eggs, which can be found in food contaminated by a definitive host’s feces (usually dogs). Larvae released from these eggs cross-intestinal mucosa and reach the liver through portal circulation. Occasionally, larvae can reach the right heart, and subsequently, the general circulation. Consequently, although CE mainly affects the liver, lungs or other organs may be affected [1]. However, portal invasion is an exceptional complication of CE.

A 69-year-old man presented with non-febrile epigastric pain, which appeared a few weeks after returning from Turkey. Clinical examination revealed epigastric pain without guarding. Initial laboratory tests showed elevated C-reactive protein (16 mg/L), hepatic cytolysis and cholestasis. Ultrasound revealed a heterogenous hepatic cyst at the junction of liver segments V and I, containing echoic areas and daughter anechoic cysts, associated with right portal branch compression. In accordance with the WHO classification, this cyst was classified CE3b [2]. Computed tomography (CT) examination obtained before and after intravenous administration of iodinated contrast material showed a 50-mm lesion of the liver with water attenuation values containing thin wall calcification, that compressed the right portal branch. Magnetic resonance imaging (MRI) confirmed these findings (Fig. 1). Positive Echinococcus serology confirmed the probability of a CE infection. Considering the portal compression, surgical treatment was suggested after 1 month of albendazole antiparasitic treatment. During surgery, the cyst was impossible to palpate, even after cholecystectomy. Intraoperative ultrasound confirmed the cyst location, close to the inferior vena cava and right portal branch, which was difficult to reach surgically. Consequently, it was decided to perform transhepatic PAIR (puncture, aspiration, injection and re-aspiration) treatment instead of a resection. An intracystic injection of 20% NaCl protoscolicide was successfully performed. The patient underwent 3 more weeks of albendazole antiparasitic treatment with uncomplicated postoperative recovery. The patient did not attend the third month planned consultation and was lost to follow-up. Six years later, the patient presented with abdominal pain. Enhanced-CT showed a 60-mm large cyst with internal clearly defined septa. Portal occlusion extended to both main portal vein and left portal branch with intraportal enhancing material, associated with portal cavernomatosis (Fig. 2). CT also showed moderate signs of portal hypertension, with esophageal varices and spleen enlargement. MRI then confirmed intraportal septa delimiting multiple daughter cysts. Review of initial MRI examination showed a slight cystic content into the occluded right portal branch, in favor of an

Figure 1. a–c: initial cyst imaging (a: enhanced axial CT image; b: non-contrast coronal CT image; c: T2-weighted axial image). Cystic lesion of the I–V hepatic segments junction (arrow head) with wall calcification (thin arrow) is associated with right portal branch compression (thick arrow).
initial intraportal cystic rupture (Fig. 3). Serological tests remained in favor of an evolutive echinococcosis. An albendazole antiparasitic treatment was started but the patient was lost to follow-up before the end of this treatment.

Complicated forms of CE involve one-third to approximately 60% of patients. Usual complications include superinfection, biliary communication and intrathoracic or abdominal rupture [3]. Portal involvement is an unusual CE complication, mostly represented by portal thrombosis. To date, only very few cases of portal invasion have been reported [4–6]. This complication could be easily mistaken for the more frequent biliary communication, considering that their CT appearances are very similar. However, in the case of biliary communication, MRI can show cyst-biliary connection and biliary tree dilatation that would not be present in intraportal invasion. Differential diagnoses may also include other biliary tract dilatation causes including lithiasic or tumoral origin as hilar cholangiocarcinoma or cystic biliary neoplasms as biliary tract papillary mucinous neoplasm, cystadenoma or cystadenocarcinoma. However, multiseptated cystic portal aspect with no biliary tract dilatation is an important finding in favor of portal inva-

![Figure 2](image_url)

**Figure 2.** a and b: six-year follow-up enhanced-CT (a: enhanced axial CT image; b: enhanced coronal CT image). Cystic lesion moderately increased, now multi-septated (arrowhead). Right portal branch occlusion extended to main portal vein and left portal branch (thin arrow). Portal occlusion is associated with cavernomatosis (thick arrow).

![Figure 3](image_url)

**Figure 3.** a–c: six-year follow-up MRI (a: axial T2-weighted image; b: axial fat-suppressed T2-weighted image; c: coronal MRCP); d: initial MRI axial fat-suppressed T2-weighted image. Portal cystic T2-weighted hypersignal combined with multiple T2-weighted hyposignal septa define portal invasion (thick arrow), confirmed by initial MRI rereading (d). MRCP does not reveal intra- or extrahepatic dilatation (thin arrow).
sion. History of hydatid liver cyst that happened several years before are in favor of a slow developing complication, as well as portal cavernomatosis, already described in portal CE invasion [4]. However, anaphylactic reaction has been reported in one patient [5], and even if portal hypertension signs were moderate in our case, more severe complications can be present like gastrointestinal bleeding secondary to the rupture of esophageal varices [6]. Surgery with pericystectomy and perioperative benzimidazole treatment is the first choice for complicated cysts. In hydatid intraportal invasion, isolated pericystectomy could not be intended alone as infected portal system would remain in place. Considering that portal invasion is very rare, few treatments and results have been reported to date with only one partial hepatectomy with a successful immediate outcome, but in this case portal invasion only involved left portal branch [5]. In an extended invasion, liver transplant could be considered but has not be reported so far. Medical antiparasitic therapy combined with variceal bleeding prophylaxis has also been proposed in one patient with successful outcome [6].

Disclosure of interest

The authors declare that they have no competing interest.

References


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