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Case Report

Atypical manifestation of a fistula between a hydatid cyst of a liver and a gallbladder: A case report ☆

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ABSTRACT

The hydatid cyst of the liver is the most common location of hydatid disease. Complications in this form are dominated by superinfection of the cyst and rupture into the bile ducts or the peritoneal cavity. We report the case of a 54-year-old female patient with a hydatid cyst of the liver complicated by a rupture in the gallbladder revealed by urinary symptoms.

Introduction

Hydatidosis or hydatid cyst is a parasitic disease caused by the accidental ingestion of dog's taenia egg by humans: echynoccocus granulosus. It is a disease that is mainly found in developing countries. Hepatic involvement is by far the most frequent and the evolution can be marked by the occurrence of complications. The aim of this paper is to report the case of complicated hydatid cyst of the liver, revealed by an atypical symptomatology followed by a review of the literature on this pathology.

Case report

A 54-year-old female patient with a history of renal lithiasis and recurrent nephritic colic without any specific medical follow-up. She was brought back to the emergency room by his family due to the onset of intense right lumbar pain radiating laterally to the abdomen and the groin suggesting an episode of nephritic colic. The initial assessment revealed a conscious patient with a heart rate of 105 beats/minute, blood pressure of 110/59 mm Hg, polypnea of 25 cycles/min, and saturation of 98% on room air. There was also a fever of 38.9°C, normal skin

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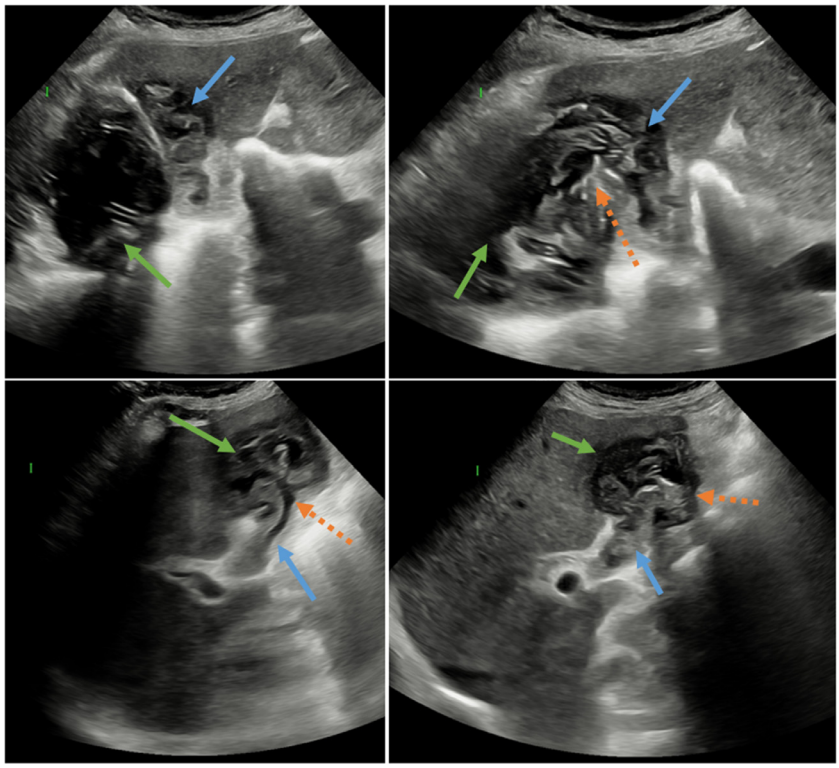

Fig. 1 – Abdominal ultrasound showing a cystic formation in segment VI of the liver (hydatid cyst: green arrow) with detached membrane, communicating with the gallbladder (blue arrow) via a fistula (orange arrowhead).

and mucosal coloration with diffuse sensibility of the right flank. The diagnosis of nephritic colic complicated by acute pyelonephritis was suspected and a renal-vesical ultrasound was performed. The ultrasound showed well-differentiated kidneys, without focal areas of hypo echogenicity to suggest pyelonephritis or dilatation of the excretory tract. Ultrasound examination of the rest of the abdomen showed a cystic formation of segment VI of the liver communicating with the gallbladder via a fistula, as well as a discrete dilatation of the main bile duct (Fig. 1). In the lab work up, we found an increase in infectious parameters (C-reactive protein at 210 mg/l, white blood cells at 18,000 elements/μL); and in addition an increase in cholestasis markers (total bilirubin at 65 mg/l including a direct bilirubin at 58 mg/l, alkaline phosphatases at: 250 IU/l, gamma glutamyl transferase at 100 IU/l) associated with moderate cytolysis. An emergency abdominal and pelvic CT scan revealed a well-limited cystic formation in segment VI of the liver with a wall split and detached membrane realizing the "**water lily sign**". This formation is ruptured at the level of the gallbladder associated with a discrete dilatation of the main bile duct. The diagnosis of hydatid cyst (classified type II of Gharbi and CE3 of WHO classification) ruptured in the gallbladder and the main bile duct, responsible for angiocholitis was retained. The patient's therapeutic management was based on broad-spectrum antibiotic therapy initiated as an emergency to control sepsis, with correction of fluid and electrolyte disorders followed by cholecystectomy, biliary tract drainage, and delayed emergency peri-cystectomy. An antiheminthic treatment of 04 weeks was also prescribed. The evolution was favorable allowing the patient to return home during the second week after surgery with the organisation of a consultation schedule for the care of the surgical scar and a serological assessment for follow-up (Fig. 2).

Discussion

Hydatid disease is an anthropozoonosis secondary to ingestion of dog's taenia egg (ecchynoccocus granulosus), which is still endemic in some regions [1]. In humans, the liver is the most common site, followed by the lung, but other sites, although rare, have been described, such as the peritoneum, kidney, muscle, brain and heart [2]. The hydatid cyst is formed by an endocyst composed of an internal single-cell germ layer secreting fluid that keeps the cyst under tension, surrounded on the outside by an external acellular layer. The endocyst forms the cyst wall. Around it, the peri-cyst develops, which corresponds to a fibrous reaction of the body. From the germinal layer, proligeral vesicles are formed which contain protoscolex. These vesicles can detach from the inner layer or even rupture, releasing the protoscolex into the fluid. These vesicles and the free protoscolexes constitute the hydatid sand, which is only visible to the naked eye. The proligeral vesicles are different from the daughter vesicles. The latter develop from protoscolexes and have a wall identical to that of the mother vesicle except for the peri-cyst, which they will only develop if they come into direct contact with the host tissue. The clinical manifestations of the disease depend on the organ affected. In the case of liver disease, it may be discovered incidentally, during the work-up of pain or sensation

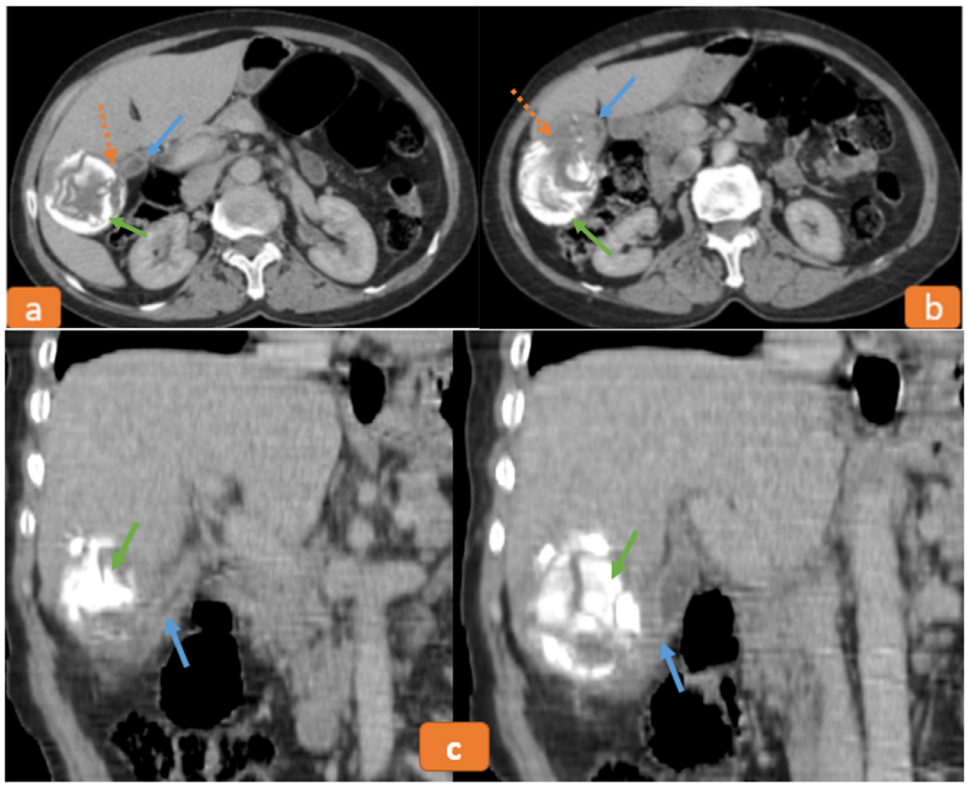

Fig. 2 – Abdominal CT scan enhanced at portal time, in axial section (a and b) and coronal reconstruction (c) showing a well-limited fluid-dense formation in segment VI of the liver with a duplicated wall and detached membrane realizing the "water lily sign". There is also a rupture of the wall with a fistula communication (orange arrowhead) between hepatic hydatid cyst (green arrow) and the gallbladder (blue arrow).

of heaviness in the area, or when complications arise. The main complications are: compression of surrounding structures (bile ducts, duodenum) in relation to an increase in the volume of the cyst and rupture of the cyst in the bile ducts, followed by bacterial superinfection. In this form, the clinical pattern is dominated by angiocholitis with right hypochondrial pain, cutaneous-mucosal jaundice and fever. The fistula of a hydatid cyst of liver interests the right bile duct in 55%, the left bile duct 25% and rarely the gallbladder with only a few cases reported so far (0.3-0.4%).

However, atypical manifestations can be found as in our patient. Imaging, in particular ultrasound and CT scans, are important for diagnosis and classification. MRI can also be used, especially in atypical locations. There are several classifications of hydatid cysts, mostly based on the natural history of the cyst. The most commonly used are the WHO and GHARBI classifications. According to the WHO classification, we distinguish [3]:

WHO	GHARBI	Ultrasound characteristics	Activity
CE1	Type 1	Unilocular hydatid cyst with a clean wall	Active
CE2	Type 3	Multivesicular hydatid cyst	Active
CE3	Type 2	Hydatid cyst with membrane detachment	Transitional
CE4	Type 4	Pseudotumoral hydatid cyst	Inactive
CE5	Type 5	Hydatid cyst with calcified wall	Inactive

The hydatid cyst with a calcified wall, represents the last phase of the cyst's life and corresponds to an inactive form. Complications in this form are uncommon and because of the parietal calcifications, conservative treatment is difficult to achieve. In the absence of complications it does not require medical or surgical management if it size is less than 5 cm with negative immunological tests [4]. In case of rupture in the gallbladder and bile ducts, the therapeutic strategy consists of emergency control of sepsis by medical means followed by delayed emergency cholecystectomy and surgical drainage of the bile ducts with removal of the cyst either by peri-cystectomy, or resection of the liver with the introduction of antihelminthic therapy.

Conclusion

Hydatid cysts of the liver can be marked by the occurrence of serious complications with an often atypical symptomatology. Although calcified forms are mostly inactive and do not require specific management, complications such as ruptures in the bile ducts are still possible.

Patient consent statement

The patient declares his consent for the publication of his case.

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