

Management of a hepatic hydatid cyst ruptured into the retroperitoneum and the abdominal wall muscles: A case report

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ABSTRACT

Introduction. Hydatid disease is a zoonosis caused by the tapeworm *Echinococcus granulosus*. The liver is the most commonly involved organ, followed by lungs. The most common complications of hepatic hydatid cyst are rupture into the biliary tract and secondary bacterial infection. However, rupture into the retroperitoneum and the abdominal wall muscles is exceptional.

Case presentation. We describe an unusual case of a 27-year-old female who was referred to our department for right flank pain for four months. Abdominal ultrasound and CT scan revealed a huge hydatid cyst in liver segment VI fistulizing to the retroperitoneum and the lateral and posterior abdominal wall muscles, causing anterior displacement of the right kidney, with two other cysts in segment II and VIII. Surgery was performed associated with perioperative antiparasitic chemotherapy. The cysts were unroofed and a resection of the protruding domes was performed. The hydatid material in the retroperitoneum and the muscles were completely evacuated by aspiration. Follow-up showed no recurrence.

Discussion. Hepatic hydatid cyst rupture usually occurs into biliary tract, pleural cavity, bronchial tree and intraperitoneal cavity. Rupture into the retroperitoneum and the lateral and posterior abdominal wall muscles is exceptional. To our knowledge, this complication has never been documented before. The hydatid disease may involve insidiously for a long time to lead to such a complication. Surgery was inevitable in our case. It allowed to treat both the hydatid cyst and its complication.

Conclusions. A rupture into the retroperitoneum and the abdominal wall muscles as a complication of a liver hydatid cyst is exceptional. In this presentation, we noticed that hydatid cysts can reach an extremely large size while remaining for a long time asymptomatic. We aim to highlight the significance of preventive measures and public health education to fight against the hydatid disease in endemic areas.

Keywords: hepatic hydatid, cyst, retroperitoneum

INTRODUCTION

Echinococcosis (hydatid cyst) is a zoonosis caused by the tapeworm *Echinococcus granulosus*. It is commonly encountered in endemic areas such as Mediterranean countries, Australia, New Zealand, South America and South Africa [1]. Although it can involve any organ in the human body, the liver and the lung are the most commonly involved

[1]. The most common complications of hepatic hydatid cyst (HHC) are rupture into the biliary tract and secondary bacterial infection [2]. However, rupture into the retroperitoneum and the lateral and posterior abdominal wall muscles is exceptional. We report herein a rare case of a HHC with retroperitoneal and abdominal wall fistulization.

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CASE PRESENTATION

A 27-year-old female, with no past medical or surgical history, was referred to our department for right flank pain evolving progressively for four months. On physical examination, her temperature was 37.2°, and right flank and upper quadrant tenderness was found. Laboratory studies showed normal white blood cell count of $7.64 \times 10^9/l$ (normal range, $3.50\text{--}9.50 \times 10^9/l$), normal blood levels for C-reactive protein (7 mg/l; normal range, 0-10 mg/l) and normal kidney and liver tests. Abdominal ultrasound showed a voluminous cystic mass in liver segment VI, with two other cystic lesions in liver segment II and VIII, suggestive of hydatid disease. Abdominal and chest computed tomography was performed in order to assess the intra and extra abdominal extension (figure 1). It revealed a 150 x 100 x 80 mm hydatid cyst with a thickened wall in liver segment VI, with exophytic growth, fistulizing to the retroperitoneum, the paraspinal muscle, the latissimus dorsi muscle and the lateral abdominal wall muscles, causing anterior displacement of the right kidney. The two other cystic lesions described in ultrasound were identified in liver segment II (13 mm) and segment VIII (32 mm). The patient was given preoperative antiparasitic chemotherapy based on albendazole 400 mg twice daily for 3 weeks. Then, surgical treatment was performed by right subcostal incision. Intraoperative findings were similar to CT description. The surrounding area was isolated with packs soaked with hypertonic saline solution. After freeing the hepatic flexure of the right colon and carrying out the Kocher maneuver, the posterior parietal peritoneum and the right kidney appeared to have been displaced anteriorly by the large ruptured hydatid cyst of segment VI (figure 2). The 3 cysts and the hydatid material

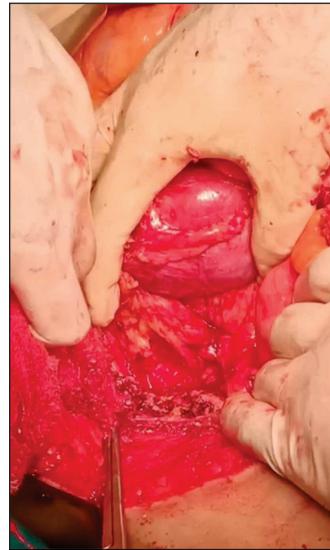


FIGURE 2. Intraoperative findings showing the liver segment VI (arrow head), the retroperitoneum (arrow) and the right kidney (asterisk) after complete evacuation of the hydatid cyst

contained in the retroperitoneum and the muscles were evacuated by aspiration with a closed system suction device. After complete aspiration of cyst contents, the cysts were unroofed and the germinal layer and the remaining daughter vesicles were removed. A resection of the protruding domes was performed and a meticulous search found no evidence of biliocystic fistula. The cysts cavities, the retroperitoneum and the abdominal wall muscles infected by the hydatid material were filled with hypertonic saline solution immersed sponges for a few minutes. Then drainage of the remnant cavities and the retroperitoneum was finally done. The post-operative course was uneventful and the patient was discharged on the 5th day after surgery, with oral albendazole 400 mg twice daily for two months. On follow up after 1 month and 4 months, he was fine and had no evidence of recurrence of the disease.



FIGURE 1. Axial (A) and sagittal (B) abdominal CT scan showing the hydatid cyst in liver segment VI fistulizing to the retroperitoneum and abdominal wall causing anterior displacement of the right kidney

DISCUSSION

Hydatid cyst is a worldwide zoonosis caused by the parasitic tapeworm *Echinococcus granulosus*. Humans are accidental hosts of this parasite, usually becoming infected when they accidentally ingest tapeworm eggs. The larvae cross the intestinal wall via the portal system and reach the liver, where they form cysts [1,2]. It is a major public health problem in endemic countries. Although cysts may develop in almost any part of the body, the location is most likely hepatic (70%). Less common sites are the lungs (15%), spleen, peritoneum, kidneys, brain etc. [3].

HHC may be responsible for a wide spectrum of complications, and rupture of the cyst, compression and secondary bacterial infection are the most frequent [2].

Several theories have been described in concern of the mechanism of the rupture, including the degeneration of the parasitic membranes due to chemical or host defense mechanisms, the ageing of the hydatid cyst and trauma [3]. HHC rupture usually occurs into biliary tract, pleural cavity, bronchial tree and intraperitoneal cavity [4]. Other rare modes of rupture have been described by several authors, such as fistulization into the colon or duodenum, and rupture into the vascular and urinary systems [5-8].

In our case, we reported an exceptional presentation of a HHC ruptured into the retroperitoneum and the lateral and posterior abdominal wall muscles.

Although several authors reported similar cases of HHC with subcutaneous rupture, or presenting with retroperitoneal growth [9,10], to our knowledge, this complication has never been documented before.

The HHC would have to remain asymptomatic for years, to lead to such a complication. Indeed, the hydatid disease may involve insidiously for a long time and diagnosis may be delayed. Furthermore, the current world health situation in relation to the COVID-19 global pandemic may explain, in part, the delays in consultations, and thus the occurrence of such complications [11]. Several studies have noted that psychological status of patients should be the major factor for postponing diagnosis or treatment [12].

On questioning, our patient claimed that she started complaining of abdominal pain only for four months. Before that time, she was completely asymptomatic.

The computed tomography was of great use for diagnosis. Indeed, it plays a crucial role to determine the exact site, number and size of the cysts and to detect the potential complications. Currently, it is strongly recommended to perform a chest and abdomen CT scan before any HHC surgery [13].

Multiple treatment options are now available including pharmacotherapy with albendazole, percutaneous aspiration injection reaspiration (PAIR), endoscopic treatment (for ruptured hepatic cyst communicating with the biliary tree) and surgery. HHC management is nonconsensual, but surgery remains standard treatment [14].

Several surgical techniques could be carried out, ranging from conservative approach including resection of the protruding dome to radical approach including pericystectomy and hepatectomy [14].

It is mainly based upon patient's condition and the liver cyst characteristics.

In all instances, the main purpose of surgery is to remove the parasitic tissues completely [15]. Also, some general principles must be used in any hydatid cyst surgery, such as prevention of the cysts content spillage and isolation of surrounding areas with packs soaked with scolicidal agent, in order to avoid recurrences [16].

In our case, surgery was inevitable. It allowed to treat both the hydatid cyst and its complication. The cysts were unroofed and a resection of the protruding domes was performed. The hydatid material contained in the retroperitoneum and the muscles were completely evacuated by aspiration.

Surgery should be covered by antihelmintics of the benzimidazole family, which decrease the viability of cysts at the time of surgery and significantly reduces the chances of cyst recurrence [17].

CONCLUSIONS

A rupture into the retroperitoneum and the abdominal wall muscles as a complication of a liver hydatid cyst is exceptional. In this presentation, we noticed that hydatid cysts can reach an extremely large size while remaining for a long time asymptomatic. We wish to highlight the importance of early diagnosis using abdominal ultrasound and CT scan to avoid such life-threatening complications. Finally, we would like to insist on the significance of preventive measures and public health education to fight against the hydatid disease in endemic areas.

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