

First case report of retroperitoneal metastasis of fascioliasis after surgery

Jun-Ke Wang, MD^a, Wen-Jie Ma, MD^a, Qiang Lu, MD^b, Er-Liang Zheng, MD^c, Qin Yang, MD^a, Hai-Jie Hu, MD^a, Fei Liu, MD^a, Quan-Sheng Li, MD^{a,*}, Fu-Yu Li, MD, PhD^{a,*}

Abstract

Rationale: Fascioliasis is a rare cause of liver abscesses, and its clinical course consists of hepatic phase and biliary phase.

Patient concerns: We describe a 58-year-old female patient who presented with a 2-month history of intermittent fever and abdominal pain. An abdominal computed tomography (CT) revealed confluent low-density lesions in the liver. Complete surgical resection of these abscesses was performed, and postoperative pathological examination and serological tests confirmed a diagnosis of fascioliasis. However, 4 months after the surgery, follow-up CT revealed a lesion in the retroperitoneal area. Meanwhile, ultrasonography-guided percutaneous needle biopsy of the retroperitoneal lesion was performed, and a parasitic infection was suspected.

Diagnoses: Retroperitoneal metastasis of hepatic phase fascioliasis.

Interventions: The patient received parasitic resistance treatment with triclabendazole at a dose of 10 mg/kg/d for 2 consecutive days.

Outcomes: After 2 courses of triclabendazole therapy, the retroperitoneal metastasis regressed to a minor lesion.

Lessons: To the best of our knowledge, this is the first case report of retroperitoneal metastasis of fascioliasis, aimed at helping recognize the clinical features and treatment options of this rare disease.

Abbreviations: CT = computed tomography, ERCP = endoscopic retrograde cholangiopancreatography.

Keywords: fascioliasis, hepatic phase, retroperitoneal metastasis, triclabendazole

1. Introduction

Fascioliasis is a rare cause of liver abscesses, attributed to *Fasciola hepatica* infection.^[1] The clinical course of fascioliasis has 2 phases—hepatic phase and biliary phase.^[2] Symptoms in hepatic phase lack specificity, and it is difficult to distinguish from a pyogenic liver abscess. The definitive diagnosis of this phase relies on the identification of live parasites or eggs in the feces, but the positivity rate with this detection method is low. Serological tests

are reliable methods and widely used in the diagnosis of fascioliasis. However, when the diagnosis is undefined or delayed, surgical resection of these abscesses is feasible to avoid the progression of the infection. Triclabendazole is currently the most effective agent for treating fascioliasis.

Here, we present a case of retroperitoneal metastasis of hepatic phase fascioliasis to help recognize the clinical features and treatment options of this rare disease. Retroperitoneal metastasis of fascioliasis has not been reported in the existing literature.

2. Case presentation

A 58-year-old female patient presented with a 2-month history of intermittent fever (up to 39.5°C) and abdominal pain in the upper right quadrant. The patient had a significant weight loss of 6 kg within 2 months. In the past year, she had traveled to rural areas in the west of China. Hepatomegaly and mild tenderness in the right upper abdomen were detected during physical examination. The results of the pertinent laboratory investigations were within their normal ranges, except for significant peripheral eosinophilia, which was initially ignored. A contrast-enhanced computed tomography (CT) of the abdomen revealed confluent irregular low-density lesions with obscure boundaries in the subcapsular portion that were 7.5 × 5.2 cm (Fig. 1). Pyogenic liver abscess was the initial diagnosis, and antibiotics were used for 4 weeks to treat the patient. However, her symptoms of abdominal pain and fever did not improve. Ultimately, complete surgical resection of these abscesses was performed for the patient.

The postoperative specimen examination revealed an eosinophilic abscess, which suggested a parasitic infection. Then, serological tests were performed, and results were positive for

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^aDepartment of Biliary Surgery, ^bDepartment of Ultrasound, ^cDepartment of Gastrointestinal Surgery, West China Hospital of Sichuan University, Chengdu, Sichuan Province, China.

*Correspondence: Fu-Yu Li and Quan-Sheng Li, Department of Biliary Surgery, West China Hospital of Sichuan University, Chengdu 610041, Sichuan Province, China (e-mails: lfy_74@hotmail.com; liquansheng@medmail.com.cn).

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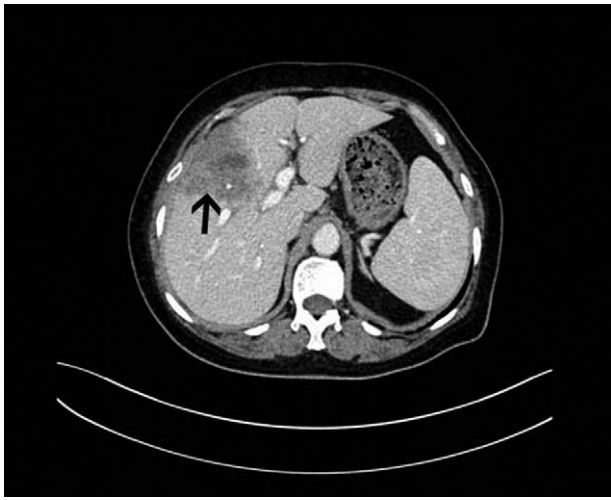


Figure 1. Contrast-enhanced CT scan showed an enlargement of the liver, irregular low attenuation lesions (black arrow) in segment VIII, and invasion of the liver capsule. CT=computed tomography.

F hepatica antibodies and negative for other parasites. The patient received a routine follow-up every 2 months after the surgery. Four months after the surgery, a follow-up CT in a local hospital revealed a 5.4×3.2 cm retroperitoneal low-density lesion that surrounded the abdominal aorta and inferior vena cava (Fig. 2). The lesion was also verified through an abdominal ultrasonographic examination, which showed a 4.6×2.3 cm hypoechoic lesion in a retroperitoneal location (Fig. 3). Meanwhile, an ultrasonography-guided percutaneous needle biopsy of the lesion was performed. Microscopic examination revealed that the lesion contained numerous eosinophilic cells, lymphocytes, and Charcot-Leyden crystals. These findings were consistent with an eosinophilic abscess, and fascioliasis metastasis was suspected. The patient received parasitic resistance treatment with triclabendazole at a dose of 10 mg/kg/d for 2 consecutive days for diagnostic and therapeutic purposes. After 2 courses of the treatment, an ultrasonographic examination revealed that the lesion was reduced to a minor lesion in the same location,



Figure 2. Follow-up abdominal CT scan showed a retroperitoneal lesion with low density (black arrow) surrounding the abdominal aorta and inferior vena cava. CT=computed tomography.

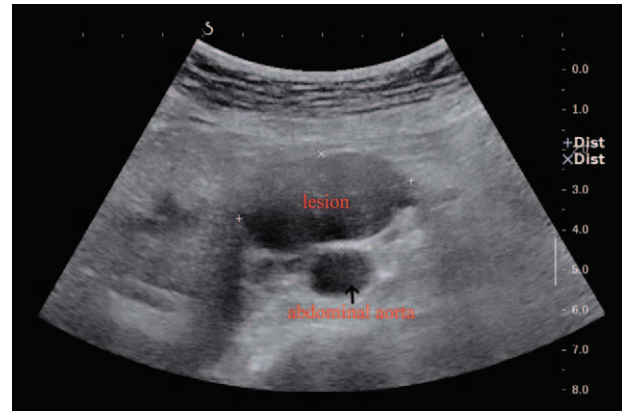


Figure 3. Abdominal ultrasonographic image showed a hypoechoic lesion with clear boundaries near the abdominal aorta.

measuring 1.4×0.6 cm (Fig. 4). On the basis of the results of the pathological examination and the curative effect of triclabendazole, retroperitoneal metastasis of fascioliasis was eventually confirmed.

2.1. Ethical review and written informed consent

The ethical approval from the Ethics Committee was not necessary to report in this case because the case was reviewed retrospectively. Written informed consent was obtained from the patient for publication of this case report and all accompanying images.

3. Discussion

Fascioliasis is a parasitic infection in the liver caused by *F hepatica*, which is a trematode that infects the liver.^[3] This trematode is common in goats, sheep, and cattle, but is rare in humans.^[4] However, the incidence of this disease in humans has shown a gradual increase in recent years, mainly in developing countries such as Africa and Latin America.^[5] An estimated several million people are infected in more than 51 countries.^[6] Although rare in humans, this disease should be considered in the differential diagnosis, especially in highly endemic areas.

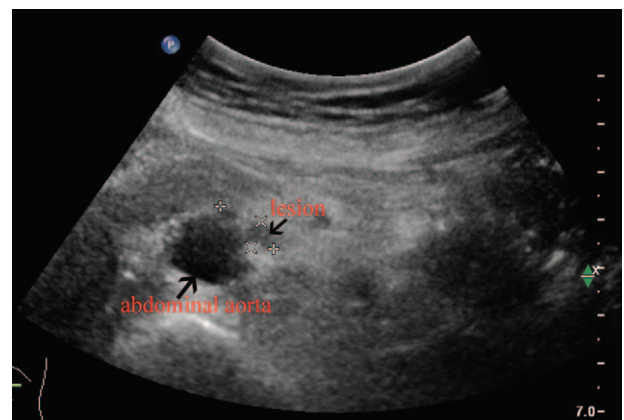


Figure 4. Ultrasonographic image showed that the lesion had regressed after drug treatment with triclabendazole.

Human fascioliasis manifests in 2 phases—hepatic phase and biliary phase. In the hepatic phase, the immature larvae penetrate the liver capsule into the liver parenchyma, which clinically manifests as a liver abscess.^[7] Clinical symptoms of hepatic phase fascioliasis can be nonspecific and manifest as fever with chills, abdominal pain, and weight loss. Before the symptoms develop, most patients reported to have traveled to an endemic area. Hepatomegaly and epigastric tenderness are often detected during the physical examination. Peripheral eosinophilia and abnormal liver function are the most frequent laboratory abnormalities.^[8] Our patient had significant peripheral eosinophilia, but this did not draw our attention because cases of parasitic infection are relatively rare in our hospital. In the biliary phase, *F hepatica* can be directly visualized in the bile duct on endoscopic retrograde cholangiopancreatography.^[9]

The diagnosis of fascioliasis is difficult and often delayed because of nonspecific clinical manifestations and extensive differential diagnoses of liver or bile duct diseases. Contrast-enhanced CT may provide information relevant to the diagnosis because the characteristic finding is often multiple tortuous, tubular hypoattenuating tracts that extend from the liver capsule into the parenchyma. However, with imaging examination alone, fascioliasis would be difficult to distinguish from liver abscesses of other etiologies, such as the more common pyogenic and amoebic infections, paragonimiasis, and hydatid causes. Definitive diagnosis relies on the identification of fluke eggs in the feces. However, this method is not reliable and has a low sensitivity.^[10] The positivity rate has been reported to be only 0.45% (1/22).^[8] Therefore, fascioliasis cannot be ruled out when the stool examination result is negative. In our patient, no eggs were detected on fecal examination. Pathological examination of the lesion was helpful for the initial diagnosis of a parasitic infection because it revealed an eosinophilic abscess. However, pathological examination cannot distinguish *F hepatica* from other parasites, such as *Paragonimus westermani* and *Paragonimus miyazaki*. Under these circumstances, serological tests are widely used for diagnosing fascioliasis, although they take a relatively long time to complete. Our patient was definitively diagnosed on the basis of serological tests after surgery.

Surgical resection of the abscesses is not the first choice of treatment for hepatic phase fascioliasis. However, for a large degree of liver involvement or if the definitive diagnosis will be delayed, it can be a suitable curative treatment. Currently, triclabendazole is a highly effective agent and is the first-line treatment for fascioliasis.^[11] In addition, bithionol was also reported to be effective for this disease.^[12] Praziquantel is

effective against almost all species of trematodes and cestodes, except fascioliasis.^[13] When a parasitic infection is suspected, these antiparasitic drugs can be used for diagnostic and therapeutic purposes.

4. Conclusions

To the best of our knowledge, the present case is the first reported case of retroperitoneal metastasis after surgical resection of hepatic phase fascioliasis. However, the migratory pathway of the parasite remains unclear and needs further evaluation. Our case is unique because of the rare retroperitoneal location of the fascioliasis and supports the necessity of thorough clinical and radiological follow-ups after surgery.

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