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CASE REPORT

Surgically resected hepatic mass caused by fascioliasis

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Abstract

Fascioliasis is a parasitic infestation caused by the digenetic trematodes *Fasciola hepatica* and *F. gigantica*. It is not commonly seen in developed countries, so diagnosis there is always difficult as a result of confusion with other hepatic or biliary disorders. A 56-year-old man presented at our hospital with a hepatic mass that had been inadvertently discovered by ultrasonography. Abdominal computed tomography revealed a multi-cystic lesion distributed along the branch of the right bile duct. Endoscopic retrograde cholangiopancreatography showed serrated changes ranging from the upper level of the common bile duct to the right hepatic bile duct. Eosinophilia was not observed and tumor marker levels were within normal ranges. Following right lobectomy combined with bile duct reconstruction, a histological examination revealed cholangitis with inflammatory cell infiltration accompanied by parasite egg-like structures and Charcot–Leyden crystals. An additional serologic test was positive for *F. hepatica* antibodies. A diagnosis of fascioliasis was thus confirmed by histopathology and serology. Fascioliasis should be suspected if imaging findings such as multiple small hypodense lesions in the liver are observed, and serologic tests can be useful for differential diagnosis.

Keywords Fascioliasis · Differential diagnosis · Serology · Radiologic findings

Abbreviations

CT	Computed tomography
MRI	Magnetic resonance imaging
DWI	Diffused weighted imaging
FDG	^{18}F-fluorodeoxyglucose
PET–CT	Positron emission tomography-computed tomography
ERCP	Endoscopic retrograde cholangiopancreatography

Introduction

Fascioliasis is a parasitic infestation caused by the digenetic trematodes *Fasciola hepatica* and *F. gigantica* [1], and the human fascioliasis has increased worldwide in recent years [2]. Since contaminated water is the main source of infestation, which is correlated with public health conditions, fascioliasis is more commonly seen in developing countries and is very rare in developed countries such as the United States and Japan [3]. Indeed, in nonendemic areas, diagnosis can be difficult and is usually delayed because fascioliasis is not often encountered and symptoms may be confused with other hepatic or biliary disorders [4]. Herein, we report a hepatic mass caused by fascioliasis, which was treated by surgical resection.

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Case report

A 56-year-old man with only a past medical history of rheumatoid arthritis presented at our hospital with an asymptomatic hepatic mass that had been inadvertently discovered by ultrasonography during a routine physical examination. He had no alcohol abuse and was negative for hepatitis B and hepatitis C infections. He stayed several years in China at young age before moving to Japan. The white blood cell count was slightly elevated (9750/μL) but eosinophilia was not observed. His liver function was normal and there was no elevation of serum tumor markers such as α-fetoprotein (2.1 ng/ml), protein induced by vitamin K absence or antagonist-2 (33 mAU/ml), carbohydrase antigen 19–9 (9.7 U/ml), and carcinoembryonic antigen (2.6 ng/ml). Other routine biochemical profile variables were within normal limits.

The patient underwent abdominal computed tomography (CT), which revealed a multi-cystic lesion with distorted shape and without solid components, distributed along the branch of the right bile duct in segment VI of the liver, but the continuity with the bile duct was not clear (Fig. 1a–c). The fluid content was suggested to be highly proteinous or hemorrhagic. In addition, wall thickening of the right hepatic bile duct was also observed (Fig. 1a). Abdominal magnetic response imaging (MRI) showed high intensity on T1-weighted (Fig. 2a) and T2-weighted images (Fig. 2b). Diffusion-weighted imaging (DWI) also showed hyperintensity (Fig. 2c). As seen in CT imaging, wall thickening of the right hepatic bile duct was observed and the tumor was shown to be distributed along the bile duct; however, continuity with the bile duct could also not be confirmed by MRI. [^{18}F]-fluorodeoxyglucose-positron emission tomography (FDG-PET) showed a slightly elevated accumulation of [^{18}F]-FDG (maximum standard uptake value = 4.0) in the posterior segment of the liver (Fig. 2d). Therefore, differential diagnoses included parasitic disease, IPNB with hemorrhage and MCN. Endoscopic retrograde cholangiopancreatography (ERCP) was also performed, showing irregular serrated changes over 15 mm in the right hepatic duct without dilation (Fig. 3). Inflammatory disease such as secondary sclerosing cholangitis could not be ruled out, but malignant tumors such as cholangiocarcinoma were more likely. Slight serrated changes were also observed in the upper level of the common bile duct, suggesting the possibility of tumor extension. However, rosary dilation of intrahepatic bile ducts, seen in CT and MRI, was not confirmed by ERCP. We performed a biliary biopsy of the right hepatic duct and no malignant cells were observed. Serology test was also performed preoperatively and showed a positivity result for *F. hepatica*.

Based on blood tests and radiological examinations, a biliary inflammatory disease such as fascioliasis was suspected but neoplastic lesions such as intraductal papillary neoplasm of the bile duct could not be ruled out. Considering the young age of the patient and the potential of malignancy, right lobectomy combined with bile duct reconstruction was performed after adequate informed consent. The operative time was 331 min, and the estimated blood loss was 259 ml.

Macroscopically, the resected specimen showed dilation and wall thickening of the intrahepatic bile ducts, which were partially filled by necrotic and inflammatory material (Fig. 4a). Microscopically, parasite egg-like structures, actinomycetes, and Charcot–Leyden crystals were observed in the necrotic contents of bile duct (Fig. 4b–d). Cholangitis and inflammatory infiltration of neutrophils to the bile duct epithelium were observed at the right hepatic duct. There was no evidence of malignancy. These features were compatible with parasite infection of the liver. Since total resection of the liver mass was performed, no addition treatment such as triclabendazole was additionally administered.

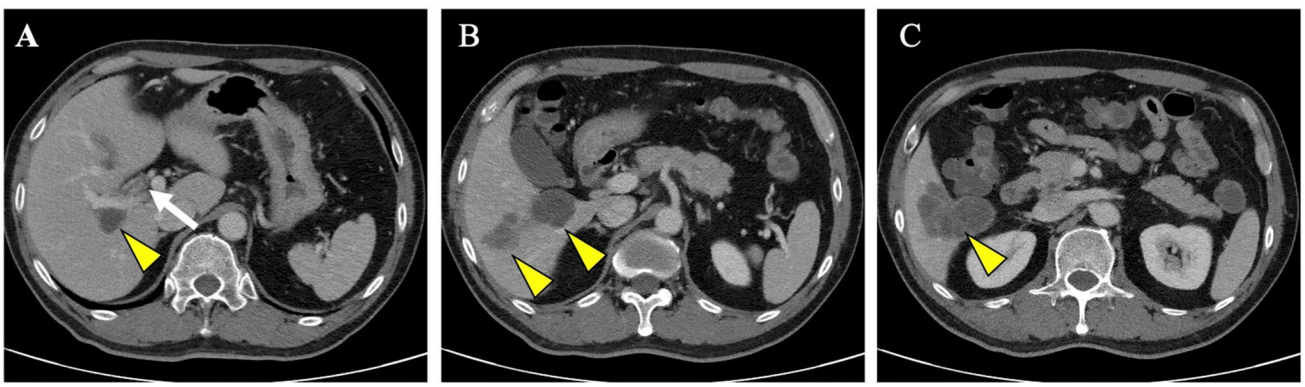

Fig. 1 Contrast-enhanced abdominal computed tomography (CT). A multi-cystic lesion was shown to be distributed along the branch of the right bile duct, but continuity with the bile duct was not clear (**a–c**, arrow head). Wall thickening of the right hepatic bile duct was also observed (**a**, arrow)

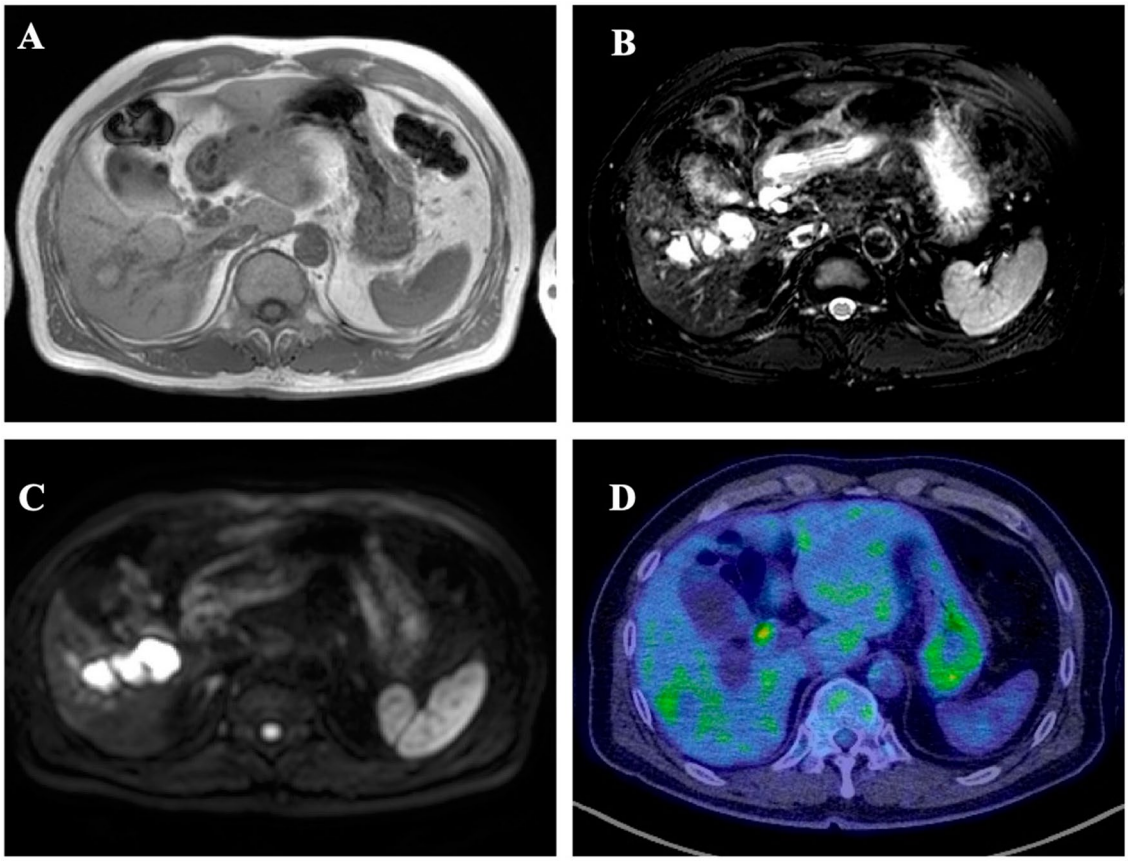

Fig. 2 Magnetic resonance imaging and [^{18}F]-fluorodeoxyglucose-positron emission tomography (FDG-PET) findings. **a** T1-weighted MRI showing a slightly high signal intensity. **b** T2-weighted MRI showing a high signal intensity. **c** Diffusion-weighted imaging (DWI) also showing hyperintensity. **d** FDG-PET showing a slightly elevated accumulation of [^{18}F]-FDG (SUVmax = 4.0)

Follow-up is regularly performed at the outpatient and no recurrence was observed.

Discussion

We report a surgically resected case of liver tumor caused by fascioliasis. Fascioliasis is a trematode flatworm infection of *F. hepatica* or *F. gigantica*. *F. hepatica* has a worldwide distribution, while *F. gigantica* occurs predominantly in the tropics. Although globally distributed, *F. hepatica* infection is more endemic in Central and South America (especially Bolivia and Peru), Europe (especially Portugal, France, Spain, and Turkey), Asia (especially China, Vietnam, and Thailand), Africa, and the Middle East [5]. Sporadic cases have also been reported in the US [6], Australia [7], Japan [8], and elsewhere [1]. An estimated 2.4 to 17 million people are infected in more than 51 countries while 91 million are at risk worldwide [9]. The patient in this case stayed several years in China at young before moving to Japan. Since infection of fascioliasis is more endemic in China compared with in Japan, it is suspected that the stay in China might be the route of the infection.

The infection course of fascioliasis is mainly divided into two phases: the acute phase and chronic phase. In the acute phase, symptoms occur when the parasite migrates from the intestine to the liver, which always occurs within 6–12 weeks of infection. Symptoms of the acute phase include fever, right upper quadrant pain, and hepatomegaly. Jaundice is occasionally observed and marked peripheral eosinophilia is almost always present. The acute phase can also be complicated by hemobilia or subcapsular hematomas of the liver [10]. During the chronic phase, most cases become asymptomatic although right upper quadrant pain, diarrhea, nausea, vomiting, and jaundice can occur. Since adult trematodes are already located in the bile ducts during this phase, bile duct obstruction can occur in some cases, leading to cholangitis, cholelithiasis, and pancreatitis [11–13]. In the present case, the patient had no symptoms such as abdominal pain, jaundice, or any other digestive symptoms, and eosinophilia was not observed. Since *F. hepatica* eggs were seen in the patient's

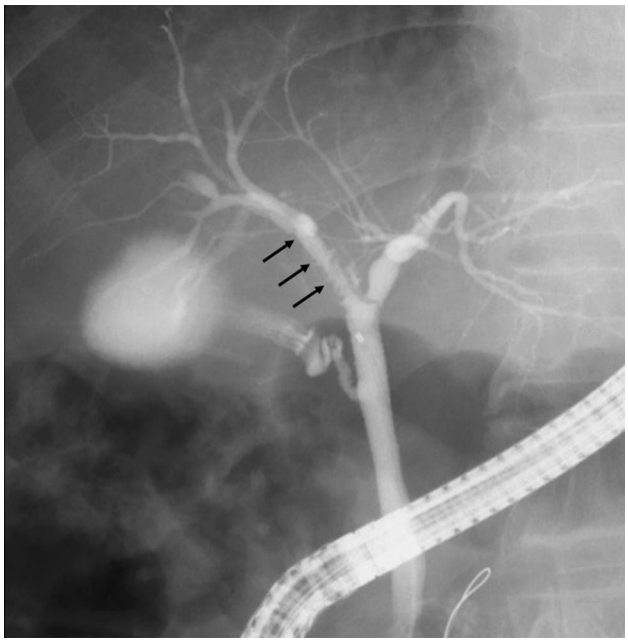

Fig. 3 Endoscopic retrograde cholangiopancreatography (ERCP). Irregular serrated changes are shown, ranging from the right hepatic duct to the upper level of the common bile duct. The left hepatic duct is intact

bile ducts, we speculate that this case was in the chronic phase of infection.

Although confirmation of the diagnosis is necessary and should be based on serology and parasitology tests, radiologic findings can also aid diagnosis [14]. Useful radiographic tools include CT, ultrasonography, and MRI [15]. A review of radiographic findings in fascioliasis identified multiple small nodular and branching lesions as the most common findings, which were frequently seen in the subcapsular area of the liver parenchyma [16]. These appear hypoechoic on ultrasonography, hypodense on CT, and T2 hyper and T1 hypointense on MRI. Necrosis may also be seen in large lesions [17]. In this case, CT findings showed multicystic and branching lesions, while MRI findings revealed high intensity on T2-weighted images. These findings were compatible with previous reports. ERCP is also a useful tool if dilation of the intrahepatic biliary tree is observed [18]. However, rosary dilation, which was shown in CT and MRI, was not observed by ERCP in this case. Instead, serrated changes, such as those frequently seen in cholangiocarcinoma, were observed. Since ERCP findings suggested the potential of malignancy rather than inflammatory disease, the decision to undergo surgery was made, resulting in overtreatment for the patient.

Triclabendazole has been proven to be highly effective against fascioliasis with a cure rate of > 90% [19]. Dosage of 10 mg/kg orally for 1 or 2 days is recommended and the drug is relatively well tolerated. Since triclabendazole has been proven to be highly effective against fascioliasis, we admitted that right lobectomy combined with bile duct reconstruction might be over indicated for this patient and treatment of drug administration should be considered first. In this case, because total resection of the liver mass was first performed and the patient had no symptoms implying an active infection, no additional treatments such as triclabendazole administration were added after surgery.

Conclusions

We herein present a rare case of a hepatic mass caused by fascioliasis which was treated by surgical resection in Japan. Since fascioliasis is not common in developed countries, confirmation of a diagnosis can be difficult. However, we suggest that fascioliasis should be suspected if certain imaging findings such as multiple small hypodense lesions in the liver are observed, and conclude that serologic tests can be useful for diagnosis.

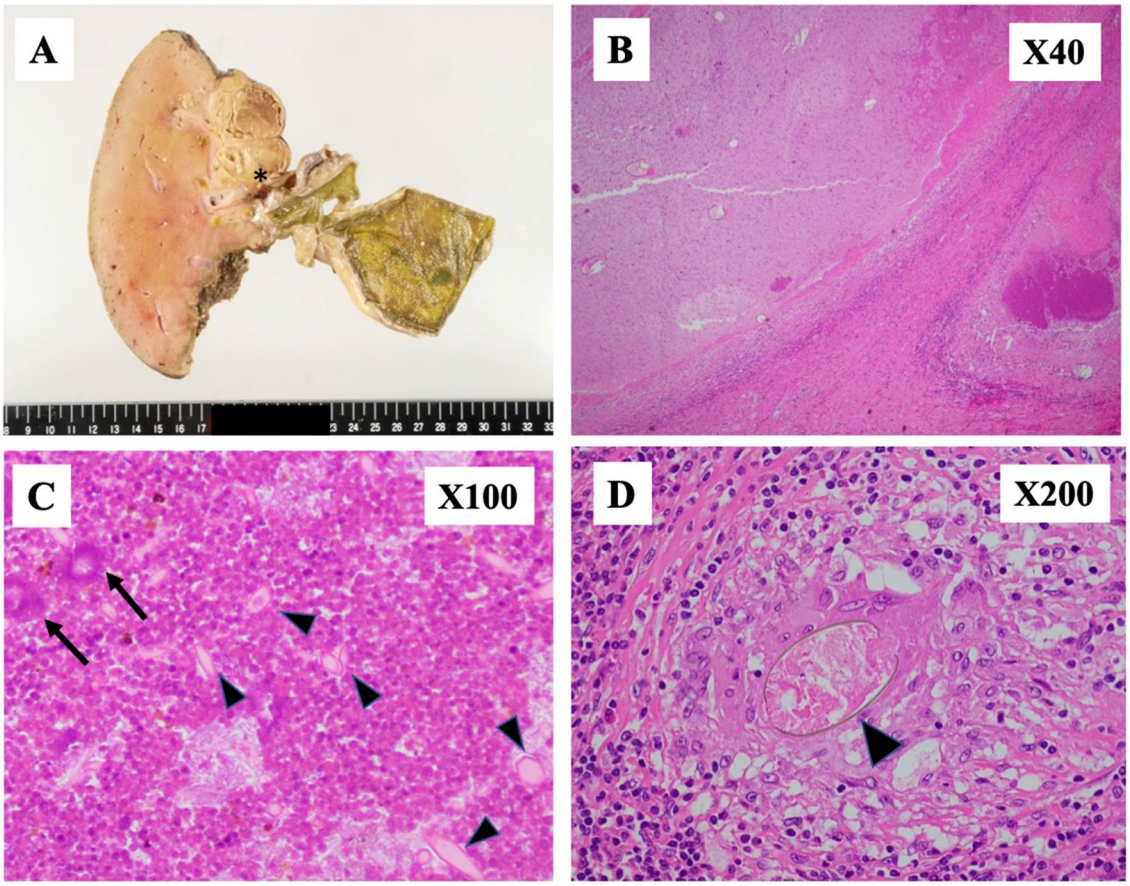

Fig. 4 Macroscopic and microscopic findings of the lesion. Macroscopically, the cut surface of the hepatic tissue shows dilation and wall thickening of the intrahepatic bile ducts, which are partially filled by necrotic and inflammatory material (**a**). Microscopically, cholangitis and inflammatory infiltration of neutrophils are seen (**b**, hematoxylin and eosin (H&E) staining, magnification ×20), accompanied by actinomycetes (**c**, H&E staining, magnification ×100, arrow), and Charcot–Leyden crystals (**c**, arrow head). Parasite eggs are also apparent (**d** H&E staining, magnification ×200, arrow head)

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Compliance with ethical standards

Conflict of interest Huanlin Wang, Shinji Itoh, Yuji Matsumoto, Akihiro Nishie, Takeshi Kurihara, Tomonari Shimagaki, Yoshihiro Nagao, Takeo Toshima, Noboru Harada, Kenichi Kohashi, Yoshinao Oda, Kousei Ishigami, Haruhiko Maruyama, Tomoharu Yoshizumi and Masaki Mori declare that they have no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from all patients for being included in the study.

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