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Finding of Biliary Fascioliasis by Endoscopic Ultrasonography in a Patient with Eosinophilic Liver Abscess

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Key Words

Magnetic resonance cholangiopancreatography · Endoscopic ultrasonography · Endoscopic retrograde cholangiopancreatography · *Fasciola hepatica*

Abstract

Fascioliasis is an endemic zoonotic disease in Iran. It occurs mainly in sheep-rearing areas of temperate climates, but sporadic cases have been reported from many other parts of the world. The usual definitive host is the sheep. Humans are accidental hosts in the life cycle of *Fasciola*. Typical symptoms may be associated with fascioliasis, but in some cases diagnosis and treatment may be preceded by a long period of abdominal pain and vague gastrointestinal symptoms. We report a case with epigastric and upper quadrant abdominal pain for the last 6 months, with imaging suggesting liver abscess and normal biliary ducts. The patient had no eosinophilia with negative stool examinations, so she was initially treated with antibiotics for liver abscess. Her clinical condition as well as follow-up imagings showed appropriate response after antibiotic therapy. Finally, endoscopic ultrasonography revealed *Fasciola hepatica*, which was then extracted with endoscopic retrograde cholangiopancreatography.

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Introduction

Fascioliasis, as a zoonotic disease, mostly affects sheep, goats and cattle and is caused by a 2–4 cm flat trematode of the liver named *Fasciola hepatica* [1]. The number of reports of

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humans infected with *F. hepatica* has significantly increased since 1980, and several geographic areas have been described as endemic regions for the disease in humans, with different prevalence and incidence rates [1–4]. In endemic regions, very young children and women are most likely to be infected, and a high incidence of co-infection with other parasites, especially echinococcosis, is found [2]. In humans, the infection begins with the ingestion of watercress or contaminated water containing encysted larvae. The larvae excyst in the stomach, penetrate the duodenal wall, escape into the peritoneal cavity and then pass through the liver capsule to enter the biliary tree [5]. Eosinophilia is a variable finding in this phase of the disease [2]. Serologic tests and imaging studies of the hepatobiliary tract are of great importance in diagnosis [6–9].

Here, we report a 64-year-old woman with epigastric and upper quadrant abdominal pain for the last 6 months, with imaging suggesting liver abscess and normal biliary ducts. Finally, endoscopic ultrasonography (EUS) revealed *F. hepatica*, which was then extracted with endoscopic retrograde cholangiopancreatography (ERCP).

Case Report

A 64-year-old female with epigastric and right upper quadrant pain was referred to Taleghani Hospital in Tehran, Iran. She complained of intermittent pain attacks radiating to the back, exacerbated after eating. There had been 3–4 episodes of pain on most days, lasting about 1 h and then decreasing gradually for at least 6 months before presentation. She also stated weight loss of about 3 kg (from 66 to 63 kg) within 6 months without fever, icterus or any change in bowel habit. In her past medical history, there were no important medical problems.

Her previous laboratory evaluation revealed an increased erythrocyte sedimentation rate (64 mm/h), while total bilirubin was 3.2 mg/dl (normal <2 mg/dl), aspartate aminotransferase 31 IU/l (normal <30 IU/l), alanine aminotransferase 37 IU/l (normal <30 IU/l), alkaline phosphatase 473 IU/l (normal <306 IU/l) and γ -glutamyltranspeptidase 82 IU/l (normal <60 IU/l). Complete blood count showed hemoglobin 12.1 g/dl, platelets 190,000/mm³ (normal 100,000–450,000/mm³) and white blood cells 4,030/mm³ (normal 4,000–10,000/mm³) with 12% eosinophils (484/mm³) on peripheral smear. Blood chemistry tests were in the normal range. Hydatid antibody titer was requested and was within normal limits. The results of stool examinations at three different times for detecting ova and parasites as well as occult blood were normal.

After admission, ultrasonography and abdominopelvic computed tomography (CT) scan showed normal intrahepatic bile ducts, but an ill-defined large heterogeneous mass lesion which contained multiple hypodense portions in the right and caudate lobes of the liver was noted (fig. 1). There were no other remarkable findings on ultrasonography and CT imaging. After that, the patient underwent evaluation to rule out possible malignant and non-malignant etiologies as follows: Endoscopy and colonoscopy were performed to rule out probable sources of liver metastasis, which were both unremarkable. The patient also underwent CT-guided core needle biopsy for pathologic and microbiological evaluation, which showed predominant eosinophil infiltration of liver sections with microabscess (eosinophilic abscess) formation without neoplastic or granulomatous process. Microbiological cultures of aspiration fluid of the liver mass showed no growth after 72 h.

Empirically, initial treatment with intravenous ceftriaxone and metronidazole was initiated, followed by oral ciprofloxacin and metronidazole. Following antibiotic therapy, the patient's symptoms alleviated gradually, and erythrocyte sedimentation rate decreased to

10 mm/h within 3 weeks. The patient was discharged from hospital and on follow-up visits mentioned improvement in her general condition. Ultrasonography performed 2 months after discharge showed resolution of the liver mass with normal intra- and extrahepatic biliary ducts, but second follow-up ultrasonography revealed a small (8 × 10 mm) cystic lesion in the right lobe of the liver with dilation of the common bile duct (CBD) (9 mm) that was confirmed by magnetic resonance imaging (fig. 2). Magnetic resonance cholangiopancreatography showed a slightly dilated CBD (9.5 mm) and normal intrahepatic bile ducts, with irregularity of the wall of the distal third of the CBD with suspicious filling defect (fig. 3). Finally, EUS with a linear echoendoscope showed a 10 mm CBD with long, linear, hyperechogenic structures, moving freely in the lower third of the CBD, in favor of *F. hepatica* (fig. 4). After diagnosis of *F. hepatica* by EUS, ERCP was performed, and after sphincterotomy, an extraction balloon was passed and removed a live Fasciola (fig. 5).

Discussion

Fascioliasis is an emerging disease in humans, while sheep and cattle are the most important definitive hosts of *F. hepatica*; goats, buffalo, horses, camels, hogs, deer, and rabbits can also be infected. Snails are intermediate hosts and humans are incidental hosts [10]. Its eggs are oval and yellow-brown, with a dimension of approximately 130–150 × 60–90 μm. The life span of adult *F. hepatica* flukes within humans is not known but is estimated to be 9–13 years [11]. It occurs mainly in sheep-rearing areas of temperate climates, particularly in certain parts of Central and South America, Europe, China, Africa and the Middle East [2]. Sporadic cases have also been reported from the United States [1]. In Iran Gilan, Ardabil, Khuzestan and Kerman are most affected [12].

The number of adult flukes that reach the biliary tree and fully develop to sexual maturity is usually small. During wet years, the degree of animal infection and the incidence of human infection rise because of an increased number of snails and longer survival of encysted cercariae [13]. Human hepatobiliary infection with *F. hepatica* has two phases: acute (hepatic) and chronic (biliary). The early acute phase, which usually begins within 6–12 weeks of metacercariae ingestion, is often associated with fever, right upper quadrant pain and hepatomegaly. General non-specific symptoms and signs including anorexia, nausea, vomiting, myalgia, cough and urticaria as well as peripheral eosinophilia are common in this phase [10, 13]. The biliary phase is usually asymptomatic, but may present with intermittent right upper quadrant pain with or without cholangitis or cholestasis [11]. Occasionally, the adult flukes can obstruct the CBD and thus induce chronic infection leading to biliary colic, cholangitis, cholelithiasis, obstructive jaundice and even secondary pancreatitis [11, 14–17]. Morbidity can increase with heavier fluke burdens [12].

Eosinophilia is a variable finding in this phase of the disease [2]. Serologic tests and imaging studies of the hepatobiliary tract are of great importance in diagnosis [6–9]. Due to diverse presentations of fascioliasis, there is often a delay in diagnosis, particularly in patients in the hepatic phase [18]. The diagnosis can be established by serology or by identifying eggs in duodenal aspirates, bile specimens or stool. Adult worms may be identifiable in endoscopic or surgical specimens. Alternative approaches to diagnosis include imaging studies, particularly CT scan of the liver which may show characteristic hypodense nodules or tortuous tracks resulting from migration of the parasite through the liver [6, 7]. Magnetic resonance cholangiography, EUS and ERCP are useful in identifying the flukes in the bile ducts and gallbladder in the biliary phase.

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We report this case to emphasize that there might be several ambiguous aspects in diagnostic evaluations of *F. hepatica*, and fascioliasis should be kept in mind in patients with preceding vague gastrointestinal symptoms, especially in endemic areas. Our case did not have significant peripheral eosinophilia or typical clinical presentation, while eosinophilia is very common in the acute phase, and there were no biliary obstruction symptoms. Besides, in comparison to other cases in the literature, the initial imagings did not show any abnormality in the biliary ducts, despite the patient having eosinophilic liver abscess [14–17, 19–21]. Stool examination may be negative for fascioliasis, and there is no obvious suggestive diagnostic clue, so this highlights the fact that *F. hepatica* should be considered in the differential diagnosis of ambiguous hepatobiliary symptoms, especially in endemic areas.

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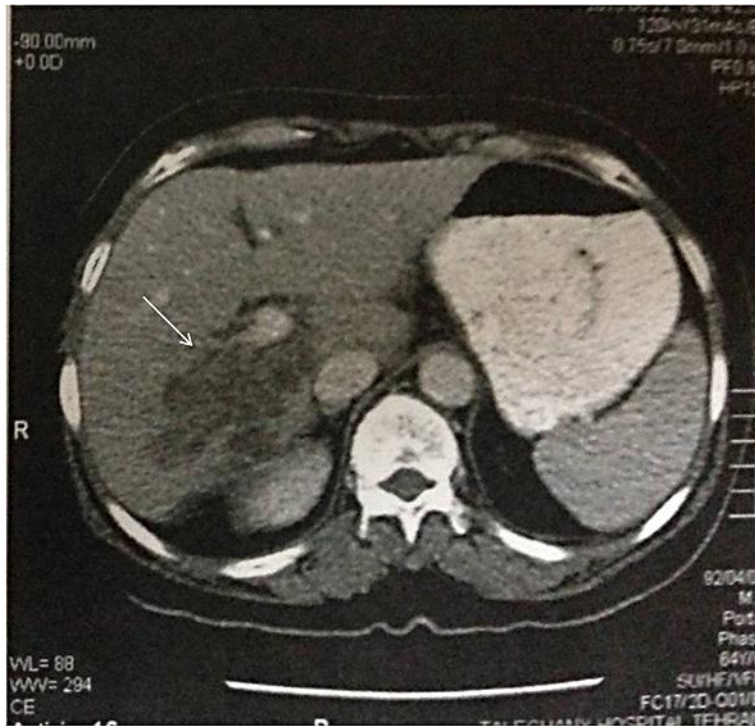


Fig. 1. CT scan of the liver showed a liver abscess. An ill-defined large heterogeneous mass lesion which contained multiple hypodense portions in the right and caudate lobes of the liver was noted (arrow).

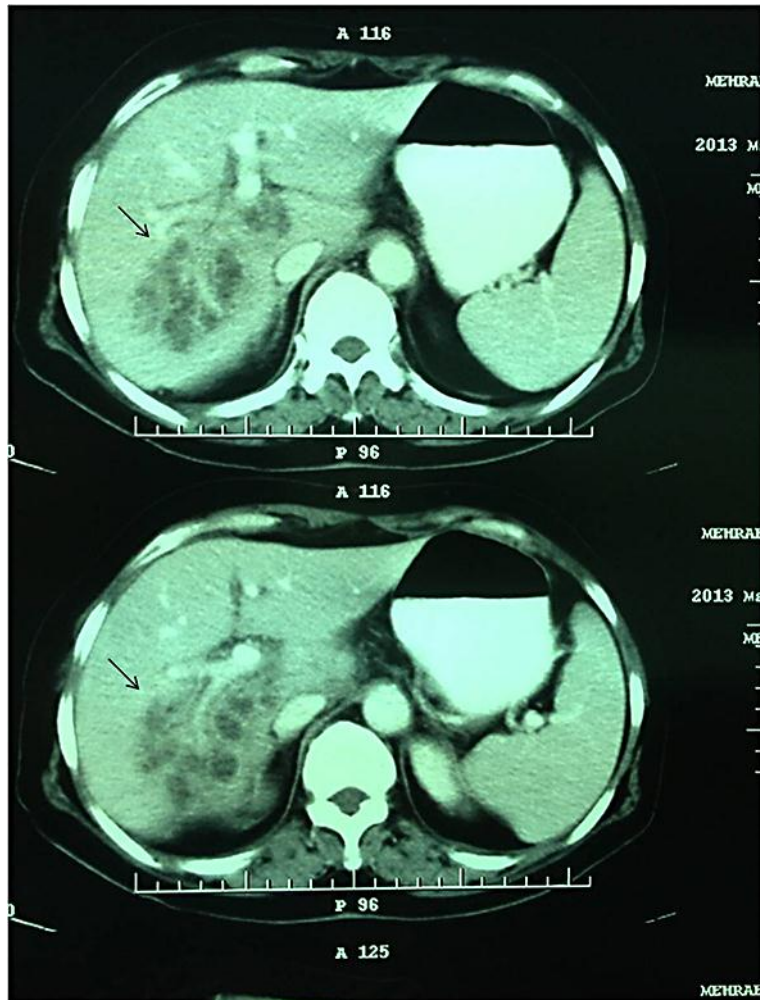


Fig. 2. Magnetic resonance imaging of the liver. After 6 months, follow-up imaging showed relapse of the liver cystic masses (arrows).



Fig. 3. Magnetic resonance cholangiopancreatography showed a slightly dilated CBD (9.5 mm, arrow) with normal intrahepatic bile ducts, with irregularity of the wall of the distal third of the CBD with suspicious filling defect.

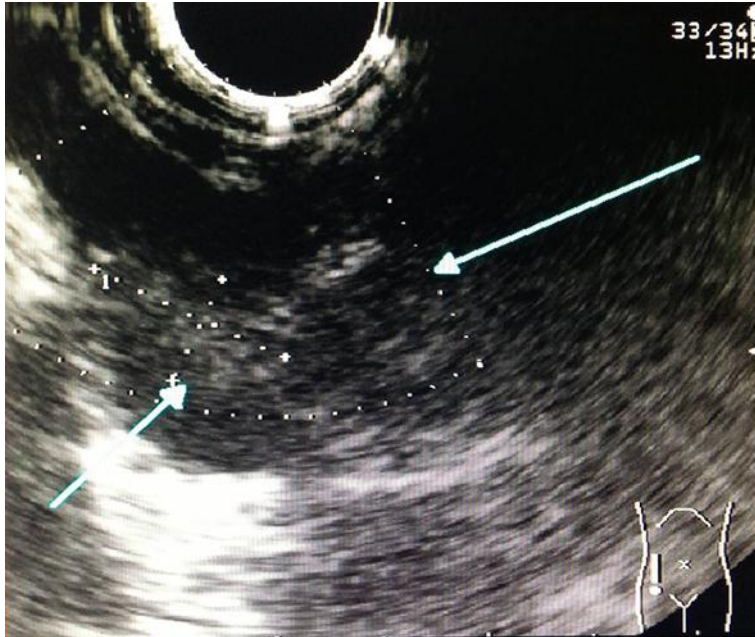


Fig. 4. EUS showed a 10 mm CBD with long (arrows), linear, hyperechogenic structures, moving freely in the lower third of the CBD, in favor of *F. hepatica*.

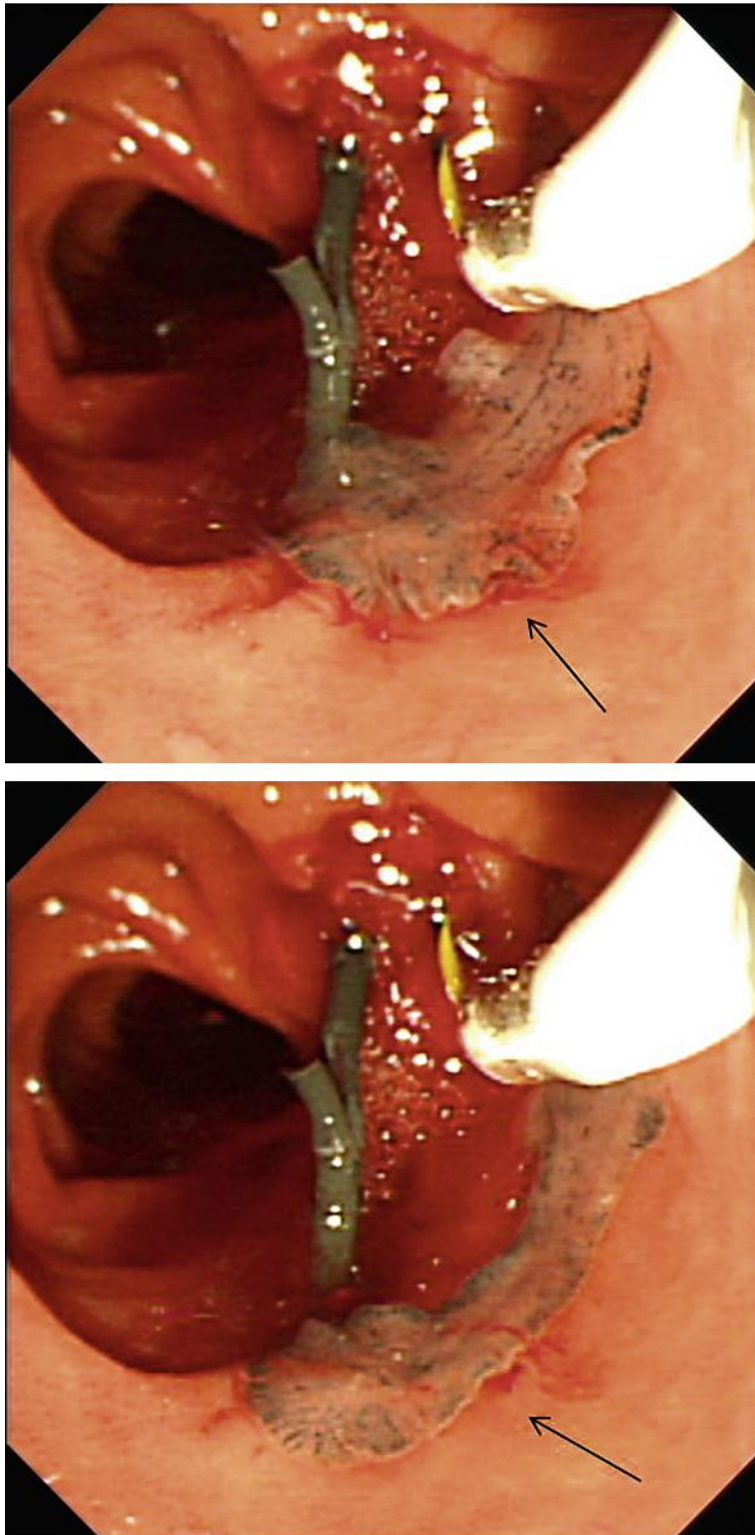


Fig. 5. Live Fasciola (arrows) was extracted from the CBD after sphincterotomy via ERCP.