

Endoscopic extraction of living fasciola hepatica: Case report and literature review

Canlı fasiola hepaticanın endoskopik çıkarılması: Vaka raporu ve literatür derlemesi

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Fasciola hepatica infestation is known to cause bile duct inflammation and biliary obstruction. Endoscopic retrograde cholangiopancreatography shows distinct features in some patients with fascioliasis, but the condition may be overlooked in chronic cases. The endoscopic retrograde cholangiopancreatography images must be carefully examined to rule out other possible causes of irregularity and thickening of the common bile duct wall. Parasite removal during endoscopic retrograde cholangiopancreatography is one therapeutic option in patients with acute obstructive cholangitis due to F. hepatica. We present a case of fascioliasis-induced acute cholangitis that was diagnosed and treated via endoscopy. A review of the literature on extraction of living parasites is also included.

Key words: Fasciola hepatica, cholangitis, endoscopic retrograde cholangiopancreatography.

Fasciola hepatica infestasyonunun biliyer obstrüksiyon ve safra kanallarında inflamasyona neden olduğu bilinmektedir. Fasiolazisli hastalarda endoskopik retrograd kolanjiyopankreatografi'de bazı belirgin görünümler izlenebilir, ancak bu durum kronik vakalarda gözden kaçabilir. Endoskopik retrograd kolanjiyopankreatografi görüntüleri, ortak safra kanalı duvarında kalınlaşma ve düzensizlik yapabilen diğer Biyolojilerin dışlanması açısından çok dikkatli değerlendirilmelidir. Parazitin endoskopik retrograd kolanjiyopankreatografi sırasında çıkarılması fasiola hepaticaya bağlı akut obstrüktif kolanjit olan hastalarda tedavi imkanı sağlar. Endoskopik yolla tanı ve tedavisini uyguladığımız fasiolazise bağlı bir akut kolanjit vakası ve bununla ilgili literatürün gözden geçirilmesini sunuyoruz.

Anahtar kelimeler: Fasiola hepatica, kolanjit, endoskopik retrograd kolanjiyopankreatografi

INTRODUCTION

Infestation with the liver fluke *Fasciola hepatica* is a common zoonosis in sheep-raising areas of the world (1). Most reports of human infection have come from South America, Europe, Africa, China, Australia, and the Middle East, but sporadic cases have also been reported in the United States (2). The sheep is the usual definitive host, and man is an accidental host. The non-embryonated eggs of *F. hepatica* are passed in the feces of the host and contaminate fresh water. The miracidium hatches and penetrates the first intermediate host, a snail. The developed cercariae leave the snail and attach themselves to herbage growing close to the water in sheep pastures. Infestation in man occurs via ingestion of contaminated water or raw green vegetables with encysted metacercariae attached. The infective metacercariae excyst, and the larvae

penetrate the intestinal wall to enter the peritoneal cavity. From there, they usually pass through the liver capsule and hepatic tissue, and the adult form finally invades the biliary tract. It takes approximately 12 weeks from infection to oviposition in humans (1).

Infection with *F. hepatica* has two distinct clinical phases, one corresponding to the hepatic migratory phase of the fluke's life cycle, and the other to the parasite's presence in their final location in the bile ducts (1). *F. hepatica* infestation may be suspected in patients who show tender hepatomegaly, fever, and eosinophilia. These features arise approximately 14 days after ingestion of contaminated water, and may last for several weeks. Adult flukes can cause obstructive jaundice or predispose the patient to cholelithiasis (3).

The diagnosis can be confirmed by serology, and by identification of ova in the stool or in duodenal aspirates. Severe hemobilia and death have been described. Associated lithiasis of the bile ducts or gallbladder is also common, as the eggs or fragments of dead parasites can form nuclei for calculi (4). Another rare complication of fascioliasis is acute pancreatitis (5). We describe a case of *F. hepatica* infestation that was endoscopically diagnosed and treated with endoscopic extraction and anti-parasitic medication.

CASE REPORT

A 52-year-old woman presented with a three-year history of repetitive abdominal pain simulating biliary colic. Three years before presentation, another center had made an ultrasound (US) diagnosis of acute cholecystitis secondary to biliary stone disease, and cholecystectomy had been performed. During the operation, numerous *F. hepatica* had been observed in the gallbladder lumen. The patient stated that she had received two doses of an unknown medication postoperatively and that she had experienced no complaints since then until five days prior to admission. On admission to our clinic, the patient had complaints of epigastric pain, nausea, and vomiting. Laboratory investigations revealed the following results: aspartate aminotransferase: 1137 IU/L, alanine aminotransferase: 554 IU/L, alkaline phosphatase: 311 IU/L, gamma glutamyl transferase: 654 IU/L, total bilirubin: 1.8 mg/dL, conjugated bilirubin: 1.6 mg/dL, hemoglobin: 13.7 g/dL, white blood cell count: 12.1 K/mm³, platelet count: 297.0 K/mm³ and eosinophil count 0.2 K/mm³. Magnetic resonance cholangiopancreatography (MRCP) revealed obstruction of the distal choledochus. Endoscopic retrograde cholangiopancreatography (ERCP) was performed due to extrahepatic cholestasis. This revealed a maximum choledochus diameter of 11 mm, and showed multiple filling defects and irregular choledochal wall margins (Figure 1). During balloon extraction after endoscopic sphincterotomy, three live *F. hepatica* were forced through the choledochus and into the lumen of the duodenum (Figure 2). Occlusion cholangiography with a balloon catheter was performed to evaluate for remaining flukes, and this showed no other parasites. One of the three parasites was physically removed from the patient by applying continuous suction with a duodenoscope (Figure 3).



Figure 1. Endoscopic retrograde cholangiography images: The patient's bile ducts were slightly dilated, and a radiolucent, roughly crescent-shaped shadow is visible in the common bile duct.

The patient was asked to attend follow-up and was discharged with a prescription for bithionol, with a total 30mg/kg dose given three times on alternate days. At a follow-up visit two months later, the patient stated she was well and her clinical and laboratory assessments were within normal limits. Repeat US revealed a normal-sized choledochus.



Figure 2. An endoscopic image of the single fluke that was physically removed from the biliary tract.

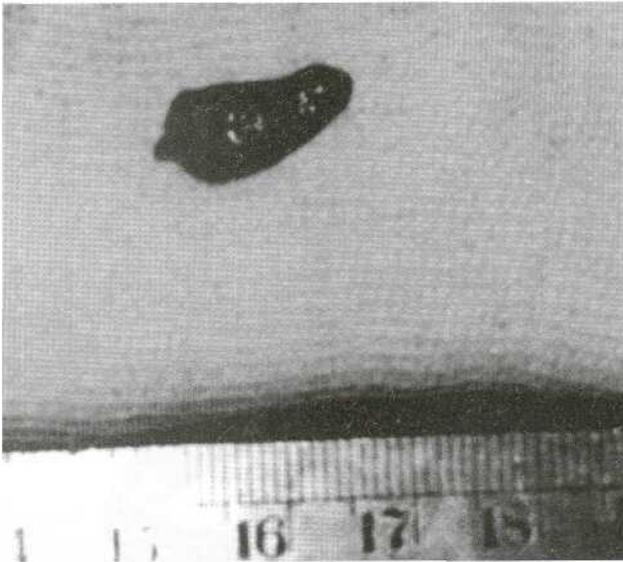


Figure 3. A macroscopic image of adult *Fasciola hepatica* extracted outside the body

DISCUSSION

The liver fluke *F. hepatica* is one of the few parasites that can cause recurrent cholangitis. Human hepatobiliary infection with this organism includes two stages: an acute, invasive, hepatic phase that starts one to three weeks after infestation, and a chronic biliary phase that starts three to four months after the contaminated material is ingested. Although some overlap may occur in endemic areas, where repetitive infection occurs and acute lesions are superimposed on chronic disease, the clinical and pathologic features of these two phases are different and pose distinct diagnostic challenges (6,7).

The clinical manifestations of hepatic fascioliasis vary according to the stage of the disease. In the initial hepatic invasion, fever, pain, hepatomegaly, general malaise, dyspepsia, eosinophilia, and positive serologic testing may be observed for three months. During the second phase, when the parasite is in the main biliary duct, the disease may feature episodes of biliary colic with or without cholangitis, obvious signs of biological cholestasis, or may remain silent. In some instances, lack of eosinophilia in combination with the absence of manifestations of the disease can make diagnosis quite difficult (8). As noted earlier, fascioliasis may be overlooked in chronic cases. This condition should always be included in the differential diagnosis when US or MRCP images show irregular and thickened com-

mon bile duct walls. The ERCP images typical of *F. hepatica* suggest biliary fascioliasis (9).

Our patient had been diagnosed with cholangitis and cholecystitis at another hospital and underwent surgery at that center. Flukes were observed at the time of the operation, and we suspect that some form of anti-parasitic treatment was given. After three years without problems, the patient suffered another cholangitis attack. This indicates either incomplete response to treatment or a second infestation. As noted above, in patients with biliary fascioliasis, ERCP frequently demonstrates typical features of *F. hepatica* in the gallbladder (8,10), dilated bile ducts with small, radiolucent linear or crescent-like shadows, suggesting parasites and with jagged, irregular margins (11,12). ERCP is an important tool for direct diagnosis and for providing bile drainage.

Chronic biliary fascioliasis may be asymptomatic and the proportion of these cases compared to the acute form is not known. When the condition is left untreated, biliary complications of iron deficiency anemia, biliary obstruction and related pain, cholangitis, or portal fibrosis may result (9,13). In order to prevent the development of irreversible complications such as secondary biliary cirrhosis, it is vital to perform a thorough investigation for parasite eggs in patients with suspected biliary fascioliasis.

The technique of endoscopic sphincterotomy was initially introduced to treat common bile duct stones; however, the indications have been expanded to include other biliary disorders. Currently, this method is considered the optimal approach for biliary parasitosis, including biliary ascariasis and biliary hydatid disease (14,15). Previous reports on two patients have noted success with the combination of ERCP and sphincterotomy for extracting *F. hepatica* from the biliary tree (5,9). This treatment is adequate for providing biliary drainage and resolving the cholangitis attack, but complementary therapy with an appropriate anti-parasitic regime is mandatory. Our case emphasizes the need for sufficient treatment and thorough follow-up in patients with fascioliasis. This is the only way to prevent acute or irreversible chronic complications.

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