CASE REPORT

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A rare cause of obstructive jaundice: Fasciola hepatica mimicking cholangiocarcinoma

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Fasciola hepatica is an endemic zoonotic disease in Turkey and neighboring countries. The usual definitive host is the sheep; humans are accidental hosts in the life cycle of the Fasciola. There are two disease stages: the hepatic (acute) and biliary (chronic) stages. When the flukes enter the bile ducts, the symptoms of cholestasis and cholangitis may present, which can easily be misdiagnosed as obstructive jaundice of other causes. We present a case of fascioliasis, which was difficult to differentiate from cholangiocarcinoma. A 47-year-old woman from Eastern Turkey presented with fever, right upper quadrant abdominal pain, and jaundice. Total bilirubin was 4.2 mg/dl, aspartate aminotransferase 55 IU/L, alanine aminotransferase 65 IU/L, alkaline phosphatase 325 IU/L, and gamma-glutamyl transpeptidase 172 IU/L. All tumor markers including carcinoembryonic antigen and Ca19-9 were in normal values. After extended evaluation, an explorative laparotomy with cholecystectomy, choledochostomy and T-tube drainage was performed. Multiple flukes were removed from the choledochus. One of the parasites was sent to the parasitological clinic for identification. The result of an indirect hemagglutination test for F. hepatica was 1/320 (+). In conclusion, the chronic phase of this zoonotic infection can be easily misdiagnosed as any other cause of obstructive jaundice. Thus, F. hepatica should be considered in the differential diagnosis of common bile duct obstruction, especially in endemic areas.

Key words: Fasciola hepatica, obstructive jaundice, endemic zoonotic disease

Tıkanma sarılığının nadir sebebi: Kolanjiyokarsinomu taklit eden Fasiola hepatika

Fasiola hepatika, Türkiye ve komşu ülkelerinde görülen endemik bir hayvan hastalığıdır. Parazitin hayat siklusunda insanlar rastlantı olarak konakçı olurken asıl konakçı koyunlardır. Hastalığın hepatik (akut) ve biliyer (kronik) olmak üzere iki evresi vardır. Parazitler biliyer sisteme ulaştıklarında kolestaza bağlı kolanjit semptomları görülür ve bu durum kolaylıkla tıkanma sarılığının diğer nedenleri ile karıştırılabilir. Biz bu çalışmada kolanjiyokarsinomdan güçlükle ayırt edilebilen Fasiola hepatikalı bir olguyu sunuyoruz. Türkiyenin doğu kesiminde yaşıyan 47 yaşında kadın hasta ateş, sarılık ve karın sağ üst kadranda ağrı şikayeti ile kliniğimize başvurdu. Total bilirubin değeri 4.2 mg/dl, aspartat aminotransferaz 55 iu/L, alanin aminotransferaz 65 iu/L, alkalen fosfataz 325 iu/L, ve gamma-glutamil transpeptidaz 172 iu idi. Ca19.9 ve karsinoembriyonik antijen dahil tüm tümör belirteçleri normal sınırlardaydı. Detaylı bir değerlendirmeden sonra laparatomi yapılarak, kolesistektomi, koledokotomi ve T-tup drenaj yapıldı. Koledoktan çok sayıda parazit çıkartıldı. Çıkartılan parazitlerden biri parazitoloji laboratuvarına gönderildi. İndirekt hemaglünitasyon testi 1/320 (+) bulundu. Sonuç olarak bu paraziter hastalığın kronik evresi tıkanma sarılığının diğer nedenleri ile kolaylıkla karışabilir. Bu nedenle Fasiola hepatika, özellikle endemik bölgelerde, tıkanma sarılığının ayırıcı tanısı yapılırken akılda tutulmalıdır.

Anahtar kelimeler: Fasiola hepatika, tıkanma sarılığı, endemik hayvan hastalığı

INTRODUCTION

Fascioliasis, caused by *Fasciola hepatica,* is a zoonotic disease that is seen rarely in Turkey and neighboring countries. It is most common in Africa, Western Europe and Latin America. It is thought that approximately 2.4 million people are infected by fascioliasis and 180 million are at risk of

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infection (2,4,5). Human hepatobiliary infection with *F. hepatica* has two phases: the acute (hepatic) and chronic (biliary) phases. The signs and symptoms of fascioliasis differ in the two phases. While fever, abdominal pain, headache, pruritus, urticaria, weight loss, and eosinophilia are seen in the acute phase, the chronic phase is usually asymptomatic (2-5). However, *F. hepatica* may occasionally cause extrahepatic obstruction and cholestasis (8-12). In our study, we present a case of fascioliasis that was difficult to differentiate from cholangiocarcinoma.

CASE REPORT

A 47-year-old woman from Eastern Turkey presented with fever, right upper quadrant abdominal pain, and jaundice. Total bilirubin was 4.2 mg/dl, aspartate aminotransferase (AST) 55 IU/L, alanine aminotransferase (ALT) 65 IU/L, alkaline phosphatase 325 IU/L, and gamma-glutamyl transpeptidase 172 IU/L. All tumor markers including CEA and CA19-9 were in normal values.

Ultrasonography revealed intrahepatic duct dilatation of the left hepatic lobe, and on magnetic resonance imaging (MRI), a filling defect was detected at the left main hepatic duct (Figure 1). After extended evaluation, an exploratory laparotomy with cholecystectomy and choledochoduodenostomy was performed. Multiple flukes were removed from the choledochus wall (Figure 2) and biopsies were taken for frozen section for excluding neoplasm; there was no evidence of malignancy. The bile ducts were irrigated with saline and a T-tube applied. The postoperative course was uneventful; one of the parasites was sent to the parasitological clinic for identification. The result of an indirect hemagglutination test for *F. hepatica* was 1/320 (+).

Our patient was given triclabendazole to eradicate the parasite with a single oral dose of 10 mg/kg, and repeat fecal examination was done in order to confirm the eradication. There were no side effects, and a single dose proved to be effective in eradicating the parasite. Her symptoms and the eosinophilia disappeared, other laboratory values returned to normal, and no eggs were detected in the feces or duodenal aspirate six months after treatment.

DISCUSSION

Fasciola hepatica is an endemic zoonotic disease in Turkey. In recent studies from Eastern Turkey,

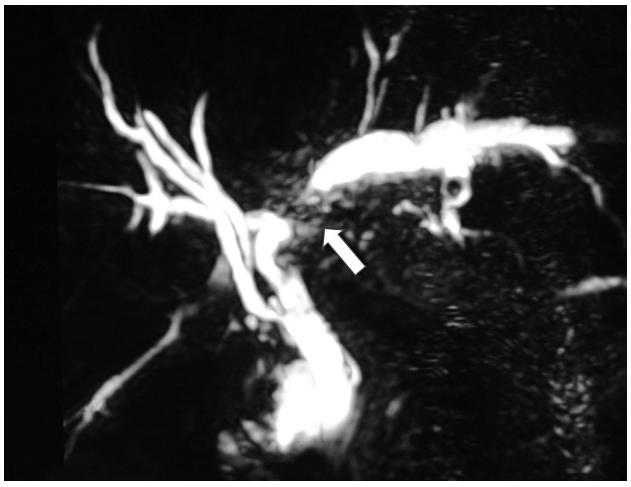

Figure 1. A white arrow shows bile duct dilatation due to filling defect at the left main hepatic duct.

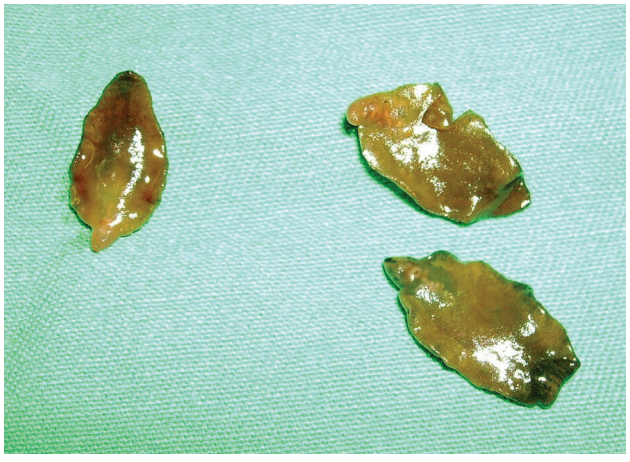

Figure 2. Multiple Flukes, which are taken out of the choledoccus.

Turhan (1) and Ozturhan et al. (2) reported the seroprevalence of fascioliasis as 2.8% (1,2). In the life cycle of the Fasciola, the usual definitive host is the sheep; humans are accidental hosts. There are two disease stages: the hepatic (acute) and biliary (chronic) stages. Infection is usually by ingestion of water or raw aquatic vegetables contaminated with the metacercariae form of *F. hepatica* (3,5-7). After ingestion, larvae excyst in the small intestine, penetrate through the wall into the peritoneal cavity, and after six to nine weeks, migrate into the liver. The young flukes then penetrate the intrahepatic bile ducts, grow in the biliary system and produce eggs. Mature *F. hepatica* flukes penetrating the bile duct are usually leaf-shaped and measure about 30x13 mm. In humans, maturation and excretion of the eggs takes about 3-4

months. In the acute phase, the patient may have prolonged fever, right upper quadrant pain, hepatomegaly, and eosinophilia (2-5), symptoms which can also present in the chronic phase. When the flukes enter the bile ducts, the symptoms of cholestasis and cholangitis may present (8-11), which can easily be misdiagnosed as obstructive jaundice of other causes of (12,13).

The biliary stage of *F. hepatica* infestation usually results in eosinophilia (2-5). Sezgin et al. (3) determined eosinophilia in five of nine patients. Ozturhan et al. (2) reported that 43% of seropositive patients had eosinophilia. Our re-examination of the laboratory values revealed mildly elevated eosinophils (23%). However, the diagnosis is based on microscopic identification of the characteristic eggs in the feces (13); stool examination is not sufficient for the diagnosis (2,3). In recent study from Turkey, Ozturhan et al. (2) found *F. hepatica* eggs in the stool in only one of seven cases who were seropositive (2). Immunoserological tests are valuable in the early hepatic stage, but enzyme linked immunosorbent assay (ELISA) testing is more rapid and reliable (14,15). The typical radiological imaging of fascioliasis is more likely to be correctly reported by radiologists in areas in which the disease is prevalent (16,17). The infection can be shown more easily by endoscopic retrograde cholangiopancreatography (ERCP), during which a filling defect can be identified in the common bile duct. After sphincterotomy and balloon extraction, live organisms can be physically removed (3,18,19). In our case, magnetic resonance cholangiography revealed a lesion in the left hepatic duct, and thus we did not perform ERCP for the diagnosis and treatment. We performed exploratory laparotomy with cholecystectomy and choledochoduodenostomy. Multiple flukes were removed from the choledochus wall and biopsies were taken for exclusion of neoplasm. There was no evidence of malignancy. The bile ducts were irrigated with saline and a T-tube applied.

Several drugs can be used during the hepatic stage. As with other liver flukes, *F. hepatica* is treated with praziquantel, bithionol or triclabendazole. Bithionol in the recommended dose of 30-50 mg/kg every other day for 10-15 doses or repeated doses is effective. Our patient was given triclabendazole, another safe drug for fascioliasis, which can eradicate the parasite with a single oral dose of 10 mg/kg (15,20-22). Consecutive fecal examination was done in order to confirm eradication. There were no side effects, and a single treatment dose was effective; the symptoms and eosinophilia disappeared (4.3%), other laboratory values returned to normal, and no eggs were detected in the feces or duodenal aspiration six months after therapy.

In conclusion, the chronic phase of this zoonotic infection can easily be misdiagnosed as any other cause of obstructive jaundice. Thus, F. hepatica should be considered in the differential diagnosis of common bile duct obstruction, especially in endemic areas.

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