Caso clinico

Case reports

An incidental case of biliary fascioliasis mimicking cholangiocellular carcinoma

Un caso accidentale di fascioliasi biliare che simula un carcinoma colangiocellulare

Ebubekir Şenateş[1], Abdullah Doğan[2], Banu Erkalma Şenateş[3], Erdal Bodakçi[2], Muhammet Bekçibaşi[4]

[1]Dicle University Medical Faculty, Department of Gastroenterology, Diyarbakir, Turkey;
[2]Dicle University Medical Faculty, Department of Internal Medicine, Diyarbakir, Turkey;
[3]Diyarbakir Education and Research Hospital, Department of Internal Medicine, Diyarbakir, Turkey;
[4]Dicle University Medical Faculty, Department of Infectious Diseases, Diyarbakir, Turkey

INTRODUCTION

Human fascioliasis is now a rare zoonotic disease caused by *Fasciola hepatica* and *Fasciola gigantica* [1]. In certain countries of South America (especially Bolivia), the Middle East (especially Egypt, Turkey), and Far East, the disease is still endemic [2]. The two phases of human fascioliasis are defined as the acute and chronic phases. In the acute phase, which involves hepatic invasion, abdominal pain, mild fever, hepatomegaly, and sometimes eosinophilia, occur. The biliary phase, characterized by cholestasis and cholangitis symptoms, is the chronic phase [3]. The diagnosis is typically based on clinical data, the presence of ova or parasites in feces or duodenal aspirates, the results of serological tests, and pathological findings [4]. Because of the non-specific disease symptoms and findings, the diagnosis and treatment are commonly delayed. Indeed, because these symptoms and findings may also be seen in other hepatobiliary system diseases, for accurate and timely diagnosis, radiology and invasive techniques are useful, as are non-invasive methods [5].

CASE REPORT

A 44-year-old female patient was admitted to our hospital with nausea, loss of appetite, weight loss, and intermittent abdominal pain over the previous 3 years. Physical examination revealed a temperature of 36.7°C, a pulse of 85/min, blood pressure of 110/70 mmHg, and minimal tenderness in the right upper quadrant. Her laboratory results revealed the following: hemoglobin 12 g/dL, hematocrit 37%, WBC 5,150 mm^3, platelets 221,000/mm^3 (eosinophils 7%), erythrocyte sedimentation rate (ESR) 19 mm/h, alanine aminotransferase (ALT) 16 IU/L, aspartate aminotransferase (AST) 15 IU/L, alkaline phosphatase (ALP) 53 IU/L, gamma glutamyltransferase (GGT) 17 IU/L, total bilirubin 0.4 mg/dL, direct bilirubin 0.15 mg/dL, indirect bilirubin 0.25 mg/dL, and lactate dehydrogenase (LDH) 175 IU/L. Abdominal ultrasound revealed wall thickening in the main hepatic duct and common bile duct, reaching 3.5 mm, and soft tissue echogenicity was observed in the lumen of the common bile duct. Because the patient presented with loss of appetite and weight loss, initially, cholangiocellular carcinoma was suspected. However, tumor markers were CEA 1.03 (negative), CA-19-9 7.5 (negative), CA-125 13.2 (negative), and AFP 1 (negative). Abdominal computed tomography revealed that the common bile duct was dilated (8.5 mm), its wall thickness was increased, and a hyperdense appearance was detected in the lumen of the common bile duct. Then, magnetic resonance cholangiopancreatography (MRCP) was performed. The findings showed intrahepatic bile duct (IHBD) dilatation with hydropic gallbladder, involvement of the proximal common bile duct wall with

Corresponding author
Ebubekir Şenateş
E-mail: ebubekirsenates@yahoo.com

contrast substance, and a filling defect in the common bile duct lumen. Thus, a malignancy could not be ruled out. Colonoscopy findings were normal. Then, ERCP was performed: the common bile duct and the IHBD were dilated and a linear filling defect in the common bile duct was detected (Figure 1). An endoscopic sphincterotomy was performed and the common bile duct was swept with a balloon. Surprisingly, eight adult *F. hepatica* were extracted consecutively (Figure 2). After removal of the parasites from the common bile duct, the patient was treated with a single dose of 10 mg/kg triclabendazole. After treatment, the patient's complaints resolved. After 2 months, abdominal ultrasonographic findings were all normal (there was no dilatation of the intrahepatic bile ducts, the gallbladder was of normal size, and the common bile ducts appeared normal). Laboratory findings also returned to normal.

DISCUSSION

Fasciola passes to people through contaminated fresh water plants. *F. hepatica* and *F. gigantica* are the most common type. The parasite's larvae penetrate the intestinal wall to enter the peritoneal cavity. In the peritoneal cavity, larvae develop into the juvenile form. They then usually pass through the liver capsule into hepatic tissues where, after reaching adulthood, they finally invade the biliary tract [2]. The diagnosis of fascioliasis can be missed due to non-specific findings, such as mild hepatomegaly, abdominal pain, and fever during the acute stage. Patients may present with cholangitis and cholestasis symptoms and signs due to obstruction of the bile duct during the chronic phase [3]. The most common laboratory findings are elevation of liver enzymes (ALT, AST) and cholestasis enzymes (ALP, GGT) [6]. However, some patients even in the chronic phase may remain asymptomatic [7]. In our case, although suffering from chronic biliary fascioliasis, the cholestasis enzymes and bilirubin were normal, there was no fever, and CRP and ESR were normal. There was no clinical cholangitis. The patient's complaints and physical examination findings were not sufficiently specific to bring the illness to mind. Initially, peripheral eosinophilia may be detected in patients infected with Fasciola [8]. However, in our case, the peripheral blood eosinophil count was normal. As noted in the literature, hepatomegaly and fever are common findings [9].

Figure 1 - Dilated common bile duct and IHBD, linear filling defects.

While our patient had mild abdominal pain, there was no fever or hepatomegaly. Although the definitive diagnosis of fascioliasis can be made by detecting the eggs of the parasite in the stool or duodenal aspirates, the egg detection rate is not high because of the low egg production rate of the parasite, so it is not an effective or convenient method [9]. Despite high specificity for a diagnosis, serological tests are not routinely performed in non-endemic areas. ELISA tests give faster and reliable results [9]. In our case, because of the initial suspicion of a malignancy, no stool examination or serological tests were

Figure 2 - Extracted adult Fasciola hepatica.

performed. In one study, it was suggested that a dilated common bile duct and the determination of material that did not have an isoechoic shadow in the bile duct with ultrasound might be specific for fascioliasis [10]. In our case, with abdominal ultrasound, in the hepatic duct and common bile duct, there was wall thickening, to ~4 mm, and this was considered to favor a malignancy. For imaging the biliary tract, ultrasonography, CT, and MRCP were used, but ERCP remains the gold standard, because USG and CT findings can sometimes be confused with malignancies and stone diseases [11, 12]. In a recent review article, *F. hepatica* and some other helminth infections were reported to mimic different malignancies [13]. Indeed, in our case, the bile duct wall thickening and contrast enhancement of the bile duct wall image with abdominal CT and MRCP strengthened the suspicion of malignancy. The common bile duct and IHBD were dilated and there were linear filling defects in the common bile duct on ERCP (Figure 1). At the end of the process, eight adult *Fasciola hepatica* were extracted from the common bile duct (Figure 2).

In conclusion, in patients who present with non-specific complaints, such as abdominal pain, nausea, fever, and weight loss, if there is common bile duct dilatation and wall thickening, before making further investigations, especially in endemic areas, the differential diagnosis of *Fasciola hepatica* should be considered, and serological tests would be wise. For both timely diagnosis and providing treatment, ERCP should be performed in these cases [14].

Keywords: *Fasciola hepatica*, ERCP, triclabendazole, cholangiocellular carcinoma.

SUMMARY

Fascioliasis is a zoonotic infestation caused by *Fasciola hepatica* that usually attacks mammals, such as goats, sheep and cattle. The parasites can infect humans via freshwater plants contaminated with encysted metacercariae. In the acute phase, which involves hepatic invasion, the disease may present with abdominal pain, mild fever and hepatomegaly. In the chronic phase, the parasites settle into the biliary tracts, and then cause cholangitis and cholestasis. Sometimes, the disease may mimic malignancies, creating a mass appearance. Endoscopic retrograde cholangiopancreatography (ERCP) is an important diagnostic and treatment method because it allows simultaneous diagnosis and treatment. Here, we present a 44-year-old female patient who presented to our hospital with complaints of abdominal pain, nausea, vomiting, anorexia and weight loss. After diagnostic investigations with laboratory and imaging methods, she was initially hospitalized with a preliminary diagnosis of cholangiocellular carcinoma (CCC). However, after a full work-up, the patient was diagnosed with *Fasciola hepatica* via ERCP and parasites were extracted with ERCP at the same time and then treated with a single dose of triclabendazole 10 mg/kg. Two months later, the clinical status of the patient had improved markedly, with resolution of all symptoms and all laboratory and imaging tests returning to within normal limits.

RIASSUNTO

La fascioliasi è una infestazione zoonotica causata da Fasciola hepatica *che abitualmente colpisce mammiferi quali capre, pecore e bovini. I parassiti possono infettare l'uomo attraverso l'ingestione di ortaggi contaminati (irrigati con acque infestate) da metacercarie incistate. Nella fase acuta, di invasione epatica, la malattia può manifestarsi con dolore addominale, febbre di lieve entità ed epatomegalia. Nella fase cronica, i parassiti si insediano nelle vie biliari provocando colangite e colestasi. In taluni casi, la malattia può mimare patologie maligne, determinando l'aspetto di una massa. La colangio-pancreatografia endoscopica retrograda (ERCP) è uno strumento considerevole sia per la diagnosi sia per il trattamento in quanto consente di effettuare entrambi, simultaneamente. In questo articolo presentiamo il caso di una paziente di 44 anni giunta alla nostra osservazione lamentando dolore addominale, nausea, vomito, anoressia e calo di peso. In seguito a diverse indagini di laboratorio e tecniche per immagine, la donna veniva inizialmente ricoverata con diagnosi preliminare di carcinoma colangiocellulare (CCC). Tuttavia, successivamente a un controllo completo, la ERCP consentiva di porre diagnosi di infestazione da* Fasciola hepatica *e, nel contempo, di estrarre i parassiti. La paziente è stata quindi trattata con una dose singola di 10 mg/kg di triclabendazolo. Dopo due mesi, le condizioni cliniche della paziente erano notevolmente migliorate, con risoluzione di tutti i sintomi e ritorno ai valori normali degli indici di laboratorio e delle indagini per immagine.*

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