

CASE REPORT

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Ectopic intra-abdominal fascioliasis

Abstract: Human fascioliasis, caused by *Fasciola hepatica*, is emerging as an important chronic zoonotic disease in many areas of the world, including Turkey. It primarily involves the liver and may also cause severe damage in the tissue. Herein we report on a patient with ectopic intraabdominal fascioliasis that presented to our clinic with abdominal pain and distention. Physical and radiological examination as well as an exploratory laparotomy revealed a 10×10 -cm mass in the splenic flexura of the colon and multiple cystic formations in the liver, spleen, and omentum, which mimicked peritoneal carcinoma. Histopathological examination revealed a granulomatous reaction due to parasites. Left hemicolectomy and partial omental resection were performed. Subsequent serological and histological examinations resulted in the diagnosis of fascioliasis. Laparotomy and treatment with triclabendazole were well tolerated by the patient. Ectopic cases of fascioliasis cannot be easily distinguished from peritoneal carcinoma involving the colon, mesocolon, and omentum, and physicians must therefore be aware of this form of infestation.

Key words: Ectopic fascioliasis, Fasciola hepatica, Turkey, clinic, diagnosis, treatment

Ektopik İntraabdominal Fascioliasis

Özet: Fasciola hepatica'nın neden olduğu insan fascioliasisi; son yıllarda dikkat çeken, Türkiye de dâhil dünyanın birçok yerinde görülen önemli bir kronik zoonotik hastalıktır. Genellikle karaciğeri etkiler ve dokuda ciddi hasara yol açabilir. Çalışmamızda kliniğimize karın ağrısı ve distansiyon ile gelen bir hasta rapor edilmiştir. Fiziksel ve radyolojik tanı yanısıra eksplaratuvar laparotomy uygulanmış, kolonun splenik fleksurasında 10 × 10 cm boyutlarında bir kitle ve karaciğer, dalak ve omentumda peritoneal karsinomayı andıran multipl kistik oluşumlar saptanmıştır. Histopatolojik incelemede parazitlere bağlı granulamatoz reaksiyon belirlenmiştir. Sol hemikolektomi ve parsiyel omental rezeksiyon uygulanmıştır. Ardı sıra fascioliasis tanısına yönelik serolojik ve histolojik incelemeler yapılmıştır. Laparotomi ve triklabendazol ile tedavi hasta tarafından iyi tolere edilmiştir. Ektopik fascioliasis olgularının kolon, mezokolon ve omentumu kapsayan peritoneal karsinomadan ayırt edilmesi kolay değildir. Bu nedenle hekimler bu tür enfestasyonlar hakkında duyarlı olmalıdır.

Anahtar sözcükler: Ektopik fascioliasis, Fasciola hepatica, Türkiye, klinik tanı, tedavi

Introduction

Fasciola hepatica is a common helminthic parasite of ruminants, especially sheep, goats, and cattle. In recent years the public health importance of fascioliasis has increased. The parasite is widespread in some European countries, including Turkey, where some regions are considered hypo- or mesoendemic areas (1-4).

The major findings of human fascioliasis are liver granuloma and biliary obstruction caused by adult worms in the bile ducts, and in heavy infections death may occur (2). Juvenile worms frequently migrate to extrahepatic organs, mainly in abdominal organs and subcutaneous tissues.

Although fascioliasis is a well-known human parasite, diagnosis of the disease is quite problematic. In Turkey, between 1932 and 1999 more than 50 cases were detected accidentally by finding adult or juvenile worms during surgical procedures or, more rarely, by parasitological methods, e.g. detection of eggs in the stool or bile (2). With the development of more sensitive serological and radiological tests, and due to increased awareness by physicians, an increase in the number of reported cases from Turkey has been observed (1,4-6). Between 1998 and 2003, 53 cases were reported in the city of Antalya alone (3).

We present a very rare case of ectopic intraabdominal fascioliasis that mimicked peritonitis and carcinoma, involving the colon, mesocolon, and omentum.

Case report

A 46-year-old woman from Kırıkkale, a rural area in central Anatolia, was admitted to the hospital in August 2006 with diffuse abdominal pain, abdominal distention, and malaise. Her symptoms at that time, i.e. abdominal pain, nausea and vomiting, had started 2 years earlier when she lost about 12 kg within 18 months. She used to consume watercress.

The physical examination revealed abdominal tenderness and a mass on her upper left quadrant. Laboratory values were as follows: hemoglobin: 10.9 g dL⁻¹ (13.2-17.3); hematocrit: 32.1% (39-49); white blood cell count: 5700 (4500-11,000); eosinophil count: 1.2% (0-3); erythrocyte sedimentation rate: 83 mm h⁻¹ (0-21); alkaline phosphatase (ALP): 99 U L⁻¹ (30-120); aspartate aminotransferase (AST): 19 U L⁻¹ (0-35); alanine aminotransferase (ALT): 12 U L⁻¹ (0-45); gamma glutamyl transferase (GGT): 22.3 U L⁻¹ (0-55).

Ultrasonography (US) and computed tomography (CT) scans of the abdomen showed the presence of an approximately 84×63 -mm amorphous cystic lesion. The lesion was located between the pancreas and spleen, and exteriorly exhibited a heterogeneous density, while its central part was hypodense. This growth was spread over parts of the stomach, spleen, and left kidney. Its borderline could not be distinguished from the wall of the splenic flexure of the colon (Figure 1). In the liver, multiple cystic structures up to 4×4 mm could be seen. In addition, a 15×13 -mm cyst was visible in the parenchyma of the spleen. Mild dilatation and wall thickening were observed in the biliary system. Both kidneys had multiple cystic formations.



Figure 1. Appearance of the mass in the colon.

Repeated laboratory examinations did not show the presence of parasites or their eggs, or blood in the stool. Indirect hemagglutination assay (IHA) and RAST-ELISA tests were negative for echinococcosis.

In the search for a potential malignancy, diagnostic laparotomy was performed, which revealed the presence of a 10×10 -cm structure on the splenic flexure of the colon that invaded the surrounding tissues, as well as multiple grayish-white miliary nodules (approximately 0.5-1 cm in diameter). Because the clinical picture resembled a metastasis located in the abdomen, omentum, and mesocolon, specimens multiple biopsy were taken. Histopathologic examination of the specimens showed granulomatous inflammation due to parasites, as well as lymphoid hyperplasia. Because the mass caused obstruction and compression, urgent excision was required. Thereafter, a subtotal left hemicolectomy and a partial omental resection were performed.

Histopathological examination of the specimens showed the presence of a colonic granulomatous lesion due to *Fasciola hepatica* (Figures 2-4). The results were serologically confirmed by IHA (Distomatose Fumouze IHA, France), with an antibody titer against fascioliasis above 1/1280 (normal values are less than 1/160). The results of other serologic tests for amebiasis and toxocariasis were negative. No complications were observed after the operation; however, liver function test results were slightly different: ALP (279 U L⁻¹) and GGT (225 U L⁻¹) increased, and ALT (12 U L⁻¹) and AST (17 U L⁻¹)



Figure 2. Macroscopic appearance of Fasciola hepatica granulomas on colon wall (marked with arrows).



Figure 3. Microscopic view. 40×, stained with Hematoxylin-Eosin, relationship between *Fasciola hepatica* granulomas and bowel wall.

were normal. Postoperative colonoscopy revealed no mucosal pathology; however, chronic colitis and mild epithelial dysplasia were visible. The patient was discharged without the use of antiparasitic medications because of increased liver function test results.

Regular postoperative follow-up was performed and 40 days postoperatively the liver function test results were nearly normal: ALP: 108 U L⁻¹, ALT 43 U L⁻¹, AST 26 U L⁻¹, and GGT 102 U L⁻¹. Three months later the patient was free of clinical symptoms; the antibody titer for fascioliasis IHA was ca. 1/320 and repeated stool examinations for parasites were negative. Despite the lack of clinical symptoms and



Figure 4. Appearance of *Fasciola hepatica* egg in our case by immersion microscopy, stained with Hematoxylin-Eosin 100×.

decrease in antibody level, triclabendazole (10 mg kg⁻¹) was prescribed for the patient as soon as her liver function test results were normal. Only one course of therapy was administered to the patient.

Discussion

Human *F. hepatica* infestation is reported worldwide. The disease has been reported in 19 European countries, including Turkey (2). In a recent study from the eastern part of Turkey, fascioliasis seroprevalence was 2.8% in the survey population and was independent of age, and educational and socioeconomic status (1). The diagnosis of fascioliasis is complicated by the fact that physicians rarely encounter this disease. Due to the different manifestations of fascioliasis— depending on the clinical stage of the disease and location of the parasite—it is difficult to describe specific clinical symptoms. In the initial phase of the disease, which lasts 1-3 months after infestation with metacercariae, the symptoms include epigastric pain (57%), fatigue (46%), fever (43%), and right quadrant abdominal pain (36%) (3).

Patients may also show extrahepatic abnormalities, such as pulmonary infiltrates, pleuropericarditis, meningitis, and lymphadenopathy (2). After the fluke reaches the lumen of the bile ducts, the biliary or latent phase of the disease begins and eggs can be found in the stool. The adult fluke may produce obstructive jaundice or predispose to cholecystitis and cholelithiasis; therefore, human fascioliasis should be considered in all cases of cholangitis (7).

The diagnosis of a fascioliasis usually fails due to the difficulty in finding eggs in the stool (8). The high prevalence of fascioliasis among patients with eosinophilia highlights the importance of this sign (9).

The fact that our patient had a chronic form of fascioliasis and ectopic location of the parasite may explain the lack of blood eosinophilia and ova in the stool. In the chronic phase of fascioliasis only 43.5% of children had eosinophilia, and as the sedimentation rate was normalizing anemia increased (2,9). Our patient also had anemia and an increased sedimentation rate, which may have been related to the existing diffuse hemorrhage due to infiltration of the tissues.

Abnormal liver function test results are characteristic of fascioliasis. In the chronic phase, while an increase in ALP and GGT indicates cholestasis, an increase in ALT and AST is a sign of tissue damage (2). In our case, normal ALT and AST values increased slightly after surgery, which most probably indicated the chronic phase of infestation, rather than tissue damage caused by surgery.

US is useful in the diagnosis of fascioliasis in the biliary stage, while CT scanning is the most useful technique in the diagnosis of biliary and ectopic stages. MRI is widely used for confirmation and follow-up of the disease. Invasive techniques, such as percutaneous cholangiography, endoscopic retrograde cholangiography, and liver biopsy could aid in the diagnosis, but are not essential (2). With CT, single or multiple hypodense areas caused by deposition of the parasite (also seen in our case) and tunnel-like branching caused by migration of the parasite through the liver are highly suggestive of fascioliasis. CT and US can also be helpful in evaluating the response to treatment (10).

Immunoserological tests have become the cornerstone for the diagnosis of fascioliasis, especially in the early stage or in cases of ectopic infection. ELISA and IHA are rapid and sensitive methods for detecting antibodies against the excretory-secretory antigens of *Fasciola* (1,2,4-6,9).

Ectopic infestations of the parasite often occur in abdominal organs and subcutaneous tissues, though rarely in the lungs, heart, or brain (2).Histopathological examination is crucial for fascioliasis, especially for ectopic and laparoscopically observed cases. Granulomatous lesions of fascioliasis are quite distinct from those of tuberculosis or sarcoidosis, as they are characterized by a predominance of eosinophils, and the lack of lymphocytes and giant cells. Microscopic examination of nodules reveals necrosis, eosinophilic infiltration, hemorrhage, and tunnels within which the parasite may be observed (2). During explorative laparotomy, multiple grayish white miliary nodules were observed in our patient, which resembled metastases of peritoneal carcinoma. After histopathological examination and serological testing the diagnosis of fascioliasis was confirmed.

Therapy for fascioliasis has been difficult, both in terms of efficacy and toxicity. Triclabendazole is a highly efficient fasciolicide for use in the acute and chronic phases of infestation. A dosage of 12 mg kg⁻¹ daily for 1-2 days has been reported to be effective in humans, and no side-effects were observed (2). In our case, the patient was treated with triclabendazole in order to prevent an eventual recurrence of the parasite and was well tolerated.

In conclusion, fascioliasis should be considered in patients with abdominal growths, especially where *Fasciola* is endemic. As clinical and laboratory findings of fascioliasis may easily be confused with those of other conditions, correct diagnosis is crucial. Both serological and imaging techniques can be helpful in diagnosing and monitoring the disease.

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