CASE REPORT

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A case of ectopic intraabdominal fascioliasis presented with acute abdomen

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Human fascioliasis with Fasciola species occurs worldwide and is most common among rural people who tend sheep and eat uncooked water vegetables, particularly watercress. The natural history of the acute phase begins with ingestion of metacercariae encysted on various kinds of aquatic vegetation such as watercress. Fascioliasis primarily involves the liver, bile ducts, gallbladder, and occasionally ectopic sites. We describe herein a case of ectopic fascioliasis. This uncommon form of disease was peritonitis; both visceral and parietal peritoneal layers were affected with the formation of multiple nodules and ascites.

Key words: Fascioliasis, ectopic, child, acute abdomen

Akut karın tablosu ile başvuran bir ektopik intraabdominal fasioliazis

Fasciola türleri ile fasioliazis tüm dünyada ortaya çıkar ve koyun bakan kırsal bölge insanlarında ve pişmemiş su sebzeleri, özellikle su teresi yiyenlerde daha sıktır. Akut dönemin doğal öyküsü, su teresi gibi çeşitli su bitkilerinin üzerindeki enkiste metacercariae yutulmasıyla başlar. Fasioliazis esas olarak karaciğeri, safra kanalları ve safra kesesini, nadiren ektopik bölgeleri tutar. Biz, bir ektopik fasioliazis olgusunu sunduk. Hastalığın bu alışılmamış formu, multipl nodüller ve assit oluşumu ile giden, hem visseral, hem paryetal tabakaları tutan peritonitti.

Anahtar kelimeler: Fasioliazis, ektopik, çocuk, akut karın

INTRODUCTION

Fascioliasis is a parasitic disease caused by the trematode liver flukes *Fasciola hepatica* and *Fasciola gigantica*. It has been reported worldwide. The fluke infects primarily sheep, goats and cattle. Humans are accidental hosts. Infection occurs due to ingestion of water or raw aquatic plants contaminated with metacercariae (2). Metacercaria exists in the intestine, perforates the intestinal wall, enters the peritoneum, and passes through the liver capsule to enter the biliary tree. In the biliary tract, the mature fluke releases eggs, which are once again excreted in feces to complete the life cycle. It primarily involves the liver but may also cause extrabiliary manifestations (3). In this report, we present a case with a diffuse peritoneal and omental fascioliasis as an unexpected manifestation of the fasciola infection.

CASE REPORT

A previously healthy six–year-old boy, whose family were seasonal workers, was admitted to the hospital following two days of increasing intense abdominal pain with fever, diarrhea and a 2-kilogram weight loss within 10 days. Physical examina-

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tion revealed fever (38.7°C), diffuse abdominal tenderness and rigidity. The remaining physical findings were normal. Laboratory investigations were as follows: white blood cells (WBC): 8900/mm^3; eosinophil 12%; erythrocyte sedimentation rate: 120 mm/hour; and C-reactive protein: 113 mg/L; serum alanine aminotransferase, aspartate aminotransferase, alkaline phosphatase, and gamma glutamyl transferase levels were normal. Parasitological investigation of his stool was negative. A plain abdominal X-ray showed air fluid level in the right lower quadrant. An abdominal ultrasound examination showed intra-abdominal fluid collection. An exploratory laparotomy was performed by a right paramedian infraumbilical incision. During the exploration, a fragile and thickened omentum widely adhered to the abdominal wall (Figure 1) and a large amount of serous intra-abdominal fluid were found. Nodular lesions were detected on the serosal surface of the bowel and all peritoneal surfaces (Figure 2). Based on this finding, incisional biopsy of the omental and peritoneal lesions was performed. After irrigation of the abdominal cavity with warm saline, the abdominal wall was closed. Oral feeding was started on the second postoperative day. The postoperative course was uneventful. Histopathologic examination of the specimens showed granulomatous lesions containing multinuclear giant cells and histiocytes. The leading infectious and noninfectious causes of granulomatous inflammation were investigated. Tuberculin skin test of the patient, who had a visible BCG scar, was 7 mm. Chest radiography and thorax computerized tomography (CT) were normal. The results of serologic tests for cytomegalovirus, Epstein-Barr virus, human immunodeficiency virus, salmonella, and brucella were negative. Quantitative serum immunoglobulin (Ig) levels were as follows: IgA 269 mg/dl (normal: 68.7-332 mg/dl), IgM 322 mg/dl (normal: 69-345 mg/dl), IgG 3180 mg/dl (normal: 701-2140 mg/dl), and IgE 1300 U/ml (normal: 0-90 U/ml). Isohemagglutinin titers were found to be positive, at a higher titer of 1/1024. Nitroblue tetrazolium test and dihydrorhodamine assay were normal for chronic granulomatous disease. Antinuclear antibody and anti dsDNA were negative, perinuclear antineutrophilic cytoplasmic antibody (p-ANCA) and cytoplasmic ANCA (c-ANCA) were positive, and rheumatoid factor (RF) was 26.9 (normal <20). Because of the increased serum IgE and eosinophilia, Fasciola indirect hemagglutination (IHA) test was performed and was found to be positive at a titer of 1/1280. The results of other serologic tests for amebiasis and toxocariasis were negative. Triclabendazole at a dose of 12 mg/kg was given to the patient per orally with a 12-hour interval. Eosinophilia and positivity of p-ANCA, c-ANCA and RF ameliorated after one week of treatment. Increased titers of serum IgE levels normalized thereafter. Three months later the patient was free of clinical symptoms, and the antibody titer for fascioliasis IHA was 1/160.

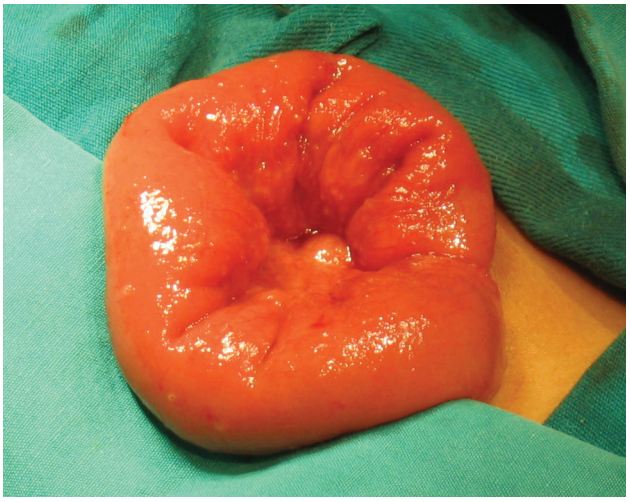

Figure 1. Nodules in the fragile and thickened greater omentum.

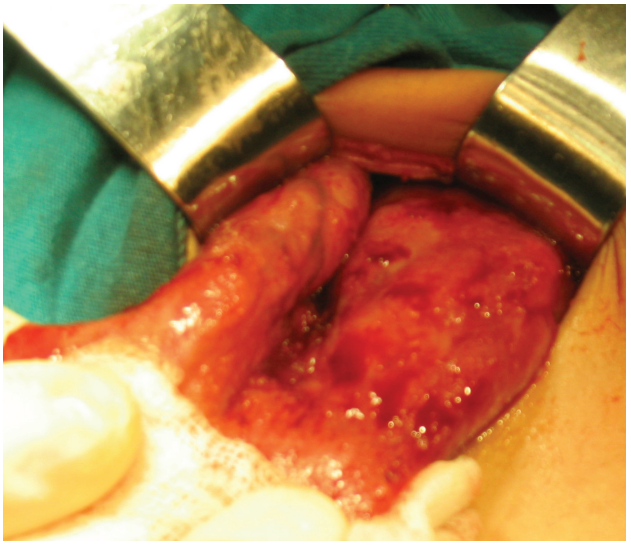

Figure 2. Nodular lesions on the serosal surface of the bowel.

DISCUSSION

Fascioliasis is a widespread disease of sheep caused by the liver fluke *Fasciola hepatica,* and less commonly by *Fasciola gigantica,* the life cycles

and clinical presentations of which are similar (1). Human fascioliasis can be divided by phases of the disease as acute and chronic according to the duration of the symptoms and the ultrasonographic findings. If the duration of symptoms is <4 months and there are no motile echogenic images in the gallbladder on admission, it is classified as acute. If symptoms persist for >4 months or there are motile echogenic images in the gallbladder, it is classified as chronic. The acute or hepatic phase of the illness occurs when the organism perforates the liver capsule and begins to migrate towards the biliary system. The acute phase is characterized by fever, abdominal pain, headache, pruritus, urticaria, weight loss, and eosinophilia; however, 15% of the patients are asymptomatic (1,2,4-6). The acute phase usually lasts for 3 months after ingestion of the metacercaria, after which, as the parasite enters the bile canaliculi, the symptoms may decline or disappear completely. Because the parasites cannot produce eggs before invasion of the biliary tract, stool studies for ova during the acute phase are unhelpful (6). This case had acute symptoms and negative stool examinations. Serological tests for *F. hepatica* are useful in establishing an early diagnosis (6,7). The chronic phase is usually asymptomatic, and the prominent sign may only be intermittent cholangitis. The definitive diagnosis of fascioliasis in this phase is based on the presence of Fasciola eggs in a stool or gallbladder sample, or on a positive serological test plus radiological findings indicating fascioliasis (6).

The manifestations of fascioliasis depend not only on the clinical stage of the disease but also the location of the parasite. While adult worms cause liver granuloma and biliary obstruction, juvenile worms may migrate to extrahepatic organs, mainly abdominal organs and subcutaneous tissues (8). The migratory route of *F. hepatica* in a human host helps to explain the ectopic location of the fluke in our case. A juvenile fluke located in the peritoneal cavity, instead of penetrating into the liver, might have deviated in its course towards the bowel, and reached the intestinal wall by direct migration. Many cases of ectopic fascioliasis have been reported in the literature, and ectopic sites include subcutaneous tissue, lung, heart, eye, brain, stomach, cecum, epididymis, and lymph nodes; however, the peritoneal cavity, omentum and intestinal wall are extremely rare sites of ectopic fascioliasis (9,10). Ectopic fascioliasis can cause various clinical symptoms. Ultrasonography, CT scans and magnetic resonance imaging are complementary modalities (3,5,11). Histopathological examination is crucial for fascioliasis, especially for ectopic cases (8). The biopsy in this case revealed granulomatous inflammation.

Eosinophilia is the predominant laboratory finding, especially in patients with the acute form of the disease. Eosinophilia may be absent in half of the chronic patients, so normal eosinophil count does not rule out the infection (12). Our case had eosinophilia. Fascioliasis may be diagnosed during the investigation for eosinophilia detected in routine screening, or during investigation of a patient's family members, and has been classified as latent fascioliasis (6).

Interestingly, our case had c-ANCA, p-ANCA and RF seropositivity. We considered that these are related with cross-reactions because the classical features of a systemic vasculitis (skin, kidney, spleen, ophthalmic, and multisystemic neurological signs) were not present in our patient. Furthermore, these tests normalized after treatment of the fascioliasis. A case of severe vasculitis associated with *F. hepatica* infection was described previously. That case was treated with steroids without clinical response. The authors reported that after treatment with triclabendazole, all symptoms and systemic manifestations of the patient resolved within weeks (13).

Triclabendazole has been the drug of choice for treating liver fluke infections. More recently, it has been used successfully to treat human cases of fascioliasis; however, resistance may develop (14). Bithionol is an alternative drug for F. hepatica (11). We used triclabendazole in our case with success.

In conclusion, human fascioliasis is currently expanding as an important public health problem. The mode of presentation and sites of involvement of ectopic fascioliasis vary widely and are unpredictable. Although medical treatment is the mainstay of therapy in fascioliasis, the varied clinical presentations of fascioliasis remain confusing for physicians, and surgery is sometimes required for the diagnosis, as in our case. We describe a case of peritonitis; both visceral and parietal peritoneal layers were affected with the formation of multiple nodules and ascites. The present case, to the best of our knowledge, is the first case with presentation as acute abdomen in childhood as well as the first report of ectopic fascioliasis in the greater omentum in a child.

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