

Biliary fascioliasis – an uncommon cause of recurrent biliary colics: Report of a case and brief review

Biliäre Fasciolose – eine ungewöhnliche Ursache für wiederholte Gallenkoliken: Bericht eines Falles mit einer kurzen Übersicht

Abstract

Biliary parasitosis is one of the important causes of biliary obstruction in endemic areas, however due to migration and travel the disease is known to occur in non endemic zones as well. The spectrum of biliary fascioliasis ranges from recurrent biliary colics to acute cholangitis. The long term complications are gall stones, sclerosing cholangitis and biliary cirrhosis. We describe fascioliasis as a cause of recurrent biliary colics in a young male necessitating multiple hospitalizations over a period of four years. Investigative profile had been non-contributory every time he was hospitalized for his abdominal pain prior to the current presentation. He never had cholangitis due to the worm in the common bile duct. It was only at endoscopic retrograde cholangiopancreatography (ERCP) biliary fascioliasis was discovered to be the cause of his recurrent biliary colics. After removal of the live *Fasciola hepatica* from the common bile duct he became symptom free and is attending our clinic for last 11 months now. Clinical spectrum of biliary fascioliasis is discussed in this report.

Zusammenfassung

In endemischen Gebieten ist der Parasitenbefall von Gallengängen eine häufige Ursache für Gallenkoliken. Aufgrund von Migration und Reise-tätigkeiten kommen derartige Erkrankungen auch außerhalb endemischer Gebiete vor. Das Spektrum der Symptome bei Gallengangsfascio-lose erstreckt sich von wiederholten Gallenkoliken bis zur akuten Cho-langitis. Als Langzeitkomplikationen werden Gallensteine, sklerosierende Cholangitis und biliäre Zirrhose beschrieben. Wir berichten über eine Fasciolose als Ursache für Gallenkoliken bei einem jungen Mann, der über vier Jahre mehrfach wegen Gallenkoliken stationär behandelt wurde. Das Untersuchungsprofil brachte bisher jedes Mal keinen Erfolg, wenn er stationär wegen Bauchschmerzen aufgenommen wurde. Er hatte vorher niemals eine Cholangitis auf der Basis eines Befalls mit dem Leberegel. Nur durch endoskopisch retrograde Cholangiopankrea-tikographie (ERCP) wurde die biliäre Fasciolose als Ursache für die wiederholten Gallenkoliken entdeckt. Nach Entfernen der lebenden *Fasciola hepatica* aus dem Gallengang wurde der Patient, nunmehr seit 11 Monaten, symptomfrei. Das klinische Spektrum der biliären Fascio-lose wird in diesem Bericht diskutiert.

Introduction

Fascioliasis is endemic in sheep raising and cattle breeding countries but reports from non-endemic areas have been described in the literature. In a recent meta analysis by Thomas Furst et al. [1] it has been estimated that more than 2.6 million people are infected by the *Fasciola hepatica* globally with highest infestations being in Latin America, Andean, followed by North Africa, middle

east, Asia and Europe. In the same meta analysis authors reported an increase of 11% for fascioliasis over the last decade. These estimates are 35% higher than previous World Health Organization (WHO) figures [2]. The life cycle of *Fasciola hepatica* is complex and human beings are the accidental hosts. Humans get infected by eating raw vegetables especially watercress grown in sheep raising areas. Other fresh water plants like lettuce, mint, alfalfa are known to transmit the infection. Occasionally drinking

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unboiled water contaminated by metacercariae may cause the infection [3]. The eggs hatch into larvae and penetrate the gut lumen, finally migrate to the liver where they lodge into the biliary system. Adult worm is large, flat, brownish and leaf-shaped. Its size is about 2.5×1 cm. It is estimated to live for 9–13 years in the human biliary system [4]. There are three phases in the life cycle of the parasite in human beings. First phase is the hepatic phase which reflects an acute phase of infection. Second phase is the biliary phase reflecting the chronic phase and the third is the pharyngeal phase which is considered to be an ectopic phase in its life cycle. Damage to the liver is proportional to the parasitic load during hepatic phase. Endoscopic ultrasound has emerged as an important investigational tool in the biliary phase of the parasite but endoscopic retrograde cholangiopancreatography (ERCP) has the advantage of therapeutic potential in the management of fascioliasis. Biliary phase requires ERCP for management as the drugs are effective only in the hepatic phase of the parasite. Reports of cholangitis, hemobilia [5] even death caused by fascioliasis have been described in the literature [6].

Case report

A 27-year-old Egyptian male with no significant comorbidities presented with history of episodic, recurrent abdominal pain of 4 years duration to our clinic at King Abdul Aziz specialist hospital Taif, a tertiary care centre in the western region of Saudi Arabia. Patient described pain as colicky, remaining for 1–2 hours necessitating intravenous analgesics predominantly in the upper abdomen. His abdominal pain had no reference or radiation and there was no jaundice associated with it. Patient had been admitted four times in various hospitals during this period and every time basic laboratory evaluation including liver function tests and serum amylase were within normal limits. His ultrasound examination had been within normal limits on each occasion he was hospitalized for his abdominal pain. Patient denied any high risk behavior or drug abuse. Over this period he had stable appetite and constant weight. On examination he was conscious oriented and he had stable vitals. There was no icterus, or lymphadenopathy. His systemic examination was unremarkable. Laboratory data revealed normal hemogram, normal liver function tests and his abdominal ultrasound was also within normal limits. There were no eggs or ova in his stool examination. Keeping in view recurrent biliary colics magnetic resonance cholangiopancreatography (MRCP) was done which showed a doubtful filling defect in the common bile duct but intra-hepatic biliary radicals were not dilated. There were no gall stones and the rest of the viscera were within normal limits. Endoscopic retrograde cholangiopancreatography (ERCP) was undertaken which revealed normal papilla. Selective common bile duct (CBD) cannulation was done and cholangiogram revealed a filling defect in the lower end of common bile duct. There was mild dilatation of the

common bile duct (Figure 1) however, biliary radicals were not dilated. Sphincterotomy was done and CBD was swept with biliary balloon and a live *Fasciola hepatica* was seen coming out of the common bile duct (Figure 2, Figure 3). Later using biopsy forceps the worm was taken out of duodenum and confirmation of the species, *Fasciola hepatica*, was made by the microbiology department of the hospital. Patient was given two tablets of triclabendazole 250 mg (manufactured by Novartis) after the procedure. Following therapeutic ERCP patient became symptom free and is attending our clinic for last 11 months now.

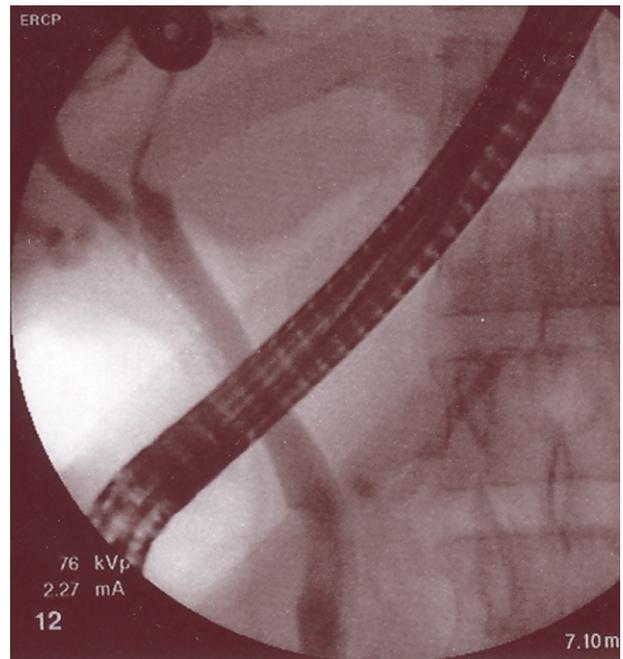


Figure 1: ERCP showing filling defect at lower end of CBD



Figure 2: Fasciola coming out of CBD

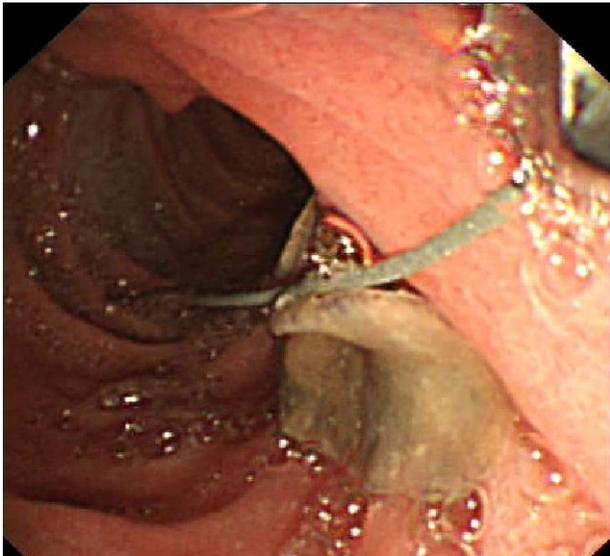


Figure 3: Complete extraction of *Fasciola* in duodenum by ERCP

Discussion

The index case had recurrent biliary colics without having cholangitis due to *Fasciola hepatica*. Although hailing from an endemic area (Egypt) he has been living in non-endemic zone (Saudi Arabia) for the last 8 years now implying that even brief exposure to an endemic zone can lead to infestation of the parasite. Various cases of biliary fascioliasis described in the literature [4], [7] have demonstrated obstructive jaundice to be the commonest presentation, however, the index case had no jaundice or cholangitis during four years of his intermittent illness. It is possible that there was a low worm load in the biliary system of the index case which allowed free flow of bile. There has to be high degree of suspicion to diagnose biliary fascioliasis as a cause of recurrent biliary colics especially in non-endemic areas. Although painful cholestasis associated with eosinophilia should make high suspicion of parasitosis as a case of biliary obstruction, the index case had no eosinophilia. Eosinophilia is striking in the hepatic phase and may not be observed in the biliary phase of the life cycle of the parasite as in the index case. During the first 3–4 months of acute infection, immunologic techniques play an important role in the diagnosis of fascioliasis. An enzyme-linked immunosorbent assay (ELISA) has a sensitivity of 100% and a specificity of 97.8% [2], [4]. During the early larval stage of infection, eggs are not found in the stool. The diagnosis can be made by finding characteristic ova in feces, duodenal aspirates, or bile specimens in the biliary phase of the parasite. Eggs are non-embryonated, ovoid, and large (130–150 × 60–90 μm), with a small operculum. Eggs being released sporadically it may be necessary to examine number of concentrated stool specimens to reach the diagnosis. The stool analysis in the index case was negative for ova and parasites, possibly we didn't repeat the analysis multiple times. We had low presumption of

fascioliasis before ERCP and no serology was done in the index case. In the biliary phase of the parasite CT scan may demonstrate hypodense nodules or tortuous tracks resulting from migration of the parasite, in addition to dilatation of biliary radicals [8]. Endoscopic ultrasound and ERCP may demonstrate irregular thickening of the common bile duct and ERCP having therapeutic potential has been preferred by most of the workers in the management of biliary fascioliasis [3], [4], [7]. The radiological picture of fascioliasis may even mimic other biliary disorders. For example Mohammad et al. [9] reported a 34-year-old male with obstructive jaundice having features of cholangiocarcinoma on magnetic resonance cholangiopancreatography (MRCP). Their patient proved to have fascioliasis on ERCP. The adult parasite mainly inhabits the biliary system and causes biliary complication but there are reports of pancreatitis caused by *Fasciola hepatica* [10], [11]. Maryo et al. [11] postulated that the eggs laid by the adult parasite cause obstruction at the papilla leading to pancreatitis and hence risk of pancreatitis is proportional to the worm load in a given case. Biliary phase of the parasite mimicking sphincter of Oddi dysfunction was reported by Keshishian et al. [12]. Thus biliary phase of the parasite has a complex clinical spectrum. It may present as biliary colics, cholangitis or pancreatitis and requires ERCP for removal of the parasite. In contrast the hepatic phase usually presents as abdominal pain, fever and jaundice. The hepatic phase is treated with triclabendazole which can be given as a single oral dose of 10 mg/kg or, in cases of severe infection, in two doses (10 mg/kg) given 12 h apart. Tablet triclabendazole 250 mg (manufactured by Novartis) is related to the benzimidazole–2–carbamate anthelmintics and has been approved for human biliary fascioliasis. This drug is shown to penetrate liver flukes by transtegumentary absorption followed by inhibition of the parasite's motility by causing the destruction of the microtubular structure, resulting in the death of the parasite [13].

Conclusion

Fascioliasis is one of the unusual causes of recurrent biliary colics and it warrants a high degree of clinical suspicion especially in non-endemic areas. The current report emphasizes that ERCP may be considered for management of recurrent biliary colics even though classical features of biliary obstruction may not be present.

Notes

Competing interests

The authors declare that they have no competing interests.

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