Case Report

Endoscopic Removal of One Live Biliary *Fasciola gigantica*

Wasana Kanoksil MD*,
Duangkamon Wattanatranon MD*, Chumpon Wilasrusmee MD**,
Somkit Mingphruedh MD**, Sukhum Bunyaratvej MD*

** Department of Pathology, Faculty of Medicine, Ramathibodi Hospital, Mahidol University*
*** Department of Surgery, Faculty of Medicine, Ramathibodi Hospital, Mahidol University*

Biliary fascioliasis was diagnosed in a woman from Angthong province, who presented herself with anorexia, weight loss, and jaundice for one month. Intraoperative retrograded cholangiopancreatography revealed a filling defect considered as a stone and bile sludge. By the sphincterotomy and balloon-extraction technique, one live fluke was removed and identified as Fasciola gigantica. A single dose of praziquantel, 25 mg/kg of body weight/day, was given although the postoperative stool examination revealed no Fasciola spp. egg. The patient was doing well after the fluke removal. According to a previous review, in this country, Ayutthaya is the southernmost province for the distribution of fascioliasis.

Keywords: *Fasciola gigantica, Fasciola hepatica, Hepatic fascioliasis, Biliary fascioliasi*

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Biliary fascioliasis denotes the infection of the large liver flukes i.e. *Fasciola hepatica* Linnaeus, 1758 or *Fasciola gigantica* Cobbold, 1855 (syn. *Fasciola indica*) in the biliary phase of their life cycles with the adult fluke(s) residing mainly in the extrahepatic biliary tract or under pancreatic fascioliasis in the pancreatic duct of the definitive host[1-9]. Due to their large sizes, 0.8-1.3 x 2-4 cm for *F. hepatica* and 0.5-1.3 x 2.4-7.6 cm for *F. gigantica*, biliary fascioliasis commonly cause obstruction of the extrahepatic biliary tract[10-12].

Fascioliasis is prevalent almost worldwide i.e. in Europe the Americas Oceania Africa Asia including Thailand[10-12]. The ranges of both *Fasciola* spp. overlap in tropical countries. Both of them have similar aquatic diheteroxenous life cycles requiring man, monkey, pig, dog, rodents and ruminants acting as the definitive hosts and fresh-water snails of the genus *Lymnaea* representing the intermediate hosts[10-12]. Man is the accidental host in the natural life cycle involving several mammalian species and snails[13].

During the past decade, recent advances in the surgical techniques allow the fluke removal from extrahepatic biliary tract or pancreatic duct by the endoscopic extraction[1-7,9]. In 2005, the authors experienced a case of biliary fascioliasis with a patient from Angtong province. She was treated by endoscopic sphincterotomy and balloon extraction of one live fluke identified as *F. gigantica.*

Correspondence to : Kanoksil W, Department of Pathology, Ramathibodi Hospital, Rama VI Rd, Bangkok 10400, Thailand.

Case Report

A 67-year-old female with underlying hypertension presented herself with anorexia and weight loss for one month. She was living in Angtong and Bangkok. Physical examination revealed jaundice and hepatomegaly. Ultrasound study of the upper abdomen showed a dilated gall bladder filled with gallstones and bile sludge. The common bile duct was 8 mm in diameter. The intrahepatic bile ducts were not dilated. Hepatomegaly with fatty liver was noted.

The liver biochemical profiles included alkaline phosphatase 180 U/l (normal 50-136), aspartate aminotransferase 33 U/l (15-37), alanine aminotransferase 28 U/l (30-65), gamma glutamyltranspeptidase 42 U/l (5-55), total protein 76.6 g/l (64-82), albumin 38.7 g/l (43.1-53.4), total bilirubin 78.3 μmol/l (0-17.1), direct bilirubin 41.2 μmol/l (0-5). Complete blood count included white blood cell count of 8.08 x 10^3/μl (4.8-10.8), neutrophils 59%, lymphocytes 30%, monocytes 8%,

eosinophils 2%, basophils 1%, red blood cell count 4.32 x 10^6/ml (4.2-6.1), platelet count 259 x 10^3/ml (140-450), hematocrit 32.5% (37-52), and hemoglobin 10.59 g/dl.

Intraoperative endoscopic retrograded cholangiopancreatography revealed mild bile-duct dilatation with a filling defect considered as a stone and bile sludge. The sphincterotomy was performed and by the balloon-extraction technique one fluke was removed alive (Fig. 1).

Post operative stool examination revealed no parasitic egg. Praziquantel (600 mg) 1 tablet three times a day (25 mg/kg of body weight/day) was given for one day. She was doing well up to six months at her last visit.

Fig. 1 A fluke being removed from the terminal bile duct into the duodenum by the balloon-extraction technique (arrow). Both of the anterior and ventral suckers can be seen

Fluke identification

The fluke received by the authors' surgical-pathology laboratory in recent formalin fixation was 0.6-1 x 2.4 x 0.1 cm in dimension. The anterior part was narrower than the posterior portion. The cephalic cone was distinct. The oral sucker was 1 mm and the ventral sucker or acetabulum was 1.5 mm in diameters (Fig. 2). The fluke was squeezed between two glass slides tightly bound by rubber bands for one week in formalin fixative, and then stained by Masson's trichrome method, and mounted on a glass slide [14].

Microscopic examination of the whole mount revealed tiny spines covering the tegument. There were 10 unembryonated eggs, ranging 88-96 x 136-140 mm (Fig. 3) in the branched uterus occupying the anterior half. The operculum could not be identified in similarity to the previous observation of the intrauterine eggs[12].

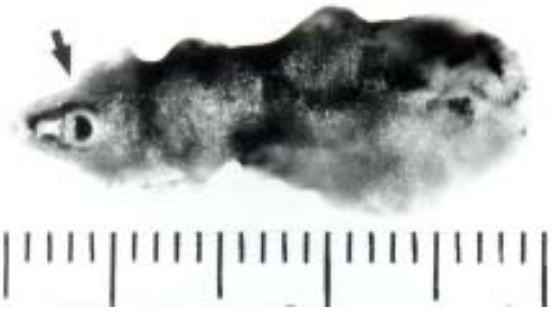

Fig. 2 The fluke after recent formalin fixation seen with a distinct cephalic cone (arrow), oral sucker and ventral sucker (acetabulum). Unstained. Scale in centimeters

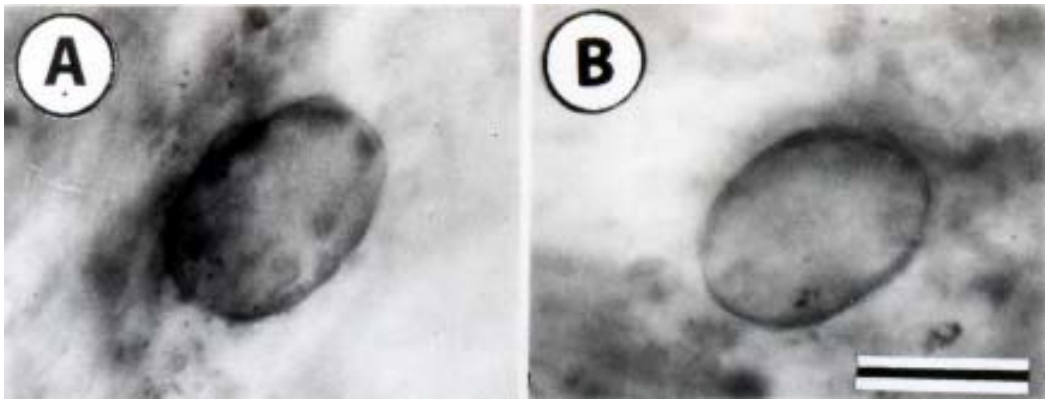

Fig. 3 A and B intrauterine eggs. Bar = 100 mm. Masson's trichrome stain

The presence of cephalic cone, the diameter proportion between the oral sucker and acetabulum of 1:1.5, the egg sizes and the fluke size fitted in for the identification of *Fasciola* spp.[10-12]. The length/width ratio of 1:2.4-1:4 indicated the entity of elongate fluke i.e. *Fasciola gigantica* [12].

Discussion

The life cycles of *Fasciola* spp. begin with egg deposition of the adult flukes residing in the biliary tract of the definitive host. The unembryonated eggs pass through the bile duct to the intestinal tract and exit with the feces. The eggs have to reach fresh water with an optimal temperature of 22-25 C before each developing a miracidium, which escapes the egg and penetrates an amphibious snail. The miracidia become cercariae through two radial stages in the snails. The swimming cercariae escape the snails and after a few hours encyst on the water plants or the surface of water as metacercariae. The metacercarial cysts remain viable for long periods. Humans and other definitive hosts mentioned prior contract fascioliasis by consuming metacercaria-contaminated water plants or drinking contaminated water[10-12].

Within the definitive host the metacercariae become excysted, probably by the host's bile and pancreatic trypsin similar to the *Sarcocystis* spp. sporocysts[15,16], and release the immature flukes in the duodenum and jejunum. The young flukes penetrate the host's intestinal wall, get into the peritoneal cavity and some find their way to the liver to complete their life cycles. Some flukes may penetrate the anterior abdominal wall and blood vessels in blind pathways. This stage is referred to as an initial phase in the acute phase extending for 3 weeks after the infection[11,12]. Some young flukes may reach the liver from the duodenal lumen by wandering up the bile duct[12].

The young flukes in the peritoneal cavity reach the liver, penetrate into the organ, and migrate through the liver parenchyma to find their way to the bile ducts. The stage is known as a middle phase or hepatic phase in the acute phase, which lasts for another 8 weeks[11,12]. Beyond this period of 11 weeks after infection, fascioliasis is recognized as chronic, latent, final, or biliary phase when the flukes establish residences in the bile ducts. A continuum of the disease with an overlapping of the hepatic phase over the biliary phase may occur and should be referred to as hepatobiliary fascioliasis[13]. The young flukes in the bile duct grow up to mature stage in another 3-4 months and can survive up to 13 years in humans[11,12].

Symptoms of the hepatic phase, which begin about 1 month after the exposure to metacercariae, are fever, general malaise, fatigue, hepatomegaly, anorexia, weight loss, urticaria with dermatographism and peripheral blood eosinophilia. The symptoms may be absent in cases of light infection. The biliary phase may be asymptomatic or there may be symptoms related to cholangitis and obstruction of the biliary tract due to the enlarging fluke(s). The biliary phase may last for months or years. Peripheral blood eosinophilia during this interval suggests hepatobiliary fascioliases[11-13]. Absence of peripheral blood eosinophilia in the biliary phase is likely due to the lack of active migration of the flukes in the tissue.

Extrahepatobiliary fascioliasis in the chronic stage may occur by worm transmission through the blood and lymph circulations. The condition leads to pulmonary infiltration, pleuropericarditis, meningitis, intraocular location, and lymphadenopathy. In addition, the young flukes may migrate to an ectopic site i.e. abdominal skin, intramuscular or intraperitoneal locations. In the Middle East adult worms ingested in raw liver from sheep or goat may attach themselves directly to the mucosa of pharynx and may cause "Halzoun" (suffocation) syndrome[11].

Sawangkit[17] had reviewed and reported cases of fascioliasis in Thai patients in both Thai and English literatures and found 25 cases during the year of first report in 1970 to 1990. These 25 cases included nine cases of hepatic phase, eight of biliary phase, seven of ectopic sites and one of pancreatic fascioliasis. Seventeen cases were identified as *F. gigantica* infection, seven as *F. hepatica* infection, and the remaining as the infection by unidentified species. Two additional cases of hepatic fascioliasis due to *F. hepatica* were recently reported[18]. The distribution of fascioliasis is confined mainly to the northeastern and northern regions of the country. The southernmost province of the distribution is Ayutthaya[17]. The presented patient and one patient in the recent report were from Angtong province just north of Ayutthaya[18]. The limited distribution of fascioliasis is not clear. Possibly, there is no available intermediate host, the snails of genus *Lymnaea*, in the remaining country south of Ayutthaya. Although, one patient in Bangkok with chronic ectopic fascioliasis was mentioned in the review[17], the patient might migrate from the endemic area similar to the patient in a recent report[18].

In animals, *F. gigantica* infection is the most important parasitic disease especially of adult cattle and water buffalos with the prevalence ranging 0-85%

in various villages in this country[19]. The prevalence is particularly high in the northeastern part, where there are several water reservoirs and ponds containing calm fresh water and *Lymnaea* spp. snails as well as various water plants allowing completion of the *F. gigantica* life cycle, which favors the natural conditions of aquatic environments and low elevations[12,19].

The conclusive diagnosis of biliary facioliasis can be made by the direct identification of fluke(s) obtained from surgical removal, or detection of eggs in the bile from duodenal tube. By stool examination the *F. hepatica* eggs, 63-90 x 130-150 μm, in the feces cannot be differentiated from those of the intestinal fluke, *Fasciolopsis buski*, with similar aquatic life cycle but requiring different species of snails and prevailing in Eastern Asia including the northeastern and central parts of Thailand[20-26]. The *F. hepatica* eggs differ from those of *F. gigantica* only by the slightly smaller size[12].

Serological methods include a complement fixation test, indirect immunofluorescence test, enzyme-linked immunoabsorbent assay (ELISA), immunoelectrophoresis and some others support the clinical diagnosis. However, cross-reactivity of *Fasciola* spp. with other trematode antigens has limited the specificity of the serological tests[11]. Various molecular techniques for distinguishing *F. hepatica* from *F. gigantica* are in developments[12].

For the medical treatment of hepatic or hepatobiliary fascioliasis, praziquantel 25 mg/kg/day three times daily for 3-7 days is ineffective[13]. Bithionol, 30-50 mg/kg/day on alternate days for 10-15 days, was recommended with a very high cure rate[13,27]. Recently, a highly effective fasciolicidal drug, triclabendazole in the dose of 10 mg/kg in a single dose after an overnight fast was described[11-13,27]. The use of prednisolone in the dose of 5-10 mg/day has been advocated prior to the administration of the fasciolicidal drug in acutely ill or toxic appearing children[13]. For biliary or pancreatic fascioliasis, the technique of endoscopic fluke removal is presently the treatment of choice[4,7].

Fascioliasis is not a major health problem in this country and not related to any hepatobiliary malignancy. The efforts in controlling human fascioliasis can be achieved by a) eradication of the disease in livestock, b) using molluscicides, and c) avoiding uncooked water plants and contaminated water in the endemic area[11].

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ฟาสซิโอเลียซิสในท่อน้ำดีรักษาด้วยการเอาตัวเป็นของพยาธิใบไม้ ฟาสซิโอลา ไยแกนติกา ออกด้วยกล้องส่องทางเดินอาหาร: รายงานผู้ป่วยหนึ่งราย

วาสนา กนกศิลป์, ดวงกมล วัฒนาตรานนท์, จุมพล วิลาศรัศมี, สมคิด มิ่งพฤติ, สุขุม บุณยะรัตเวช

โรคฟาสซิโอเลียซิสในท่อน้ำดีได้ถูกวินิจฉัยในผู้ป่วยหญิงจากจังหวัดอ่างทอง ผู้ป่วยมาโรงพยาบาลด้วยอาการเบื่ออาหาร น้ำหนักตัวลด และดีซ่านมา 1 เดือน จากการฉีดสารทึบรังสีเข้าสู่ท่อน้ำดีในห้องผ่าตัดพบว่ามีการพร่องของสารทึบรังสีคาดว่าจากนิ่วและน้ำดีข้น โดยวิธีการกรีดปลายท่อน้ำดีให้กว้างออกและใช้การดึงด้วยถุงลมโป่ง พยาธิใบไม้ที่ยังมีชีวิตอยู่หนึ่งตัวถูกลากออกมาจากท่อน้ำดีและได้รับการตรวจวิเคราะห์ว่าเป็น ฟาสซิโอลา ไยแกนติกา ผู้ป่วยได้รับยาพราซิควานทอลขนาด 25 มก./กก. ของน้ำหนักตัว/วัน นานหนึ่งวัน แม้ว่าการตรวจหาไข่พยาธิในอุจจาระเป็นผลลบหลังการผ่าตัด ผู้ป่วยมีอาการดีขึ้นภายหลังการเอาพยาธิใบไม้ออกจากท่อน้ำดี จังหวัดพระนครศรีอยุธยาเป็นพื้นที่ใต้สุดของประเทศ ที่พบการกระจายโรคฟาสซิโอเลียซิสในคนจากการศึกษารวบรวมรายงานก่อนหน้า