CASE REPORT

GRANULOMATOUS, HEPATOLITHIASIS AND HEPATOMEGALY CAUSED BY *CAPILLARIA HEPATICA* INFECTION: FIRST CASE REPORT OF THAILAND

S Tesana[1], A Puapairoj[2] and O-T Saeseow[3]

[1]Food-Borne Parasite Research Group, Department of Parasitology,
[2]Department of Pathology, [3]Department of Surgery, Faculty of Medicine,
Khon Kaen University, Khon Kaen, Thailand

Abstract. This is the first case report in Thailand of a *Capillaria hepatica* infection causing a granulomatous hepatic lesion, bile duct dilatation, hepatolithiasis and hepatomegaly. The patient's chief complaint was abdominal pain with fever and chills. Imaging of the liver revealed a 3-cm mass in the postero-inferior sub-segment of the right lobe of the liver with bile duct dilatation. Right hepatectomy and cholecystectomy were performed. Gross pathology of the right hepatectomy revealed focal intrahepatic duct dilatation with prominent periductal fibrosis. The histopathological section revealed chronic inflammation and some granuloma formation surrounding the bile ducts, generalized portal infiltration, prominence of eosinophils and hepatolithiasis. Histopathotogical section revealed oblique sections of *C. hepatica* egg (size 35.4±6.38 µm in width) and brown amorphous pigment.

INTRODUCTION

The rodent hepatic nematode, *Capillaria hepatica,* is rarely found infecting humans although sporadic human cases throughout the world have been reported. *C. hepatica* is a member of the family Trichuridae, known for its stichosomal esophagus.

The delicate adult worms are thread-like. The males are ~10 mm long and the females are ~20 mm long. The eggs of this parasite are similar to the eggs of *Trichuris trichiura* but their two plugs taper, *ie,* they do not protrude as do *T. trichiura* eggs. The eggs are 51 to 68x30 to 35 µm in size and the outer shell is striated (Beaver *et al,* 1984). Eggs are laid unembryonated and develop over a month in the soil until mature. The eggs are highly resistant to cold weather and remain viable in the soil for several months (Miyazaki, 1991).

The life-cycle of *C. hepatica* requires no intermediate host but two group of accidental final hosts have been found. The first group is predators of rats. When the predators eat infected rats and the rat's liver tissue is digested, the nematode eggs are liberated in the predator's feces (Farhang-Azad, 1977; Miyazaki, 1991).

Man is the second accidental host and acquires the infection by ingesting contaminated foods with the mature parasite eggs. When embyonated eggs are ingested, the larvae hatch in the cecum then migrate through venules to the liver. The parasite normally inhabits the liver parenchymal tissue where adult nematodes lay eggs. Fibrosis occurs around

Correspondence: Smarn Tesana, Food-Borne Parasite Research Group, Department of Parasitology, Faculty of Medicine, Khon Kaen University, Khon Kaen 40002, Thailand.
Tel: +66 (043) 348387; Fax: +66 (043) 202475
E-mail: smarn_te@kku.ac.th

trapped eggs and granulomatosis forms around adults. Infection causes fever, nausea, vomiting, hepatomegaly and eosinophilia (Arean, 1975). Many cases of spurious infection have been reported after finding eggs in the feces (Fan *et al,* 2000).

CASE REPORT

The subject was a married, 33-year-old, Thai, merchant woman. She was born in Champasak Province, Lao PDR. She moved to Thailand after marrying a Thai. She was admitted to a hospital in Chiang Mai Province with a chief complaint of abdominal pain in the right upper quadrant for two weeks. She had fever and chills at night.

Ultrasonography and computerized tomography of her abdomen revealed a 3-cm mass in the postero-inferior sub-segment of the right lobe of the liver with bile duct dilatation.

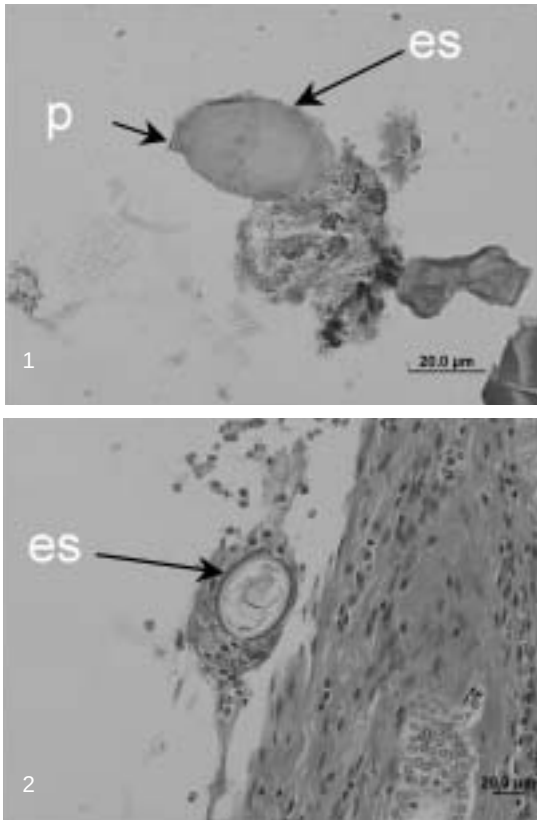
The laboratory findings included: Hb 13 g% (12-18), WBC 12,900 cells/mm^3 (5,000-10,000), neutrophils 95% (45-75), lymphocytes 2% (20-50), monocytes 2% (2-5) and eosinophils 1% (0-1). The liver function test was normal, ALT 28 U/l (4-36), AST 18 U/l (12-32) and alkaline phosphatase 87 U/l (42-121). Her blood sugar was 94 mg/dl. The tumor markers examined were: CA19-9 383 U/ml (0-37), CEA 0.8 ng/ml (0-2.5) and AFP 2.56 IU/ml (0-10).

She was given antibiotics for three days without improvement in her symptoms, which grew worse so she was referred to Srinagarind Hospital, Faculty of Medicine, Khon Kaen University. On admission, the patient appeared tired, but was neither pale nor jaundiced.

Physical examination revealed mild tenderness of the right upper quadrant of the abdomen with no guarding. Palpable hepatomegaly was found and the liver span was 10 cm. The other organs were normal. From her previous history, laboratory and image findings, she was diagnosed as having cholangiocarcinoma of the right lobe of the liver.

The patient underwent right hepatectomy and cholecystectomy. Gross examination of the right lobe revealed focal intrahepatic duct dilatation with prominent periductal fibrosis and hepatolithiasis. The gall-bladder was normal in appearance.

The histopathological examination revealed generalized portal infiltration, prominent eosinophilia and hepatolithiasis. Chronic inflammation and some granuloma formation surrounded the bile ducts and some bile ducts were dilated. A cross-section of the biliary passage contained 10 round parasite egg 28-45 µm in width (average, 40.31±4.96 µm). Some were seen on the oblique-section and presented with flattened polar plugs (Figs 1, 2) and brown amorphous pigment was seen



Figs 1 and 2–Histopathological sections of liver tissue showing oblique *C. hepatica* eggs (es) with a flattened polar plug (p).

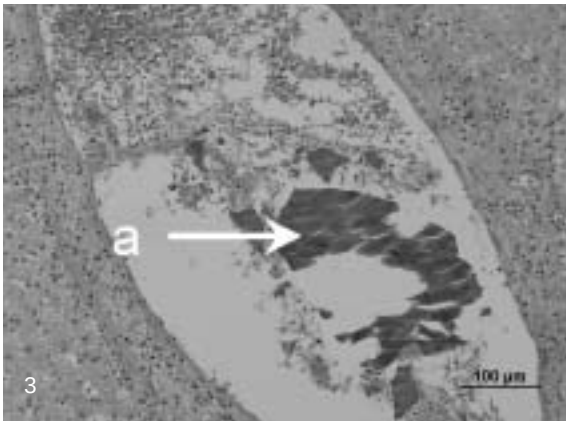


Fig 3–Histopathological section of liver tissue showing brown amorphous pigment (a).

on histhopathological section (Fig 3).

After the operation, she recovered and was discharged.

DISCUSSION

C. hepatica has a world-wide distribution with variable prevalence of infection in rats. Infection in man is rare and case reports are mostly in children (Ward and Dent, 1959; Attah *et al,* 1983; Pannenbecker *et al,* 1990; Choe *et al,* 1993; Sawamura *et al,* 1999) that may have acquired the infection by accidental ingestion of contaminated mature *C. hepatica* eggs in the soil. In Thailand, infection in rodents in nature has been found.

The prevalence of *C. hepatica* infection in rat in the Chiang Mai moat, Thailand, was 7.89% (Namue and Wongsawad, 1998); however, there have been no reported human cases of this infection in Thailand. This is, therefore, the first reported case (adult female) in Thailand, whose chief problems were fever, hepatomegaly and granulomatous pathological changes as has been reported in many previous cases.

Our patient was diagnosed with cholangiocarcinoma based on her history, laboratory (CA19-9: 383 U/ml) and image findings. A right hepatectomy and cholecystectomy were performed after finding granulomatosis of the liver and hepatolithiasis. A cross-section of the liver revealed egg, as the worms may have disintegrated into a brown amorphous material.

Infected patients usually have signs and symptoms of fever, nausea, vomiting, hepatomegaly, eosinophilia and weight loss (Arean, 1975; Sawamura *et al,* 1999; Klenzak *et al,* 2005). The symptoms may be confused with cholangiocarcinoma in northeast Thailand, where the incidence is the highest in the world. Marked fibrosis of the liver causes granulomatous lesions surrounding the eggs and infiltration of eosinophils (Attah *et al,* 1983; Pannenbecker *et al,* 1990; Kohatsu *et al,* 1995; Govil and Desai, 1996).

The parasite may have a recognizable structure or be partially disintegrated or appear as a bright, amorphous acidophilic material (Arean, 1975; Kohatsu *et al,* 1995). In our patient, the eggs had no clear shape or structure, only eggshells were found.

The parasite invaded the liver tissue causing eosinophil infiltration. Damage to the liver correlated with disordered liver function resulting in fever and chills at night similar to a liver abscess. Many other cases have had eosinophilia (Berger *et al,* 1990; Choe *et al,* 1993; Kohatsu *et al,* 1995) but in our case, the eosinophilia was not high. The infection may have been chronic but the worm had already disintegrated. Eosinophil infiltration was prominent on the histhopathological section. There are case reports without eosinophilia (Gonzalez Barranco *et al,* 1996).

The prominent granulomatous lesions in the liver may be confused with schistosomiasis mansoni (Govil and Desai, 1996). In most reported cases eggs or worms have been found trapped in the liver parenchymal tissue. The width of some eggs in our case were larger than reported elsewhere (*ie,* 30-35 µm) but the average width was comparable. No

pitted egghells were observed possibly because this may have been destroyed by host immune response. Eggs found on fecal examination are usually spurious parasites (Fan *et al,* 2000), eggs contaminating food or water.

By comparison, in *Ascaris lumbricoides* there is migration through the bile or pancreatic ducts resulting in damage and the formation of a hepatic abscess, acute cholecystitis, acute cholangitis, biliary colic and/or acute pancreatitis, and its eggs, if found, are larger (fertile egg, 40x60 µm, infertile egg, 40x90 µm) than those of *C. hepatica* (Beaver *et al,* 1984; Khuroo *et al,* 1990; Bellini *et al,* 1992; Bahu Mda *et al,* 2001).

C. hepatica infection is affected by treatment with thiabendazole, mebendazole, albendazole and ivermectin (Berger *et al,* 1990; Pannenbecker *et al,* 1990; Cheetham and Markus, 1991; Choe *et al,* 1993; Sawamura *et al,* 1999; Klenzak *et al,* 2005). Successful treatment of a man with a massive *C. hepatica* infection was achieved with prednisone, disophenol and pyrantel tartrate (Pereira *et al,* 1983).

This parasitic infection caused dilatation of bile ducts, fibrosis and granulomatous changes due to disintegrated worms and their eggs resulting in abdominal pain, fever and chills. These symptoms and imaging of the liver are similar to cholangiocarcinoma. Physicians in endemic areas should be aware of this when making a diagnosis.

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