

## HEPATIC SCHISTOSOMIASIS JAPONICA IN A PATIENT WITH GALLSTONES AND BILE DUCT STONES – A CASE REPORT

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Schistosomiasis, a common parasitic disease in many countries, is found as imported cases in Taiwan. Responsible for human infections are five species, one of which, *Schistosoma japonicum*, is currently endemic in China and South-east Asia. Chronic infection with *S. japonicum* may lead to the development of liver fibrosis, calcification and portal hypertension. Under investigation by sonography and computed tomography (CT) scan, a peculiar “turtle-back” appearance of liver fibrosis and calcification may be found. Herein, we report a case referred to our department due to jaundice. The sonography of liver showed typical “turtle-back” appearance. Gallstones and bile duct stones were also found in this case. Surgical interventions with percutaneous transhepatic biliary drainage (PTBD), cholecystectomy and choledocholithotomy were performed to relieve the obstructive jaundice and remove the stones. There were no parasitic eggs in the extracted stones or in drained bile juice. However, deposits of calcified *S. japonicum* eggs in liver parenchyma and portal tracts were identified in liver biopsy. No special treatment was given for the schistosomiasis japonica because the calcified parasitic eggs were the sequelae of past infection.

**Key words:** hepatic schistosomiasis japonica, gallstones, bile duct stones

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Schistosomiasis is a common parasitic infection. Five species, including *Schistosoma masoni*, *S. japonicum*, *S. haematobium*, *S. intercalatum*, and *S. mekongi* are responsible for human infections. It is endemic in at least 75 countries and affects approximately 2 million people in the world [1]. Among these five species, *S. japonicum* is endemic in China, Indonesia, the Philippines and other parts of South-east Asia [2]. Chronic infection with *S. japonicum* may lead to the development of liver fibrosis, calcifi-

cation and portal hypertension. A peculiar fibrosis and calcification pattern of the liver may be found by sonography and computed tomography (CT) [3, 4].

We report a case of chronic hepatic schistosomiasis japonica presenting with the typical findings of liver fibrosis. Gallstones, common hepatic duct stone (CHD) and common bile duct (CBD) stone with obstructive jaundice were also found in this case.

### CASE PRESENTATION

A 74-year-old male patient was referred to our department with the chief complaint of jaundice for five months. He was born in Hwu-Pei Province, China and came to Taiwan when he was 28 years old. Six years prior to admittance to our hospital, he had moved to Cambodia and come back to Taiwan

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the year before. Unfortunately, he suffered from jaundice later. At our emergency department, abnormal liver function test was found. The total serum bilirubin was 8.7 mg/dL with direct bilirubin 5.5 mg/dL. The serum aspartate aminotransferase (AST) and alanine aminotransferase (ALT) were 119 IU/L and 98 IU/L, respectively. The serum alkaline phosphatase was 674 IU/L. Sonography of liver showed "turtle-back" appearance of fibrosis (Fig. 1). Abdomen CT revealed gallstones and CBD stones along with intrahepatic ducts and CBD dilatation.

After admission, percutaneous transhepatic biliary drainage (PTBD) was performed to relieve obstructive jaundice. The cholangiography showed CHD stone and distal CBD stone (Fig. 2). The surface antigen of hepatitis B virus (HBV) and antibody of hepatitis C virus (HCV) yielded positive and negative results, respectively. Cholecystectomy and choledocholithotomy with T-tube drainage were done later. Mild liver cirrhosis was found during operation. Intra-operative wedge liver biopsy showed moderate portal inflammation with focal granulomatous reaction and fibrosis. Deposits of *S. japonicum* eggs with calcified shells in liver parenchyma and portal tracts were noted (Fig. 3). No parasitic eggs could be identified in the extracted stones or drained bile juice. No special treatment was given for the schistosomiasis japonica because these calcified parasitic eggs represented the sequelae of past infection.

## DISCUSSION

Schistosomiasis has two major stages in its life cycle: an asexual stage occurring in the snail as intermediate host and a sexual stage, in the human. After development in the snail, the cercariae penetrate human skin and migrate through the venous circulation to the heart and lung. The worms migrate to the hepatic portal circulation and mesenteric venous plexus later [1].

The intermediate host of *S. japonicum* is *Oncomelania* spp, distributed across China, Philippines and other parts of Southeast Asia [1]. In China, the snails are mainly distributed along the Yantze River. Wiest *et al.* [5] reported the prevalence of active infection to be 39.4% in the Jiangxi Province. Although the active infection among domestic animals is rarely seen in Taiwan now, the infection rate of susceptible snails was high. Lin *et al.* [6] in 1987 found 17% of snails in central Taiwan harbored cercariae of *S. japonicum*. The parasitic infection is

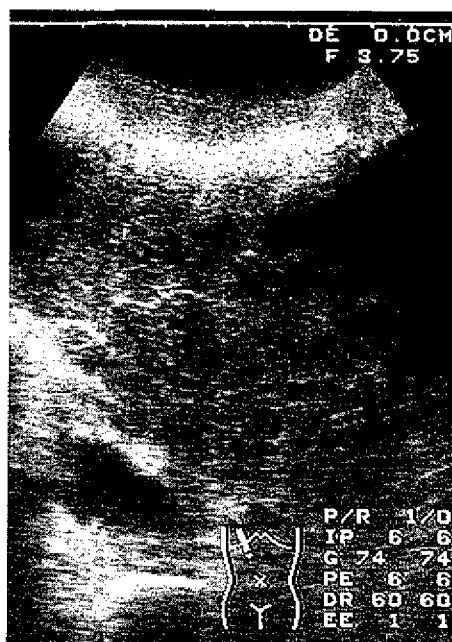


Fig. 1. Abdomen sonography shows "turtle-back" appearance of liver fibrosis.



Fig. 2. Two filling defects meaning stones were found over the common hepatic duct (black arrow) and common bile duct (white arrow) in the cholangiography.

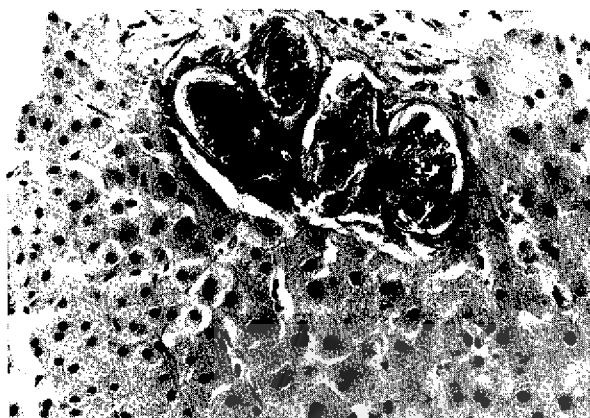


Fig. 3. Liver biopsy reveals deposits of *S. japonicum* eggs on liver parenchyma. H&E stain, reduced from  $\times 400$ .

also prevalent among the foreign laborers in Taiwan. In 1993, Peng *et al.* [7] reported the overall infection rate of the parasite was 18% among 1,364 Thai laborers and *S. japonicum* infection was found in one laborer. Therefore, schistosomiasis may become an important issue of public health.

In this case, he was born in an endemic area of schistosomiasis japonica, Hwu-Pei Province, China, and lived there for around 28 years. The patient had also lived in Cambodia for 6 years prior to his return to Taiwan one year before, where *S. mekongi*, not *S. japonicum*, is endemic [2,8]. Therefore, deposits of calcified eggs of *S. japonicum* could indicate a remote infection from his childhood or youth in China.

The clinical manifestations of chronic schistosomiasis are mainly caused by liver cirrhosis and portal hypertension. Schistosome eggs, which reside in the mesenteric venous plexus, can induce granulomatous responses. If the released eggs move to the liver by portal flow, granulomatous reactions could lead to pyelophlebitis, peripyelophlebitis and periportal fibrosis [1]. On sonography, the peculiar type of hyperechoic fibrotic septae separates lobules of liver parenchyma into a network pattern. Furthermore, this fibrosis could undergo dystrophic calcifications. Araki *et al.* [3] described the characteristics of the calcifications, which occur at the outermost layer to form capsular calcification or in the parenchyma of the liver perpendicular to the capsular calcification, resembling a septal calcification. The combined capsular and septal calcifications produce a characteristic "turtle-back" appearance of liver. However, in this case, the sonography revealed typical hepatic fibrosis, but no calcifications. This may be due to fewer deposited eggs to induce the dystrophic calcification.

Gallstones and bile ducts stones are associated with liver flukes such as *Clonorchis* and *Opisthorchis* [9,10], while urolithiasis is also considered to be associated with *S. haematobium* infection [11]. However, there are no studies describing the relationship between the schistosomiasis japonica and cholelithiasis. In this case, we did not find parasitic eggs in the bile juice or in the gallstones or CBD stones. Hence, there is no evidence proving the cholelithiasis related to *S. japonicum* infection.

Patients with schistosomiasis have been found to have a higher rate of HBV and HCV infection [12,13]. This association may be due to the use of contaminated needles for parenteral therapy of schistosomiasis. These coinfections, obviously, could increase the destruction of the liver. However, the

association of the schistosomiasis and the hepatocellular carcinoma (HCC) remains unconfirmed. Iida *et al.* [13] reported that there was no evidence of a significant correlation between schistosomiasis and HCC. On the other hand, Kojiro *et al.* [14] found *S. japonicum* infection to be a risk factor. Further studies are necessary to establish this relationship.

In conclusion, while hepatic schistosomiasis japonica is currently rarely seen in Taiwan, infected individual could bring it to Taiwan from overseas. Sonography and CT scan are excellent tools in the diagnosis of hepatic schistosomiasis with the characteristic "turtle-back" fibrosis or calcification.

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## 肝臟日本住血吸蟲感染合併 膽囊結石與膽管結石—病例報告

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住血吸蟲在很多國家是一種常見的感染，然而台灣的報告均屬於境外感染的病例。日本住血吸蟲目前流行於中國大陸以及東南亞地區。慢性感染日本住血吸蟲可能導致肝臟纖維化、鈣化及造成門靜脈高壓。在超音波及電腦斷層上，有時可以見到肝臟如「龜殼樣」的纖維化或鈣化。我們報告一位在中國大陸出生，於28歲遷移來台的74歲男性，因黃疸至本院求診。其腹部超音波檢查顯示肝臟具有「龜殼樣」的纖維

化，並且合併有膽道擴張、膽管結石及膽囊結石。入院後，該病人接受「經皮穿肝膽汁引流術」以改善阻塞性黃膽，之後並且接受膽管取石術及膽囊切除術，在膽汁及結石中並未發現蟲卵。但在病人的肝臟切片上可以發現已經鈣化的日本住血吸蟲蟲卵。這種鈣化的蟲卵代表過去感染所留下的後遺症，因此並未給予住血吸蟲症的特殊治療。

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