Iranian Journal of Pathology (2007)2 (2), 74-76

Case Report

Cholangiocarcinoma Associated with Liver Fluke Infection in an Iranian Patient

Mitra Heidarpour[1], Parvin Rajabi[1], Shervin Pejhan[1]

1.Dept. of Pathology, Alzahra Hospital, Isfahan University of Medical Sciences, Isfahan, Iran

ABSTRACT

Biliary liver flukes are common parasites in some regions of the world where consumption of raw fresh-water fish is a cultural practice. Clonorchis sinensis and Opisthorchis species are the major biliary liver flukes associated with human disease. Some of these parasites are highly associated with cholangiocarcinoma. This report describes a clinically and epidemiologically unsuspected liver fluke infection associated with cholangiocarcinoma in a middle aged woman from the south shores of Iran, and emphasizes the importance of efforts to discourage raw freshwater fish intake as the route of infection in high risk populations.

Key words: Cholangiocarcinoma, Liver fluke, Clonorchis sinensis

Introduction

Well-documented epidemiological, histopathological, and experimental studies of Clonorchis sinensis and Opisthorchis viverrini have provided convincing evidence of a relationship between these trematodes infections and the tendency for malignant transformation of the biliary epithelium in humans and experimentally infected animals (1- 4).

Clonorchiasis and opisthorchiasis Infection with the Chinese or oriental fluke, is endemic among fish-eating mammals in Southeast Asia. Human is an incidental host; the prevalence of human infection is highest in China, Vietnam, and Korea (5,6).In this paper we report the diagnosis of liver fluke infection associated with cholangiocarcinoma in a middle aged woman from southern area of Iran, which is a non endemic country.

Case report

A 55 year old woman from Genaveh port referred to our hospital for a one month history of jaundice, itching and abdominal pain. In her laboratory exams, total bilirubin was 36.5 mg/dl, conjugated bilirubin was elevated at 24 mg/dl, AST at 310 IU/L, ALT at 382 IU/L and Alkaline Phosphatase at 2287 IU/L.

On abdominal CT scan an isodence lesion causing biliary obstruction was seen. Intrahepatic ducts, adjacent to the mass were dilated (Figure 1). Cholangiography showed focal stricture of the bile ducts and abnormal enhancement of the ductal wall. The radiologic findings were in favor of intrahepatic cholangiocarcinoma(7,8).

Received: 24 February 2007
Accepted: 24 April 2007
Address communications to: Dr. Mitra Heidarpour, Department of Pathology, Alzahra Hospital, Isfahan University of Medical Sciences, Isfahan, Iran.
Email: pejhaan@yahoo.com

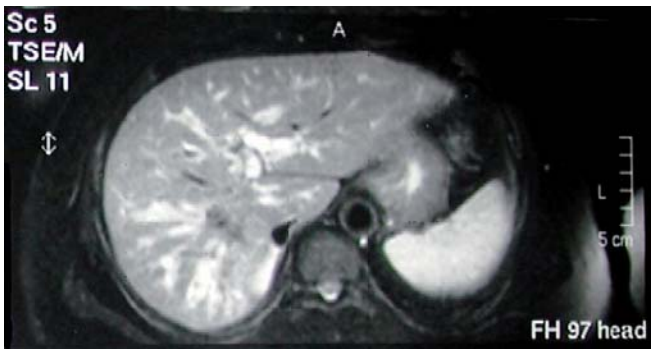

Figure 1. Contrast-enhanced transverse hepatic computed tomography: Hilar cholangiocarcinoma. Peripheral intrahepatic ducts are dilated.

The patient underwent surgery. A tumoral mass was found in the hilum of the liver which was resected. Cholecystectomy and Roux-en-Y hepatojejunostomy were performed.

At operation, tumoral seedings over the small intestine and peritoneum were suspicious, therefore biopsies from these areas were sent for pathologic study. The gross appearance of the tumor mass was that of a gray-white, relatively firm tumor circumscribed by hepatic tissue. The gall bladder had a normal appearance except for the fibrous thickening of the wall. Microscopically, the hilar mass was an adenocarcinoma with perineurial invasion. The tumoral glands were spreading between hepatocyte plates and along duct walls (Figure 2). Peritoneal biopsies showed metastatic cholangiocarcinoma.

The gallbladder specimen revealed a chronic cholecystitis but within the dilated bile ducts with periductal fibrosis, sections of mature liver flukes were observed (Figure 2 left). Of course it is not easy to distinguish clonorchis Sinensis from Opisthorchis flukes morphologically.After surgery her cholestatic jaundice was improved both clinically and paraclinically.Then the palliative therapy started.

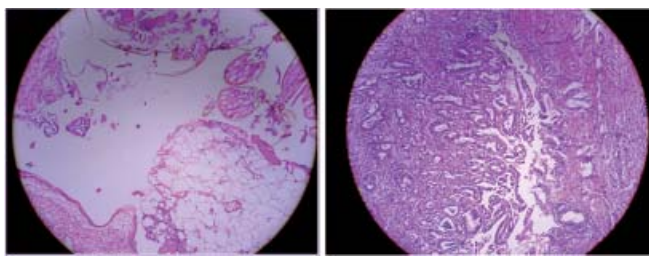

Figure 2. Left- Liver fluke within the dilated bile duct Thin arrow. Note the section of its oral sucker Thick arrow. Right- Tumoral glands of cholangiocarcinoma. (original magnification ×400)

Discussion

Clonorchiasis is endemic in the Far East, especially in southern and northeastern China, eastern Russia, Vietnam, and Korea (9). The custom of eating raw freshwater fish harboring metacercariae contributes to the high incidence of infection in these areas (10, 11). These organisms excyst in the duodenum, releasing larvae that travel through the ampulla of Vater and mature into adult worms in the bile canaliculi. The long life span of the worm, as long as 30 years, and the frequent asymptomatic infection, prevent us from an accurate diagnosis and treatment (12- 14).

Some of these flukes have been commonly seen in our neighborhood like Turkey, India and southern parts of Russia (15, 16). Although Iran is not known as an endemic area of clonorchiasis, this case report might warn us of an old and silent carcinogen in some parts of our country.

The liver fluke infection results from the consumption of fresh-water fish containing the encysted metacercariae. Fish may be eaten raw, pickled, smoked or dried (13). As we found retrospectively, in Genaveh port, like some other parts of our country, ingesting raw or inadequately cooked fish has been an old tradition, even sometimes with therapeutic goal.

Conclusion

Presence of special snail species as the first intermediate host of these worms could be investigated in suspected areas of Iran. Epidemiological studies may reveal the presence of infection in these areas. Although biliary liver fluke is an infrequent cause of infection in Iran, because of the potential long-term consequences of chronic infection, educational information highlighting routes of infection and the fact that asymptomatic infection is common and may eventually lead to cancer should be aimed at high-risk community members.

References

1. Stauffer WM, Sellman JS, Walker PF. Biliary liver flukes (Opisthorchiasis and Clonorchiasis) in immigrants in the United States: often subtle and diagnosed years after arrival. J Travel Med. 2004 May-Jun;11(3):157-9.

2. K. Okuda, D. G. Mitchell, Y. Itai, J. Ariyama. Hepatobiliary disease pathophysiology and imaging. London: Blackwell Science; 2001.

3. HOU PC. The relationship between primary carcinoma of the liver and infestation with Clonorchis sinensis. J Pathol

Bacteriol. 1956 Jul;72(1):239-46.

4. Monogr. Eval. Carcinog. Risks Hum. International Agency for Research on Cancer Working Group. Infection with liver flukes (Opisthorchis viverrini, Opisthorchis felineus and Clonorchis sinensis). 1994. P.121-175.

5. Monroe L. S, Haubrich W. S., Schaffner F., Berk J. E., editors. Bockus gastroenterology Gastrointestinal parasites. Philadelphia: Saunders; 1995.

6. Rim HJ. The current pathobiology and chemotherapy of clonorchiasis. Kisaengchunghak Chapchi. 1986 Apr;24 Suppl:1-141.

7. Valls C, Gumà A, Puig I, Sanchez A, Andía E, Serrano T, et al. Intrahepatic peripheral cholangiocarcinoma: CT evaluation. Abdom Imaging. 2000 Sep-Oct;25(5):490-6.

8. Kim YH. Extrahepatic cholangiocarcinoma associated with clonorchiasis: CT evaluation. Abdom Imaging. 2003 Jan-Feb;28(1):68-71.

9. Ona FV, Dytoc JN. Clonorchis-associated cholangiocarcinoma: a report of two cases with unusual manifestations. Gastroenterology. 1991 Sep;101(3):831-9.

10. Choi BI, Han JK, Hong ST, Lee KH. Clonorchiasis and cholangiocarcinoma: etiologic relationship and imaging diagnosis. Clin Microbiol Rev. 2004 Jul;17(3):540-52.

11. Lun ZR, Gasser RB, Lai DH, Li AX, Zhu XQ, Yu XB, et aL. Clonorchiasis: a key foodborne zoonosis in China. Lancet Infect Dis. 2005 Jan;5(1):31-41.

12. Sun T. Clonorchiasis and opisthorchiasis. Pathology and clinical features or parasitic diseases. New York: Masson; 1982.

13. David T.,John William A, Petri Jr, Markell ,Voge's. Medical Parasitology. Philadelphia: Saunders; 2006.

14. Abdel-Rahim AY. Parasitic infections and hepatic neoplasia. Dig Dis. 2001;19(4):288-91.

15. Rim HJ. Clonorchiasis: an update. J Helminthol. 2005 Sep;79(3):269-81.

16. Berger B, Vierbuchen M. Opisthorchiasis simulating a malignancy. Z Gastroenterol. 2001 Feb;39(2):173-5.