

Recurrent pyogenic cholangitis in a white woman

Simon Bergman, MD;* Antonio Di Carlo, MD;† Prosanto Chaudhury, MD;* Carl Blum, MD;† Ayoub Nahal, MD;‡ Peter Metrakos, MD*

A healthy 68-year-old white woman of eastern European origin who had lived her entire life in Canada presented with ascending cholangitis. She had undergone cholecystectomy many years earlier and had suffered recurrent bouts of abdominal pain over the last few years, for which multiple investigations demonstrated only common bile duct (CBD) dilatation without a clear etiology. CT of the abdomen (Fig. 1) now showed dilated left-sided central intrahepatic bile duct, and endoscopic retrograde cholangiopancreatography (ERCP) confirmed the presence of multiple small stones in those segments. A diagnosis of recurrent pyogenic cholangitis (RPC) was made. Because the large number of stones could not all be retrieved by ERCP and because of the recurrent nature of her disease, surgical management was indicated.

The patient underwent a left hepatectomy. Multiple, small, friable, pigmented stones were found in the left intrahepatic bile ducts; intraoperative choledochoscopy confirmed the absence of any disease in the right biliary tree. A Roux-en-Y hepaticojejunostomy was performed with pexy of the blind-ended limb to the anterior abdominal wall for future percutaneous cannulation of the biliary tree in the event of recurrent stones or strictures (a modification of the technique described by Hutson and colleagues¹ for the treatment of recurrent biliary strictures in sclerosing cholangitis). The excised speci-

men contained multiple intrahepatic pigmented stones. The biliary mucosa showed marked chronic inflammatory and fibrotic changes with a minimally inflamed hepatic parenchyma (Fig. 2).

Discussion

Recurrent pyogenic cholangitis, also known as Hong Kong disease and oriental cholangiohepatitis, is characterized by

recurrent bouts of abdominal pain, jaundice and fever secondary to intrahepatic ductal dilatation and strictures, and subsequent intrahepatic stone formation. Most prevalent in the East, it is seen increasing in the West mainly owing to immigration.^{2,3} This is the ninth case of RPC in a non-Asian and only the third documented case in North America. Western doctors should be familiar with this disease as it differs significantly both in diagnosis and



FIG. 1. CT scan of the abdomen demonstrating left-sided dilatation of the central intrahepatic bile duct.

From the Departments of *Surgery, †Radiology and ‡Pathology, McGill University, Montréal, Que.

Accepted for publication Jul. 19, 2007

Correspondence to: Dr. Peter Metrakos, 687 Pine Ave. W, Rm. S10-26, Montréal QC H3A 1A1; fax 514 843-1503; Peter.Metrakos@muhc.mcgill.ca

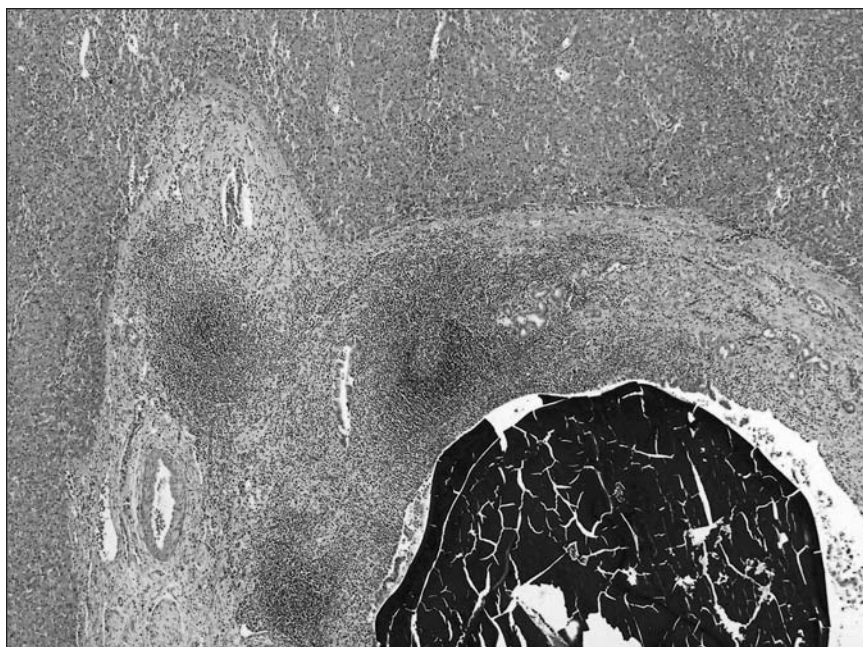


FIG. 2. Excised specimen of the left hepatic lobe showing chronic inflammatory and fibrotic changes of the biliary mucosa with minimal inflammation of the hepatic parenchyma.

management as compared with biliary disease in that region of the world. If left untreated, RPC is associated with a high morbidity and mortality as it progresses to biliary cirrhosis and liver failure.

Recurrent pyogenic cholangitis is thought to occur in patients suffering from chronic infestation of the biliary tree by *Clonorchis sinensis* and *Ascaris lumbricoides*, liver flukes endemic to Southeast Asia that may obstruct the intrahepatic biliary ducts with resultant bile stasis, pigment stone formation and bacterial superinfection. The *Clonorchis* parasite has a lifespan of 20 years and can therefore manifest itself many years after initial infestation.⁴ Patients with RPC most often present with symptoms of cholangitis, and bile cultures are almost always positive and usually yield

Escherichia coli, *Enterococcus fecalis*, or other enteric bacteria, but *Clonorchis* or *Ascaris* parasites or ova are recovered from the stool in only 25% of cases. Biliary disease involves the CBD and left hepatic lobe in 37% of cases and includes the right hepatic lobe in 31%. It is confined to the CBD in 27%, and only rarely confined to the right lobe. The gallbladder is usually disease-free.²

Acutely, RPC is treated with broad-spectrum antibiotics and ERCP decompression. CBD exploration and T-tube drainage are reserved for failure of medical management. Further attacks may be prevented by elective CBD exploration with choledocopy for stone retrieval, irrigation of the biliary tree and treatment of strictures. Biliary-enteric bypass is required for distal extrahepatic stric-

tures; however, this is associated with cholangitis and intrahepatic abscess in up to 30% of patients.⁵ Hepatic resection should be reserved for patients with significant hepatic atrophy and fibrosis, multiple liver abscesses or concurrent intrahepatic cholangiocarcinoma, which may occur in 3%–5% of patients treated conservatively.

Conclusions

Recurrent pyogenic cholangitis should be suspected in anyone with a history of recurrent right upper quadrant pain despite past cholecystectomy. Its diagnosis and treatment require the cooperation of surgeon, endoscopist, and diagnostic and interventional radiologists. Surgical interventions still carry a high morbidity and must be tailored to the individual patient, depending on the severity of disease.

Competing interests: None declared.

References

1. Hutson DG, Russell E, Schiff E. Balloon dilatation of biliary strictures through a choledochojejunocutaneous fistula. *Ann Surg* 1984;199:637-47.
2. Harris HW, Kumwenda ZL, Sheen-Chen S. Recurrent pyogenic cholangitis. *Am J Surg* 1998;176:34-7.
3. Federle MP, Cello JP, Laing FC. Recurrent pyogenic cholangitis in Asian immigrants. Use of ultrasonography, computed tomography, and cholangiography. *Radiology* 1982;143:151-6.
4. Sun T. Clonorchiasis: a report of four cases and discussion of unusual manifestations. *Am J Trop Med Hyg* 1980;29:1223-7.
5. Kusano T, Isa T, Muto Y. Long-term results of hepaticojejunostomy for hepatolithiasis. *Am Surg* 2001;67:442-6.