4. Chappet V. Cancer epithelial primitif du canal choledoque [French]. Lyon Chirurgic 1894;76:145-57.
5. Nakanuma Y, Sasaki M, Ishikawa A, et al. Biliary papillary neoplasm of the liver. Histol Histopathol 2002;17:851-61.
6. Hoang TV, Bluemake DA. Biliary papillomatosis: CT and MR findings. J Comput Assist Tomogr 1998;22:671-2.
7. D'Abrigeon G, Blanc P, Bauret P, et al. Diagnostic and therapeutic aspects of endoscopic retrograde cholangiography in papillomatosis of the bile ducts: analysis of five cases. Gastrointest Endosc 1997;46:237-43.
8. Kim YS, Myung SJ, Kim SY, et al. Biliary papillomatosis: clinical, cholangiographic and cholangioscopic findings. Endoscopy 1998;30:763-7.
9. Barnett JL, Knol J. Use of a novel, "adoptable" baby cholangioscope to diagnose a biliary papillary adenema. Gastrointest Endosc 1995;41:70-2.
10. Zen Y, Fujii T, Nakamura K, et al. Biliary papillary tumors share pathological features with intraductal papillary mucinous neoplasm of the pancreas. Hepatology 2006;44:1333-43.
11. Rambaud S, Nores J, Meeus F, et al. Malignant papillomatosis of the bile ducts: a new indication for liver transplantation? Am J Gastroenterol 1989;84:448-9.
12. Christiaens P, Decock S, Buchel O, et al. Endoscopic trimming of metallic stents with the use of argon plasma. Gastrointest Endosc 2008;67:369-71.

Division of Digestive and Liver Diseases, Department of Internal Medicine (S.F.J., D.N., S.-J.T.), Department of Surgery (C.B.), The University of Texas Southwestern Medical Center, Dallas, Texas, USA.

Reprint requests: Shou-jiang Tang, MD, Director of Endoscopy, Parkland Memorial Hospital, Division of Digestive and Liver Diseases, UT Southwestern Medical Center, 5323 Harry Hines Boulevard, Dallas, TX 75390-9151.

0016-5107/$36.00
doi:10.1016/j.gie.2008.03.1095

Sclerosing cholangitis from microscopic polyarteritis: an 8-year follow-up case report

Sina Alexander, MBBS, FRACP, Michael J. Bourke, MBBS, FRACP, Jonard Co, MD, Stephen J. Williams, MBBS, FRACP

Sydney, Australia

Disruption to the arterial blood supply of the biliary epithelium can lead to ischemic cholangitis, with ensuing strictures, cholestasis, and, if unresolved, progression to secondary sclerosing cholangitis. Reported causes include iatrogenic hepatic arterial injury, hepatic artery thrombosis in liver transplantation recipients, or intra-arterial administration of chemotherapeutic drugs.[1] Rarely, ischemic cholangitis can develop as a result of vasculitis affecting the mesenteric blood vessels. We present a case that shows the progression of ischemic cholangitis in a patient with microscopic polyarteritis (MPA) who developed symptomatic sequential common bile duct (CBD) and hilar strictures. Side-by-side plastic stents and bilateral hilar stents were used for the treatment of CBD and hilar strictures, respectively, in conjunction with cytotoxic and immunosuppressive therapy.

CASE REPORT

A 37-year-old woman with no medical history presented with epigastric and right upper-quadrant pain. She underwent a series of investigations that included upper and lower endoscopies, US, abdominal CT, hepatobiliary iminodiacetic acid scan, and laparoscopy, all of which were unremarkable. Subsequently, a further severe episode of abdominal pain was associated with mild mixed abnormalities of liver function tests (LFT) and biliary dilatation. An

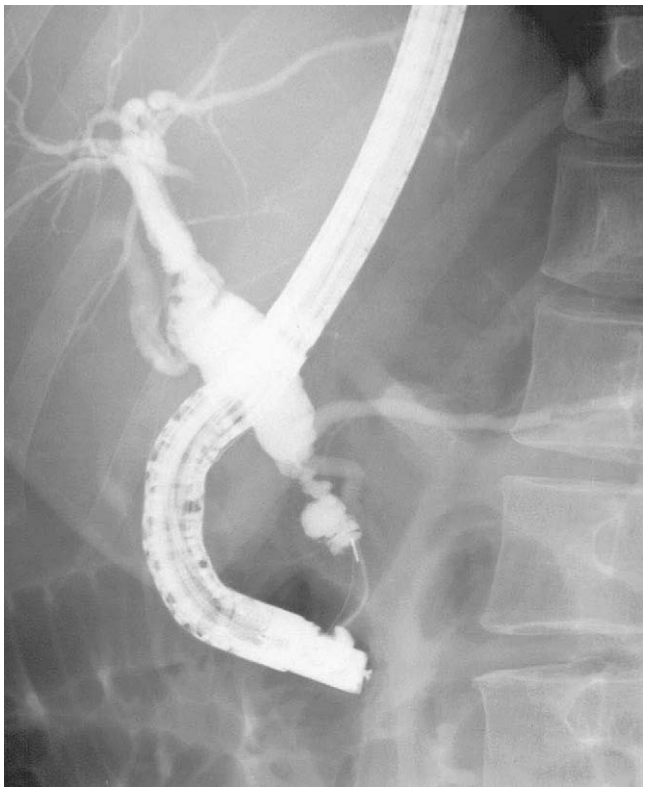

Figure 1. Finding on ERCP, showing a distal CBD stricture with proximal biliary dilatation.

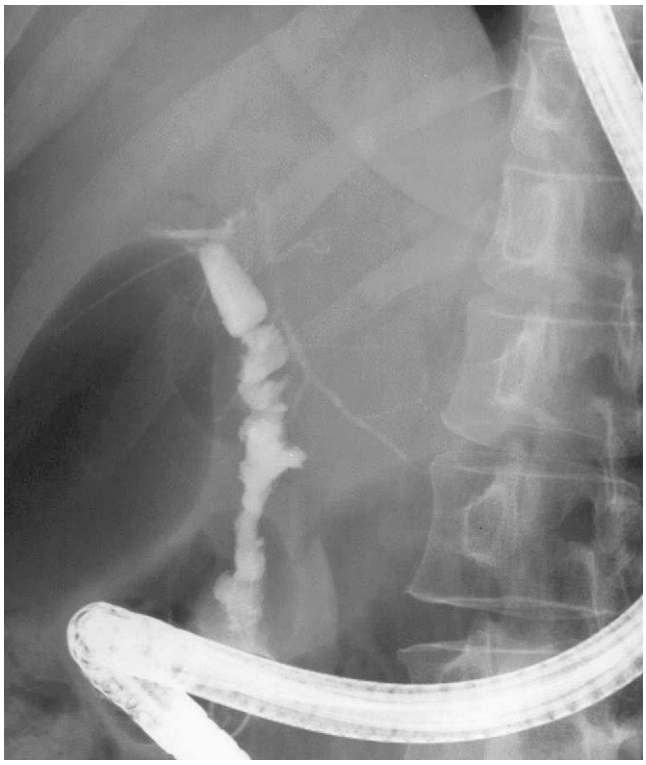

Figure 2. Finding on ERCP, showing resolution of distal CBD stricture but diffuse abnormality with irregular ridging of the biliary wall and pseudodiverticuli.

ERCP was performed. The papilla and pancreatic duct were normal. The bile duct had a pleated configuration at the distal end, with a stricture in the mid CBD and proximal dilatation. The intrahepatic ducts were normal (Fig. 1). A 10F, 7-cm-long plastic stent was inserted. A subsequent exhaustive investigation, including an EUS, failed to discern an etiology. A subsequent ERCP at 3 months showed improvement of the biliary stricture, and she began a program of side-by-side stenting. The stricture and the proximal dilatation resolved, although the cholangiogram revealed an irregular ridged biliary wall and pseudodiverticuli (Fig. 2). The patient was now asymptomatic and declined further follow-up.

Four years after the initial ERCP, she developed polyarthritis, peripheral neuropathy, and cutaneous vasculitis, with symptoms of cholestasis and abnormal LFTs. An ERCP revealed bilateral hilar strictures with dilatation, which was more pronounced on the right. There was also retraction of the biliary tree, with shortening of the CBD (Fig. 3A and B). Antinuclear antibodies were positive (1 in 320). Perinuclear antineutrophil cytoplasm antibodies (p-ANCA) were also positive. Erythrocyte sedimentation rate was elevated at 96 mm/h (normal = 0-26 mm/h). Abdominal angiogram performed on 2 separate occasions revealed microaneurysm formation. The changes predominantly involved the renal and hepatic arteries, although

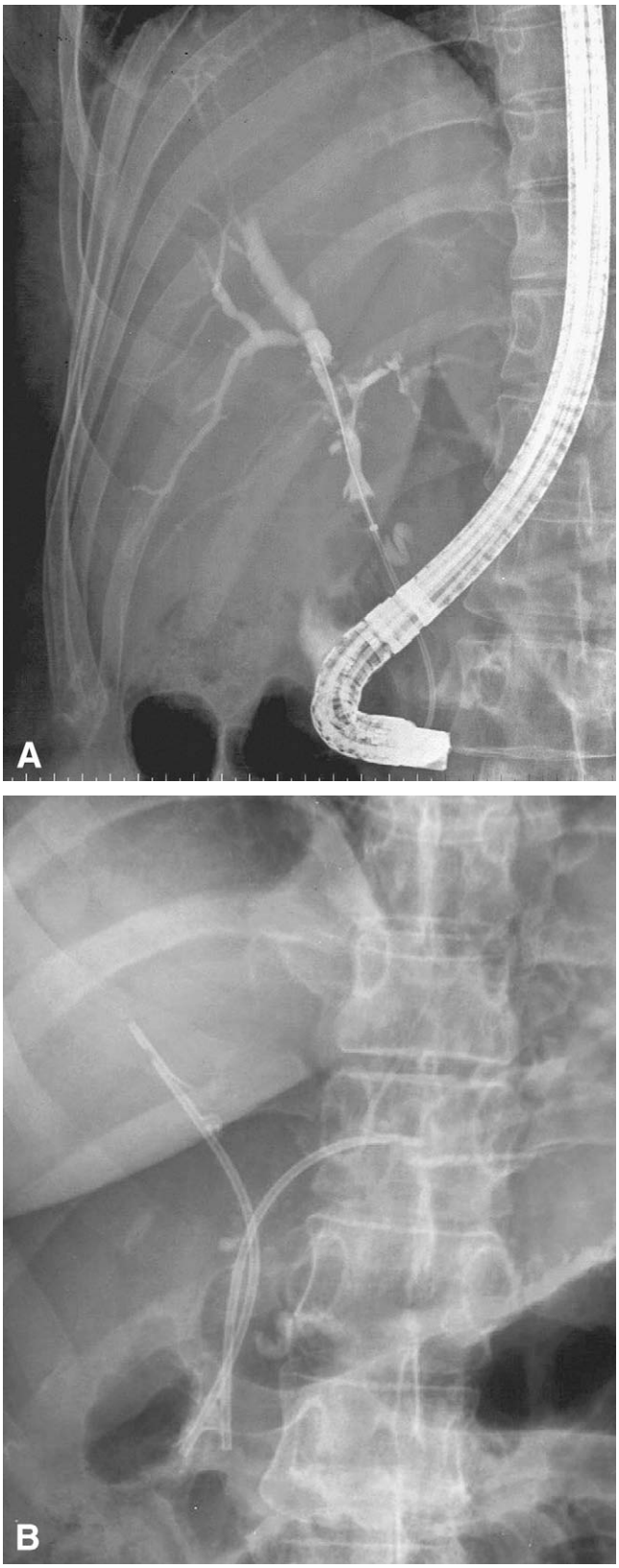

Figure 3. A, Balloon occlusion cholangiogram, showing shortening of the bile duct, bilateral hilar strictures, and proximal dilatation. **B**, Bilateral 7F hilar stents were placed.