BMC Gastroenterology

Case report

Bacterial cholangitis causing secondary sclerosing cholangitis: A case report

Pieter CJ ter Borg[1], Henk R van Buuren*[1] and Annekatrien CTM Depla[2]

Address: [1]Department of Gastroenterology and Hepatology, Erasmus MC, Rotterdam, The Netherlands and [2]Department of Internal Medicine and Gastroenterology, Slotervaart Ziekenhuis, Amsterdam, The Netherlands

E-mail: Pieter CJ ter Borg - terborg@mdl.azr.nl; Henk R van Buuren* - vanbuuren@mdl.azr.nl;
Annekatrien CTM Depla - bartelsman.depla@chello.nl

*Corresponding author

Published: 3 June 2002

BMC Gastroenterology 2002, **2**:14

Received: 28 March 2002
Accepted: 3 June 2002

This article is available from: http://www.biomedcentral.com/1471-230X/2/14

Abstract

Background: Although bacterial cholangitis is frequently mentioned as a cause of secondary sclerosing cholangitis, it appears to be extremely rare, with only one documented case ever reported.

Case presentation: A 48-year-old woman presented with an episode of acute biliary pancreatitis that was complicated by pancreatic abcess formation. After 3 months she had an episode of severe pyogenic *(E. Coli)* cholangitis that recurred over the subsequent 7 months on a further two occasions. Initially, cholangiography suggested the presence of extra-biliary intrahepatic abcesses while repeated investigations demonstrated development of multiple segmental biliary duct strictures. After maintenance antibiotic treatment was started, no episodes of cholangitis occurred over a 14-month period.

Conclusions: Sclerosing cholangitis can rapidly develop after an episode of bacterial cholangitis. Extra-biliary involvement of the hepatic parenchyma with abcess formation may be a risk factor for developing this rare but particularly severe complication.

Background

Secondary sclerosing cholangitis following pyogenic cholangitis is usually listed among the many potential causes of biliary stricture formation (Table) [1,2]. However, in the literature indexed in Medline we could find only one such case, indicating that this is an extremely rare condition. We hereby report a second case, documented by cholangiography, of rapidly progressive sclerosing cholangitis secondary to bacterial cholangitis.

Case presentation

A previously healthy 48 year-old woman of Moroccan origin was admitted because of abdominal pain and nausea. A diagnosis of acute pancreatitis was made, based on elevated urinary and serum amylase levels and ultrasound imaging. Endoscopic retrograde cholangiopancreatography (ERCP) showed normal bile ducts, but suggested presence of biliary sludge, and biliary sphincterotomy was performed. In the following weeks four laparotomies were required for drainage of multiple pancreatic abcesses. Three months after the initial presentation, the patient was readmitted with jaundice, right upper quadrant ab-

dominal pain and fever, and a clinical diagnosis of cholangitis was made. Laboratory tests demonstrated a total serum bilirubin level of 53 μmol/l (normal: <17 μmol/l), and a serum alkaline phosphatase level of 1326 U/l (normal: <117 U/l). Blood cultures yielded E. Coli. ERCP (Figure 1) showed pus draining from the papillary orifice, multiple abcesses in connection with the biliary tree and several stones. Multiple soft stones were removed, and nasobiliary drainage and antibiotics were started. A subsequently performed CT-scan was compatible with the presence of these tiny abcesses. After two weeks ERCP (Figure 2) demonstrated narrowing of the left and right main hepatic ducts which was not present on the previous examinations. Six weeks later there was another episode of E. Coli cholangitis; cholangiography showed similar abnormalities. Ten months after the initial presentation the patient had a third episode of cholangitis; ERCP (Figure 3) then showed hilar biliary stenoses as well as multiple filliform strictures of segmental intrahepatic bile ducts associated with peripheral duct dilatation. These abnormalities were considered not to be amenable for endoscopic or percutaneous transhepatic treatment. Antibiotic treatment was continued and cholangitis did not recur during 14 months of follow-up. The patient is currently asymptomatic and has been evaluated for future liver transplantation. The most recent laboratory tests show a serum alkaline phosphatase level of 424 U/l and a total serum bilirubin level of 13 μmol/l.

Table: Causes of biliary strictures

Primary sclerosing cholangitis
Biliary ischaemia
Previous biliary surgery
Graft-versus-host-disease
Rejection after liver transplantation
Hepatic arterial infusion of antineoplastic agents
Cholangiocarcinoma
Choledocholithiasis
Bacterial cholangitis
AIDS-cholangiopathy
Sclerosing pancreatocholangitis
Portal vein thrombosis / cavernoma
Parasitic biliary infection
Oriental cholangiohepatitis
Lymphoma / hepatic or metastatic carcinoma
Amyloidosis

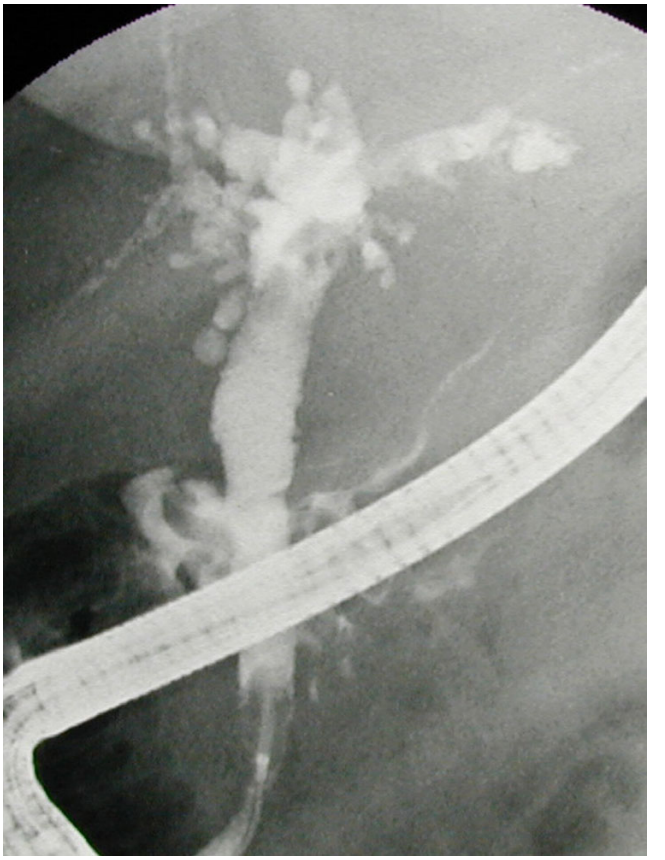

Figure 1
ERCP during the first episode of cholangitis This ERCP was performed during the first episode of cholangitis. There are multiple abcesses in connection with the biliary tree and several stones.

Discussion

This case clearly demonstrates the rapid development of sclerosing cholangitis after an episode of severe bacterial cholangitis. Although previous bacterial cholangitis is usually included in the differential diagnosis of sclerosing cholangitis, we have been unable to find more than a single reported case [3]. Tanaka et al. described a patient who developed multiple intrahepatic biliary strictures after pyogenic cholangitis caused by Klebsiella and Enterococcus, that occurred as a complication of colonoscopic polypectomy. In both this and our case no obvious predisposing factors were identified. In particular, they were previously, healthy, immunocompetent patients without evidence to suggest pre-existing bile duct abnormalities or hepato-biliary disease. In our patient however, one can speculate about a role of the previous pancreatic abcesses in causing bacterial infestation of the biliary tree, as well as a possible role for the previous sphincterotomy in facilitating retrograde colonization of the biliary tree. Since the patient had a normal biliary anatomy except for the previous sphincterotomy, and had no signs or symptoms suggesting the presence of persistent infection of the biliary tree, we consider the presence of chronic cholangitis unlikely. The isolated bacterial organisms are commonly involved in bacterial cholangitis [4,5]. At the time our patient presented with bacterial cholangitis, cholangiography and CT-imaging showed a picture suggesting the presence of

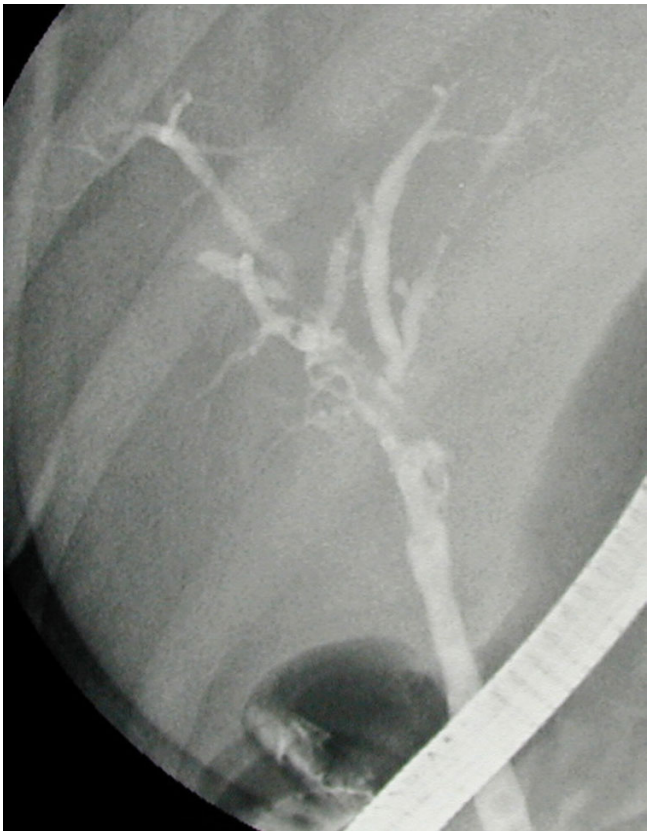

Figure 2
ERCP performed two weeks later This ERCP was performed two weeks after the first ERCP. The left and right hepatic ducts are narrowed.

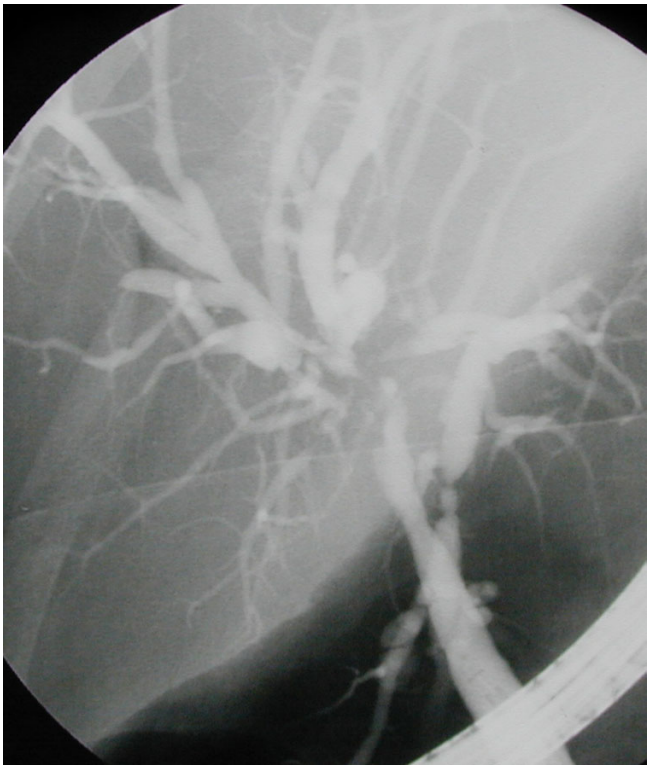

Figure 3
ERCP after 10 months This ERCP was performed ten months after the first ERCP. Hilar biliary stenoses and multiple filliform strictures of segmental intrahepatic ducts, associated with peripheral bile duct dilatation are shown.

multiple tiny parenchymal abcesses, communicating with the biliary tract. We speculate that this unusual extension of pyogenic cholangitis into the liver parenchyma may have played an etiological role in the subsequent development of biliary strictures, although another possibility is that the formation of these abcesses merely indicates the presence of severe cholangitis.

Our patient had repeated bouts of cholangitis once biliary strictures had developed. Antibiotic treatment and drainage led to rapid improvement on each occasion, but did not prevent new episodes. After maintenance antibiotic treatment was started, no further episodes of cholangitis occurred, and the patient remains in good clinical condition untill the moment of this writing. However, she seems to carry a considerable risk for developing future complications, including renewed bacterial cholangitis and development of secondary biliary cirrhosis.

Conclusions

Biliary strictures can develop as a complication of bacterial cholangitis. Extra-biliary hepatic involvement in the inflammatory process may be a risk factor for developing this rare complication.

Competing interests

None declared.

Authors' Contributions

PCJtB reviewed the patients history and the available literature, and drafted the manuscript. HRvB reviewed the patients history and the available literature, and critically reviewed and modified the manuscript. ACTMD was closely involved in the patients management, and critically reviewed the manuscript.

All authors read and approved the final manuscript.

Acknowledgements

Written consent was obtained from the patient for publication of the patient's details.

References

1. Sherlock S, Dooley J: **Diseases of the Liver and Biliary System.** *9th edn. London: Blackwell Scientific Publications* 1994, 237-48
2. Vleggaar FP, van Buuren HR, Lameris JS: **Bile duct lesions in portal vein thrombosis.** *Ned Tijdsch Geneesk* 1999, **143:**2057-2062

3. Tanaka Y, Koshiyama H, Nakao K, Makita Y, Kobayashi Y, Yoshida Y, Kimura M, Adachi Y: **Rapid progress of acute suppurative cholangitis to secondary sclerosing cholangitis sequentially followed-up by endoscopic retrograde cholangiography.** *Endoscopy* 2001, **33:**633-5
4. Maluenda F, Csendes A, Burdiles P, Diaz J: **Bacteriological study of choledochal bile in patients with common bile duct stones, with or without acute suppurative cholangitis.** *Hepatogastroenterology* 1989, **36:**132-5
5. Shimada K, Noro T, Inamatsu T, Urayama K, Adachi K: **Bacteriology of acute obstructive suppurative cholangitis of the aged.** *J Clin Microbiol* 1981, **14:**522-6

Pre-publication history

The pre-publication history for this paper can be accessed here:

http://www.biomedcentral.com/1471-230X/2/14/pre-pub