

Case report

Spontaneous perforation of the bile duct

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Background

Spontaneous perforation of the bile ducts is a rare condition in adults. It is commonly secondary to gallstones, and the site of the perforation is nearly always extrahepatic. Intrahepatic perforation has only once been described in the literature to date.

Case outline

We report an unusual presentation of this condition with perforation occurring at an intrahepatic site. The manage-

ment of perforation and the possible predisposing factors are described.

Discussion

Adequate management of this problem requires an awareness of its existence and prompt, appropriate investigation to discover the cause and site of perforation.

Keywords

bile duct, perforation, cholelithiasis, periampullary diverticulum.

Introduction

Spontaneous perforation of the bile ducts is a rare event. It is most commonly described in children. In adults the site of perforation is nearly always extrahepatic. We report the presentation and subsequent management of an adult woman with a bile duct perforation within the liver; this is only the second such case reported in the literature to date.

Case report

An 80-year-old woman presented to her local hospital with a 5-day history of epigastric pain and obstructive jaundice, which was confirmed biochemically with a bilirubin of 118 $\mu\text{mol/L}$ (normal range <17) and alkaline phosphatase of 208 IU/L (normal range <130). Serum transaminases were normal. She had previously been well with no relevant past medical history and in particular no symptoms suggestive of cholelithiasis. An ultrasound scan demonstrated a thickened gallbladder wall, multiple gallstones, a dilated common bile duct (12 mm diameter) and distal obstructing choledocholithiasis. In addition, there was a peri-splenic fluid collection, which was drained under computed tomography (CT) guidance. The fluid was heavily bile-stained, and a pigtail drainage catheter was left in situ. A magnetic resonance cholangiopancreatogram (MRCP) did not add any extra information, and endoscopic retrograde cholan-

giopancreatogram (ERCP) was unsuccessful because of a periampullary diverticulum.

She was transferred to this unit. On arrival she was well, afebrile and not jaundiced, with no signs of peritonitis. Repeat CT scan confirmed extrahepatic biliary dilatation, a small left hepatic lobe, distal common bile duct stone and continued peri-splenic fluid collection (Figure 1). ERCP was repeated and, although technically difficult, it successfully cannulated the bile duct. A 12-mm stone was demonstrated in the distal common bile duct with a bile leak from the parenchyma of the left lobe of the liver

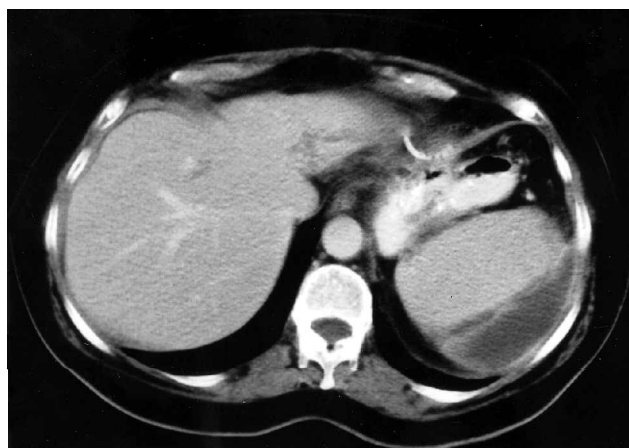


Figure 1. CT scan demonstrating the peri-splenic fluid collection and the small left lateral segments of the liver.

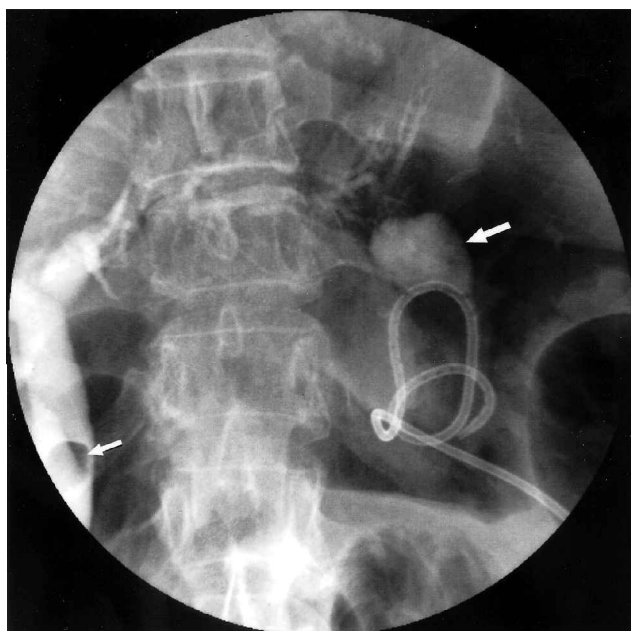


Figure 2. ERCP demonstrating the distal common bile duct stone (small arrow), bile duct dilatation and left lobe leak from the hepatic parenchyma (large arrow).

(Figure 2). She subsequently progressed to an open cholecystectomy with exploration of the common bile duct, distal stone retrieval and insertion of a T-tube. Dense inflammatory reaction under the left lobe of the liver prohibited any formal repair of the parenchymal leak. The gallbladder was noted to be inflamed and full of stones.

She made a full recovery from operation. T-tube cholangiography performed 3 weeks later demonstrated no filling defects, a normal calibre biliary system, free flow into

the duodenum and no evidence of any continuing bile leak from the left lobe parenchyma (Figure 3). The T-tube was removed uneventfully.

Discussion

Spontaneous perforation of the bile duct has been reported in small series in children. Chardot reported 11 patients over a 23-year period, and Spigland reviewed the literature on <100 patients worldwide in 1996 [1,2]. Most cases are not actually spontaneous but occur secondary to underlying pathology. Predisposing factors have included stone disease, protein plugs, distal atresia and possibly viral gastro-enteritis. Approximately 50 cases have been reported in the literature among adults [3], nearly all being perforation of the extrahepatic biliary system. This is only the second report of parenchymal (i.e. intrahepatic) perforation. Most adult cases (70%) are associated with obstruction to the biliary tract by stone disease [3]. In the extrahepatic bile ducts the common bile duct and cystic/hepatic duct junction are the commonest sites of perforation (excluding the gallbladder), but this phenomenon has been described everywhere in the biliary tract including congenital anomalies such as choledochal cysts [5].

All reported patients have either been treated surgically or have been diagnosed post mortem. Not all cases lead to overt biliary peritonitis, and the diagnosis is sometimes made at laparotomy. Surgical treatment has been drainage of the biliary tract, drainage plus exploration or exploration plus repair. Roux-en-Y loop bilio-enterostomy, although performed in children, is rarely carried out for spontaneous perforation in adults. In our patient the perforation appears to have been precipitated by a rise in intraductal pressure following obstruction by a distal common bile duct stone. This sequence may have been influenced by a high resting intrabiliary pressure due to the presence of a periampullary diverticulum [6,7]. Another possible contributing factor was the congenitally small left lateral segments. Alternative pathological explanations suggested in other patients have included necrosis of the duct wall due to direct erosion by stones, reflux of potentially damaging pancreatic secretions, spasm of the sphincter of Oddi, intramural infection or necrosis secondary to intramural vascular thrombosis [8].

Adequate management requires an awareness of the possibility of spontaneous bile duct perforation in adults. ERCP can demonstrate the site of the leak and also may diagnose and treat any obstructing cause. It offers advan-



Figure 3. Postoperative T-tube cholangiogram showing resolution of the parenchymal bile leak.

tages over MRCP in this situation as it offers a therapeutic option of duct drainage and stone retrieval. In the present case, the first ERCP failed because of difficulty cannulating the biliary system through a periampullary diverticulum. A surgical approach was performed once stone disease was established by a second ERCP. Stone retrieval, stent insertion or endoscopic sphincterotomy may all be potential alternative treatments in patients considered unfit for biliary tract surgery.

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