Cystic duct directly joining the main pancreatic duct

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Abstract
A unique anomaly of the direct union between the cystic duct and the main pancreatic duct is presented. A 19-year-old man with a history of repeated epigastralgia underwent endoscopic retrograde cholangiopancreatography that showed a direct union between the cystic duct and the main pancreatic duct. No pancreaticobiliary maljunction was noticed. Cholecystectomy accompanied by resection of the long cystic duct was performed. The excised gallbladder showed cholesterolosis, chronic cholecystitis, and hyperplasia of the pseudopyloric glands microscopically. The patient has been well for 3 years since surgery.

Key words Cystic duct anomaly · Pancreaticobiliary maljunction · Pancreatic bladder

Introduction
Anatomical variations of the cystic duct are significant for hepatobiliary surgery. They are usually associated with an anomalous union with the hilar bile ducts or a low junction to the extrahepatic bile duct, but hardly ever with the pancreatic duct. In this report, we present a case of a unique anomaly of the cystic duct that joined the main pancreatic duct directly, whereas the extrahepatic bile duct presented a normal anatomy.

Case report
A 19-year-old Japanese man presented with sudden epigastralgia. He had experienced the same symptom once a year since he was 12 years old. He had been diagnosed with cholecystolithiasis by ultrasonography (US) 2 years earlier. However, no further examination or surgical treatment was performed because the symptoms disappeared spontaneously. On admission, a physical examination showed tenderness and muscular defense in the epigastric region. A series of laboratory tests revealed normal values in the liver enzymes, white blood cell count, and C-reactive protein. US detected a slightly distended gallbladder with high echoic lesions without acoustic shadow. Computed tomography showed small high-density lesions in the gallbladder. Endoscopic retrograde cholangiopancreatography (ERCP) was conducted for a close examination of the bile duct. This first revealed the main pancreatic duct, from which the gallbladder was shown through a long cystic duct (Fig. 1A). Cannulation into the cystic duct was possible without passing through the bile duct. There was no maljunction between the main pancreatic duct and the bile duct (Fig. 1B). Based on the imaging information, a diagnosis of maljunction between the cystic duct and the main pancreatic duct was made.

In order to prevent acute cholecystitis and carcinogenesis of the gallbladder in the future, a cholecystectomy was performed. Laparotomy showed an almost normal gallbladder. Both the extrahepatic bile duct and the gallbladder were punctured to collect bile juice for biochemical analysis. The cholecystectomy was performed, accompanied by dissecting the long cystic duct into the pancreas parenchyma. The cystic duct was dissected as far as possible down to the pancreatic duct. The cystic duct was parallel to the dorsal side of the extrahepatic bile duct. Intraoperative contrast medium infusion into the cystic duct showed the main pancreatic duct, but the extrahepatic bile duct was not shown (Fig. 2). Pancreatic amylase isozyme levels in a whitish fluid in the gallbladder and bile juice in the extrahepatic bile duct were 664524 IU/l and 13 IU/l, respectively. These findings strongly suggested a direct connection between the cystic duct and the pancreatic duct without an anomalous union between the extrahepatic bile duct and the pancreatic duct. Histopathological examination

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Infrared spectrophotometry (Herschel FT/IR-410; JASCO, Tokyo, Japan) by SRL Inc. showed fatty acid calcium and protein in the debris of the gallbladder.

Discussion

We often encounter anatomical variations in the extrahepatic bile duct during hepatobiliary surgery. Recognition of these variations is extremely significant, because they often result in intraoperative bile duct injuries. Pancreaticobiliary maljunction (PBM), a complicated anomalous union between the extrahepatic bile duct and the pancreatic duct, is a well-known pathological condition, but it is rarely associated with the cystic duct only. Anatomical variations of the cystic duct are usually associated with the hilar bile ducts or a low joint to the extrahepatic duct. Only two cases, of an anomalous direct union between the cystic duct and the pancreatic duct, are available in the literature. These two cases presented the same direct union between the cystic duct and the main pancreatic duct as in the present case and the authors named this anomaly “pancreatic bladder.”

The developmental mechanism of the anomaly in the present case cannot be fully explained, but the report suggested an embryonic disorder. The anomalous union between the cystic duct and the pancreatic duct in the ventral pancreas develops when an embryo is 8mm in size. Thereafter, the ventral pancreas rotates and completes fusion into the dorsal pancreas when the embryo grows to 12mm. The two pancreatic ducts then join with each other, and this unique anomaly is finally developed.