Anatomic variations

Double common bile duct with ectopic drainage into the stomach. Case report and review of the literature

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Abstract

Abstract: A rare abnormal biliary tract consisting in a double common bile duct with an ectopic biliary tree draining into the stomach is described. This congenital anomaly, associated with lithiasis in the ectopic duct, was detected for the first time on MR-cholangiopancreatography. Only 23 cases of abnormal biliary drainage into the stomach have been reported in the literature. Embryogenesis and potential risks, such as lithiasis in the ectopic duct and the development of gastric carcinoma, are discussed.

Anatomical variations of the biliary tree are numerous and frequent [1, 2, 4, 6]. However, biliary drainage into the stomach is an extremely rare congenital anomaly, which may be visualized on non-invasive MR-cholangiopancreatography (MRCP).

Case report

A seventy-year-old woman was admitted for epigastric pain and dyspepsia which had persisted for 18 months. A review of her medical history showed that primary sclerosing cholangitis (PSC) was suspected in 1991 after the visualization of multifocal biliary strictures, involving both the intrahepatic and extrahepatic ducts on endoscopic retrograde cholangiopancreatography (ERCP). She was treated by ursodesoxycholic acid at that time undergoing laparoscopic cholecystectomy and choledocotomy for acute cholecystitis and choledocholithiasis in 1997. In 1999, upper gastrointestinal endoscopy revealed the presence of bile juice in the stomach and intense hyperemia of the gastric mucosa no hole or mucosal ulceration was observed. Biopsy specimens showed hyperplastic gastritis. MRCP showed an atypical communication (2 cm wide, 10 cm long) containing stones between the left biliary duct and the lesser curvature of the stomach (Fig. 1). Unlike the 1991 findings there were no major alterations of the main biliary tract, which ended normally in the duodenum pancreatic structures were normal. Laparotomy (May 18, 1999) confirmed the presence of an abnormal duct located at the upper part of the hepatogastric ligament (Fig. 2). Peroperative catheterization and opacification of the atypical duct confirmed a large communication between the left hepatic duct and stomach (Figs. 3, 5). Resection of the abnormal duct revealed five stones intramurally on the gastric side. Cholangiojejunostomy was performed between the hilar side of the atypical duct and a Roux-Y loop the postoperative course was uneventful. Histopathologic examination confirmed that the resected specimen had the same structure as a normal biliary duct, but without a sphincter at the gastric junction. Pathological findings on liver biopsy were consistent with low-grade PSC. Upper gastrointestinal endoscopy one year later confirmed the disappearance of the gastric mucosal lesions.

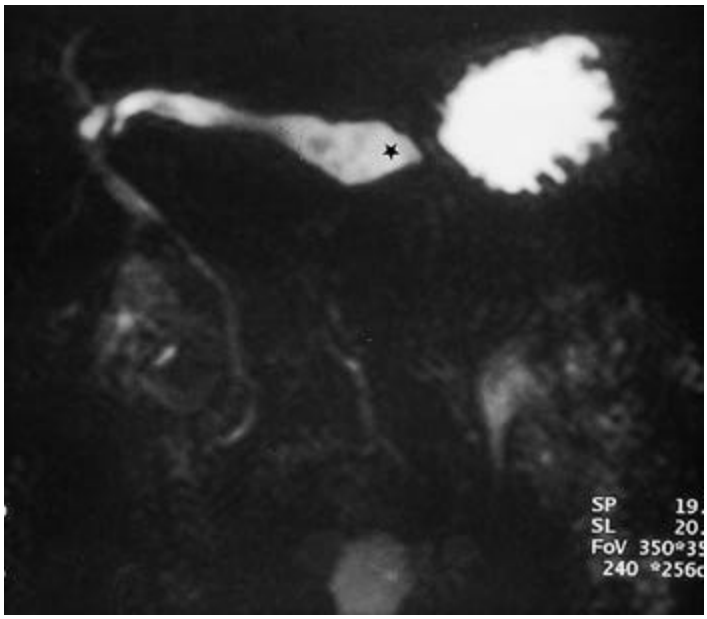

Fig. 1 MR-cholangiopancreatography Atypical communication (*) containing stones, between the left biliary duct and the lesser curvature of the stomach

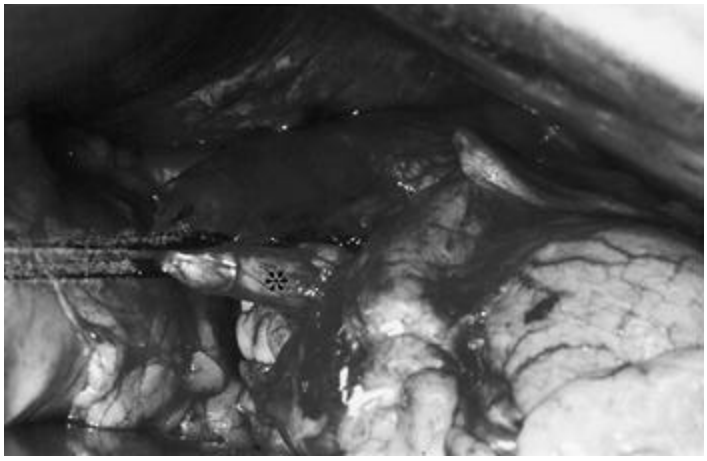

Fig. 2 Peroperative view showing the atypical duct (*)

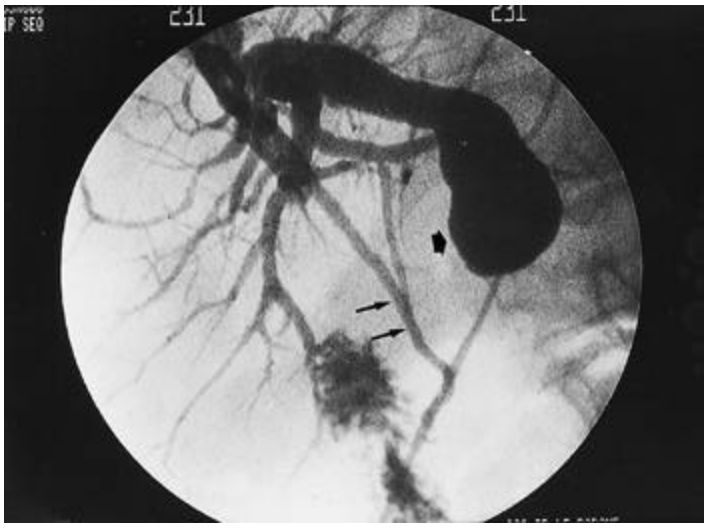

Fig. 3 Peroperative opacification of the abnormal biliary duct between the left biliary duct and stomach (*ﬁ*).The normal biliary tract ending in the duodenum is indicated by 2 arrows

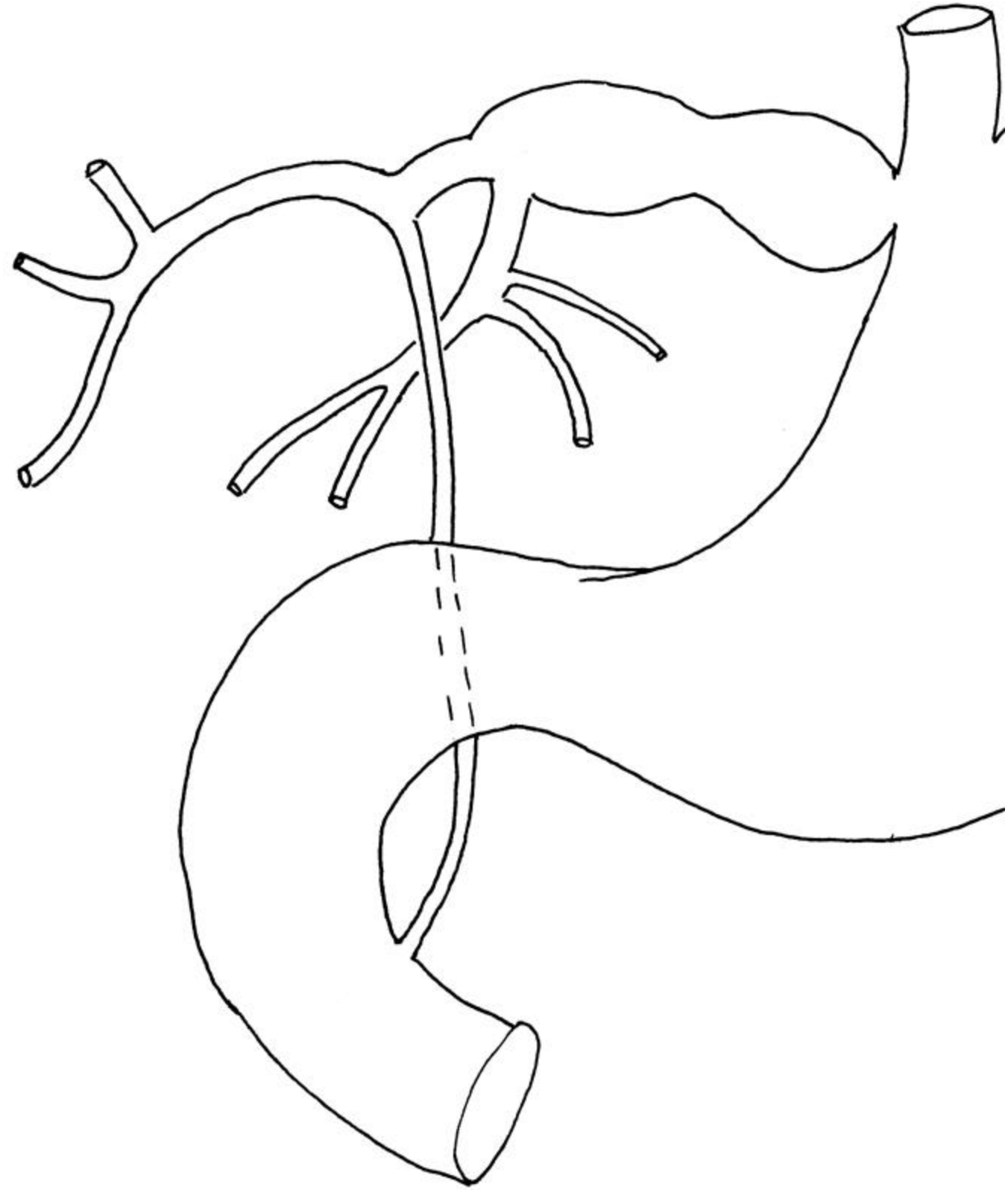

Fig. 5 Drawing of the findings in the present case

Discussion

Ectopic drainage of the biliary tract is a very uncommon congenital anomaly which includes abnormal junction of the common bile duct, a cystic duct or intrahepatic duct with the duodenum, stomach, esophagus, trachea and main pancreatic duct [7, 8, 9, 18]. In 1992 Kanematsu et al [9] reported on 56 cases of drainage of the common bile duct into the gastrointestinal tract, with 52% (n = 25) being into the duodenum, 46% (n = 22) into the stomach and 2% (n = 2) into the main pancreatic duct. In cases with gastric drainage there appear to be two configurations, either a single bile duct ending in the stomach [3, 12] or a double common bile duct [8, 10, 14, 17]. Double common bile duct is characterized by the coexistence of normal drainage into the duodenum and ectopic drainage into the stomach, the ectopic duct being continuous with the left hepatic duct, as in the present case. Horsmans et al [7] published a review of 12 cases of double common bile duct in 1996. The location of the gastric junction has been determined in 20 cases of gastric drainage [9] and found to be in the antrum (n = 4), lower and middle body (n = 8), and, as in the present case, upper body (n = 3). Histological examination of 11 ectopic ducts showed the absence of a sphincter structure at the gastric junction [9], as in the present case. There may also be other associated congenital biliary anomalies absence of the gallbladder, development of a bile duct cyst on the non-ectopic duct, and duct stricture [9, 14]. Congenital duodenal anomalies such as duodenal atresia or multiple diverticuli of the first portion of the duodenum have also been described [9, 12]. The development of the pancreatic ducts appears to be independent and they remain unmodified in cases of gastric drainage of the biliary tract [7].

The normal development of the biliary tract is related to the diverticulum hepaticum, the cranial part (pars hepatica) of which gives rise to the intrahepatic and common hepatic ducts. The anomalies of biliary tree drainage may be due to disruption of the diverticulum hepaticum [5]. If a subdivision of the diverticulum hepaticum occurs very early, the pars hepatica will lie above the growth zone between the stomach and duodenum (Fig. 4). Consequently, the pars hepatica will develop into a duct emptying into the stomach [7]. Obliteration of the common bile duct during fetal life may explain the persistence of a single ectopic duct, but the concept of a solid stage of endodermal occlusion of the common bile duct lumen is controversial [15]. A classification of congenital anomalies associated with biliary drainage has been proposed, specifying four types [4, 9] Type I, septum in the lumen of the common bile duct Type II, division of the common bile duct into two separate junctions, as in the present case Type III, double independent drainage and Type IV, double drainage with one or more intercommunications (Fig. 6).

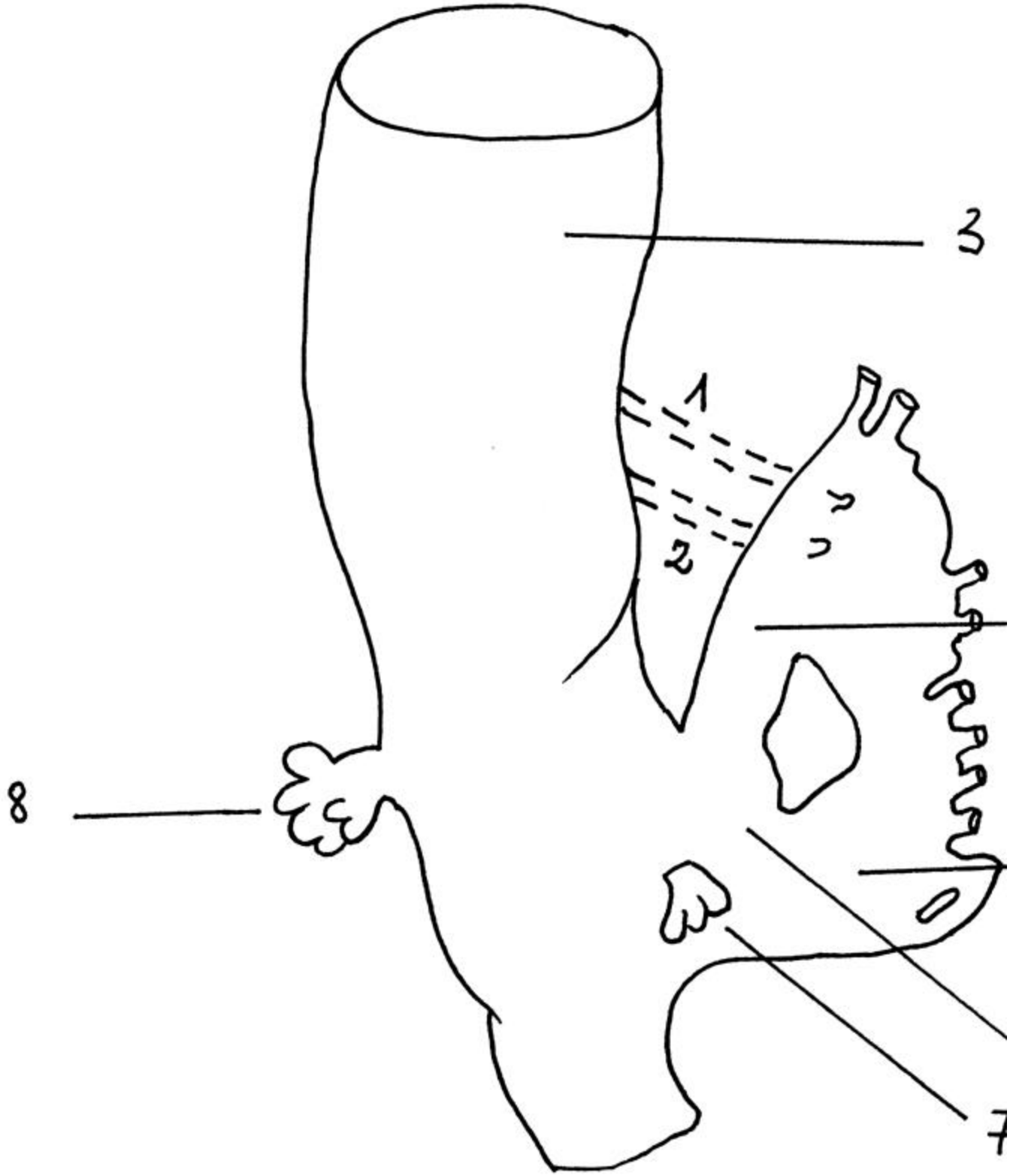

Fig. 4 Reconstruction of the biliary system in a 5 weeks human embryo (from Hayes et al [5]). *1* and *2*, possible anomalies wih ectopic drainage into the stomach *3*, stomach *4*, common hepatic duct *5*, gallbladder *6*, cystic duct *7*, ventral pancreas *8*, dorsal pancreas

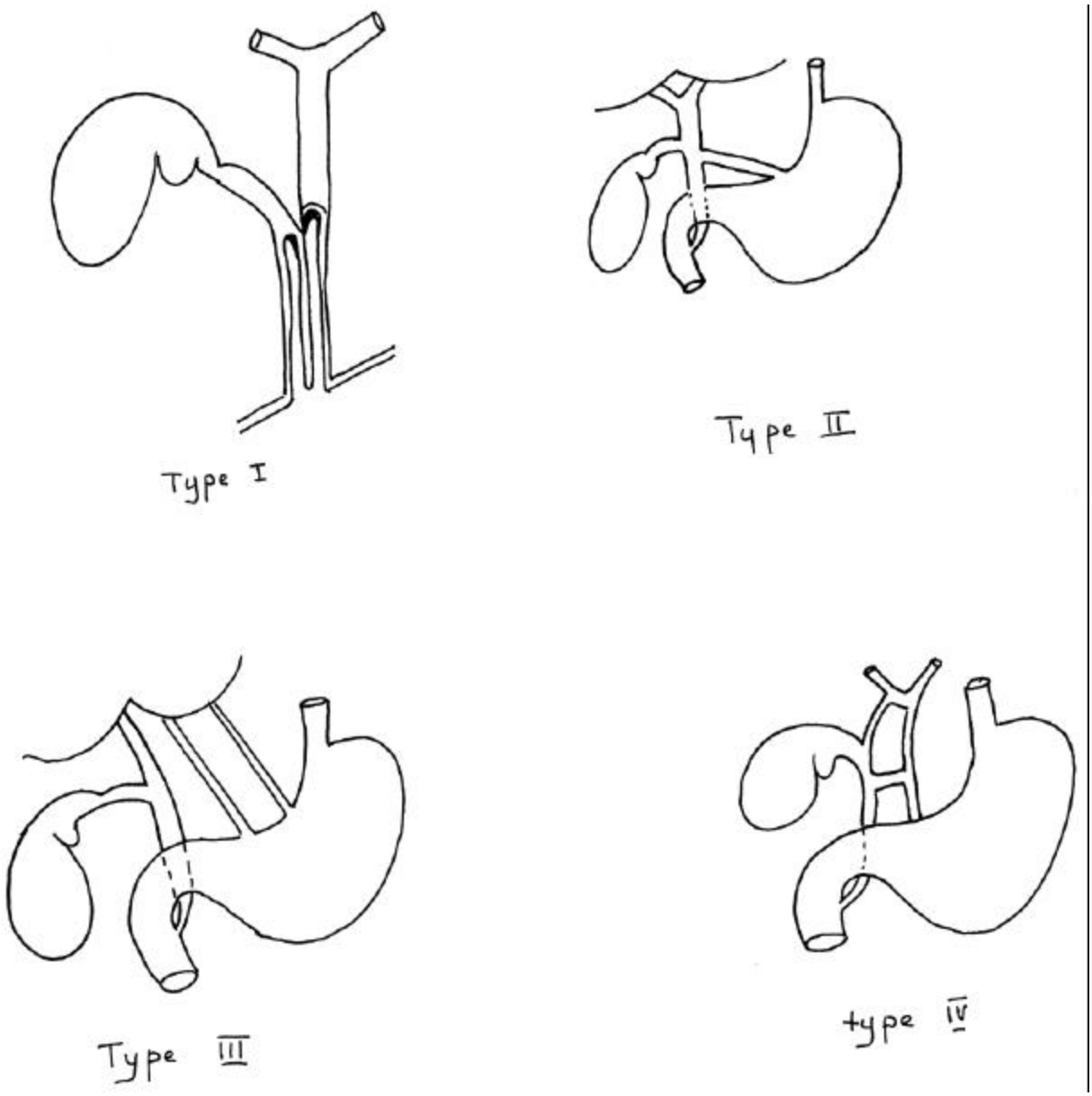

Fig. 6 Classification of congenital anomalies of the common bile duct (4 types taken from Kanematsu et al [9])

The most common presenting symptom is mild to moderate epigastric abdominal pain, followed by dyspepsia, fever and jaundice. Preoperative diagnosis is based on radiological and endoscopic findings. In some cases, gastroscopy shows an intragastric biliary opening with or without bile flow [9, 18]. The first case detected by ERCP was reported in 1966 [7], however the anomaly in the present case was not detected in an ERCP performed 8 years earlier in another centre. MRCP is an non-invasive investigation particularly adapted to the assessment of biliary and pancreatic duct anatomy. The case reported here appears to be the first of ectopic drainage into the stomach diagnosed on MRCP.

There are some complications associated with gastric drainage of the biliary tract. Biliary lithiasis with stones in the abnormal duct, as in the present case, may be complicated by cholangitis and jaundice, as well as pancreatitis and liver abcess [9, 10]. Prolonged exposure of the gastric mucosa to bile juice may be responsible for atrophic gastritis and predispose to the development of gastric cancer. Five cases of gastric carcinoma have been reported (4 in the Japanese literature) one case of exclusive drainage of the common bile duct into the stomach [12], two cases with double common bile duct [11], and two cases published in Japanese and cited in [9]. Therefore, if double common bile duct with ectopic drainage into the stomach is confirmed on MRCP, a careful endoscopic examination and biopsy of the gastric mucosa surrounding the opening should be performed [11].

Management of this anomaly is controversial, with most symptomatic patients being treated surgically [9]. In cases of an

ectopic biliary duct ending in the stomach the surgical procedure employed has involved conservation [7, 9], ligation [11, 12], or duodenal reimplantation [13]. Resection of the ectopic duct and cholangiojejunostomy between the biliary section and the Roux-Y loop, as in the present case, appears to be the best procedure for the prevention of biliary complications and potential risk of gastric carcinoma.

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