

Duplication of the Extrahepatic Bile Duct in Association with Choledocholithiasis as Depicted by MDCT

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We report here on an extremely rare case of duplicated extrahepatic bile ducts that was associated with choledocholithiasis, and this malady was visualized by employing the minimum intensity projection images with using multi-detector row CT. The presence of duplicated extrahepatic bile ducts with a proximal communication, and the ducts were joined distally and they subsequently formed a single common bile duct, has not been previously reported.

Index terms:

Bile ducts, abnormalities
Bile ducts, calculi
Computed tomography (CT),
minimum intensity projection

DOI:10.3348/kjr.2008.9.6.550

Korean J Radiol 2008; 9: 550-554

Received February 11, 2008; accepted
after revision May 23, 2008.

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Congenital duplication of the extrahepatic bile duct is a very rare anomaly and this has been reported in only 24 cases in the clinical literature during the last 500 years until 1986 (1).

Recognition of this anomaly is clinically important as it can lead to complications such as choledocholithiasis, cholangitis, pancreatitis and upper gastrointestinal malignancies (2). In addition, this anomaly is often accompanied with anomalous union of the pancreaticobiliary ductal system (AUPBD) and the presence of a choledocal cyst (2). Most cases of duplicated extrahepatic bile ducts that have been reported up to now were diagnosed based on the abnormalities that were found by endoscopic retrograde cholangiography (ERC), MR cholangiopancreatography (MRCP) or operative cholangiography (1-8).

We present here a case of an adult patient who had duplication of the extrahepatic bile duct along with choledocholithiasis, as was revealed by the minimum intensity projection (MinIP) images of multi-detector row CT (MDCT). To the best of our knowledge, this is the first reported case of the Vb type, according to the modified classification of duplication of the common bile duct (CBD) as proposed by Choi et al. (5).

CASE REPORT

An 81-year-old woman presented with a 2-day history of chills and fever. She had been admitted two years ago with jaundice and fever. On a CT scan, the distal CBD, a large gallbladder (GB) and cystic duct stones were revealed. Urgent endoscopic retrograde cholangiopancreatography (ERCP) to remove the distal CBD stone was performed. Multiple CBD stones were extracted using a basket and balloon sweep. After the stones were removed, there was no filling defect on the CBD during balloon cholangiography. The patient refused cholecystectomy. She was not followed up after discharge from the hospital. On the next presentation, the laboratory tests revealed a slightly elevated aspartate aminotransferase level (45 IU/L) and an increased value of C-reactive protein (15.3 mg/l). The total bilirubin level, the alkaline phosphatase level,

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the gamma-glutamyltransferase level and the WBC count were all within the normal ranges. The presumed diagnosis was recurrent CBD stones, and an ERCP for recurrent CBD stones was planned. Transabdominal ultrasonography demonstrated a dilated extrahepatic bile duct filled with echogenic lesions that showed posterior acoustic shadowing, which was suggestive of bile duct stones (Fig. 1A). On the more medial view of the sagittal sonogram, another mildly dilated extrahepatic bile duct was seen, but no stone was noted (Fig. 1B). The sonographic findings led

us to presume that the stone-filled aberrant right hepatic duct drained into the common hepatic duct. A contrast-enhanced CT scan was performed using a 64-MDCT scanner (LightSpeed VCT, GE Healthcare, Milwaukee, WI) and it showed two separate extrahepatic bile ducts (Fig. 1C). Multiple stones were noted in a posterolaterally located bile duct and these stones extended to right intrahepatic bile ducts. There was a large stone in the GB and the cystic duct joined to a posterolaterally-located extrahepatic bile duct. The MinIP images were obtained on



Fig. 1. 81-year-old woman with duplication of extrahepatic bile duct.
A. Sagittal sonography shows dilated extrahepatic bile duct (white arrows) filled with shadowing echogenic materials (between electronic calipers).
B. On more medial view, another extrahepatic bile duct is seen (white arrows). No intraluminal lesion is found in this duct.
C. Contrast-enhanced axial CT scan shows two separate extrahepatic bile ducts (arrows), one of which was filled with stones (white arrow).
D. Minimum intensity projection image shows proximal and distal unions of duplicated extrahepatic bile ducts (white arrows). Also note stones in laterally located extrahepatic bile duct and right intrahepatic bile ducts. These ducts show higher attenuation than that of water (black arrows).

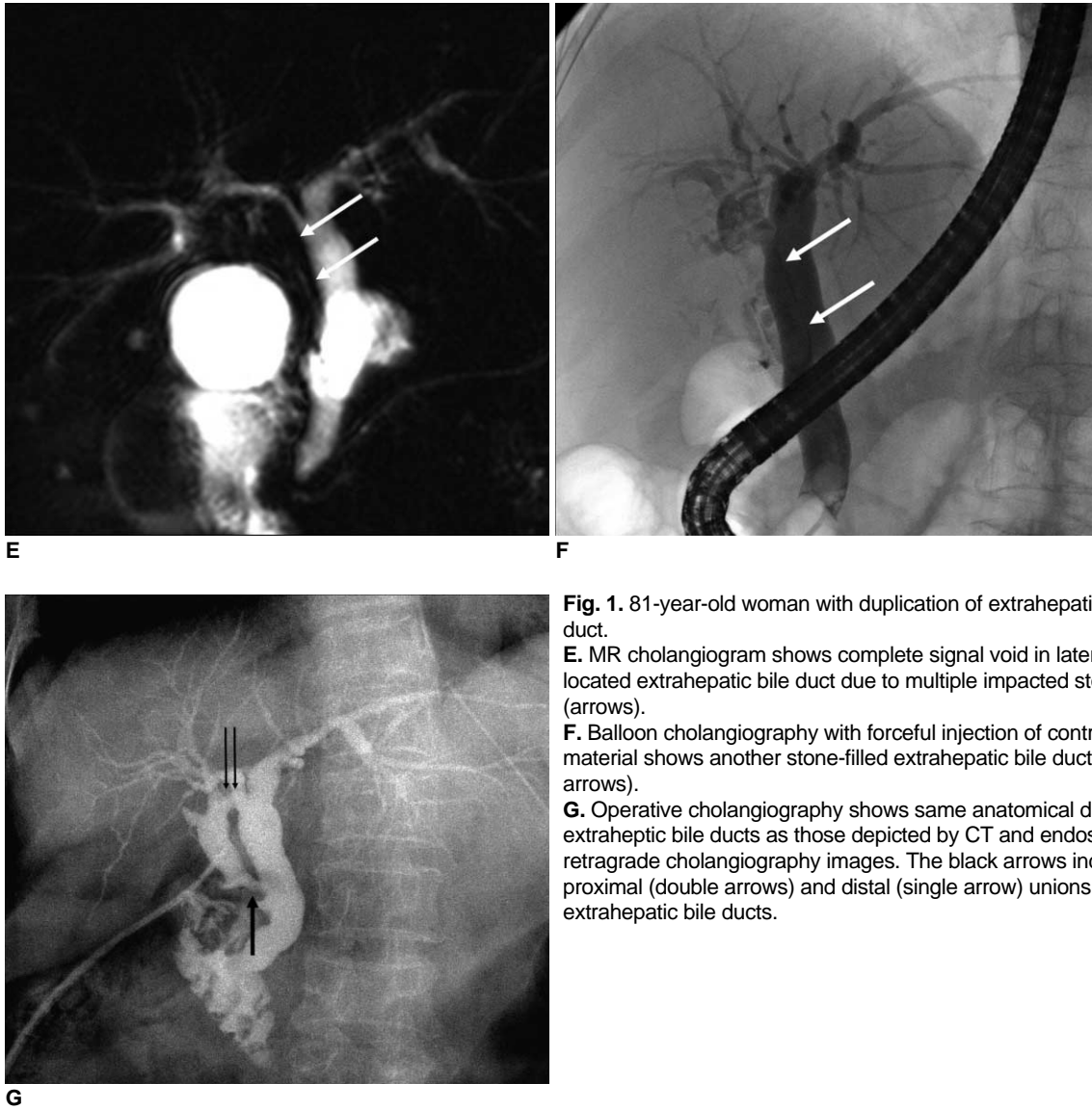


Fig. 1. 81-year-old woman with duplication of extrahepatic bile duct.

E. MR cholangiogram shows complete signal void in laterally located extrahepatic bile duct due to multiple impacted stones (arrows).

F. Balloon cholangiography with forceful injection of contrast material shows another stone-filled extrahepatic bile duct (white arrows).

G. Operative cholangiography shows same anatomical details of extrahepatic bile ducts as those depicted by CT and endoscopic retragrade cholangiography images. The black arrows indicate proximal (double arrows) and distal (single arrow) unions of double extrahepatic bile ducts.

a postprocessing workstation (Advantage Windows workstation [version 4.3], GE Healthcare, Milwaukee, WI). The MinIP image (coronal oblique with a 5.3 mm thickness slab) revealed double extrahepatic bile ducts that combined to create a CBD in the pancreatic head portion with a communicating channel in the hilar portion (Fig. 1D). MRCP performed with a 1.5-T Intera scanner (Philips Medical Systems, The Netherlands) failed to depict the precise anatomy of this anomaly due to the impacted stones (Fig. 1E). There was no AUPBD on the MRCP. ERC was subsequently performed to remove the multiple stones in one of the double extrahepatic bile ducts. An ERC image also disclosed duplication of the extrahepatic bile duct with multiple stones in the posterolaterally located duct and the right intrahepatic ducts (Fig. 1F). However, we failed to gain access to the right CBD and to remove the CBD stones.

To remove the remnant stones, duct exploration with insertion of a T-tube and cholecystectomy was performed. On a cholangiogram obtained during the subsequent operation, there was no evidence of remnant stones in the bile ducts (Fig. 1G). The patient's postoperative recovery was uneventful.

DISCUSSION

Duplication of the extrahepatic bile duct is an extremely rare condition (2). The developmental failure for the double biliary system to regress, and this double system is present in early normal embryogenesis, is considered to be the mechanism of this anomaly (5). The morphological classification of a double extrahepatic bile duct has been modified because the newly reported cases could not be included in the existing classification system. Choi et al.

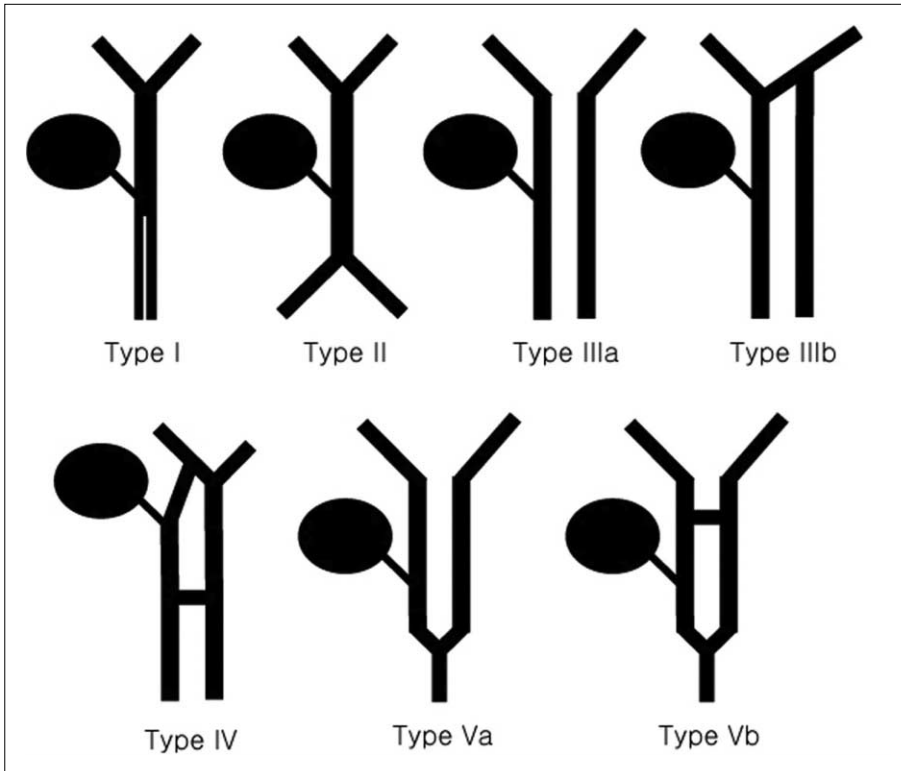


Fig. 1. 81-year-old woman with duplication of extrahepatic bile duct.
H. Modified classification system of duplicated extrahepatic bile duct as proposed by Choi et al. (5).

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(5), when reporting on a type Va case, added types Va and Vb to the classification system that was modified by Saito et al. (9). The individual subtypes of the modified classification system are as follows (5) (Fig. 1H): type I, a CBD with a septum in the lumen; type II, a CBD that bifurcates and drains separately; type III, double biliary drainage without extrahepatic communicating channels (without [a] or with intrahepatic communicating channels [b]); type IV, double biliary drainage with one or more extrahepatic communicating channels; type V, single biliary drainage of double extrahepatic bile ducts without (a) or with communicating channels (b). Our case corresponded with a type Vb anomaly.

The clinical issues for these anomalies are the combined complications and the concomitant AUPBD. In a review of the Japanese clinical literature by Yamashita et al. (2), the investigators found cholelithiasis in 28% of the cases, a choledochal cyst in 11% of the cases, AUPBD in 30% of the cases and cancer in 26% of the cases. These investigators also emphasized that the opening site of the accessory bile duct was associated with a type of cancer and the concomitant presence of AUPBD. Our case is a form of single biliary drainage in the second portion of the duodenum; no evidence of AUPBD was found by MRCP. Although multiple stones were impacted in one of the double extrahepatic bile ducts, there was no evidence of cholestatic findings based on the laboratory results nor was

intrahepatic biliary dilatation found in our case. This may be ascribed to the presence of a communicating channel between the two extrahepatic bile ducts that provided another way for biliary excretion.

Making a correct diagnosis of these anomalies prior to biliary surgery is clinically important due to the risk of biliary injury during the operation. Among the seven cases that were recently reported (2–8), four cases were detected intraoperatively (2–4, 8). The three remaining cases were diagnosed by means of MRCP or ERCP. MRCP is a safe, noninvasive imaging technique and it provides similar diagnostic information as compared with ERCP, which is the standard of reference for biliary imaging (10). Although MRCP has also played an important role in the diagnosis of double extrahepatic bile ducts (5, 6), it did not provide detailed anatomical information about the biliary tree due to the multiple impacted stones in the current case. Instead, MDCT imaging using the MinIP technique was very helpful for precisely depicting the complex anatomical details of this anomaly.

Recent advances in MDCT technology enable physicians to acquire a large volume of images rapidly and these advances also facilitate postprocessing applications with high spatial resolution due to the thin collimation. MinIP is one of the postprocessing techniques, and it displays the lowest attenuated voxel within a slab and so it is useful for imaging the biliary tree that has a water-density. The

usefulness of MinIP for making the diagnosis of biliary obstruction has been reported in the radiological literature (11, 12). This technique was more useful than axial CT scanning or MRCP to demonstrate the detailed anatomical information such as communicating channels or a stone-filled duct those in our case.

In conclusion, we report here on a type Vb case of the duplication of the extrahepatic bile duct that was associated with choledocholithiasis. In this case, obtaining the MDCT images using the MinIP technique played a decisive role in the preoperative diagnosis and therapeutic planning.

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