Using MR Cholangiopancreatography to Reveal Anomalous Pancreaticobiliary Ductal Union in Infants and Children with Choledochal Cysts

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OBJECTIVE. The purpose of this study was to determine whether MR cholangiopancreatography can accurately depict anomalous pancreaticobiliary ductal union in children with choledochal cysts.

SUBJECTS AND METHODS. Twenty children (age range, 1 month–13 years; mean age, 4.6 years; all girls) who were diagnosed with choledochal cyst by sonography underwent MR cholangiopancreatography with a single-shot fast spin-echo sequence. The type of choledochal cyst and anomalous pancreaticobiliary ductal union were characterized on the basis of MR cholangiopancreatographic findings and were compared with the finding of intraoperative cholangiography.

RESULTS. The type of choledochal cyst (type Ia, $n = 4$; type Ic, $n = 7$; type IVa, $n = 7$; type IVb, $n = 2$) determined on MR cholangiopancreatography correlated with that identified on intraoperative cholangiography in each patient. Anomalous pancreaticobiliary ductal union was detected by MR cholangiopancreatography and intraoperative cholangiography in 12 (60%) and 16 (80%) of 20 patients, respectively. The types of anomalous pancreaticobiliary ductal union as determined on MR cholangiopancreatography (type A, $n = 2$; type B, $n = 7$; type C, $n = 3$) were concordant with those of intraoperative cholangiography in 11 of 12 patients. In five of eight patients with choledochal cyst (type Ia, $n = 1$; type IVa, $n = 5$; type IVb, $n = 2$) in whom MR cholangiopancreatography could not depict anomalous pancreaticobiliary ductal union, anomalous pancreaticobiliary ductal union was documented on intraoperative cholangiography that was performed after choledochal cyst resection.

CONCLUSION. MR cholangiopancreatography provides diagnostic information about anomalous pancreaticobiliary ductal union in children with choledochal cyst.

Choledochal cyst, which is usually diagnosed in infants and children, requires preoperative radiologic evaluation to be resected completely. Sonography is the method of choice in the initial evaluation of choledochal cyst. However, sonography cannot reveal anomalous pancreaticobiliary ductal union, which is generally believed to be a cause of choledochal cyst [1, 2]. Surgeons need an exact anatomic map of the pancreaticobiliary ductal union because it is essential that the choledochal cyst be completely resected without pancreatic ductal injury. In terms of preoperative evaluation of anomalous pancreaticobiliary ductal union, endoscopic retrograde cholangiopancreatography (ERCP) is regarded as the most definitive and reliable diagnostic method of revealing anomalous pancreaticobiliary ductal union [3–6]. However, ERCP is contraindicated in patients with acute pancreatitis and cholangitis and requires the administration of general anesthetic in children [7, 8].

MR cholangiopancreatography is a noninvasive procedure and provides the best available projection image for revealing the extent of choledochal cyst in children and adults [8–15]. Researchers have reported that anomalous pancreaticobiliary ductal union can be depicted in adult patients on MR cholangiopancreatography with a half-Fourier single-shot fast spin-echo or turbo spin-echo sequence [12, 13, 15], but that this finding was not shown in children when a turbo spin-echo or an inversion recovery turbo spin-echo sequence with maximum intensity projection was used [9, 10]. The limited efficacy of the technique in pediatric patients seems to result, in part, from insufficient spatial resolution in terms of depicting the relatively small bile

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ducts and pancreatic ducts [11–14]. Moreover, the size of the choledochal cyst and the stones impacted in the common channel might influence the visualization of anomalous pancreaticobiliary ductal union [8, 13].

The purpose of this study was to evaluate whether MR cholangiopancreatography can accurately depict anomalous pancreaticobiliary ductal union and other abnormalities of the biliary tract in children with choledochal cysts.

Subjects and Methods

Patients

Twenty infants and children with symptoms of abdominal pain, mass, or jaundice who were diagnosed with choledochal cyst by sonography were included in this study consecutively from August 1997 to April 1999. All the patients were girls and ranged in age from 1 month to 13 years (mean age, 4.6 years). Acute pancreatitis was diagnosed on the basis of the clinical findings of acute abdominal pain accompanied by upper abdominal tenderness and increased serum amylase and lipase levels. Ten patients presented with acute pancreatitis, four of whom had a history of recurrent pancreatitis. In four patients, cholangitis was diagnosed on the basis of the clinical findings (fever and jaundice) and laboratory data (elevated serum levels of aspartate aminotransferase, alanine aminotransferase, and γ-glutamyltransferase). All patients underwent MR cholangiopancreatography, intraoperative cholangiography, and cyst resection with choledochojejunostomy.

Imaging Technique

The MR imaging protocols for our study were approved by the institutional review board at our institution. Informed consent was obtained from the parents of all the patients before MR cholangiopancreatography was performed. MR imaging was performed with a 1.5-T magnet (Signa Horizon; General Electric Medical Systems, Milwaukee, WI) and head or torso coils. Patients fasted for 3–5 hr before the MR examination, and infants and young children were sedated with oral chloral hydrate (50 mg/kg of Pocral; Hanlym, Seoul, South Korea). To localize the hepatobiliary system, we acquired an axial T1-weighted fast multiplanar spoiled gradient-recalled MR image (TR range/TE, 220–250/4.2; flip angle, 90°; slice thickness, 7 mm; matrix, 256 × 128; and scanning time, 23–26 sec). MR cholangiopancreatography was mainly performed with a non–breath-hold single-shot fast spin-echo sequence. Images were acquired using the breath-holding technique in the older children if possible. A respiratory trigger was not used because the small bellows for infants and young children was not available in our institution. To maintain parenchymal signals surrounding the ductal structure and detect stones within the bile duct on T2-weighted images, we acquired axial and coronal multislice single-shot fast spin-echo MR images with the following parameters: TR/effective TE range, infinite/80–100; slice thickness, 3–4 mm; slice gap, 0–1 mm; field of view, 16–24 cm; matrix, 256 × 192; and scanning time, 36–53 sec. MR cholangiopancreatography was then performed using single-slab projection imaging using the following parameters: TR/effective TE range, infinite/1000–1400; slab thickness, 20–30 mm; field of view, 16–20 cm; matrix, 256 × 256; and scanning time, 2 sec. For multiple projection images, coronal and oblique coronal (–45°, –30°, 30°, 45° to the axis) images were acquired.

Image Analysis

MR cholangiopancreatographic images were evaluated independently by two pediatric radiologists who were unaware of the clinical and sonographic findings. The type of choledochal cyst and anomalous pancreaticobiliary ductal union, the location of the stone, and the anatomic variations of the biliary tree were evaluated on the basis of thin-slice and thick-slab images. The MR cholangiopancreatographic findings were compared with those of intraoperative cholangiography. The mean interval of time between the two imaging studies was 6.6 days (range, 1–17 days).

The classifications of the type of choledochal cyst and anomalous pancreaticobiliary ductal union (Figs. 1 and 2) devised by Todani et al. [16, 17] were used. Anomalous pancreaticobiliary ductal union was diagnosed when the union between the common bile duct and pancreatic duct was located far from the duodenum and the length of the common channel exceeded 5 mm [18] (Fig. 3A). Because MR imaging cannot delineate the fine, intricate network of ducts in children, type C union was judged to be present when the accessory pancreatic duct could be identified.

Stones were diagnosed when round or irregular-shaped signal voids were found in the choledochal cyst and the common channel (Fig. 4A). Finally, we determined whether anatomic variations of the biliary tree were present. These variations included a low cystic duct insertion, a medial insertion of the cystic duct, a long parallel course of the cystic and common hepatic ducts, and an aberrant right posterior hepatic duct draining to the cystic duct or to the common hepatic duct [19–21].

Statistical Analysis

All statistical analyses were performed using statistical software (MedCalc 6.11; MedCalc Software, Mariakerke, Belgium). The degree of agreement between observers in the interpretation of the type of choledochal cyst and anomalous pancreaticobiliary ductal union, the location of the stone, and the anatomic variation of the biliary tree was determined using the kappa coefficient. The kappa values were interpreted as follows: less than 0.20, poor agreement; 0.21–0.40, fair; 0.41–0.60, moderate; 0.61–0.80, good; and 0.81–1.00, excellent. The mean age of the patients who had an anomalous pancreaticobiliary ductal union that was visible on MR cholangiopancreatography and the mean age of those who did not were tested using the unpaired Student's *t* test. A *p* value of less

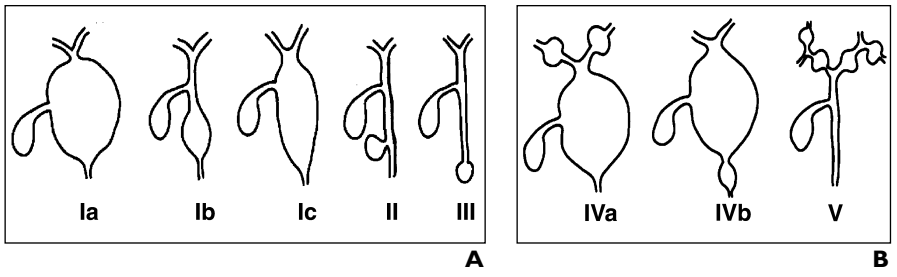

Fig. 1.—Classifications of types of choledochal cysts. (Adapted with permission [16])
A and **B,** Drawings depict types Ia, Ib, Ic, II, and III (**A**) and types IVa, IVb, and V (**B**) choledochal cysts.

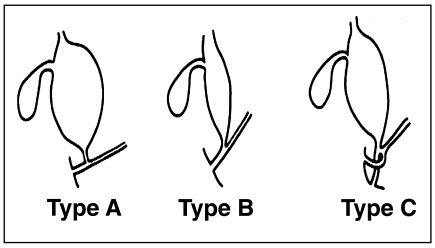

Fig. 2.—Drawings show classifications of types of anomalous pancreaticobiliary ductal union. In type A anomalous pancreaticobiliary ductal union, narrowed common bile duct joins pancreatic duct at right angle. In type B union, pancreatic duct joins common bile duct at acute angle. Type C union is complicated by patent accessory pancreatic duct and fine, intricate network of ducts.

than 0.05 was considered to indicate a statistically significant difference.

Results

On MR cholangiopancreatography, the types of choledochal cysts were determined as follows: four cases of type Ia, seven of type Ic, seven of type IVa, and two of type IVb. Interobserver agreement for the type of choledochal cyst was excellent ($\kappa = 1.0$). The types of choledochal cyst determined on MR cholangiopancreatography correlated with those identified on intraoperative cholangiography in all patients.

MR cholangiopancreatography showed the type of anomalous pancreaticobiliary ductal union in 12 patients (age range, 2.4–13 years; mean age ± SD, 5.9 ± 2.9 years) as follows: two cases of type A (Fig. 3), seven of type B (Fig. 4), and three of type C (Fig. 5). Interobserver agreement for the type of anomalous pancreaticobiliary ductal union was good ($\kappa = 0.704$). In 11 of these 12 patients, the types of anomalous pancreaticobiliary ductal union revealed by MR cholangiopancreatography were concordant with those identified on intraoperative cholangiography. In the remaining patient, MR cholangiopancreatography depicted a type C anomalous pancreaticobiliary ductal union, but intraoperative cholangiography failed to depict the anomalous pancreaticobiliary ductal union.

MR cholangiopancreatography could not show the common channel and anomalous pancreaticobiliary ductal union in eight patients (age range, 0.1–9 years; mean age ± SD, 2.5 ± 3.4 years) who had a markedly dilated choledochal cyst (type Ia, $n = 1$; type IVa, $n = 5$; type IVb, $n = 2$) (Fig. 6). In five of these eight patients, the type of anomalous pancreaticobiliary ductal union was confirmed by intraoperative cholangiography performed after resection of the choledochal cyst as follows: four cases of type A and one case of type B. In the remaining three patients with type Ia and IVa choledochal cysts, intraoperative cholangiography failed to depict the anomalous pancreaticobiliary ductal union.

The mean age of the patients with anomalous pancreaticobiliary ductal union shown on MR cholangiopancreatography was higher than that of the patients in whom anomalous pancreaticobiliary ductal union could not be detected by MR cholangiopancreatography ($p = 0.034$). According to the type of choledochal cyst, anomalous pancreaticobiliary ductal union was revealed in three of the four patients with a type Ia cyst and in all of the patients with a type Ic choledochal cyst, whereas this union was not visible in five of the seven patients with a type IVa cyst and in all of the patients with a type IVb choledochal cyst.

Stones were located in the common channel in three patients and in the choledochal cyst in five patients. In one of the three patients with stones in the common channel (Fig. 5), the stones were recognized only on the thin-slice axial images. In the remaining seven patients, stones were identified on either the thin-slice or the thick-slab images. An aberrant right posterior hepatic duct inserted into the distal common bile duct was shown by MR cholangiopancreatography in each patient with a type Ic choledochal cyst and in each patient with a type IVa choledochal cyst (Fig. 4); low insertion of the cystic duct was revealed in one patient with a type Ia choledochal cyst. A dilated cystic duct draining into an aberrant right posterior hepatic duct was identified in one patient with a type IVa choledochal cyst. Intraoperative cholangiographic findings verified the anomalous insertion of the right posterior hepatic duct or the cystic duct. Interobserver agreement for the location of the stones and for the anatomic variations of the biliary tree was excellent ($\kappa = 1.0$).

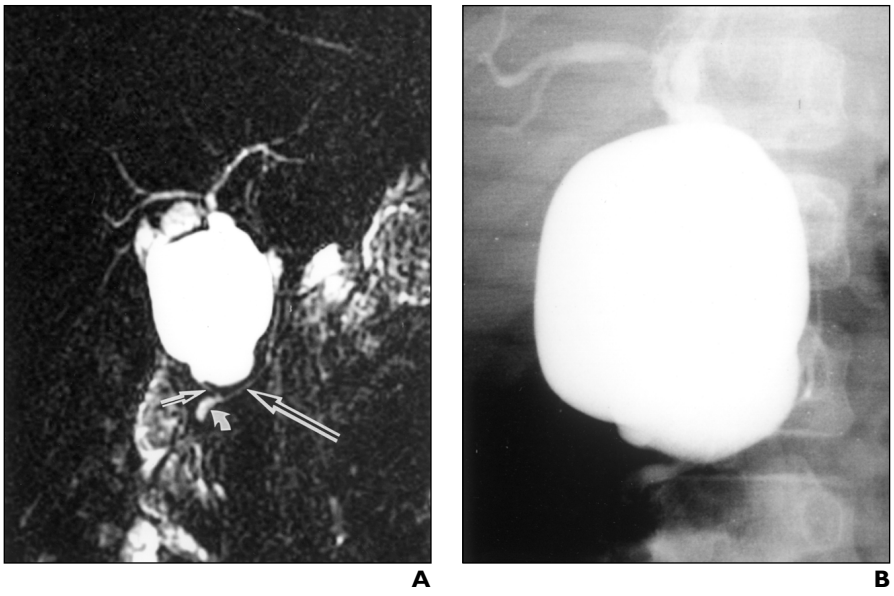

Fig. 3.—9-year-old girl with abdominal pain and recurrent pancreatitis.
A, Coronal MR cholangiopancreatographic image (TR/effective TE, infinite/1055) obtained using single-shot fast spin-echo sequence shows type Ia choledochal cyst with type A anomalous pancreaticobiliary ductal union. Common channel (*curved arrow*) is dilated, and angle between distal common bile duct (*short straight arrow*) and pancreatic duct (*long straight arrow*) is obtuse.
B, Intraoperative cholangiogram shows cystic dilatation of common bile duct and ectatic common channel. Overdistended choledochal cyst obscures union between common bile duct and pancreatic duct.

Discussion

Anomalous pancreaticobiliary ductal union is usually associated with choledochal cyst and is generally accepted as a cause of it. Cheney et al. [22] proposed that choledochal cyst represented a spectrum of embryonic malformations of the pancreatobiliary system, one of which might be an anomalous pancreaticobiliary ductal union. Craig et al. [23] suggested that dysfunction of the sphincter of Oddi might play a role in choledochal cyst formation.

Complete excision of the extrahepatic bile duct has been adopted as the standard treatment for patients with a choledochal cyst. Repeated cholangiography is usually performed to evaluate the distance between the site of dissection and the main pancreatic duct so that iatrogenic injury to the main pancreatic duct can be avoided. If anomalous pancreaticobiliary ductal union is detected at preoperative radiologic evaluation, then it would be helpful for the surgeon to decide the surgical margin and perform cholangiography carefully. Some children who have symptoms similar to those associated with choledochal cyst have anomalous pancreaticobiliary duc-

tal union without dilatation of the common bile duct [24, 25]. For these patients, complete excision of the common bile duct is also recommended to prevent symptoms from recurring and to decrease the risk of carcinoma of the bile duct, which becomes high for these patients as they age [25]. For the diagnosis of anomalous pancreaticobiliary ductal union with or without dilatation of the common bile duct, ERCP has been recommended; however, ERCP is invasive and requires the administration of general anesthetic in children. MR cholangiopancreatography is considered a safe substitute for ERCP. The development of the single-shot fast spin-echo sequence made it possible to evaluate the biliary tree in infants and children unable to breath-hold [8–15]. In our study, 12 patients had symptoms of pancreatitis or cholangitis, which further justified the use of MR cholangiopancreatography instead of ERCP.

The type and extent of choledochal cyst determined on MR cholangiopancreatography correlated well with the findings on intraopera-

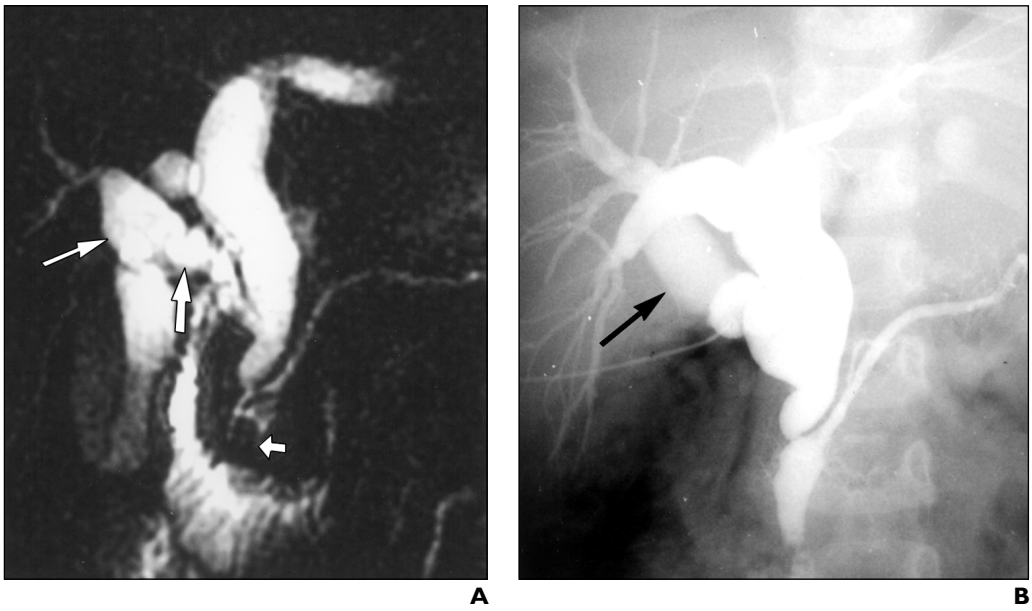

Fig. 4.—3-year-old girl with abdominal pain and pancreatitis.
A, Coronal MR cholangiopancreatographic image (TR/effective TE, infinite/1339) obtained using single-shot fast spin-echo sequence shows type IVa choledochal cyst with type B anomalous pancreaticobiliary ductal union. Dilated pancreatic duct and common channel containing stones (*short arrow*) are also evident. Dilated aberrant right posterior hepatic duct (*long thin arrow*) draining cystic duct (*long thick arrow*) inserts into common bile duct.
B, Intraoperative cholangiogram reveals findings that correlate well with MR cholangiographic findings. Arrow indicates aberrant right posterior hepatic duct.

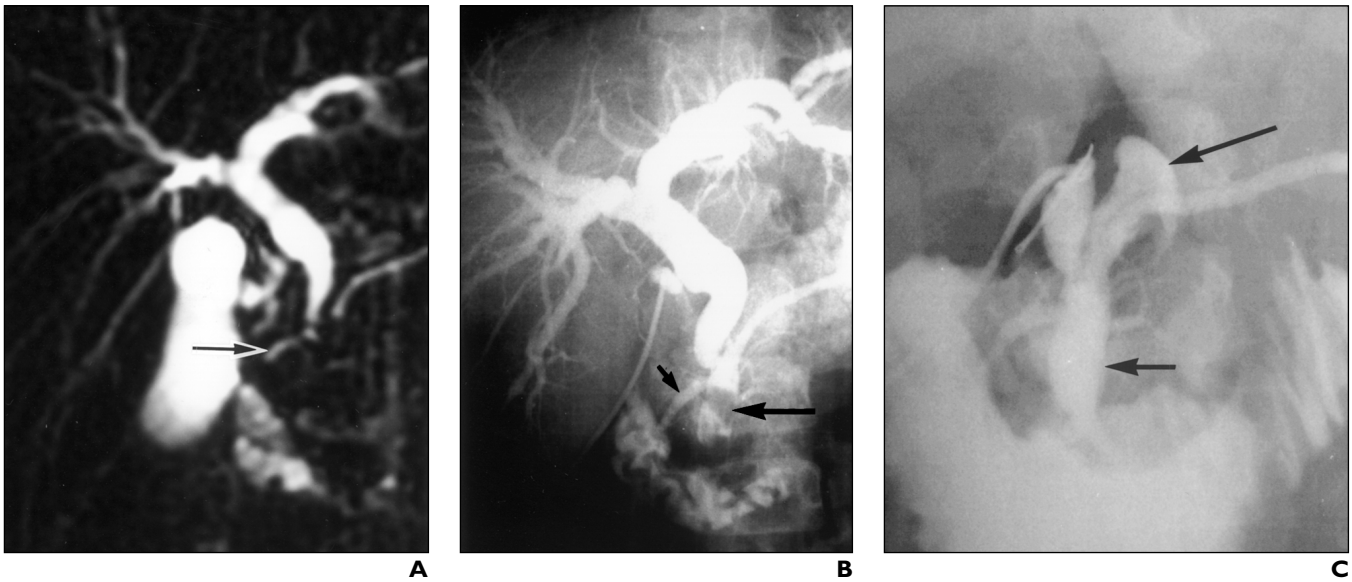

Fig. 5.—5-year-old girl with abdominal pain, jaundice, and fever.
A, Coronal MR cholangiopancreatographic image (TR/effective TE, infinite/1055) obtained using single-shot fast spin-echo sequence shows mild fusiform dilatation of common bile and hepatic ducts and of right and left intrahepatic ducts. Dilated main and accessory (*arrow*) pancreatic ducts are visible, whereas common channel cannot be seen.
B, Intraoperative cholangiogram shows stones (*long arrow*) impacted in common channel as well as dilated main and accessory (*short arrow*) pancreatic ducts.
C, Intraoperative cholangiogram obtained after resection of choledochal cyst shows type C anomalous pancreaticobiliary ductal union. In addition to dilated common channel (*short arrow*), ectatic communicating channel (*long arrow*) is apparent between main and accessory pancreatic ducts.

tive cholangiography in all 20 patients (100%), although types Ib, II, III, and V choledochal cysts were not included in our study. In 12 (60%) of the 20 patients, anomalous pancreaticobiliary ductal union was clearly visible on MR cholangiopancreatography. In nine of 12 patients, the type of anomalous pancreaticobiliary ductal union (A or B) as determined on MR cholangiopancreatography correlated well with the findings on intraoperative cholangiography. Thin-slice and thick-slab MR images enabled visualization of the accessory pancreatic duct and the common channel in the remaining three patients with type C anomalous pancreaticobiliary ductal union. However, the fine, intricate network of type C anomalous pancreaticobiliary ductal union was observed only on intraoperative cholangiography.

The main drawback of using MR cholangiopancreatography for the determination of anomalous pancreaticobiliary ductal union seems to be that large choledochal cysts obscure the common channel, which occurred in eight of our patients. Yamataka et al. [8] also reported that MR cholangiopancreatography and ERCP failed to detect anomalous pancreaticobiliary ductal union in one patient with a large choledochal cyst and that the size and location of the large choledochal cyst prevented the visualization of the anomalous pancreaticobiliary ductal union. In five patients, the type of anomalous pancreaticobiliary ductal union was not revealed by initial intraoperative cholangiography performed by cystic duct cannulation, but it was depicted by intraoperative cholangiography repeatedly performed after resection of the choledochal cyst. In the remaining three infants, intraoperative cholangiography failed to show anomalous pancreaticobiliary ductal union because of poor image quality or nonopacification of the pancreatic duct.

Results of previous studies undertaken to determine whether MR cholangiopancreatography can depict anomalous pancreaticobiliary ductal union associated with choledochal cyst have not been consistent [9–15]; these differences might be attributed to the different ages of the patients involved in the studies or to the different imaging techniques used. Even in pediatric patients, the mean age of the patients with anomalous pancreaticobiliary ductal union shown on MR cholangiopancreatography was higher than that of the patients in whom anomalous pancreaticobiliary ductal union could not be detected by MR cholangiopancreatography; this finding proved to be statistically significant in our study.

The global quality of images obtained with single-slab images is better than that of maximum-intensity-projection images in adults with suspected biliary or pancreatic abnormalities [26]. In a study by Chan et al. [10], anomalous pancreaticobiliary ductal union was not visualized on MR cholangiopancreatography using a two-dimensional turbo spin-echo sequence with the maximum-intensity-projection technique. However, in pediatric patients, MR cholangiopancreatography with a half-Fourier acquisition of a single-shot fast spin-echo or a turbo spin-echo sequence might increase the sensitivity of anomalous pancreaticobiliary ductal union detection [11–14]. A comparison of single-slab imaging and imaging with the maximum-intensity-projection technique was not possible in our study because we did not use the maximum-intensity-projection technique to show anomalous pancreaticobiliary ductal union.

Stones in the common channel were seen in two patients with a type Ic choledochal cyst and in one with a type IVa choledochal cyst. In these patients, the body and tail portions of the pancreatic ducts were more clearly visualized than those in patients without stones in the common channel. In one patient with a type Ic choledochal cyst, multiple stones occupied al-

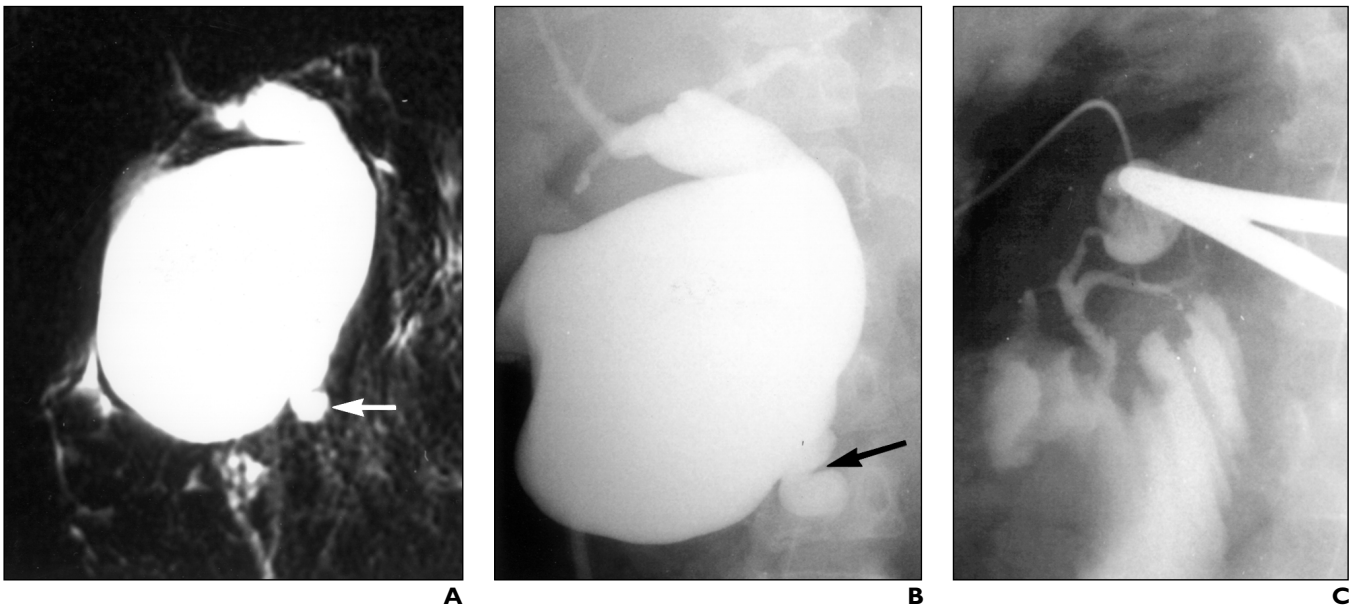

Fig. 6.—6-year-old girl with jaundice.
A, Coronal MR cholangiopancreatographic image (TR/effective TE, infinite/1004) obtained using single-shot fast spin-echo sequence shows type IVb choledochal cyst that is causing dilatation of proximal common bile duct and short segmental dilatation of distal common bile duct (*arrow*). Type of anomalous pancreaticobiliary ductal union cannot be determined.
B, Intraoperative cholangiogram shows type of choledochal cyst that is concordant with that shown on MR cholangiopancreatography. Arrow indicates stenotic segment between cysts. Type of anomalous pancreaticobiliary ductal union cannot be determined in this image.
C, Intraoperative cholangiogram obtained after resection of proximal part of choledochal cyst reveals that type of anomalous pancreaticobiliary ductal union is type B.

most the entire portion of the common channel and were not outlined by bile; thus, the common channel was hard to detect on MR cholangiopancreatography. The mechanism of stone formation in the common channel is uncertain; however, Kaneko et al. [27] proposed that increased mucin production from the bile duct in patients with a pancreaticobiliary maljunction might play an important role.

An aberrant right posterior hepatic duct inserting into the distal common bile duct can be observed on MR cholangiopancreatography. Normal anatomic variations of the biliary tree in patients with a choledochal cyst can occasionally be detected using intraoperative cholangiography or are visible at the time of surgery. Preoperative detection of this anomaly, which is difficult to recognize on sonography or CT, reduces the risks associated with the ligation of the aberrant duct, which may result in biliary infection and subsequent atrophy of the posterior segment of the right lobe [19–21].

Our study has a number of limitations. Although anomalous pancreaticobiliary ductal unions were shown mainly in patients with a type I choledochal cyst and not in those with a type IV choledochal cyst, there were too few patients in our study to evaluate the association between the visibility of anomalous pancreaticobiliary ductal union on MR cholangiopancreatography and the type of choledochal cyst. Moreover, types Ib, II, III, and V choledochal cysts were not included in our study. Another limitation is that anomalous pancreaticobiliary ductal union seen on MR cholangiopancreatography could not be confirmed by intraoperative cholangiography in four patients because of poor image quality or nonopacification of the pancreatic duct.

In conclusion, MR cholangiopancreatography with the non–breath-hold technique is an accurate, noninvasive method of evaluating anomalous pancreaticobiliary ductal union in children with choledochal cysts.

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