A Large Common Bile Duct Stone Migrated from the Gallbladder through a Cholecystohepaticodochal Fistula: an Unusual Complication of Mirizzi Syndrome Type II

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Mirizzi syndrome with a biliobiliary fistula (Mirizzi syndrome type II) is a rare complication of a long-standing gallbladder stone disease. It is even rarer for a gallbladder stone to migrate through a biliobiliary fistula into the common duct. We encountered this interesting complication of Mirizzi syndrome type II in an 86 year-old female patient. A large gallbladder stone migrated into, and impacted into the distal common bile duct through a cholecystohepaticodochal fistula. The stone was resistant to mechanical lithotripsy and was treated with biliary endoprosthesis and oral bile acids.

Key Words: Mirizzi syndrome type II, biliobiliary fistula, bile duct stone

Gallbladder (GB) stones can produce complications by erosion into the adjacent structures by the formation of fistulas. The common sites for fistulization are the duodenum, colon, and stomach (Glenn et al. 1981). Also in a small number of patients, the stones in the GB can erode into the common duct and result in the formation of cholecystocholedochal or cholecystohepaticodochal fistulas (Mirizzi syndrome type II) (McSherry et al. 1982). This type of biliobiliary fistulas has been collecting interest because of its diagnostic and therapeutic difficulties (Baer et al. 1990; Mishra et al. 1990; Yip et al. 1992). Reported incidences of biliobiliary fistulas range from 0.7% to 1.43% of the patients undergoing cholecystectomy (Coffeltte and Bismuth 1975; Mishra et al. 1990; Yip et al. 1992). Almost all stones in the biliobiliary fistulas have been reported to sit astride the fistula and obstruct the common hepatic duct. (Venkatesh Rao et al. 1988; Yip et al. 1992).

We experienced an interesting complication of Mirizzi syndrome type II: a large GB stone migrated into the common duct through a cholecystohepaticodochal fistula and impacted in the distal common bile duct. The stone in the common duct was resistant to mechanical lithotripsy, and was subsequently treated with biliary endoprosthesis and oral bile acids considering the patient’s old age and fragility. We herein report this rare complication of Mirizzi syndrome type II, and discuss the clinical significance of a combined treatment of biliary endoprosthesis and oral bile acids in patients with non-extractable bile duct stones.
CASE REPORT

An 86-year-old female patient was admitted to Yongdong Severance Hospital on December 23rd, 1993 with a 7-day history of postprandial epigastric pain, and a 1-day history of jaundice and fever. About 20 years prior to admission, the patient was diagnosed as having a symptomatic GB stone, but refused to take her GB out. From then on, she had been asymptomatic until this admission. Upon examination, the patient was thin (height 150 cm, weight 39 kg) and appeared acutely ill-looking. She was afebrile although she had felt fever and chilling before admission. The skin and the sclerae were icteric. The examination of the abdomen revealed only mild right upper quadrant tenderness. Upon laboratory examinations, leukocytosis (24,400/mm$^3$) and mild anemia (hematocrit 32.3%) were found. Total protein was 5.6 g/dl, albumin 2.9 g/dl, total bilirubin 11.4 mg/dl, alkaline phosphatase 229 IU/L, aspartate transaminase 40 IU/L, alanine transaminase 43 IU/L, and gammaglutamyltransferase 259 IU/L. Viral markers for hepatitis B and C were negative. The serum and bile CEA values were 0.8 and 6.9 ng/ml, respectively.

An abdominal computed tomography (CT) showed marked dilatation of both intrahepatic bile ducts and minimal dilatation of the common duct. The GB wall was markedly thickened and a non-calcified large stone filled almost the entire GB lumen (Fig. 1A). This disproportional bile duct dilatation made us suspect a pathology at the level of the common hepatic duct (CHD). An abdominal sonography also displayed a large stone in a contracted GB with a thickened wall, but there was neither stenosis nor a mass lesion at the CHD level. A duodenoscopy for an endoscopic retrograde cholangiopancreatography (ERCP) displayed a slightly bulged, and pus discharging papilla. A pancreatography revealed that the main pancreatic duct was dilated, but the tail portion was stenotic. Because of the difficulty in selectively cannulating the bile duct, an infundibulotomy with a needle-knife sphincterotome was performed. A cholangiography revealed that the intrahepatic bile ducts were irregularly dilated and stiff. The common duct was also dilated (13 mm in diameter). A couple of small floating stones

![Fig. 1. Abdominal CT scan taken during the first admission (A) and the second admission (B)]. Initially (A), the gallbladder wall was thickened and the lumen of the gallbladder was filled with heterogeneously increased density suggesting a large stone. The extrahepatic bile duct was minimally dilated. About 5 months later (B), the gallbladder markedly shrunk, whereas the extrahepatic bile duct was markedly dilated with heterogeneously increased density in it (closed arrow: gallbladder, open arrow: extrahepatic bile duct).
Fig. 2. A cholangiogram through the nasobiliary drainage tube shows dilated intra- and extrahepatic bile ducts. The gallbladder with a large filling defect (closed arrow) was visualized in contact with the right hepatic duct near the porta hepatis. At this time, since the cystic duct (open arrow) was also well visualized, the presence of a cholecystohepatic ductal fistula was not noticed.

Fig. 3. Endoscopic retrograde cholangiography performed during the second admission showed a large oval-shaped stone impacted at the distal common bile duct.

were seen in the common bile duct. The GB was not visualized at this time. An endoscopic nasobiliary drainage (ENBD) tube (7 French) was placed, and an ENBD tube cholangiogram depicted a cystic duct and a small GB lumen filled with a filling defect (Fig. 2). At this time, although the GB lumen was in contact with the main right hepatic duct, we did not suspect a cholecystohepatic ductal fistula. The bile fluid culture revealed the growth of Acinetobacter Iwoffi. While waiting for symptoms and signs of acute cholangitis to subside, we performed stone removal through the previous infundibulotomy opening from where three small pigment stones were extracted. Uneventfully, the patient was discharged on the 17th hospital day.

About 5 months later (May 26th, 1994), the patient visited our emergency department because of an abruptly developed, severe 7-day postprandial epigastric pain. Upon examination, the patient appeared acutely ill-looking, yet the vital signs remained stable.

The sclerae were anicteric and the abdominal examination revealed only mild right upper quadrant abdominal tenderness. Upon laboratory examinations, the white blood cell count was 12,600/mm³, total protein 7.0 g/dl, albumin 3.6 g/dl, total bilirubin 1.5 mg/dl, alkaline phosphatase 292 IU/L, aspartate transaminase 94 IU/L, alanine transaminase 72 IU/L.

In consideration of the previously acknowledged ductal stenosis of the pancreas tail, an abdominal CT scan was performed to eliminate the possibility of a pancreatic cancer. Compared with the results in previous abdominal CT films, both intrahepatic ducts and the common duct were more markedly dilated. However, the GB shrunk. The CT density of intraluminal content in the distal CBD was slightly high, which suggested a large stone (Fig. 1B). On the 5th hospital day, an ERCP was performed. The ampulla of Vater was markedly bulged and the previous infundibulotomy fistula shrunk and discharged a whitish pus. A cholangiography showed a large (3.0 cm in the longest diameter) oval-
shaped stone impacted in the distal CBD (Fig. 3). A complete sphincterotomy was performed through the infundubulotomy stoma. Since it was impossible to retrieve this impacted stone, an ENBD was performed. The bile fluid culture revealed the growth of multiple organisms. An ENBD tube cholangiogram revealed a small GB lumen without filling defect, which resembled a diverticulum. The GB lumen was filled through a small fistula tract from the right main intrahepatic duct and the part of the cystic duct was retrogradely (from the GB toward the common duct) visualized (Fig. 4). These findings were conclusive of a cholecystohepaticochoanal fistula, and the migration of a large GB stone into the common bile duct through this fistula. Since an intraoperative cholangioscope with electrohydraulic lithotripsy was only available in our hospital, we were considering a cholangioscopic electrohydraulic lithotripsy through a percutaneous route. However, access was difficult to obtain because of the contracted intrahepatic ducts. A follow-up cholangiogram through the ENBD tube revealed that the stone was floating in the common duct (Fig. 5A). An endoscopic stone removal was repeated using a mechanical lithotriptor. The stone

![Fig. 4. Endoscopic retrograde cholangiography through a balloon catheter showed markedly dilated intra, and extrahepatic bile ducts and a small diverticulum-like gallbladder filled through a cholecystohepaticochoanal fistula (open arrow). The cystic duct (closed arrow) was partially visualized through retrograde filling (from the aspect of the gallbladder toward the extrahepatic bile duct).](image)

![Fig. 5. A cholangiography through the nasobiliary drainage tube during the second admission showed that the stone floated in the common bile duct (A). Endoscopic stone removal with a mechanical lithotriptor was tried and the stone was successfully captured (B).](image)
was successfully captured (Fig. 5B), but the steel wire broke at the handle portion. Subsequently, emergency lithotripsy was attempted, but resulted in a ragged fracture of the basket (Fig. 6). At this time, we decided to perform endoscopic retrograde biliary drainage (ERBD) for the purpose of palliative treatment of this non-extractable stone. At first, a 10 French 7cm plastic tube was inserted, but it appeared quite unstable. For the tight anchoring of the endoprosthesys, another 10 French 10cm plastic tube was placed (Fig. 7). On the 17th hospital day, the patient was discharged uneventfully with oral bile acids (combined ursodeoxycholic acid and chenodeoxycholic acid). The patient is now in good health (8 months later after discharge).

**DISCUSSION**

Based on ERCP findings, McSherry et al. (1982) suggested a subclassification of the Mirizzi syndrome into two types. Type I is the classical Mirizzi syndrome which designates external compression of the common hepatic duct by a large stone impacted in the cystic duct or Hartmann’s pouch (Mirizzi 1948). In the Mirizzi syndrome type II, a calculus in the GB erodes partly or completely into the bile duct, resulting in the formation of a cholecystocholedochal or of a choledochocystohepaticodochal fistula, depending on the site of the fistula formation.

Although internal biliary fistula is a common complication of gallstone disease, the Mirizzi syndrome type II (biliobiliary fistula) is a relatively rare complication, constituting 2.7% to 16% of all internal biliary fistulas (Corlette and Bismuth 1975; Morrissey and McSherry 1988), and 0.7% to 1.43% of the patients undergoing cholecystectomy (Corlette and Bismuth 1975; Mishra et al. 1990; Yip et al. 1992).

Biliobiliary fistulas are believed to form as a result of chronic inflammation with the presence of long-standing gallstone diseases (Corlette and Bismuth 1975). Venkatesh Rao et al. (1988) suggested that gallstone impaction in
the ampulla of the GB with compression of the common duct was the initiating event in the formation of a biliobiliary fistula. With repeated bouts of inflammation, the GB adhered to, and fused with the common duct. As the process continued, the GB contracted around the stone. Finally, the gallstone produced pressure necrosis of the intervening walls, and ulcerated into the common duct resulting in a fistula formation. This suggested pathogenetic mechanism of fistula formation is supported by the findings that in most cases of biliobiliary fistula, the site of the fistula is occupied by a large gallstone which is impacted at the fistula opening, and that the dense fibrous adhesions are found between the GB and the common duct (Yip et al. 1992).

Patients with biliobiliary fistulas usually have a history of long-standing biliary tract disease like other cases of internal fistulas. Because gallstone disease is more common among women, these fistulas also occur more often among women (Yip et al. 1992).

Mishra et al. (1990) suggested that the history of a clinical triad of jaundice, fever, and pain in the presence of GB stones indicate the possibility of the Mirizzi syndrome or biliobiliary fistula. However, these clinical features are not diagnostic of biliobiliary fistulas especially in cases where GB stones combined with common duct stones. Noninvasive imaging methods such as ultrasonography and computed tomography are helpful, but not always diagnostic, because they cannot depict the exact biliary anatomy. Upon ultrasound examination, the combination of cholelithiasis in a small contracted GB having an echogenic nodule or calculus in the common duct and with a mild to moderate dilation of proximal biliary radicals, and with a normal-sized or untraced common bile duct distal to the calculus or nodule is an indication of biliobiliary fistula (Mishra et al. 1990).

The biliary abnormality can be demonstrated only by the use of direct cholangiography such as ERCP and percutaneous transhepatic cholangiography (Cornud et al. 1981; Fan et al. 1985; Mishra et al. 1990; Yip et al. 1992). A unique observation found in the cholangiogram is that the common duct below the fistula remains small and non-dilated, whereas the portion of the common duct above the site of obstruction is dilated with an abrupt change in size at the fistula level (Yip et al. 1992). However, despite all of these diagnostic efforts, biliobiliary fistulas can only be found at the time of operation (Kelly and Benson 1980).

Although our case was presented with typical clinical features, the findings from the imaging studies were somewhat confusing. The reasons of failure to diagnose the biliobiliary fistula during the initial presentation of our patient are as follows: First, although the initial CT scans revealed disproportionate dilations in the intrahepatic and extrahepatic bile ducts, the abdominal sonography and the ERC displayed both intra, and extrahepatic bile ducts dilated without a significant narrowing at the common hepatic duct. Second, the cystic duct seemed to be patent on the cholangiogram. Finally, the presence of small floating stones in the common duct was thought to be the sole cause of acute supplicative cholangitis in this patient. We assumed that multiple bile duct stones might have been entrapped in the upper bile duct when the initial abdominal CT scan was performed, and may have passed downward thereafter when abdominal ultrasonography and ERCP were performed.

Biliobiliary fistulas pose great technical difficulties during surgery. The GB is small and contracted with obliterated Calot's triangle and with intense inflammatory adhesions of the surroundings. The junction between the cystic duct and the common duct is not clearly demarcated. These findings may mimic the condition of biliary tract malignancy (Baer et al. 1990; Mishra et al. 1990). The common duct is prone to operative injury if an usual retrograde cholecystectomy is to be performed (Baer et al. 1990; Mishra et al. 1990). In order to clearly define the distorted anatomy, it is recommended that the GB be opened at its fundus and the stones evacuated (Yip et al. 1992). Exploration of the common duct can then be performed through the fistula. Intraoperative cholangiogram may be performed to confirm the presence of a fistula.
(Sutton and Sachatello 1967; Mishra et al. 1990). A partial cholecystectomy is then performed and the remaining flaps of the GB are used to cover the fistula over a T-tube (Corlette and Bismuth 1975; Fan et al. 1985; Mishra et al. 1990; Yip et al. 1992). However, Baer et al. (1990) advocated performing a biliary-enteric bypass to the wall of the fistulous defect by either duodenostomy or Roux-en-Y jejunostomy since flap techniques using thickened tissue, already damaged by the inflammatory process would be prone to failure.

Because most patients with Mirizzi syndrome type II are elderly and frail, minimally invasive treatment may be better in managing these patients. Binnie et al. (1992) reported a case of Mirizzi syndrome type II treated with endoscopic stenting and subsequent laparoscopic subtotal cholecystectomy. With the advent of laparoscopic surgery, this might be one of the alternative treatment modalities only if the diagnosis is precisely made before operation. Also some authors (Cairns et al. 1987; Binmoeller et al. 1993) described successful treatment of Mirizzi syndrome by electrohydraulic lithotripsy under percutaneous or retrograde cholangioscopic guidance.

The present case was interesting in that the large GB stone spontaneously migrated into and impacted in the common bile duct. To our knowledge, only one of the cases reported by Yip et al. (1992) depicted an impacted stone in the common duct at the level of the cystic duct, which was confirmed and treated by operation. The stone in the present case floated later and we attempted endoscopic stone removal. However, the stone was resistant to mechanical lithotripsy. We performed ERBD as a palliative treatment of this non-extractable stone.

Despite widely available technology for the removal of bile duct stones, endoscopists currently encounter approximately 3% of patients with stones that resist extraction (Johnson et al. 1993). Because of limited availability, and high cost of equipment used to destroy non-extractable bile duct stones, such as extracorporeal shock-wave lithotriptors, tunable dye lasers, Nd:YAG laser, and mother-baby endoscopes, most gastrointestinal endoscopy units cannot afford their acquisition. However, treatment of non-extractable CBD stones with biliary stent placement can be performed without excessively expensive or complex equipment (Johnson et al. 1993). Many authors have reported the relative efficacy and safety of this method (Siegel and Yatto 1984; Peters et al. 1992; Van Steenbergen et al. 1992; Bergman et al. 1994; Navicahrren et al. 1994; Raijman et al. 1994). In the meantime, the efficacy of oral dissolution therapy for bile duct stones has been investigated, and several previous reports suggest that chenodeoxycholic acid (CDCA) (Sue et al. 1981), a combination of CDCA or ursodeoxycholic acid (UDCA) with proprietary terpene, or UDCA alone (Somerville et al. 1985) may be effective in treatment. A controlled study conducted by Salvioli et al. (1983) showed statistically significant improvement in a group of patients with retained stones after they received UDCA for 3 to 24 months when compared to results in a placebo group. These observations have facilitated combined use of endoprosthesis and oral bile acids treatment. Johnson et al. (1993) reported the results of combined treatment of non-extractable CBD stones with endoprostheses plus UDCA. They found that the UDCA group showed a higher rate of bile duct stones clearance than in the control group. Moreover this group concluded that oral UDCA facilitated extraction of non-extractable bile duct stones. Furthermore, high dose (15–20 mg/kg) UDCA treatment plus biliary stenting was reported to be effective in reducing the size and the number of the retained CBD stones (Gallo et al. 1994). Thus, we are administering a combined product of UDCA and CDCA in the hope of extraction or spontaneous passage of the stone in future.

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