

[CASE REPORT]

Large Gallstones Stacked Together in the Common Bile Duct after Choledochojejunostomy

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Abstract:

A 65-year-old man had a history of cholecystectomy and treatment for cholelithiasis with a common bile duct incision. Owing to frequent cholangitis, he underwent choledochojejunostomy. Twenty years after the surgery, he was hospitalized for cholangitis and was suspected of having hilar cholangiocarcinoma based on imaging findings. Percutaneous transhepatic cholangioscopy using a SpyGlass™ DS (Boston Scientific, Marlborough, USA) showed gallstones and bile sludge in the bile ducts, but no tumors were noted. Electrohydraulic shockwave lithotripsy with double-balloon enteroscopy enabled complete stone removal; a direct visual biopsy with peroral cholangioscopy showed no malignancy in the bile duct.

Key words: choledochojejunostomy, choledocholithiasis, endoscopic retrograde cholangiopancreatography, double-balloon enteroscopy, electrohydraulic shockwave lithotripsy, gallstones

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Introduction

Cases of common bile duct gallstones are often diagnosed by imaging before treatment; however, some cases of stacked gallstones that fill the bile duct are difficult to distinguish from neoplastic lesions. Endoscopic retrograde cholangiopancreatography does not lead to a diagnosis if sufficient imaging is not possible. In the present case, direct observation with cholangioscopy was considered, but until recently, cholangioscopy had not been widely used due to shortcomings, such as issues with operability. Disposable digital cholangioscopy has been performed since 2016, and its usefulness is being recognized again. In addition, it is difficult to perform endoscopic mechanical lithotripsy for the removal of large or stacked gallstones, since the basket catheter cannot be expanded. In such cases, electrohydraulic shockwave lithotripsy may be more effective.

We herein report a patient with a history of choledochojejunostomy who was diagnosed with multiple, large gallstones in the common bile duct using percutaneous transhepatic cholangioscopy. All stones were removed by electrohydraulic shockwave lithotripsy using double-balloon en-

teroscopy.

Case Report

A 65-year-old man underwent cholecystectomy for cholelithiasis in his 20s, a common bile incision for cholelithiasis in his early 40s, and choledochojejunostomy at 45 years old. Twenty years after the surgery, he was admitted to the hospital with symptoms of obstructive cholangitis. Abdominal ultrasonography (Fig. 1) showed significantly dilated intrahepatic bile ducts and a slightly hyperechoic 3×2-cm mass. Dynamic computed tomography (CT) of the abdomen (Fig. 2) showed a coarse, substantial mass with almost no contrast effect filling the hilar region of the bile duct from the hepaticojejunostomy site to the right and left hepatic ducts. Abdominal magnetic resonance imaging (MRI) (Fig. 3) showed a mass with a high signal intensity on T1-weighted imaging, a mildly high signal intensity on T2-weighted imaging, a mildly high signal intensity on diffusion-weighted imaging, and a heterogeneous low signal intensity on apparent diffusion coefficient maps. Magnetic resonance cholangiopancreatography showed a defect in the hilar bile duct. Based on the above imaging findings, cho-

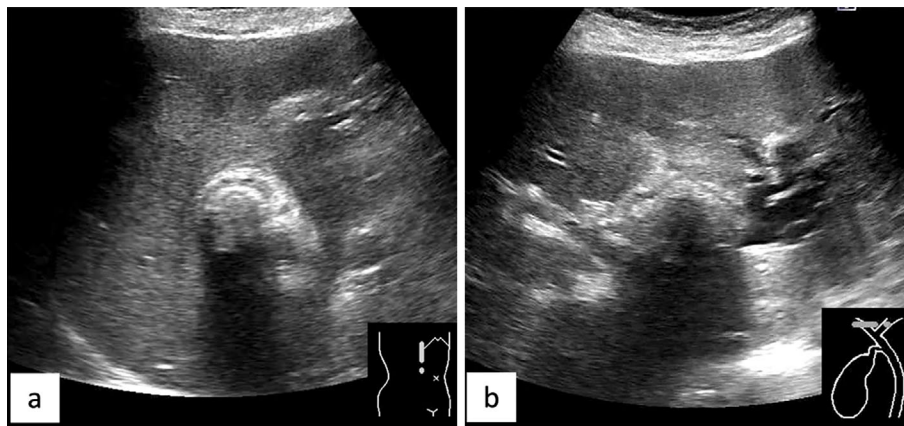


Figure 1. Abdominal ultrasonography. a: A slightly hyperechoic 3×2-cm mass. b: The significantly dilated intrahepatic bile duct.

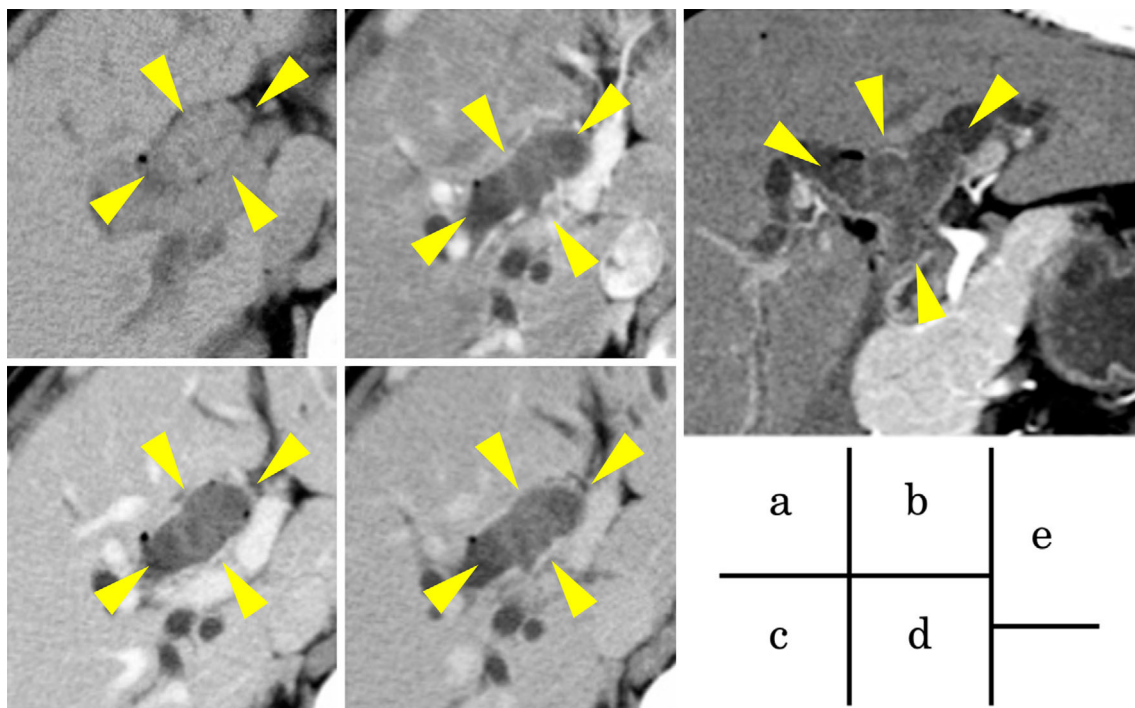


Figure 2. Dynamic enhanced CT showing a coarse, substantial mass with almost no contrast effect, filling the hilar region of the bile duct from the hepaticojejunostomy site to the right and left hepatic ducts. CT: computed tomography. a: Horizontal section, plain. b: Horizontal section, arterial phase. c: Horizontal section, portal phase. d: Horizontal section, equilibrium phase. e: Coronal section, equilibrium phase.

langiocarcinoma with hemorrhaging was first suspected. Percutaneous transhepatic biliary drainage was performed for the treatment of cholangitis because the previous physician did not use a double-balloon endoscope. Bile cytology was performed several times, but a diagnosis could not be made. An 8-Fr drainage tube was initially used, but the patient was transferred to our hospital after being frenched up to 10-Fr and 12-Fr every week for percutaneous transhepatic cholangioscopy. At the time of admission to our hospital, the cholangitis had improved, and the carcinoembryonic antigen and cancer antigen 19-9 levels were within the reference

ranges (Table).

When percutaneous transhepatic cholangioscopy was performed using the percutaneous transhepatic biliary drainage route with a cholangioscope (SpyGlass™ DS; Boston Scientific, Marlborough, USA), the bile duct was found to be filled with gallstones of different sizes, and bile sludge (Fig. 4). No tumor was seen up to the hepaticojejunostomy site, and the shadows that were suspected to be a tumor on the images were determined to be gallstones and bile sludge. There was no stenosis at the hepaticojejunostomy site. At a later date, the stones were removed in the antegrade direc-

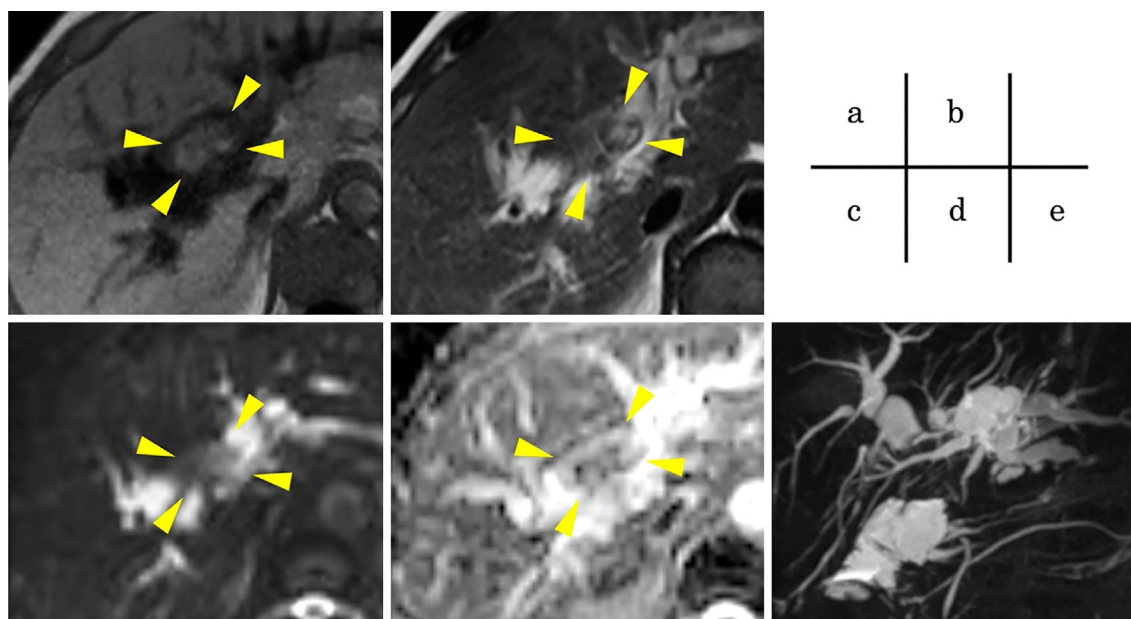


Figure 3. Magnetic resonance imaging showing a mass with a high signal intensity on T1-weighted imaging, a mildly high signal intensity on T2-weighted imaging, a mildly high signal intensity on diffusion-weighted imaging, and a heterogeneous low signal intensity on apparent diffusion coefficient maps. Magnetic resonance cholangiopancreatography showed a defect in the hilar bile duct. a: T1-emphasized image. b: T2-emphasized image. c: Diffusion-weighted image. d: ADC mapping. e: Magnetic resonance cholangiopancreatography. ADC: apparent diffusion coefficient

Table. Blood Test.

WBC	5,400 / μ L
RBC	4.07×10^6 / μ L
Hb	12.9 g/dL
Plt	20.4×10^4 / μ L
TP	6.3 g/dL
T-Bil	1.4 mg/dL
AST	53 U/L
ALT	80 U/L
ALP	666 U/L
γ -GTP	229 U/L
LDH	105 U/L
AMY	99 U/L
BUN	7.1 mg/dL
Cr	0.69 mg/dL
Na	140 mEq/L
K	4.2 mEq/L
Cl	106 mEq/L
CRP	0.44 mg/dL
CEA	2 ng/mL
CA19-9	19.4 U/mL

WBC: white blood cell, RBC: red blood cell, Plt: platelets, TP: total protein, T-Bil: total bilirubin, AST: aspartate transaminase, ALT: alanine transaminase, ALP: alkaline phosphatase, γ -GTP: γ -glutamyl transpeptidase, LDH: lactate dehydrogenase, Amy: amylase, BUN: blood urine nitrogen, Cr: creatinine, CRP: C-reactive protein, CEA: carcinoembryonic antigen, CA19-9: carbohydrate antigen 19-9

tion using a balloon catheter (Selecon MP Catheter II; Terumo, Tokyo, Japan) through the percutaneous transhepatic biliary drainage route. Some stones were extruded to the jejunum, but most stones were difficult to push out.

The next day, increased percutaneous transhepatic biliary drainage, a fever, and mild elevation of bilirubin and biliary system enzymes were observed, and cholangitis due to bile duct obstruction was suspected. Since gallstones were expected to be inlaid at the hepaticojejunostomy site, and a large number of stacked stones remained in the bile duct, we considered stone removal by transgastrointestinal treatment to be preferable to percutaneous treatment.

We performed the gallstone removal surgery using a short-type double-balloon enteroscope (Fig. 5). An impacted gallstone was found when the hepaticojejunostomy site was reached. Since the guidewire could not pass through the stone and endoscopic mechanical lithotripsy was difficult, electrohydraulic lithotripsy was performed with water stored in the afferent loop to disintegrate the stones. Almost all of the stones were removed by repeated electrohydraulic shock-wave lithotripsy using the basket catheter. After stone removal, the bile duct was observed by peroral cholangioscopy, but no obvious tumor lesions were found. One week later, percutaneous transhepatic cholangioscopy was performed using a SpyGlass™ DS, which revealed edematous mucosa at the confluence of the hepatic ducts and the common bile duct. A biopsy was performed, and no malignancy was found.

Ursodeoxycholic acid was prescribed to prevent recurrence, and the patient has been followed up on an outpatient

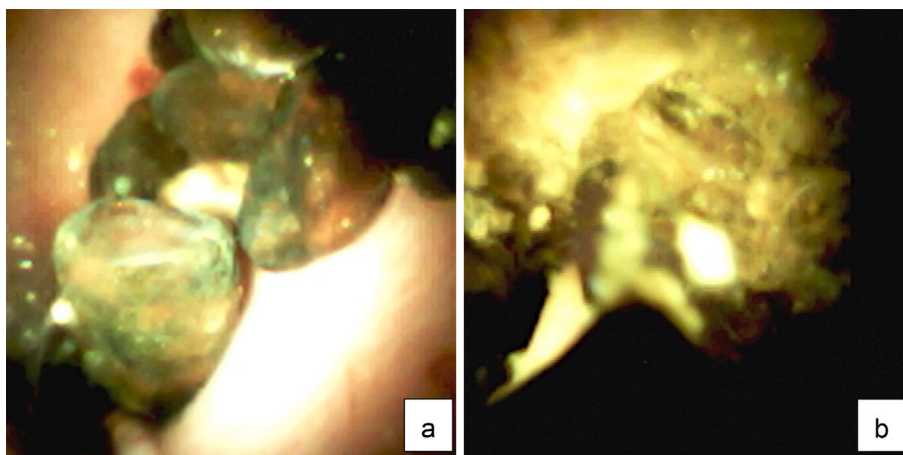


Figure 4. Percutaneous transhepatic cholangioscopy. Bile duct filled with gallstones of different sizes and bile sludge. a: left intrahepatic bile duct. b: common bile duct.

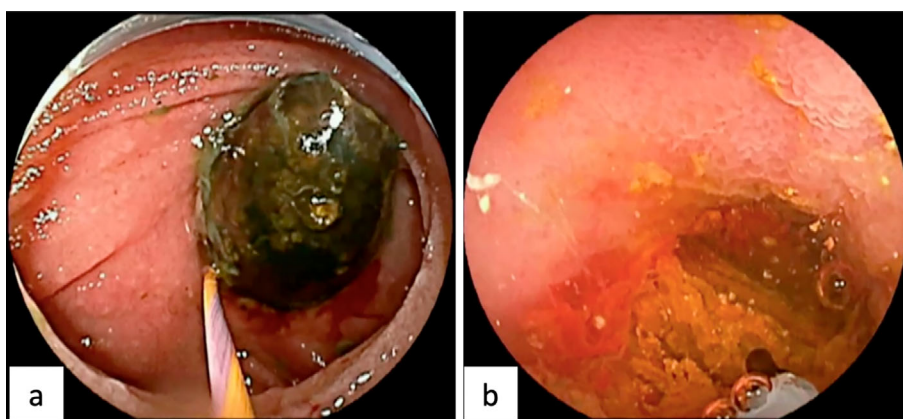


Figure 5. Double-balloon enteroscopy. a: An impacted gallstone at the hepaticojejunostomy site. b: Electrohydraulic lithotripsy, with water stored in the afferent loop to break the stones.

basis since then; no recurrence has been observed in the past four years.

Discussion

The sensitivity of CT for the diagnosis of common bile duct gallstones is 65%, while that of MRI is 96% (1). The diagnosis of bile duct gallstones by CT is affected by the calcium content in the stones, with stones less likely to be detected as the cholesterol content increases (2). In the present case, CT showed a mass in the bile duct. It was not possible to determine that the mass was a large number of gallstones of unequal sizes because the gaps between the stones were filled with bile sludge, and the stones appeared as a single mass. However, dynamic CT showed no contrast effect on the mass, and the findings were not clearly definable as a tumor. Although MRI is useful for differentiating bile duct gallstones from cholangiocarcinoma, it may show atypical signals depending on the composition, size, and density of the stones (3). In the present case, T2- and diffusion-weighted MRI showed a high signal intensity in the mass area, suggesting malignancy. Since there was a

mismatch between the CT and MRI findings and malignancy could not be ruled out, we planned to examine the patient following the protocol for hilar cholangiocarcinoma management.

Although a short-type double-balloon endoscope has been introduced in recent years, endoscopic retrograde cholangiopancreatography has not been widely used for surgically altered anatomies due to procedural difficulties (4). In the present case, the patient had undergone choledochojejunostomy, and although cholangiography and bile cytology had been performed by the percutaneous transhepatic cholangioscopy route by the previous physician, a definitive diagnosis could not be made.

In patients with a surgically altered anatomy, cholangioscopy has been reported to be useful for investigating recurrent cholangitis and treating bile duct gallstones unable to be detected by other imaging modalities (5). The current devices available for percutaneous transhepatic cholangioscopy have the ability to bend in two directions and do not have independent water delivery channels. However, the SpyGlass™ DS is essentially a parent-child/dual operator system cholangiopancreatography device that can be bent in four

directions and has independent water channels, allowing for detailed observation (6). In the present case, percutaneous transhepatic cholangioscopy using the SpyGlass™ DS allowed us to observe the bile ducts filled with bile sludge between many stones of different sizes while cleaning the bile sludge. When performing direct visualization bile duct biopsy, multiple biopsies are recommended due to the small sample size and low accuracy (7). Multiple biopsies were performed in the present case, all of which showed that the specimens were benign.

Endoscopic cholangiolithotripsy is used to remove gallstones up to 15 mm in diameter using a balloon catheter or basket catheter, and larger gallstones can be removed using either endoscopic mechanical lithotripsy, electrohydraulic shockwave lithotripsy, or laser lithotripsy (8). The removal rate of gallstones by electronic mechanical lithotripsy is good, ranging from 79% to 94.5% (9, 10). However, electronic mechanical lithotripsy may be difficult if the stones are very hard. It is difficult to grasp stones >20 mm with a basket, and it is also difficult to deploy the basket to release the gallstones when they are accumulated in the bile duct (11). Electrohydraulic shockwave lithotripsy and laser lithotripsy are mainly performed under peroral cholangioscopy, using electrohydraulic shock waves, or lasers to break up stones. Peroral cholangioscopy-guided lithotripsy is highly useful in cases with intrahepatic stones and stones in the gallbladder duct that are difficult to treat with conventional lithotripsy; the complete removal rate of stones is reported to be 77-92% (12, 13). It is important to note that there is a risk of damage to the bile ducts by shock waves or lasers and that the use of a biliary speculum may cause saline to flow backwards and increase the intraductal pressure, resulting in bacteremia or shock due to bile duct vagal reflex. Another problem is that the equipment is expensive, and the number of facilities that can perform this procedure is limited. In the present case, we used a double-balloon enteroscope to reach the anastomotic site after choledochojejunostomy. Since the gallstones were embedded at the site of anastomosis, the electrohydraulic shockwave lithotripsy probe was inserted directly into the endoscope, and lithotripsy was performed. After the impacted stone was removed, lithotripsy was continued. Electrohydraulic shockwave lithotripsy was used for the large gallstones, and almost all of the stacked stones were removed.

Although many studies have been conducted on the prevention of recurrence of bile duct gallstones, effective measures against recurrence remain unclear. There is no evidence concerning the efficacy of ursodeoxycholic acid, but it was reportedly effective in preventing the recurrence of common bile duct cholelithiasis in randomized controlled trials (14). Our patient had had gallstones since his 20s, and cholangitis

had recurred even after surgical treatment. The possibility of recurrence is high; thus, the patient has been carefully observed and followed up under treatment with oral ursodeoxycholic acid.

The authors state that they have no Conflict of Interest (COI).

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