Bangladesh Journal of Medicine (BJM)
ISSN : 1023 - 1986
eISSN : 2408 - 8366

CASE REPORT

RECURRENT CHOLANGITIS FROM COMMON BILE DUCT STONES AFTER CHOLECYSTECTOMY IN A PATIENT WITH DUODENAL PERIAMPULLARY DIVERTICULA

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Abstract:

Choledocholithiasis is a commonly encountered hepatobiliary disease. The gallstones may migrate into the common bile duct (CBD) or develop de novo in the CBD. Endoscopic Retrograde Cholangiopancreatography (ERCP) is the primary treatment for choledocholithiasis with reduced morbidity and similar efficacy compared to surgical exploration of the common bile duct. Some anatomic factors that promote biliary stasis include CBD dilation, periampullary diverticulum, and CBD stricture. We reported on a 61-year-old patient who underwent multiple ERCPs for recurrent cholangitis due to CBD stones. Each time she had fever, abdominal pain and jaundice. Periampullary duodenal diverticula were a risk factor for recurrent stone formation. Choledochoduodenostomy was performed to prevent recurrent CBD stone formation. At endoscopic follow-up, healed diverticula were noted.

Key words: *Recurrent cholangitis, duodenal periampullary diverticula, ERCP, choledochoduodenostomy*

Date of submission: 29.07.2025 *Date of acceptance: 30.08.2025*

DOI: https://doi.org/10.3329/bjm.v36i3.84038.

Citation: *Shrestha R, Shrestha M, Bhattarai K, Jha IK, Khadka D. Recurrent Cholangitis from Common Bile Duct Stones after Cholecystectomy in a patient with Duodenal Periampullary Diverticula. Bangladesh J Medicine 2025; 36(3): 155-159.*

Introduction:

The presence of stones in the common bile duct (CBD) defines choledocholithiasis. Stones can either form in the CBD or migrate to it from the gallbladder. Symptoms include episodic right upper quadrant (RUQ) abdominal pain, nausea, fever, jaundice, and weight loss. The patient may also develop biliary pancreatitis and cholangitis with increased morbidity and mortality due to severe complications. They may also develop within the CBD de novo due to bacterial infections of the bile ducts and biliary stasis.[1]

Since its introduction in 1974, initially as a procedure of choice for elderly unfit patients for surgical management of CBD stone (CBDS), ERCP with endoscopic sphincterotomy has become the procedure of choice for the treatment of CBDS in all categories of patients worldwide[2]. Although the procedure is usually considered safe and effective, it is not an innocuous one, and complications (4-16%), either early or late, may develop.

Periampullary diverticulum (PAD) refers to a mucosal or submucosal outpouching of the duodenum commonly caused by a defect of the local muscular layer and arising within a 2-3 cmradius of the major papilla[3,4]. Prevalence of PAD increases with age, and

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overall prevalence among the elderly is reportedly 65%[5]. The presence of duodenal periampullary diverticulum had also been reported as a risk factor for CBDS recurrence. Duodenal diverticulum, firstly recognized in 1710 by Chomel, is uncommon, with an incidence rate of 15-22% by post-mortem studies[6,7]. Periampullary diverticula are the most common type of duodenal diverticulum, usually located within two to three centimeters from the papilla of Vater[8]. Three different types of periampullary diverticulum are known, depending on the location of the major papilla: type 1 - inside the diverticulum, type 2 - adjacent to the diverticulum, and type 3 - outside the diverticulum.

Case report:

A sixty-one-year-old lady with a history of cholecystectomy one week prior presented to the Gastroenterology Department of National Academy of Medical Sciences, Kathmandu with a history of fever, abdominal pain, and jaundice. She had undergone open choledocholithotomy with cholecystectomy. Common bile duct exploration with T-tube exploration was done. Operative findings revealed multiple stones in the common Bile duct and dilated cystic duct. Postoperative T-tube cholangiogram showed a filling defect. near the tip of the T-tube in the common bile duct. Total leukocyte count was 13000 with neutrophilia. Her liver function test revealed total bilirubin: 3.1mg/ dl, direct 1.4mg/dl, SGOT (AST):184 U/L, alkaline phosphatase 853 IU/L, Hb% %:12gm/ dl. Amylase and lipase were within normal limits. Abdominal ultrasound demonstrated a dilated biliary tree with the presence of calculi in the distal common bile duct, along with upstream dilatation of the biliary tree. Endoscopic Retrograde Cholangio-pancreatography (ERCP) was performed. Periampullary diverticula noted. Initially, cannulation was difficult. Two diverticula, one on each side of the ampulla of different sizes. Larger diverticulum was around 1.5 cm in diameter and smaller diverticulum of 5 mm in diameter. Cholangiogram showed few filling defects. After selective cannulation over a guide wire technique, sphincterotomy was done. Two stones measuring up to 1 cm were extracted with the extraction balloon. A temporary plastic stent was placed in the common bile duct. T-tube cholangiogram showed no filling defect. She was discharged home. After two months of follow-up, her symptoms resolved, and the comprehensive metabolic panel was normalized (Bilirubin (total): 0.8 mg/dl, conjugated 0.3 mg/dl, SGPT:37 U/l, Alkaline phosphatase: 111IU/l. During repeat ERCP, the previous stent was removed, and a few debris and sludge were successfully extracted. Distal balloon occlusion cholangiogram revealed no remaining stone. On subsequent follow-up, the patient was symptom-free with no calculus in imaging. Her comprehensive metabolic panel was within normal range.

Two years later, the patient presented to the hospital with symptoms suggestive of cholangitis in the form of fever, abdominal pain, and yellowish discoloration of the body and high colored urine. Patient had conjugated hyperbilirubinemia along with transamnitis and raised alkaline phosphatase (Bilirubin was 8.7 mg/dl with Conjugated bilirubin 3.9mg/dl, aspartate transaminase 148 IU/l, and alanine transaminase:240 U/l. Alkaline phosphatase 366 U/l. She also showed a raised total leukocyte count with neutrophil predominance. MRCP again showed a CBD (1.2cm) with recurrence of choledocholithiasis. During ERCP, a single stone was extracted. No filling defects were confirmed on occlusion cholangiogram. Free flow of bile noted, and the common bile duct was not stented. Three days later, the patient had fever and abdominal pain, suggestive of cholangitis. It was thought that there might be retained stone, and decompression was done with the placement of a double pigtail stent.

Within nine months, she had recurrent cholangitis four times and was admitted at ICU. During her last ERCP, the patient had multiple stones (largest one 1 x 0.5 cm) and was extracted with mechanical lithotripsy. Frank discharge of pus was noted suggesting cholangitis. Double pigtail plastic stent deployed. The patient was referred to surgical Gastroenterology for the surgical management. The patient was re-evaluated along with radiological imaging and had normal tumor markers, with evidence of periampullary diverticula and choledocholithiasis. ECBD with choledocho-duodenostomy was done for biliary decompression with no diverticulectomy. Operative findings revealed dense omental adhesion. CBD diameter is approximately 2 cm. CBD stent in situ. Few microliths were present in CBD. Liver and peritoneum were normal during the operative evaluation.

The patient was followed up for five years with a six-monthly interval. The latest endoscopic image showed choledochoduodenostomy in the first part of the duodenum. The papilla was intact with no evidence of diverticula [Figure *5,6*]. Diverticula healed spontaneously after decompression.

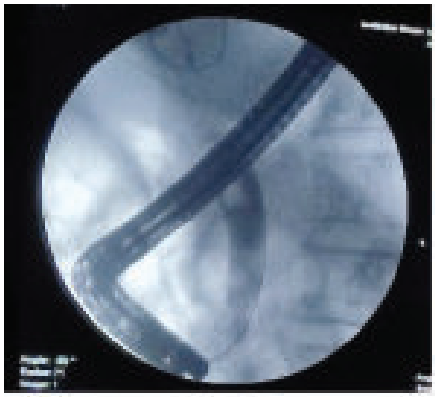

Fig.1: *Filling defects (stones) On cholangiogram*

Fig. 2: *Extraction of stones*

Fig.3: *Papilla with two diverticulum*

Fig.4:*Papilla with diverticulum*

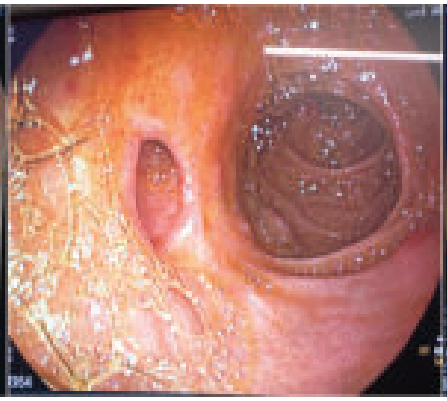

Fig.5: Choledochoduodenostomy in the first part of duodenum diverticula

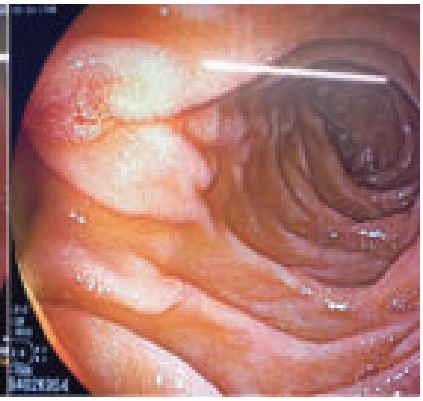

Fig.6: *Papilla with healed*

Discussion:

Five to thirty percent of patients have cholelithiasis along with choledocholithiasis (common bile duct stones (CBDS)) at the time of diagnosis[9]. CBDS are usually classified as primarywhen detected two years or more following cholecystectomy. When CBDSare detected up to two years following cholecystectomy, it is labelled as secondary CBDS. Recurrent CBDS are defined as stones found six months or more following initial endoscopic retrograde cholangiopancreatography (ERCP) with complete duct clearance[10,11].

Deng et al[9] found that the risk of common bile duct stone recurrence after ERCP was 1.599 times higher for patients with a stone diameter ≥10 mm compared to those with a stone diameter < 10 mm. A study involving 1148 patients found that stone diameter > 12 mm was more likely to recur.[12]

Several risk factors have been reported in various studies[13-18].This study showed that advanced age more than 65 years, history of choledocholithotomy, endoscopic papillary balloon dilatation(EPBD), endoscopic sphincterotomy (EST), CBD stent implantation, multiple ERCP procedures (more than two 2), stones in the intrahepatic bile duct, periampullary diverticulum, choledocholithiasis diameter ≥10 mm, bile duct-duodenal fistula, biliary tract infection, and no preoperative antibiotic use were independent risk factors for the recurrence of choledocholithiasis after ERCP.

Duodenal diverticulum was diagnosed radiologically by Case in 1913[19]. Duodenum is the second most common site of diverticula in the alimentary tract after colon, followed by jejunum, ileum, and stomach[20,21]. Several studies confirmed that the second part is the most common site, followed by the third and fourth parts of the duodenum. Periampullary diverticulum is usually asymptomatic, with only five percentage of patients presenting with complications. These complications include pancreatitis, biliary obstruction, choledocholithiasis, and, rarely tumors. PAD was classified as type 1, 2, or 3 according to the position of the major papilla from the endoscopic view: type 1, the major papilla was located inside of the diverticula; type 2, the major papilla was located at the edge of the

diverticula; type 3, the major papilla was located outside of the diverticula. PAD, especially type 1 PAD, is associated with an increased risk of acute pancreatitis as well as the occurrence and recurrence of CBD stones[22]. The relatively high prevalence of PAD (around 5–23%) documented in previous studies underscores the significant influence of these luminal defects on pancreaticobiliary diseases or the procedure of endoscopic retrograde cholangiopancreatography (ERCP)[7].In a study by Lapin et al. periampullary diverticulum was reported as the cause of massive gastrointestinal bleeding [23] .Venishetty N, et al. (July 20, 2023) reported periampullary diverticula causing recurrent pancreatitis[24].Grant Boileau identified two diverticula in 11 female subjects, compared to 13 in 122 male subjects[25]. Mackenzie et al. have also reported a female preponderance in the ratio from 1.6 to 1[26].

In cases of biliary obstruction, excision of the diverticulum (diverticulectomy) is anappropriate procedure, but it can be difficult and is associated with significant mortality andmorbidity[27]. Bypass procedures such as choledocho-duodenostomy, Roux-en-Ycholedocho,jejunostomy, and procedures such as duodenal diverticulization and duodenojejunostomy, aiming at duodenal decompression and inversion or reconstruction of the diverticular sac through aduodenotomy, may also be useful [28].

Conclusion:

Primary recurrent choledocholithiasis is usually uncommon. Frequent episodes of primary choledocholithiasis post-cholecystectomy increase the likelihood of future recurrence. Several risk factors are well known. In the absence of therapeutic consensus, a multidisciplinary team management approach is highly recommended. The patients with risk factor like periampullary diverticulum should be identified and referred for early surgical intervention if a second symptomatic episode cholangitis develop.

Conflict of Interest:

The authors stated that there is no conflict of interest in this study.

Funding:

This research received no external funding.

Consent for publication:

Informed written consent was taken from the parents of the patient to publish details relevant to the disease and management.

Acknowledgments:

The authors were grateful to the staffs of the Department of Gastroenterology, National Academy of Medical Sciences, Kathmandu, Nepal

Authors' contributions:

All authors were involved in the management of the patient and all authors contributed to the conception, writing, and editing of the case report.

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