

Recurrent Pancreatitis Caused by Stones in Remnant Intrapancreatic Choledochal Cyst Treated with Complete Re-Excision: Case Report

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1. Abstract

Recurrent pancreatitis due to stones in remnant intrapancreatic Choledochal Cyst (CC) is rare condition of postoperative course. We report the case of 55-year-old man with a history of recurrent abdominal pain who showed clinical and biochemical evidence of recurrent pancreatitis. He had received cyst excision with hepaticojejunostomy due to CC. We performed ERCP and operative stone removal in remnant intrapancreatic CC. But symptom was recurred. The abdominal pain and biochemical abnormality disappeared after complete re-excision of remnant intrapancreatic CC.

2. Keywords: Choledochal cyst; Pancreatitis; Stone

3. Introduction

It is well known that Choledochal Cyst(CC) is congenital dilatation of the biliary tree associated with Anomalous Pancreatico Biliary Ductal Union(APBDU), and complete excision is gold standard treatment. [1] CC is one of the risk factors of biliary tract malignancy and complete resection of CC is recommended for the treatment. Surgeons should be careful to avoid injury of the pancreatic duct when removing intrapancreatic portion. For this reason, some part of CC can remain in the intrapancreatic portion and lead to stone formation, recurrent pancreatitis, and cholangiocarcinoma in remnant intrapancreatic CC. In previous studies, protein stones or plugs developed in remnant IPCCs in 5% and 30% patients who presented with abdominal pain or recurrent acute pancreatitis during follow-up. [2,3] Complete surgical resection is recommended for such patients because of the risk of developing cholangiocarcinoma in remnant intrapancreatic CC. [4] However, some studies reported successful outcomes

for several patients with remnant intrapancreatic CC by using Endoscopic Retrograde Cholangiography (ERCP). [5-7] Herein, we report our treatment experience for the case of the stone formation in the remnant intrapancreatic CC with complete excision after stone extraction with ERCP and operation.

4. Case

45-year-old male was underwent cyst excision and Roux-en-Y hepaticojejunostomy for the treatment of CC in June, 1999. After the surgery, he discharged uneventfully and did not intake alcohol. However, he was referred to our institution due to acute pancreatitis in 2004. Abdominal Computed Tomography(CT) showed that there were 12mm and 5mm sized stones in remnant CC in intrapancreatic portion. We performed Endoscopic Retrograde Cholangio Pancreatography(ERCP) and tried to remove the stones. But, eventually we failed to remove them totally (**Figure 1**).

After then, he had been follow-up loss. In Sep. 2014, pancreatitis was recurred and we found that the size of the stones were increased to 20mm and 16mm in both abdominal CT and magnetic resonance cholangiopancreatography findings (**Figure 2**). We performed ERCP again, but we failed to remove the stones, could not find the common bile duct and decided to perform exploratory laparotomy. In the operation, we found suture material of the end of intrapancreatic common bile duct, opened the end and removed whitish stone. And then, we performed intraoperative choledochoscopy and identified no remnant stone in the remnant intrapancreatic CC. After confirmed chronic inflammation in frozen section, we finished the operation remaining remnant intrapancreatic CC. The patient was discharged on postoperative day 8

without any complication. In checked the follow-up CT, the size of the remnant intra pancreatic CC was decreasing from 50mm to 30mm. But, new stone developed in the remnant cyst with pancreatitis after 2 years (**Figure 3**). At Nov. 2016, we did re-exploration and complete re-excision of remnant intrapancreatic CC. (**Figure 4**) The patient was discharged on postoperative day 8 without any complication. The abdominal pain and biochemical abnormality have been disappeared after complete re-excision of remnant intrapancreatic CC for 3 years.



Figure 1. Dilated remnant intrapancreatic CC and hyperdense stone after 1st operation(at 2004)

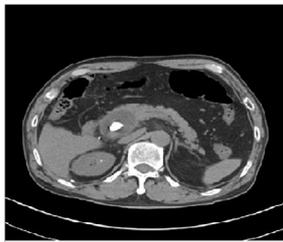


Figure 2. Increasing remnant intrapancreatic CC and stone (at 2014)

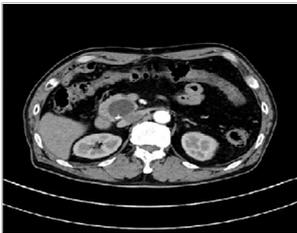


Figure 3. Recurred stone in remnant CC after extraction in remnant CC (at 2016)

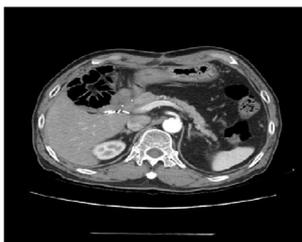


Figure 4. After complete resection of remnant CC (at 2018)

5. Discussion

Complete resection of the extrahepatic bile duct and hepaticojejunostomy is considered as a standard treatment modality for CC and provide excellent long-term prognosis with few complications. However, most of the reports have focused on the surgical treatment for CC and issues about long-term complications were overlooked. Ono et al. presented late complications postoperative liver dysfunction, persistent intrahepatic duct dilatation, recurrent abdominal pain, repeated cholangitis and intrahepatic lithiasis [8]

It is impossible that biliary stone develops in remnant CC after surgical biliary diversion. Therefore, it can be explained as the mechanism of pancreatic stone formation in chronic pancreatitis or increased size of unidentified sludge or stone that exist at the time of the initial operation. Protein plugs and stones were detected in 28%–36% of intrapancreatic CCs by routine intraoperative endoscopy during cyst excision and the incidence of stone formation reduced to 5.3%. [9,10] In our patients, he experienced recurrent pancreatitis with stone formation after complete removal the stones using intraoperative choledochoscope during 2nd operation. The formation of pancreatic stones must have two basic conditions, change of pancreatic fluid component and pancreatic obstruction.[11] In our case, the cause of stone formation was considered that flow disturbance of the pancreatic juice into remnant CC and accumulation of calcium protein plug could develop the stone formation because of no pancreatic duct dilatation. The remnant CC in the patient with Anomalous Pancreatico Biliary Ductal Union (APBDU) can compress pancreatic duct and disturb the flow of pancreatic juice causing pancreatitis and stone formation.

The treatment options for residual intrapancreatic CC are reoperation for complete resection, close observation, and endoscopic treatment. [12] Because of malignant risk of intrapancreatic remnant CC, most studies showed that radical excision of the dilated cystic remnant, buried in the pancreatic head, was associated with low morbidity rates and yielded favorable long-term outcomes. ERCP can be performed to remove remnant stones in intrapancreatic CC and resolved pancreatitis symptom. [5-7] However, pancreatitis can be recurred after stone removal because of protein plugs and stone. We thought that complete re-excision of the remnant intrapancreatic CC could be the best way in recurrent pancreatitis due to stones in remnant intrapancreatic CC.

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