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Original article

Symptomatic pancreatic duct stones in the disconnected bile duct: A case series

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ABSTRACT

Background: Pancreaticobiliary maljunction (PBM) refers to the union of the pancreatic and biliary ducts outside of the duodenal wall. Patients are at increased risk of bile duct and gallbladder cancer, likely secondary to pancreatic juice refluxing into the biliary tree, and it is recommended that they undergo biliary diversion.
Methods: This is a case series of all patients in our institution with PBM and bilioenteric anastomosis who presented with symptomatic pancreatic duct stones in a disconnected bile duct. IRB approval was obtained prior to the initiation of the study.
Results: We describe eight cases of this finding. All patients underwent ERCP, with stones successfully removed from the disconnected bile duct in seven patients and from the pancreatic duct in one patient.
Conclusion: This novel finding has not been described in the medical literature, and may become more prevalent as more patients with PBM undergo bilioenteric anastomosis.

1. Introduction

Pancreaticobiliary maljunction (PBM) is a rare congenital anomaly where the pancreatic duct and bile duct join outside the duodenal wall forming a long common channel [1]. The prevalence has been estimated at 0.03% of the population in a Japanese autopsy study, though it is likely lower in Western countries [2,3]. Several studies have shown that patients with PBM are at increased risk for choledochal cysts as well as bile duct and gallbladder carcinoma, likely secondary to reflux of pancreatic juice into the biliary tree leading to metaplasia, dysplasia and eventually carcinoma [4–8]. Although there are currently no U.S. guidelines on this topic, in Japan, where PBM is more common, it is recommended that all patients with PBM and biliary dilation undergo prophylactic cholecystectomy and flow-diversion surgery [9]. Surgical management of patients with PBM without biliary dilation is more controversial, though some surgeons choose to perform biliary diversion given that the risk of biliary cancer is higher than in the general population [10,11].

In addition to cancers, PBM is associated with pancreatitis and bile duct stones, which affect 17.9% of patients with biliary dilation and 27.3% of patients without biliary dilation [12]. To date, however, the presence of pancreatic duct stones in the remnant bile duct has not been described. Pancreatic duct stones are easily recognized during ERCP by their characteristic white appearance. We present eight cases of pancreatic duct stones removed from the remnant bile duct during ERCP in patients who have undergone prior bilioenteric anastomosis.

2. Patients and methods

This was a retrospective, observational, consecutive case study conducted at a single tertiary care center. Only patients with a history of biliary diversion surgery who presented with pancreatic duct stones in a disconnected bile duct were included (Figs. 1 and 2). This study was approved by the Institutional Review Board at our institution.

3. Results

Eight patients, median age 51 years (range 6–71), were identified. All patients had PBM and had undergone bile duct resection and bilioenteric anastomosis, 7 with hepaticojejunostomy and one with end-to-side choledochoduodenostomy. Two of the 8 patients

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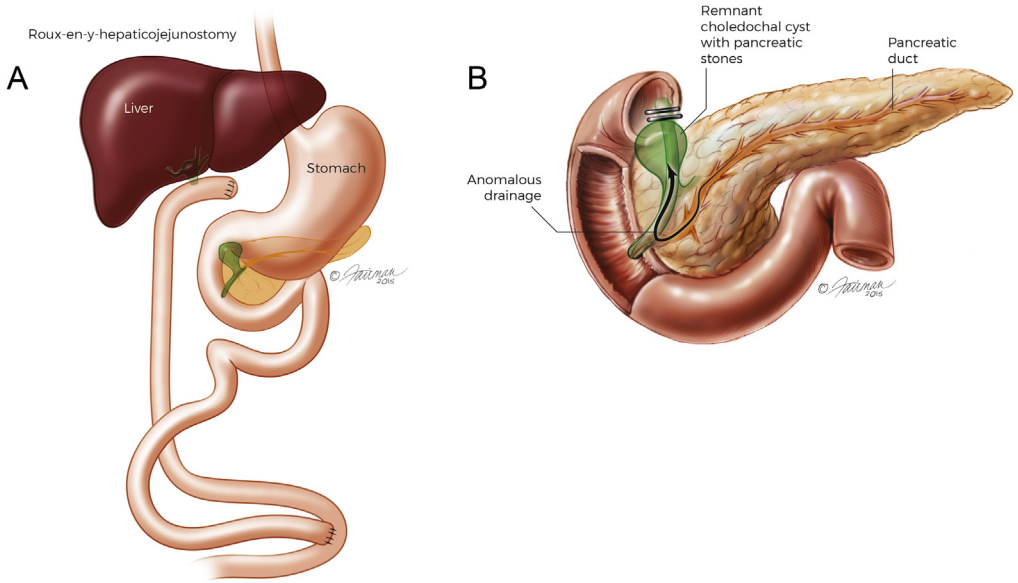

Fig. 1. (a) Anatomy after Roux-en-y-Hepaticojejunostomy; (b) pancreatic juice refluxes into the remnant choledochal cyst, where stasis leads to stone formation.

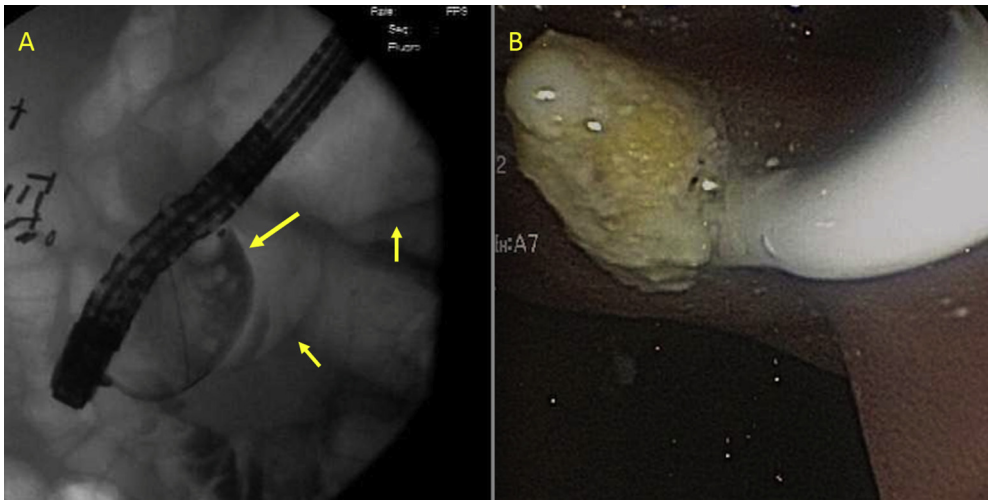

Fig. 2. Note dilated remnant choledochal (CD) cyst (long arrow) filled with pancreatic duct stones in patient who had previously undergone Roux-en-Y hepaticojejunostomy for CD cyst and pancreatic biliary maljunction (a). Short arrows depict pancreatic duct following a large sphincterotomy. Note pancreatic duct stone delivery using an extraction balloon (b).

had undergone biliary diversion surgery for choledocholithiasis. Three underwent surgery for choledochal cyst, 2 because of recurrent pyogenic cholangitis, and 1 for resection of an indeterminate biliary stricture (Table 1). Clinical presentation was abdominal pain in all patients, 4 of whom were found to have acute on chronic pancreatitis. In 3 of these cases, pancreatitis preceded biliary diversion surgery; one case developed afterwards. All patients had abnormal imaging prior to ERCP, 6 with biliary dilation in the disconnected distal bile duct or filling defects, and 2 with pancreatic duct dilation.

Seven patients had ERCP with sphincterotomy or balloon sphincteroplasty with successful removal of pancreatic stones from the remnant bile duct. One patient had her CBD-PD junction enter a peripheral PD side branch, precluding access to the disconnected CBD, and underwent extracorporeal lithotripsy to fragment the pancreatic stones and fragment retrieval from the PD as opposed to the remnant CBD. The size of the remnant bile ducts ranged from 5.4 × 22.9 mm to 17.7 × 36.2 mm. The number of stones in the remnant bile ducts ranged from one to eight and from 5.4 mm to 22.9 mm in greatest diameter. One patient developed mild post-ERCP pancreatitis.

Four of the 8 patients required a second ERCP at a later date for recurrent symptoms. However, the frequency of admissions for pancreatitis dropped from a median of 4.5/year to 1.5/year. Three of the 4 patients with pancreatitis had improvement in frequency of pancreatitis episodes on follow-up (range 6 months to 6 years). One patient was lost to follow-up.

4. Discussion

The finding of symptomatic pancreatic duct stones in a disconnected bile duct has not been previously described in the literature. The etiology is likely stasis of pancreatic juice that has refluxed into the remnant bile duct or sump, which may behave as a “cyst” once

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