



Preoperative endoscopic extraction of a huge pancreatic protein plug in a choledochal cyst: A case report



Noboru Oyachi^{a,*}, Kazuko Obana^a, Takeyuki Suzuki^a, Tazue Ochiai^b, Hitoshi Mochizuki^b

^a Department of Pediatric Surgery, Yamanashi Prefectural Central Hospital, 1-1-1 Fujimi, Kofu-city, Yamanashi 400-8506, Japan

^b Department of Gastroenterology, Yamanashi Prefectural Central Hospital, Kofu-city, Japan

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ABSTRACT

A 4-year-old female was referred to our institution with acute pancreatitis due to a choledochal cyst. A protein plug was found to be deeply impacted in the main pancreatic duct. She underwent preoperative endoscopic retrograde cholangiopancreatography (ERCP) followed by sphincteroplasty and extraction of the protein plug with a basket extractor. After the ERCP procedure, the patient successfully underwent total choledochal cyst excision with Roux-en-Y hepaticojejunostomy. In cases in which a huge protein plug is located deep in the pancreatic duct, therapeutic ERCP is useful as preoperative management for choledochal cyst excision with hepaticojejunostomy.

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An intraductal pancreatic protein plug is a worrisome problem in patients with a choledochal cyst. However, its surgical treatment has not been well discussed. We describe a 4-year-old female with a choledochal cyst that was accompanied by a huge protein plug that was impacted in the distal main pancreatic duct, and which was extracted endoscopically.

1. Case report

A 4-year-old female presented with abdominal pain and vomiting, and was diagnosed with a choledochal cyst by abdominal ultrasound and acute pancreatitis based on elevated serum pancreatic enzymes (amylase 216 IU/L, lipase 323 IU/L) and LFTs (TBil 1.55 mg/dL, DBil 0.64 mg/dL, AST 260 IU/L, ALT 499 IU/L, GGTP 761 IU/L).

Abdominal ultrasound, CT and MR-cholangiopancreatography (MRCP) revealed cystic dilation of the common bile duct (maximum diameter 17 mm) accompanied by a slightly dilated intrahepatic duct. The choledochal cyst was equivalent to Todani classification IV-A. Furthermore, a radiolucent filling defect (2 cm in length) was

detected in the main dilated pancreatic duct to the common channel by MRCP and abdominal ultrasound. While her symptoms of pancreatitis resolved by conservative therapies within one week after admission, the pancreatic substance did not disappear and remained lodged in the main pancreatic duct.

We performed diagnostic and therapeutic endoscopic retrograde cholangiopancreatography (ERCP) under general anesthesia preoperatively. The scope used for this patient was manufactured for adult use (Olympus JF-240, Olympus, Ltd., Tokyo, Japan: scope diameter 12.6 mm with a 3.2 mm channel). In ERCP, the common bile duct was expanded and the patient had a long common channel (30 mm in length) with a pancreaticobiliary junction. The pancreatic intraductal filling defect was a huge protein plug (20 mm × 9 mm) that was located in the main pancreatic (Wirsung's) duct (Fig. 1). The substance was unmovable despite the washout-effect of the contrast agent. Therefore, endoscopic extraction was performed following sphincteroplasty. A guide wire was placed in the pancreatic duct and a balloon dilator (6 mm in diameter) was inserted into the duct over the wire. After sphincteroplasty, the substance was retrieved with a basket lithotripter as a mass via the papilla of Vater and it was proved to be a protein plug (Fig. 2). Complete clearance was confirmed by cholangiography.

She did not develop post-ERCP pancreatitis and underwent surgical treatment consisting of excision of the choledochal cyst and Roux-en-Y hepaticojejunostomy. The collected gallbladder bile demonstrated elevated amylase (41,236 IU/L), which was due to the

* Corresponding author. Tel.: +81 55 253 7111; fax: +81 55 253 8011.

E-mail address: oyachi-ampw@ych.pref.yamanashi.jp (N. Oyachi).

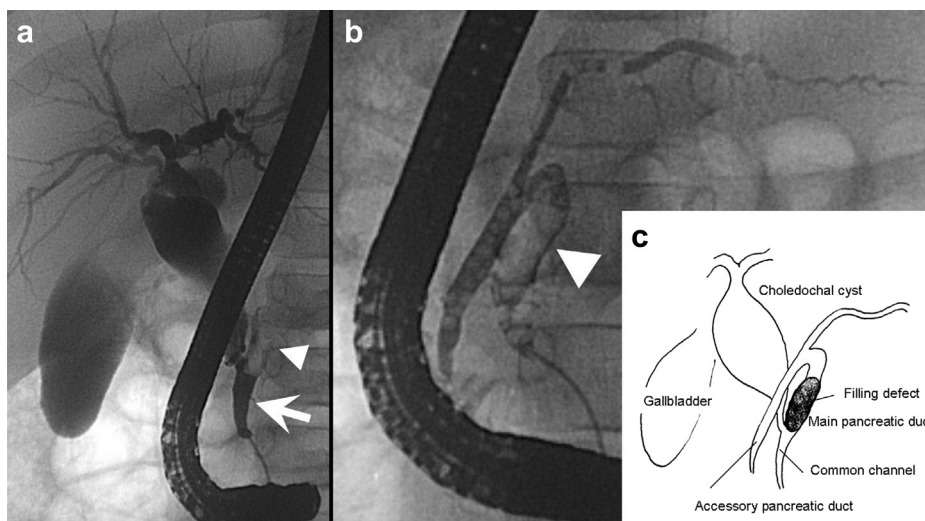


Fig. 1. ERCP showing a choledochal cyst with a pancreaticobiliary maljunction complicated with a huge obstructing protein plug in the main pancreatic duct. a) cholangiography, b) pancreatography, c) gross scheme. ERCP shows a pancreaticobiliary maljunction forming the common channel (arrow) for a length of up to 30 mm. The pancreatic protein plug was detected as a huge filling defect (arrowhead: 20 mm × 9 mm), which impacted and caused dilatation of the main pancreatic (Wirsung's) duct.

reflux of pancreatic enzymes into the common duct. The post-operative course was uneventful.

2. Discussion

In the common form of choledochal cyst, a pancreaticobiliary maljunction permits the reflux of pancreatic enzymes into the common bile duct through the maljunction, and it has been proposed that the associated symptoms are caused by this reflux [1]. On the other hand, a protein plug may also be found in the common channel or pancreatic duct and has been proposed to be responsible for the symptoms due to obstruction of the bile and pancreatic ducts [2,3].

If a radiographic investigation shows a filling defect in the pancreaticobiliary system, a protein plug should usually be suspected and the simultaneous occurrence of symptoms may be explained by the consequent disturbance of biliary pancreatic secretory flow. This finding may be supported by analysis at the time of surgery [3]. A common channel protein plug complicating choledochal cyst is found in 10%–40% of cases, however, the

association of a pancreatic protein plug is considered rare [3,4]. The mechanism of protein plug formation in choledochal cysts has been gradually elucidated [5], but does not still become clear. The protein plug may act as a ball valve and produce a transient and abrupt elevation in intraluminal pressure in both the bile and pancreatic duct [6].

Kamisawa and co-workers reported that a filling defect disappeared spontaneously in 50% of patients [7]. However, if protein plugs remain incarcerated or persist until the time of surgery, a preoperative or intraoperative maneuver for protein plug extraction is chosen. Although the treatment of protein plugs located in the pancreatic duct has not been discussed well, the representative English language literatures on management of pancreaticobiliary protein plugs associated with choledochal cyst in children are displayed in Table 1. Preoperatively, endoscopic short-tube stenting was effective when ERCP showed the common channel protein plug impaction in patients with persistent or worsening symptoms [8] and endoscopic protein plug extraction or irrigation for the washout in the common channel is adequate and effective for the management of small protein plugs as well [9,10]. On the other

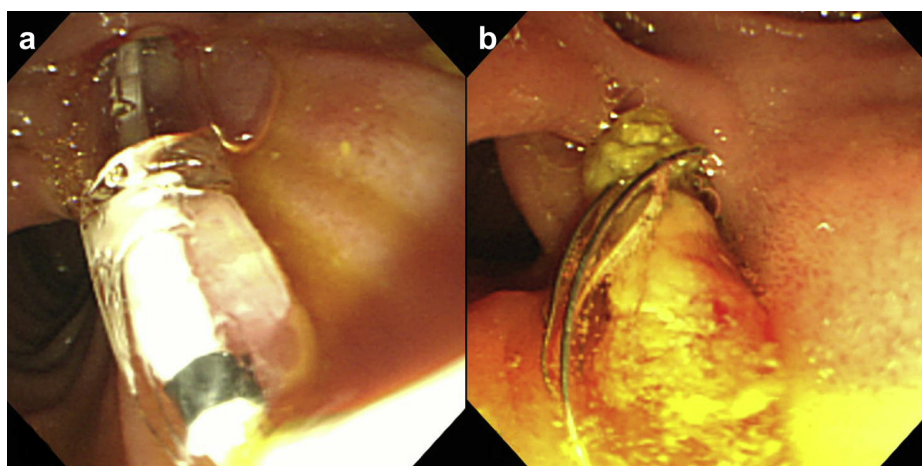


Fig. 2. a) Endoscopic sphincteroplasty and b) protein plug extraction. The protein plug was stagnant despite the washout-effect by contrast medium. Endoscopic sphincteroplasty was followed by extraction of the protein plug with a basket lithotripter. The plug was crushed but extracted as a mass via the papilla of Vater.

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