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CASE REPORT

내시경역행담췌관조영술 후 발생한 복강 가성동맥류 파열

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Visceral Artery Pseudoaneurysm Rupture after Endoscopic Retrograde Cholangiopancreatography

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A visceral artery pseudoaneurysm after ERCP is a rare adverse event that is potentially life-threatening. Most cases reported previously originated from the peripancreatic arteries, including the splenic artery, gastroduodenal artery, or pancreaticoduodenal artery. The mechanism of the occurrence of visceral artery pseudoaneurysms after ERCP has not been elucidated until now. Recently, a pseudoaneurysm rupture originating from the superior mesenteric artery after ERCP was observed in a patient without a history of pancreatitis. This paper reports this case with a review of the relevant literature. **(Korean J Gastroenterol 2020;75:162-166)**

Key Words: Endoscopic retrograde cholangiopancreatography; Pseudoaneurysm; Rupture

INTRODUCTION

A visceral artery pseudoaneurysm is a rare vascular complication caused by peripancreatic vascular erosion or vascular wall component disruption induced by pancreatic enzymes from acute or chronic pancreatitis, necrotizing pancreatitis, pancreatic tumor, vascular anomaly, and trauma.[1] To date, most reported visceral artery pseudoaneurysms have been associated with acute, chronic, or necrotizing pancreatitis.[1,2] The splenic artery, gastroduodenal artery, and pancreaticoduodenal artery are frequently reported locations of pseudoaneurysms.[3] A visceral artery pseudoaneurysm after ERCP without pancreatitis is extremely rare.[4] A pseudoaneurysm after ERCP usually occurs in the gastroduodenal artery or pancreaticoduodenal artery, which supplies blood to the major papillary area. The rupture of a pseudoaneurysm from the pancreaticoduodenal artery or gastroduodenal artery is potentially life-threatening.[5,6] This paper reports a case of pseudoaneurysm rupture originating from the superior mesenteric artery (SMA), which is a very rare location, with a review of the literature.

CASE REPORT

A 55-year-old man presented with severe abdominal pain and visited the emergency room. He had a prior medical history of diabetes that had not been managed. He was a current smoker and a social drinker. The abdominal pain was asso-

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ciated with fever and chills. An examination revealed a body temperature of 38.3℃ with his other vital signs unremarkable. Tenderness was noted in the epigastric area, but there was no rebound tenderness. Icteric sclera was noted.

The laboratory findings revealed the following: leukocytosis of 23,220/mm^3, hemoglobin level of 14.7 g/dL, total bilirubin of 8.0 mg/dL, an AST level of 316 IU/L, ALT level of 350 IU/L, ALP level of 317 IU/L, GGT level of 1,191 IU/L, and hemoglobin A1c level of 8.6%. An abdominal CT scan revealed diffuse gallbladder wall edema with pericholecystic fluid collection and subtle calcification in the distal common bile duct (CBD) (Fig. 1). MRCP failed due to claustrophobia.

Intravenous third-generation cephalosporin was administered to control the acute cholangitis, and therapeutic ERCP was performed for CBD stone removal and biliary drainage. Written informed consent for ERCP was obtained from the patient before the procedure. ERCP was performed under conscious sedation using midazolam and propofol. After selective cannulation of the CBD, the suppurative discharge was drained through the papilla, but the patient showed a severe paradoxical response. Therefore, the procedure was stopped, and the endoscope was withdrawn for the safety of the patient. After stabilizing the paradoxical activity, ERCP was performed without sedation. On cholangiography, a filling defect was noted in the distal CBD (Fig. 2A). The endoscopic sphincterotomy (EST) was performed until the hooding fold of the ampulla of vater using a pull type sphincterotome (Ultratome^{TM} XL; Boston Scientific Co., Natick, MA, USA). The mode of electrocautery used in EST was ENDO CUT I mode (upmax, 550 Vp; effect, 2; cut duration, 2; cut interval, 2) (ERBE$^{®}$; Erbe Elektromedizin GmbH, Tuebingen, Germany). A papillary balloon dilator or papillary large balloon dilator was not used in sphincter management. After EST, a round-shaped stone was removed using a retrieval balloon catheter (Fig. 2B). Six Fr endoscopic nasobiliary drainage was then performed.

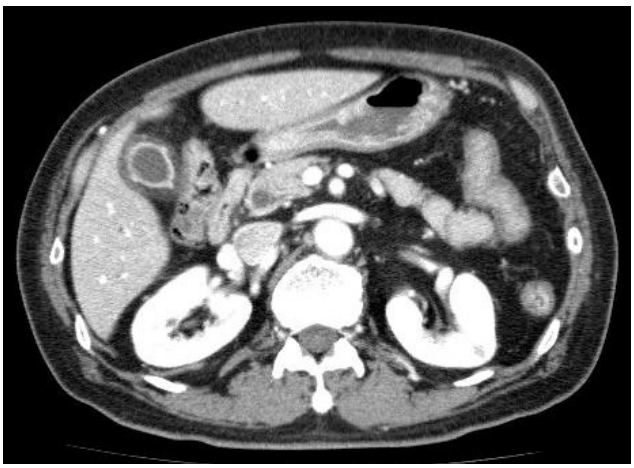

Fig. 1. Diffuse GB wall edema with pericholecystic fluid collection and subtle calcification in the CBD were noted in the abdominal CT scan. GB, gallbladder; CBD, common bile duct; CT, computed tomography.

After ERCP, there was no evidence of adverse events, such as pancreatitis, bleeding, or bowel perforation. After 1 week, the liver function test result normalized, and the patient was transferred to the department of general surgery for a laparoscopic cholecystectomy. On the day of the elective operation, the patient complained of sudden abdominal distension and dizziness before the operation, and the procedure was cancelled. The vital signs showed 80/60 mmHg, and the he-

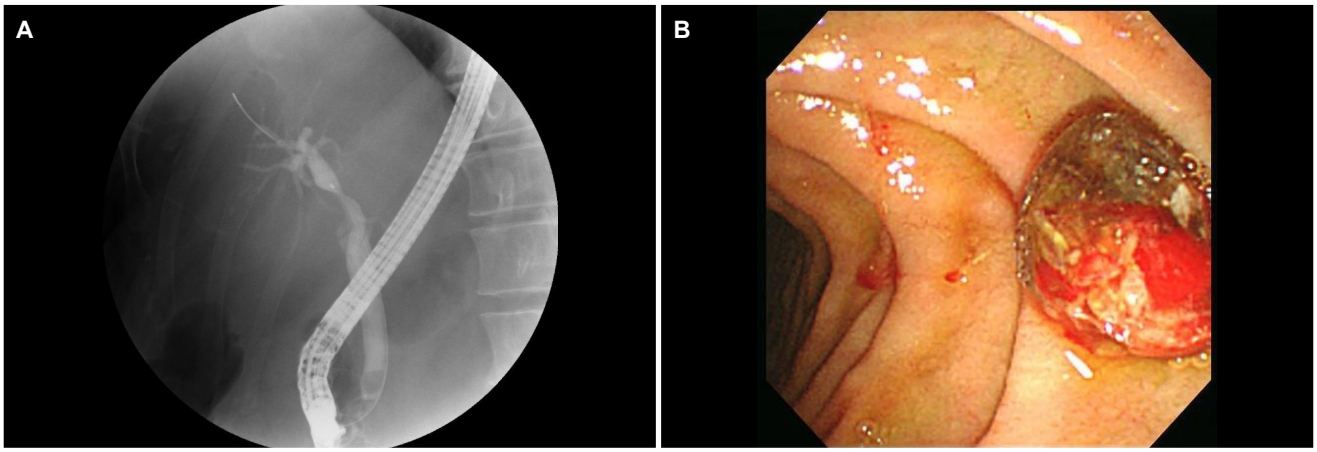

Fig. 2. Endoscopic retrograde cholangiopancreatography. (A) A filling defect was noted in the distal CBD during cholangiography. (B) After EST, a round stone was removed using a retrieval balloon catheter. CBD, common bile duct; EST, endoscopic sphincterotomy.

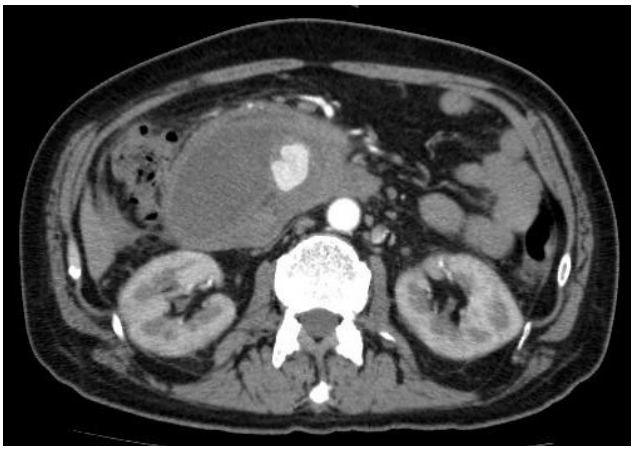

Fig. 3. 2.5 cm sized heart-shaped contrast leakage was noted inside the hematoma on enhanced abdominal CT. CT, computed tomography.

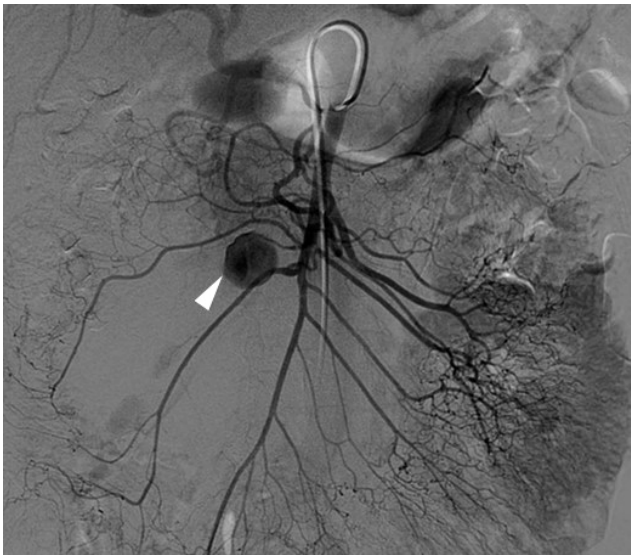

Fig. 4. Contrast leakage (arrowhead) from the right side wall of the first jejunal branch originating from the SMA was noted on angiography. SMA, superior mesenteric artery.

moglobin level was 7.5 g/dL. No symptoms of gastrointestinal bleeding, such as hematemesis, melena, or hematochezia, were noted. On an abdomen CT scan, a 12×9 cm sized well-capsulated hematoma was observed in the retroperitoneal area, and 2.5 cm heart-shaped contrast leakage was noted inside the hematoma (Fig. 3). As a result, the rupture of a visceral artery pseudoaneurysm was diagnosed. Angiography was performed to evaluate and manage the pseudoaneurysm. On angiography, contrast leakage was confirmed at the right side wall of the first jejunal branch originating from the SMA, and embolization was performed using 0.5 mL of Histoacryl® (N-butyl-2-cyanoacrylate; Braun, Melsungen, Germany) (Fig. 4). Four weeks after angioembolization, the size of the retroperitoneal hematoma was decreased to 8×7 cm, and the Histoacryl® (N-butyl-2-cyanoacrylate; Braun) and lipiodol agglomerate were noted inside the pseudoaneurysm. No contrast leakage from the embolized first jejunal branch of the SMA was noted on enhanced abdominal CT (Fig. 5). The patient was discharged without additional adverse events. The Institutional Review Board of the Inje University Seoul Paik Hospital approved this case report (IRB No. PAIK 2019-02-002).

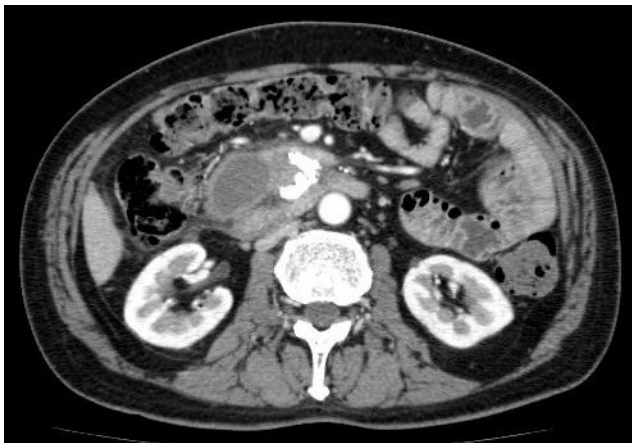

Fig. 5. No contrast leakage from the embolized first jejunal branch of the SMA was observed on enhanced abdominal CT. SMA, superior mesenteric artery; CT, computed tomography.

DISCUSSION

Visceral artery pseudoaneurysm can be caused by vascular wall damage from pancreatitis, malignancy, vascular malformation, or trauma.[1,2] A visceral artery pseudoaneurysm is a rare adverse event, but it can be potentially life-threatening. A visceral artery pseudoaneurysm is clinically associated with pancreatitis, and most cases originate from the peripancreatic arteries, including the splenic artery, gastroduodenal artery, or pancreaticoduodenal artery.[3] The mortality of visceral artery pseudoaneurysm rupture has been reported to be 20% in cases caused by chronic pancreatitis and up to 60% in acute pancreatitis.[7] The mortality can be higher in the rupture of pseudoaneurysms originating from the pancreaticoduodenal artery than in the rupture of pseudoaneurysms originating from the splenic artery or gastroduodenal artery.[7]

Among cases previously reported, a visceral artery pseudoaneurysm after ERCP without the association of acute or

Table 1. Clinical Features of Visceral Artery Pseudoaneurysm Rupture after ERCP

Study	Sex/ age	Underlying disease	Anatomy	Indication for ERCP	Details of procedure	Post-ERCP pancreatitis	Location of pseudoaneurysm	Treatment
Al-Jeroudi et al. (2001)[8]	F/76	None	Normal	Pancreas cancer	Precut sphincterotomy	No	Pancreaticoduodenal artery	Embolization
Gaduputi et al. (2013)[4]	M/74	Chronic hepatitis C	Billroth II gastrectomy	CBD stone	Unintended PD cannulation	Yes	Gastroduodenal artery	Embolization
Kurita et al. (2015)[5]	F/71	Chronic renal failure	Normal	CBD stone	EPLBD	No	Gastroduodenal artery	Embolization
Priya et al. (2016)[6]	M/64	None	Normal	Acute cholangitis with CBD stone	EST, plastic stent	No	Gastroduodenal artery	Embolization
	M/72	None	Normal	Periampullary mass with jaundice	EST, plastic stent	No	Gastroduodenal artery	Embolization
Mohapatra et al. (2017)[11]	M/53	None	Normal	Acute cholangitis with CBD stone	EST, plastic stent	No	SMA	Embolization
El Hajj et al. (2017)[10]	F/55	None	Normal	Acute cholangitis with CBD stone	EST, plastic stent	No	Right hepatic artery	Stent graft
Ding et al. (2017)[9]	M/56	Chronic hepatitis B	Liver transplantation	CBD stricture	EST, plastic stent	No	Left hepatic artery	Embolization
Current case	M/55	DM	Normal	Acute cholangitis with CBD stone	EST, ENBD	No	SMA	Embolization

ERCP, endoscopic retrograde cholangiopancreatography; F, female; M, male; CBD, common bile duct; PD, pancreatic duct; EPLBD, endoscopic papillary large balloon dilation; EST, endoscopic sphincterotomy; SMA, superior mesenteric artery; DM, diabetes mellitus; ENBD, endoscopic nasobiliary drainage.

chronic pancreatitis is very rare. Until now, the mechanism of occurrence of visceral artery pseudoaneurysms after ERCP has not been investigated. Several hypotheses have been proposed, including direct mechanical injury from the accessories or endoscope, ischemic injury from electric stimulus, and secondary injury from pancreatitis.[4,6] The rupture of a visceral artery pseudoaneurysm after ERCP occurs in diverse patients with a range of characteristics, such as gender; middle to advanced age; surgically altered anatomy, including Billroth II gastrectomy and liver transplantation; association with pancreatitis; sphincter therapy, including EST; endoscopic papillary large balloon dilation; and precut sphincterotomy.[4-6,8-11] In most cases, fatal hemorrhage occurred, and the clinical presentations were diverse, including gastrointestinal bleeding, hemobilia, intra-abdominal hematoma, and hypovolemic shock. In the current case, there were several risks of adverse events according to the previously reported cases. These included middle age, an indication of ERCP for CBD stone, and the use of EST as sphincter management. Among them, the extent of the sphincterotomy and mode of electrocautery in performing the EST can be a procedure-related risk of this event. Therefore, technical issues should be considered as a cause of this adverse event. Table 1 lists the detailed clinical features of cases of visceral artery pseudoaneurysm rupture after ERCP.

In conclusion, this paper reports a case of a pseudoaneurysm rupture of the SMA after ERCP, which is a very rare location for a pseudoaneurysm. The pseudoaneurysm rupture of the SMA was managed successfully with angioembolization. The current case reminds the ERCP endoscopist always to be alert to the occurrence of unexpected and rare but possibly lethal adverse events after ERCP.

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