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Single Case

Effectiveness of Endoscopic Pancreatic Stenting for Pancreatic Pseudocyst-Portal Vein Fistula

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Keywords

Pancreatic pseudocyst-portal vein fistula · Endoscopic pancreatic stenting · Pancreatitis

Abstract

Pancreatic pseudocyst-portal vein (PP-PV) fistula, mostly occurring after pseudocyst formation following acute/chronic pancreatitis, is a rare but life-threatening condition. The majority of treatments are based on conservative or surgical interventions. We report the case of a 70-year-old man with a PP-PV fistula and PV thrombosis. We adopted conservative treatment at first due to his mild symptoms. However, after resuming food intake, the patient had severe abdominal pain. Following endoscopic retrograde cholangiopancreatography, we found that the pseudocyst was connected with the PV through the fistula. Subsequently, an endoscopic nasopancreatic drainage (ENPD) catheter was inserted into the main pancreatic duct to establish pancreatic drainage, which resulted in a decrease in the abdominal pain. After the ENPD tube had been exchanged for endoscopic pancreatic stenting, his abdominal pain did not recur. Therefore, this case demonstrated endoscopic treatment as an effective treatment option for PP-PV fistula.

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Introduction

Pancreatic pseudocysts (PPs) are common complications occurring in the course of acute/chronic pancreatitis, which can result in bleeding, infection, and compression. However, PP rupture into the portal vein (PV) is an extremely rare and serious complication. Twenty-nine cases of PP-PV fistula have been reported to date in the literature since 1966 (Table 1) [1–14]. Out of 29 patients, 13 underwent conservative therapy, 12 required surgical intervention, and 4 improved with endoscopic treatment (pancreatic stent, $n = 2$; cyst-gastrostomy, $n = 2$). Out of the 29 patients, 7 died (conservative therapy, $n = 6$; surgical treatment, $n = 1$). All patients who had undergone endoscopic treatment were alive. Although the majority of the treatments are based on conservative or surgical approaches, conservative therapies show poor patient prognosis, whereas surgical methods are often invasive. Here, we report a case of PP-PV fistula which was managed effectively with endoscopic pancreatic duct stenting.

Case Report

A 70-year-old alcoholic man had a history of occasional stomachache over several years. He was admitted previously to another hospital with severe abdominal pain. An abdominal computed tomography (CT) scan demonstrated severe pancreatitis and a pseudopancreatic cyst (diameter 15 mm) in the pancreatic head region (day 1) (Fig. 1a). The patient had a history of gastrectomy with Billroth II reconstruction for stomach cancer. After 3-week pancreatitis treatment, a CT scan showed a PV thrombosis (day 21) (Fig. 1b). For the PV thrombosis treatment he was first administered heparin (20,000 U/day). One week later, a CT scan showed a slightly high-attenuation fluid within the pseudocyst; under the suspicion of rupture of the aneurysm he was transferred to our hospital (day 28) (Fig. 1c).

At the time of admission, the patient had clear consciousness with vital signs within the normal range. Physical examination revealed epigastric pain and anemia. No gastrointestinal bleeding was found on endoscopy. Laboratory blood test results included red blood cells $287 \times 10^4/\text{mm}^3$ (normal range $438\text{--}555 \times 10^4/\text{mm}^3$), hemoglobin 9.2 g/dL (normal range 13.7–16.8 g/dL), pancreatic amylase 95 U/L (normal range 16–52 U/L), and lipase 277 IU/L (normal range 9–59 IU/L). Following admission, abdominal angiography was performed immediately, which demonstrated no active bleeding source and aneurysm within the cyst. At first, conservative treatment was adopted for the pseudocyst. The patient was transfused with red blood cells for the treatment of anemia and administered antithrombin-III for PV thrombosis. Follow-up CT showed a decrease in the size of the thrombus in the PV (day 31) (Fig. 2a). As his symptoms gradually improved, he resumed food intake; however, this led to severe abdominal pain (day 38). Endoscopic retrograde cholangiopancreatography demonstrated that the pseudocyst was connected with the PV by a fistula (Fig. 2b). A 5-French endoscopic nasopancreatic drainage (ENPD) catheter was inserted into the main pancreatic duct to establish pancreatic drainage (day 42) (Fig. 2b), which resulted in a decrease in the abdominal pain. A three-dimensional image of the PP-PV fistula is shown in Figure 3. Follow-up CT 2 weeks later showed a significant decrease in the size of the pseudocyst, with pancreatography through the ENPD tube demonstrating resolution of the PP-PV fistula (day 56) (Fig. 2c). After the ENPD tube had been exchanged for endoscopic pancreatic stenting, his abdominal

pain did not recur, and the patient was discharged on day 80. Up to now, the endoscopic pancreatic stent has been changed every 3 months.

Discussion

Mechanism of Fistula Formation

The mechanism of fistula formation between a pseudocyst and the PV is still theoretical. Several reports indicated that activated pancreatic enzymes in the pseudocyst might cause inflammation, directly contributing to cyst wall fragility and failure [3, 9]. Another report indicated that the pressure in pseudocyst is higher than that in the PV and that hence activated pancreatic enzymes flow into the PV [12]. This might be the reason why none of the cases reported hemorrhage after the PP-PV fistula. In our case, the patient presented with acute pancreatitis and overlapping chronic pancreatitis symptoms, as observed in previous reports, and it is believed that fragility of the cyst and PV walls induced by activated pancreatic enzymes due to the pressure difference probably contributed to the fistula formation.

PV Thrombosis

Previous studies proposed two mechanisms of PV thrombus formation. It is hypothesized that activated pancreatic enzymes cause PV wall erosion, inducing venous thrombosis [3, 9]. Moreover, it has also been postulated that PV thrombosis may result due to mass effect and compression by the pseudocyst, along with associated peri-pseudocyst inflammation [13, 15]. We believe that these two mechanisms together led to thrombus formation in our case. In this patient, the PV thrombus was dissolved by the administration of heparin and increased blood flow into the cyst.

Treatment

Although PP-PV fistulas were managed in previously reported cases through conservative, surgical, or endoscopic therapy, the most effective treatment approach remains controversial. Conservative management can be effective in patients who are asymptomatic or have mild symptoms [10, 12]. Conversely, some cases required aggressive surgical treatment, including pancreatectomy, pseudocyst-enterostomy, or pancreaticoenterostomy [2, 5, 6, 8, 9, 13]. Interestingly, the endoscopic pancreatic duct stent insertion approach, which is less invasive than surgical treatment, improved the flow of pancreatic secretion into the pancreatic duct and blocked the flow into the pseudocyst. We adopted the treatment algorithm proposed by Ng et al. [13] for the management of the PP-PV fistula. First, we evaluated for PV patency. CT demonstrated that there was not PV patency. Subsequently, we assessed the relationship between the pseudocyst and the pancreatic duct and confirmed the fistula. Therefore, by following their decision-making flow chart for the treatment strategies, we considered the endoscopic approach as the optimal management strategy.

The clinical management of PP-PV fistulas remains controversial. Though treatment strategies vary with patients, we have described a case of successful treatment of a PP-PV fistula, which, together with previous reports, showed the endoscopic approach to be a less invasive and effective treatment method.

Statement of Ethics

The patient provided informed consent; the study design was exempt from ethics review board approval.

Conflict of Interest Statement

The authors declare no conflict of interest. There was no grant support.

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Author Contributions

A. Kimura and K. Hayashi wrote the manuscript. A. Kimura, K. Hayashi, C. Oda, K. Hosaka, N. Kimura, K. Tominaga, S. Ikarashi, A. Tsuchiya, and S. Terai diagnosed and treated the patient. All authors read and approved the final version of the manuscript.

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Fig. 1. a Abdominal CT demonstrating a pseudopancreatic cyst in the pancreatic head region (arrow). b Low-attenuated fluid thrombus within the portal vein (arrow). c Slightly high-attenuated fluid within the pseudocyst (arrow). CT, computed tomography.

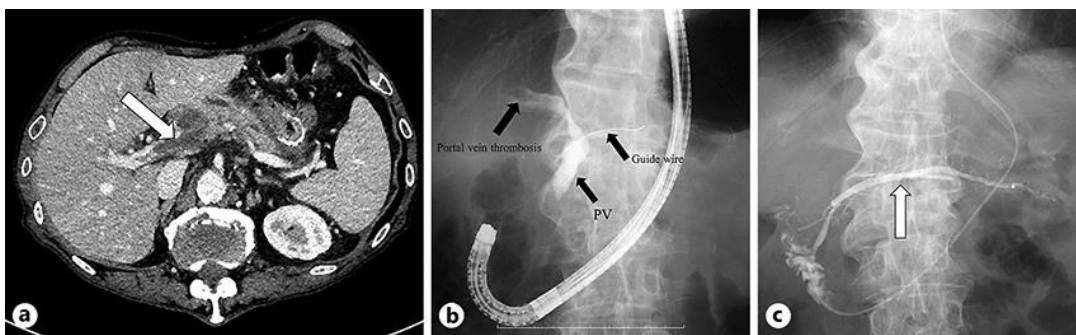


Fig. 2. a Contrast-enhanced CT showing a decrease in the size of the PV thrombus (arrow). b Endoscopic retrograde cholangiopancreatography demonstrating that the pseudocyst was connected with the PV by a fistula. c The PP-PV fistula was resolved, with the arrow showing the endoscopic nasopancreatic drainage tube. CT, computed tomography; PP, pancreatic pseudocyst; PV, portal vein.

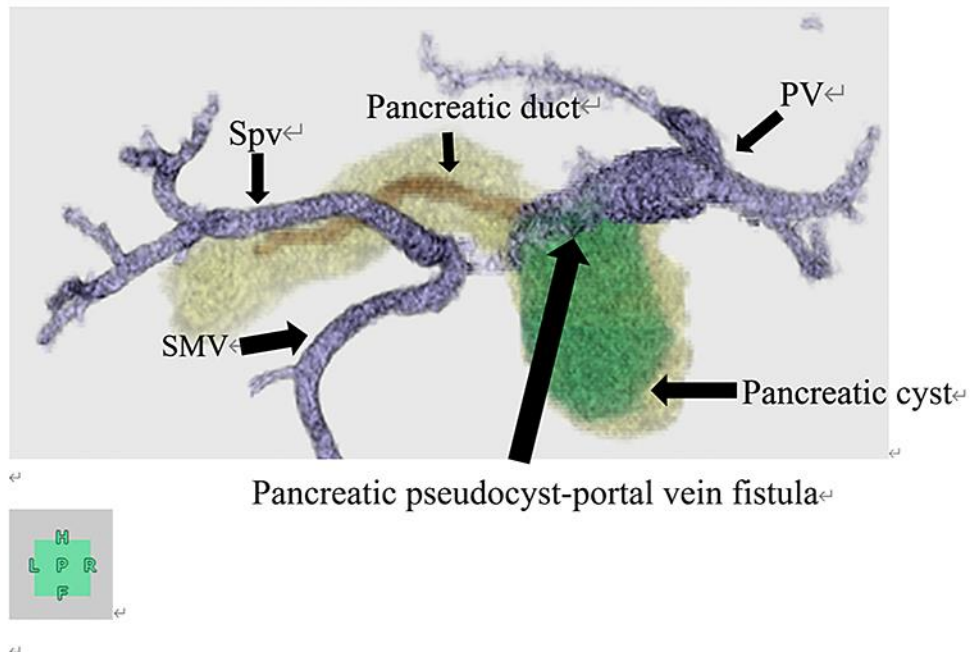


Fig. 3. Three-dimensional image of the PP-PV fistula. PP, pancreatic pseudocyst; PV, portal vein; SMV, superior mesenteric vein; Spv, splenic vein.

Table 1. Literature review of reported cases of PP-PV fistula

Ref. No.	Year	First author	Age, years	Sex	Diagnostic modalities	Treatment	PVT	Outcome
1	1966	Zeller	57	M	autopsy	conservative therapy, autopsy	no	deceased
2	1986	Pedrazzoli	30	M	surgery	surgery (pancreatico-cysto-jejunostomy)	yes	survived
3	1986	Takayama	52	M	percutaneous transhepatic portogram, CT	surgical drainage of ascites	yes	survived
3	1986	Takayama	82	M	percutaneous pancreatic ductogram, CT	conservative therapy	NR	deceased
4	1988	Sørensen	60	M	autopsy	conservative therapy, autopsy	no	deceased
5	1991	Potts	62	M	surgery	surgery (pancreaticoduodenectomy)	no	survived
6	1995	Procacci	36	F	CT, guided portography	surgery (pancreaticojejunostomy)	yes	survived
7	1995	Skarsgard	47	F	autopsy	conservative therapy, autopsy	yes	deceased
8	2002	Hammar	29	M	surgery	surgery (pancreaticojejunostomy)	no	survived
9	2008	Yoon	43	M	CT, MRCP, ERCP	surgery (pancreaticoenterostomy)	yes	survived
10	2010	Noh	54	M	CT, portography, ERCP	EPS	yes	survived
11	2012	Horino	57	M	US, CT, ERCP	EPS → US-guided drainage	yes	survived
12	2013	Raza	45	M	tubogram, angiography, ERCP	EPS → surgery (pancreaticoduodenectomy)	yes	survived
13	2015	Ng	58	F	CT, MRI	surgery (cyst-gastrostomy)	yes	deceased
14	2019	Eccles	64	M	CT	EUS-guided pancreatic cyst-gastrostomy	yes	survived
14	2019	Eccles	54	M	CT	EUS-guided pancreatic cyst-gastrostomy	yes	survived
This case	2020	Kimura	70	M	ERCP, CT	EPS	yes	survived

CT, computed tomography; EPS, endoscopic pancreatic stent; ERCP, endoscopic retrograde cholangiopancreatography; EUS, endoscopic ultrasound; MRCP, magnetic resonance cholangiopancreatography; MRI, magnetic resonance imaging; NR, not recorded; PP, pancreatic pseudocyst; PV, portal vein; PVT, portal vein thrombosis; US, ultrasound.