Recurrence of Pancreatic Pseudocyst following Endoscopic Ultrasonography-Guided Drainage

Ahmed SELIM[1,2], Nobuhiko FUKUBA[1], Ichiro MORIYAMA[3], Hiroki SONOYAMA[1], Chika FUKUYAMA[1], Shohei SUMI[1], Yoshiyuki MISHIMA[1], Shunji ISHIHARA[1], Yoshikazu KINOSHITA[1]

[1] *Department of Internal Medicine II, Shimane University Faculty of Medicine, Izumo, 693-8501, Japan*

[2] *Department of Gastroenterology and hepatology, Tanta University, Faculty of Medicine, Egypt*

[3] *Cancer Center, Shimane University Hospital, Izumo, 693-8501, Japan*

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A 71-year-old female came to our hospital with upper abdominal pain, fever, and jaundice, and was diagnosed with severe acute biliary pancreatitis. Upon admission, the patient underwent emergency endoscopic retrograde cholangiopancreatography and endoscopic sphincterotomy procedures, as well as endoscopic retrograde biliary drainage. Although pseudocyst formation was observed, no symptoms were presented and she was discharged. One month after, she was admitted again due to pain caused by pseudocyst enlargement and the symptoms persisted following conservative treatment. Although we performed endoscopic ultrasonography-guided drainage for the pancreatic pseudocyst, abdominal pain continued and abdominal computed tomography revealed that the pseudocyst was again enlarged. Since insufficient drainage seemed to be the cause, additionally balloon dilatation and placement of plastic stents for the fistulae were performed. There are no guidelines available for selection of drainage period or number of stents in pseudocyst cases, and additional studies regarding management of affected patients are needed.

Keywords: EUS, drainage, pancreatic pseudocyst, recurrence

Corresponding author: Nobuhiko Fukuba, MD, Ph.D
Department of Internal Medicine, Izumo City General Medical Center, 613 Nadabun-cho, Izumo, Shimane 691-0003, Japan
Tel: +81-853-63-5111
Fax: +81-853-63-4228
E-mail: fnat98@yahoo.co.jp

INTRODUCTION

According to the revised Atlanta criteria for inflammatory pancreatic fluid collection, a pancreatic pseudocyst is defined as an encapsulated collection of fluid with a well-defined inflammatory wall usually outside the pancreas, with minimal or no necrosis. Usually, more than 4 weeks after onset of interstitial edematous pancreatitis is required for this pseudocyst to mature [1].

The prevalence of pancreatic pseudocyst in acute pancreatitis cases has been reported to range from 6% to 18.5% [2, 3], while that in cases of chronic pancreatitis ranges from 20% to 40% [4]. A pancreatic pseudocyst most commonly arises in patients with alcoholic chronic pancreatitis (70% to 78%) [5], while the second most common cause is idiopathic chronic pancreatitis (6% to 16%), followed by biliary pancreatitis (6% to 8%). This type of pseudocyst may be asymptomatic or can present a variety of symptoms, such as abdominal pain, satiety, upper gastrointestinal bleeding, infection, nausea, and vomiting that necessitates drainage [6, 7]. Traditionally, a pancreatic pseudocyst is managed by open surgical internal drainage. However, with continued medical technology development, less invasive options including percutaneous, endoscopic, and laparoscopic drainage are now possible [8]. Hereby we described a case of pancreatic pseudocyst recurrence following endoscopic ultrasonography-guided drainage.

CASE STUDY

We present here a 71-year-old female who was

repeatedly admitted to our hospital over a period of 6 months. Her significant past medical history included dyslipidemia, hyperuricemia, and gall bladder stones. The patient denied alcohol intake. The initial hospital admission was due to an acute attack of severe biliary pancreatitis (Fig. 1A) that was managed by an emergency endoscopic retrograde cholangiopancreatography (ERCP) procedure with endoscopic sphincterotomy (EST) and endoscopic retrograde biliary drainage (ERBD) (Fig. 1B). She also received medical treatment for pancreatitis and pancreatitis parameters were improved. However, pseudocyst formation was observed (Fig. 1C).

One month after discharge, she was admitted again due to exacerbation of pancreatitis and pseudocyst enlargement (Fig. 1D). The symptoms were mild and then disappeared on the second day after admission. We decided to perform conservative treatment based on her general condition and findings from endoscopic ultrasonography (EUS) scanning of the pseudocyst, and the patient was discharged on hospital day 10. Unfortunately, 1 day after that second discharge, epigastric pain started, which was improved with acetaminophen, though she again visited our hospital because of its persistence. The initial clinical evaluation at that time showed that the patient was hemodynamically stable with a low grade fever. Computed tomography (CT) showed that the pseudocyst around the pancreatic head had slightly increased, and laboratory investigations revealed elevated WBC and CRP levels. We decided to use EUS-guided drainage for treatment of the pancreatic pseudocyst.

Initial EUS-guided pseudocyst drainage

Under routine procedural sedation with midazolam and pentazocine, as well as vital sign monitoring and a continuous nasal oxygen supply, a curvilinear array echoscope (GF TYPE UCT260, EU ME-1, OLYMPUS) was passed to the stomach, and echo scanning of the pseudocyst through both the gastric and duodenal walls was carefully performed.

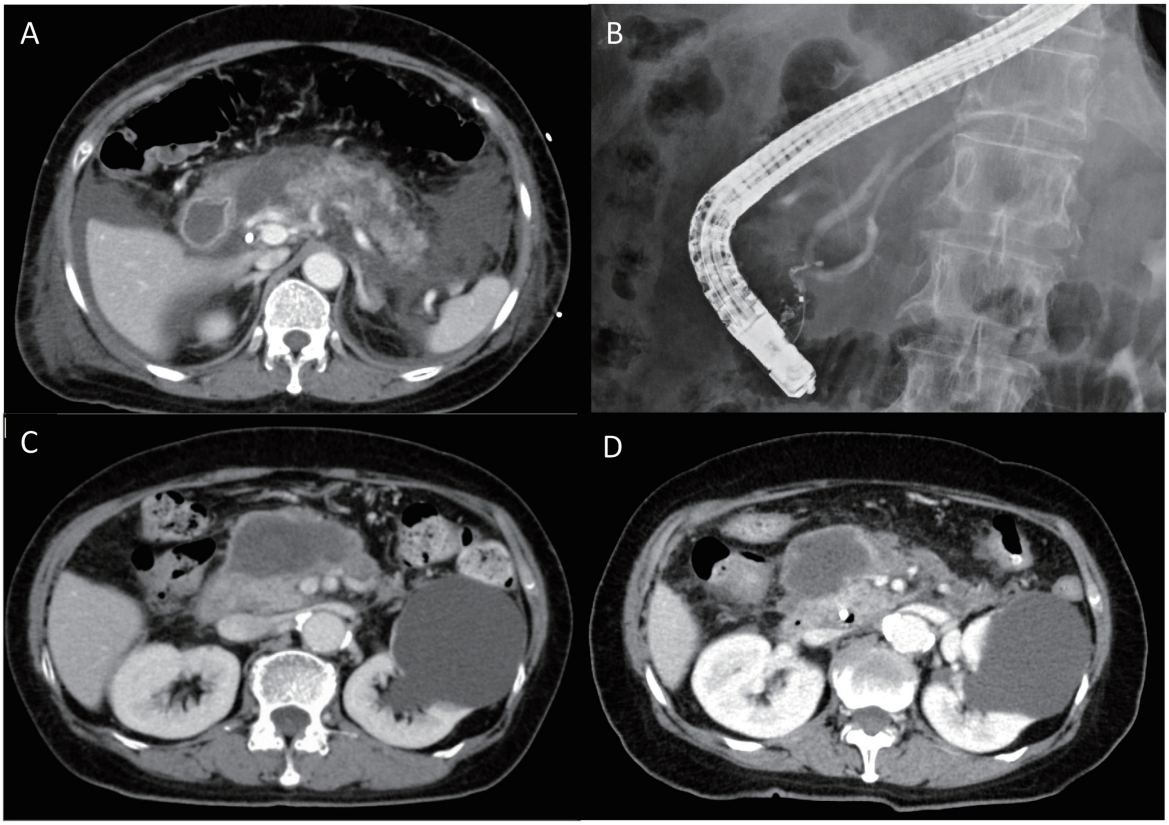

Fig. 1. The initial hospital admission was due to an acute attack of severe biliary pancreatitis (A) that was managed by ERCP with EST (B). (C) shows CT imege of edematous pancreas with fluid collection following the first admission. Following the final admission, a pancreatic pseudocyst enlarged (D).

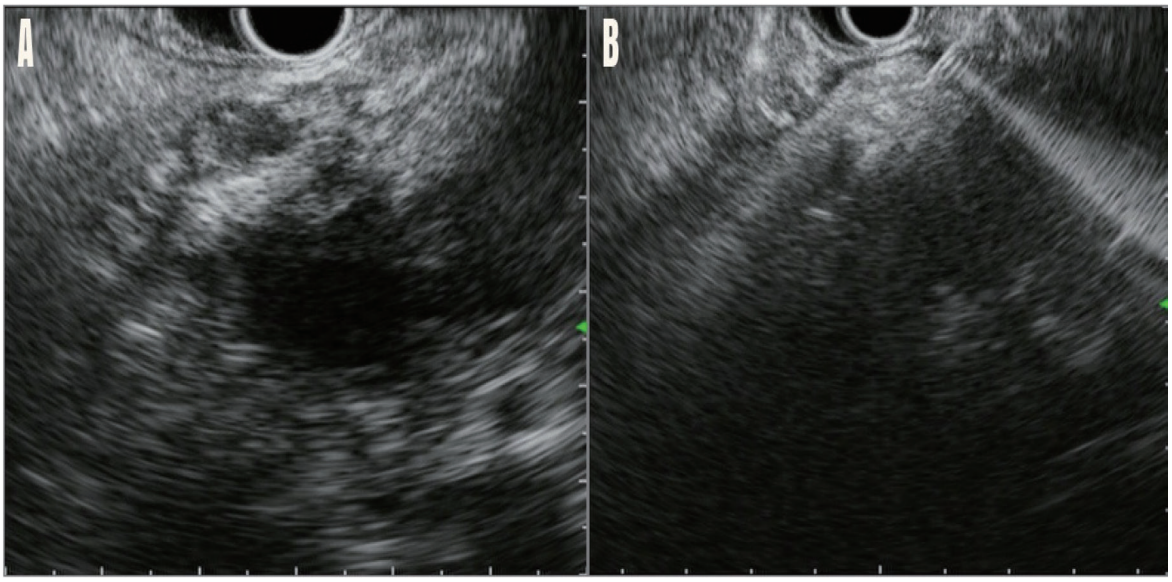

Fig. 2. EUS scanning and pseudocyst puncture.

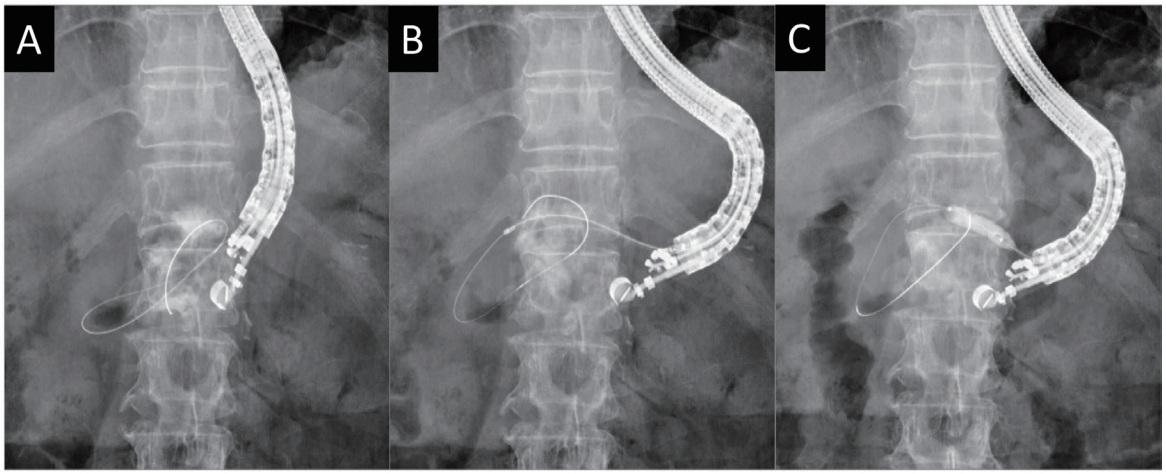

Fig. 3. Fluoroscopic images of EUS-guided（A）guidewire placement,（B）biliary ballon dilation, and（C）placement of dilatation catheter.

We chose the gastric position for drainage because of the straight position of the scope, and small gap between the pseudocyst and gastric wall. Following application of color Doppler and determination of the puncture site, a 19-G needle（Echo-Tip, ULTRA, Wilson-Cook Medical）was introduced through the working channel of the endoscope and used to puncture the cyst wall under EUS guidance（Fig. 2）. Next, the stylet was removed and replaced by a guidewire（VisiGlide 2, 0.025” OLYMPUS）, which was coiled inside the cyst cavity under cystography with fluoroscopic guidance（Fig. 3A）. We then slowly removed the needle, leaving the wire inside the cystography device. Dilation of the fistula was done in 3 steps. First, we performed diathermic dilation using a Cysto-Gastro-Set（ENDO-FLEX, DEU）passed above the already placed wire based on observations of endoscopic and fluoroscopic images（Fig. 3B）. The second step was mechanical dilation by use of a biliary balloon dilator（REN 8 Fr, KANEKA, JPN）based on observations of fluoroscopic images（Fig. 3C）. Finally, mechanical dilation was performed using a Soehendra Dilation Catheter（6-9 fr, Cook Medical, NC）. During this third step, an additional guidewire（the Soehendra dilator can accommodate 2 guidewires）was inserted

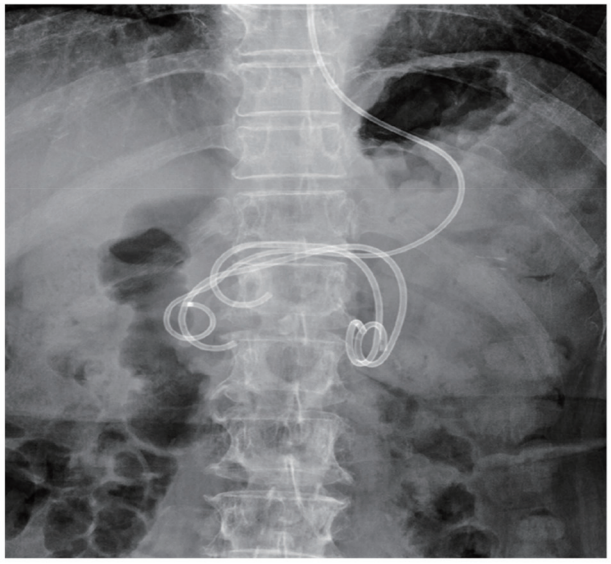

Fig. 4. Two double pigtail stents and nasocystic tube placed inside the pancreatic pseudocyst.

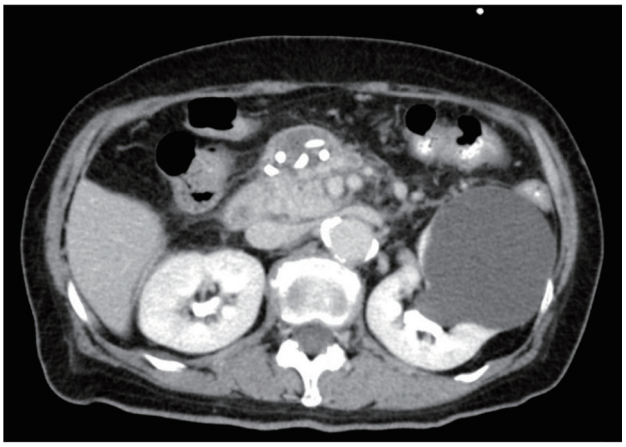

Fig. 5. Enhanced CT after EUS-guided drainage, showing that the pseudocyst size has been reduced.

Fig. 6. Enhanced abdominal CT showing recurrence of pseudocyst with 2 stents inside.

into the cyst cavity under fluoroscopy guidance, which was repeated to finally have 3 guidewires placed inside the cyst. Thereafter, we placed 2 double pigtail plastic stents (Through Pass 7 Fr, 7 & 10 cm, GADELIUS, JPN) and a nasocystic tube using a Nasal Biliary Drainage Set (NB-BRAID 6 Fr, PIOLAX, JPN) (Fig. 4). The procedure was finished uneventfully and antibiotic administration was prescribed. Four days after the procedure, enhanced abdominal CT showed that the pancreatic pseudocyst was reduced in size.

One week after drainage, cystography was performed with injection of a contrast agent through the nasocystic tube, which showed that the cyst cavity was shrunken. The pseudocyst content obtained by the nasocystic tube was small and submitted for a bacteriological examination, which revealed neutrophilia and candida. Furthermore cytological examination had no findings of malignancy.

Second pseudocyst drainage

After confirmation of pseudocyst shrinkage in abdominal CT results and abdominal pain improvement (Fig. 5), we removed the nasocystic tube 10 days after first drainage and left the 2 double pigtail stents in place. However, on the planned day of discharge 6 day after removal of nasobiliary drainage, she suffered again from abdominal pain. Abdominal CT was performed, which showed that the pseudocyst had enlarged again with the 2 stents in place (Fig. 6). Following diagnosis of pseudocyst recurrence, we decided to perform another drainage procedure. An upper endoscope (GIF-260J, OLYMPUS, JPN) was passed to the stomach (Fig. 7A), the 1 of the double pigtail stents was removed by a snare, followed by insertion of a guidewire inside the pseudocyst cavity with widening of the fistula again by biliary balloon dilation (REN 10mm, KANEKA, JPN) (Fig.7B). We then changed the ultra-slim scope to a nasal type (GIF-XP260, OLYMPUS, JPN) and carefully passed over the wire inside the pseudocyst cavity under fluoroscopic guidance (Fig. 7C). Irrigation was performed with 500 ml of normal saline, then we switched back to an upper endoscopy with great care so as to not lose the wire position (Fig. 8). Then, additionally 2 double pigtail stents (Through Pass 7 Fr, 7 and 10 cm, GADE-

LIUS, JPN) and a nasocystic tube (NB-BRAID 6 Fr, PIOLAX, JPN) were placed. As a result, the patient had placed a total of 3 double pigtail stents and a nasocystic tube (Fig. 9). The pseudocyst condition improved following this additional therapy. She discharged 7 days after the final treatment.

DISCUSSION

A pseudopancreatic cyst is a known complication of both acute and chronic pancreatitis, and shows a wide variety of clinical presentations, ranging from completely asymptomatic lesions to multiple pseudocysts with pancreatic and/or bile duct obstruction, which may require immediate intervention to prevent secondary complications. Other indications for drainage include infection, hemorrhage, and presence of symptoms [9]. The probability of spontaneous resolution of a pancreatic pseudocyst ranges widely from 8% to 85%, depending on etiology, localization, and predominantly size [10]. Warshaw and Rattner reported that a pseudocyst is unlikely to resolve spontaneously if: a) it persists for more than 6 weeks, b) chronic pancreatitis is evident, c) there is a pancreatic duct anomaly (except for communication with the pseudocyst), or d) the pseudocyst is surrounded by a thick wall [11]. The pseudocyst in our patient was a sequel of biliary pancreatitis, relatively large

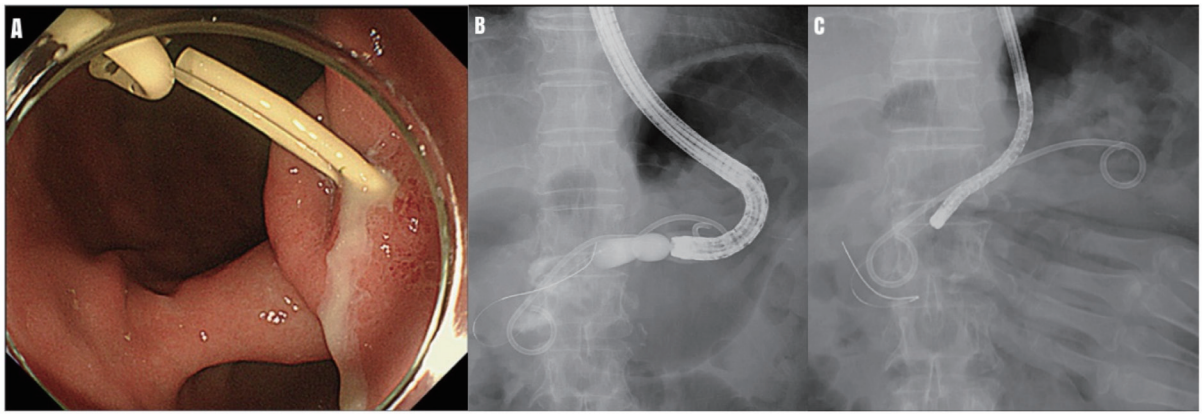

Fig. 7. Images obtained during endoscopic-guided drainage. (A) Narrow fistula opening. (B) Dilation with biliary ballon. (C) Nasal-type scope above the wire inside the cyst cavity.

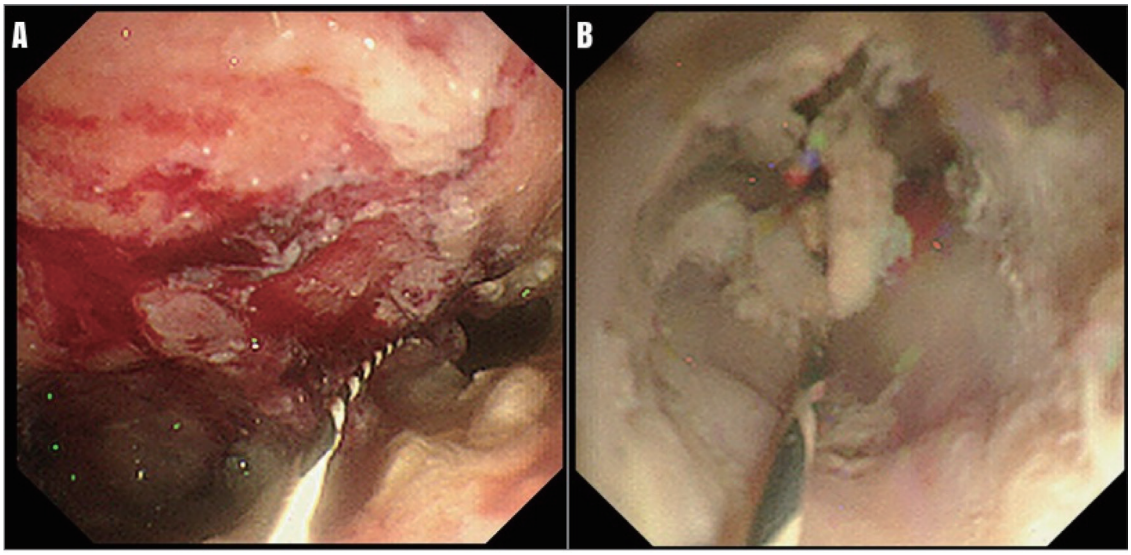

Fig. 8. (A) Image of pseudocyst lumen by ultra-slim scope, (B) Image of irrigation for pseudocystic lumen with saline by ultra-slim scope

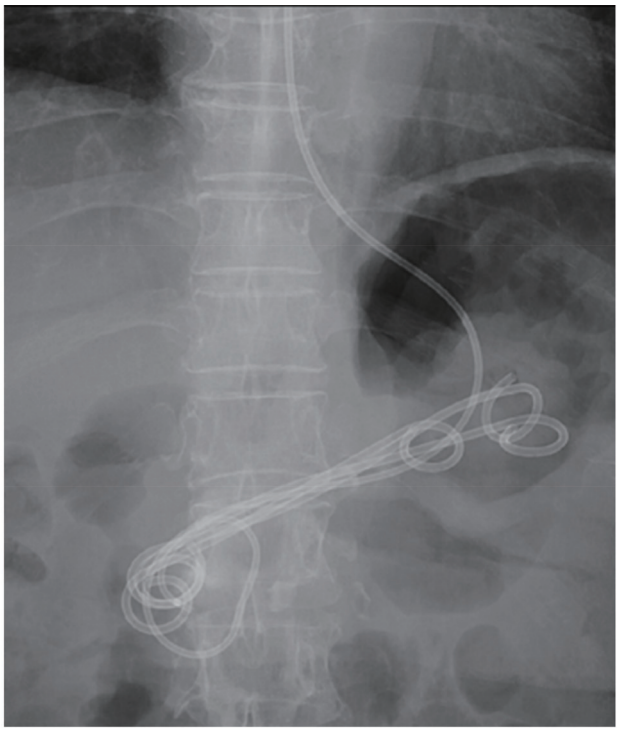

Fig. 9. Three double pigtail stents and nasocystic tube inside the pseudocyst.

in size and had persisted more than 6 weeks. Thus, spontaneous resolution was considered to be rather difficult. In general, the pseudocyst was, initially, asymptomatic, then later the patient complained of abdominal pain. Although conservative treatment reduced the symptoms, evidence of pseudocyst infection was an important clue to attempt drainage.

Drainage options include percutaneous, endoscopic transpapillary, endoscopic transmural (with/without EUS guidance), and surgical drainage, the latter by either open or laparoscopic surgery [12, 13]. However, there are no widely accepted guidelines or international consensus for dealing with a pseudocyst. As such, a classification system based on individual cyst characteristics would offer physicians some guidance for therapeutic decision making [14, 15]. The relationship between a pancreatic pseudocyst and the pancreatic duct must be evaluated before treatment. If communication exists, endoscopic drainage via a transpapillary approach can be achieved [16]. If there is a less than 1-cm gap between the pseudocyst and gastrointestinal wall, endoscopic drainage via cystogastrostomy or cystoduodenostomy should be performed [17, 18]. If neither of those conditions exist, patients should be treated with surgery for internal drainage. In our strategy for pancreatic pseudocyst, we will prioritize trans-papillary drainage for patients that were evidenced to have both a stenosis of main pancreatic duct and a communication between pancreatic pseudocyst and pancreatic duct. On the other hand, we will select EUS-guided trans gastric approach to pancreatic pseudocyst without both or one. In the present patient, the pseudocyst was related to the body and tail of pancreas, with a gap of less than 1 cm between it and the gastric wall. However, we could not evaluate the main pancreatic duct even by MRCP and it was unclear whether the pseudocyst communicated to pancreatic duct. Therefore we performed EUS-guided trans gastric drainage.

A variety of stents have been used to maintain patency of the fistulous tract between the gut lumen and pseudocyst cavity, including single plastic stents (straight or double pigtail), multiple plastic stents, nasocystic drainage catheters, enteral metal stents, and biliary metal stents [19-21]. Until now, there has been no clear evidence to suggest that expandable metallic stents are superior to plastic stents or whether different plastic stents offer a technical advantage [22-24]. In this present case, a plurality of plastic stents was placed as internal fistula. Plastic stents are easy to be exchanged compared with expandable metallic stents, and a plurality of plastic stents is effective for forming gaps and sustaining the drainage effect. Furthermore, in order to monitor the properties of cystic drainage and to follow up cystography, an additional nasocystic drainage tube for temporally external fistula was placed. For these reasons, we performed placement of 2 double pigtail plastic stents and a nasocystic tube in the present case.

Despite confirmation of pseudocyst shrinkage by CT scanning and cystography, recurrence occurred within about 1 week after removal of the nasocystic tube. One of the reasons for relapse after a short period may have been related to our finding that the infectious fluid in the pseudocyst was quite sticky. When the viscosity of the liquid in the cyst is high, the lumen and the gap of stents are likely to clog and the drainage tends to be inadequate. That was solved by dilating the fistula tract and increasing the number of pigtail stents from 2 to 3. In cases with

high viscosity cystic fluid, a larger fistula expansion and placement of more stents might be required as compared to cases with cystic fluid of low viscosity.

In a systemic review of current guidelines for minimally invasive management of pancreatic pseudocyst cases published in 2008 [25], the American Gastroenterological Association stated that an infected pseudocyst should be termed an abscess and can be treated with percutaneous drainage [26]. Also, guidelines presented by the Society for Surgery of the Alimentary Tract (SSAT) [27] and Sociétié Nationale Francais de Gastro-Entérologie (SNFG) [28] recommend that infected pancreatic fluid be drained in a percutaneous manner, though they do not differentiate between a pancreatic abscess and infected pseudocyst. Japanese guidelines for management of acute pancreatitis presented in 2015 state that during therapeutic intervention for infected pancreatic necrosis, percutaneous (retroperitoneal) drainage or endoscopic transluminal drainage should be given first, then if no improvement is achieved, a necrosectomy should be performed. In addition to a necrosectomy, an endoscopic or retroperitoneal approach is recommended. However, there is no mention regarding selection of the period of drainage or number of stents in the current guidelines. Each guideline recommends drainage for infectious pancreatic pseudocysts. In recent years endoscopic drainage has been developed and becoming the first choice treatment for it. However, there are no evidences regarding selection of the period of drainage or number of stents. Until evidence accumulates, we think that it is necessary to have as many stents as possible and a long term stent placement period. Based on our experience, we consider that a key factor for success of endoscopic-guided drainage of a pseudocyst may be the patency of the draining tubes and fistula tract. Further investigations for management of patients with a pseudocyst are needed.

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