A Case of Mediastinal Pancreatic Pseudocyst Successfully Treated with Somatostatin Analogue

HIDEYA SUGA*, OSAMU TSURUTA*, YOSHINOBU OKABE*, FUMIHIKO SAITOH*,
TETSUROU NODA*, HIKARU YOSHIDA*, NAOFUMI ONO^{††},
HISAFUMI KINOSHITA**, ATSUSHI TOYONAGA^{†,*}
AND MICHIO SATA*

Departments of Medicine*, Surgery**, and Gastroenterology and Endoscopy[†], Kurume University School of Medicine, Kurume 830-0011 and ^{††}Eguchi Hospital, Saga 845-0032, Japan

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Summary: A 57-year-old man with a 3-year history of chronic pancreatitis was admitted to our hospital with upper abdominal pain. Based on examination findings, the patient was diagnosed as having pseudocysts in the pancreatic body and the mediastinum that were associated with acute aggravation of chronic pancreatitis. Because of the patient refused an operation, he was submitted to conservative management including intramuscular injection with somatostatin analogue of 100 μ g/day. On the 14th day of the treatment, pleural effusion and pseudocyst in the pancreatic head were additionally diagnosed based on the findings of computed tomography, magnetic resonance imaging and other examinations, and the dose of somatostatin analogue was increased to 200 μ g/day. As a result, on the 28th day of the treatment, pancreatitis was inactivated, and the pseudocysts in the mediastinum and the pancreas disappeared. The patient has been followed up for 15 months, and there has been no recurrence.

Key words mediastinal pancreatic pseudocyst, somatostatin analogue, chronic pancreatitis

INTRODUCTION

Mediastinal pancreatic pseudocyst as a complication of pancreatitis is a rare disease. To date, only 56 patients had been reported. Most cases were treated with drainage or surgical resection of pseudocysts. This is a report of a patient, who had chronic pancreatitis associated with mediastinal pancreatic pseudocyst, achieved remarkable improvement by somatostatin analogue administration.

CASE REPORT

A 57-year-old man with chronic pancreatitis visited a local clinic for upper abdominal pain. The patient had consumed alcohol approximately 120 g

per day for the past 30 years, diagnosed as having 3 year history of chronic pancreatitis and had been clinically followed-up on the out patient basis. At the visit, serum amylase level increased, and abdominal ultrasonography revealed cystic lesion in the body of the pancreas. Acute aggravation of chronic pancreatitis was suspected and conservative treatment was initiated. The symptoms were not improved and the patient was referred to our hospital for a further medical work-up.

At the admission, the patient's abdomen was flat and soft, but there was tenderness from the epigastrium to the right hypochondrium lesion. There were no sign of peritoneal irritation. Hematological examination revealed increased white blood cell count and biochemical examination showed decreased serum

Correspondence: Hideya Suga, MD, Second Department of Internal Medicine, Kurume University School of Medicine, 67 Asahi-machi, Kurume 830-0011, Japan. Tel: +81-942-31-7561 Fax: +81-942-34-9413 E-mail: suga516@med.kurume-u.ac.jp

Abbreviations: CT, computed tomography; ERP, endoscopic retrograde pancreatography; MRCP, magnetic resonance cholangiopancreatography.

albumin, and increased C-reactive protein 4.71 mg/dl, pancreas type amylase 643 U/L, and lipase 409 U/L. Computed tomography (CT) images obtained by us depicted a mediastinal cystic lesion in the longitudinal area between the esophageal hiatus and the posterior mediastinum, right pleural effusion (Fig. 1a), and a cystic lesion in the body of pancreas (Fig. 1b). The pancreas was reduced in size with calcification.

The patient was diagnosed as having acute aggravation of chronic pancreatitis associated with pseudocyst in the mediastinum and in the pancreatic body. Patient refused to have surgical treatments, and chose to have conservative management. Oral food intake was stopped and central venous nutrition with

Fig. 1. Abdominal CT at hospital admission. a. There was a long vertical cystic lesion in the mediastinum from around the esophageal hiatus to the posterior mediastinum (arrow head). Small amount of pleural effusion was detected in the right thoracic cavity. b. A multilocular cystic lesion (arrow) with 50 mm in diameter was found in the body of pancreas. The pancreas was contracted and had calcification.

Fig. 2. Abdominal CT at chest pain episode. a and b. Enlargement of the mediastinal pancreatic pseudocyst and large amount of pleural effusion in the right thoracic cavity were depicted (arrow head). c. The pseudocyst in the pancreatic body was on a mild reduction.

the administration of gabexate mesilate, famotidine, sulbactam, and cefoperazone was started. Subjective symptoms improved but serum amylase increased and co-administration of ulinastatin was started. Since then, serum amylase became close to normal level, but exertional dyspnea and upper back pain appeared. At that time, serum pancreatic enzyme levels increased again. Abdominal CT depicted the growth of mediastinal pancreatic pseudocyst, large amount of right pleural effusion (Fig. 2a, b), shrinkage of pseudocyst in the pancreatic body (Fig. 2c), and new development of pseudocyst in the pancreatic head. Magnetic resonance cholangiopancreatography (MRCP) revealed the presence of several cysts in the pancreatic head and body and a longitudinal cystic lesion reaching to the pseudocyst in the pancreatic body. This longitudinal lesion was diagnosed as mediastinal pancreatic pseudocyst. However, MRCP images did not demonstrate the communication between the cystic lesions (Fig. 3). Sample of the pleural effusion was light yellow exudative fluid, and enzyme contents were at quite high levels pancreas type amylase 5,944 U/L and lipase 5,250 U/L. The patient was diagnosed as having pancreatic pleural effusion and mediastinal pancreatic pseudocyst, but he refused to have thoracostomy drainage and requested conservative management. The patient was started on a somatostatin analogue, octreotide 100 μ g/day, and then subjective symptoms together with the pancreatic enzyme levels improved. On the 14th day of octreotide treatment, the improvement of pleural effusion was

Fig. 3. Magnetic resonance cholangiopancreatography (MRCP). There were several cystic lesions in the head and body of the pancreas and a long cystic lesion extended longitudinally and reaching to near the pancreatic body (arrow head). Communication between the cysts was not depicted.

Fig. 4. Abdominal CT on the 28th day of treatment or on the 14th day of octreotide 200 $\mu\text{g}/\text{day}$ administration. a. Mediastinal pancreatic pseudocyst and pleural effusion in the right thoracic cavity disappeared. b. Pseudocysts in the body and head of the pancreas disappeared.

imaged by abdominal CT, but the pseudocysts in the mediastinum and the pancreatic body became larger. Endoscopic retrograde pancreatography (ERP) was performed as a means by trans papillary drainage. However, as the ERP images depicted irregular dilatation of pancreatic duct-branches and numerous pancreatic calculi in the main pancreatic duct, pancreatitis was suspected to be aggravated if contrast medium was injected and insertion of guiding wire was thought to be difficult because of the calculi. Trans-papillary cyst drainage was canceled, and the conservative treatment was continued by increasing the daily dose of octreotide to 200 $\mu\text{g}/\text{day}$. The patient's pancreatic enzyme levels were normalized, and on the abdominal CT taken 28th day of the treatment the pseudocysts in the mediastinum and the body of pancreas disappeared (Fig. 4). Food intake started on 30th day of the treatment but pancreatitis did not recur. Dosage of octreotide was gradually decreased every week. 54th day of the treatment, the patient discharged from the hospital, and the octreotide discontinued. The patient has been followed up for 15 months after the discontinuation of octreotide, but there have been no recurrence of pancreatitis, pancreatic pseudocysts and pleural effusion.

DISCUSSION

Mediastinal pancreatic pseudocyst is a rare complication of pancreatitis, and there have been only 56 reported cases including ours since the first case of Edlin in 1951 [1-9]. Most of the 56 patients had alco-

hol drinking habit, their major basic disease was alcoholic chronic pancreatitis, and many had pleural effusion. Major clinical symptoms were dyspnea, epigastric pain, chest pain, swallowing difficulty and upper back pain [1-10]. Our patient also developed upper back pain and dyspnea during the conservative management and they were thought to be attributable to pleural effusion and mediastinal pancreatic pseudocyst.

Cameron [11] described the development process of mediastinal pancreatic pseudocyst as the following: pancreatitis causes rupture of pancreatic duct on the posterior side, pancreatic juice exudes into the retroperitoneum and then goes into the mediastinum through the esophageal hiatus and aortic hiatus, and then pseudocyst is formed. This could explain the higher incidence of mediastinal pancreatic pseudocyst in the posterior mediastinum.

The pseudocyst of our patient would be also formed by the exudation of pancreatic juice through the esophageal hiatus and reaching to the posterior mediastinum.

Mediastinal pancreatic pseudocyst is relatively easily diagnosed with imagings such as CT and MRI [12]. Definite diagnosis is made when ERP using contrast medium depicts communication between mediastinal pseudocyst and internal fistula. In our patient, however, the communication was not proven with contrast medium because ERP was discontinued by considering the risk of aggravation of pancreatitis due to contrast medium and the difficulty of inserting a guiding wire due to the numerous pancreatic calculi. The communication was neither depicted in MRCP. For the definite diagnosis on such patients as ours, advancement of diagnostic imaging techniques such as multidetector-row CT that allows non-invasive monitoring of the communication is awaited. We consider that there was a communication between the pancreatic duct and mediastinal pseudocyst in our patient because suppression of mediastinal pancreatic exocrine function with somatostatin analogue resulted in the reduction of pancreatic pseudocyst.

Mediastinal pancreatic pseudocyst is usually treated with surgical resection. Rose et al. [10] proposed that patients with good overall conditions are at first managed with conservative treatment, because the cyst of 5 cm or smaller in diameter or immature cyst that are associated with acute pancreatitis often disappear spontaneously or react well to conservative treatment; and for the cyst of 5 cm or larger in diameter that are associated with chronic pancreatitis, the

timing of surgical treatment should be carefully determined because the risk of complications is high. In our patient, a possible cause of the pseudocysts in the pancreatic body and the mediastinum was the distressed secretion of pancreatic juice due to pancreatic calculi in the main pancreatic duct. This was the indication of surgical treatment, however somatostatin analogue treatment was prescribed based on the patient's request for conservative management.

Somatostatin is a peptide hormone and its hormonal activities include inhibition of the release of insulin, glucagon, gastrin, TSH, ACTH, secretin, pancreaticozym, cholecystokinin, pepsin, and renin. Octreotide is a somatostatin analog that possesses the same pharmacological properties as somatostatin, and recent studies reported clinical efficacy of somatostatin analogues in treatment for pancreatic pseudocyst that is refractory to conservative therapies [13-15]. We also administered octreotide to our patient and obtained significant reduction of the cyst and improvement of pancreatitis. Somatostatin analogue has been administered to 6 cases of mediastinal pancreatic pseudocyst [6-9], and 3 of them [7-9] and our patient had good clinical results with conservative therapy alone. However, none of them have been followed up for a long period and this point should be examined and reported in future studies. In addition, no consensus has been reached in regards to the dosage of somatostatin analogues, therefore cases must be accumulated for further discussion.

Chronic pancreatitis with mediastinal pancreatic pseudocyst is refractory to conservative therapies. As noted above, administration of a high dose somatostatin analogue for a long period would be a therapeutic option for these patient, and dosage and treatment method should be investigated and discussed in future studies.

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