J Gastroenterol 1998; 33:289–294

Journal of Gastroenterology

Case report

Segmental groove pancreatitis accompanied by protein plugs in Santorini's duct

Ryushi Shudo, Takeshi Obara, Satoshi Tanno, Tsuneshi Fujii, Noriyuki Nishino, Miho Sagawa, Hitoshi Ura, and Yutaka Kohgo

Third Department of Internal Medicine, Asahikawa Medical College, 4-5-3-11 Nishikagura, Asahikawa, Hokkaido 078, Japan

Abstract: "Groove pancreatitis", a form of segmental pancreatitis affecting the head of the pancreas, is localized within the "groove" between pancreas head, duodenum, and common bile duct. Differentiation between groove pancreatitis and pancreatic head carcinoma is often difficult. We report a case of groove pancreatitis in which a hypoechoic mass between the duodenal wall and pancreas was clearly imaged, and narrowing of the second portion of the duodenum and bile duct stenosis were also found. The diagnosis was confirmed by surgery (pylorus-preserving pancreato duodenectomy). The patient was relieved from abdominal pain post operation. Up to the present, the patient has been good condition. We review the clinicopathologic and radiologic features of groove pancreatitis in the Japanese literature and discuss the possible role of Santorini's duct in its pathogenesis. We consider that impacted protein plugs in Santorini's duct are a pathogenic factor in the development of groove pancreatitis. Therefore, the findings of Santorini's duct on endoscopic retrograde pancreatography are very important in the diagnosis of groove pancreatitis. Groove pancreatitis presents various clinical features, such as biliary stenosis, duodenal stenosis, and pancreatic mass, and often masquerades as pancreatic head carcinoma. This condition should be kept in mind in the differential diagnosis of pancreatic head carcinoma.

Key words: groove pancreatitis, Brunner's gland, protein plug, Santorini's duct

Introduction

The term "groove pancreatitis" is employed to describe a specific form of pancreatitis that results in scarring which extends, in particular, into the "groove" between pancreas head, duodenum, and common bile duct. Since this entity was first described by Becker[1] in 1973, there have been only a few case reports, probably because of a lack of awareness.[2] The largest series was described by Stolte et al.[3] in 1982, who reviewed in detail the histopathologic features of 30 patients with groove pancreatitis found in a series of 123 patients who had undergone pancreatoduodenectomy for chronic pancreatitis.

The particular feature of groove pancreatitis is its topographical anatomy. In the management of this form of pancreatitis, it is important to differentiate it from pancreatic carcinoma. The groove represents a theoretical gap formed in the "sliding plane" interfacing between duodenum and pancreas. It serves as a "bed" for the vessels and the lymph nodes. Apart from obliterating the plane of relative movement by fusion, the interstitial scarring also leads to compression of vessels, lymphatics, common bile duct, and duodenum. The X-ray image findings of duodenal wall first observed are duodenal wall rigidity with loss of motility, with subsequent progression to unilateral or concentric narrowing of the lumen, more pronounced plication of the wall, and a flattening of the C-loop.

The differentiation of groove pancreatitis from pancreatic carcinoma is often difficult and may be impossible in some patients.[4,5] We report a case of groove pancreatitis and review this disease entity. A possible role of Santorini's duct in the pathogenesis of groove pancreatitis is also discussed.

Case report

A 53-year old man was admitted to our hospital with upper abdominal pain. He had consumed about 110 g ethanol per day for 30 years. Serum pancreatic and hepatic enzymes were slightly elevated: amylase,

Offprint requests to: R. Shudo
(Received Apr. 17, 1997; accepted Sept. 26, 1997)

486 IU/l (normal range; 30–160 IU/l); GOT, 46 IU/l (normal range; 8–40 IU/l); GPT, 88 IU/l (normal range; 5–40 IU/l); and ALP, 508 IU/l (normal range; 80–260 IU/l). Tumor markers were within the normal range: carcinoembryonic antigen, 3.2 ng/ml (normal; <5.0 ng/ml); carbohydrate antigen 19-9, 13 U/ml (normal; 37 U/ml). Transcutaneous abdominal ultrasonography (US) showed a hypoechoic mass measuring 40 mm in diameter, including small cystic lesions in the pancreas head. Endoscopy revealed irregular polypoid bulging adjacent to the minor papilla with narrowing of the duodenal lumen. Endoscopic biopsy showed only inflammation of the duodenal wall without evidence of malignancy. Hypotonic duodenography demonstrated marked stenosis of the descending part of the duodenum (Fig. 1). Computed tomography (CT) revealed thickening of the duodenal wall, a cystic lesion between pancreas head and duodenum, and swelling of pancreas head, with a low-density area that was nonhomogenously enhanced, suggesting groove pancreatitis (Fig. 2). Endoscopic retrograde cholangiopancreatography (ERCP) showed smooth stenosis of the intrapancreatic segment of the common bile duct. Cannulation of the pancreatic duct revealed stenosis of Wirsung's duct in the head, without irregularity, and Santorini's duct was not visualized (Fig. 3). Histological examination of specimens obtained by endoscopic pancreatic biopsy, did not show any evidence of malignancy. Endoscopic

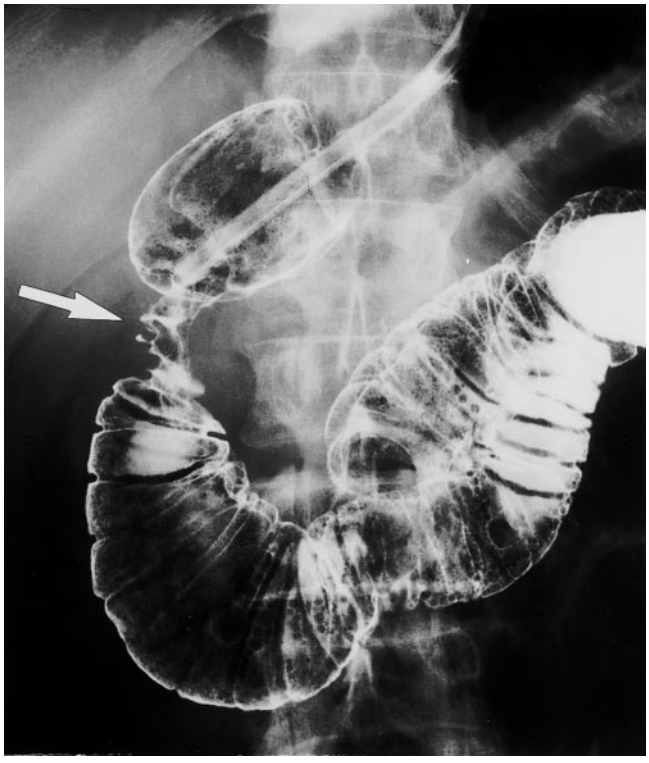
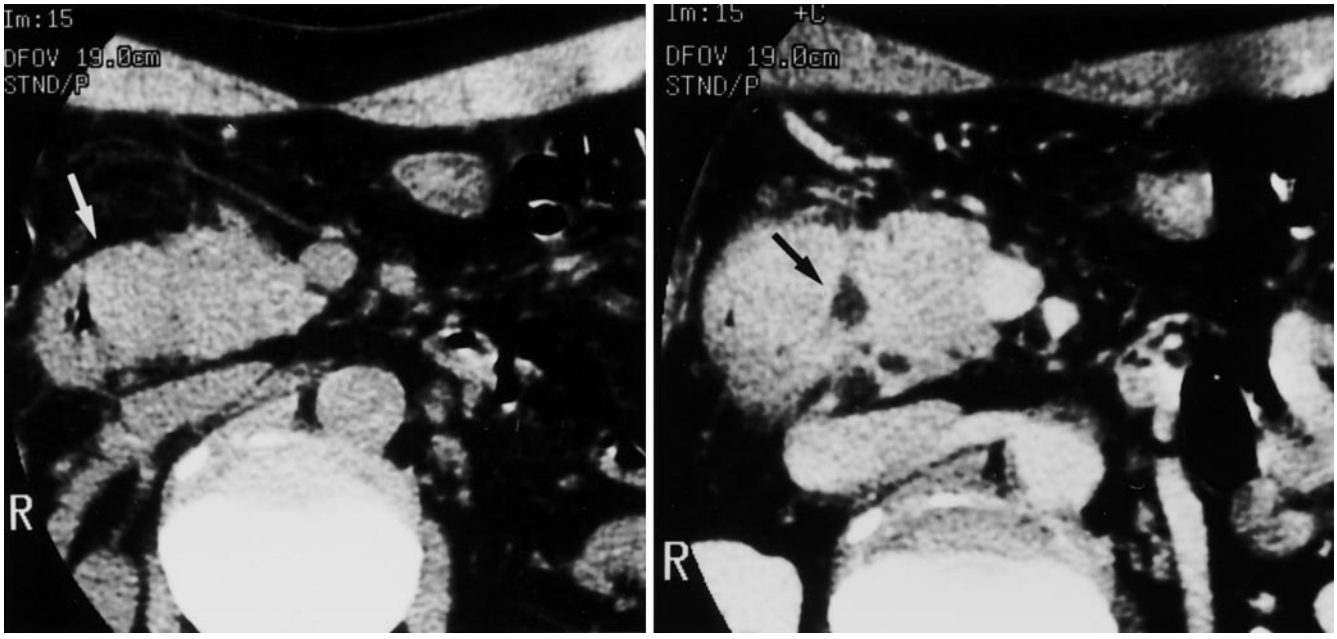


Fig. 1. Hypotonic duodenography demonstrating marked stenosis (*arrow*) of the descending part of the duodenum



a b

Fig. 2. a Abdominal computed tomography (CT) scan showing thickening (*arrow*) of the duodenal wall. **b** On high-dose enhanced CT scan, the head of the pancreas shows nonhomogeneous enhancement and a cystic lesion (*arrow*) between the head of the pancreas and the duodenum

ultrasonography (EUS) revealed a hypoechoic mass situated mainly between pancreas head and duodenum, with a penetrating duct sign, which suggested the tumor was caused by chronic pancreatitis rather than by pancreatic carcinoma (Fig. 4). Celiac angiography showed no encasement or occlusion. These abnormalities appeared consistent with the diagnosis of groove pancreatitis.

Consequently, we treated the patient conservatively, by fasting and intravenous hyperalimentation. After 7 weeks of this conservative treatment we evaluated its effects in terms of clinical signs, laboratory data, and imaging findings. Laboratory data e.g., amylase, 157 IU/l; GOT, 20 IU/l; GPT, 23 IU/l; ALP, 257 IU/l were almost normal. However, the abdominal pain was still present and as soon as he ate a meal, he had symptoms of pancreatitis. Imaging findings did not improve during follow-up. We considered that the condition would not improve with further conservative treatment, and a pylorus-preserving pancreatoduodenectomy (PpPD) was performed.

The cut surface of the resected specimen revealed a hard white tumor in the pancreatic head, and the high-

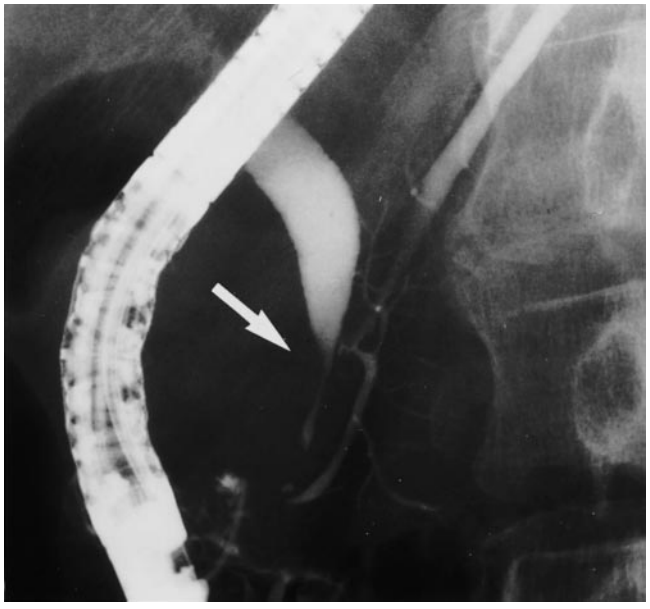


Fig. 3. Endoscopic retrograde cholangiopancreatography showing smooth stenosis of the intrapancreatic segment of the common bile duct and stenosis of the main pancreatic duct in the head, without irregularity. Note that Santorini's duct is not visualized (*arrow*)

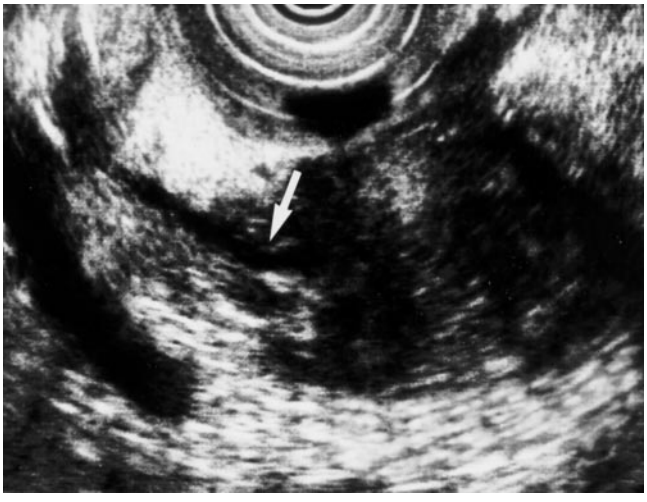
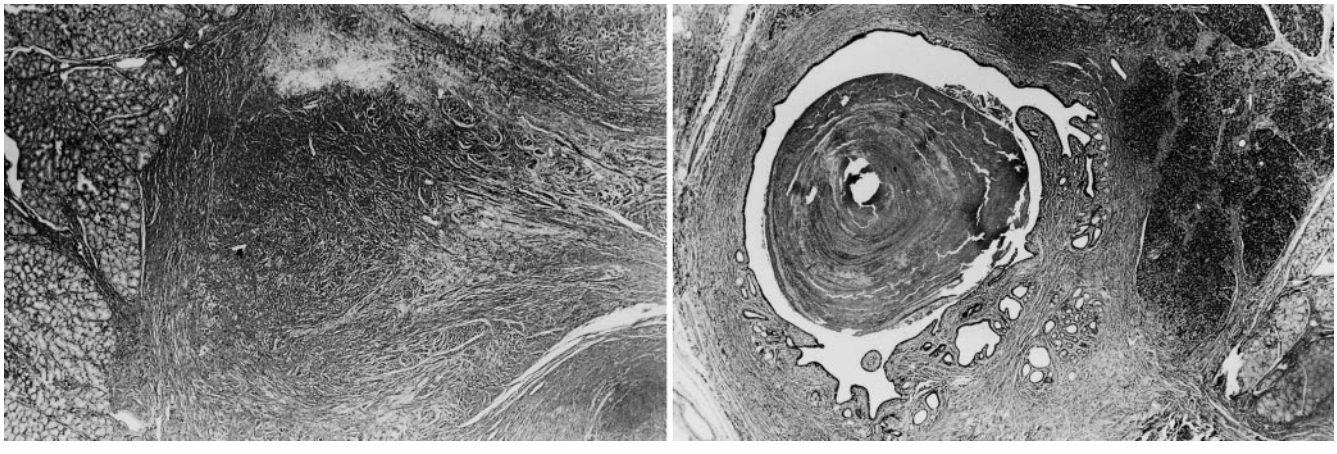


Fig. 4. Endoscopic ultrasonography revealing a hypoechoic mass situated mainly between the pancreas head and duodenal wall with some extention into the pancreatic parenchyma, with penetrating duct sign (*arrow*)



a **b**

Fig. 5a,b. Photomicrographs showing **a** marked hyperplasia of Brunner's glands and extensive scarring between the duodenum and the head of the pancreas, with high-grade duodenal wall cicatrization and **b** protein plugs in Santorini's duct. Hematoxylin-eosin stain, ×20

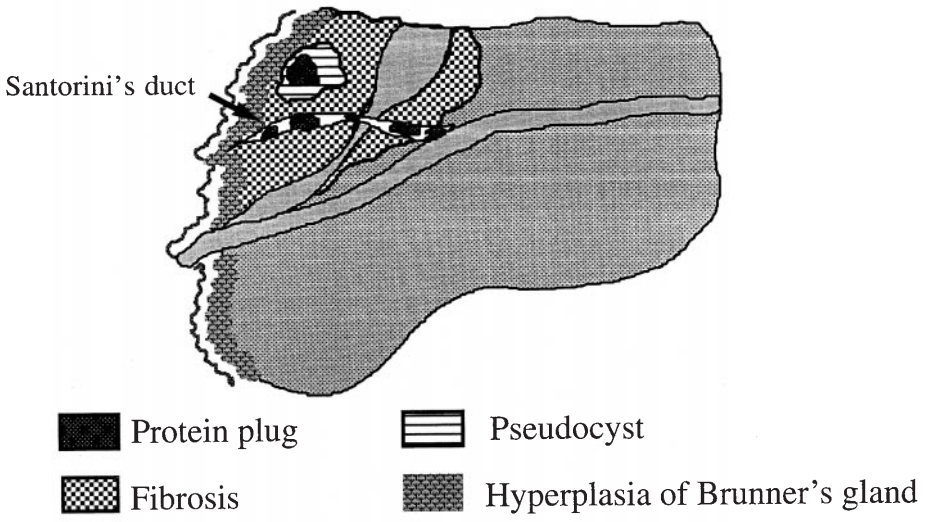


Fig. 6. Schematic diagram of histopathological findings

grade supra-ampullary stenosis of the duodenum was caused by a periduodenal cicatricial plate. The tumor was contiguous with the duodenum. Marked fibrosis was observed in the area of Santorini's duct, but not in the area of Wirsung's duct.

Histologically, extensive fibrosis with round cell infiltration affected both the dorso-cranial portion of pancreas head and the groove. The duodenal wall was thickened, with hyperplasia of Brunner's gland. In addition, protein plugs in Santorini's duct and pseudocysts in pancreas head were observed (Figs. 5 and 6). The patient was relieved from abdominal pain post operation. Up to the present, the patient has been good condition.

Discussion

Groove pancreatitis, a variant of chronic pancreatitis in which scarring is found mainly in the groove between pancreas head, common bile duct, and duodenum, sometimes masquerades clinically as pancreatic head carcinoma. This disease is rare[1,2] and its low frequency of detection may be caused partly by lack of familiarity. Stolte et al.[3] reported that the etiology of groove pancreatitis in its various forms did not differ in any way from that of other types of pancreatitis.

In pure groove pancreatitis, scarring is found only in the groove, while no cicatrization of the pancreatic parenchyma is seen; the pancreatic duct is intact and the condition is seldom detected. In segmental groove pancreatitis, scarring is found not only in the groove but also in pancreatic parenchyma. The replacement of parenchyma by scar tissue is extended to the dorso-cranial portion of the pancreas head. Segmental groove pancreatitis often shows stenosis or obstruction of Santorini's duct, while Wirsung's duct is intact. The sole peculiarity of groove pancreatitis is its topography with respect to degree and spread. The anatomical and clinical symptomatology is derived from its particular topography. The main symptom is often severe upper abdominal pain, and subsequently, impaired motility and stenosis of the duodenum, disordered gastric emptying, postprandial vomiting, and postprandial pain often lead to rapid loss of weight.

Preoperative differentiation between groove pancreatitis and pancreatic carcinoma is difficult and sometimes impossible.[4,5] In most cases reported previously, surgery was performed because pancreatic carcinoma was suspected. However, we were able to diagnose groove pancreatitis preoperatively in our patient. We differentiated this case from pancreatic carcinoma as follows. The patient had a long history of alcohol abuse, a feature often found in groove pancreatitis. Endoscopic biopsy specimens obtained from the edematous mucosa of the second portion of duodenum showed only inflammation. CT revealed the duodenal wall thickening and swelling of the pancreas head that correspond to the histological characteristics of this disease, and the cystic lesions often found in this disease. Generally, as Luetmen et al.[6] report, CT reveals most anatomical findings of the pancreas and the surrounding tissues: duodenal wall thickening, mass, and cysts. However, the EUS approach allows clearer imaging of the extent and location of the inflammatory abnormality[7] and it is also possible to evaluate to which layer of the duodenum the inflammation extends. Thus, we used both CT and EUS for evaluating the extent and location of tumor. Dynamic CT demonstrated an enhanced mass, which was considered suggestive of groove pancreatitis. The mass of pancreatic carcinoma

is seen as non-enhanced.[8] This difference may be the critical finding for the differentiation between two diseases. But it should be noted that groove pancreatitis often reveals poor enhancement, and this may be due to the delayed circulation caused by proliferation of fibrous tissue and the secondary constriction of arteries.

ERCP and/or EUS findings of the common bile duct are also useful in the differentiation between the two diseases. Although a smooth, tubular stenosis of the common bile duct is a frequent and characteristic occurrence in groove pancreatitis, genuine obstructive jaundice is found only rarely.[9,10] On the other hand, irregular ductal stenosis or obstruction with subsequent jaundice is found in carcinoma. In our patient, ERCP showed smooth stenosis of the common bile duct and Wirsung's duct in the head, and no visualization of Santorini's duct. Histological examination showed protein plugs in Santorini's duct. We emphasize this finding of Santorini's duct as one of the factors in the development of groove pancreatitis. EUS revealed a hypoechoic mass with penetrating duct sign, without nodular margin, characteristic of groove pancreatitis, and celiac angiography showed no encasement or occlusion. These findings suggested a tumor caused by groove pancreatitis rather than by pancreatic carcinoma. Thus, we diagnosed this present case as groove pancreatitis according to the mass location, the imaging characteristics, and the pathogenesis.

However, the pathogenesis of groove pancreatitis is still unclear. On the basis of our present case and previous reports,[9,10] we speculate that an important factors is whether the minor papilla is not present at all, or is present only in a rudimentary form. That is to say, when the minor papilla happens to be closed for some reason, the flow of pancreatic juice in Santorini's duct is stagnant. For example, chronic consumption of alcohol leads to an increase in the cholinergic tone of ganglionic synapses.[11] This effect stimulates a functional, trophic effect on Brunner's glands and leads to hyperplasia of these glands. The secretion from the minor papilla is disturbed because of this hyperplasia of Brunner's glands. Pancreatic juice in Santorini's duct flows toward the pancreas body and encounters an acute angle in the so-called Wirsungian knee, the result being interference with the flow and a temporary back-up of secretion in the peripheral sections of pancreas head. This phenomenon may be exacerbated by a more viscous and protein-rich pancreatic juice caused by chronic excessive ingestion of alcohol.[5] This may be a possible explanation for the segmental form of the disease. In the present patient, we considered that the chronic stagnancy of pancreatic juice in Santorini's duct caused by hyperplasia of Brunner's glands led to the formation of eosinophilic protein plugs, and that the subsequent disturbance of pancreatic juice drainage caused by the protein plugs led to the damming up of the juice and development of the localized pancreatitis in the cranial part of pancreas head. The present case suggests that the presence of impacted protein plugs in Santorini's duct is a pathogenic factor in the development of groove pancreatitis. Therefore, the findings of Santorini's duct on ERCP are very important in the diagnosis of groove pancreatitis.

As shown in Table 1, we reviewed the clinicopathologic and radiologic features of groove pancreatitis

Table 1. Cases of groove pancreatitis reported in Japan

Author	Patient Age/Sex	Symptom	Alcoholic	Duodenal stenosis	Operation	Biliary tract findings	ERCP findings of Santorini's duct	Ca
1. Yamashita	52 M	Vomiting	+	−	PD		Stenosis	−
2. Sugiyama	53 M	Nausea	+	+	PD			−
3. Saitoh	69 M	Epigastralgia	+	+	Distal gastrectomy	Gallstone		−
4. Okayama	37 F	Abd.fullness	+	+	PD	n.p.	Not demonstrated	−
5. Watanabe	51 M	Epigastralgia	+	+	PD	n.p.	Pancreatic stone	−
6. Oikawa	53 M	Epigastralgia	+	+	Pancreatojejunostomy	Stenosis (Bi)	Not demonstrated	−
7. Ozeki	62 F	Nausea	−	+	PD	n.p.		−
8. Matsumoto	57 M	Jaundice	+	+	PD	Stenosis (Bi)		
9. Taya	54 M	Epigastralgia	+	+	PD	Stenosis (Bi)		+
10. Fukuhara	40 M	Vomiting	+	−	PD	Wall rigidity	Slight irregularity	−
11. Fukahori	42 M	Back pain	+	+	—	Stenosis (Bi)	Irregularity	
12. Okabe	40 M	Vomiting	+	−	PpPD		Irregularity	−
13. Kobayashi	59 M	Vomiting	+	+	Duodenoduodenostomy	Left shifted	Not demonstrated	−
14. Uchizono	56 M	No symptoms	+	+	—	Left shifted	Not demonstrated	
15. Kiyohara	47 M	Epigastralgia	+	+	PD	Stenosis (Bi)	Cystic dilatation	−
16. Kaneko	43 F	Jaundice		+	PD			+
17. Present study	53 M	Epigastralgia	+	+	PpPD	Stenosis (Bi)	Not demonstrated	−

PD, pancreatoduodenectomy; PpPD, pylorus-preserving pancreatoduodenectomy; Abd., Abdominal; n.p., not particular; Bi, bile duct inferior; ERCP, endoscopic retrograde cholargio pancreatography; Ca, cancer.

reported in the Japanese literature. The 17 patients (14 men and 3 women) had a median age of 51 years (range, 37–69 years). Sixteen of the 17 patients had symptoms (6, abdominal pain; 4, vomiting; 2, jaundice; 2, nausea; 1, abdominal fullness; and 1, back pain) and only 1 patient was asymptomatic. All but one patient were alcoholics. Radiologically, duodenal stenosis was evident in 14 patients and biliary stenosis in 6, and left-shifted common bile duct in 2. The biliary stenosis was characterized by smooth tapering. ERCP was so difficult to perform because of the marked duodenal stenosis that it was unsuccessful in 6 of the 17 patients. In all 11 patients in which ERCP was successful, Wirsung's duct was observed intact or slight change with regularity, whereas Santorini's duct was not demonstrated in 5 patients, irregularity in 3, stenosis in 1, cystic dilatation in 1, and intraductal stone in 1. Preoperatively, almost all patients were suspected of having pancreatic head carcinoma. Pancreatic head carcinoma could not be ruled out clinically, thus leading the surgeons to operate. Of note, 2 patients with groove pancreatitis with adenocarcinoma were found.

Although follow-up of the clinical course, and various combinations of diagnostic imaging lead to a high probability of a correct diagnosis, resection is often the final arbiter. Groove pancreatitis should be kept in mind as a differential diagnosis of pancreatic head tumor. Awareness of this disease may prevent excessive surgical intervention.

References

1. Becker V. Bauchspeicheldruse. In: Doerr W, Seifert G, Uhlinger E (eds) Spezielle pathologische Anatomie, Bd. VI. Berlin Heidelberg New York: Springer, 1973.
2. Falck J, Voigt K. Normal pancreatic duct system in severe chronic pancreatitis of the head of the pancreas. Endoscopy 1975;7:173–175.
3. Stolte M, Weiß W, Volkholz H, et al. A special form of segmental pancreatitis: "Groove Pancreatitis". Hepatogastroenterology 1982;29:198–208.
4. Moosa AR. Surgical treatment of chronic pancreatitis: An overview. Br J Surg 1987;74:661–667.
5. Yamaguchi K, Tanaka M. Groove pancreatitis masquerading as pancreatic carcinoma. Am J Surg 1992;163:312–318.
6. Luetmer PH, Stephens DH, Ward EM. Chronic pancreatitis: Reassessment with current CT. Radiology 1989;171:353–357.
7. Tio TL, Luiken GJHM, Tytgat GNJ. Endosonography of groove pancreatitis. Endoscopy 1991;23:291–293.
8. Freeny PC, Marks WM, Ryan JA, et al. Pancreatic ductal adenocarcinoma: Diagnosis and staging with dynamic CT. Radiology 1988;166:125–133.
9. Becker V, Mischke U. Special article. Groove pancreatitis. Int J Pancreatol 1991;10:173–182.
10. Newton BB, Rittenbury MS, Anderson MC. Extrahepatic biliary obstruction associated with pancreatitis. Ann Surg 1983;197:645–653.
11. Sarles H, Sahel J. Cholestasis and lesions of the biliary tract in chronic pancreatitis. Gut 1978;19:851–857.
12. Yamashita Y, Bekki N, Nohtomi M, et al. A case of groove pancreatitis presenting as duodenal submucosal tumor (in Japanese with English abstract). Nippon Rinshou Geka Igakkai Zasshi 1986;46:1350–1354.
13. Sugiyama K, Nakano S, Takeda I, et al. A case of groove pancreatitis and review of the pertinent literature in Japan (in Japanese with English abstract). Suizou (J Japn Pancreas Soc) 1989;4:78–86.
14. Saitoh S, Kimura T, Yoda M, et al. A case of groove pancreatitis (in Japanese with English abstract). The Journal of Ushioda general Hospital 1990;5:25–35.
15. Okayama Y, Miyaji M, Hayakawa T, et al. A case of duodenal stenosis due to segmental chronic pancreatitis: "Groove Pancreatitis" (in Japanese with English abstract). Nippon Shoukakinaishikyo Gakkai Zasshi (Jpn J Gastroenterol Endoscopy) 1990;32:1986–1993.
16. Watanabe H, Fujita N, Lee S, et al. A case of preoperatively diagnosed groove pancreatitis (in Japanese with English abstract). Nippon Shoukakinaishikyo Gakkai Zasshi (Jpn J Gastroenterol Endoscopy) 1990;32:2272–2277.
17. Oikawa I, Hirata K, Akino Y, et al. A case of the groove pancreatitis appearing as the cystadenocarcinoma (in Japanese with English abstract). Nippon Rinshou Geka Igakkai Zasshi 1990;51:2506–2509.
18. Ozeki Y, Onitsuka A, Hayashi M, et al. Idiopathic duodenal obstruction resembling pure groove pancreatitis: case report (in Japanese). Nippon Shoukakibyo Gakkai Zasshi (Jpn J Gastroenterology) 1991;88:1140–1144.
19. Matsumoto M, Goshima H, Tomita R, et al. A case of Recurrent Duodenal Stenosis due to Segmental Groove Pancreatitis (in Japanese). Tan to sui 1991;12:1397–1402.
20. Taya N, Okamoto M, Shirota K, A case of segmental groove pancreatitis accompanied by minute carcinoma of Santorini's duct (in Japanese with English abstract). Suizou (J Japn Pancreas Soc) 1993;8:449–455.
21. Fukuhara T, Fujie S, Nakajima S, et al. A Case of Groove Pancreatitis (in Japanese with English abstract). Fukubu Gazou Shindan 1993;13:1073–1078.
22. Fukahori T, Muranaka H, Ueno M, et al. A Case of Groove Pancreatitis (in Japanese). Tan to sui 1994;15:281–285.
23. Okabe Y, Itani T, Murata Y, et al. A case of groove pancreatitis (in Japanese). Shoukakika 1994;18:479–481.
24. Kobayashi M, Araki K, Nakamura S, et al. A case of Groove Pancreatitis with Duodenal Stenosis (in Japanese with English abstract). Nippon Shoukaki Geka Gakkai Zasshi (Jpn J Gastroenterol Surg Soc) 1995;28:2290–2294.
25. Uchizono Y, Itou T, Migita R, et al. A case of groove pancreatitis with duodenal stenosis and duodenal ulcer (in Japanese). Rinshou to kenkyu 1996;73:118–122.
26. Kiyohara K, Kosugi M, Ietsugu K, et al. A case of groove pancreatitis with duodenal stenosis (in Japanese). Nippon Shoukakibyo Gakkai Zasshi (Jpn J Gastroenterology) 1996; 93:681–687.
27. Kaneko T, Nakao T, Nomoto S, et al. A case of carcinoma of the head of the pancreas mimicking groove pancreatitis duct (in Japanese with English abstract). Suizou (J Japn Pancreas Soc) 1996;11:328–333.