

Endovascular Management of Ruptured Pancreaticoduodenal Artery Aneurysms Associated with Celiac Axis Stenosis

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Abstract The purpose of this study was to assess the efficacy of transcatheter arterial embolization for ruptured pancreaticoduodenal artery (PDA) aneurysms associated with celiac axis stenosis (CS). Seven patients (four men and three women; mean age, 64; range, 43–84) were treated with transcatheter arterial embolization between 2002 and 2007. They were analyzed with regard to the clinical presentation, radiological finding, procedure, and outcome. All patients presented with sudden epigastric pain or abdominal discomfort. Contrast-enhanced CT showed a small aneurysm and retroperitoneal hematoma around the pancreatic head in all patients. The aneurysms ranged from 0.3 to 0.9 cm in size. In one patient, two aneurysms were detected. The aneurysms were located in the pancreaticoduodenal artery ($n = 5$) and the dorsal pancreatic artery ($n = 3$). Embolization was performed with microcoils in all aneurysms ($n = 8$). *N*-Butyl 2-cyanoacrylate ($n = 1$) and gelatine particle ($n = 1$) were also used. Complete occlusion was achieved in four patients. In the other three patients, a significantly reduced flow to the aneurysm remained at final angiography. However, these aneurysms were thrombosed on follow-up CT within 2 weeks. And

there was no recurrence of the symptoms and bleeding during follow-up (mean, 28 months; range, 5–65 months) in all patients. In conclusion, transcatheter arterial embolization for PDA aneurysms associated with CS is effective. Significant reduction of the flow to the aneurysm at final angiography may be predictive of future thrombosis.

Keywords Pancreaticoduodenal artery · Rupture · Aneurysm · Celiac axis stenosis · Embolization

Introduction

Splanchnic artery aneurysms are relatively rare disease with an incidence of 0.01–0.2% in autopsies. Specifically, pancreaticoduodenal artery (PDA) aneurysms account for only 2% of all splanchnic artery aneurysms [1]. The causes of PDA aneurysms are usually arteriosclerosis, celiac axis stenosis (CS), and congenital vascular disease. PDA pseudoaneurysms, which are caused by pancreatitis, trauma, and infection, are more often seen. Therefore, PDA aneurysms associated with CS are the subgroup of all PDA aneurysms. The subtotal of CS-associated aneurysms of all PDA aneurysms is about 7% [2]. These aneurysms are caused by a high-flow state of the pancreatic arterial arcade due to CS. Therefore, the clinical management of CS associated aneurysms should be discussed separately from that of other PDA aneurysms without CS and PDA pseudoaneurysms. However, many reviews about PDA aneurysms treated with embolization include both true aneurysms (with and without CS) and pseudoaneurysms in their patient databases [3–5]. In addition, most studies addressing PDA aneurysms associated with CS are published as case reports [6–12].

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The purpose of this study was to review and assess the efficacy of transcatheter arterial embolization for PDA aneurysms associated with CS.

Materials and Methods

A retrospective analysis was conducted of all patients with PDA aneurysms who had been treated with transcatheter arterial embolization at four medical centers, from January 2002 to December 2007. PDA aneurysms without CS and PDA pseudoaneurysms were excluded.

A total of seven patients (four men, three women) with a mean age of 64 years (range, 43 to 84 years) were identified. Two patients had a medical history of systemic hypertension, and one patient had cholelithiasis. The summary of the results is presented in Table 1. They were analyzed with regard to the clinical presentation, radiological findings, procedure, and outcome. To discuss the effect of transcatheter arterial embolization, the results were assessed with regard to technical success and clinical success. Technical success was defined as nonvisualization of the aneurysm at final angiography. Clinical success was defined as freedom from symptoms and bleeding during the follow-up. Radiological findings of CT and angiography and the cause of CS were analyzed by the consensus of two radiologists. CS was defined as luminal narrowing of the celiac axis orifice exceeding 50%. The cause of CS was determined according to the following findings of CT and angiography. The criteria for diagnosis of celiac axis compression by the median arcuate ligament included the diaphragmatic crura surrounding the origin of the celiac axis on CT, poststenotic dilatation of the distal portion of the celiac axis, and superior

notching, and acute downward angulation of the proximal celiac axis on angiography [13, 14]. Criteria for diagnosis of atherosclerosis included calcification or atheromatous plaque of the celiac axis on CT and concentric stenosis of the orifice of the celiac axis on angiography.

Results

Clinical Presentation

At the time of hospital admission, six patients had sudden, severe abdominal pain, and one patient had only nausea and abdominal discomfort. No hematemesis was observed in any patient. Six patients were hemodynamically stable. One patient with epigastric pain was in hypovolemic shock and required immediate volemic resuscitation. They were admitted based on the symptoms of a ruptured aneurysm. There was no symptom associated with the aneurysm before rupture in all patients. Median arcuate ligament syndrome, which shows abdominal angina-like symptoms, was not clinically suspected in any of the patients.

Radiological Findings

Contrast-enhanced CT was performed in all patients. CT angiography was also reconstructed in five patients. Contrast-enhanced CT images showed retroperitoneal hematoma around the pancreatic head. In addition, a small aneurysm around the pancreatic head could be detected in all patients. There was no calcification at the wall of the aneurysms. The aneurysms ranged from 0.3 to 0.9 cm in diameter (mean, 0.6 cm) on CT. Stenosis of the celiac axis also could be detected on CT in all patients. Complete

Table 1 Summary of patient data

Patient no., age/gender	Location of aneurysm	Presentation	Size (mm)	Cause of stenosis	Embolization	Approach route	Embolic material	Technical success	Clinical success
1. 55/F	APDA	Rupture	9	MAL	Aff, Eff	SMA CA	Microcoils	No	Yes
2. 84/M	PPDA	Rupture	6	Atherosclerosis	Packing Aff, Eff	SMA	Microcoils	Yes	Yes
3. 60/F	DPA	Rupture	5	MAL	Packing Aff, Eff	SMA	Microcoils	Yes	Yes
4. 79/M	DPA	Rupture	8	MAL	Aff, Eff	SMA CA	Microcoils NBCA	No	Yes
5. 43/M	DPA	Rupture	5	MAL	Aff	SMA	Microcoils	No	Yes
6. 56/M	PPDA	Rupture	3	MAL	Packing Aff, Eff	SMA CA	Microcoils Gelatin	Yes	Yes
7. 73/F	PPDA APDA	Rupture	7 6	MAL	Packing Aff, Eff	SMA CA	Microcoils	Yes	Yes

Note: APDA, anterior pancreaticoduodenal artery; PPDA, posterior pancreaticoduodenal artery; MAL, median arcuate ligament; Aff, afferent artery embolization; Eff, efferent artery embolization; SMA, superior mesenteric artery; CA, celiac axis

occlusion of the celiac axis was seen in one patient on celiac axis angiography. Superior mesenteric artery (SMA) angiography showed a PDA aneurysm and retrograde flow in the gastroduodenal artery with the collateral pancreatic arterial arcade connecting to the branch of the celiac axis in all patients. The causes of celiac axis stenosis were diagnosed as extrinsic compression from the median arcuate ligament of the diaphragm in six patients and as atherosclerosis in one patient.

The locations of the embolized aneurysms were the anterior PDA ($n = 2$), the posterior PDA ($n = 3$), and the dorsal pancreatic artery (DPA)/branch of the DPA ($n = 3$).

Embolization Technique

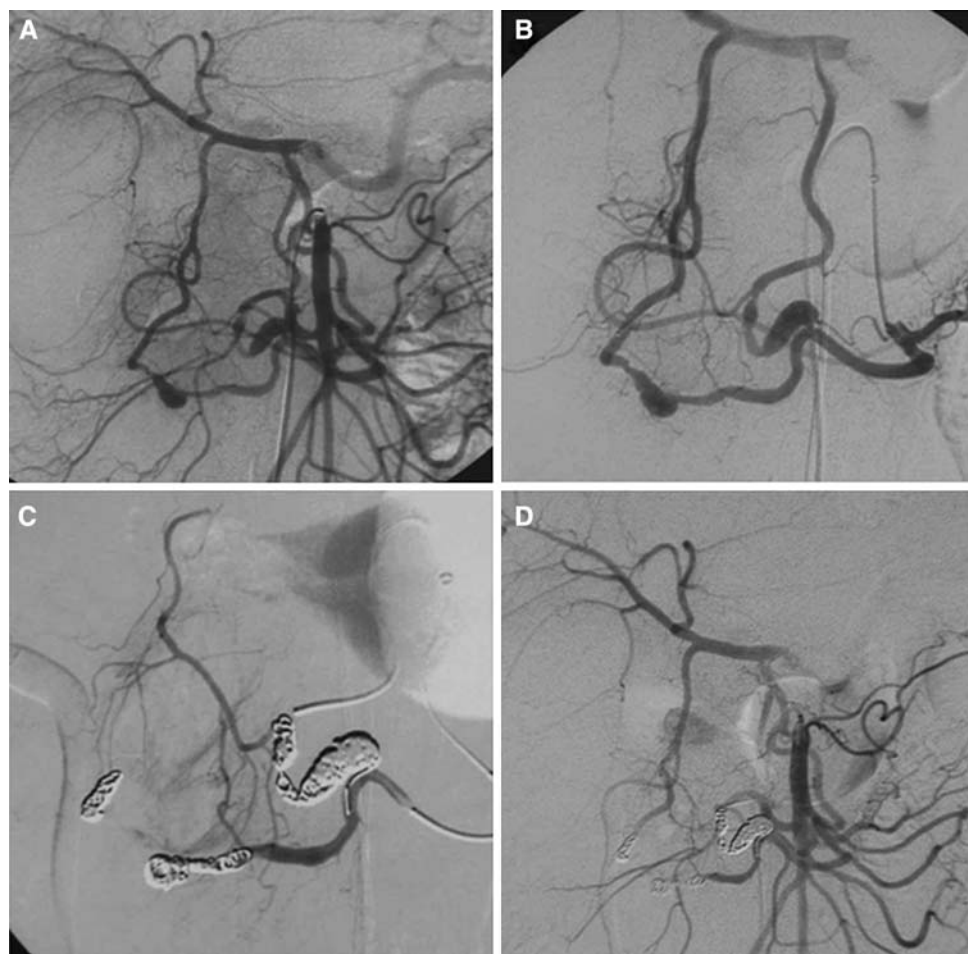
Transcatheter arterial embolization was performed 0 to 24 days (median, 2 days) after hospital admission. In the patient of 24 days, the only symptom was nausea and abdominal discomfort, and the general state of the patient was stable. At the first admission to another hospital, a pancreatic head tumor was suspected. Then the patient was introduced to our hospital to treat the mass, which was later diagnosed as retroperitoneal hematoma. Therefore, it took

time to reach the exact diagnosis of PDA aneurysm. In that patient, transcatheter arterial embolization was performed 6 days after the diagnosis. The coaxial catheterization technique (2.0- to 2.3-Fr microcatheter and 4-Fr catheter) was used in all patients. In one patient, a 5-Fr guiding sheath, which was advanced to the SMA, was also used. Methods of embolization were afferent and efferent artery embolization and packing of the aneurysm ($n = 5$), afferent and efferent artery embolization ($n = 2$), and afferent artery embolization only ($n = 1$). In all patients, transcatheter arterial embolization was performed under hemodynamically stable status, and there was no active contrast extravasation during the procedure. Microcoils were used in all aneurysms ($n = 8$). *N*-Butyl cyanoacrylate (NBCA; $n = 1$) and gelatin particles ($n = 1$) were also used in addition to the microcoils. Approach routes to the aneurysms were via SMA and CA ($n = 5$) and via SMA ($n = 3$).

Outcome

In this study, four of the seven patients (five of the eight aneurysms) were successfully embolized without residual flow to the aneurysm at final angiography (Fig. 1). The

Fig. 1 Patient 7: (A) SMA angiography shows retrograde flow to the branch of the celiac axis via the dilated pancreatic arterial arcade and the DPA. (B) Selective inferior PDA angiography shows two aneurysms, at the posterior PDA and the anterior PDA. (C) Inferior PDA angiography after embolization shows complete occlusion of the two aneurysms. (D) Flow to the branch of the celiac axis is preserved by the developed collateral arterial network



immediate technical success rate was 57% (four of seven patients). In the other three patients (three of the eight aneurysms), significantly reduced flow to the aneurysm remained at final angiography (Fig. 2). The remaining artery was extremely narrow and tortuous. Therefore, it was not possible to advance the microcatheter to the aneurysm through the vessel. However, all these aneurysms were observed to be thrombosed on contrast-enhanced CT which was performed 1 to 2 weeks after embolization. There was no recurrence of symptoms or bleeding after embolization in all patients. The follow-up period was 5 to 65 months (mean, 28 months). Therefore, complete occlusion of the aneurysm was achieved in all seven patients (eight of the eight aneurysms), and the clinical success rate was 100%. There was no complication associated with embolization.

In one patient with a posterior PDA aneurysm, the branch of the DPA was also irregular and slightly dilated (it was diagnosed to be a microaneurysm) on SMA angiography during posterior PDA embolization. SMA angiography was performed to embolize the DPA microaneurysm 23 days after the first embolization. However, SMA angiography showed improvement of the DPA microaneurysm (Fig. 3).

Discussion

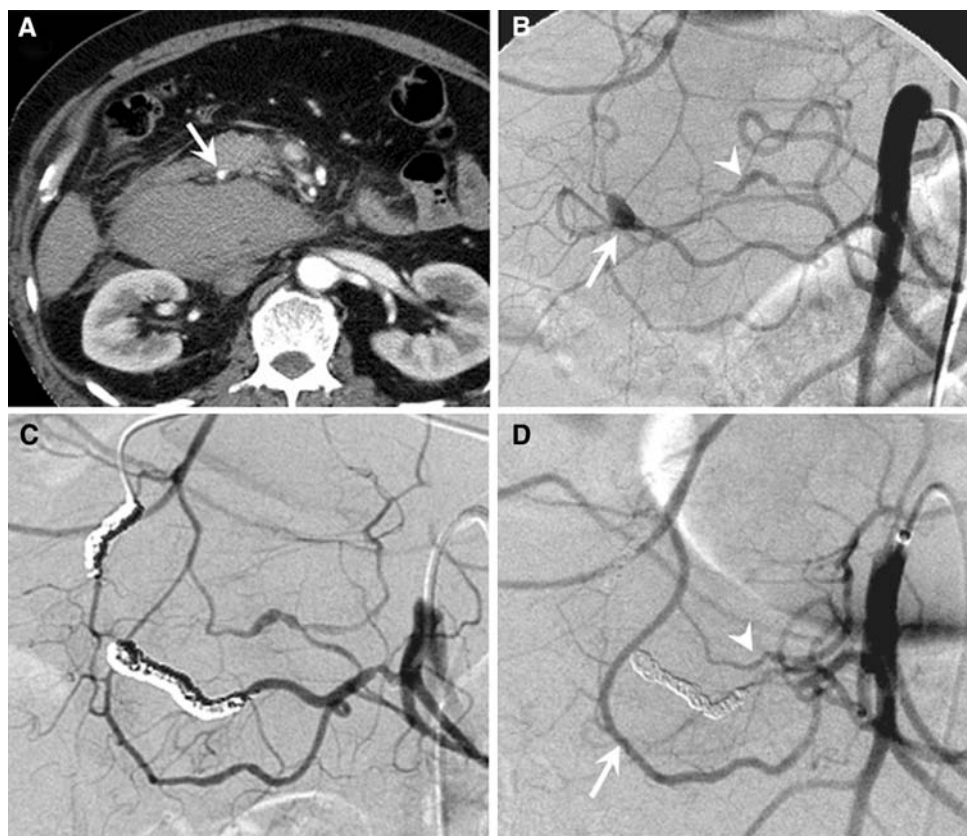
According to previous publications, 50–80% of reported true PDA aneurysms have been accompanied by CS [3, 6, 7]. Therefore, CS is considered to be the most common underlying cause of PDA aneurysm formation. The relationship of PDA aneurysms and CS was first described by Sutton and Lawton in 1973 [15]. They suggested that a high-flow state in the pancreatic arterial arcade as a collateral supply from the SMA to the branch of the celiac axis could lead to aneurysm formation.

The incidence of CS without a PDA aneurysm is comparatively as high as 7.3–12.5% [16, 17], and asymptomatic stenosis of the celiac axis is a common finding. However, PDA aneurysms are clinically quite rare. Uher et al. [7] reported that weakness of the arterial wall such as atherosclerosis or congenital dysplasia was also a determining factor for aneurysm formation, in conjunction with CS. Indeed, some histological examinations of surgically resected specimens of CS-associated aneurysms showed fibromuscular dysplasia [7], segmental arterial mediolysis [8], and atherosclerosis [18]. On the other hand, some reports stated that there was no abnormality at the arterial

Fig. 2 Patient 4: (A) SMA angiography shows retrograde flow to the branch of the celiac axis and a DPA aneurysm. (B) Selective dorsal pancreatic arterial angiography shows the DPA aneurysm connecting to multiple vessels. (C) Final angiography after five-vessel embolization shows residual aneurysmal perfusion (arrow) through the narrow collateral vessel (arrowhead). However, the aneurysm had thrombosed on follow-up CT at 10 days after embolization



Fig. 3 Patient 6: (A) Contrast-enhanced CT shows a small aneurysm (arrow) and retroperitoneal hematoma. (B) SMA angiography shows retrograde flow in the gastroduodenal artery with a posterior PDA aneurysm (arrow). It also shows a microaneurysm at the branch of the DPA (arrowhead). (C) The posterior PDA aneurysm was embolized via the celiac axis and the SMA. (D) SMA angiography 23 days after embolization shows complete occlusion of the posterior PDA aneurysm and an enlarged anterior PDA (arrow) which has developed as a collateral artery. In addition, the DPA microaneurysm has improved



wall of the PDA aneurysms [4]. In many cases, including our study, pathological specimens of the aneurysms which were treated with embolization were not available. Therefore, it remains undetermined whether weakness of the arterial wall existed in these cases.

Most PDA aneurysms associated with CS are asymptomatic before rupture. Therefore, it is difficult to make an accurate diagnosis of unruptured PDA aneurysm except for cases that are found accidentally in other examinations [2, 18]. All patients in this study were also detected by symptoms of rupture. In contrast to pseudoaneurysms, PDA aneurysms rarely present with gastrointestinal hemorrhage. They usually rupture into the retroperitoneal space and cause acute abdominal pain. Some patients show hypotension and shock. In addition, other symptoms are mainly related to the mass effect of the hematoma such as vomiting, jaundice, or gastric outlet compression [4, 19].

Unlike other splanchnic artery aneurysms, PDA aneurysms show no correlation between size and risk of rupture. There is no evidence that smaller aneurysms are safe because 18% of ruptured aneurysms are <1 cm in diameter [2, 4]. In this review, the mean size of the ruptured aneurysms was 0.6 cm and the smallest one was only 0.3 cm. Therefore, all PDA aneurysms should be treated, whatever their size, at the time of diagnosis.

The operative mortality rate of ruptured PDA aneurysms is relatively high (12–50%) [3]. Recent advances in

interventional materials and techniques have made it possible to perform transcatheter arterial embolization safely and effectively. Therefore, embolization has been more commonly performed as the initial therapy for PDA aneurysms in the last decade. The ideal embolization is complete occlusion of the aneurysms. However, the difficult point of embolization is that the peripancreatic artery (especially the branch of the DPA) is sometimes narrow and tortuous. In addition, flow to the hepatic artery and the mesenteric artery must be preserved to prevent ischemic dysfunction. Moreover, other small arteries of the pancreatic arterial arcade are easily advanced as a collateral vessel after embolization. Even if the afferent and efferent vessels are selectively embolized, flow to the aneurysm may remain via small collateral vessels. However, even if the reduced gradual flow to the PDA aneurysm remains at final angiography, the possibility of complete occlusion or shrinkage of the aneurysm is high. Because the PDA aneurysm is a flow-related aneurysm, the high-flow status is the most aggravating factor. In this study, all PDA aneurysms were small aneurysms (<1 cm). Therefore, the small size of the aneurysms may also have been advantageous for the thrombosis.

In patient 6 in this study, the DPA microaneurysm improved after posterior PDA aneurysm embolization (Fig. 3). After PDA embolization, the flow of the pancreatic arterial arcade changed and the anterior PDA

developed. Therefore, the change must be the cause of the shrinkage of the DPA microaneurysm.

Some studies have reported satisfactory results by revascularization of CS (resection of the median arcuate ligament or stent placement) without local treatment of the aneurysms [20, 21]. In such cases, the treatment strategy was to reduce the retrograde flow of the pancreatic arterial arcade and to prevent possible aneurysm rupture. This theory is consistent with the argument that the reduced residual flow after embolization may not cause enlargement of the aneurysm.

In addition to embolization of the aneurysm, the necessity for revascularization of the celiac axis remains controversial. Some reports recommend revascularization of the celiac axis to prevent aneurysm recurrence, by reestablishing the normal pancreatic arterial circulation [19–24]. In such cases, the celiac axis was restored by either arterial reconstruction, aortohepatic bypass, division of the causative arcuate ligament, or stenting. However, in the majority of the reported cases including our study, only local treatment was performed and the high-flow state of the pancreatic arterial arcade remained. These studies showed good outcomes without recurrence or rupture of the PDA aneurysms [5, 6, 9–12]. To our knowledge, no study has reported the recurrence of PDA aneurysms caused by residual CS after embolization. Therefore, if the risk of ischemic dysfunction of the liver and the duodenum is not so high, additional treatment of the CS may not necessarily be required. However, it is still a matter of fact that these aneurysms are flow related. Therefore more long-term follow-up is necessary to clarify this controversial point.

In conclusion, embolization therapy for PDA aneurysms associated with CS is safe and effective. It should be performed as the initial therapy for CS-associated aneurysms. And even when significantly reduced flow to the aneurysm remains at final angiography, it may be predictive of future thrombosis.

References

- Stanley JC, Wakefield TW, Graham LM et al (1986) Clinical importance and management of splanchnic artery aneurysms. *J Vasc Surg* 3:836–840
- Neschis DG, Safford SD, Golden MA (1998) Management of pancreaticoduodenal artery aneurysms presenting as catastrophic intraabdominal bleeding. *Surgery* 123:8–12
- Mandel SR, Jaques PF, Mauro MA et al (1987) Nonoperative management of peripancreatic arterial aneurysms. *Ann Surg* 205:126–128
- Perrot MD, Berney T, Deleaval J et al (1999) Management of true aneurysms of the pancreaticoduodenal arteries. *Ann Surg* 229:416–420
- Murata S, Tajima H, Fukunaga T et al (2006) Management of pancreaticoduodenal artery aneurysms: results of superselective transcatheter embolization. *AJR* 187:290–298
- Weber CH, Pfeifer KJ, Tato F et al (2005) Transcatheter coil embolization of an aneurysm of the pancreatico-duodenal artery with occluded celiac trunk. *CardioVasc Interv Radiol* 28:259–261
- Uher P, Nyman U, Ivancev K et al (1995) Aneurysms of the pancreaticoduodenal artery associated with occlusion of the celiac artery. *Abdom Imaging* 20:470–473
- Jibiki M, Inoue Y, Iwai T et al (2005) Treatment of three pancreaticoduodenal artery aneurysms associated with coeliac artery occlusion and splenic artery aneurysm: a case report and review of the literature. *Eur J Vasc Endovasc Surg* 29:213–217
- Ogino H, Sato Y, Banno T et al (2002) Embolization in a patient with ruptured anterior inferior pancreaticoduodenal arterial aneurysm with median arcuate ligament syndrome. *CardioVasc Interv Radiol* 25:318–319
- Guijt M, van Delden OM, Koedam NA et al (2004) Rupture of true aneurysms of the pancreaticoduodenal arcade: treatment with transcatheter arterial embolization. *CardioVasc Interv Radiol* 27:166–169
- Kobayashi T, Uenoyama S, Isogai S (2004) Successful transcatheter arterial embolization of an inferior pancreaticoduodenal artery aneurysm associated with celiac axis stenosis. *J Gastroenterol Hepatol* 19:599–601
- Ikeda O, Tamura Y, Nakasone Y et al (2007) Coil embolization of pancreaticoduodenal artery aneurysms associated with celiac artery stenosis: report of three cases. *CardioVasc Interv Radiol* 30:504–507
- Patten RM, Coldwell DM, Ben-Menachem Y (1990) Ligamentous compression of the celiac axis: CT findings in five patients. *AJR* 156:1101–1103
- Horton KM, Talamini MA, Fishman EK (2005) Median arcuate ligament syndrome: evaluation with CT angiography. *Radio Graphics* 25:1177–1182
- Suton D, Lawton G (1973) Celiac stenosis or occlusion with aneurysm of the collateral supply. *Clin Radiol* 24:49–53
- Bron KM, Redman HC (1969) Splanchnic artery stenosis and occlusion: incidence, arteriographic and clinical manifestations. *Radiology* 92:323–328
- Park CM, Chung JW, Kim HB et al (2001) Celiac axis stenosis: incidence and etiologies in asymptomatic individuals. *Korean J Radiol* 2:8–13
- Iyomasa S, Matsutaka Y, Hiei K et al (1995) Pancreaticoduodenal artery aneurysm: a case report and review of the literature. *J Vasc Surg* 22:161–166
- Bageacu S, Cuilleron M, Kaczmarek D et al (2006) True aneurysms of the pancreaticoduodenal artery: successful non-operative management. *Surgery* 139:608–616
- Tien YW, Kao HL, Wang HP (2004) Celiac artery stenting: a new strategy for patients with pancreaticoduodenal artery aneurysm associated with stenosis of the celiac artery. *J Gastroenterol* 39:81–85
- Proud G, Chamberlain J (1978) Aneurysm formation on the small pancreatic arteries in associated with celiac axis compression. *Ann R Coll Surg Engl* 60:294–297
- Suzuki K, Kashimura H, Sato M et al (1998) Pancreaticoduodenal artery aneurysms associated with celiac axis stenosis due to compression by median arcuate ligament and celiac plexus. *J Gastroenterol* 33:434–438
- Quandalle P, Chambon JP, Marache P et al (1990) Pancreaticoduodenal artery aneurysms associated with celiac axis stenosis: report of two cases and review of the literature. *Ann Vasc Surg* 4:540–545
- Ducasse E, Roy F, Chevalier J et al (2004) Aneurysm of the pancreaticoduodenal arteries with a celiac trunk lesion: current management. *J Vasc Surg* 39:906–911