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CASE REPORT

A successful case of pancreaticoduodenectomy with resection of the hepatic arteries preserving a single aberrant hepatic artery for a pancreatic neuroendocrine tumor: report of a case

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Abstract A 65-year-old male with a pancreatic neuroendocrine tumor presenting with a duodenal ulcer was referred to our department. The tumor involved the common hepatic artery, gastroduodenal artery, left hepatic artery and the right posterior hepatic artery, but not the right anterior hepatic artery originating from the superior mesenteric artery. The hepatic arteries, except the aberrant right anterior hepatic artery, were embolized using coils 18 days before the surgery. The patient underwent pancreaticoduodenectomy with resection of the tumor-encased hepatic arteries, while preserving the aberrant artery. The patient was discharged uneventfully on postoperative day 13 with no ischemic complications. A histopathological examination revealed a grade 2 pancreatic neuroendocrine tumor according to the classification of the World Health Organization, and the surgical margin was negative. The patient developed hepatic metastases 16 months after surgery; hence, hepatic resection was performed. The present surgical strategy is applicable in patients with relatively low-grade pancreatic malignancies involving major hepatic arteries.

Keywords Gastrinoma · Pancreaticoduodenectomy · Arterial resection · Aberrant hepatic artery

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Introduction

Invasive pancreatic cancer involving the hepatic arteries is generally contraindicated for surgery, because surgical resection is technically demanding, associated with high morbidity and the expected patient survival rate is no better than that of patients with locally advanced pancreatic cancer undergoing chemotherapy [1, 2]. However, a pancreatic neuroendocrine tumor (pNET) grows relatively slowly, and the 10-year survival rate following surgery for pNETs with major vascular involvement is reportedly 60 % [3]. Therefore, the surgical strategy for pNETs involving hepatic arteries should be different from that for invasive pancreatic cancer.

We herein report a case of pNET involving all of the hepatic arteries, with the exception of a single aberrant right anterior hepatic artery (RAHA). The tumor-encased hepatic arteries were embolized prior to surgery. Thereafter, the patient successfully underwent pancreaticoduodenectomy (PD) with resection of the hepatic arteries, while preserving the aberrant hepatic artery. Cases of PD with resection of an aberrant hepatic artery [4–6] or splenic artery [7] were previously reported; however, this report is the first to describe a case of PD with resection of the common hepatic artery (CHA), and the present surgical strategy is applicable in patients with relatively low-grade pancreatic malignancies involving major hepatic arteries.

Case report

A 65-year-old male with a pancreatic head tumor presenting with a duodenal ulcer was referred to our department. An endoscopic ultrasound-guided fine needle aspiration examination performed at the previous hospital had revealed a grade 2 (G2) pNET according to the World Health Organization (WHO) Classification of Tumors of the Digestive System 2010 [8]. The patient experienced epigastralgia, vomiting and diarrhea, and also reported a body weight reduction of 7 kg over the previous 3 months. One day after his first visit to our hospital, he arrived at our emergency department complaining of severe upper abdominal pain. Following a diagnosis of a perforated duodenal ulcer, he underwent peritoneal lavage and omental implantation. Although the calcium infusion test was negative, a gastrinoma was suspected on the basis of the clinical course.

Dynamic computed tomography (CT) revealed a well-demarcated, enhanced tumor on the pancreatic head, measuring 40 mm in diameter (Fig. 1). The CHA, gastroduodenal artery (GDA), left hepatic artery (LHA) and right

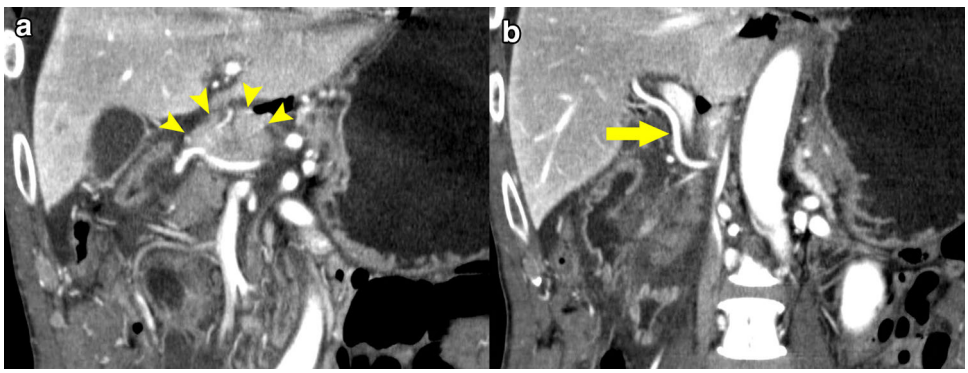

Fig. 1 Preoperative CT. **a** A coronal-view CT scan demonstrated a tumor (*arrow head*) in the pancreatic head and complete encasement of the common hepatic artery by the tumor. **b** A coronal-view CT scan showed that the right anterior hepatic artery (*arrow*) running behind the pancreatic head was free from the tumor

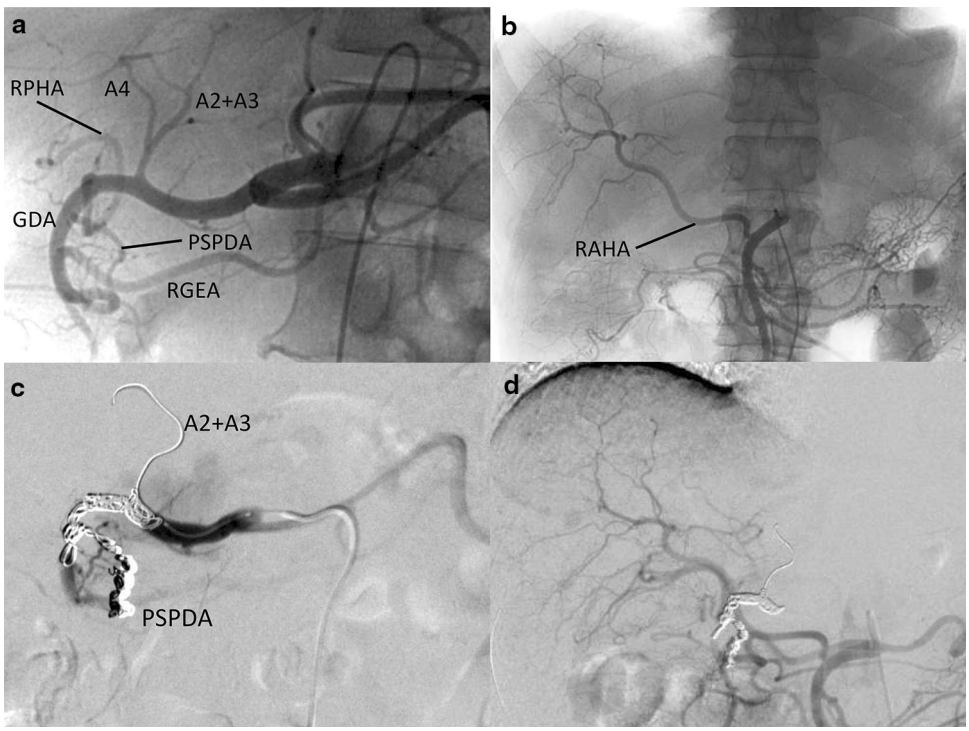

Fig. 2 Arteriography. **a** Celiac arteriography revealed the hepatic arterial anatomy. **b** SMA arteriography revealed the RAHA arising from the SMA. **c** The CHA, GDA, LHA, RPHA and PSPDA were embolized with coils. Post-embolization celiac arteriography demonstrated that the embolized arteries were not enhanced. **d** Post-embolization SMA arteriography visualized the enhanced intrahepatic branches of the LHA and RPHA. *GDA* gastroduodenal artery, *A2+A3* the left lateral segmental branches of the hepatic artery, *A4* the medial segmental branch of the hepatic artery, *RPHA* right posterior hepatic artery, *PSPDA* posterior superior pancreaticoduodenal artery, *RGEA* right gastroepiploic artery, *RAHA* right anterior hepatic artery, *SMA* superior mesenteric artery, *CHA* common hepatic artery, *LHA* left hepatic artery

posterior hepatic artery (RPHA) were surrounded by and involved with the tumor. Only the RAHA originating from the superior mesenteric artery (SMA) was free from the tumor. Therefore, we scheduled a PD with resection of the involved hepatic arteries, while preserving the aberrant RAHA. Preoperative embolization of the CHA, GDA, LHA and RPHA was performed using an interventional approach with mechanical detachable coils (IDC, Boston, Mass) and microcoils (Tornado; Cook Medical, Bloomington, Ind) (Fig. 2). After embolization, the intrahepatic branches of the LHA and RPHA were well visualized by SMA arteriography (Fig. 2d), and the serum transaminase levels were not elevated (Fig. 3). Dynamic CT scans taken 13 days after embolization confirmed uniform intrahepatic arterial blood flow in the whole liver.

On post-embolization day 18, we performed PD with resection of the LHA, RPHA and CHA, while preserving the RAHA. As the tumor involved the nervous plexus around the CHA, the CHA was divided at its origin. The LHA was divided distal to the bifurcation of the left lateral segmental branches (A2+A3) and the medial segmental branch (A4). In addition, the RPHA was divided in the middle, and the RAHA was the only hepatic artery preserved (Fig. 4). Doppler ultrasonography confirmed sufficient blood flow in the intrahepatic arteries, and a partial hepatectomy for a solitary liver metastasis was performed simultaneously. The estimated blood loss was 1700 ml, and the length of the operation was 786 min. The duration of hepatic vascular clamping (Pringle time) was 5 min.

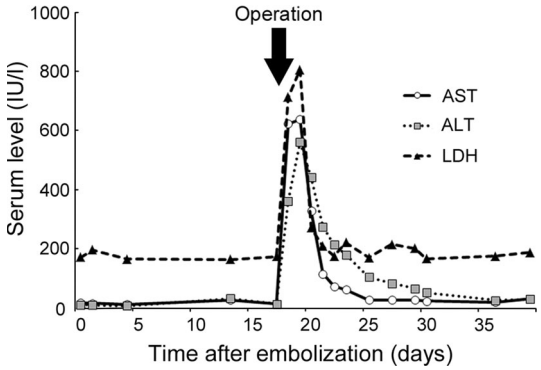

Fig. 3 The trends in the serum levels of AST, ALT and LDH. This graph shows the trends in the serum levels of AST, ALT and LDH after embolization. The serum levels of AST, ALT and LDH were not elevated after embolization. The operation was performed 18 days after embolization. The serum levels of AST, ALT and LDH increased to 638, 561 and 805 IU/L, respectively, after the operation, but normalized by postoperative day 18. *AST* aspartate aminotransferase, *ALT* alanine aminotransferase, *LDH* lactate dehydrogenase

On postoperative day 1, the serum aspartate and alanine transaminase levels increased to 638 and 561 IU/L, respectively, but normalized by day 18 (Fig. 3). As no pancreatic fistula or hepatic abscess formation occurred, the patient was discharged on postoperative day 13. CT scans taken 3 months after surgery demonstrated the development of a collateral system via the hilar plate arterial plexus (Fig. 5).

Following surgery, the preoperative clinical symptoms (i.e., epigastralgia, vomiting and diarrhea) resolved completely, and the patient's quality of life dramatically improved. A histopathological examination revealed a G2 pNET according to the WHO classification [8]. Nine to 11 mitoses per 10 high-power fields were observed, and the Ki-67 labeling index was 20 %. The surgical margin was negative for the tumor, but six of the examined 14 lymph nodes were positive, and there was severe perineural and lymphovascular invasion. As immunohistochemical staining revealed that some of the tumor cells were gastrin positive, the tumor was finally diagnosed as a gastrinoma. Sixteen months after surgery, dynamic CT revealed hepatic metastases; thus, nine small nodules of the liver and a swollen lymph node in the lesser omentum have just been resected 20 months after the initial surgery.

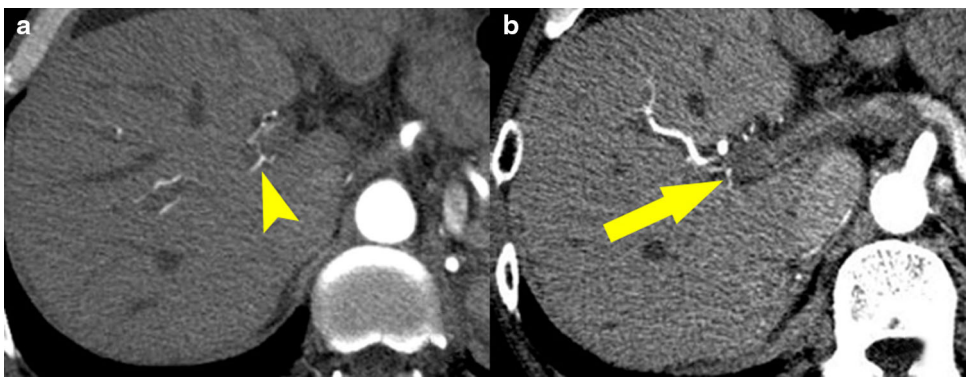

Fig. 4 A schematic diagram of the PD procedure with en bloc resection of the hepatic arteries. The CHA, A2+A3, A4 and RPHA were divided as shown. The pancreatic transection line was also drawn in the diagram. *LGA* left gastric artery, *LGV* left gastric vein; A2+A3 m the left lateral segmental branches of the hepatic artery, *A4* the medial segmental branch of the hepatic artery, *RAHA* right anterior hepatic artery, *RPHA* right posterior hepatic artery, *PD* pancreaticoduodenectomy, *CHA* common hepatic artery

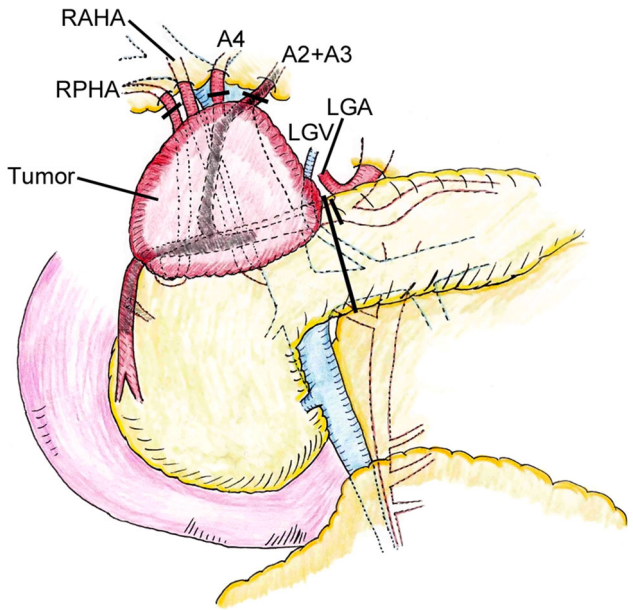

Fig. 5 A CT scan taken 3 months after surgery. The development of a collateral system at the hilar plate was observed. **a** CT demonstrated the communicating arcade (*arrow head*) from the right anterior hepatic artery to the intrahepatic branches of the left hepatic artery. **b** CT demonstrated the communicating arcade (*arrow*) from the right anterior hepatic artery to the intrahepatic branches of the right posterior hepatic artery

Discussion

We successfully performed PD for pNET, accompanied by *en bloc* resection of the encased hepatic arteries following preoperative embolization, while preserving the aberrant RAHA. Invasive pancreatic cancer involving the hepatic arteries is generally contraindicated for surgery [1, 2]; however, surgery can be performed for pNET even with major vascular involvement, because pNETs grow relatively slowly [3]. In the present case, the aberrant RAHA originating from SMA was free from the tumor, and we had preoperatively embolized the encased hepatic arteries. With the aid of this procedure, an *en bloc* resection of the encased arteries was performed safely without any ischemic complications. A curative (R0) resection was achieved, and the preoperative clinical symptoms completely resolved. Therefore, this surgical strategy may be recommended for patients with relatively low-grade pancreatic malignancies, especially when symptomatic.

PD with *en bloc* resection of the major arteries for pancreatic cancer remains controversial, as Marangoni et al. [1] argued that arterial resection during pancreatic surgery was associated with higher morbidity and mortality rates. Moreover, Nakao et al. [2] described that procedure-related death (within 30 days after surgery) occurred in five of 14 (35.7 %) patients undergoing portal vein plus arterial resection, and the survival rate of those patients was no better than that of an unresectable group.

On the other hand, aggressive surgical resection has been reported in patients with pNET involving the major vessels. Norton et al. [3] argued that neuroendocrine tumors should be aggressively resected even when there is major vascular abutment, involvement or encasement, because the 10-year and disease-free survival rates of patients who underwent surgery for pNETs involving major vessels were 60 and 30 %, respectively. These data are encouraging, because patients with vascular involvement who did not undergo surgery had a 5-year survival rate of 30–40 % [3].

Ischemic complications, such as hepatic infarction, multiple liver abscesses and breakdown of the bilioenteric anastomosis may occur even with meticulous attention to radical resection of the hepatic arteries during pancreatic surgery. The occurrence of hepatic infarction and multiple liver abscesses is significant because it sometimes lead to hepatic failure [9, 10]; moreover, a sufficient blood supply to the biliohepatic system is mandatory to ensure a safe bilioenteric anastomosis [11].

In the present case, the CHA, GDA, LHA and RPHA were completely encased by the tumor, and only the aberrant RAHA originating from SMA was intact. Vellar et al. [12] reported that right and left hepatic arteries communicate freely via the hilar plate arterial plexus, and this collateral system plays an important role when the right or left hepatic artery is injured during hepatobiliary procedures. As we planned to divide the LHA and RPHA, the blood supply through this communicating arcade was presumed to be crucial in the present situation for managing the encased arteries and aberrant RAHA. Therefore, we decided to perform preoperative embolization to increase the blood supply via this arcade. Indeed, CT scans taken 3 months after the operation showed the development of this collateral system (Fig. 5).

The usefulness of preoperative embolization was first reported in cases of extended distal pancreatectomy with *en bloc* resection of the celiac arteries for invasive pancreatic cancer. Kondo et al. [13] argued that preoperative embolization of the celiac artery can reduce the risk of ischemic gastritis in such cases; in addition, Miyamoto et al. [4] reported that preoperative embolization was useful during PD with *en bloc* resection of the hepatic artery in a patient with pancreatic head cancer involving the CHA originating from the SMA (Table 1; Fig. 6a). Furthermore, Sasaki et al. [5] reported a case of cholangiocarcinoma involving a replaced CHA from the SMA, in which they embolized the replaced CHA and performed PD with a right hepatic lobectomy, while preserving the collateral blood supply via the accessory left hepatic artery (Fig. 6b). In these previous reports, the aberrant hepatic arteries were embolized before being sacrificed; however, in the present case, all but the future preserved aberrant hepatic arteries were embolized

Table 1 Characteristics of the previous reports

First author	Year	Disease	Embolized artery	Collateral pathway	Type of pancreatic resection	Complications
Miyamoto [4]	2004	Pancreatic cancer	Replaced CHA	Left and right GA	PD	–
Sasaki [5]	2011	Bile duct cancer	Replaced CHA	Accessory LHA	PD with right hepatic lobectomy	–
Cloyd [6]	2012	Bile duct cancer	Replaced RHA	LHA	PD	–
		Pancreatic cancer	Replaced RHA	LHA	PD	Pancreatic fistula, bleeding

CHA common hepatic artery, *RHA* right hepatic artery, *GA* gastric artery, *LHA* left hepatic artery, *PD* pancreaticoduodenectomy

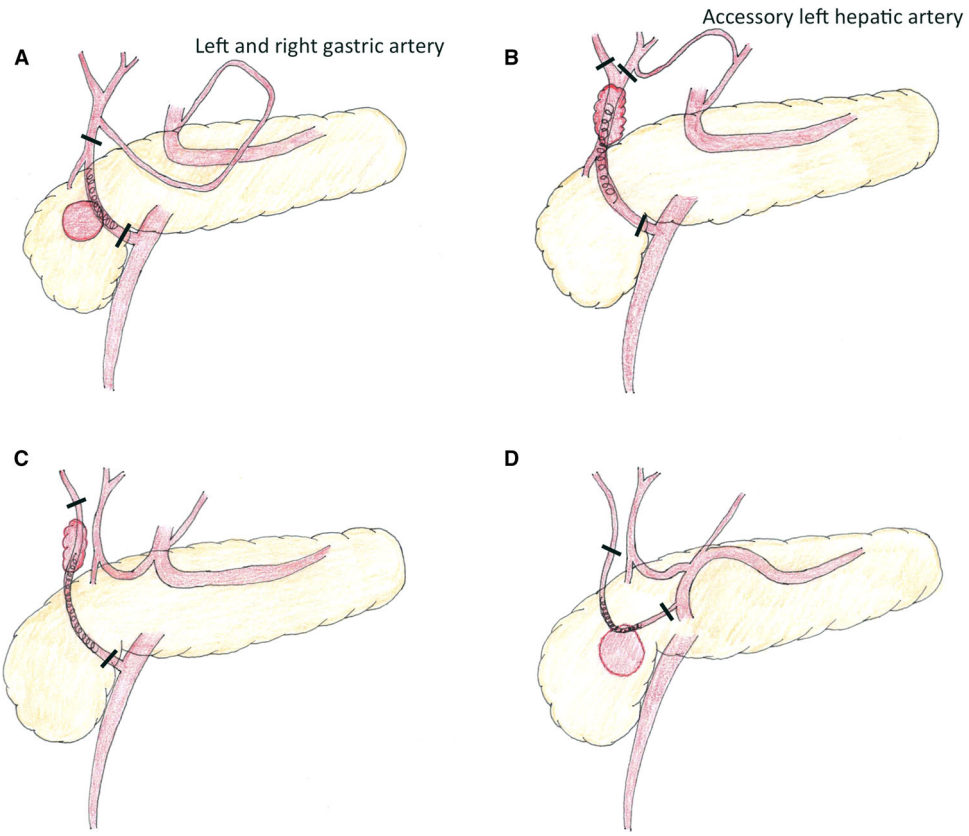

Fig. 6 Diagrams showing the embolized hepatic arteries and collateral pathways presented in the previous reports. **a** In the case reported by Miyamoto et al. [4], the replaced common hepatic artery from the superior mesenteric artery was embolized and resected, while the left and right gastric arteries were preserved as a collateral pathway. **b** In the case reported by Sasaki et al. [5], the replaced common hepatic artery from superior mesenteric artery was embolized and resected, while the accessory left hepatic artery was preserved as a collateral pathway. **c** In the first case reported by Cloyd et al. [6], the replaced right hepatic artery from the superior mesenteric artery was embolized and resected, while the left hepatic artery was preserved as a collateral pathway. **d** In the second case reported by Cloyd et al. [6], the replaced right hepatic artery that separately branched off from the origin of the celiac artery and ran behind the head of the pancreas was embolized and resected, while the left hepatic artery was preserved as a collateral pathway

and resected. To our knowledge, this is the first report documenting such a procedure.

Recent advances in molecular-targeted agents have demonstrated that everolimus, an inhibitor of mammalian target of rapamycin, and sunitinib, a multitargeted tyrosine kinase inhibitor, had antitumor activities in patients with advanced pNETs [14, 15]. However, the clinical significance of adjuvant therapy for pNETs has not been established yet, and thus, we did not administer adjuvant targeted therapy in the present patient.

In conclusion, we successfully performed PD accompanied by *en bloc* resection of tumor-encased hepatic arteries with preoperative embolization while preserving an aberrant RAHA for a pNET. As R0 resection was achieved, the symptoms derived from the gastrinoma resolved completely. The present surgical strategy should be considered for patients with relatively low-grade pancreatic malignancies involving the major hepatic arteries with the presence of an intact aberrant hepatic artery.

Conflict of interest The authors declare that they have no conflicts of interest to report.

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