

Distal Pancreatectomy for a Solid-Pseudopapillary Neoplasm of the Pancreas with the Preoperative Suspicion of Major Arterial Involvement: A Case Report

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Abstract

Background: Solid-pseudopapillary neoplasms (SPNs) of the pancreas are a relatively rare type of tumor with low-grade malignant potential. Although surgical resection provides a cure in most of cases, the safety and efficacy of surgical treatment for tumors with major arterial involvement remains unknown.

Case presentation: A 30-year-old man with a preoperative diagnosis of SPN was referred to our department. Abdominal computed tomography (CT) revealed a lobulated mass located at the pancreatic body, with a maximum diameter of 11 cm. The celiac artery (CA) and splenic artery (SA) were surrounded (by approximately 270° and 360°, respectively) by an expansively growing tumor, while the common hepatic artery (CHA), superior mesenteric artery (SMA) and superior mesenteric vein (SMV) were contacted by the tumor for a distance of 26.6, 42.8, and 43.5 mm, respectively on CT. The arterial walls of the CHA and SMA were smooth without any irregularity; however, narrowing and irregularity of the SA was present. The imaging findings strongly suggested an SPN of the pancreas; the involvement of the SA, CA, SV and SMV by the tumor was suspected. Distal pancreatectomy with celiac axis resection was planned. After laparotomy, the CHA, SMA, CA, and SMV were safely preserved, and the tumor was resected by distal pancreatectomy alone. A pathological examination confirmed that the tumor was an SPN with a malignant component. Although the tumor pathologically invaded the retropancreatic tissue, splenic artery and splenic vein, the resected margin was negative.

Conclusion: Although the preoperative examination delineated the gross expansion of an SPN around the CA, CHA, and PV, these vessels were safely preserved during pancreatectomy. This case suggests that the surgical resection of the SPN may occasionally be attempted; even in cases with compressed and deviated by the tumor.

Keywords: Solid-pseudopapillary neoplasm; Pancreas; Major arterial involvement; Major vessel involvement; Distal pancreatectomy

Abbreviations: SPN: Solid-Pseudopapillary Neoplasm; PV: Portal Vein; SMA: Superior Mesenteric Artery; CA: Celiac Artery; CHA: Common Hepatic Artery; DA: Descending Aorta; SA: Splenic Artery; CT: Computed Tomography; SV: Splenic Vein; SMV: Superior Mesenteric Vein; LRV: Left Renal Vein

Case Report

A 30-year-old man presented to another hospital with epigastric pain. He had a huge tumor of 11 cm in size that was located on the pancreatic body. Endoscopic ultrasound-guided fine-needle aspiration revealed the tumor was an SPN. He was referred to Shizuoka Cancer Center for treatment.

A physical examination revealed a huge mass in the left side of his upper abdomen with mild tenderness. Laboratory tests showed a normal hemoglobin level of 15.0 g/dl. Abdominal computed tomography (CT) revealed a large lobulated mass with maximum diameter of 11 cm at the pancreatic body. The use of contrast media revealed the heterogeneous enhancement of a hypodense, round, well-circumscribed tumor mass (Figures 1a-1c). Magnetic resonance imaging also showed a huge tumor and identified a mixture of solid and cystic components, and T2-weighted imaging showed a high-intensity area, which suggested that the tumor had a necrotic or cystic pathology. Positron emission tomography-CT only revealed a positive uptake (SUVmax 10.3) in the solid component of the tumor.

With regard to the relationship with the major vessels around tumor, approximately 270° of the CA and 360° of the SA were surrounded by the expansively growing tumor, which compressed and caused the deviation of the upper side (Figure 1a). The CHA and SMA were contacted for a distance of 26.6 mm and 42.8 mm, respectively

by the tumor and compressed and deviated to the upper right side of the abdomen (Figure 1b and 1c). The arterial walls of the CHA and SMA were smooth without any irregularity or constriction. On the other hand, narrowing and irregularity of the SA was observed on CT angiography (Figure 1d). The splenic vein (SV) was involved by the tumor, and apparently obstructed (Figure 1e). The superior mesenteric vein (SMV) was severely contacted for a distance of 43.5 mm. The SMV was deviated to the right side of the abdomen and was stretched and narrowed by the tumor. The DA and the left renal vein (LRV) were also broadly contacted for distances of 33.9 mm and 30.7 mm, respectively, by the tumor. There was no sign of distant metastasis or adjacent organ invasion. The portal collateral vessels had developed due to left-sided portal hypertension. The imaging findings strongly suggested a SPN of the pancreas, with the suspected involvement of the SA, CA, SV and SMV. Considering the expansive nature of SPN, we expected that the CHA, SMA, DA, and SMV would be able to be preserved.

Distal pancreatectomy with combined celiac axis resection was planned for the curative resection of this SPN, and combined resection of the PV may be necessary in some situations. No distant metastasis

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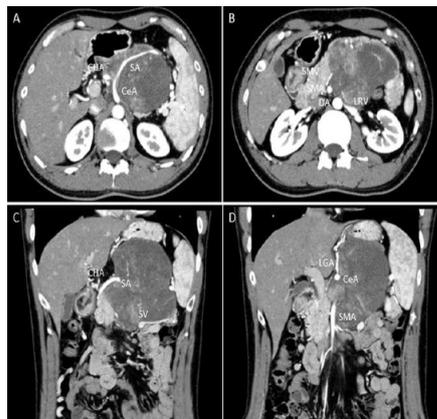


Figure 1: Abdominal computed tomography images in axial (a, f) and coronal (b, c) views show a huge lobulated mass of 11 cm in diameter, with a well-circumscribed capsule on the pancreatic body. **a** Approximately 270° of the celiac artery (CA) and 360° of the splenic artery (SA) were surrounded by the tumor. **b** The common hepatic artery (CHA) was contacted for a distance 26.6 mm by the tumor and compressed to the upper side of the abdomen (arrow). **c** The superior mesenteric artery (SMA) was contacted for 42.8 mm and was compressed to the right side (arrow). **d** CT angiography showed the narrowing and irregularity of the SA.

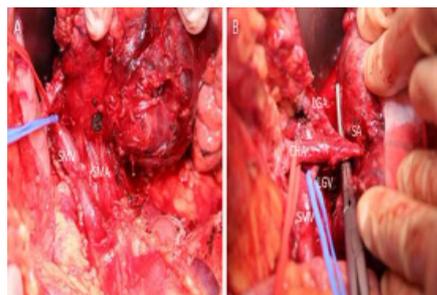


Figure 2: **a** The tumor was encapsulated by a thick wall. The CHA was encircled and tunnelling was performed at the pancreatic neck. **b** The origin of the SA was exposed, ligated and divided. The left gastric artery (LGA) and left gastric vein (LGV) were preserved.

was found after laparotomy. The tumor was encapsulated by a thick wall, and invasion to the SMA or DA was not found. The CHA was first encircled and dissected toward the celiac axis (Figure 2a). The origin of SA was not involved by the tumor. The SA was exposed, ligated and divided (Figure 2b). After cutting the pancreatic neck, it was found that the tumor did not involve the SMV or SMA. The tumor was dissected from these vessels. Furthermore, the DA and LRV were not involved by the tumor. These vessels were preserved. Finally, distal pancreatectomy preserving the left adrenal gland was performed without celiac axis resection or the combined resection of any other organs.

Macroscopically, the lobulated tumor had cystic and solid components with thick walls of fibrous tissue. The cystic component demonstrated hemorrhage and necrosis. The histological examination of the tumor revealed a pseudopapillary pattern with moderate nuclear atypia. Immunohistochemically, the tumor cells were found to be positive for β -catenin and CD10, which was consistent with the diagnosis of SPN. No lymph node metastasis was detected microscopically. The tumor was found to have directly invaded the retropancreatic tissue, the SV and SA. The resected margin, including the stumps of the SA and SV, was negative for tumor cells. Finally, the tumor was diagnosed as an SPN with a malignant component. Postoperatively, pancreatic fistula occurred, which was treated conservatively. He was discharged

on postoperative day 30. At 10 months after the operation, there have been no signs of tumor recurrence.

Discussion

The surgical resection of SPN is the only treatment that provides the chance of cure. SPNs have been subdivided by the WHO classification into: SPNs with borderline malignant potential, and solid-papillary carcinoma. Approximately 15% of SPNs have a malignant nature, which is evidenced by the development of metastasis or recurrence. It was reported that SPNs of more than 6 cm in diameter are more likely to display aggressive behavior, such as metastasis and recurrence [1-9]. Moreover, no cases of SPNs smaller than 6 cm were categorized as having capsular invasion in their group. On the other hand, Kang et al. [10] and Kim et al. [9] reported that a diameter of more than five centimeters is a significant feature for predicting SPNs and a finding that suggests malignant potential. It has been difficult to establish the criteria that suggest aggressive behavior, including the development of recurrence and metastasis [2,10].

In cases in which SPNs involving the SMV have been resected, the prognosis has been relatively good, with an acceptable survival rate. Kanai et al. [5] reported that pancreatoduodenectomy with PV resection was performed for an SPN with the involvement of the PV for a distance of approximately 8 cm; however, there was no direct pathological invasion of the PV by the tumor. In our case, preoperative CT revealed that the SPN involved the PV for a distance of 43.5 mm; however, we could easily detach the tumor from the PV. Wang et al. [6] reported the narrowing and irregularity of the PV, which was involved by the SPN, which indicated the pathological invasion of the PV. Meanwhile, pathological tumor invasion may be unlikely in cases in which there is a smooth margin between tumors and vessels. Even when the PV is compressed for a length of more than 40 mm, the tumor may be detached from the PV-in some cases-due to the basically low-grade malignant nature of SPN.

In our case, the tumor widely contacted the celiac axis and CHA for distances of 26.6 mm and 42.8 mm, respectively. We preoperatively planned distal pancreatectomy with combined celiac axis resection; however, these arteries were successfully preserved and curative resection was achieved by distal pancreatectomy alone. There are two possible reasons why the combined resection of major arteries could be avoided. Firstly, the SPN was well encapsulated by thick fibrous layers, despite it having a malignant component. Secondly, the neural plexus around the arteries provided another layer between the tumor and the arteries. Even if radiological imaging suggests that an SPN widely contacts the major arteries, it may be possible to avoid the combined resection of these arteries because it is difficult for the SPN to involve the major arteries due to the basically low-grade malignant nature of the tumor. It is true that the SPN pathologically invaded the SA in this case. In our case, the narrowing and irregularity of the peripheral part of the SA was observed in the preoperative CT imaging. This case suggests that the irregularity of major arteries involved by SPN will be the key finding to indicate the pathological invasion of these arteries and that the preservation of these arteries will be impossible. On the other hand, huge SPN may actually involve the major branches of the abdominal aorta. In the case involving major arteries, aggressive resection may cause severe complications after surgery. Surgeons should carefully consider the surgical indications to avoid misjudging SPNs as unresectable when an SPN widely contacts the major arteries.

In conclusion, we surgically treated a huge SPN of the pancreas. Even if the SPN widely contacts major arteries, it may be possible to

curatively resect the tumor with the preservation of the major arteries in cases of SPN that does not develop irregularity of the major arteries on preoperative CT.

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