

Case Series

Two Cases of a Portal Annular Pancreas in Patients Undergoing Pancreatoduodenectomy

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Keywords

Portal annular pancreas · Pancreatoduodenectomy

Abstract

Introduction: Portal annular pancreas (PAP) is a congenital anomaly resulting from aberrant fusion of the ventral and dorsal pancreatic buds around the portal vein (PV). PAP was classified into three types by Joseph et al., based on the location of the main pancreatic duct around the PV. The presence of PAP is important for the surgical procedure because it is associated with the postoperative pancreatic fistula. There are no standardized surgical procedures of resection and reconstruction for PAP. **Case Presentation:** We report 2 cases of subtotal stomach-preserving pancreatoduodenectomy in patients with PAP. One case of PAP was discovered coincidentally intraoperatively, and the other case was diagnosed before surgery. The first case was an 84-year-old male patient who underwent surgery for distal bile duct cancer. PAP was noticed intraoperatively when the uncinate process of the pancreas was detached from behind the PV. The second case was an 84-year-old female patient who also underwent surgery for distal bile duct cancer. We recognized PAP from preoperative computed tomography images. In both cases, the ductal anatomy was consistent with type IIIA PAP, and the dorsal pancreas was resected using a stapling device. During the postoperative period, there was no clinically relevant postoperative pancreatic fistula. **Conclusion:** PAP is rarely encountered intraoperatively; however, it is important to recognize it before surgery and take it into consideration when deciding upon the procedures for resection and reconstruction.

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Introduction

Portal annular pancreas (PAP) is a rare anomaly characterized by portal vein (PV) encasement in the pancreatic parenchyma resulting from aberrant fusion of the ventral and dorsal pancreatic buds [1]. There are no symptoms, and it is often found incidentally on computed tomography (CT) examination. The reported incidence ranges from 1.1% to 2.5% [2, 3]. The rarity of PAP means that it is sometimes overlooked on preoperative CT images, and surgeons may face challenges in dealing with this anomaly [4]. Surgery in patients with PAP is associated with higher rates of postoperative pancreatic fistula (POPF) and overall morbidity. The appropriate surgical strategy is yet to be established because of the complexity of varying main pancreatic duct (MPD) and two pancreatic resection surfaces. Anticipation of PAP when performing pancreaticoduodenectomy (PD) and adequate preoperative and intraoperative planning can help minimize postoperative complications [5, 6]. We present 2 patients with PAP who underwent subtotal stomach-preserving PD (SSPPD) (1 case was discovered incidentally intraoperatively, and the other was diagnosed preoperatively) and describe our successful management of PAP.

Case Presentation

Case 1

An 84-year-old man was referred to our hospital for investigation of liver dysfunction. Enhanced CT examination revealed a protruding tumor in a distal bile duct (Fig. 1a), and adenocarcinoma was detected from cytopathology using endoscopic retrograde cholangiopancreatography. We planned SSPPD with lymph node dissection for distal bile duct carcinoma. During surgery, the anteportal pancreas was transected just above the PV; however, the pancreas could not be separated from the uncinate process and the PV (Fig. 1b). We confirmed PAP from CT images (Fig. 1c). The retroportal pancreas was resected using a Signia stapler (Medtronic, Japan). The MPD was confirmed in the anteportal pancreas, and pancreaticojejunostomy was performed using the modified Blumgart method. Drains were placed on the Winslow hole, near the ventral and dorsal sides of the pancreatojejunostomy. The pathological specimen showed a hard, flat-infiltrative mass of 60 × 22 mm and revealed a moderately differentiated adenocarcinoma with negative resection margins. Based on the 8th Union for International Cancer Control (UICC) TNM staging, the pathological diagnosis was T1N0M0, stage I. The amylase level in the drain inserted into the dorsal side of the pancreatojejunostomy was 1,855, 302, and 41 IU/L on postoperative days 1, 3, and 5, respectively. The drain became chyle-like, but it recovered spontaneously and was removed on postoperative day 20. The patient was discharged on postoperative day 27 without clinically relevant POPF (CR-POPF).

Case 2

An 84-year-old woman was referred to our hospital for investigation of liver dysfunction. A low-density mass was found in a distal bile duct by CT examination (Fig. 2a), and adenocarcinoma was diagnosed by scraping cytology of the endoscopic retrograde cholangiopancreatography. After insertion of a self-expandable metallic stent, we planned SSPPD. PAP was recognized from CT (Fig. 2b), and magnetic resonance cholangiopancreatography (MRCP) (Fig. 2c) performed before bile drainage. Because we recognized the presence of PAP and the main or accessory pancreatic duct was not contained in retroportal pancreas, the anteportal pancreas was first dissected just above the PV. The dorsal pancreas was sufficiently dissected from the dorsal side of the PV and transected using the stapler (Fig. 2d). The

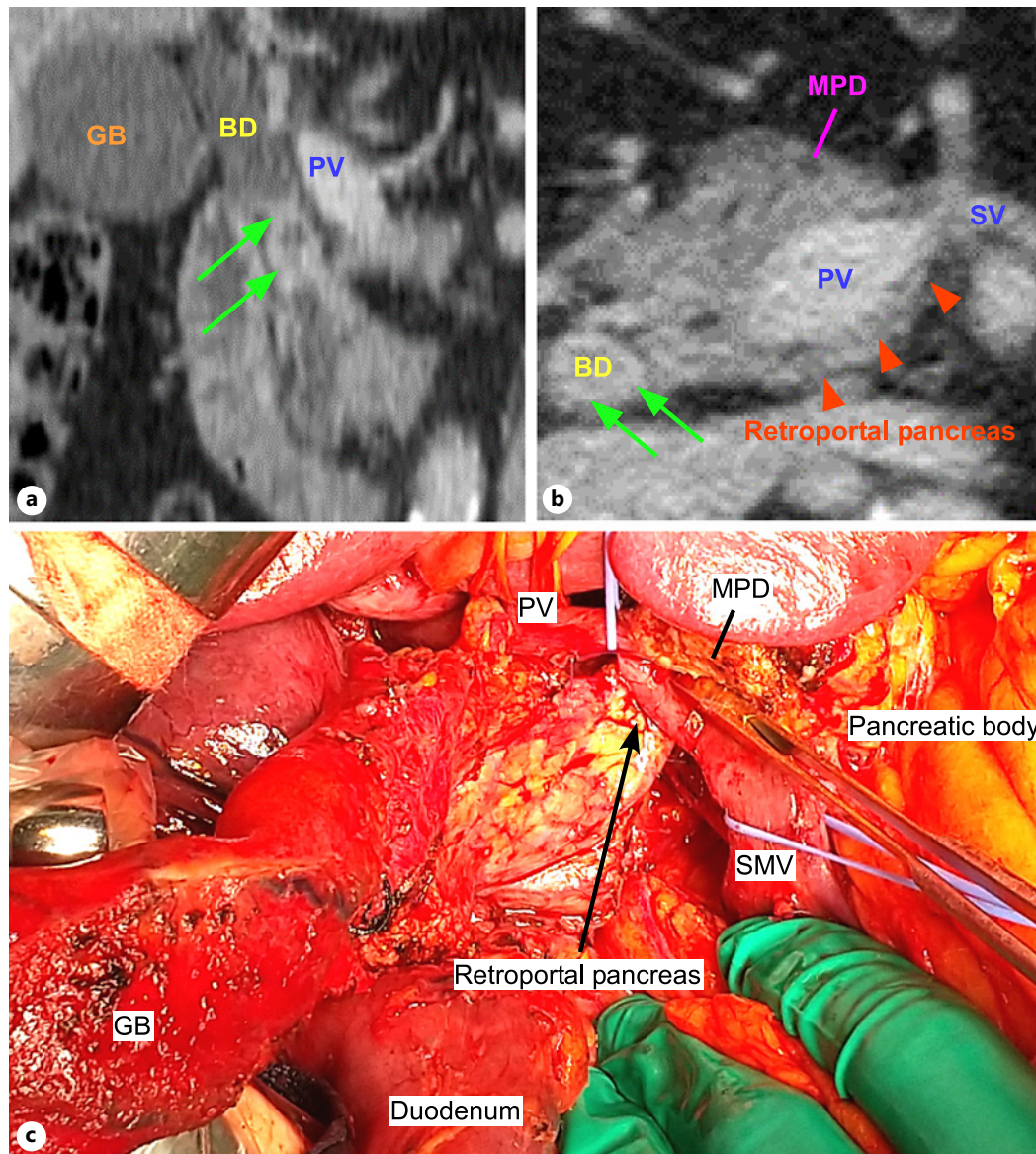
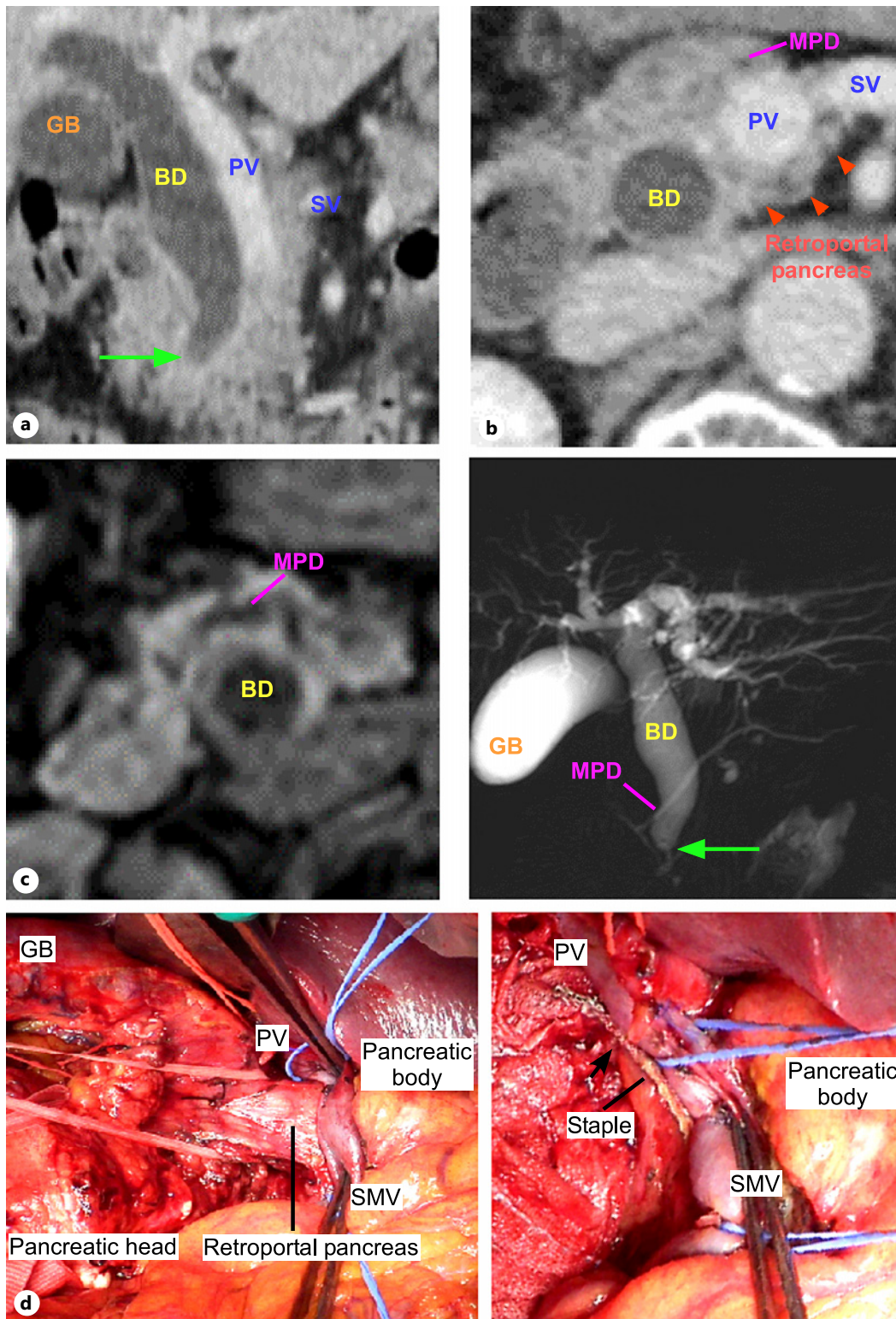


Fig. 1. CT images and intraoperative findings in case 1. **a** Axial view. CT imaging showed enhancement of the tumor in a distal BD (arrows). **b** Intraoperative finding of PAP after resection of the anteportal pancreas just above the PV. The retroportal pancreas could not be separated from the uncinate process. **c** Sagittal view. An existence of the PAP found from the CT image. BD, bile duct; CT, computed tomography; GB, gallbladder; MPD, main pancreatic duct; PAP, portal annular pancreas; PV, portal vein; SMV, superior mesenteric vein.

pathological specimen showed a hard flat mass of 70 × 25 mm and revealed a well-differentiated adenocarcinoma with invasion of the duodenum, with negative resection margins. Based on the 8th UICC TNM staging, the pathological diagnosis was pT2N1M0, stage IIB. The amylase level in the drain inserted into the dorsal side of the pancreaticojejunostomy was 2432, 923, and 898 IU/L on postoperative days 1, 3, and 5, respectively. The drain was removed on postoperative day 14. The patient had no complications and was discharged on postoperative day 27.



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(For legend see next page.)

Discussion

PAP was first reported in 1987 by Sugiura et al. [1] as a hypertrophic uncinated process that encircled the superior mesenteric vein. During embryonic development, the ventral pancreatic bud rotates posteriorly to fuse with the dorsal pancreatic primordium. The malformation is generated by abnormal fusion between the ventral and dorsal pancreatic buds. Variants were classified by Joseph et al. [7] according to the positional relationship between the MPD, PV, and splenic vein. In type I, the ventral bud of the pancreas fuses with the body and the MPD runs through the retroportal pancreatic tissue. In type II, type I pancreatic divisum is present and the MPD runs through the retroportal pancreas. In type III, the uncinata process alone fuses with the pancreatic body and the MPD runs through the anteportal pancreatic tissue. The positional relationship between PAP, PV, and MPD can be recognized by CT or magnetic resonance imaging. It is crucial to preoperatively identify this anomaly to assess the ductal anatomy better, preferably by MRCP [4]. Although the presence of PAP is widely known, it is sometimes overlooked before surgery [8]. Preoperative recognition of PAP is important for well-planned surgery and avoidance of potential complications [9, 10].

The most commonly reported adverse outcome associated with PAP is POPF after PD [10]. PD generates two cut pancreatic surfaces; therefore, the position of resection, resection method, and reconstruction method must be considered. At present, an appropriate surgical strategy is not defined according to the PAP types [11]. When the MPD runs through the retroportal pancreas (type I) or the pancreatic duct runs through both sides of the PV (type II), extended resection seems to be beneficial. The anastomosis part becomes one surface, so it can be considered a normal anastomosis. The incidence of CR-POPF decreased from 71% to 16% when a single pancreatic stump was obtained by extended resection [12–14]. However, deterioration of the remnant pancreatic functions is assumed. In the case of type III PAP, as in our case, there is no standardized method of resection of the dorsal pancreas [12].

At our institution, in total, 162 cases of pancreatic surgery were performed between January 2013 and December 2022, of which only 2 cases had PAP. The incident rate is 1.2%, and it is coincident with the reports [2, 3]. Two cases of PAP were diagnosed as type III PAP (one case was preoperatively diagnosed, and the other case was intraoperatively diagnosed) by using CT and MRCP images. We opted to use a stapling system because the retroportal pancreatic tissue was thin. This method was similar to the pancreatic resection method used for distal pancreatectomy (DP), and most of the distal pancreas could be preserved. The operation was successful, and there was no CR-POPF. Postoperative remnant pancreatic function was maintained, and no symptoms such as diarrhea, malnutrition, or worsening of diabetes were observed. Although it took some time for them to regain physical strength through rehabilitation, they were discharged without any complications.

In cases of type III PAP, suturing, stapling, and extended resection are used for resection of the dorsal pancreas, but there are few reports of this [13, 14]. These reports indicated a

Fig. 2. CT images and intraoperative findings in case 2. **a** Axial view. CT imaging showed common bile duct dilation and indicated a tumor in a distal BD (arrow). **b** Sagittal view. PAP was recognized from the CT image before surgery (arrowhead). **c** MRCP imaging of the T2 fat-suppressed image (left) and maximum intensity projection image (right). The MPD ran through the anteportal pancreas, and no pancreatic duct passed through the retroportal pancreas. **d** Left panel shows intraoperative finding of PAP after resection of the anteportal pancreas just above the PV. The right panel shows the stump of the retroportal pancreas after resection by stapling system. BD, bile duct; CT, computed tomography; MPD, main pancreatic duct; MRCP, magnetic resonance cholangiopancreatography; PAP, portal annular pancreas; PV, portal vein.

lower rate of CR-POPF with suturing (12.5%) as opposed to stapling (71.4%). There were no differences in primary tumor or surgical method (PD or DP) in these patients. Predicted clinically relevant fistula rates based on intraoperative findings of cases 1 and 2 were 8.7% and 13.8%, respectively [15]. It is thought that POPF did not arise from the retroportal pancreas because the pancreatic stump was sufficiently closed with staples. Recently, a new type of a triple-row stapler reinforced with preloaded bioabsorbable polyglycolic acid felt has been widely used for DP. The results showed a decrease in the incidence of POPF compared with the standard stapler [16, 17]. Advances in devices might have contributed to reduce the incident rate of CR-POPF in our cases.

The surgical strategy differs depending on the anatomical type of PAP; therefore, it is important to consider the method before surgery. We prepared our case report following the CARE Guidelines [18], where the completed CARE checklist has been uploaded as online supplementary material (for all online suppl. material, see <https://doi.org/10.1159/000538370>).

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Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from both the patients for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors declare no conflict of interests.

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Author Contributions

All the authors contributed to the concept of this case report. Mineto Ohta drafted the manuscript. Mineto Ohta, Keisuke Fukushima, Hiroyasu Nishimaki, Takayuki Morita, and Yuta Ito performed the surgery and conducted postoperative management. Keisuke Fukushima and Kenji Namiki supervised the writing of the report.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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