



Case report

Resection of a recurrent solid pseudopapillary neoplasm of the pancreas after duodenal sparing pancreaticoduodenectomy: A case report

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ABSTRACT

Introduction: Solid Pseudopapillary Neoplasm (SPN) is a rare pancreatic neoplasm with low malignant potential and a relative indolent course. Complete resection of the SPN is curative for most cases and has a high survival rate. Recurrences, though rare, can still occur despite adequate resection. Pancreaticoduodenectomy is commonly performed to treat pancreatic head SPNs. In recent years, duodenum-preserving pancreatic head resection (DPPHR) has been reported as a less radical and acceptable alternative.

Case presentation: We are reporting a case of 26-year old female who presented with a 7 month history of epigastric pain and increasing abdominal girth. She was diagnosed by MRI to have a huge but resectable pancreatic head mass and subsequently underwent duodenum-preserving pancreatic head resection (DPPHR) with pancreaticojejunostomy. Histopathologic examination revealed a solid pseudopapillary tumor (SPN) with lymphovascular invasion and negative margins of resection. The patient underwent hepaticojejunostomy 5 months after resection for biliary stricture. Surveillance imaging revealed tumor recurrence warranting re-exploration for recurrence 3 years after the initial surgery. Intraoperative findings revealed the mass at the distal pancreatic remnant, requiring distal pancreatectomy and splenectomy.

Discussion: Solid pseudopapillary neoplasms are rare pancreatic neoplasms. Surgical resection of SPNs affords long term cure with good 5-year survival rates for localized tumors. Despite the low malignant potential of SPNs, relapse after resection can still occur.

Conclusion: Complete local resection of the tumor is the treatment of choice in SPNs. DPPHR should be considered as an alternative in young patients with a localized SPN in the pancreatic head.

1. Introduction

Solid pseudopapillary neoplasm (SPN) of the pancreas was first described by Frantz [1] in 1959, predominantly affecting young women with a mean age of 24 years, with a 10:1 female predominance [2]. SPN has a rare occurrence and only comprises 1–2% of exocrine pancreatic tumors, and around 5% of cystic pancreatic tumors [3].

Clinically, SPN manifest as slow growing abdominal masses which may be asymptomatic or present with nonspecific abdominal pain. Histologically, these are solid tumors with pseudopapillary and pseudocystic components with rich microvasculature [4]. The origin of these tumors is still unknown and is a matter of controversy [5].

SPNs are classified as low grade neoplasms and thus usually have a favorable prognosis. En bloc resection (R0) is considered as curative

management. The majority of resected SPN patients have a 5-year survival rate of approximately 95% for tumors confined in the pancreas [7]. Due to the indolent nature of this disease, recurrences are very rare. Although SPNs are classified as low-grade neoplasms, they still have the potential for metastasis. Distant spread, when present, is usually confined to the liver and/or peritoneum [5–7].

Due to the low incidence of SPN, there are still no established recommendations regarding the extent of resection of the primary neoplasm and the management of recurrences [8,9]. We are reporting a case of a pancreatic head solid pseudopapillary neoplasm initially treated with duodenal-preserving pancreatic head resection presenting with a recurrence on the pancreatic tail one year post operatively. This case is being presented for the rarity of the condition and the complexity in the management it posed both for the initial tumor and the

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recurrence. This is being reported in accordance with the SCARE 2020 guidelines [10].

2. Case summary

A 26-year old female initially presented with a 7 month history of burning epigastric pain and reflux symptoms. Four months prior to admission, the patient then presented with increasing abdominal girth. There was no history of weight loss, vomiting, jaundice, or bowel movement changes. She had no previous surgeries nor co-morbidities. There is no history of malignancy within her immediate family. Physical examination revealed normal stable vital signs, no jaundice, with abdomen nondistended, soft and non-tender with a palpable right hemiabdominal mass. Subsequent Magnetic Resonance Imaging (MRI) of the Abdomen (Fig. 1) showed a large heterogenous, well circumscribed mass measuring $10 \times 11.3 \times 10.8$ cm (AP x T x CC) predominantly demonstrating T1 hyperintense and T2 mildly hyperintense signal centrally with peripheral areas of T1 hypointensity and restricted diffusion which is consistent with findings of mixed solid components, highly proteinaceous fluid or subacute blood products, and internal septations. The primary consideration for the MRI was that of a pancreatic head solid pseudopapillary neoplasm. The mass was located at the right subhepatic space and was displacing the second and third segments of the duodenum with no signs of obstructions. The common bile duct draped along the superior margin of the mass, with the distal segment and ampullary insertion was not visualized. The main portal vein and superior mesenteric vein also draped along the periphery of the mass with no infiltration. There was also a large tubular cystic focus with highly proteinaceous fluid or subacute blood components intraluminally measuring 3 cm in diameter and 12 cm in length seen along the expected location of the pancreatic body and tail likely a dilated pancreatic duct. CA 19-9 of the patient at that time was elevated to 84.45 u/ml with serum amylase, lipase and other laboratory results unremarkable.

The patient underwent elective laparotomy with findings of a large firm, well circumscribed, cystic mass with hemorrhagic and necrotic regions (Fig. 2) noted at the head of the pancreas with no infiltration of duodenum or the adjacent structures. The mass however was intimately located to the common bile duct, superior mesenteric vein and its branches, as well as the mesentery of the transverse colon and displaces said structures. Surgery then proceeded with duodenum-preserving pancreatic head resection with approximately 70% of the pancreas preserved. A roux-en Y pancreatico-jejunosotomy was performed (Fig. 2) to maintain the continuity of the proximal and distal pancreatic duct to the gastrointestinal tract. The opening of the pancreatic duct (proximal) to the duodenum was ligated, leaving the distal CBD intact in the

common Channel as it opens in to the papilla of Vater.

Histopathology (Fig. 3) showed evidence of a malignant round cell neoplasm with lymphovascular invasion possibly (1) a solid pseudopapillary tumor, (2) an acinar cell carcinoma, or (3) a pancreatic neuroendocrine tumor. Immunohistochemical stains (Fig. 4) revealed negative staining for chromogranin, low Ki-67 staining (5%), and positive staining for progesterone receptors, vimentin, beta catenin, synaptophysin, and cytokeratin which confirmed diagnosis of solid pseudopapillary neoplasm. All margins of resection were free of tumor.

Patient was discharged stable but was readmitted due to episodes of cholangitis secondary to a benign common bile duct stricture 4 ½ months after the first surgery. The patient then underwent immediate percutaneous transhepatic biliary drainage with resolution of cholangitis. Five months after the first surgery, the patient underwent roux-en Y hepaticojejunostomy with cholecystectomy. In this procedure, the roux limb of the established roux-en Y pancreaticojejunostomy was used to construct the hepaticojejunostomy. The stricture was documented intraoperatively as benign.

The patient was sent home improved and was unremarkable until one year postoperatively, on surveillance MRCP (Fig. 5) noted a 0.7×1.4 cm irregular soft tissue thickening in the left anterior pararenal space with tumor implant cannot totally be ruled out.

Close observation was advised. Repeat MRCP the following year (2 years postoperatively) showed increase in size of soft tissue thickening now with measurement of $1.7 \times 1.6 \times 1.6$ cm (AP/T/CC). Three years postoperatively, the patient still has no associated symptoms but follow up MRCP (Fig. 6A) showed further increase in size of irregular enhancing soft tissue lesion involving the left anterior pararenal space now measuring $2.7 \times 2.8 \times 2.9$ cm (AP/T/CC). The rest of the findings were unchanged.

The patient underwent laparotomy. Intraoperatively, there a firm, irregularly shaped mass (Fig. 6B) measuring $3 \times 1.5 \times 1$ cm located at the distal pancreas and is intimately related to the transverse colon splenic flexure, spleen and left anterior kidney. There were noted dense adhesions surrounding the mass at the area of the splenic flexure, transverse colon omentum and left Gerota's fascia. Surgery then proceeded with distal pancreatectomy, resecting approximately 20% of the pancreas and leaving behind around 50% of the original pancreatic tissues (pancreatic body and proximal tail). Histopathology again revealed positive for round cell neoplasm compatible with solid pseudopapillary neoplasm (recurrence) of the pancreas. Patient's post-operative course was generally unremarkable and was discharged without complications.

Four months post operatively, patient had a 1 kg weight gain, is non-insulin requiring and has a normal HBA1c (5.6). She also has completed

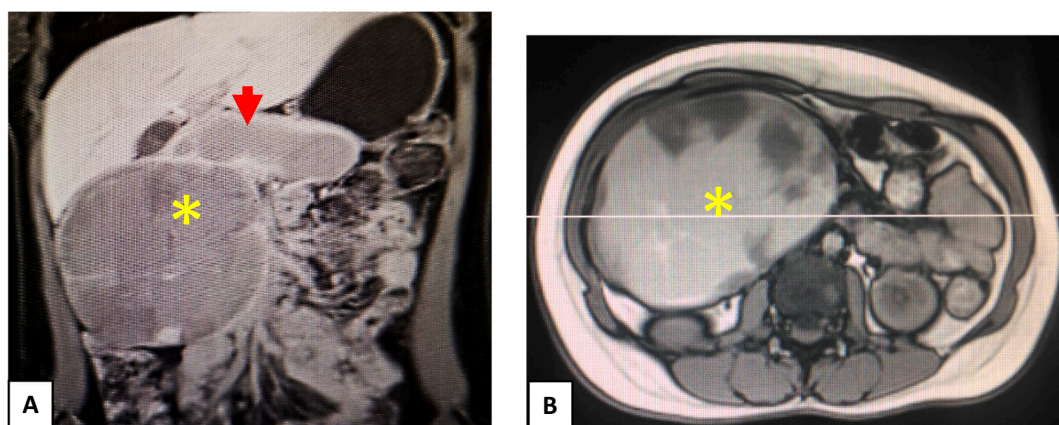


Fig. 1. MRI with Liver Specific Contrast (A) coronal and (B) axial views showing a $10 \times 11.3 \times 10.8$ cm pancreatic head mass (yellow asterisk) with a 3×12 cm tubular cystic focus (red arrow) located superior to the mass likely the pancreatic duct. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

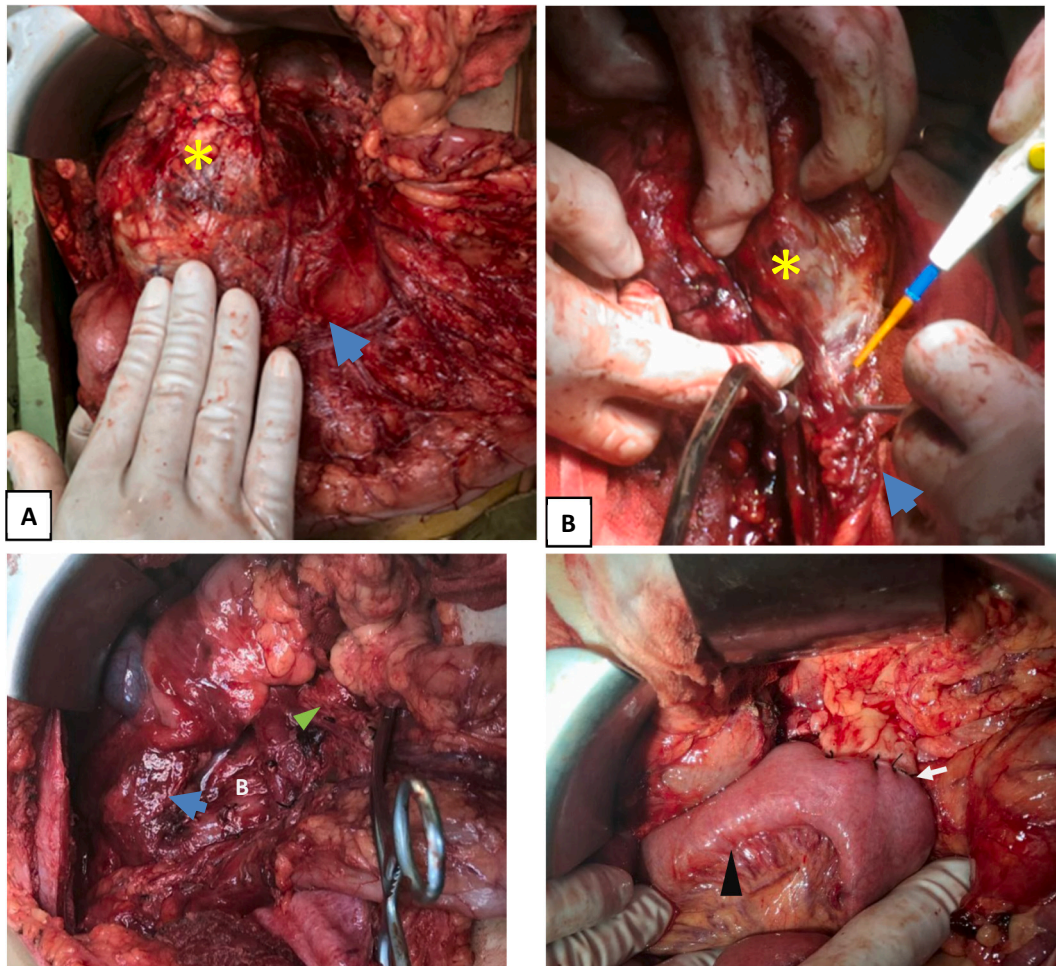


Fig. 2. (A) & (B): A 12 × 12 × 10 cm pancreatic head mass (yellow asterisk) displacing surrounding structures with no duodenal (blue arrow) infiltration. (C): The tumor bed (white “B”) post removal of the pancreatic head mass with the duodenum (blue arrow) and the distal pancreas (green arrowhead) preserved. D: Pancreaticojejunostomy (white arrow) with the roux limb indicated by black arrowhead. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

her post splenectomy vaccination with Hib, Pneumococcal and Meningococcal vaccines. She verbalized her gratitude for the successful management of her case despite the complexity of the surgery.

3. Discussion

Solid pseudopapillary neoplasm is a rare pancreatic tumor that accounts for 2–3% of primary pancreatic malignancies, found mainly in women during their second and third decades of life [11].

Some patients are asymptomatic with the tumor only detected via imaging or routine physical examination. Our patient presented with increasing abdominal girth and abdominal pain which are some of the nonspecific symptoms of SPN. Other symptoms include abdominal fullness, nausea and vomiting if the tumor compresses on local intestinal structures [7]. There is usually no evidence of pancreatic insufficiency, cholestasis nor of elevated pancreatic enzymes. Tumor markers for SPNs usually show values within normal limits [12]. This patient also did not exhibit any biochemical abnormalities which is consistent with the findings of other authors.

Imaging studies can be used to diagnose SPNs. Abdominal ultrasound and CT scan usually show a well encapsulated, complex mass with solid and cystic components. Peripheral calcifications and intravenous contrast enhancement within the mass can also be seen which would suggest hemorrhagic necrosis [13]. CT scan however has an inferior contrast resolution compared with MRI. Furthermore, compared with

MRI, CT has limitations in showing tissue characteristics specific for SPNs such as hemorrhage, cystic degeneration and presence of capsule. SPNs on MRI exhibit high signal intensity areas consistent with hemorrhagic substance and thin hypo-intense rims which are identified as the tumor capsule [14]. These MRI findings were seen in our patient.

Since preoperative diagnosis is difficult because of the similarities of SPNs to other cystic lesions, some authors advocate endosonographic guided fine-needle aspiration biopsy of the tumor. This procedure however is controversial due to the possibility of tumor spread and the low yield in diagnosis [15]. The surgical team elected to forego biopsy of the tumor since the MRI findings were already diagnostic for SPN.

Eighty five to 90% of primary tumors are localized to the pancreas at the time of diagnosis and most are localized to the tail [12]. This is in contrast to our patient where the initial presentation of the mass is at the head of the pancreas.

SPNs have a low grade of malignancy and are usually encapsulated thus R0 local resection is the therapy of choice. For tumors in the body and tail of the pancreas, distal pancreatectomy with or without splenic preservation can be performed. On the other hand, for tumors located in the pancreatic head, pancreatoduodenectomy has been a common procedure of choice [16]. This procedure however is considered to be too radical and less aggressive procedures such as duodenum preserving pancreatic head resection (DPPHR) has been proposed as an alternative for pancreatic head SPNs as long as there is no compromise in oncologic radicality. DPPHR spares the pylorus, duodenum and extrahepatic bile

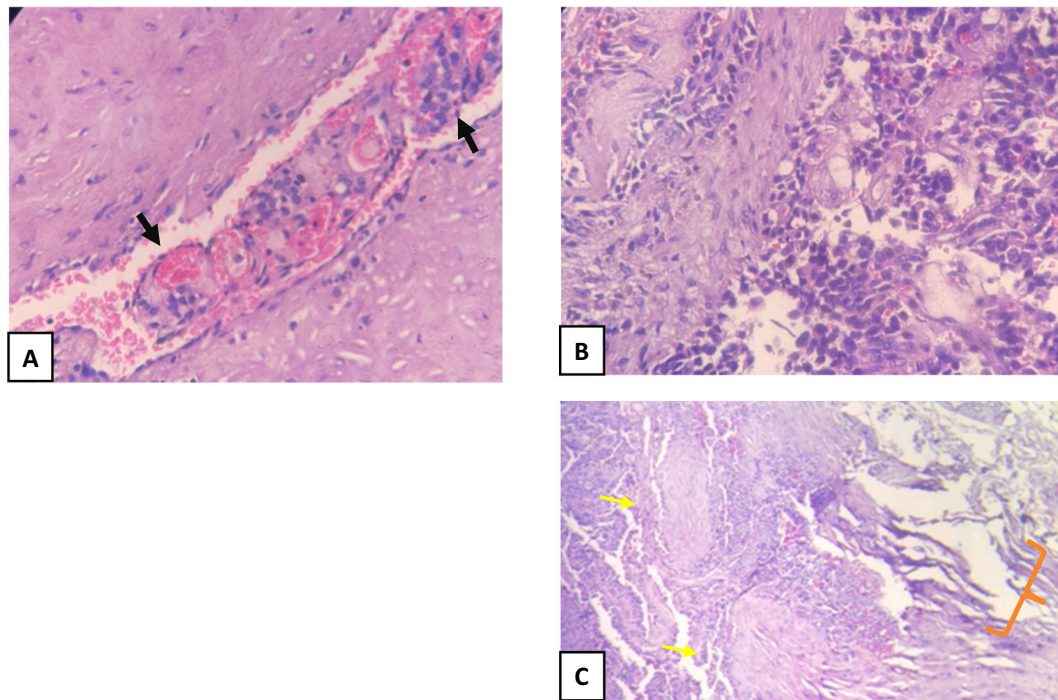


Fig. 3. A, B, C & D: Hematoxylin and Eosin stain. Representative section of the pancreatic head mass with (A) lymphovascular invasion of tumor cells (black arrows), (B) solid sheets of tumor cells oriented around fibrous tissues. (C) Tumor cells (yellow arrows) adjacent to the tumor capsule (orange bracket). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

duct which reduces complications and preserves pancreatic exocrine and endocrine functions. Lymph node metastasis rarely occurs for SPNs thus regional lymphadenectomy is not routinely recommended [11]. The surgical team elected to perform DPPHR due to the localized nature of the disease and to avoid performing a radical procedure on a young patient.

This patient developed a benign common bile duct stricture after her first surgery which required hepaticojejunostomy to restore biliary continuity. Bile duct stricture along with pancreatic fistula and duodenal stricture have been noted complications of DPPHR for benign and low-grade malignant tumors [17]. Despite these documented complications, authors still advocate for DPPHR as a suitable alternative to traditional surgery [17,18]. In a study by Buchler et al., DPPHR fares favorably compared to Whipple operation in postoperative complications, glucose tolerance, postoperative pain and quality of life 6 months post operation in the treatment of chronic pancreatitis [18]. It should be noted that despite the patient undergoing both DPPHR and distal pancreatectomy, her pancreatic endocrine and exocrine functions were preserved which is consistent with the benefits associated with DPPHR.

Surgical resection of SPNs affords long term cure with a reported 5-year survival rate of 95% for tumors confined in the pancreas [7]. Despite the low malignant potential of SPNs, relapse after resection can still occur with a risk of recurrence of around 3–9% [19]. Factors that increase the risk of recurrences include tumor size >5 cm, lymphovascular invasion, lymph node metastasis, synchronous metastasis and positive margin [20]. SPNs usually displace surrounding structures rather than invade them thus recurrences are mostly resectable [19]. Treatment for recurrent disease is still complete resection of tumor [20,21]. Currently, there is still limited data regarding the role of chemotherapy and radiotherapy in the management of SPNs [21].

For this patient, the risk factors that predisposed this patient for recurrence are large tumor size and lymphovascular invasion. Tumor recurrence was managed with distal pancreatectomy with splenectomy since the tumor is located at the tail of the pancreas.

Intraoperatively, the findings of the patient were consistent with the

typical gross appearance of SPNs which is a well circumscribed mass with areas of hemorrhage, necrosis and cystic degeneration. Microscopic examination of this patient showed sheets of uniform tumor cells with eosinophilic cytoplasm surrounding fibrous and vascular stalks which is consistent with the hallmark histologic pattern of SPNs [12]. Initial histopathologic exam of this patient also revealed findings of malignant round cell neoplasm which has a similar histological pattern with neuroendocrine tumors and acinar cell carcinomas. Immunohistochemical studies can be used to distinguish between the different pancreatic tumors [22]. In studies, SPNs in general are shown to stain positive for vimentin, β -catenin, and progesterone receptor [22,23]. Immunohistochemical staining for chromogranin is usually negative in SPNs and synaptophysin is only detectable in a small percentage [21]. Furthermore, Ki-67 labelling of cells is scarcely evident in SPN sections [23]. All of these are evident in the patient thus confirming the diagnosis of solid pseudopapillary neoplasm.

4. Conclusion

Solid pseudopapillary neoplasm is a rare pancreatic neoplasm. Complete local resection of the tumor is the treatment of choice. Duodenum preserving pancreatic head resection should be considered as an alternative in young patients with a localized non-invasive SPN in the pancreatic head provided that there no compromise in oncologic radicality. Despite its low malignant potential, recurrence can still occur and complete resection of tumor recurrence is the ideal management. Large tumor size (>5 cm in diameter) and lymphovascular invasion are notable risk factors for SPN recurrence. Due to its similar presentation with other pancreatic tumors, immunohistochemical staining is used to confirm diagnosis.

Provenance and peer review

Not commissioned, externally peer-reviewed.

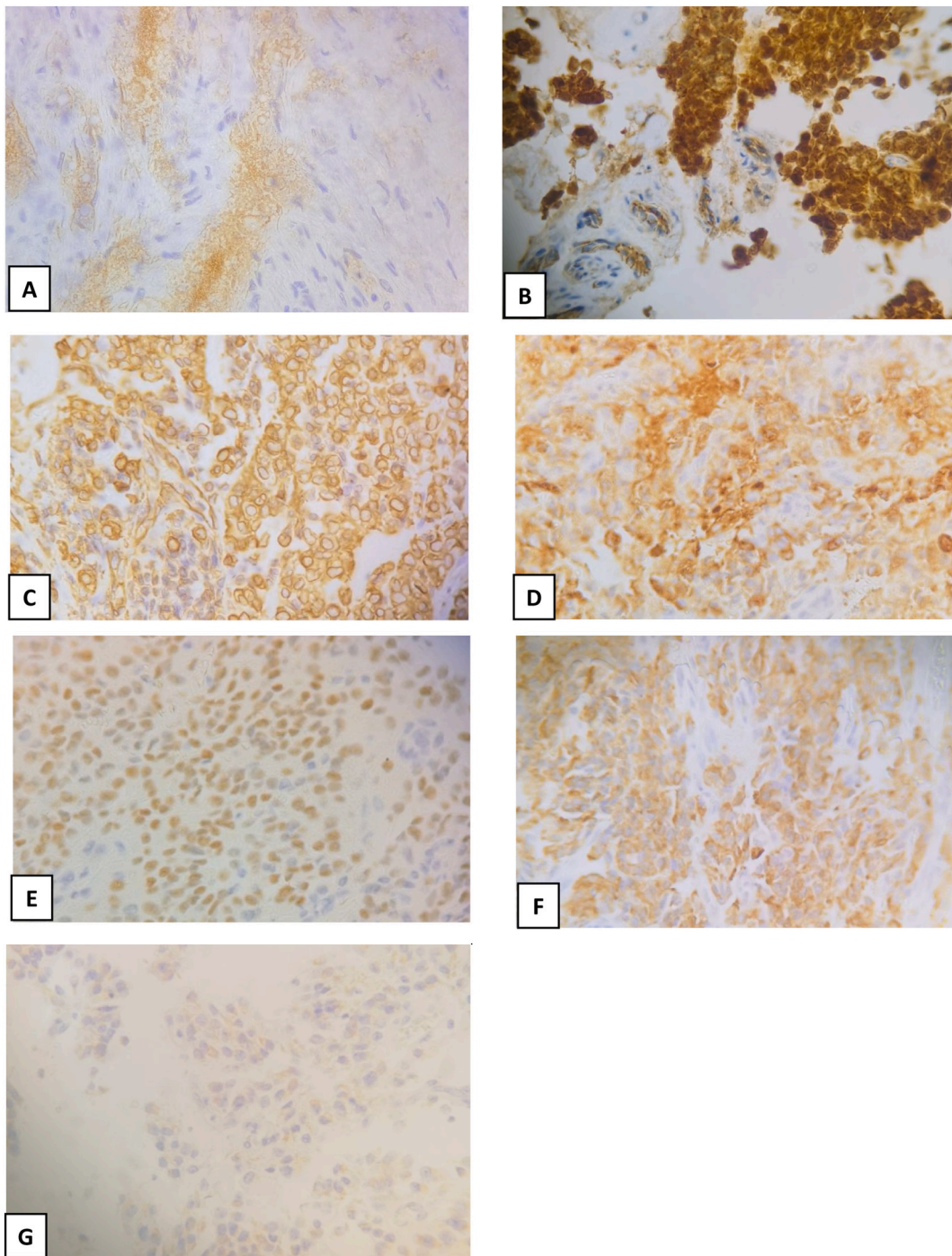


Fig. 4. Immunohistochemical staining of representative sections of pancreatic head mass 40 \times . (A) very low Ki-67 score. Positive staining for (B) β -catenin, (C) Vimentin, (D) Synaptophysin, (E) Progesterone receptor and (F) cytokeratin. Negative staining for (G) chromogranin.

Consent

Written informed consent was obtained from the patients next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Guarantor

Anthony R. Perez, MD, MHA.

Ethical approval

Ethics approval obtained from University of the Philippines Ethics Review Board.

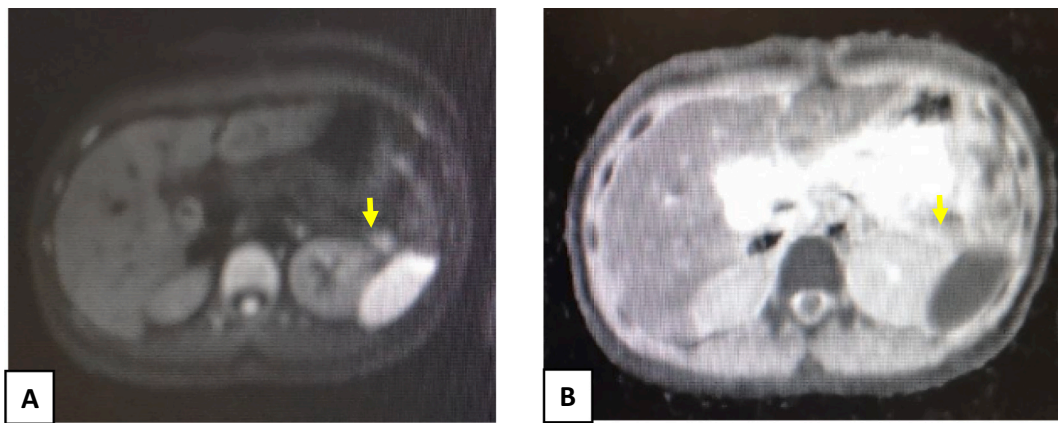


Fig. 5. MRI with MRCP axial views (A) TRACE and (B) ADC (apparent diffusion coefficient) diffuse weighted images showing a 0.7×1.4 cm soft tissue thickening (yellow arrow) at the left anterior pararenal space. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

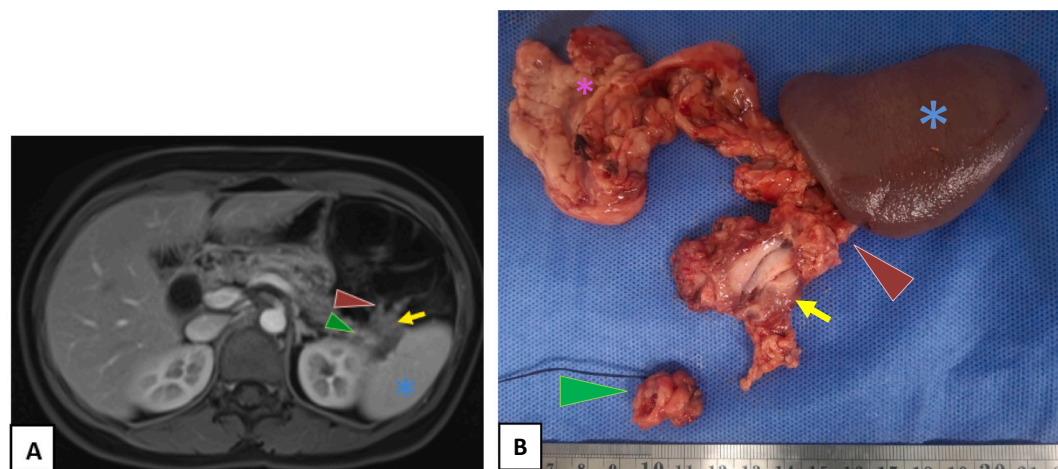


Fig. 6. (A) MRI with MRCP axial view and (B) Intraoperative findings revealing a distal pancreatic mass (yellow arrow) which is closely adherent to the spleen (blue asterisk), transverse colon omentum (pink asterisk) and segment of the splenicocolic ligament (red arrow head) with invasion of the anterior Gerota's Fascia and perirenal fat (green arrowhead). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Registration of research studies

Not applicable.

CRedit authorship contribution statement

Anthony Perez, MD- writing the paper, final editing.
 Crisostomo Arcilla Jr. MD: Data collection, study design, manuscript editing.
 Maria Raisa Katrina Fontanilla, MD_- Data collection, review of literature, writing the paper.
 Apolinario Ericson Berberabe, MD: Data collection, review of literature, final draft.

Declaration of competing interest

There were no conflicts of interest.

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