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PANCREATOBLASTOMA, PREVAILING IN ALL AGE GROUPS: A CASE REPORT.

Arnetta Naomi Louise LALISANG[1,2] , SO Jeong Yoon[3], IN Woong Han[3], JIN Seok Heo[3], Nathaniel Jason ZACHARIA[2], Davin Nathan WIJAYA[2].

[1]Digestive Surgery Division, Department of Surgery, Cipto Mangunkusumo Hospital, Jakarta, Indonesia.
[2]Faculty of Medicine Universitas Indonesia, Cipto Mangunkusumo Hospital, Jakarta, Indonesia.
[3]Division of Hepatobiliary-Pancreatic Surgery, Department of Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, Republic of Korea.

ABSTRACT

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Pancreatoblastoma is an exceptionally rare tumor that primarily affects the young population, with even fewer cases reported in adults. The rarity of this disease often leads to underdiagnosis and misdiagnosis, resulting in delayed treatment and a worsened prognosis. We report two cases of pancreatoblastoma, one in a child and one in an adult, to highlight the challenges in reaching a definitive diagnosis prior to surgery despite extensive laboratory and imaging examinations. This is to raise awareness and improve understanding of pancreatoblastoma, facilitating earlier diagnosis and prompt treatment for better patient outcomes.

Key Words: **Adult, Chemotherapy, Pancreatoblastoma, Pancreatic Neoplasm, Pediatrics.**

INTRODUCTION

Pancreatoblastoma is an extremely rare tumor disease, primarily affecting infants and young population, accounting for less than 1% of all pancreatic tumors and 0.5% of all pancreatic non-endocrine tumors.[1,2] The occurrence rate is reported to be 0.004 cases per 100,000 persons per year.[3] Its rarity in adults led to its previous designation as infantile carcinoma of the pancreas before 1977.[4,5]

Due to its rare occurrence, especially in adults, pancreatoblastoma is often underdiagnosed and misdiagnosed as other pancreatic tumors, further contributing to delayed treatment. Although pancreatoblastoma is less aggressive in children, local invasion, recurrence, and metastasis are still common.[5] Meanwhile, in adults, this tumor exhibits a highly aggressive behavior, worsening the prognosis.[3]

We report two cases of pancreatoblastoma, one in a 4-year-old child and another in a 64-year-old adult. In both cases, the diagnosis of pancreatoblastoma could not be confirmed preoperatively with laboratory tests nor imaging studies. The definitive diagnosis could be made only by intraoperative histopathological examination.

Corresponding author: Arnetta Naomi Louise Lalisang, Jl. Pangeran Dipenogoro No. 71, Jakarta, Postal Code: 10430, Indonesia.
Telephone number: (+62)8119590124; Email: arnetta.naomi01@ui.ac.id

CASE REPORT

Case 1

A male patient, aged 4 years and 10 month, presented with complaints of a mass in the upper abdominal area accompanied by abdominal pain and vomiting occurring every two weeks since last month. The patient denied experiencing diarrhea, constipation, hematemesis, or melena. Physical examination revealed a mass in the left upper quadrant (LUQ) with tenderness. Bowel sound was within normal limit.

Abdominal ultrasound (USG) revealed a cystic and solid tumor in the LUQ with a suspected diagnosis of solid pseudopapillary tumor in the pancreatic tail, with a differential diagnosis of cystadenoma or cystadenocarcinoma. Contrast-enhanced magnetic resonance cholangiopancreatography (MRCP) also showed a large lobulated solid and cystic mass in the pancreatic tail, most likely representing a solid pseudopapillary neoplasm of the pancreas. A small amount of fluid was observed in the perisplenic and perihepatic spaces, indicating a focal rupture, without evidence of distant metastasis. Abdominal CT scan confirmed the presence of a large lobulated solid and cystic mass with egg-shell calcification in the pancreatic tail. It also revealed a small amount of fluid in the perisplenic space and pelvic cavity, suggestive of focal rupture of the tumor, with no demonstrable distant metastasis (Figure 1). Laboratory examination found elevated alpha-fetoprotein, neuron-specific enolase, lactate dehydrogenase, and C-reactive protein.

The patient underwent distal pancreatectomy, splenectomy, and segmental resection of the colon one month after presentation. Segmental resection of the colon was carried out due to the mass invading the transverse mesocolon and splenectomy was performed due to the proximity of the anastomosis to the spleen.

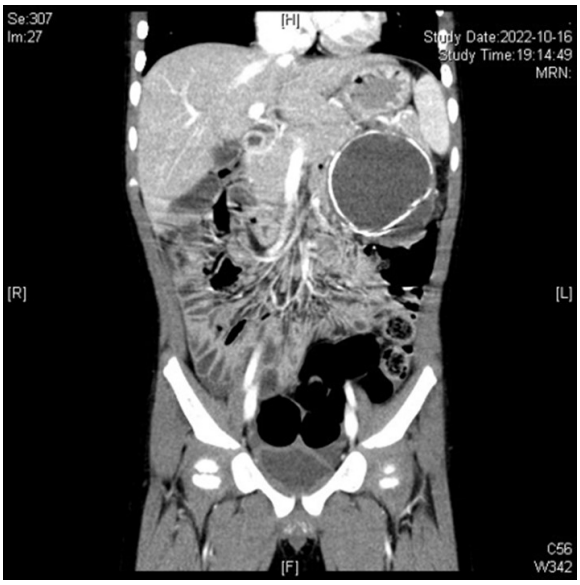

Figure 1. Abdominal CT scan of Case 1 showed a large lobulated mass in the pancreatic tail. **Click on image to enlarge)**

The excised tumor measured 9.5 cm x 6.9 cm (Figure 2). Histopathological examination revealed a diagnosis of stage pT3 pancreatoblastoma, with no involvement of the spleen, superior mesenteric vein, or portal vein. There was no evidence of regional lymph node metastasis or neural invasion. The resection was complete, with grossly and microscopically negative resection margins (R0).

Following the surgery, the patient was hospitalized for 8 days and was discharged in a stable condition. The patient received amoxicillin prophylaxis and vaccinations, including Menveo, PPSV23, and influenza vaccines. He continued with outpatient care. Subsequently, the patient was readmitted several times to undergo chemotherapy with CC321P2 (cisplatin) and ICE (Ifosfamide, Carboplatin, Etoposide) regimens. The patient's condition remained stable, and to date, six months after surgery, the patient continues regular follow-up in the outpatient setting.

Case 2

A 64 years old male presented with jaundice, epigastric discomfort, and nausea for the past

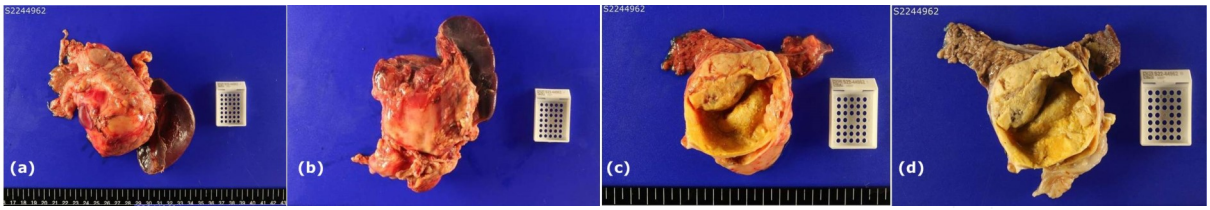

Figure 2: (a-c) The excised tumor of Case 1, above: measured 9.5 cm x 6.9 cm, below: sectioned tumor mass, and (d) Gross pathology of the tumor of Case 1. (Click on image to enlarge)

2 weeks. The patient also experienced decreased appetite and a weight loss of 24 kg over the past 4 months. Physical examination revealed epigastric tenderness with no palpable mass and normal bowel sounds.

CT scan revealed a well-defined hypervascular mass measuring 5.8 cm with a necrotic/cystic portion in the pancreatic head, which was suspected to be a pancreatic neuroendocrine tumor with a differential diagnosis of solid pseudopapillary tumor and gastrointestinal stromal tumor. Diffuse dilation of the biliary tree with obstructive cholangiohepatitis was also observed, along with a possible focal pancreatitis at the tail of the pancreas. Borderline-sized lymph nodes were seen in the hepatoduodenal, paraaortic, and left gastric areas.

ERBD (endoscopic retrograde biliary drainage) showed external compression of the second part of the duodenum, so biliary sphincterectomy (BST) and bile duct cannulation were done for decompression. EUS-FNA (endoscopic ultrasound-guided and fine-needle aspiration) was performed and revealed a large mass with relatively clear borders in the head of the pancreas, accompanied by cystic fluid and blood. Biopsy results indicated a solid pseudopapillary neoplasm of the pancreas.

PET scan showed a hypermetabolic mass involving the head of the pancreas, probably pancreatic cancer, with no evidence of regional lymph node or distant metastasis. It raised suspicion of a solid pseudopapillary neoplasm with low-grade malignancy. Mild heterogeneous uptake in the liver was also observed, possibly due to cholangiohepatitis. Laboratory examination found elevated leukocyte as well as CRP and procalcitonin. Immunohistochemisty staining showed positive for CK7,cytokeratin AE1/AE3, EMA, Ki-67; and negative result for P63, CEA, and S-100 protein.

Two months after his admission to the hospital, the patient underwent pylorus-preserving pancreatoduodenectomy. Bluish appearance of the liver was observed, probably due to biliary obstruction, along with severe inflammation around the common bile duct (CBD) and pancreas due to mass obstruction, and pus discharge from the gallbladder. A mass measuring 7.6 cm was present in the pancreatic head (Figure 3). The tumour was excised completedly with clear margin visually and microscopically. Duct-to-mucosa end-to-side pancreatojejunostomy with type II reconstruction was performed.

Histopathologic examination confirmed the diagnosis of pancreatoblastoma with the staging of pT3 (size 6.7 cm x 6.4 cm; Figure 4), N2 (regional lymph node metastasis $\geq$ 4 with extranodal extension), accompanied by lymphovascular, neural, and perineural invasion. Immunohistochemistry examination showed diffuse positive staining for MLH1 (80%) and MSH2 (60%).

The patient was hospitalized for 7 days and discharged in stable condition, with

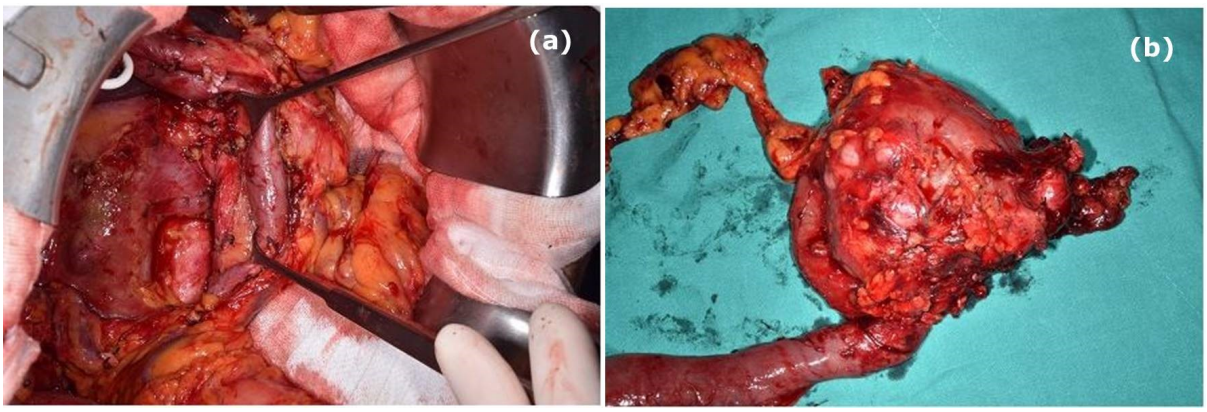

Figure 3: (a) Operation field of Case 2 after the tumor was removed showed margin free of tumor macroscopically. (b) The excised tumor of Case 2 ex-vivo. (Click on image to enlarge)

regular outpatient follow-up. Two months later, a contrast-enhanced CT scan of the pancreas showed no tumor recurrence. The patient then underwent adjuvant chemotherapy with FOLFIRINOX.

DISCUSSION

As an extremely rare disease, with approximately 0.004 cases per 100,000 persons per year, pancreatoblastoma (PB) is commonly found in the pediatric population, with a mean age of 4 years.[3,6.] Only 25% of them are adult PB, accounting for more than 80 cases reported with a median age of 41, including this case report.[6] Clinicians and radiologists often experience delay or misdiagnoses based on preoperative findings due to diverse, non-specific, and similar clinical manifestations of PB in comparison to other pancreatic tumors. Despite PB being a slow-growing tumor, it is nonetheless an aggressive tumor that exhibits local invasion and systemic metastasis. Diagnostic challenges may delay the initiation of proper surgical treatment, thereby reducing the survival rates of patients.[7,8]

There are no significant differences in characteristics between adult and child PB.[9] In our case, the pediatric patient presented with abdominal pain and a mass, while the adult patient presented with abdominal discomfort and jaundice. Generally, adult PB patients presented with abdominal pain (41-44%) as the most frequent symptom, followed by weight loss (29-32%), obstructive jaundice (22-26%), abdominal mass (17-21%), and other digestive system manifestations such as diarrhea, vomiting, GI bleeding.[7,10,11] On the other hand, child patients typically present with abdominal masses (47.6%) and abdominal pain (44.4%), followed by ab-

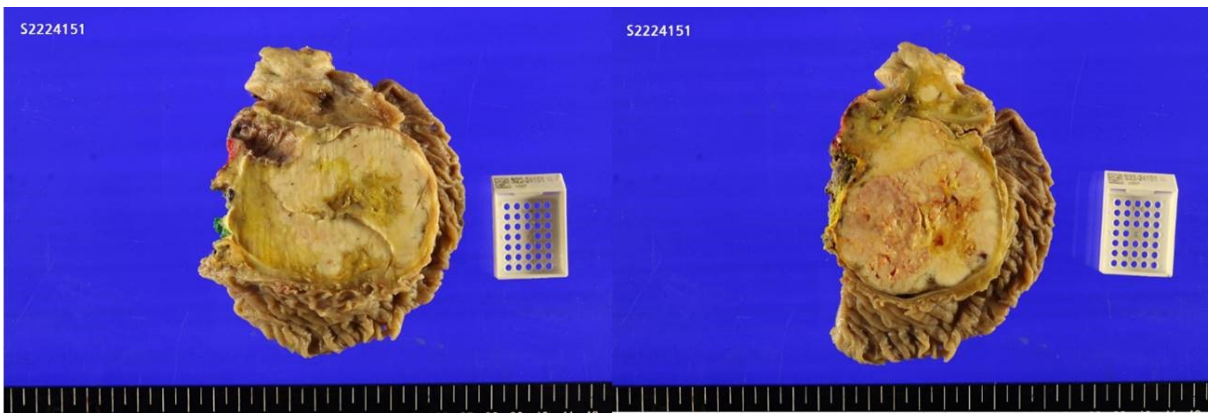

Figure 4: Gross pathology of the tumor of Case 2 showing solid tissue throughout the tumour, measured 6.7 cm x 6.4 cm, . (Click on image to enlarge)

as diarrhea, vomiting, GI bleeding.[7,10,11] However, child patients typically present with abdominal masses (47.6%) and abdominal pain (44.4%), followed by abdominal distension, vomiting, anorexia, fever, and less commonly failure to thrive (1.5-5%).[2] These symptoms usually manifest when the mass has reached a considerable size, often leading to delayed diagnosis.[3] The most common tumor location is in the head of the pancreas (48.4%) for adult patients and in the body-tail, head-body -tail, and head-body of the pancreas (30%) for pediatric patients.[1,7,10] However, PB can be found in any part of the pancreas at any age, as in our cases, the adult case in the head and the pediatric case in the tail.

No studies have reported specific PB laboratory findings in pediatrics or adults, and tumor markers are often not helpful. In some rare cases, adult PB may cause elevated serum levels of CA19-9, CEA, or alpha-fetoprotein (AFP). On the other hand, AFP and CEA serum levels are frequently elevated in pediatric PB, often exceeding 1000 µg/L.[6,10] An elevated serum AFP can be used as a tumor recurrence marker or to monitor disease progression, as AFP levels should be within the normal range when treatment succeeds. However, it is important to understand that AFP is not specific to PB cases, as hepatoblastoma, pancreatic ductal adenocarcinomas, and pancreatic acinar cell carcinomas can also cause elevated AFP serum levels.[1,6] Theere may be a slight increase in AST/ALT, GGT, as well as conjugated and total bilirubin when there is an extrahepatic obstruction.

A definitive diagnosis of PB must always be made after a thorough histological examination characterized by heterogeneous cellularity with acinar differentiation, squamoid nests, and foci of ductal, squamous, and endocrine cells.[2,12,13] Preoperative diagnosis is difficult because of the tumor's cellular heterogeneity with multiple cellular lines of differentiation.[14] However, imaging modalities such as ultrasound, CT, and MRI could sometimes be useful to demonstrate the radiologic characteristic of PB.[2,15] No studies have reported differences between adult and pediatric PB regarding their imaging characteristics, as they are usually characterized by a solid, cystic, large, well-defined mass (may appear blurry in adults) with partially circumscribed and lobulated margins without a cleavage plane. Pancreatoblastoma has typical characteristics on imaging as a mixed echogenicity solid mass on ultrasound, which manifests as a mass with low attenuation multiloculated on CT and a mass with low to intermediate signal intensity on T1-weighted and hyperintensity on T2 -weighted MRI images. The contrast-enhanced examination will show a typical enhancement in arterial and portal venous phases with a late washout. At the same time, MRI better delineates intratumoral hemorrhage and necrotic areas. Calcifications are more clearly found with CT as small, punctate, clustered, or rim-like Ca^{2+}.[14]

Sometimes, diagnosis of PB is challenging because it is difficult to distinguish squamoid corpuscles from typical histologic findings. This condition means that preoperative cytology, such as fine needle aspiration biopsy, is not helpful due to some sampling alternation.[7] On the other hand, establishing a differential diagnosis is as important as making a definitive diagnosis to treat and correctly predict the patient's prognosis. Pancreatoblastoma has some differential diagnoses through imaging findings such as pancreatic acinar cell carcinomas, solid-pseudopapillary neoplasms, neuroendocrine tumors, intraductal papillary mucinous neoplasm, mucinous cystic neoplasm, serous cystadenoma, pseudocyst, and ductal adenocarcinoma.[6,7,14] Therefore, a definitive diagnosis of PB can only be made by the presence of squamous cell nests on pathological examination.[13] Immunohistochemical examination may help to a certain extent with positive findings of AFP, B72.3, CEA, DuPan-2, EMA,

cytokeratins, trypsin, and chymotrypsin.[9,14]

There are no widely applied standard guidelines for pancreatoblastoma, except for the management recommendations published by Glick et al. (2012) for child and young adult PB.[2] Whenever anatomically possible, complete surgical resection is the only curative treatment unless distant metastases necessitate neoadjuvant therapy.[2,9,12,14,15] Systemic neoadjuvant chemotherapy may be indicated in PB patients with large tumors involving major blood vessels or surrounding organs and metastatic findings.[6] Following chemotherapy and radiotherapy after complete resection may also play a role in recurrent, residual, or metastatic cases.[6,15] However, the optimal chemotherapy regimen remains controversial and subject to discussion.[9,12] The most commonly recommended chemotherapy agents for PB are a combination of cisplatin (80 mg/m2 as a continuous 24-hour intravenous infusion) and doxorubicin (60mg/m2 over 48 hours) regimens. These regimens have shown positive results in reducing the tumor size and have been beneficial in cases of vascular invasion with additions of ifosfamide and etoposide.[2] In adults with advanced PB cases, oxaliplatin-containing chemotherapy may be a reasonable option, as it benefits for overall survival rates although does not have an impact on surgical aspects.[16] The effectiveness of radiotherapy is not fully known but it is suggested only for some unresected cases, incomplete resection, positive margins, or instances of tumor spillage.[2]

Based on the limited reported data, the adult population has a worse prognosis than younger cases with frequent local tissue infiltration and metastasis, as found in our adult PB case report.[6,10,12,14,15] Pancreatoblastoma may invade surrounding structures such as the portal vein, mesenteric vessels, common bile duct, nerves, spleen, duodenum, and colon. Distant metastases frequently happen with the liver being the most affected organ, followed by lymph nodes, lungs, bones, and peritoneum.[14,15] In our adult PB cases, lung metastasis was observed nine months after completed resection and the patient underwent FOLFIRINOX chemotherapy. Around 42% of adult patients died to the disease, with a mean interval of 27 months (0.8-348 months) after chemoradiotherapy and surgery.[6,13]Conversely, some patients showed no evidence of disease recurrence and were alive with disease, accounting for 38% and 16%, respectively.[6] Adult PB patients with unresected tumors had a reported median survival of 5 months.[14]

Our case report provided additional data on the imaging findings of PB and highlights the similarities between patients at extreme age differences, a child (4 years 10 months) and an elderly (64 years) individual. We reported our successful management of PB patients with adjacent organ invasion through a complete radical resection followed by chemotherapy regiments (Cisplatin, Ifosfamide, Carboplatin, and Etoposide). Our child patient has remained stable and continues regular follow up six months after surgery. On the other hand, our adult patient who underwent complete surgical resection and received chemotherapy (FOLFIRINOX), experienced lung metastases after a nine-months period.

CONCLUSION

Pancreatoblastoma, a rare disease, can occur primarily in pediatric and some adult populations. A definitive diagnosis can only be made by intraoperative histopathological examination, which means laboratory and imaging modalities only help to determine the clinical and management preoperatively. Complete surgical resection is the best option for operable cases, followed by chemotherapy regimens using cisplatin and doxorubicin to improve the treatment outcome. Neoadjuvant

chemotherapy plays a role in increasing the operability of unresectable cases. All patients must be monitored routinely after surgery to recognize residual tumors, recurrence, or metastasis.

DISCLOSURE STATEMENT

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REFERENCES

1: Liu T, Zhao T, Shi C, Chen L. Pancreatoblastoma in children. Clinical management and literature review. Transl Oncol. 2022; 18: 101359.
2: Glick RD, Pashankar FD, Pappo A, LaQuaglia MP. Management of pancreatoblastoma in children and young adults. J Pediatr Hematol Oncol. 2012; 34: S47-50.
3: Thera R, Souied O, Ahmed S, Hagel K, Chalchal H, Iqbal, M, Angeles R. Pancreatoblastoma in adulthood: Case report and literature review. Int Case Rep Jour; 2022; 2(7): 1-11.
4: Savastano S, d'Amore ESG, Zuccarotto D, Banzato O, Beghetto M, Famengo B. Pancreatoblastoma in an adult patient: A case report. J Pancreas. 2009; 10(2): 192-5.
5: Argon A, Celik A, Oniz H, Ozok G, Barbet FY. Pancreatoblastoma, a rare childhood tumor: A case report. Turk Patoloji Derg. 2017; 33:164-7.
6: Omiyale AO. Adult pancreatoblastoma: current concepts in pathology. World J Gastroenterol. 2021;27(26):4172–81.
7: Chen M, Zhang H, Hu Y, Liu K, Deng Y, Yu Y, et al. Adult pancreatoblastoma: a case report and clinicopathological review of the literature. Clin Imaging; 2018;50:324–9.
8: Chisholm KM, Hsu CH, Kim MJJ, Rangaswami A, Gray Hazard FK. Congenital pancreatoblastoma. J Pediatr Hematol Oncol; 2012;34 (4):310–5.
9: Li J, Peng C, Fan X, Wang L, Wang J.. Adult pancreatoblastoma: a case report. The J Int Med Res. 2021;49(8):03000605211039565.
10: Terino M, Plotkin E, Karagozian R. Pancreatoblastoma: an atypical presentation and a literature review. J Gastrointest Cancer; 2017;49 (3): 361–4.
11: Salman B, Brat G, Yoon YS, Hruban RH, Singhi AD, Fishman EK, et al. The diagnosis and surgical treatment of pancreatoblastoma in adults: a case series and review of the literature. J Gastrointest Surg. 2013;17(12):2153–61.
12: Morrissey G, Cohen P, Julve M. Rare case of Adult Pancreatoblastoma. BMJ Case Rep. 2020;13(4).
13: Wu M, Lin J, Liu Z, Huang Z, Wang R. CT, MRI, and 18F-FDG PET/CT imaging features of seven cases of Adult Pancreatoblastoma. BMC Med Imaging. 2022;22(1).
14: Vilaverde F, Scigliano H, Rodrigues P, Carvalho A, Reis A. Adult Pancreatoblastoma - case report and review of literature. J Radiol Case Rep. 2016;10(8).
15: Huang Y, Yang W, Hu J, Zhu Z, Qin H, Han W, et al. Diagnosis and treatment of Pancreatoblastoma in children: A retrospective study in a single pediatric center. Pediatr Surg Int. 2019;35(11):1231–8.
16: Berger AK, Mughal SS, Allgäuer M, Springfeld C, Hackert T, Weber TF, et al. Metastatic adult pancreatoblastoma: Multimodal treatment and molecular characterization of a very rare disease. Pancreatology. 2020;20(3):425–32.