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Extrapancreatic Primary Retroperitoneal Solid Pseudopapillary Neoplasm: A Case Report

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Solid pseudopapillary neoplasm (SPN) is a rare pancreatic neoplasm. This case reports a 63-year-old female previously diagnosed with bilateral adrenocortical carcinoma post resection who presents with persistent epigastric pain. Suspicions for recurrence prompted diagnostics revealing retropancreatic and left suprarenal foci, both suspicious for malignancy. Resection of both tumors yielded a moderately to poorly differentiated retropancreatic carcinoma with differentials not limited to recurrence and pancreatic neuroendocrine tumor with a left suprarenal lymph node. Immunohistochemistry was then done and was consistent with SPN having a strong expression for β-catenin, vimentin, pancytokeratin. It was non-specific to melan-A and negative for chromogranin A, synaptophysin, inhibin-α and CD10. At 6 months' follow up, patient is clinically well and abdominal CT scan showed no recurrence. SPNs can masquerade as other neoplasms having similar clinical, radiologic and histopathologic features. Immunohistochemistry thus plays a crucial role for accurate diagnosis and management. Surgical resection still remains the treatment of choice and can provide a 95% overall survival rate, while limited evidence supports the use of adjuvant chemotherapy or radiation.

Key words: Solid pseudopapillary neoplasm, extrapancreatic, immunohistochemistry

Solid pseudopapillary neoplasm (SPN) of the pancreas is a rare pancreatic neoplasm that accounts for only 0.1% to 3% of all pancreatic exocrine tumors.[1] Predominantly, patients are young women with a mean presentation age of 35 years.[2] Numerous well-documented cases of SPNs have been published in the English literature and only a few on extrapancreatic SPNs. These tumors have been found anatomically separate from the pancreas, most commonly in the ovarian[3] and testicular/paratesticular areas.[4] Some of these cases have occurred in the context of an ectopic pancreas.[1] The present case details a primary extrapancreatic solid pseudopapillary neoplasm of the retroperitoneum albeit on a patient previously diagnosed with bilateral adrenocortical carcinoma.

The Case

This is the case of a 63-year-old female previously diagnosed with bilateral adrenocortical carcinoma. Ten years prior to presentation, she underwent left radical nephrectomy with en bloc distal pancreatectomy, splenectomy, and right partial nephrectomy. Interval history was unremarkable until eight months prior to consult when she presented with recurrent epigastric pain and bloatedness. She denied any vomiting, febrile episodes, chills, weight loss, abdominal distention or changes in bowel habits. She was treated as a case of acid related disorder; however, proton pump inhibitors afforded no relief. She was also diagnosed with hypertension and diabetes mellitus, which were controlled with maintenance medications. Physical examination at presentation was unremarkable. Vital signs were stable. Patient had no pallor, jaundice or any palpable lymphadenopathies. Heart and lungs findings were unremarkable The abdomen was flabby with a midline laparotomy scar, non-distended, normoactive bowel sounds, tympanitic, soft and non-tender with no palpable mass. Patient had no bipedal edema. Basic laboratory workup revealed a normal complete blood count and creatinine.

Given her medical history of bilateral adrenocortical carcinoma, there was a high suspicion for recurrent

disease. An abdominal computed tomography scan revealed a large, well-defined, round focus at the retropancreatic area measuring 4.6 cm x 3.9 cm x 3.8 cm with heterogenous enhancement on contrast study. There was minimal compression of the pancreas and surrounding vessels anterosuperiorly (Figure 1). Another well-defined enhancing nodule was noted in the left suprarenal area measuring 2.4 cm x 1.2 cm x 1.3 cm. The distal part of the pancreas was not visualized. The pancreatic duct was not dilated. The rest of the pancreas was unremarkable with homogenous parenchyma.

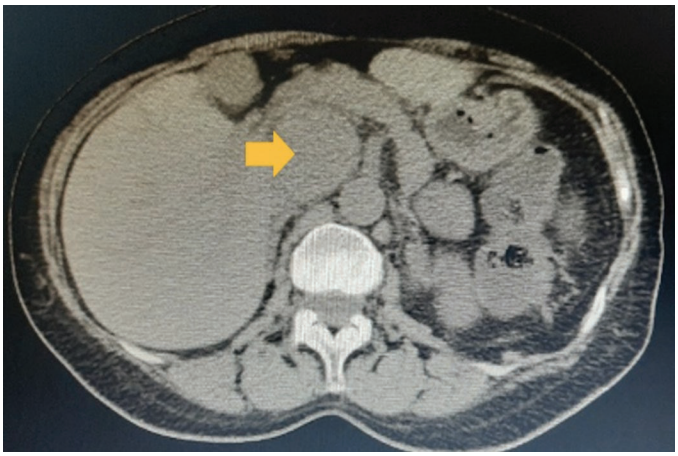

Figure 1. Plain abdominal CT scan showing a well-defined, round focus at the retropancreatic area (arrow) with minimal compression of the pancreas and surrounding vessels anterosuperiorly.

About 12 days after the CT scan, a positron emission tomography (PET) scan demonstrated a round, well-defined retropancreatic focus with intensely avid fluorodeoxyglucose (FDG) uptake approximately measuring 4.8 cm x 4.4 cm x 4.6 cm (Figure 2). This focus was mildly compressing on the pancreatic head and surrounding vessels anterosuperiorly. There was also a well-defined, enhancing, left suprarenal nodule approximately measuring 2.8 cm x 1.2 cm x 2.6 cm, but FDG activity was low. The spleen, adrenals, left kidney and distal pancreas were surgically absent. Post-surgical changes were likewise evident in the right kidney which was otherwise unremarkable. The intensely hypermetabolic retropancreatic density was consistent with recurrent disease and the left suprarenal density was possibly indicative of metastatic disease.

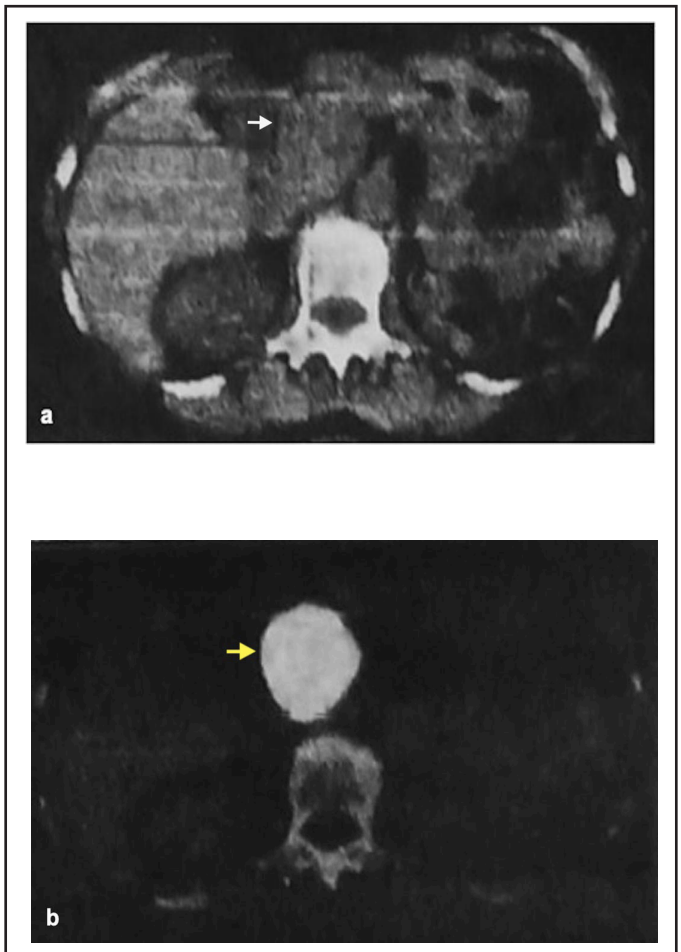

Figure 2. Positron emission tomography scan revealed a well-defined retropancreatic round focus (arrow) measuring around 4.8cm x 4.4cm x 4.6cm that was mildly compressing on the pancreatic head a nnd surrounding vessels (a). The focus is noted to be intensely FDG-avid consistent with recurrent disease (b).

The patient underwent exploratory laparotomy. Intraoperatively, postoperative adhesions were encountered. Adhesiolysis was performed and dissection was carried out in the area of the retroperitoneum. The retropancreatic tumor was identified, which measured about 5 cm x 5cm x 3 cm. It was noted to be anatomically

separate from the pancreas with venous drainage into the inferior vena cava (Figure 3). A left suprarenal mass measuring about 2 cm x 1.5 cm x 1 cm was noted to be posterior to the stomach. Adjacent organs were not involved. Both tumors were resected. The patient tolerated the procedure well, had an uneventful recovery period and was discharged four days, postoperatively.

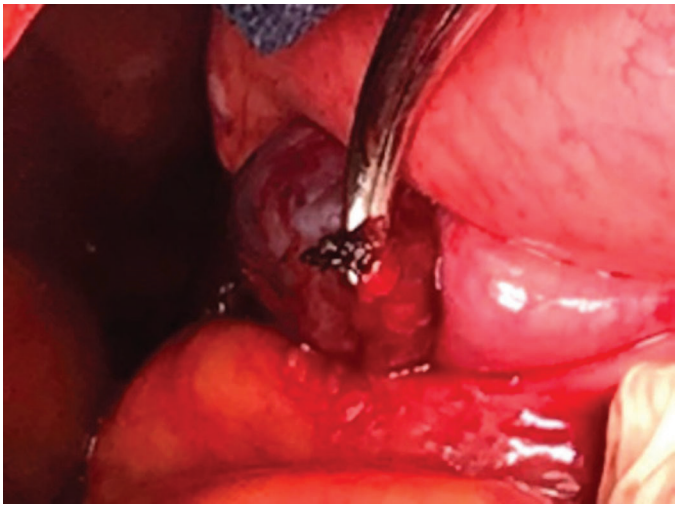

Figure 3. Intraoperatively, a retropancreatic tumor anatomically separate from the pancreas was resected measuring 5 cm x 5 cm with noted draining vein (ligated vessel held by mosquito forceps) into the inferior vena cava.

On gross histopathologic examination, the retropancreatic tumor measured 6.0 cm x 4.4 cm x 3.6 cm. Cut sections were tan to gray-brown, with firm tissue peripherally and central areas of hemorrhagic necrosis. Histomorphologic features were characterized as sheets, clusters, and pseudoacinar patterns of medium-to-large sized tumor cells with a moderate amount of eosinophilic, granular cytoplasm. Cells showing abundant, vacuolated cytoplasm and marked nuclear atypia and pleomorphism and prominence of nucleoli were present (Figure 4). These tumor cells were seen separated by scanty fibrocollagenous stroma. There were 0-3 mitotic figures seen per 50 high power fields. Extensive areas of hemorrhage and necrosis were noted. This was consistent with a moderately to poorly differentiated carcinoma with histomorphologic features suggestive of adrenal cortical carcinoma, oncocytic type, low grade. However, other neuroendocrine tumors could not be totally ruled out so immunohistochemical stains were suggested for a more definitive diagnosis. The left suprarenal mass was consistent with a lymph node with noted sinus hyperplasia and was negative for tumor.

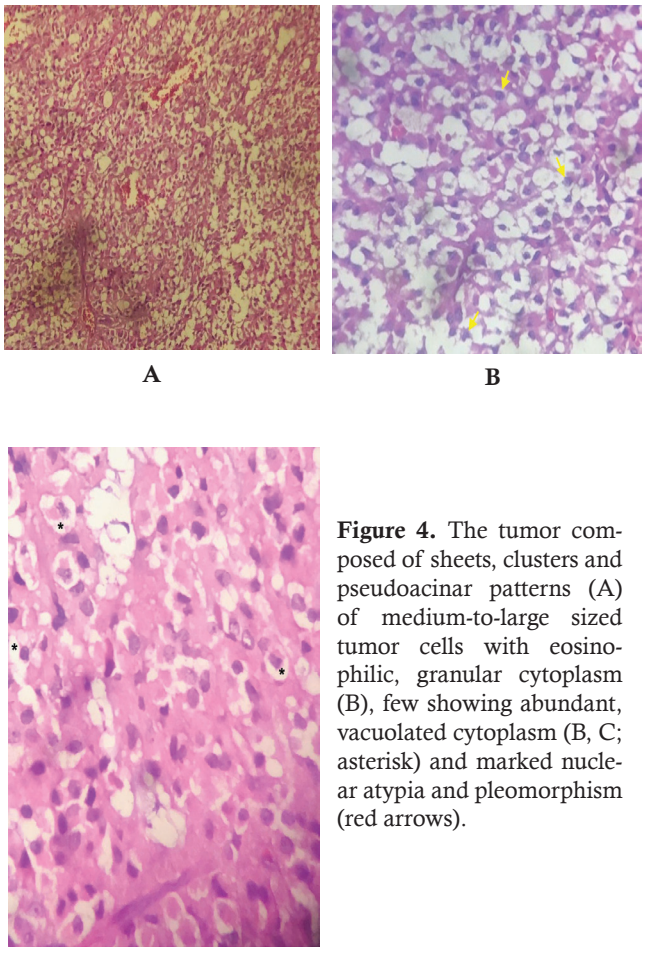

Figure 4. The tumor composed of sheets, clusters and pseudoacinar patterns (A) of medium-to-large sized tumor cells with eosinophilic, granular cytoplasm (B), few showing abundant, vacuolated cytoplasm (B, C; asterisk) and marked nuclear atypia and pleomorphism (red arrows).

Immunohistochemical staining results (Figure 5) revealed a strong and diffuse cytoplasmic expression for vimentin strong and diffuse nuclear and cytoplasmic expression for β-catenin, strong and patchy cytoplasmic expression for pancytokeratin, and nonspecific nuclear expression for melan-A. The patient had negative expression for inhibin α, chromogranin A,

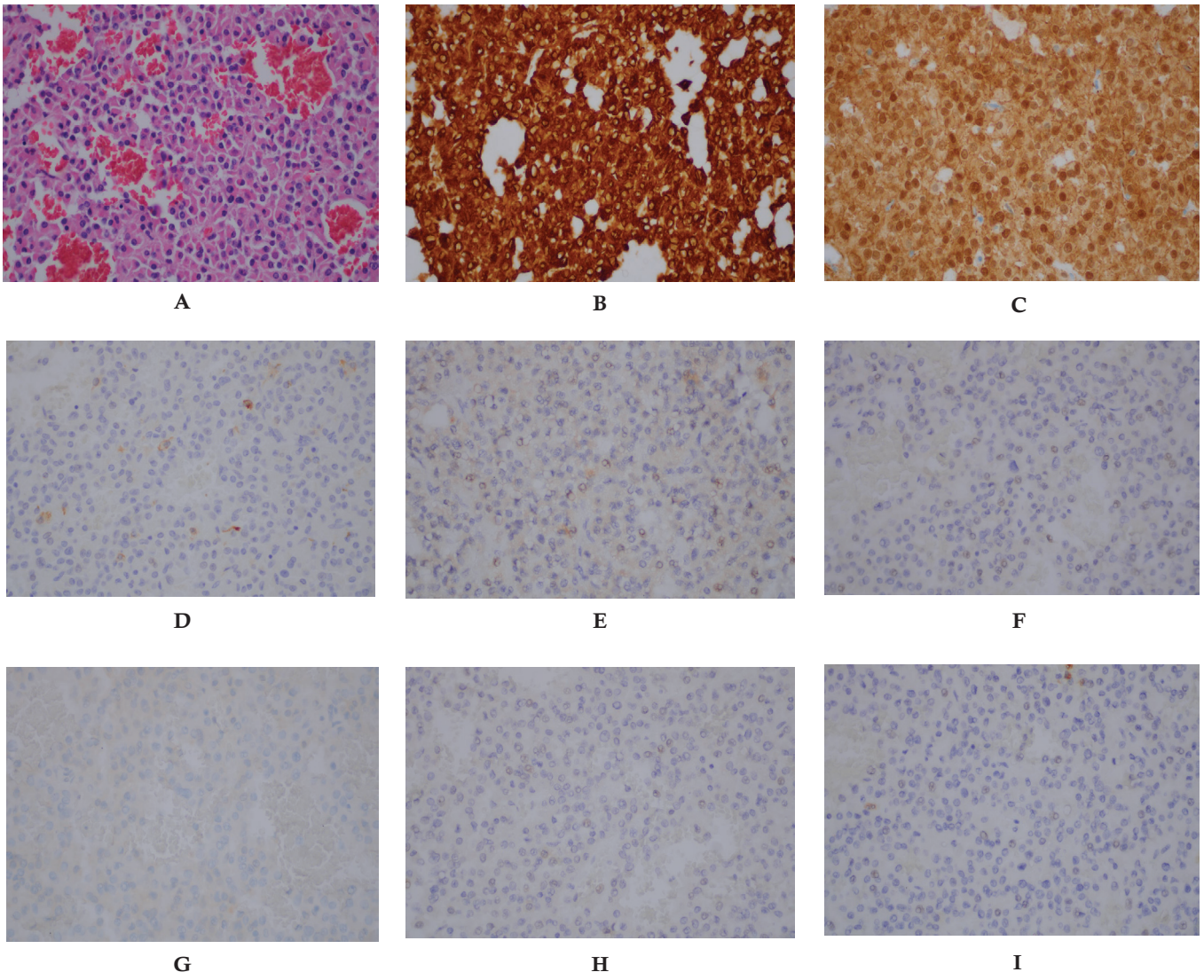

Figure 5. Microphotographs of routine and immunohistochemical stains of the specimen. Hematoxylin and eosin stain (A). Vimentin, β-catenin, and pancytokeratin were strongly positive (B, C, D), and nonspecific for melan-A (E). Stains for inhibin α, chromogranin A, synaptophysin, and CD10 were negative (F, G, H, I).

synaptophysin, and CD10. The histomorphologic and immunohistochemistry findings were consistent with a solid pseudopapillary neoplasm.

Discussion

Solid pseudopapillary neoplasm is a rare pancreatic neoplasm with known low-malignant potential. It is a rather slow-growing tumor with a reported doubling time of 765 days.[5] In about 60% of cases, it is located in the pancreatic body or tail with the remainder developing in the pancreatic head and neck.[6] The incidence of this tumor has been increasing over the past years. It is predominantly seen in young women, with a male-to-female ratio of 1:9.78[7], and an average presentation age of 35 years (range of 8-67 years).[2]

This neoplasm has been reported under various names such as solid and papillary neoplasm, papillary epithelial

neoplasm, solid and cystic acinar tumor, papillary-cystic carcinoma, Gruber-Frantz tumor, but the most widely accepted terminology by the World Health Organization is a *solid pseudopapillary neoplasm*, designating both solid and pseudopapillary histologic features of the tumor.[8] Majority of cases were reported from Europe, Japan, and North America, and only a few in the Caribbean region.[9-10]

In contrast to pancreatic SPNs, the reported incidence of extrapancreatic SPN is only around 1% to 1.8% and has decreased to 0.62% from 2004 to 2018.[5] These tumors are found anatomically separate from the pancreas, arising most commonly from the ovary[3] and testis/paratesticular[4] areas, but can also be found in the retroperitoneum[1,11], mesentery[12], omentum[13], gastrointestinal tract[14], and mesocolon.[15] So far, only about 50 cases have been reported in the English literature.[5] Some of these tumors have occurred in the context of an ectopic pancreas or an ectopic pancreatic tissue having no structural connection to the normal pancreas.[15,16] In the Philippines, only one case of extrapancreatic SPN, also retroperitoneal in origin, has been documented by Atibagos, et al. in 2003.[11] In this hospital, this is the first documented case of extrapancreatic SPN.

SPNs have a clinical, radiologic, and histologic resemblance to that of pancreatic neuroendocrine tumors (PNET) with occasionally similar immunohistochemical profiles. Patients are most of the time asymptomatic. If symptoms are present, they are non-specific: abdominal pain or discomfort, or compressive symptoms from a gradually enlarging mass. A physical examination is frequently unremarkable but will sometimes reveal a palpable abdominal mass.

Laboratory examinations identify few to no abnormalities. These tumors are frequently detected incidentally on imaging. On computed tomography and magnetic resonance imaging, they are described as well-circumscribed, encapsulated, and heterogenous tumors with hemorrhagic and cystic degenerative components. On dynamic enhancement, they have heterogenous enhancement on the arterial phase and progressive centripetal enhancement on the delayed phase. This is similar for both pancreatic and extrapancreatic SPNs.[17] Just as in the present case, few studies have also reported these tumors to be particularly FDG avid on PET/CT scan despite having a low malignant potential.[18]

Grossly, SPNs are large and solitary with cross-sections ranging from solid to cystic, and often with areas of necrosis and hemorrhage, congruent with its radiologic findings. Most have a well-developed capsule described to be rarely infiltrative.[19] Histomorphologically, SPNs present a characteristic combination of solid-cystic elements having a distinctive pseudopapillary growth pattern. Solid areas are formed by nests of uniform, small-to-medium sized, polygonal cells, separated by fibrovascular septa, such features indistinguishable from PNET.[1,20] However, the presence of pseudopapillary areas with myxoid connective tissues favors the impression of SPN. Tumor cells have abundant clear to eosinophilic cytoplasm that can be vacuolated, similar to the case presented. Nuclei are usually uniform, round to oval with frequent nuclear grooves lacking the salt-and-pepper chromatin of PNET. Mitotic figures can be present, but these are noted more frequently in metastatic tumors compared to primary tumors, though their mitotic indices are not markedly different.[21]

Based on currently available data, immunohistochemical stains are invaluable in diagnosing SPNs. Tumor cells are consistently positive for nuclear expression of β-catenin (98%), E-cadherin, vimentin (88%), CD10 (63%), synaptophysin (55%), and cytokeratin (52%)[22], and characteristically negative for chromogranin A.[20] The patient tested positive for β-catenin, vimentin, and cytokeratin while synaptophysin, chromogranin A, melan-A and inhibin α were negative. Similarly, PNET and adrenocortical tumors share similar immunohistochemical findings (Table 1). In general, the IHC profile of an adrenocortical tumor of the oncocytic type shows diffuse positivity for vimentin, melan-A, synaptophysin, and inhibin α, while chromogranin A is negative.[23] PNETs will generally stain positive for markers of neuroendocrine differentiation like synaptophysin and chromogranin A.[24]

The pathogenesis of SPN remains elusive. The predilection of SPN for females, its proximity of the pancreas to the genital ridges during embryogenesis[25], and the positivity of progesterone receptors immunohistochemically have led to the hypothesis that SPN could be derived from omnipotent cells of the genital ridge being entrapped in the pancreatic anlage during early embryogenesis, while other cells follow the

normal migration pathway to the ovaries.[26] Following this theory, extrapancreatic SPNs can then occur at any point along the route of migration of these stem cells towards the ovary, which includes the retroperitoneal space. This theory has been gaining some support in recent studies. The presence of progesterone receptor markers also supports the theory that hormones may have an effect on tumor development along with the ovaries being identified as a possible site of extrapancreatic SPNs. However, this is still somewhat controversial as 12.2% of SPNs occur in men.[10] Recent studies have also suggested a common origin for SPN and signet ring cell carcinoma of testis or ovary with both variants having a immunohistochemistry profile similar to SPN to include nuclear β-catenin with CD10 positivity.[4]

Preoperative biopsy of these tumors has shown limited accuracy as evidenced in a study by Butte, et al. with a diagnostic accuracy of only 56%.[27] The treatment of choice remains to be complete surgical excision, whether open or laparoscopic. In a study by Tan, et al, both approaches have no significant difference in terms of intraoperative blood loss, transfusion requirements, postoperative morbidity and mortality, resection margin, lymph node yield, and long-term survival, though laparoscopic approach provides a shorter postoperative length of stay.[28] Several studies reported excellent prognosis after total surgical excision with local recurrence of up to 10% in 4 years[29], similar for both pancreatic and extrapancreatic SPNs. Factors associated with recurrence as detailed by a study by Yang, et al. include vascular invasion, invasion to adjacent organs, lymph node metastasis, and Ki-67 index ≥4%.[30] Metastasis can occur in about 15% of cases, but clinical progression following metastasis is slow and most lesions can be treated by complete surgical excision of metastatic tumors.[31] Liver is the most common site of metastasis, followed by lymph nodes and peritoneum.[32] Lung metastasis was also reported.[33] Metastasis or invasion to adjacent organs is not a contraindication of surgery; however, extensive lymphadenectomy is not recommended. The overall 5-year survival of patients is around 95%.[34] Only around 5-10% of these tumors have been described to demonstrate aggressive behavior[6,35], but despite poor prognostic factors, patients still have good outcomes.

For unresectable disease, limited evidence supports the use of chemotherapy and radiotherapy with little to no reliable data.[36] Long-term survival for this subset of patients is difficult to assess because whether the high long-term survival is attributable to the benefits of chemotherapy or the natural cause of a slow aggressive tumor is questionable. There is a lack of any evidence-

Table 1. Comparison of immunohistochemistry profile between the case and its differential diagnoses.

	Case	SPN	PNET	ACC
Beta-catenin	+	++	+	+
Vimentin	+	+	-/+	+
Melan A	+/-	ND	ND	+
Inhibin alpha	-	-	LD	+
Synaptophysin	-	+/-	+	+
Chromogranin A	-	-	+	-
Cytokeratin	+	+/-	+	-/+
CD10	-	+	+	-

Abbreviations: ++, more than 95% are positive; +, usually more than 75% of cases are positive; -, less than 5% of cases are positive; +/- , usually more than 50% of cases are positive; -/+, less than 50% of cases are positive; LD, limited data; ND, no data; ACC, adrenocortical carcinoma

based guidelines for follow-up postoperatively. Bansal, et al. recommend annual imaging studies with abdominal and chest CT scans up to a minimum 10-year follow-up after resection.[35]

In summary, SPNs developing outside the pancreas are extremely rare, our present case included. Aside from that, the present case developed as a second primary in a patient previously diagnosed with adrenocortical carcinoma. The etiopathogenesis of SPN remains elusive. They are thought to originate from stem cells of the genital ridge entrapped in the pancreas. SPNs may develop at any point along the route of stem cell migration during early embryogenesis. Since most of these neoplasms demonstrate indolent behavior, prognosis is favorable after surgical resection even in the setting of metastatic disease. There is limited evidence supporting the use chemotherapy and radiotherapy in their treatment.

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