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Duodenum-preserving pancreatic head resection in solid pseudopapillary neoplasm—Report of a case

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ABSTRACT

INTRODUCTION: Solid pseudopapillary neoplasm (SPPN) was first characterized by Virginia Frantz in 1959. The duodenum-preserving pancreatic head resection (DPPHR) has been described as treatment for low-grade malignant tumors of the head of the pancreas including eight cases of SPPN.

PRESENTATION OF CASE: A 16-year-old white female patient presented with abdominal pain and dyspepsia. Computed tomography scan of abdomen showed a $10 \times 9 \times 10\,cm^3$ lesion on the pancreatic head. After radiological diagnosis of SPPN the patient was submitted to DPPHR. Resection was achieved with clear margins. Immunohistochemical study demonstrated positivity for progesterone receptor, β-catenin, cytoplasmic paranuclear dot-like CD99, negativity for chromogranin and S100 protein and Ki 67 index of 1%.

DISCUSSION: A large encapsulated pancreatic mass with well-defined borders that contains areas of calcifications and intratumoral hemorrhage on CT scan in a young female is virtually diagnostic of an SPPN. A particular dot-like intracytoplasmic expression of CD99 appears to be highly unique for SPPN

CONCLUSION: DPPHR should be considered in cases of SPPN in the pancreas head if there is no compromise with oncologic radicality.

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1. Introduction

Solid pseudopapillary neoplasm (SPPN) was first characterized by Virginia Frantz in 1959[1] and was defined by The World Health Organization (WHO) as "solid pseudopapillary tumors" of the pancreas in 1996.[2] It is a neoplasm of unknown etiology, low malignant potential and good prognosis that occurs most often in young women.[3–5] Symptoms of SPPN are often nonspecific but the radiological appearance is distinct from other pancreatic tumors.[5,6] Complete resection has a favorable prognosis.[7,8]

Pancreatoduodenectomy is the most described surgical procedure to treat SPPN in the head of the pancreas.[3–8] The duodenum-preserving pancreatic head resection (DPPHR) has been described for treatment of low-grade malignant tumors of the head of the pancreas including SPPN, if there is no compromise with oncologic radicality.[9,10]

We present a case of 16-year-old female patient with solid-pseudo papillary neoplasm submitted to duodenum-preserving pancreatic head resection. The histopathology examination and immunohistochemical study confirmed the diagnosis.

2. Presentation of case

Written informed consent was obtained from the patient for publication of this case report and accompanying images. In October 2012, a 16-year-old white female patient presented with abdominal pain and dyspepsia, which started when she was at age of 12. Computed tomography scan of abdomen showed a $10 \times 9 \times 10$ cm lesion on the pancreatic head (Fig. 1). The radiological diagnosis was of a solid pseudopapillary neoplasm. Serum tumor markers were carcinoembryonic antigen (CEA): 0.8 ng/mL and carbohydrate antigen 19-9 (CA 19.9): 50 U/mL The patient was submitted to a duodenum-preserving pancreatic head resection (Fig. 2). She had a very good recovery and was discharged on the 9th post-operative day.

Pathologic evaluation showed a well-demarcated encapsulated tumor weighing 201.6 g and measuring $7.0 \times 6.5 \times 6.0$ cm. Sectioning demonstrated a solid mass with soft areas, fibrous bands and hemorrhage focus, without cystic degeneration (Fig. 3). The histological sections showed a neoplasm characterized by polyhedral/elongated cell proliferation, with round and pleomorphic nuclei forming solid nests and papillae with fibrovascular axes

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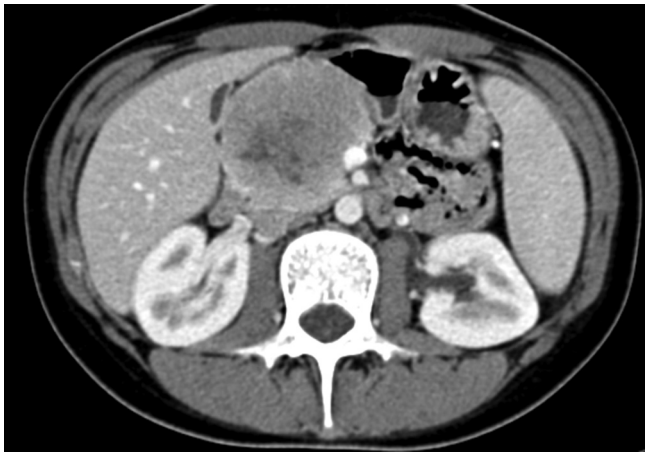

Fig. 1. Axial CT of the abdomen with intravenous contrast demonstrating a large heterogeneous mass in the ventral head of the pancreas. The mass measures approximately $10 \times 9 \times 10$ cm in its maximum anterior-posterior, transverse and craniocaudal dimensions, respectively.

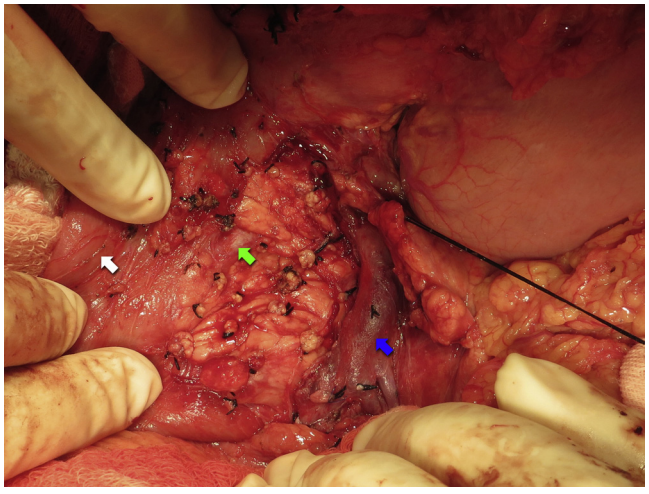

Fig. 2. Duodenum preserving pancreatic head resection. Duodenum (white arrow), superior mesenteric vein (blue arrow) and common bile duct (green arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.).

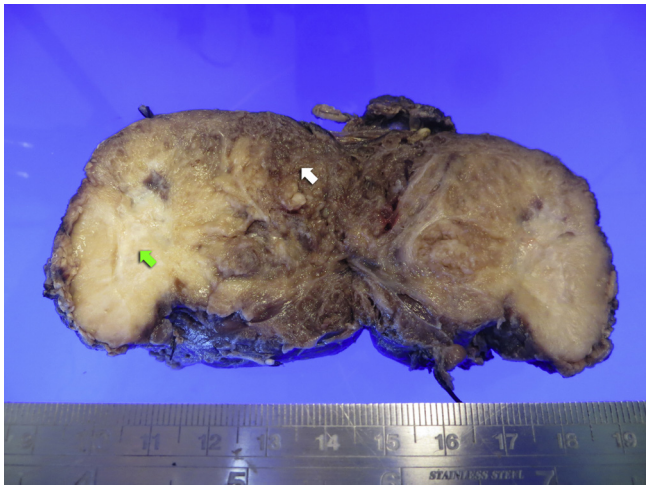

Fig. 3. Cutting surface shows a solid mass (green arrow) and soft areas (white arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.).

(Fig. 4a). Clear margin was achieved. Immunohistochemical study demonstrated strong and diffuse nuclear positivity for progesterone receptor, negativity for cromogranin and S100 protein, nuclear and cytoplasmic positivity for β-catenin and cytoplasmic paranuclear dot-like positivity for CD99 and Ki 67 index of 1% (Fig. 4 b, c, d, e and f, respectively).

3. Discussion

Solid pseudopapillary neoplasm (SPPN) otherwise known as solid and papillary tumor, solid-cystic tumor and Frantz's tumor, among others, was first characterized by Virginia Frantz in 1959 as a "papillary cystic tumor of the pancreas". The patient was a 2-year-old boy who died during an attempted duodenectopancreatectomy.[1] The World Health Organization (WHO) defines it as "solid pseudopapillary tumors" of the pancreas since 1996.[2]

SPPN is an uncommon indolent cancer of unknown etiology, low malignant potential and good prognosis that account for 2–3% of primary pancreatic malignancies. SPPN occurs most often in women during their second and third decades as a slow-growing mass, and can arise at any location within the pancreas (head, body, or tail), without a preferential site. If a complete resection is achieved, most patients have a favorable prognosis, with higher than 90% overall cure rate.[7,8] However, 10–15% of the patients with SPPN reported developing metastasis and/or recurrence.[11,12]

Symptoms of SPPN are often nonspecific and include abdominal pain, dyspepsia, early satiety, nausea and vomiting.[3–5] It is distinct from other pancreatic tumors both in epidemiologic distribution as well as radiological appearance. A large encapsulated pancreatic mass with well-defined borders that contains areas of calcifications as well as intratumoral hemorrhage on CT scan in a young female is virtually diagnostic of an SPPN.[4–6] Our patient presenting symptoms were comparable with previous literature, with the most common complaint being abdominal pain.

Grossly, the tumor is a solid mass well circumscribed with regions of necrosis, hemorrhage and cystic degeneration with a thick fibrous capsule. Histologically, the solid portions contain sheets, cords and nets of uniform, rather small, and fairly round cells in a rich and delicate vascular network. Many of the cells farthest from the vessels undergo degeneration, causing the remaining cells around the vessels to form pseudorosette or pseudopapillary patterns.[1,2,4] Rarely, the SPPN exhibit a high-grade component that consists of diffuse sheets of cells with a high nucleus to cytoplasm ratio, necrosis and elevated mitotic activity suggesting the development of a poorly differentiated carcinoma. These tumors are associated with a rapidly progressive and fatal clinical course.[13]

The tumors show immunohistochemical stain for vimentin, α1-antitrypsin, neuron-specific enolase, CD56 and CD10. Positivity staining for chromogranin (a precursor of several neuroendocrine peptides) is never seen. The abnormal immunohistochemical nuclear localization of β-catenin staining has been proposed as a diagnostic aid. Corresponding loss of the cell surface expression of E-cadherin is also seen, and these abnormalities likely contribute to reduce cell cohesion that characterizes this neoplasm.[14,15] A particular dot-like intracytoplasmic expression of CD99 appears to be highly unique for SPPN.[16,17] CD99 is a glycoprotein encoded by the pseudoautosomal gene MIC2. It is typically present in Ewing sarcomas (ES), primitive neuroectodermal tumors (PNETs), lymphoblastic lymphomas, synovial sarcoma and other solid tumors, but the immunostaining in all those cases is almost exclusively confined to the cellular membrane. The characteristic cytoplasmic paranuclear "dot-like" pattern described in SPPN has also been found in colonic adenomas/adenocarcinomas, pituicytomas and endometrial serous carcinomas with ES differentiation, but to date it has not been described in any other type of endocrine or

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