CLINICAL CASE

Pediatric solid pseudopapillary tumor of the pancreas. Case report and literature review

José Francisco Camacho Aguilera[1], Juan Carlos Duarte Valencia[2], Juan Francisco Hidalgo Corona[3], Alfonso Valenzuela Espinoza[4], Pameri Ortiz Equihua[5]

Abstract

Background. Solid pseudopapillary tumor (SPT) of the pancreas is a low-grade epithelial malignant tumor principally affecting young women and represents ~1-2% of all pancreatic neoplasms. We present a case of this type of tumor treated at the General Hospital of Tijuana in the pediatric surgery service. We also present a review of the literature.

Case. We present the case of a 12-year-old female with symptomatology of a progressively growing palpable tumor in the upper abdomen. She was diagnosed with SPT located in the tail of the pancreas. It was treated successfully with distal pancreatectomy without splenectomy. The patient was discharged on the third postsurgical day.

Discussion. SPT is a differential diagnosis with the presence of a mass at the level of the pancreas. Due to its rarity, it is not the first option to rule out, especially in pediatric patients. Surgery alone represents the best treatment for this pathological entity and should be attempted in all cases, independent of the size of the pancreatic lesion.

Key words: pediatrics, pancreatic solid pseudopapillary tumor.

Introduction

Solid pseudopapillary tumor (SPT) of the pancreas is a low-grade malignant epithelial tumor that mainly affects young women. It was first described by Frantz[1] in 1959 and has also been referred to by other names such as papillary epithelial neoplasm, solid and cystic acinar tumor, papillary cystic neoplasm, papillary cystic carcinoma, solid and cystic tumor, papillary tumor of low grade and Frantz's tumor.[2]

[1]Cirugía General, Querétaro, Qro, [2]Cirugía Pediátrica; [3]Cirugía General; [4]Patología, Hospital General de Tijuana; [5]Médico General, Universidad Xochicalco, Tijuana, Baja California, México.

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Finally, in 1996, WHO named it solid pseudopapillary tumor of the pancreas[3] and classified it within the group of "borderline" tumors of the exocrine pancreas, i.e., with uncertain malignant potential.[4] SPT is a neoplasm that comprises ~1-2% of all pancreatic neoplasms (Table 1).[5] We report a case of this type of tumor treated at Hospital General de Tijuana in the Pediatric Surgery Department, as well as a review of the literature.

Case Report

We present the case of a 12-year-old female without relevant history. There was a 4-year evolution of the presence of a mass in the epigastric region, which was gradually increasing in size to be notable

with superficial palpation. Physical examination revealed a patient in good general condition and with presence of a painless palpable tumor in the epigastrium and left hypochondrium of ~15 × 10 cm. Laboratory tests were normal including á-fetoprotein and â-human chorionic gonadotropin levels. An ultrasound was performed showing a solid lesion of ~8 × 9 × 5 cm in the left upper quadrant with a heterogeneous appearance and well-defined capsule (Fig. 1). Computed axial tomography was carried out where a heterogeneous, well-defined encapsulated tumor of ~8 × 8 cm was observed (Fig. 2). MRI axial image of the lesion (Fig. 3) revealed the tumor occupying most of the upper left quadrant.

Finally, an exploratory celiotomy was performed with an approach through a left subcostal incision. A solid tumor ~10 cm in diameter was found in the

Table 1. WHO classification of cystic tumors of the pancreas

I. Primary tumors
- *A. Exocrine pancreas*
  - a. Benign
    - Serous cystadenoma
    - Mucinous cystadenoma
    - Intraductal papillary mucinous adenoma
    - Cystic mucinous teratoma
    - "Bordeline" (malignant potential)
    - Cystic mucinous tumor with moderate dysplasia
    - Intraductal papillary mucinous tumor with moderate dysplasia
    - Solid pseudopapillary tumor
  - b. Malignant
    - Ductal adenocarcinoma
    - Undifferentiated carcinoma (anaplastic)
    - Serous cystadenocarcinoma
    - Mucinous cystadenocarcinoma: noninvasive or invasive
    - Intraductal papillary mucinous tumor: noninvasive or invasive
    - Cystadenocarcinoma of acinar cells
    - Solid papillary carcinoma
- *B. Endocrine pancreas*
  - Islet cell tumors (functional and nonfunctional)

II. Secondary tumors
- A. Cystic tumors of the exocrine pancreas
  - Pseudocyst
  - Congenital cyst
  - Lymphoepithelial cyst
  - Endometrial cyst
  - Retention cyst
  - Parasitic cyst
  - Cyst of the paraampular duodenal wall
  - Enterogenous cyst

Adapted from Valdés et al.[9]
WHO, World Health Organization.

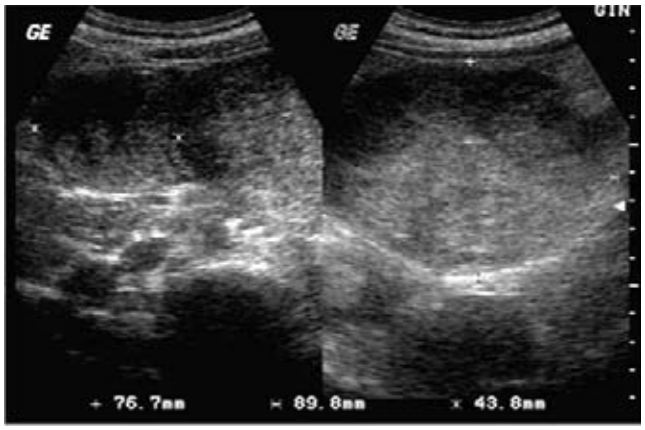

Figure 1. Abdominal ultrasound showing the encapsulated tumor with heterogeneous tissue.

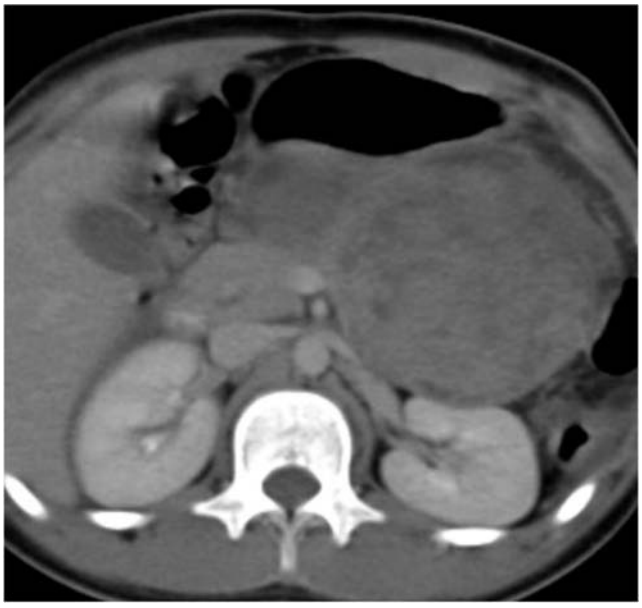

Figure 2. CT scan cross-section involving the body of the pancreas showing a mass with a thin capsule and with a heterogeneous density, apparently on the tail of the pancreas.

tail of the pancreas (Fig. 4). Distal pancreatectomy was performed with tumor resection. There was good postoperative evolution, and vital signs were normal.

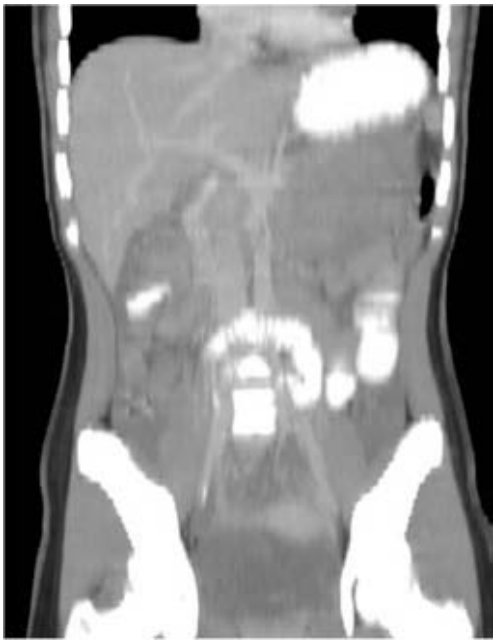

Figure 3. Magnetic resonance imaging showing a mass preventing adequate filling of the stomach with contrast media.

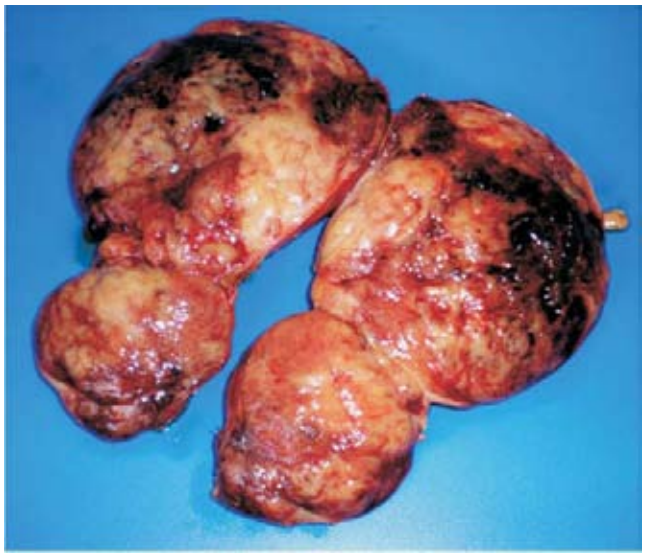

Figure 4. Macroscopic image of the resected lesion with coronal section where the presence of an encapsulated, solid and pseudocystic tumor is observed in the tail of the pancreas.

There was a small amount of discharge from the closed drainage and oral tolerance.

Histopathological analysis revealed the presence of a 9 × 8 × 4 cm grayish-white encapsulated and indurated tumor. Dissection of the mass revealed dark brown areas with alternating solid, grayish-white, granular and microcystic areas and appearance of congestive hemorrhage (Fig. 5). Histopathological diagnosis was SPT of the pancreas.

Discussion

SPT of the pancreas is an extremely rare neoplasm. Since the first report conducted by Frantz in 1959, up until 2006, 629 cases have been reported in 178 series[6-8] and only four cases have been reported in Mexico.[9-11] Until 1999, there were ~90 children (<18 years of age) reported with SPT.[12] SPT of the pancreas is very rare, accounting for 0.17-2.7% of primary nonendocrine tumors of the pancreas.[13-15] It is primarily seen in non-Caucasian women in 90-95% of the cases[5,6] (especially Asian and African- American women) who are in their second and third decades of life[5,16,17] with a peak of incidence in the third decade of life (average age: 24 years; range: 2-72 years of age).[8] Cases presented in the first decade of life are rare and <10% are

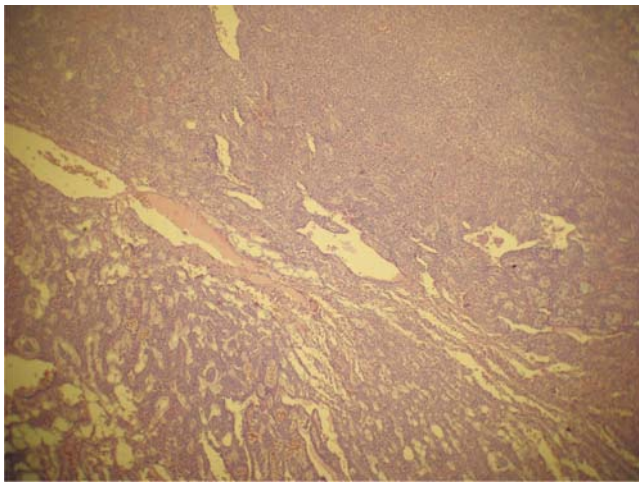

Figure 5. Microscopic image showing a solid pattern (top) and pseudopapillary formations of the SPT characteristics (below).

reported in patients >40 years of age.[18] SPT is rarely seen in males (1:9.5 M-F ratio),[15,19] even though in children we have found a 1:3 M-F ratio.[20] Average peak of incidence in males is 31 years of age[18] (average age is higher in male patients, $p < 0.05$).[21]

The etiology of SPT of the pancreas is still unknown and continues to be controversial. There have been three possible sources proposed: 1) pancreatic duct cells, 2) acinar cells or 3) endocrine cells.[16,22] Another hypothesis is that the SPT arises from pancreatic stem cells or derived from extrapancreatic TISSUE, possibly gonadal (ovarian), which may have been attached to the pancreatic parenchyma during early embryogenesis.[8,23] Presence of genetic factors has also been suspected as its origin according to the observed higher incidence in Asian women.[14] Immunohistochemical analysis reveals that in >90% of cases there is positivity for vimentin, a neuron-specific enolase, α-1-antitrypsin[3,5,24,25] α-1-antichemotrypsin,[16] to CD10 and to CD56.[24,26] We have also observed the expression of E-cadherin in the nucleus and its absence at the level of membrane and cytoplasm.[27] All SPT cells show cytoplasmic immunoreactivity for galectin-3, which serves to differentiate the SPT of endocrine tumors.[22]

The presence of estrogen receptors in these tumors has not been observed, but the presence of progesterone receptors has been indicated in many cases (80-100%).[10,16,21,28] The expression of vimentin, α-1 antitrypsin, α-1 antichemotrypsin and neuron-specific enolase confirms the suspicion of the theory of a stem cell from a primitive epithelial cell.[7] It is believed that sex hormones play a greater role in tumor growth than in its pathogenesis[14,18] as confirmed by the highest growth rate of SPT during a pregnancy.[16] On the other hand, Abraham et al. observed the presence of mutations in exon 3 of the oncogene of the β-catenin in 90% of the analyzed samples, and only 15.8% showed nuclear overexpression of p53.[13] Tanaka et al. observed 100% immunoreactivity for β-catenin in the cytoplasm and 83% in the nucleus of 18 samples of SPT.[29] Occasionally we have observed the presence of aneuploidy,[13] especially in malignant SPT. We found chromosomal abnormalities including double loss of X chromosomes, trisomy of chromosome 3 and unbalanced translocation between chromosomes 13 and 17 associated with aggressive behavior.[18] The proliferative index, evaluated by Ki-67 immunoreactivity, is <1% of the common samples of SPT. In aggressive cases it has shown 30-40% positivity.[30]

The clinical picture of these tumors is variable. In the review by Martin et al., abdominal pain was found in 58% of cases, and 29% of cases were asymptomatic,[2] similar to that observed by Nakagohri et al.[31] Moreover, Patil et al. reported the presence of abdominal pain in 72% of the patients in their report of 14 cases,[32] and Sheehan et al. in 63% of their cases.[33] Abdominal pain is also the most common symptom in children with SPT and is present in 87% of the cases in addition to palpable mass in 35%, dyspepsia in 26% and elevated serum amylase in 18%.[20] Other clinical data that may be present are vague abdominal discomfort,[16] sensation of fullness,[14] or early satiety,[7] sensation of abdominal mass, jaundice,[34] nausea and vomiting (32%),[21] and weight loss (18%).[21,25,33] A palpable abdominal mass may be a late symptom of the disease.[7] Nevertheless, these types of tumors grow slowly, which contributes to long disease-free periods even in patients with recurrences or metastases.[1] Although these tumors can grow to 20 cm, growth generally occurs around the tissues rather than within them.[35] Hence, it follows that although the SPT is situated at the head of the pancreas, only 8% of them present with jaundice.[36] In other cases, diagnosis is made incidentally while performing a complementary imaging study for another disease[14] because these patients are asymptomatic even if they are carriers of this tumor.[20] In some series, incidental diagnosis reached up to 55% of patients.[23] Diagnosis of acute abdomen when presented with hemoperitoneum due to spontaneous rupture or intratumoral bleeding is even more rare.[14,17,34,37]

Laboratory tests are usually normal (e.g., amylase levels)[16] and pancreatic cancer markers (e.g., CA19-9, CEA or α-fetoprotein) are almost always negative.[3]

Imaging studies for the diagnosis of these lesions are ultrasound, CAT scan and MRI imaging. On plain films of the abdomen we can observe displaced structures or calcifications.[9] Moreover, in the ultrasound study we observe a mass with areas of high and low echogenicity without septa in its interior,[34] and angiography demonstrate areas of low vascularity.[9,38] In the CAT scan, SPT presents as a large heterogeneous encapsulated mass.[2] Areas of hemorrhage have hyperattenuation and areas of cystic degeneration are seen with hyperattenuation. Calcifications were observed in 30% of the cases. In contrast, there is a heterogeneous peripheral reinforcement with low-density center.[39] The capsule is usually hyperdense (70%) and tumors may have a fluid level in their interior (10%).[35] Calcifications are rare, but when they exist are in the periphery of the tumor.[40] Some authors have reported a frequency of ~40% calcifications.[23,31] MRI shows heterogeneity in 73.5% of the cases and 89% are hyperintense. The presence of hemorrhage within the lesion is seen in >70% of the cases.[35] Areas of hyperintensity in the T1 series and low hypointensity or non-homogeneous in the T2 series help identify the areas of hemorrhage and also help to differentiate SPT from other pancreatic tumors.[19] Solid areas in the T1 series demonstrate hypointensity, although occasionally we observe areas of hyperintensity.[41] It is suggested to perform percutaneous CT-guided fine needle aspiration (FNA), although this practice is not widespread because of potentially serious complications.[37] Therefore, the possibility of endoscopic ultrasound FNA biopsy has been suggested because this procedure shows 81.6% sensitivity and 87.5% specificity for non-neuroendocrine tumors.[42] Complications from this diagnostic method are scarcely 2%.[43] SPT has also been detected with PET scan where we can observe elevation of F-18 fluorodeoxyglucose due to the lesion.[44]

SPT is now confined to the pancreas in 85% of the cases and 10-15% of the patients have metastases[13,16] or have tumor recurrence.[11,25] The most common site for metastasis is the liver (2-42%, average 14%),[8,45] inferior vena cava (27%), spleen (17%),[8] portal vein (5%),[17,46] peritoneum (42%), lymph nodes (25%)[17] and the remaining to other organs such as the retroperitoneum, duodenum, omentum, colon, mesentery and lung (9%).[8,46] Other authors reported lymph nodes and peritoneum[2] as common sites for metastases, after the liver. Liver metastases are usually solitary.[19] No clear criteria of malignancy has been established, but perineural or angioinvasion, with or without deep invasion into surrounding tissues,[34] as well as a high degree of cellular pleomorphism and an elevated mitotic index, may indicate an aggressive behavior.[30,46] Other pathological features probably associated with an aggressive behavior of SPT are diffuse growth pattern with extensive tumor necrosis, presence of an undifferentiated component,[30] atypial nucleus, venous invasion and the presence of mono- or multinucleated giant cells.[22] The presence of SPT in elderly patients has been associated with increased likelihood of malignancy.[22] The neoplasm is more aggressive in male patients.[21]

Histopathological analysis demonstrated the presence of encapsulated tumors composed of cystic, solid and hemorrhagic elements. The presence of a capsule and intratumoral hemorrhage are important characteristics in the diagnosis because they are rarely found in other pancreatic neoplasms.[3,15,34] Sizes of these tumors ranges from 2-20 cm. Various studies report an average between 6 and 10 cm, being larger in the body and tail of the pancreas than in the head.[12,15,20,25] Tumors can be found calcified in 30% of the cases that are generally peripheral.[8,19,34] At the microscopic level, there are solid areas alternating with pseudopapillary formations, evidence of cell degeneration including cholesterol crystals and histiocyte aggregates, nuclear grooves and hyaline cytoplasmic globule aggregates.[2,25] Degenerative changes produce a pseudopapillary pattern characteristic of epithelial cells in several layers around a central fibrovascular stalk.[3,16,25] The cells are uniform with low mitotic activity, polygonal, monomorphic and with a high concentration of eosinophils.[10,15] Solid areas contain necrosis, spongy macrophages, and granules of cholesterol and calci-

fication.[16] It is possible that the SPT is essentially solid and hypervascular and, that as the size increases, is associated with high intratumoral hemorrhage and necrosis with development of degenerative cystic components. Clinical manifestations, growth characteristics and immunoprofile are similar to the classic SPT.[47] Another growth form is called solid infiltrative papillary cystic neoplasm, which is histologically, histochemically and ultrastructurally similar to the classic SPT except for absence of a capsule and presence of an infiltrative portion.[38]

Differential diagnosis includes any solid and/or cystic pancreatic process.[17,18,34] In children we should rule out neuroblastoma, leukemia/lymphoma and lymphoproliferative disorders because they are more common than primary neoplasms. Metastases are extremely rare.[12]

The treatment of choice is surgery, preserving as much of the pancreatic tissue as possible.[8] The site of localization of the SPT in the pancreas in the head is 33-40%, in the body 14-28% and 32-50% in the tail;[2,10] hence, surgical options encompass simple enucleation, distal pancreatectomy and pancreaticoduodenectomy.[7,16,31] Splenectomy can be performed with distal pancreatectomy, which does not significantly alter morbidity and mortality.[48] In the case of enucleation, the risk of developing a fistula is high, so that it should be avoided where possible.[33] Extensive lymph node dissection or resection of adjacent structures is not justified.[14] Tumor size is not a predictor of unresectablility because lesions as large as 20-30 cm can be resected without problems.[2] With surgery alone, SPT prognosis is favorable. Five-year survival is >95%[12,49] and at 10 years was 93%[28] with a morbidity rate of up to 43%[35] and low recurrence rate (10-15%).[6,10,46] Complications that may occur after surgery were pancreatic fistula, biliary fistula, surgical wound infection, abdominal abscess, prolonged gastric emptying, intra-abdominal and ileus bleeding.[50] Even if synchronous metastases exists, resection should be attempted because the prognosis remains satisfactory with this approach. Metastases is not considered as a negative predictor of survival.[2,37] It has been suggested that age is a risk factor for development of metachronous metastases.[51]

Adjuvant chemotherapy has been used in a low number of patients without a clear picture regarding its role in this disease entity, especially in cases of liver metastases.[14] Chemotherapy regimens have been diverse without showing an adequate response.[2] Administration of chemotherapy may be attempted in patients with unresectable lesions or with multiple liver metastases, although there is no standard chemotherapy scheme.[45] Radiotherapy has been used infrequently in unresectable tumors or as an adjunctive therapy after tumor resection.[2,46] A poor response has been shown with cycles of 5-fluorouracil. Moreover, there has been a successful use of radiotherapy in locally advanced unresectable SPT. Favorable results have also been obtained with the use of ablation with percutaneous radiofrequency of the liver metastases.[45]

Our case corresponds to a 12-year-old patient, which is unusual even in this rare disease entity. There have been some reported case series of pediatric ages such as the study by Choi et al.[20] who reported an average age of 13 years. Our case presents the most common clinical data reported for the SPT: abdominal pain and presence of a slow-growing palpable mass. In studying the case report, laboratory tests do not demonstrate abnormalities. However, ultrasound, CAT scan and MRI clearly indicated the presence of the lesion. This has been confirmed by other authors who argue that these studies are of significant value for diagnosis of pancreatic SPT. In our patient, SPT treatment was a distal pancreatectomy. Complete tumor resection was achieved without any immediate postoperative complications.

We conclude that SPT is a differential diagnosis in the presence of a pancreatic mass, although due to its rarity it should not be the first option to rule out, especially in pediatric patients. Surgery alone is the optimal treatment for this disease because it demonstrates a level of excellent recovery.

Correspondence to: Dr. José Francisco Camacho Aguilera
Hospital General de Tijuana
Tijuana, Mexico
E-mail: entamoebo@hotmail.com

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