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Spontaneous rupture of solid pseudopapillary tumor of pancreas

A case report and review of literature

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Abstract

Introduction: Solid pseudopapillary tumors (SPT) account for 1% to 3% of all pancreatic tumors. They have low malignant potential with a favorable prognosis, and predominantly occur in young women. The pathogenesis and clinical behavior of SPT are still uncertain. In addition, most ruptures of SPT were associated with blunt abdominal trauma, while spontaneous ruptures seemed to be quite rare. Up to now, there have been only 3 spontaneous ruptured SPT cases reported worldwide.

Patient concerns: Here, we reported a 22-year-old female patient with left lower abdominal pain. Computed tomography (CT) showed that a hemorrhagic complex solid cystic mass located in the lesser omentum sac.

Diagnosis: According to pathological findings of tumor specimen, the diagnosis of solid pseudopapillary tumor (SPT) of the pancreas was made.

Interventions: Distal pancreatectomy and splenectomy was carried out.

Outcomes: The patient recovered to normal status within 10 days after surgery.

Conclusion: Besides, we reviewed about 50 cases in literatures to find out the clinical characteristics and differential diagnostic strategies of SPT.

Abbreviations: CT = computed tomography, EUS = endoscopic ultrasound, MRI = magnatic resonance imaging, SPT = solid pseudopapillary tumors.

Keywords: cyst, neoplasm, pancreas, rupture, solid pseudopapillary tumors

1. Introduction

Solid pseudopapillary tumor (SPT) of the pancreas is a kind of rare neoplasm, which represents less than 3% of all exocrine pancreatic tumors. SPT is prevalent among young females, with a median age of 20 to 30 years old.[1,2] When SPT represents in male, it has greater malignant potential with a worse prognosis. Besides, most of the ruptures of SPT were associated with blunt abdominal trauma, while the spontaneous ruptures seemed to be quite rare. Furthermore, the symptoms of SPT are not typical in general. Symptoms can occasionally occur due to the size and location of the tumor but usually are nonspecific.[3] Because of its unusual behavior, SPT is often associated with diagnostic and therapeutic challenges.[4] Computed tomography (CT) and magnetic resonance imaging (MRI) is beneficial for the diagnosis of this tumor.[5] Surgical resection is now considered as the most efficient treatment option for patients with SPT, because it offers a good chance of long-term survival.

2. Case report

The patient was a 22-year-old female, who presented with 2 days history of left lower abdominal pain. She was taken to the local hospital, and the computed tomography scan revealed a large occupying lesion in the peripancreatic clearance. She was treated with antibiotics and intravenous fluid therapy, but the symptom still advanced. Then she was admitted to the emergency department of our hospital. Physical examination revealed the

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XX, DC, and LC are joint first authors.

Patient has provided informed consent for publication of the case.

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Informed written consent was obtained from the patient for publication of this case report and accompanying images.

The authors have no conflicts of interests to disclose.

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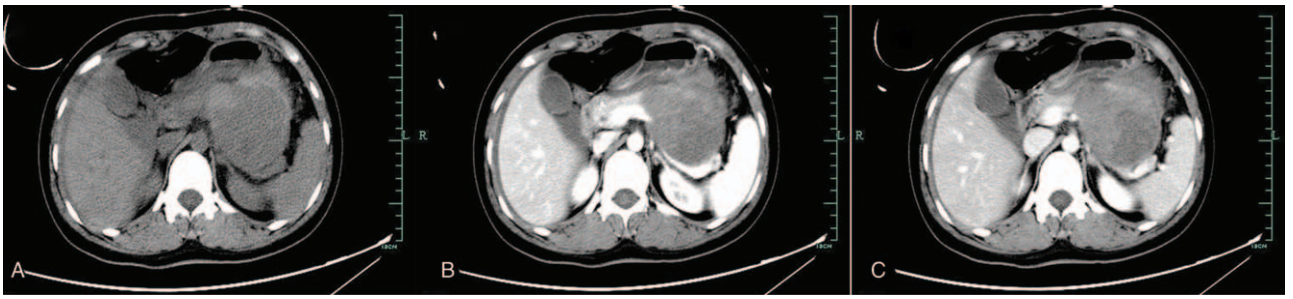

Figure 1. The CT scan demonstrated a cystic lesion (about 95 × 75 mm) located in the pancreas.

tenderness and rebound-tenderness of the whole abdomen and abdominal muscular tension was obvious. Laboratory findings revealed elevated leukocytosis (12.3×10^9/L), neutrophile granulocytes (86.3% of the leukocytes) and decreased hemoglobin (Hb) (105 g/L). Then a CT scan was performed again to assess the properties of the abdominal lesion. The review result of the CT scan showed a hemorrhagic complex solid cystic mass located in the lesser omentum sac, which was considered to be originated from pancreas (Fig. 1).

In order to stop bleeding in time, the patient underwent emergency excision laparotomy of the cyst based on clinical and radiological findings. We located the tumor in the body of the pancreas (about 8 × 7 cm), and it had invaded to the spleen. Therefore, distal pancreatectomy (including the cyst, the body and tail of pancreas) and splenectomy were performed. The pathology results reported a solid-cystic mass (8 × 6.5 × 5 cm) with heterogenesis. Histological examination indicated that a solid and vascular pattern with pseudo-papillary cores (Fig. 2).

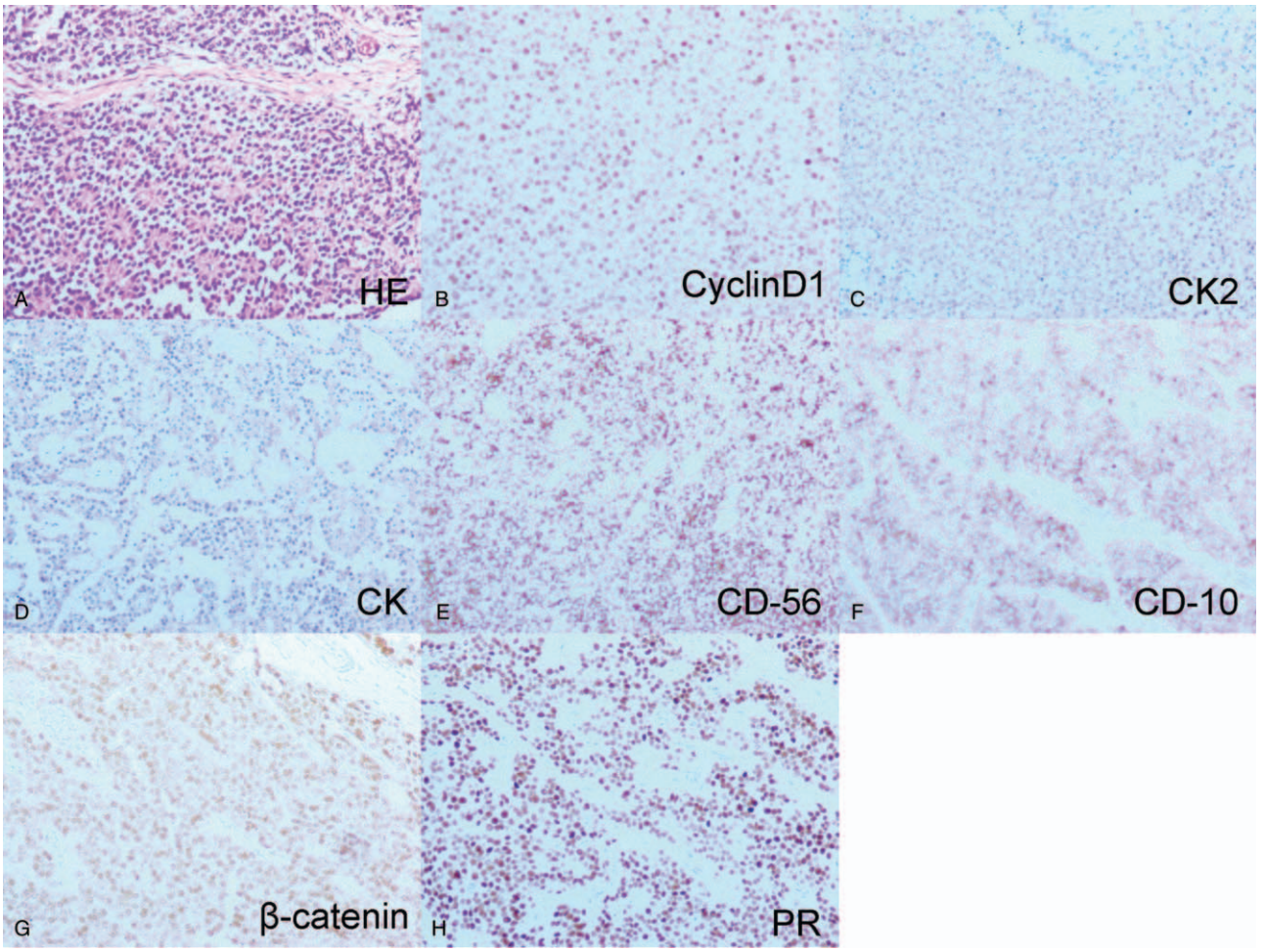

Figure 2. A. H&E staining shows that gland-like structure was lined by round tumor cells in solid pseudopapillary tumors specimen (original magnification × 100). B–H: The immunohistochemical results of solid pseudopapillary tumors specimen.

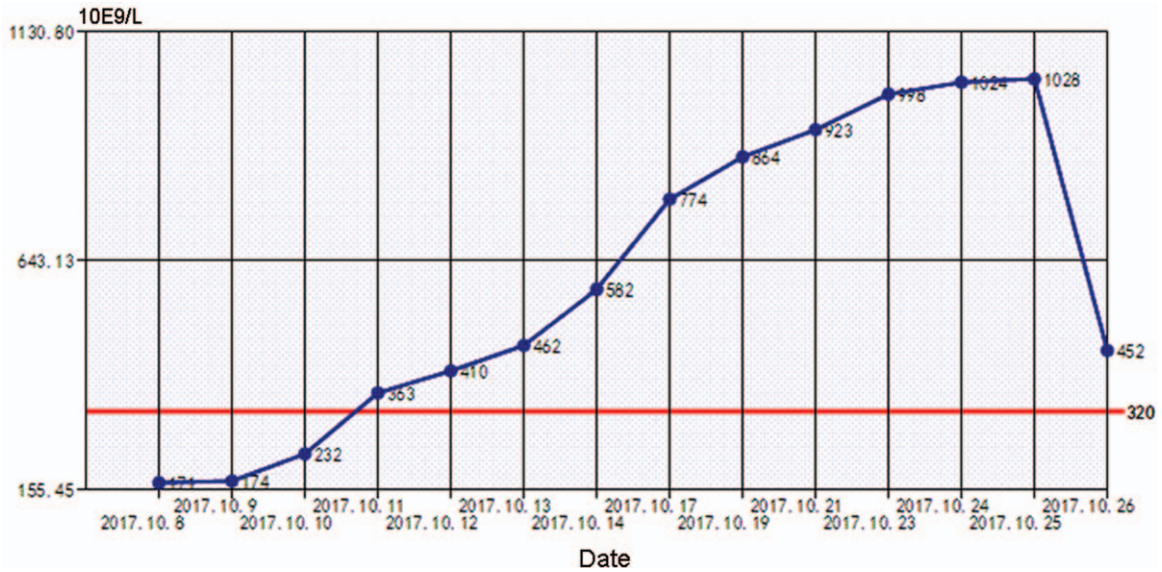

Figure 3. Trend of platelet change after surgery of this patient.

Immunohistochemical (IHC) stains for CK (pan), CD-10, PR, CyclinD1, CD-56 and β-Catenin showed positivity, but CgA and E-cadherin staining were negative. On the whole, final pathological diagnosis was solid pseudopapillary tumor of the pancreas. In the postoperative period, the patient had high level of platelet (PLT) (12.3×10^9/L, Fig. 3), and it was thought to be induced by the resection of spleen. After treated by acute preoperative plateletpheresis, PLT level came back to normal. A follow-up 10 months later showed neither signs of tumor recurrence nor endocrine and exocrine insufficiency of the pancreas (Fig. 4).

3. Discussion

3.1. Description of solid pseudopapillary tumors of pancreas

SPT is an uncommon and enigmatic pancreatic neoplasm firstly described by Frantz in 1959,[3] which is considered to be low malignant potential.[6] It represents 0.2% to 2.7% of tumors in the pancreas.[7] Frequently, it is identified as solid and cystic tumor, solid and papillary epithelial neoplasm, papillary-cystic neoplasm, papillary-cystic epithelial neoplasm, papillary-cystic tumor or Franz tumor. The concept of SPT for the international

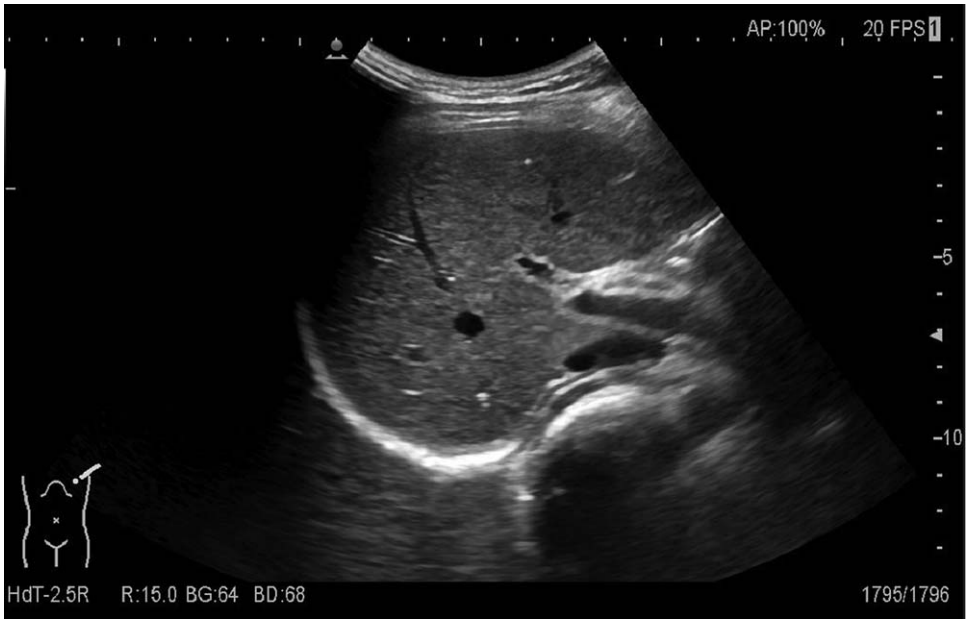

Figure 4. Abdominal ultrasound scan at 10 months after operation.

histological classification of tumors of the exocrine pancreas was firstly put forward by the World Health Organization (WHO) in 1996.[8] Then we collected 50 SPT cases from pubmed database to ensure the characters of SPT. Until now, the cases reported showed that SPT commonly occurs in the head or tail region of pancreas. As shown in Table 1, 17 of the cases had the SPT in the head of pancreas. While, 5 reported cases had SPT in the body, and 8 cases occurred in the tail. The remaining cases reported the tumor located in the head-body junction (4/50) or, in the body-tail junction of pancreas (10/50).

3.2. Spontaneous rupture of solid pseudopapillary tumors of pancreas

The symptoms of SPT are usually nonspecific, with abdominal pain being the most common. SPT which was discovered after rupture and hemoperitoneum was rare.[9] According to the cases

Table 1

Summary information of the 50 cases reviewed in literatures.

Year	Authors	Nation	Sex	Age	Size	Clinical Treatment
2018	Hooper et al[19]	Spain	Female	31	1.7 × 1.4 cm	pancreatoduodenectomy
2018	Liang B et al[20]	Spain	Female	35	2 × 2 cm	distal pancreatectomy with splenectomy
2018	Estifan et al[21]	USA	Female	15	10 × 12 cm	distal pancreatectomy
2018	Burk et al[22]	India	Female	12	NM	distal pancreatectomy with splenectomy
2018	Filatov et al[23]	USA	Female	27	2.1 x 1.8 cm	distal pancreatectomy with splenectomy
2018	Bender et al[24]	India	Female	49	8.2 × 14 × 11 cm	tumor resection
2018	Lanke et al[25]	Brazil	Female	19	3.0 × 2.2 cm	pancreatoduodenectomy
2017	Kim SS et al[26]	Brazil	Female	18	4.1 × 3.3 cm	pancreatoduodenectomy
2017	Lawlor et al[27]	Sri Lanka	Female	19	15 × 18cm	distal pancreatectomy with splenectomy
2017	Kriger et al[28]	Poland	Female	15	4.8 × 4.2 × 5 cm	pancreatoduodenectomy
2017	Gozdowska et al[29]	Poland	Female	12	5.3 × 3.2 × 5 cm	distal pancreatectomy with splenectomy
2017	Azagoh-Kouadio et al[30]	France	Female	69	6 × 4 cm	pancreatoduodenectomy
2017	Ruzzenente et al[31]	Russia	Female	31	5 × 4.5 cm	tumor resection
2017	Aikot S et al[32]	Qatar	Female	32	5.1 × 4.6 cm	distal pancreatectomy with splenectomy
2016	Yang et al[33]	Turkey	Female	13	9 × 7.2 cm	distal pancreatectomy
2016	Chinnusamy et al[34]	USA	Female	27	5.6 × 4.5 cm	pancreatoduodenectomy
2016	Michalova et al[35]	India	Female	18	10 × 8 × 6 cm	distal pancreatectomy with splenectomy
2015	Coronel et al[36]	Brazil	Female	47	11 × 6 × 9 cm	distal pancreatectomy
2015	Šnajdauf et al[37]	Argentina	Female	28	2.8 cm in diameter	distal pancreatectomy with splenectomy
2015	Xiang et al[38]	China	Female	25	4.5 cm in diameter	distal pancreatectomy
2015	Sharma et al[39]	India	Male	10	11 × 10 × 9.2 cm	tumor resection
2015	Wu et al[40]	China	Female	19	14.8 × 8.9 cm	distal pancreatectomy with splenectomy
2012	Cho et al[41]	Korea	Male	10	11 × 10 cm	NM
2012	Manfredi et al[42]	Porland	Female	39	15 cm in diameter	distal pancreatectomy
2012	Luchini et al[43]	Porland	Female	36	17 × 13 × 8 cm	distal pancreatectomy
2012	Jung et al[44]	China	Female	24	4.0 × 5.0 cm	distal pancreatectomy
2012	Park et al[45]	Portugal	Female	35	4 cm	tumor resection
2010	Sandlas et al[46]	Greece	Female	55	5 cm	distal pancreatectomy with splenectomy
2010	Lee et al[47]	Italy	Female	15	12 × 13 × 10 cm	distal pancreatectomy with splenectomy
2010	Nishida et al[48]	Japan	Female	32	1.5 cm	pancreatoduodenectomy
2009	Dumitru et al[49]	Iran	Female	17	NM	pancreatoduodenectomy
2009	Jiang et al[50]	Mexico	Female	37	NM	distal pancreatectomy with splenectomy
2006	Branco et al[51]	Japan	Female	26	5 × 4.5 × 5.2 cm	pancreatoduodenectomy
2006	Murayama et al[52]	USA	Female	20	6.1 × 6.6 cm	distal pancreatectomy with splenectomy
2006	Fukuda et al[53]	Turkey	Female	27	14 × 16 cm	distal pancreatectomy with splenectomy
2006	Dalla Bona et al[54]	Tunisia	Female	14	6.8 × 6.3 cm	distal pancreatectomy
2006	Okamoto et al[55]	Turkey	Female	29	11 cm in diameter	distal pancreatectomy with splenectomy
2004	Lakhtakia et al[56]	India	Female	22	5 × 4 × 4 cm	EUS-FNA
2004	Hanada et al[57]	Japan	Female	33	2.5 × 2.5 × 2cm	NM
2004	Kanngurn et al[58]	Italy	Female	49	10 × 10 × 9.5 cm	distal pancreatectomy
2004	Karamarković et al[59]	Greece	Female	17	6.5 × 5.4 mm	EUS-FNA
2004	Aydiner et al[60]	Turkey	Female	29	8.5 cm in diameter	pancreatoduodenectomy
2004	Ulusan et al[61]	Turkey	Female	17	NM	pancreatoduodenectomy
2004	Levy et al[62]	USA	Male	31	9 cm	pancreatoduodenectomy
2002	Mancini et al[63]	USA	Male	43	11 cm	distal pancreatectomy with splenectomy
2002	Mancini et al[63]	USA	Female	47	3.5 × 2.3 cm	distal pancreatectomy
2002	Coleman et al[64]	USA	Female	28	4.5 × 3.0 cm	distal pancreatectomy
2001	Potrc et al[2]	Slovenia	Female	14	10 × 10cm	pancreatoduodenectomy
2001	Molino et al[65]	Italy	Female	67	2 cm	pancreatoduodenectomy
2001	Casanova et al[66]	Italy	Female	44	6 cm	pancreatoduodenectomy

EUS-FNA = endoscopic ultrasonography guided fine needle aspiration, NM = not mentioned.

reported, most ruptured SPTs are induced by the blunt abdominal trauma, and spontaneous ruptures seem to be quite uncommon. Since the cystic part of SPT consisted of the degeneration after the intramural hemorrhage, SPT had a natural tendency to hemorrhage inside the tumor.[10] Abrupt massive hemorrhage and increased pressure of the tumor are considered to be the main reasons for the spontaneous rupture of SPT. When we face with such patient, the enhanced CT and emergency laparotomy would be helpful to make a correct diagnosis.

3.3. Clinical findings of solid pseudopapillary tumors of pancreas

We reviewed 50 case reports of SPT in literatures. The patients included 46 females and 4 males, the ages ranged from 10 to 69, and the mean age was 31.5 years old (Table 1). Totally, SPT, as an uncommon, typically benign tumor, is found mainly in young non-Caucasian women between the 2nd and 3rd decades of life. In addition, we found that most of the patients were Asians (20/50) and Americans (18/50), and Europeans comprised 11/50, respectively (Table 2). This observation suggests that it seems to have a predilection for Asian and American women, although rare cases have been reported in children and men. Some experts considered that female predominance may attribute to the proximity of primordial pancreatic cells to the ovarian ridge during the development of SPT.

Table 2

Epidemiological and pathological results of the 50 cases in the literature review.

Item	Statistical result
Age range (years)	10–69 (31.5)
Sex (F:M)	46:4
Location	
Head	17
Body	5
Tail	8
Body-tail junction	10
Head-body junction	4
All of pancreas	0
Not described	6
Race	
Asian	20
American	18
European	11
Other Races	1
Symptoms	
Abdominal pain	24
Abdominal pain/nause/vomitting	7
Painless mass	8
Abdominal distension	3
Jaundice	1
Not described	7
Treatment	
EUS-FNA	2
Distal pancreatectomy	11
Distal pancreatectomy with splenectomy	16
Pancreaticoduodenectomy	15
Tumor resection	2
Others	3
Not described	1

EUS-FNA = endoscopic ultrasonography guided fine needle aspiration.

Clinical presentation of solid pseudopapillary tumor in pancreas is various. Abdominal discomfort or vague pain can be the most common symptom, but the minority of patients can be also asymptomatic and the tumors are detected incidentally. However, some symptoms including vomiting, discomfort in the epigastrium or jaundice occur more rarely.[11] We collected about 50 cases of SPT between 2018 to 2001 (Table 1). As the reported cases showed (Table 2), most patients presented with abdominal pain (24/50), and others complained with the symptoms of nausea, vomiting (7/50), abdominal distension (3/50), or jaundice (1/50). However, when the tumors invade to neighboring organs, such as the adrenal glands, patients present with specific clinical manifestations, such as acute kidney injury (AKI) with rhabdomyolysis. Therefore, the clinical presentations may be the necessary clues to find the origin of the tumor and make the differential diagnosis.

3.4. Diagnosis of solid pseudopapillary tumors of pancreas

Image examination is significant in diagnosis of SPT. On X-ray, solid pseudopapillary tumors appear as large masses, which sometimes could displace adjacent structures like stomach or bowels. The majority of tumors are diagnosed through ultrasound or CT scan of the abdomen. Ultrasound shows a well-defined mass with solid and cystic components and increased vascularity.[12,13] Besides, CT imaging is superior for the diagnosis of SPT. Through contrast enhanced CT, it shows an encapsulated lesion with enhancing solid and non-enhancing cystic areas with some showing calcific foci. If spontaneous bleeding was occurred in the tumor, the hemorrhagic density can be found within the lesion. Solid pseudopapillary tumors may grow to large sizes with a mean diameter ranging from 6 cm to 10 cm.[14] Through magnetic resonance image (MRI), it reveals the hyper-vascular, well-encapsulated, round tumors with mixed cystic and solid components. Furthermore, echo-endosonography may provide FNA biopsy with the possibility of pre-operative pathologic diagnosis. So SPT requires differential diagnosis to be made with other pancreatic tumors, such as mucinous neoplasm, serous cystadenoma, pseudocyst, nonhyperfunctioning islet cell tumor and pancreatic adenocarcinoma (Table 3). Thus, to ensure a better surgical approach, a clear preoperative diagnosis of SPT is preferable.

As for the confusion between pancreatic SPTs and cystic neoplasms, histological differential diagnosis is crucial. SPT is composed of poorly cohesive, monomorphic cells forming solid, and pseudopapillary structures are lined by neoplastic cells. Moreover, cystic spaces containing blood and necrotic debris.[15] Histologically, the neoplastic cells are characteristically strongly positive for vimentin, α1-antitrypsin, α1-antichymotrypsin, CD-10, progesteron receptor (PR), neuronspecific enolase, CD-56 and cyclin D1.[6,16] In our review, we found that neoplastic cells were strongly and diffusely positive for vimentin in 12 cases, positive for CD-10 in 10 cases. Besides, a total of 9 patients in our review showed positive results for PR expression and 8 patients showed positive results for α1-antitrypsin expression (Table 4). Thus, the combination of multiple IHC markers including vimentin, CD-10, PR andα1-antitrypsin, may contribute to improving the diagnosis rate of SPT.

3.5. Treatments for solid pseudopapillary tumors of pancreas

Despite the large tumor size at the time of diagnosis, surgery is the preferred treatment option for solid pseudopapillary tumors of

Table 3

Immunohistochemical markers of the SPT specimens in the literature review.

Author	CK	Vimentin	CHROMO	NSE	CD10	CD56	SYNAPTO	Ki67	PR	β-catenin
Bender Am et al[24]	NM	NM	+	NM	NM	+	+	NM	+	+
Lanke G et al[25]	+	+	NM	NM	+	+	NM	NM	NM	+
Kim SS et al[26]	NM	NM	-	NM	NM	NM	NM	NM	NM	+
Lawlor RT et al[27]	NM	NM	NM	NM	+	NM	NM	NM	NM	NM
Kriger AG et al	NM	NM	NM	NM	+	+	NM	NM	NM	NM
Gozdowska J et al[28]	NM	NM	NM	NM	+	+	NM	NM	+	+
Azagoh-Kouadio R et al[30]	NM	+	NM	NM	NM	NM	NM	NM	NM	NM
Ruzzenente A et al[31]	NM	NM	NM	NM	NM	NM	NM	+	NM	NM
Aikot S et al[32]	NM	+	NM	NM	+	+	NM	NM	NM	+
Yang JR et al[33]	NM	NM	+	NM	NM	NM	NM	NM	+	NM
Chinnusamy P et al[34]	NM	+	NM	+	+	NM	+	+	NM	NM
Michalova K et al[35]	NM	NM	+	NM	+	NM	+	+	+	NM
Coronel M et al[36]	-	+	-	+	NM	NM	-	NM	+	NM
Šnajdauf J et al[37]	NM	+	-	NM	+	NM	NM	NM	NM	NM
Xiang D et al[38]	NM	+	+	NM	+	+	NM	NM	+	+
Sharma M et al[39]	NM	+	NM	-	NM	NM	NM	NM	+	NM
Wu H et al[40]	+	NM	NM	NM	NM	NM	+	NM	NM	NM
Cho YJ et al[41]	NM	+	NM	NM	NM	NM	NM	NM	NM	NM
Manfredi R et al[31]	+	NM	NM	+	NM	NM	NM	NM	NM	NM
Luchini C et al[43]	NM	+	NM	+	+	NM	NM	NM	NM	NM
Jung MJ et al[44]	NM	+	+	+	NM	NM	+	NM	NM	NM
Park HJ et al[45]	-	+	-	NM	NM	NM	+	NM	+	NM
Coleman KM et al[31]	NM	NM	-	NM	NM	NM	+	-	+	NM

NM = Not Mentioned, SPT = solid pseudopapillary tumors.

Table 4

Differential diagnosis of SPT.

	Local Morphology	US	CT	MRI
Solid pseudopapillary tumor	Head and tail	Wellcircumscribed, heterogeneous (hypoechoic solid and anechoic cystic components) and hypovascular	Well-circumscribed, encapsulated, round or lobulated lesion. Variable internal architecture (solid, mixed solid and cystic, thick-walled cyst) depending on the degree of hemorrhagic necrosis.	Degradation blood products: high SI onT1-wi,low or inhomogeneou s SI on T2-wi Solid component without hemorrhagic foci: low SI on T1-wi, high SI on T2-wi Fibrous capsule: low SI on T1 and T2-wi
Mucinous neoplasm	Tail (Uni/ multilocular cyst)	Large unilocular or multilocular cysts	Water HU of the cyst Distant metastasis (mucinous adenocarcinoma)	High SI on T1 and T2-wi (but variable with concentration)
Serous cystadenoma	Body and tail (small cysts)	Multiple milimetric hypoechoic or anechoic cysts	Honeycomb pattern of multiple milimetric cysts	Hypointense on T1 and hyperintense on T2-wi clustered cysts
Pseudocyst	Head 50% (Large or small Uni/ multilocular cysts)	Uni/Multiple cysts of of the upper abdomen	Round or lobulated like low-density area	High SI on T1wi and T2wi
Lymphangioma	Head and body (cystic/ cavernous mass)	Multilocular lesions with internal septa	Homogeneous cystic mass with thin walls and multiple fine intervals,and the wall of the tumour may be enhanced after intravenous contrast-administration	Cystic spaces appear hypointense on T1-weighted images and hyperintense on T2-weighted images.
Nonhyperfunctioning islet cell tumors	Small or large in size	Homogeneously hypoechoic lesion	Isoattenuating to the parenchyma Distant metastasis	Low SI or isointensity on T1-wi High to isointense on T2-wi
Pancreatic adenocarcinoma	Head (Lesion with contour deformity of the gland)	Hypoechoic lesion Dilated pancreatic duct Atrophic gland	Isodense to the parenchyma Dilated pancreatic duct and atrophic gland Obliteration of peripancreatic fat Contiguous organ invasion, vascular invasion and distant metastases	Low SI on T1wi Variable SI on T2-wi Contiguous organ invasion and distant metastases

pancreas. In addition, complete aggressive resection is necessary for SPT. Based on the morphology and size of the tumor, different surgical options, including a simple excision of the mass or a pancreatic resection, such as pancreaticoduodenectomy or distal pancreatectomy, must be considered.[17] Due to the potential malignancy, liver is the most common metastatic site of SPT. For liver metastases, other treatments include chemotherapy, alcohol injection, transcatheter arterial chemoembolization, radiotherapy, and liver transplantation could be considered.[18] However, distant or local recurrences still could occur in some cases after surgical resection. The recent studies showed that moreover repeated surgical resection for recurrences can considerable prolong survival. This case was admitted to our hospital for acute hemorrhage, emergency surgery was performed. However, as SPT is a potential benign and malignant borderline tumor, the choice of emergency surgery may cause high risk for recurrence of the patient. Therefore, when it is possible to stabilize the condition of patient with conservative treatment, elective surgery may be a better choice.

Author contributions

Investigation: Linping Cao, Xiaode Feng.
Methodology: Linping Cao.
Resources: Diyu Chen.
Writing – original draft: Xiaofeng Xu, Diyu Chen, Xiaode Feng, Jian Wu.
Writing – review & editing: Rongliang Tong, Shusen Zheng, Jian Wu.

References

[1] Mao C, Guvendi M, Domenico DR, et al. Papillary cystic and solid tumors of the pancreas: a pancreatic embryonic tumor? Studies of three cases and cumulative review of the world's literature. Surgery 1995;118:821–8.
[2] Potrc S, Kavalar R, Horvat M, et al. Urgent whipple resection for solid pseudopapillary tumor of the pancreas. J Hepatobiliary Pancreat Surg 2003;10:386–9.
[3] Papavramidis T, Papavramidis S. Solid pseudopapillary tumors of the pancreas: review of 718 patients reported in English literature. J Am Coll Surg 2005;200:965–72.
[4] Frattaroli FM, Proposito D, Conte AM, et al. Assessment of guidelines to improve diagnosis and treatment of solid pseudopapillary tumor of the pancreas. A case report and literature review. Ann Ital Ch 2009;80:29–34.
[5] Hu S, Lin X, Song Q, et al. Multidetector CT of multicentric solid pseudopapillary tumor of the pancreas: a case report and review of the literature. Cancer Imaging 2011;11:175–8.
[6] Kim CW, Han DJ, Kim J, et al. Solid pseudopapillary tumor of the pancreas: can malignancy be predicted? Surgery 2011;149:625–34.
[7] Kloppel G, Maillet B. Histological typing of pancreatic and periampullary carcinoma. Eur J Surg Oncol 1991;17:139–52.
[8] Lestelle V, de Coster C, Sarran A, et al. Solid pseudopapillary tumor of the pancreas: one case with a metastatic evolution in a Caucasian woman. Case Rep Oncol 2015;8:405–8.
[9] Huang SC, Wu TH, Chen CC, et al. Spontaneous rupture of solid pseudopapillary neoplasm of the pancreas during pregnancy. Obstet Gynecol 2013;121:486–8.
[10] Sandlas G, Tiwari C. Solid pseudopapillary tumor of pancreas: a case report and review of literature. Indian J Med Paediatr Oncol 2017;38:207–9.
[11] Permi HS, Kishan Prasad HL, Shetty BN, et al. Solid pseudopapillary tumor of pancreas with sickle cell trait: a rare case report. J Cancer Res Ther 2013;9:537–40.
[12] Kim B, Lee SS, Sung YS, et al. Intravoxel incoherent motion diffusion-weighted imaging of the pancreas: Characterization of benign and malignant pancreatic pathologies. J Magn Reson Imaging 2017;45:260–9.
[13] Jiang L, Cui L, Wang J, et al. Solid pseudopapillary tumors of the pancreas: findings from routine screening sonographic examination and the value of contrast-enhanced ultrasound. J Clin Ultrasound 2015;43:277–82.
[14] Watanabe D, Miura K, Goto T, et al. Solid pseudopapillary tumor of the pancreas with concomitant pancreas divisum. A case report. JOP 2010;11:45–8.
[15] Guo M, Luo G, Jin K, et al. Somatic genetic variation in solid pseudopapillary tumor of the pancreas by whole exome sequencing. Int J Mol Sci 2017;18.
[16] Tian G, Savell VH, Esquilin JM. Solid pseudopapillary tumor of pancreas: a case report and review of genetic features. Pediatr Blood Cancer 2018;65:e26980.
[17] Leonher-Ruezga K, Lopez-Espinosa S, Moya Herraiz A, et al. Solid pseudopapillary tumor of the pancreas: case report and review of the literature. Cir Esp 2015;93:e37–40.
[18] Miloudi N, Hefaiedh R, Marzouk I, et al. Solid pseudopapillary tumor of the pancreas with concomitant Bochdalek's hernia. The first reported case. Tunis Med 2015;93:184–6.
[19] Hooper K, Tracht JM, Eldin-Eltoum IA. Cytologic criteria to reduce error in EUS-FNA of solid pseudopapillary neoplasms of the pancreas. J Am Soc Cytopathol 2017;6:228–35.
[20] Liang B, Chen Y, Li M, et al. Total laparoscopic duodenum-preserving pancreatic head resection for solid pseudopaillary neoplasm of pancreas: a case report. Medicine 2019;98:e15823.
[21] Estifan E, Cavanagh Y, Kapoor A, et al. Pancreatic solid pseudopapillary tumor associated with elevated DHEA and testosterone. Case Rep Gastrointest Med 2019;2019:8128376.
[22] Burk KS, Knipp D, Sahani DV. Cystic pancreatic tumors. Magn Reson Imaging Clin N Am 2018;26:405–20.
[23] Filatov AV, Smolyannikova VA. A case of solid pseudopapillary tumor of the pancreas: features of the course, difficulty in diagnosis. Arkh Patol 2018;80:46–50.
[24] Bender AM, Thompson ED, Hackam DJ, et al. Solid pseudopapillary neoplasm of the pancreas in a young pediatric patient: a case report and systematic review of the literature. Pancreas 2018;47:1364–8.
[25] Lanke G, Ali FS, Lee JH. Clinical update on the management of pseudopapillary tumor of pancreas. World J Gastrointest Endosc 2018;10:145–55.
[26] Kim SS, Choi GC, Jou SS. Pancreas ductal adenocarcinoma and its mimics: review of cross-sectional imaging findings for differential diagnosis. J Belg Soc Radiol 2018;102:71.
[27] Lawlor RT, Dapra V, Girolami I, et al. CD200 expression is a feature of solid pseudopapillary neoplasms of the pancreas. Virchows Arch 2019;474:105–9.
[28] Kriger AG, Smirnov AV, Berelavichus SV, et al. Diagnosis and treatment of duodenal dystrophy in patients with chronic pancreatitis. Khirurgiia 2016;25–32.
[29] Wojciak M, Gozdowska J, Pacholczyk M, et al. Liver transplantation for a metastatic pancreatic solid-pseudopapillary tumor (frantz tumor): a case report. Ann Transplant 2018;23:520–3.
[30] Azagoh-Kouadio R, Couitchere LG, Kouyate M, et al. Rare pancreatic tumor detected unexpectedly in a child in the Ivory Coast. Pan Afr Med J 2018;29:171.
[31] Cingarlini S, Ortolani S, Salgarello M, et al. Role of combined 68Ga-DOTATOC and 18F-FDG positron emission tomography/computed tomography in the diagnostic workup of pancreas neuroendocrine tumors: implications for managing surgical decisions. Pancreas 2017;46:42–7.
[32] Aikot S, Manappallil RG, Pokkattil S, et al. Solid pseudopapillary neoplasm of pancreas: an unusual aetiology for haematochezia. BMJ Case Rep 2018;2018: bcr-2018-225332.
[33] Yang JR, Xiao R, Zhou J, et al. Endoscopic linear stapler-assisted resection of a giant solid pseudopapillary pancreatic tumor with concurrent regional portal hypertension: a case report. J Int Med Res 2018;46:3000–8.
[34] Palanisamy S, Deuri B, Naidu SB, et al. Hepatic artery reconstruction following iatrogenic injury during laparoscopic distal pancreatectomy: minimal access surgery is new horizon. J Minim Access Surg 2016;12:382–4.
[35] Michalova K, Michal M, Sedivcova M, et al. Solid pseudopapillary neoplasm (SPN) of the testis: comprehensive mutational analysis of 6 testicular and 8 pancreatic SPNs. Ann Diagn Pathol 2018;35:42–7.
[36] De Moura DTH, Coronel M, Ribeiro IB, et al. The importance of endoscopic ultrasound fine-needle aspiration in the diagnosis of solid pseudopapillary tumor of the pancreas: two case reports. J Med Case Rep 2018;12:107.

[37] Snajdauf J, Petru O, Nahlovsky J, et al. Pancreas divisum in children and duodenum-preserving resection of the pancreatic head. Eur J Pediatr Surg 2018;28:250–4.
[38] Xiang D, He J, Fan Z, et al. Situs inversus totalis with solid pseudopapillary pancreatic tumor: a case report and review of literature. Medicine 2018;97:e0205.
[39] Sharma M, Somani P, Sunkara T, et al. Endoscopic ultrasound-guided management of bleeding periampullary tumor. Endoscopy 2018;50: E192–3.
[40] Wu H, Zou WB, Zhou DZ, et al. Longer leukocyte telomere length is associated with an increased risk of chronic pancreatitis. Pancreas 2017;46:e65–6.
[41] Cho YJ, Namgoong JM, Kim DY, et al. Suggested Indications for enucleation of solid pseudopapillary neoplasms in pediatric patients. Front Pediatr 2019;7:125.
[42] European evidence-based guidelines on pancreatic cystic neoplasms. Gut 2018;67:789–804.
[43] Riva G, Pea A, Pilati C, et al. Histo-molecular oncogenesis of pancreatic cancer: From precancerous lesions to invasive ductal adenocarcinoma. World J Gastrointest Oncol 2018;10:317–27.
[44] Jung MJ, Kim HK, Choi SY, et al. Solid pseudopapillary neoplasm of the pancreas with liver metastasis initially misinterpreted as benign haemorrhagic cyst. Malays J Pathol 2017;39:327–30.
[45] Park HJ, Sung YS, Lee SS, et al. Intravoxel incoherent motion diffusion-weighted MRI of the abdomen: the effect of fitting algorithms on the accuracy and reliability of the parameters. J Magn Reson Imaging 2017;45:1637–47.
[46] Desale J, Shah H, Kumbhar V, et al. Desmosis coli - a case report and review of the literature. Dev Period Med 2017;21:390–2.
[47] Lee JS, Han HJ, Choi SB, et al. Surgical outcomes of solid pseudopapillary neoplasm of the pancreas: a single institution's experience for the last ten years. Am Surg 2012;78:216–9.
[48] Nishida T, Takeno S, Nakashima K, et al. Salvage photodynamic therapy accompanied by extended lymphadenectomy for advanced esophageal carcinoma: a case report. Int J Surg Case Rep 2017;36:155–60.
[49] Chirita D, Calita M, Grasu M, et al. Metachronous ampulla of vater carcinoma after curative-intent surgery for klatskin tumor. Chirurgia (Bucur) 2015;110:379–83.
[50] Jiang YJ, Lee CL, Wang Q, et al. Establishment of an orthotopic pancreatic cancer mouse model: cells suspended and injected in Matrigel. World J Gastroenterol 2014;20:9476–85.
[51] Branco C, Vilaca S, Falcao J. Solid pseudopapillary neoplasm-case report of a rare pancreatic tumor. Int J Surg Case Rep 2017;33:148–50.
[52] Iitaka D, Ikoma H, Kawaguchi T, et al. A case report–locally advanced pancreatic adenocarcinoma was resected after chemotherapy. Gan To Kagaku Ryoho 2010;37:2358–60.
[53] Fukuda S, Oussoultzoglou E, Bachellier P, et al. Significance of the depth of portal vein wall invasion after curative resection for pancreatic adenocarcinoma. Arch Surg 2007;142:172–9. discussion 80.
[54] Dalla Bona E, Beltrame V, Blandamura S, et al. Huge cystic lymphangioma of the pancreas mimicking pancreatic cystic neoplasm. Case Rep Med 2012;2012:951358.
[55] Okamoto H, Hosaka M, Fujii H, et al. Successful management of a blunt pancreatic trauma by endoscopic stent placement. Clin J Gastroenterol 2010;3:204–8.
[56] Lakhtakia S, Gupta R, Ramchandani M, et al. Endoscopic ultrasound-guided biliary stent placement using Soehendra stent retriever. Indian J Gastroenterol 2007;26:178–9.
[57] Hanada K, Hino F, Amano H, et al. Current treatment strategies for pancreatic cancer in the elderly. Drugs Aging 2006;23:403–10.
[58] Kanngurn S, Somran J, Art-Ong C, et al. Primary peritoneal adenosarcoma with stromal overgrowth and fetal type cartilage: a case report and literature review. J Med Assoc Thai 2005;88:849–54.
[59] Karamarkovic AR, Micev M, Culafic D, et al. Solid cystic pseudopapillary tumor of the pancreas. Srp Arh Celok Lek 2004;132:431–4.
[60] Aydiner F, Erinanc H, Savas B, et al. Solid pseudopapillary tumor of the pancreas: emphasis on differential diagnosis from aggressive tumors of the pancreas. Turk J Gastroenterol 2006;17:219–22.
[61] Ulusan S, Bal N, Kizilkilic O, et al. Case report: solid-pseudopapillary tumour of the pancreas associated with dorsal agenesis. Br J Radiol 2005;78:441–3.
[62] Levy C, Pereira L, Dardarian T, et al. Solid pseudopapillary pancreatic tumor in pregnancy. A case report. J Reprod Med 2004;49:61–4.
[63] Mancini GJ, Dudrick PS, Grindstaff AD, et al. Solid-pseudopapillary tumor of the pancreas: two cases in male patients. Am Surg 2004;70:29–31.
[64] Coleman KM, Doherty MC, Bigler SA. Solid-pseudopapillary tumor of the pancreas. Radiographics: a review publication of the Radiological Society of North America, Inc 2003;23:1644–8.
[65] Molino D, Perrotti P, Napoli V, et al. Solid pseudopapillary tumour of the pancreas. Report of a case. Minerva Chir 2003;58:815–21.
[66] Casanova M, Collini P, Ferrari A, et al. Solid-pseudopapillary tumor of the pancreas (Frantz tumor) in children. Med Pediatr Oncol 2003;41:74–6.