

# World Journal of *Clinical Cases*

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## Pancreatic panniculitis and solid pseudopapillary tumor of the pancreas: A case report

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### Abstract

Solid pseudopapillary tumor of the pancreas (SPTP), also known as solid and papillary epithelial neoplasm of the pancreas, is a rare pancreatic exocrine tumor that is difficult to diagnose before surgery. Pancreatic panniculitis is a rare type that occurs in less than 3% of all patients with pancreatic diseases. We here report a 19-year-old woman who presented with persistent left upper quadrant pain without obvious cause for 1 d. The patient also developed subcutaneous nodules involving lower abdomen bilaterally and lower limbs, and subcutaneous nodules were pathologically diagnosed as pancreatic panniculitis. Plain abdominal computed tomography revealed a soft-tissue mass in the body and tail of the pancreas, which was closely associated with the gastric wall. Contrast-enhanced ultrasound showed inhomogeneous echogenicity in the anterior pancreatic body, which had blurred parenchymal demarcation of the body and tail of the pancreas. Contrast-enhanced abdominal computed tomography revealed a mixed density mass with solid and cystic components in the body and tail of the pancreas, and the solid component was markedly enhanced. The lesion was pathologically diagnosed as SPTP after laparoscopic resection. Clinicians should be aware of the clinical manifestation, diagnosis, and treatment of pancreatic panniculitis and SPTP.



**Key words:** Case report; Pancreatic panniculitis; Solid pseudopapillary tumor of the pancreas; Subcutaneous nodules; Laparoscopy

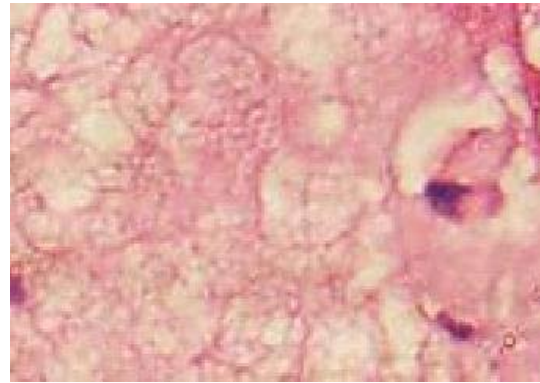
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**Core tip:** Pancreatic panniculitis is a rare complication of pancreas diseases, and solid pseudopapillary tumor of the pancreas is a rare pancreatic exocrine tumor that is difficult to diagnose before surgery. We describe the diagnosis and treatment of solid pseudopapillary tumor of the pancreas accompanied with pancreatic panniculitis in a young woman. Plain abdominal computed tomography, contrast-enhanced ultrasound, contrast-enhanced abdominal computed tomography and pathological examinations were performed.

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## INTRODUCTION

Pancreatic panniculitis is a rare complication of pancreas disorders occurring in 1%-3% of patients, most often accompanied by the pancreatic carcinoma. It often presents multiple and red-brown subcutaneous nodules. The pathogenesis is not fully understood, but it is thought to result from lipolysis and fat necrosis with secondary tissue inflammation induced by pancreatic enzymes<sup>[1]</sup>. Solid pseudopapillary tumor of the pancreas (SPTP) often occurs in young women, with an average age of 25 years. While the histogenetic origin of SPTP is still unclear, it may arise from the cells related with the gonadal ridge/ovarian primordium during embryogenesis or the embryonic neural crest cells. SPTP exhibits two histological features: Solid and pseudopapillary. In fact, the papillary structure is composed of pseudopapillae that are formed due to degeneration of tumor cells, decreased adhesion of cells, and cystic cavity formation. Clinically, it is usually manifested as an epigastric mass or epigastric pain, and some patients present with both epigastric mass and pain. Some patients may only have epigastric discomfort or fatigue, while others do not have any symptoms. In those asymptomatic patients, pancreatic masses are accidentally found only during routine examinations, and the results of routine biochemical tests are normal<sup>[2]</sup>. SPTP is unresponsive to radiotherapy or chemotherapy, and surgical resection is the primary treatment of choice. In this report, we describe the diagnosis and treatment of SPTP accompanied with pancreatic panniculitis in a young woman.

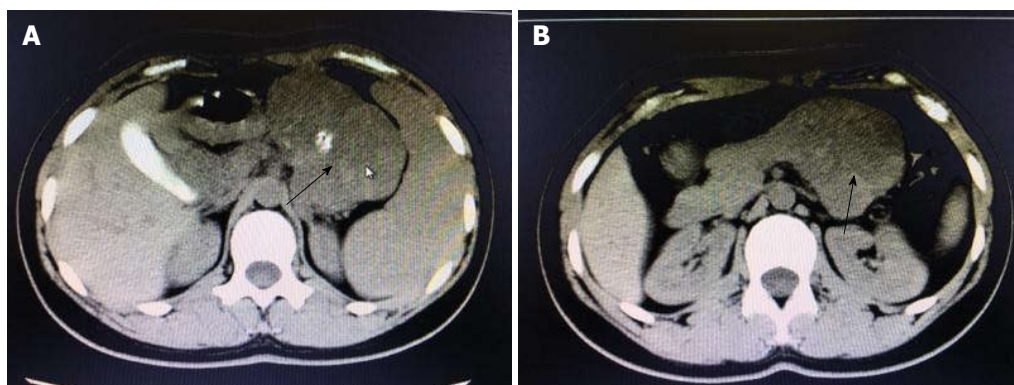


**Figure 1** Results of pathological examination. Subcutaneous nodules, fat necrosis ( $\times 200$ )

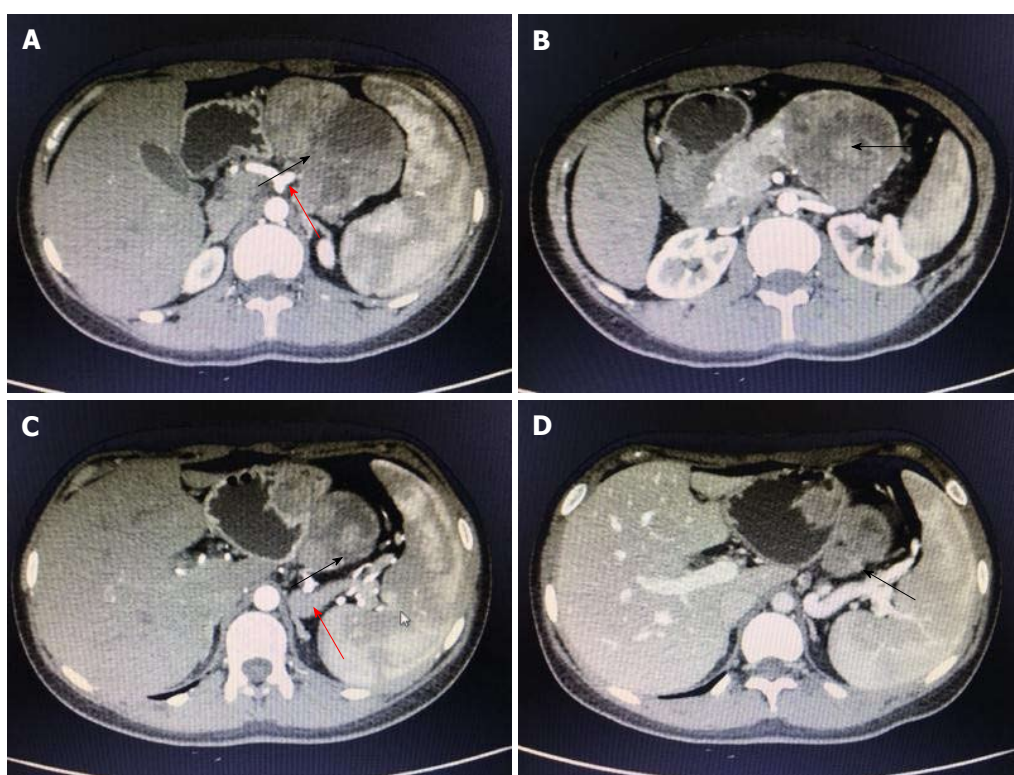
## CASE REPORT

A 19-year-old woman was presented with persistent left upper abdominal pain without obvious cause for 1 d. She also developed subcutaneous nodules involving lower abdomen bilaterally and lower limbs. She had no abdominal distention, vomiting, or radiative and referred pain. Physical examination revealed a palpable mass with slight tenderness in the left upper quadrant of the abdomen. Subcutaneous nodules were pathologically diagnosed as pancreatic panniculitis (Figure 1). The results of routine blood test at admission were as follows: white blood cell  $7.84 \times 10^9/L$  (normal range:  $4-10 \times 10^9/L$ ), neutrophil-to-lymphocyte ratio 85.5% (normal range: 40%-75%), and hemoglobin 88 g/L (normal range: 115-150 g/L). Liver function test showed albumin 30 g/L (normal range: 40-55 g/L) and alanine aminotransferase 62 U/L (normal range: 8-40 U/L). The result of serum amylase was 869 U/L (normal range: 40-100 U/L), serum lipase was 759 U/L (normal range: 0-110 U/L). Plain abdominal computed tomography (CT) revealed a mass in the body and tail of the pancreas, which required further examinations (Figure 2). After admission, antimicrobial treatment, somatostatin, and hepatoprotective therapies were administered. Contrast-enhanced abdominal CT was highly suggestive of a neoplastic lesion in the body and tail of the pancreas. A SPTP was suspected. The lesion was fed by the common hepatic artery and branches of splenic artery, along with regional portal hypertension (Figure 3). Abdominal contrast-enhanced ultrasound showed a solid mass in the body and tail of the pancreas, and a diagnosis of SPTP was considered (Figure 4).

After antimicrobial drug, somatostatin, and liver-protective drug treatments, routine blood test showed white blood cell  $5.62 \times 10^9/L$ , neutrophil-to-lymphocyte ratio 72.8%, and hemoglobin 108 g/L, and liver function test showed albumin 39 g/L and alanine aminotransferase 30 U/L. Serum amylase and lipase, however, declined slowly, showing 479 U/L and 325 U/L. Therefore, traditional Chinese medicine was added to



**Figure 2 Findings of plain abdominal computed tomography scan.** A: Plain abdominal computed tomography reveals a soft-tissue mass in the body and tail of pancreas; B: It has a well-defined border and sized 8.5 cm × 6.1 cm × 6.5 cm, with heterogeneous signal intensity patches and nodular calcifications. It is closely associated with gastric wall. Arrow indicates the mass.



**Figure 3 Findings of contrast-enhanced abdominal computed tomography scan.** A and B: Contrast-enhanced abdominal computed tomography reveals a mixed density mass with solid and cystic components, sized 6.8 cm × 7.8 cm × 8.8 cm, in the body and tail of the pancreas, with a well-defined border. Nodular calcifications are found inside the lesion; C: The solid component is obviously enhanced, although the degree of enhancement is lower than the normal pancreatic parenchyma; D: The spleen is slightly larger, and the splenic vein is compressed. Multiple collateral circulations are seen in the left upper abdomen. The lesion is fed by the common hepatic artery and a branch of splenic artery (red arrows in A and C, black arrows indicate the mass).

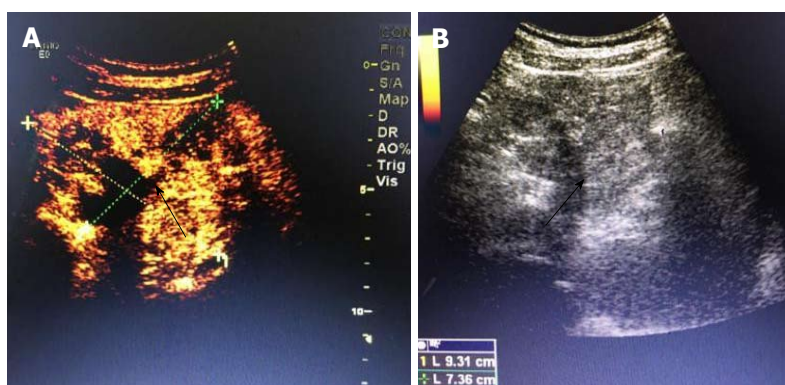
the treatment. Three days later, serum amylase and lipase declined obviously, showing 92 U/L and 98 U/L. After adequate preoperative preparation, the patient underwent laparoscopic resection of the lesion in the pancreatic body and tail with preservation of the spleen. During the operation, a mass sized about 8.5 cm × 8 cm × 7.0 cm was found at the tail of the pancreas. It had an irregular shape and a poorly-defined border with pancreatic body and tail (Figure 5). It was tightly adhered to the splenic vein, and several of its nourished vessels joined the splenic vein. Postoperative pathology

suggested that the mass was an SPTP (Figure 5).

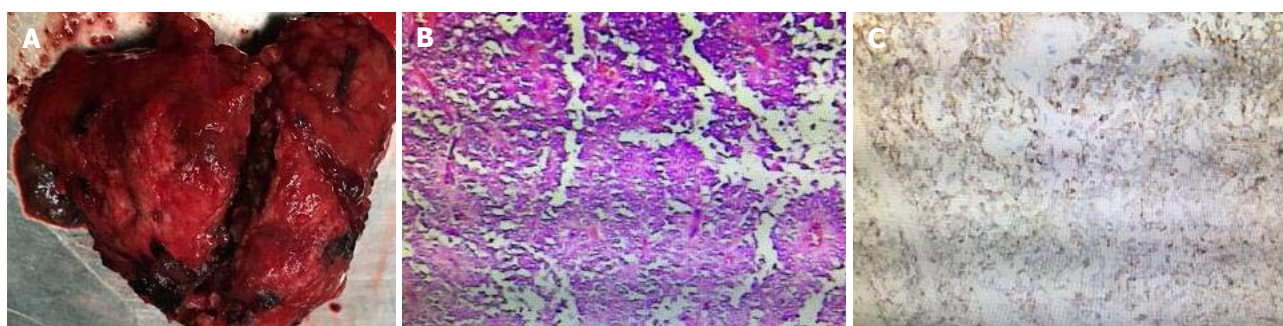
## DISCUSSION

Pancreatic panniculitis is a rare cause of subcutaneous fat necrosis secondary to elevated serum levels of pancreatic enzymes. It is most often associated with pancreatic cell carcinoma, but has also been seen in patients with pancreatitis. Systematic evidence on pancreatic panniculitis is limited<sup>[3]</sup>. SPTP is a rare tumor, accounting for about 3% of all pancreatic tumors. It is





**Figure 4 Findings of abdominal contrast-enhanced ultrasonography.** A: Abdominal contrast-enhanced ultrasonography shows an abnormal morphology of the pancreas. A mass with inhomogeneous echo, sized about 9.3 cm × 7.2 cm, is found in the anterior pancreatic body; it has a well-defined border and regular shape, but the border with the parenchyma of pancreatic body and tail is poorly defined, with flocculent echo and strong spotty or patchy echo in the central part; B: Color Doppler flow imaging showed a few low-speed arteriovenous blood flow signals, whereas the echoes are homogeneous in the remaining parenchyma. The main pancreatic duct is not dilated. Black arrows indicate the mass.



**Figure 5 Specimen of the resected pancreas and results of pathological examination.** A: Specimen of the resected pancreas; B and C: A solid pseudopapillary tumor of the pancreas, sized about 8.2 cm × 8 cm × 5 cm, showed infiltrative growth and necrosis. No vascular or neural invasion was visible. Immunophenotyping showed synaptophysin(+), CD56(+), CD10(+),  $\beta$ -catenin(+), vimentin(+), AE1/AE3(+), PR(+), plasma chromogranin A(-), E-cadherin(-), and Ki-67(+/3%). B: Hematoxylin and eosin staining ( $\times 100$ ); C: Immunohistochemistry ( $\times 100$ ).

a low-grade malignancy, and usually occurs in young women.

The clinical manifestations of SPTP are atypical. In most cases, a mass is often found within the abdomen accidentally or during physical examination, with or without gastrointestinal symptoms, such as the upper abdominal discomfort and dyspepsia<sup>[4-6]</sup>. These non-specific gastrointestinal symptoms are mostly associated with compression of nearby organs by tumors. In some special cases, SPTP can cause acute pancreatitis or spontaneous rupture of the pancreatic pseudocyst, which is clinically manifested as acute abdominal pain. Routine blood test, liver function test, renal function test, and tests for the endocrine/exocrine functions of pancreas often show normal results, and the levels of various tumor markers are usually within the normal ranges.

Imaging examination is crucial for the preoperative diagnosis of SPTP. It can determine the size and location of the tumor and its relationship with surrounding tissues, thus providing detailed information to make a surgical protocol<sup>[2,7,8]</sup>. The typical ultrasonographic finding of SPTP is a cystic/solid mass with mixed density in the pancreas. When the tumor is small, homogen-

eous hypoechoic echoes can be detected. Color doppler flow imaging often reveals a few blood flow signals on the capsule or parenchyma of the tumor. CT is the most commonly used imaging method for SPTP. On CT scans, SPTP is often round and oval, with lobulation. It has a well-defined border with the pancreas, and its margin is smooth. The solid part is often located in the periphery of the tumor, and the cystic part inside the tumor. The tumor can be located at any part of the pancreas. After enhancement, the parenchyma part is slightly enhanced during arterial phase and markedly enhanced during portal venous phase. The cystic part is hypodense before and after enhancement<sup>[9-11]</sup>.

In our case, routine blood test, serum amylase and lipase test, and liver function test showed slight or obvious abnormalities before the operation, which were significantly improved after treatment. Subcutaneous nodules involving lower abdomen bilaterally and lower limbs reduced after treatment. The diagnosis of SPTP and the differential diagnosis of this lesion from other pancreatic diseases were mainly based on abdominal contrast-enhanced CT and abdominal contrast-enhanced ultrasound. A cystic/solid mass with mixed density was found in the body and tail of the pancreas,

with a well-defined border. Nodular calcifications were seen inside the lesion, and the lesion was fed by the common hepatic artery and a branch of splenic artery. The spleen was slightly enlarged, along with regional portal hypertension. Surgery was performed after adequate preparation, during which a mass with areas of necrosis was found in the body and tail of the pancreas. The tumor had a poorly-defined border with pancreatic body and tail. It was tightly adhered to the splenic vein, and several of its nourished vessels joined the splenic vein.

Postoperative pathology confirmed the result of preoperative imaging diagnosis. Immunophenotyping showed synaptophysin (+), CD56(+), CD10(+),  $\beta$ -catenin(+), vimentin (+), and AE1/AE3(+), which met the diagnostic criteria for SPTP, and was consistent with the histopathological diversity of SPTP<sup>[12-14]</sup>. Vimentin,  $\beta$ -catenin, synaptophysin, neuron-specific enolase,  $\alpha$ 1-antitrypsin ( $\alpha$ 1-AT), S-100, neural cell adhesion molecule CD56, and cluster of differentiation 10 (CD10) can all be expressed in SPTP<sup>[15-17]</sup>. At present, surgical resection is the only radical treatment for SPTP. However, laparoscopic surgery is a challenging technique, and the selection of specific surgical approaches depends on the location and size of tumor and the findings of intraoperative pathological examination<sup>[18]</sup>. The most common postoperative complication is pancreatic fistula, followed by pancreatitis, gastrointestinal bleeding, and pseudocyst. The prognosis of SPTP is good, and the postoperative survival rate is high. If the tumor is located in the body and tail of the pancreas, resection of the body and tail of the pancreas is feasible. If the tumor invades or is closely related to the splenic vessels, pancreatectomy combined with resection of the body and tail of the pancreas can be performed<sup>[19-22]</sup>. No evidence is yet available that local resection has a higher risk of recurrence or metastasis than radical resection. Therefore, clinicians should be aware of the clinical manifestation and treatment of pancreatic panniculitis<sup>[23,24]</sup>. Although SPTP has good prognosis and the postoperative 5-year survival rate is high, efforts should be made to increase further diagnostic accuracy and optimize therapeutic methods, so as to improve the quality of life of SPTP patients.

## ARTICLE HIGHLIGHTS

### Case characteristics

A 19-year-old woman presented with persistent left upper abdominal pain without obvious cause for 1 d. The patient also developed subcutaneous nodules involving lower abdomen bilaterally and lower limbs. An irregular mass was found in the pancreatic body and tail on plain abdominal computed tomography (CT), contrast-enhanced abdominal CT, and contrast-enhanced ultrasound.

### Clinical diagnosis

Solid pseudopapillary tumor of the pancreas (SPTP), pancreatic panniculitis.

### Differential diagnosis

Pancreatic cancer.

### Laboratory diagnosis

Abnormal laboratory findings included the results of routine blood: Neutrophil-to-lymphocyte ratio 85.5% (normal range: 40%-75%), hemoglobin 88 g/L (normal range: 115-150 g/L). Liver function test showed albumin 30 g/L (normal range: 40-55 g/L) and alanine aminotransferase 62 U/L (normal range: 8-40 U/L). The result of serum amylase was 869 U/L (normal range: 40-100 U/L), and serum lipase was 759 U/L (normal range: 0-110 U/L).

### Imaging diagnosis

Contrast-enhanced abdominal CT revealed a cystic/solid mass with mixed density in the body and tail of the pancreas.

### Pathological diagnosis

SPTP, pancreatic panniculitis.

### Treatment

Laparoscopic resection of the mass in the pancreatic body and tail with preservation of the spleen.

### Related reports

Some articles have described the imaging diagnosis and treatment of SPTP and pancreatic panniculitis, as shown in the References.

### Experiences and lessons

Clinicians should be aware of the clinical manifestation and treatment of pancreatic panniculitis. Although SPTP has good prognosis and the postoperative 5-year survival rate is high, efforts should be made to increase further diagnostic accuracy and optimize therapeutic methods, so as to improve the quality of life of SPTP patients.

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