Giant Solitary Fibrous Tumor of the Pancreas: A Case Report and Review of the Literature

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Abstract

Background: Solitary fibrous tumors (SFTs) originating from the pancreas are rare, only 23 cases have been reported so far. Here, we report the largest one, which has its unique treatment characteristics.

Case report: A 42-year-old woman was admitted to the hospital with abdominal pain and abdominal mass, and the preoperative diagnosis was unclear. As the mass was rich in blood vessels, she underwent partial pancreatectomy after embolizing part of the blood vessels by preoperative intervention, which, at final pathology, showed spindle and oval cell proliferation and featured the presence of hemangiopericytoma-like blood. Immunohistochemistry showed cytoplasmic expression of CD34 and nuclear expression of STAT6, and mitotic activity was lower than 4 mitoses/10 high-power fields (HPFs). At the end, she was diagnosed with pancreatic SFT. The postoperative recovery was uneventful and she was alive and free of disease after 8 months.

Conclusion: The diagnosis of SFT of the pancreas needs to be done very cautiously. Surgical treatment and postoperative follow-up are more reasonable methods at the moment.

Introduction

SFTs were rare spindle-cell neoplasms of pluripotent fibroblastic or myofibroblastic origin.[1] Previous reports believed that this kind of tumor mostly occurred in the thoracic cavity, while SFT that originated outside the thoracic cavity was rare.[2] SFT originating in the pancreas was even rarer, and only 20 cases have been reported so far.[3, 4] Due to the low incidence, there was still no consensus on the treatment of this disease. This article reported a case of a patient with giant SFT originating from the pancreatic tail. She finally underwent pancreatic tail resection with good results.

Case Report

A 42-year-old woman was admitted to the hospital with intermittent left upper abdomen pain for 1 month. Physical examination showed a mass in the left abdomen, which was hard on palpation, and the boundary with surrounding tissues was not clear, and it was not easy to push away, with no other abnormalities. After admission, we performed some examinations. Blood biochemical and tumor markers, such as AFP, CEA, CA19-9, CA125, and CA153 were all normal. MRI showed that the left upper abdomen was a huge mass with partial necrosis, which was lobulated, and the surrounding tissues were obviously compressed. MRI also showed splenomegaly, portal hypertension, splenic varices, and liver cysts. These phenomena consider the possibility of retroperitoneal fibroids. In the arterial phase, we could see that the mass was hypervascular. The enhanced CT examination revealed that a huge mass in the liver-spleen space with partial necrosis in the left upper abdomen, and the surrounding tissues were severely compressed. CT results were considered to be exogenous liver cancer, a mass in the retroperitoneal adrenal area on the left, considering metastasis. Splenomegaly, portal hypertension, and splenic varices. Because the boundary between the mass and the surrounding organs was unclear, and

the results of MRI and CT examinations were different, to clarify the nature of the mass and determine the benefit of the operation to the patient, liver biopsy was performed before the operation.

Immunohistochemistry showed that the molecular markers of tumor cells were: CD117 (-), CD34(+), Desmin (-), DOG1(-), Ki-67(Li:5%), S-100(-), SDHB(+)), SMA(-), Bcl-2(+) and STAT6(+). Therefore the mass was considered to be a solitary fibrous tumor. But considering the contradiction between biopsy, CT, and MRI, the diagnosis of solitary fibroma was cautious and we were also not sure whether the tumor was malignant.

Because the mass was so huge and abundant in blood supply, a multidisciplinary consultation was conducted. Finally, interventional experts recommend DSA examination before surgery. DSA indicated that the splenic artery, left hepatic artery and gastroduodenal artery were involved in blood supply. During the DSA examination, part of the feeding arteries of the tumor was successfully embolized to prevent uncontrollable bleeding. After proper preparation, the patient underwent abdominal lesion resection and splenectomy, but before the operation, we still informed the family that the operation will be canceled if the mass is malignant or unresectable. Because the tumor was huge, a larger surgical incision was chosen to better expose the tumor and relieve the pressure of the abdominal cavity. The good news was that after extending the surgical incision, the tumor was separated from the liver and stomach clearly, therefore the operation could continue. During the separation, a tight adhesion between the mass and pancreatic tail was found, so we believed that the mass was derived from the pancreas. Except for the expected pancreatic fistula, the postoperative recovery was uneventful. During the 8-month follow-up, the patient did not recurrence.

Macroscopic examination showed that the tumor was a piece of 24×16×12cm grayish yellow-brown irregular tissue with a complete envelope, the cut surface was grayish-white and grayish-brown, solid to soft, and focally grayish-yellow. Microscopic examination showed necrosis, and the tumor cells were moderately atypia, mitosis was less than 4/10 per high-power fields. Proliferative index by Ki-67 was 5% and immunohistochemically, the tumor cells were positive for CD34 and STAT6. There was no metastasis in the resected greater curvature lymph nodes. The final diagnosis was consistent with SFT of the pancreas.

Discussions And Conclusions

SFT was reported by Klemperer et al. in 1931 as a tumor of the pleura.[5] According to the WHO classification, SFT is a tumor of mesenchymal tissue origin. It accounts for about 2% of soft tissue tumors.[6] However, with the development of pathology, SFT was found to be a soft tissue tumor that occurred in fibroblasts or myofibroblasts and could appear all over the body,[7, 8] and it was also pathologically diverse.[9] A solitary fibrous tumor of the pancreas was a non-epithelial tumor of the pancreas.[10] SFTs originating from the pancreas were rare, we searched several major databases and found a total of 23 English articles documenting the cases. A summary of these cases was shown in Tables 1 and Table 2. According to the collected literature, pancreatic SFT was more common in women. Sheng et al. reported a SFT originating in toddler pancreas,[11] but in general, middle-aged and elderly people are more likely to suffer from it. The most common tumor site was the pancreatic head, with 14

cases; 8 cases were reported in the pancreatic body and 3 cases in the pancreatic tail. The most common treatment for pancreatic SFT was surgical resection, as, the location and symptoms of the tumor and the modality of surgery are closely related. Most tumors were benign and did not metastasize at the end of follow-up.

Table 1
Characteristics of pancreatic solitary fibrous tumors

Author	Age/sex	Chief compliant	Size(cm)	location	treatment
Qian et al.[13]	48/M	Hypoglycemia	6.5	Body	Distal pancreatectomy and hepatic tumor resection
Geng et al.[11]	48/ M	Hypoglycemia	6.5	Body	TACE, DP, left lateral liver sectionectomy
Taguchi et al. [31]	60/M	Abnominal mass	8	Head	PD
D'Amico et al. [23]	52/M	Incidental	2	Body	Enucleation
Oana et al.[32]	73/M	Abdominal discomfort	7.5	Head	Partial pancreatectomy
Sheng et al. [10]	1/m	Jaundice	2	Head	PD
Paramythiotis et al.[33]	55/m	Abdominal pain	3.6	Body	DP
Murakami et al.[34]	82, M	Hypokalemia hypertension	6	Tail	DP
Spasevska et al.[35]	47, M	Epigastric pain jaundice	3.5	Head	PD
Han et al.[36]	77, F	Jaundice	1.5	Head	Conservative
Estrella et al. [12]	52, F	Jaundice	15	Head	PD
Chen et al.[37]	49, F	Abdominal pain	13	Head	PD
Azadi et al.[38]	57, M	Incidenta	3.1	Tail	DP
Van der Vorst et al.[39]	67, F	Abdominal pain	2.8	head	Enucleation
Tasdemir et al. [40]	24, F	Epigastric pain	18.5	Head	Enucleation
Sugawara et al.[41]	55, F	Incidental	7	Head	PD
Chetty et al. [42]	67, F	Incidental	2.6	Head	PD
Kwon et al.[43]	54, M	Incidental	4.5	Body	Median segmentectomy

M: man; F: femal PD: pancreaticoduodenectomy DP Distal pancreatectomy

TACE: transarterial chemoembolization;

Author	Age/sex	Chief complaint	Size(cm)	location	treatment
Srinivasan et al.[44]	58, F	Incidental	3	Head	PD
Miyamoto et al.[45]	41, F	Abdominal pain	2	Head–body	Enucleation
Gardini et al. [46]	62, F	Abdominal pain	3	Head	PD
Chatti et al.[47]	41/M	Abdominal pain	13	Body	Enucleation
Lüttges et al. [48]	50, F	Incidental	5.5.	Body	DP
Present case	42/F	Abdominal pain	24	Tail	Distal pancreatectomy
M: man; F: female PD: pancreaticoduodenectomy DP Distal pancreatectomy					
TACE: transarterial chemoembolization;					

Table 2
Pathological features and outcomes of pancreatic solitary fibrous tumors

Author	Histology	Immuno- histochemistry	nature	Follow- up	recurrence
Qian et al.[13]	4–5 mitoses/10 HPFs	STAT6, CD34, Bcl-2, Ki67 10%	Malignant	10	Y
Geng et al. [11]	Necrosis, 4–5 mitoses/10 HPFs	STAT6, CD34, Bcl-2, CD31, PHH-3, D2-40, Ki67 > 10%	Malignant	6	N
Taguchi et al. [31]	Hypercellularity 12/10 HPFs necrosis invasive growth	STAT6, CD34, Bcl-2, vimentin, cytokeratin AE1/AE3(focal)	Malignant	12	N
D'Amico et al. [23]	No necrosis or mitoses	STAT6, CD34	Benign	24	N
Oana et al. [32]	No necrosis or mitoses	CD34, Bcl-2	Benign	36	N
Sheng et al. [10]	2–5 mitoses/10 HPFs	CD34, SMA, vimentin Ki67 < 3%	Benign	12	N
Paramythiotis et al.[33]	No necrosis or mitoses	CD34, CD99, Bcl-2, vimentin, S100 (focal)	Benign	40	N
Murakami et al.[34]	Spindle neoplastic cells in fascicular arrangement	CD34, Bcl-2, STAT6, ACTH (focal), POMC (focal), NSE (focal)	Benign	4	N
Spasevska et al.[35]	No necrosis or mitoses	CD34, vimentin, CD99, Bcl-2 (focal), nuclear beta-catenin (focal)	Benign	NA	NA
Han et al.[36]	No necrosis or mitoses	CD34, CD99	Benign	10	No progression
Estrella et al. [12]	17 mitoses/10 HPFs, necrosis	CD34, Bcl-2, keratin (rare), p16, p53	Malignant	40	N

Y: Yes; N:No; NA: Not applicable; STAT6: signal transducer and activator of transcription 6;SMA: smooth muscle actin;CD: cluster of differentiation;

Author	Histology	Immuno- histochemistry	nature	Follow- up	recurrence
Chen et al. [37]	Necrosis,	CD34, Bcl-2, vimentin, CD68, muscle- specific actin	Benign	30	N
Azadi et al. [38]	No necrosis or mitoses	CD34, Bcl-2, Ki67 < 5%	Benign	NA	NA
Van der Vorst et al.[39]	No necrosis or mitoses	CD34, CD99, Bcl-2	Benign	NA	NA
Tasdemir et al.[40]	1–2 mitoses/10 HPFs	CD34, Bcl-2, betacatenin, vimentin, Ki67 < 2%	Benign	NA	NA
Sugawara et al.[41]	No necrosis or mitoses	CD34	Benign	NA	NA
Chetty et al. [42]	No necrosis or mitoses	CD34, CD99, Bcl-2	Benign	6	N
Kwon et al. [43]	No necrosis or mitoses	CD34, CD99, vimentin	Benign	NA	NA
Srinivasan et al.[44]	mitosis < 1 /10 HPFs, no necrosis	CD34, Bcl-2	Benign	7	N
Miyamoto et al.[45]	No necrosis or mitoses	CD34, Bcl-2	Benign	16	N
Gardini et al. [46]	NA	CD34, CD99, Bcl-2, vimentin, smooth muscle actin (focal)	Benign	16	N
Chatti et al. [47]	No necrosis or mitoses	CD34, CD99, Bcl-2, vimentin	Benign	NA	NA
Lüttges et al. [48]	No necrosis or mitoses	CD34, CD99, Bcl-2, vimentin	Benign	20	N
Present case	Necrosis, mitosis < 4 /10 HPFs	CD34, STAT6, Ki67 5%	Benign	8	N
Y: Yes; N:No; NA: Not applicable; STAT6: signal transducer and activator of transcription 6;SMA: smooth muscle actin□CD: cluster of differentiation;					

The most common symptoms of pancreatic SFT were abdominal pain, abdominal distension, weight loss, and jaundice.[11, 12] Estrella et al. reported a patient with pancreatic head SFT combined with jaundice.[13] There were also reports of other complications such as hypoglycemia in metastatic pancreatic SFT.[14] However, the largest pancreatic SFT we reported did not show these systemic symptoms, which may be a feature of pancreatic SFT, especially benign pancreatic SFT. As for imaging, Shin et al. believed that in the arterial phase of enhanced CT, the rich blood vessels next to the mass were the specific manifestations of SFT.[15] In addition, it was reported that a malignant and larger tumor may present with hemorrhage, calcifications, and cystic areas.[16] But according to the current research, there was no consensus on imaging for the diagnosis of pancreatic SFT. Our patient's tumor was too large, which severely squeezed the surrounding organs, resulting in differences in CT and MRI reports. It can be considered that the imaging features of our case were different from previous reports, so it was unique. The diameter of the mass reported in the literature was 1.5 to 18.5cm,[11] while in our report it was 24cm, which was the largest case so far. Shanbhogue et al. thought extrapleural SFTs typically manifest as slow-growing soft-tissue neoplasms.[17] But it was worth noting that our patient suffered a trauma 8 months ago, the CT scan did not reveal an abdominal mass at that time, which suggested that the growth of SFT can be very rapid, although it was benign. As to the diagnosis, It mostly relied on microscopic phenomena and several molecular markers(CD34, CD99, and Bcl-2) in the past.[18] In addition, Schweizer et al., Yoshida et al., and Doyle et al. also reported high sensitivity and specificity of STAT6 nuclear expression for SFT.[19–21] But since SFT was characterized by the presence of a NAB2-STAT6 gene fusion, the diagnosis of SFT was more accurate.[22, 23]

It was believed that the treatment of pancreatic SFT should be radical surgical resection. D'Amico et al. summarized the literature and found that the resection of pancreatic SFT was followed by tumor enucleation, resection of the pancreatic body and tail, Whipple surgery, etc.[24] Our patient finally excised part of the pancreatic tail where the tumor originated from. In our case, the tumor was too large, and enhanced CT showed that the surrounding tumor was rich in blood vessels. In addition, the splenic vein, left hepatic artery and gastroduodenal artery were involved in blood supply, so we embolized part of the blood vessels before the operation, which was not reported before. Abdominal surgical incisions were mostly midline or paramedian approach. During the operation, the initial small incision did not fully relieve the pressure in the abdominal cavity, which made us mistakenly believe that the tumor was unresectable, especially as enhanced CT suggested that it may be liver cancer. After extending the incision, it was discovered that this was not the case. This suggested that in some cases when the tumor was large and of unknown nature, a small incision may not be best. After the operation, the patient's breathing and digestive functions recovered slowly. We believed that this was due to the sudden release of the pressure of the abdominal cavity, which affected the patient's original morbid balance.

The 2013 WHO classification of soft tissue tumors defines malignant forms as hypercellular, mitotically active (> 4 mitosis/10 high-power fields), with cytological atypia, tumor necrosis, and/or infiltrative margins.[25] Gold et al. found when the tumor was larger than 10 cm, it was more likely to metastasize or recur.[6] This meant that the larger the tumor, the more likely it was to be malignant. But the case we reported is currently the largest, the postoperative pathology was benign, and during the follow-up, the

patient did not recur after 8 months. However, the nature of pancreatic SFT was not decisive for the treatment of the disease. Researchers believed that although most SFTs are benign, their behavior can be unpredictable, and assessment of the recur and metastatic risk remains a debatable topic.[10] Some clinical studies had found that even though SFT was considered benign at first, it could get metastasized later.[26–28] In addition, studies had shown that most SFTs after complete resection showed benign behavior, though with local recurrence and distant metastasis, and the 10-year survival rate after R0 resection was as high as 54%-89%.[29, 30] Surgical resection should be the most accepted treatment for SFT at present. Stacchiotti et al. thought chemotherapy was beneficial to the treatment of SFT.[31] But we should also understand that due to the rarity of cases, these conclusions may not be representative. This suggests that our understanding of SFT needs to be further strengthened, especially for pancreatic SFTs.

Abbreviations

AFP

Alpha-fetoprotein; Bcl-2:B cell CLL/lymphoma-2; CA125:Carbohydrate antigen 125;

CA153

Carbohydrate antigen 153; CA19-9:Carbohydrate antigen 19 – 9; CD:Cluster of differentiation;

CEA:Carcinoembryonic antigen; CT:Computed tomography; DSA:Digital Subtraction Angiography;

HPFs:High-power magnification fields; MRI:Magnetic resonance imaging; SFTs:Solitary fibrous tumors;

SMA:Smooth muscle actin; STAT6:Signal transducer and activator of transcription

Declarations

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Availability of the data and materials

All the data are available in the patient's medical record.

Authors' contributions

Junqiang Peng and Yanmin Liu collected and analyzed the patient data in the context of existing literature. Qian Peng and, Junqiang Peng, and Tingting Dou performed the data collection. Xiaochang Fang, Tiecheng Yang, and Huaqiao Wang performed the histological analysis. Junqiang Peng and Lijie Yang were the major contributors in writing the manuscript. The authors read and approved the final manuscript.

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Ethics approval and consent to participate

This is a case report. The Institutional Review Board at the Zhongnan Hospital of Wuhan University has confirmed that no ethical approval is required. Consent was obtained from the patient for participation in this study.

Consent for publication

We obtained the patient's consent for publication of this case report.

Competing interests

The authors have no conflicts of interest relevant to this article.

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Figures

Figure 1

(A) Contrast-enhanced MR scan showed a large heterogeneous mass in the abdominal cavity (arrow). (B) Contrast-enhanced MR scan showed a large heterogeneous mass in the abdominal cavity (arrow). (C) CTA showed that the tumor was rich in blood supply, and the branches of the splenic artery, gastroduodenal artery, and left hepatic artery were all involved in blood supply. (D) The preoperative intervention clearly showed the blood supply vessels of the mass. (E) Intervention surgery embolized part of the blood vessel. T= tumor, SA = splenic artery, GDA = Gastroduodenal artery, LHA= Left hepatic artery, STO= stomach.

Figure 2

(A) No liver lesions were observed, and the liver activity was good. (B) The tumor was smooth and did not adhere to the liver and retroperitoneum. (C and D) A macroscopic examination revealed a 24×16×12cm tumor that was oval, lobulated, well-circumscribed, and well-capsulated. The cut surface was yellowish-gray, and fish-like appearance.

Figure 3

Immunohistochemical results of SFT of the pancreas. (A): A microscopic examination showed spindle cell neoplastic proliferation, HE stain, magnification: 200, tumor cells were moderately atypia, the nuclear division was about 4/10 HPF; (B, C, and D): Immunohistochemical results were: ki-67(Li: 5%), CD34+, STAT6+. Magnification: 200. SFT=solitary fibrous tumor, HE = hematoxylin and eosin, HPF = high-power field.