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CASE REPORT

Imaging, pathology, and diagnosis of solitary fibrous tumor of the pancreas: A case report and review of literature

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Abstract

BACKGROUND

A solitary fibrous tumor (SFT) is often located in the pleura, while SFT of the pancreas is extremely rare. Here, we report a case of SFT of the pancreas and discuss imaging, histopathology, and immunohistochemistry for accurate diagnosis and treatment.

CASE SUMMARY

A 54-year-old man presented to our hospital with pancreatic occupancy for over a month. There were no previous complaints of discomfort. His blood pressure was normal. Blood glucose, tumor markers, and enhanced computed tomography (CT) suggested a malignant tumor. Because the CT appearance of pancreatic cancer varies, we could not confirm the diagnosis; therefore, we performed endoscopic ultrasound-guided fine-needle biopsy (EUS-FNB). Pathology and immunohistochemistry were consistent with SFT of the pancreas. The posto-perative pathology and immunohistochemistry were consistent with the puncture

results. The patient presented for a follow-up examination one month after discharge with no adverse effects.

CONCLUSION
Other diseases must be excluded in patients with a pancreatic mass that cannot be diagnosed. CT and pathological histology have diagnostic value for pancreatic tumors. Endoscopic puncture biopsy under ultrasound can help diagnose pancreatic masses that cannot be diagnosed preoperatively. Surgery is an effective treatment for SFT of the pancreas; however, long-term follow-up is strongly recommended because of the possibility of malignant transformation of the tumor.

Key Words: Pancreas; Neoplasm fibrous tumor; Endoscopic ultrasound-guided fine-needle biopsy; Treatment; Case report

Core Tip: We need to be more vigilant for indeterminate pancreatic masses, and then computed tomography and histopathology can play a very important role in clinical diagnosis. Surgery is an effective treatment for solitary fibrous tumor of the pancreas; however, long-term follow-up is strongly recommended because of the possibility of malignant transformation of the tumor.

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INTRODUCTION

A solitary fibrous tumor (SFT) is histologically characterized as a mesenchymal tumor, probably fibroblastic in origin, located primarily in the pleura; however, it can be found in any other extrapleural region[1-3]. Extrapleural areas include the liver, peritoneum, kidney, and salivary glands[4-7]. SFT of the pancreas is rare, with only about 30 cases reported to date[1-3,6-35]. SFT of the pancreas is usually asymptomatic, and most are detected by physical examination, computed tomography (CT), or ultrasound as pancreatic masses[6,30,32]. The final diagnosis depends on histopathology and immunohistochemistry[7,31].

Here, we report a case of SFT of the pancreas and present the radiological and pathological differential diagnosis.

CASE PRESENTATION

Chief complaints

A 54-year-old man was admitted to our hospital with a pancreatic space-occupying mass of one month's duration, identified on a physical exam.

History of present illness

A 54-year-old man had been one month before a medical CT finding of pancreas space-occupying lesions, with no adverse reactions, patients for further treatment at our hospital.

History of past illness

The patient had no other significant medical history. History of hypertension, diabetes, coronary heart disease, and other chronic disease was denied.

Personal and family history

The patient had no significant personal or family history.

Physical examination

The patient had no discomfort after the physical examination.

Laboratory examinations

There was no abnormal carcinoembryonic antigen [< 0.5 ng/mL (normal 0-5 ng/mL)], carbohydrate antigen 199 3.9 U/mL (average 0-7 U/mL), alpha-fetoprotein 2.4 ng/mL (normal 0-8.8 ng/mL), carbohydrate antigen 125 12.5 (average 0-30.2 U/mL). Fasting glucose was 5. 19 mmol/L (normal 3.89-6. 11 mmol/L).

Imaging examinations

A review of an abdominal enhanced CT showed a tumor of about 3 cm × 2 cm in the tail of the pancreatic body, showing uneven enhancement after enhancement, consistent with a malignant tumor (Figure 1).

FINAL DIAGNOSIS

A SFT of the pancreas.

TREATMENT

CT revealed a mass with mixed density and inadequate blood supply; these findings were inconsistent with a pancreatic tumor; therefore, we considered a pseudopapillary tumor and a non-functional pancreatic neuroendocrine tumor. We performed an ultrasound endoscopic tissue biopsy. The pathology and immunohistochemistry suggested SFT of the pancreas. After excluding contraindications to surgery and obtaining informed written consent, we performed a laparoscopic distal pancreatectomy with splenectomy. No significant adhesions were seen in the peripancreatic tissue. The pancreatic body was approximately 3 cm × 2 cm (Figure 2). Intraoperative frozen sections showed negative margins. Intraoperative blood loss was 100 mL and no blood transfusion was required.

The patient had no postoperative pancreatic fistula, abdominal infection, or bleeding. Ten days after surgery, he was discharged from the hospital after removing the drainage tube. One month after surgery, the patient returned to the hospital for examination. He did not complain of discomfort. The complete blood count, liver enzymes and renal function were normal.

Histopathological and immunohistochemical results of the postoperative specimen suggested an SFT of the pancreas of 3.0 cm × 2.5 cm × 1.0 cm, negative margins, no tumor involvement in the surrounding lymph nodes, and no tumor involvement in the spleen. Markers were as follows: Signal transducer and activator of transcription 6 (STAT6) (+), cluster of differentiation (CD) 34 (+), B cell CLL/lymphoma-2 (Bc1-2) (+), vimentin (+), CD99 (+), CD117 (-), Ki-67 (+40%), discovered on GIST-1 (+), transducin-like enhancer protein 1 (+), S- 100 (-), cytokeratin pan (pan) (-), somatostatin receptor 2 (-) (Figure 3).

OUTCOME AND FOLLOW-UP

No specific treatment was given after the patient was discharged from the hospital, and he had no complaints for three months after the procedure. He returned for regular follow-up. No abnormalities were found on complete blood counts, blood glucose, tumor markers, or CT.

DISCUSSION

SFT is a mesenchymal tumor comprising less than 2% of soft tissue tumors[36]. About 65% of SFTs originate from the pleura[3]; however, they can also be found in extrapleural areas[6], with only 34 cases reported to date, including the present case (Tables 1 and 2). SFT of the pancreas is extremely rare. We searched PubMed and Google Scholar for pancreatic tumors and SFT and found 34 cases. Of these, 14 (41. 1%) were male, and 20 (58.9%) were female. The mean age was 54. 17 ± 15.4, and the median age was 54; 17 patients had lesions in the pancreatic tumor head [three (17.6%) male and 13 (76.4%) female]. Seventeen had tumors in the tail of the pancreatic body [ten (58.8%) male and seven (41.2%) female]. The mean tumor diameter was 5.2 cm ± 3.8 cm. Of the 34 patients, 12 presented with pain (12/34), 12 were discovered on physical examination (12/34), four presented with jaundice (4/34), one presented with an abdominal mass (1/34), and five were detected by other means (5/34) (Table 1).

Most SFTs of the pancreas are detected by physical examination; clinical signs and symptoms include abdominal pain and jaundice. Because these are not typical symptoms, it is challenging to differentiate SFT from other pancreatic diseases. Histopathology and immunohistochemistry are the gold standards for diagnosis. We recommend ultrasound endoscopic aspiration biopsy for space-occupying pancreatic lesions that cannot be diagnosed on imaging.

Our preoperative diagnosis relied on ultrasound endoscopic puncture biopsy in the present case. The preoperative and postoperative pathological histological examination and immunohistochemistry were consistent with SFT of the pancreas with no tumor involvement in the peripheral lymph nodes, no tumor involvement in the incised margin of the pancreas, and no tumor involvement in the spleen.

The immunohistochemical differential diagnosis of SFT of the pancreas should include spindle cell tumors such as gastrointestinal stromal tumor (GIST), smooth muscle sarcoma, nerve sheath tumor, fibrous mucinous sarcoma, perivascular epithelioid cell tumor, and vascular tumors[3,16,20,37]. The immunomarkers of SFT of the pancreas include STAT6, CD34, bc1-2, vimentin, and CD99[34]. These features help to distinguish SFT from other mesenchymal tumors[34, 37]. SFT expresses CD34 and vimentin in 80%-90% of cases and CD99 and bcl-2 in 70%. SFTs are usually negative for c-kit (CD117), smooth muscle actin, junctional protein, S-100 protein, and cytokeratin (markers for GIST, smooth muscle

Table 1 Characteristics of pancreatic solitary fibrous tumors

No	Ref.	Age	Sex	Pancreatic site	Symptoms	Size (cm)	Pancreatic surgery
1	Lüttges *et al*[1]	50	F	Body	Incidental	55	DP
2	Chatti *et al*[8]	41	M	Body	Abdominal pain	13	DP
3	Gardini *et al*[9]	62	F	Head	Abdominal pain	3	PD
4	Miyamoto *et al*[10]	41	F	Head	Abdominal pain	18 × 15	Enucleation
5	Kwon *et al*[11]	54	M	Body	Incidental	76 × 6	MS
6	Srinivasan *et al*[12]	78	F	Body	Back pain, weight loss	5	DP
7	Chetty *et al*[13]	67	F	Head	Incidental	26	PD
8	Ishiwatari *et al*[14]	58	F	Head	Incidental	3	PD
9	Sugawara *et al*[15]	55	F	Head	Incidental	6 × 4	PD
10	Santos *et al*[16]	40	M	Body	Incidental	3	Enucleation
11	Tasdemir *et al*[17]	24	F	Body	Epigastric pain	11	Enucleation
12	van der *et al*[18]	67	F	Head	Abdominal pain	28 × 16	Enucleation
13	Chen *et al*[19]	49	F	Head	Abdominal pain	13	PD
14	Hwang *et al*[20]	53	F	Head	Incidental	52 × 45 × 40	PHR
15	Baxter *et al*[21]	58	F	Head	Abdominal pain	35 × 3	LPD
16	Estrella *et al*[22]	52	F	Head	Jaundice	15 × 10 × 10	LPD
17	Han *et al*[23]	77	F	Head	Jaundice	15 × 14	Biopsy
18	Murakami *et al*[24]	82	M	Body	Hypokalemia hypertension, edema	6	DP
19	Spasevska *et al*[3]	47	M	Head	jaundice	35 × 2 × 18	LPD
20	Paramythiotis *et al*[7]	55	M	Body	Abdominal pain	31 × 28	DP
21	D'Amico FE *et al*[25]	52	M	Body	Incidental	12	DP
22	Oana *et al*[26]	73	M	Head	Abdominal discomfort	65 × 55	Enucleation
23	Sheng *et al*[27]	1	M	Head	Jaundice	20	DP
24	Geng *et al*[28]	48	M	Body	Hypoglycemia	65 × 5	DP
25	Qian *et al*[29]	46	M	Body	Hypoglycemia	70 × 61	DP
26	Rogers *et al*[30]	37	F	Head	Abdominal pain	23	PD
27	Taguchi *et al*[31]	60	M	Head	Palpable mass	9 × 7 × 7	PD
28	Jariwalla *et al*[32]	64	F	Body	Abdominal pain	19	DP
29	Marotti *et al*[33]	75	F	Body	Incidental	13	Enucleation
30	Addeo *et al*[6]	59	M	Body	Incidental	4	DP
31	Rodriguez *et al*[2]	48	F	Body	Abdominal pain	13 × 10 × 95	TP
32	Jones *et al*[34]	61	F	Body	NA	27	DP
33	Liu *et al*[35]	54	F	Head	Incidental	31 × 23	LDPPHRt
34	Present case	54	M	Body	Incidental	3 × 2	DP

LDPPHRt: Laparoscopic duodenum-preserving pancreatic head resection; Ms: median segmentectomy; PHR: Pancreatic head resection; TP: Total pancreatectomy; PD: Pancreaticoduodenectomy; DP: Distal pancreatectomy.

sarcoma, nerve sheath tumor, and fibrous mucinous sarcoma, respectively) are negative[3]. NAB2-STAT6 fusion is a driver mutation in SFT, where transcriptional repressors of the cytokinesis pathway are converted into transcriptional activators[31,38,39]. STAT6 has a sensitivity of 98% and a specificity of 85% for SFT and is therefore considered the most characteristic SFT marker[40,41]. In our case, the tumor was positive for STAT6, while CD34, bc1-2, vimentin, and CD99 were positive.

Table 2 Histological features and outcomes of pancreatic solitary fibrous tumors

No	Ref.	Immunohistochemistry	Outcome	Follow-up
1	Lüttges *et al*[1]	CD34, CD99, Bcl-2, vimentin	Alive	20 months
2	Chatti *et al*[8]	CD34, CD99, Bcl-2, vimentin	Death	3 d
3	Gardini *et al*[9]	CD34, CD99, Bcl-2, vimentin, SMA	Alive	16 months
4	Miyamoto *et al*[10]	CD34, Bcl-2	Alive	7 months
5	Kwon *et al*[11]	CD34, CD99, vimentin	NA	NA
6	Srinivasan *et al*[12]	CD34, Bcl-2	Alive	7 months
7	Chetty *et al*[13]	CD34, CD99, Bcl-2	42 mo	6 v
8	Ishiwatari *et al*[14]	CD34, Bcl-2	Alive	42 months
9	Sugawara *et al*[15]	CD34	NA	NA
10	Santos *et al*[16]	CD34, betacatenin	NA	NA
11	Tasdemir *et al*[17]	CD34, Bcl-2, beta-catenin, vimentin, Ki67 < 2%	Alive	3 months
12	van der *et al*[18]	CD34, CD99, Bcl-2	NA	NA
13	Chen *et al*[19]	CD34, Bcl-2, vimentin, CD68, muscle-specific actin	Alive	30 months
14	Hwang *et al*[20]	CD34, Bcl-2, muscle-specific actin, CD10, ER, PR	Alive	30 months
15	Baxter *et al*[21]	CD34, Bcl-2	NA	NA
16	Estrella *et al*[22]	CD34, Bcl-2, keratin (rare), p16, p53	Alive	40 months
17	Han *et al*[23]	CD34, CD99	No progression	10 months
18	Murakami *et al*[24]	STAT6, CD34, Bcl-2, ACTH, POMC, NSE	Death	4 months
19	Spasevska *et al*[3]	CD34, vimentin, CD99, Bcl-2, nuclear betacatenin	Death	1 wk
20	Paramythiotis *et al*[7]	CD34, CD99, Bcl-2 vimentin, S-100	Alive	40 months
21	D'Amico FE *et al*[25]	STAT6, CD34	Alive	24 months
22	Oana *et al*[26]	CD34, Bcl-2	Alive	36 months
23	Sheng *et al*[27]	CD34, vimentin, SMA, Ki67 < 3%	Alive	12 months
24	Geng *et al*[28]	STAT6, CD34, Bcl-2, CD31, PHH-3, D2-40, Ki67 > 10%	Alive	6 months
25	Qian *et al*[29]	STAT6, CD34, Bcl-2, Ki67 10%	Alive	10 months
26	Rogers *et al*[30]	STAT6, CD34, Bcl-2, CD99	Alive	4 months
27	Taguchi *et al*[31]	STAT6, CD34, Bcl-2, vimentin, cytokeratin AE1/AE3	Alive	12 months
28	Jariwalla *et al*[32]	STAT6, CD34	NA	NA
29	Marotti *et al*[33]	STAT6, CD34	Alive	6 months
30	Addeo *et al*[6]	STAT6, CD34, Bcl-2, Ki67 7%	NA	NA
31	Rodriguez *et al*[2]	STAT6	Alive	12 months
32	Jones *et al*[34]	STAT6, CD34	Alive	1 months
33	Liu *et al*[35]	CD34, STAT6, CD99	Alive	6 months
34	Present case	TAT6, CD34, Bc1-2, Vimentin, CD99, Ki67 40%	Alive	3 months

STAT6: Signal transducer and activator of transcription 6; ER: Estrogen receptor; PR: Progesterone receptor; SMA: Smooth muscle actin; NA: Not applicable.

In this case, CT showed no enhancement in the arterial phase and heterogeneous enhancement in the venous area. We believe that it should be distinguished from neuroendocrine tumors, which show enhanced CT from the arterial phase to the portal venous phase[13,37], which makes it difficult for us to distinguish the disease, so many scholars before us also misdiagnosed it before surgery[1,10,11,13,26]. At the same time, we believe that it should also be differentiated from pancreatic cancer and solid pseudopapillary tumors of the pancreas. The imaging features of this tumor have been described in detail in our previous work on pancreatic tumors[42].

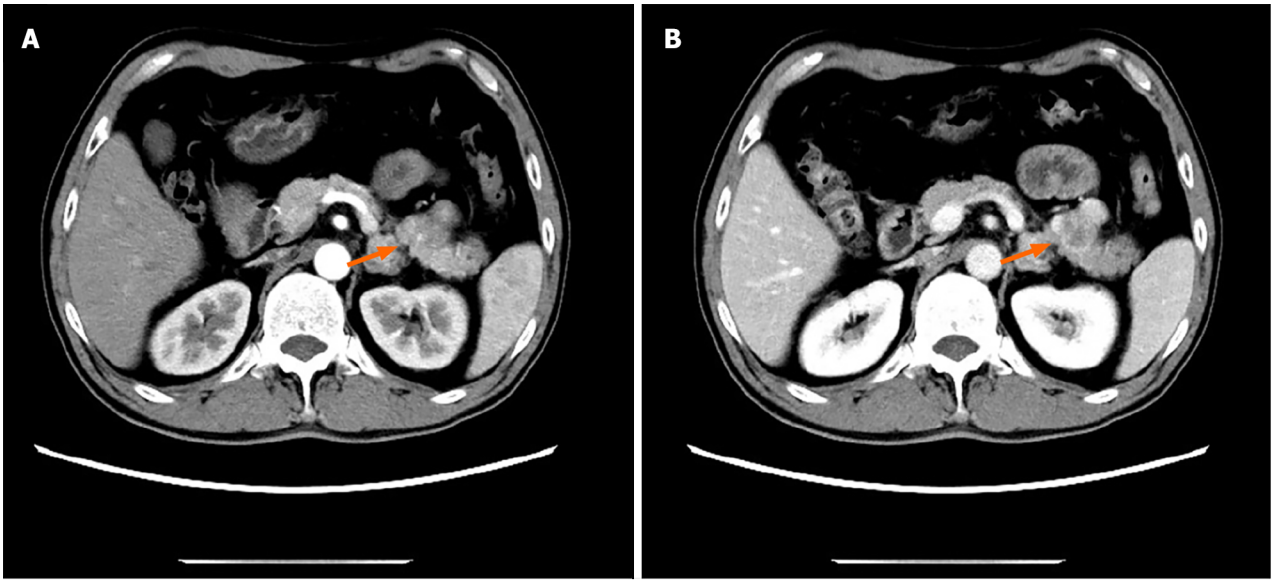

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Figure 1 Abdominal computed tomography scan showing a 5.52 cm × 2.82 cm × 2 cm mass in the pancreas (orange arrows). A: No enhancement in the arterial region. B: Heterogeneous enhancement in the venous area.

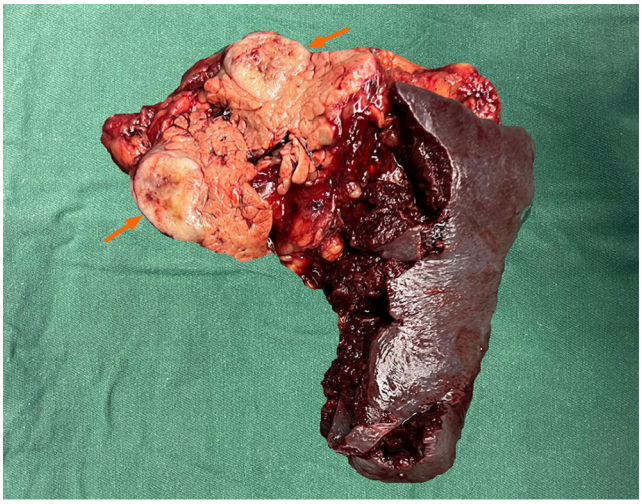

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Figure 2 Postoperative surgical specimen: Pancreatic tail and spleen (tumor cut open chart) (orange arrows).

Most SFTs are benign[43], and malignant SFTs account for 10%-15%[30,39,44,45]. The histopathological features of malignant SFT: (1) Hypercellularity; (2) more than four mitotic figures per ten high-power fields; (3) nuclear pleomorphism; (4) hemorrhage and necrosis; (5) tumor diameter ≥10 cm; and (6) positive margins[15,21,46]. Ki-67 can also differentiate benign from malignant tumors, with a cutoff value of 0%-5% (indeterminate in 5%-10%) for benign tumors and > 10% for malignant SFTs[40,47]. In our case, our patient had a Ki-67 proliferation index of 40%; therefore, the tumor was possibly malignant. Because SFT of the pancreas is rare, there are no uniform treatment criteria; nevertheless, complete resection is the treatment of choice for intra-abdominal SFTs[1,7,10-12,15], and post-surgical follow-up is critical because SFTs have a high recurrence rate. Due to the increasing number of reported cases of SFT, we believe there will be a complete system of treatment.

CONCLUSION

Because of the non-specific clinical symptoms and radiological features of SFT of the pancreas, the diagnosis is challenging with preoperative radiological and laboratory examinations alone. A definitive diagnosis relies on histopathology and immunohistochemistry. In cases where the tumor is found in the pancreas, and the diagnosis cannot be confirmed, it is recommended to obtain histopathology with ultrasound aspiration. As this presentation is rarely

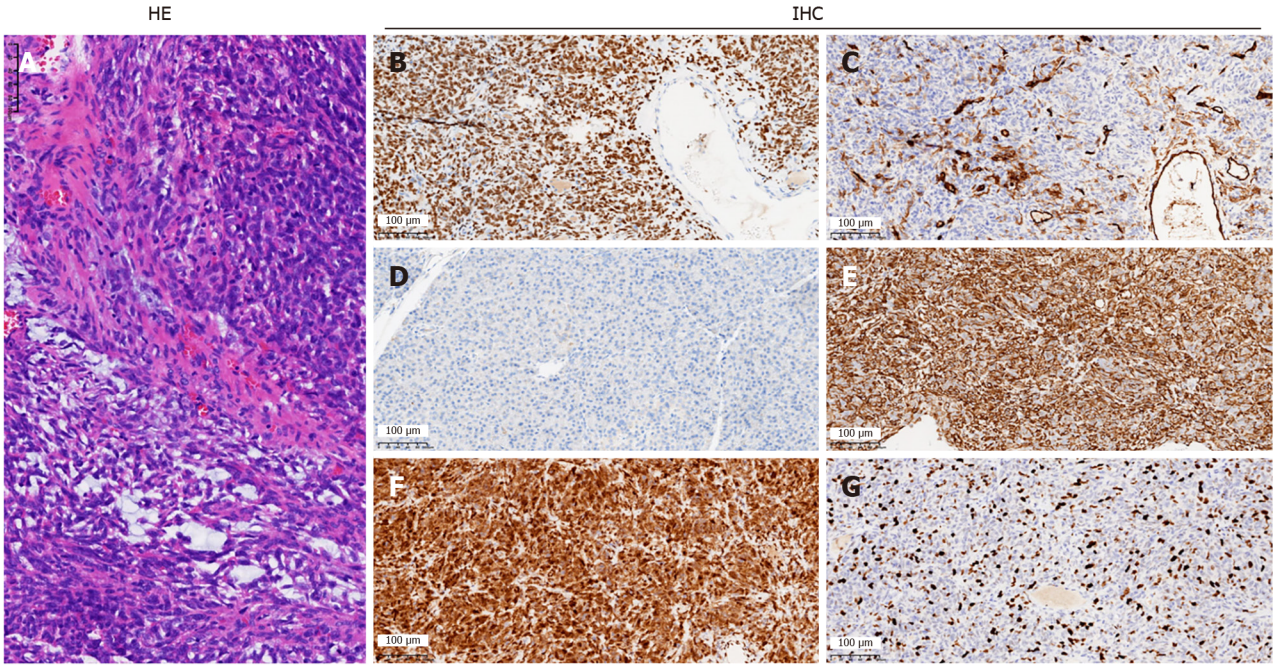

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Figure 3 Representative results of hematoxylin and eosin and immunohistochemical staining of surgical specimens of solitary fibrous tumor of the pancreas. A: Hematoxylin and Eosin staining (hematoxylin and Shuhong); B: Immunohistochemistry (original magnifcation of × 400) signal transducer and activator of transcription 6; C: CD34; D: CD99; E: Vimentin; F: Vimentin; G: Ki-67.

reported, there is a lack of uniform treatment criteria, and surgery is effective. However, the tumor may lead to potential recurrence or metastasis; therefore, long-term follow-up is recommended.

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FOOTNOTES

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Co-corresponding authors: Xin-Hua Zhou and Li-Li Qiu.

Author contributions: Wang WW, Zhou SP and Wu X, investigation, data curation, writing-original draft; Wang LL investigation, provide image pictures; Ruan Y investigation, funding acquisition, medical history collection; Lu J investigation, supervision; Li HL pathology to provide; Ni XL pathology to provide; Qiu LL and Zhou XH resources, writing-review and editing, supervision, project administration funding acquisition; all authors read and approved the final manuscript. The author contributions of Wang WW and Wu X as co-first authors, Qiu LL and Zhou XH as corresponding authors are as follows: Wang WW and Wu X are co-first authors because they made equal contributions to the research and were involved in all stages of the study, from design to data collection and analysis. They both contributed significantly to the writing of the manuscript and are responsible for the accuracy and validity of the results reported. Qiu LL and Zhou XH are corresponding authors because they supervised the research project, provided guidance and expertise throughout the study, and ensured the quality and reliability of the data presented. As senior researchers, they are responsible for the overall content and implications of the study and act as points of contact for further information or queries.

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