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Retroperitoneal cavernous hemangioma misdiagnosed as lymphatic cyst: A case report and review of the literature

Xiao-Fan Hou, Zhan-Xue Zhao, Lin-Xun Liu, Hao Zhang

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Abstract

BACKGROUND

Primary abdominal and retroperitoneal cavernous hemangioma is a vascular tumor and rarely seen in the clinic. Due to the lack of specific imaging features, retroperitoneal cavernous hemangioma cannot be diagnosed accurately. Some symptoms may develop with the enlargement of lesion volume or the occurrence of complications such as rupture or oppression. We report here a special case who was admitted with chronic abdominal pain. Admission examination suggested a retroperitoneal lymphatic duct cyst. Laparoscopic resection of the retroperitoneal mass was performed, and histological examination confirmed retroperitoneal cavernous hemangioma.

CASE SUMMARY

The patient was a 43-year-old Tibetan woman with intermittent left lower abdominal pain and discomfort 3 years ago. Ultrasonography revealed a cystic mass in the retroperitoneum with clear boundaries, internal septa, and no blood flow signal. Computed tomography (CT) and magnetic resonance imaging (MRI) showed an irregular space-occupying mass in the retroperitoneum, and retroperitoneal lymphatic cyst was considered. Plain CT scanning showed multiple cyst-like hypo-intense shadows in the retroperitoneum, partially fused into a mass, and no obvious enhancement was found on enhanced scanning. MRI showed multiple irregular clump-like long T1 and long T2 signal shadows above the pancreas, within which linear short T2 signal shadows were seen. Diffusion-weighted imaging sequence showed hypo-signal shadows, without obvious enhancement on enhanced scanning. Ultrasound, CT, and MRI all suggested the possibility of retroperitoneal lymphatic cyst. However, the patient was finally diagnosed with retroperitoneal cavernous hemangioma by pathological examination.

CONCLUSION

Retroperitoneal cavernous hemangioma is a benign lesion, and it is difficult to make a diagnosis preoperatively. Surgical resection may be the only treatment, which not only allows histopathological confirmation as a diagnostic purpose and excludes any risk of malignancy, but also avoids invasion of adjacent tissues, oppression, and other complications as a therapeutic goal.

Key Words: Cavernous hemangioma; Retroperitoneal; Diagnosis; Treatment; Case report

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Core Tip: In this report, we describe a case of retroperitoneal cavernous hemangioma misdiagnosed as retroperitoneal lymphangiod cyst preoperatively.

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INTRODUCTION

Cavernous hemangioma is a benign vascular neoplasm consisting of proliferative vascular endothelial cells characterized by abnormal angiogenesis[1], with a propensity for orbital region, liver, brain cadres, and skin involvement but rarely involving the retroperitoneum[2-6]. Cavernous hemangiomas can occur at any age[7] and grow rapidly in infancy, but may spontaneously degenerate late in life[8,9]. In the early stage, most patients tend to be asymptomatic, while as the tumor grows, patients require hospital management due to the presence of oppressive symptoms such as pain, nausea, and vomiting. Cavernous hemangioma has diverse imaging features. Ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) contribute to the initial identification and localization of pathological lesions. However, it is difficult to make a definitive diagnosis of retroperitoneal cavernous hemangioma by any non-surgical modality, and this disease is often misdiagnosed as adenocarcinoma, neuroendocrine tumors, or other solid pancreatic entities. In the case that we report here, the patient was preoperatively diagnosed with a retroperitoneal lymphatic duct cyst.

Despite the benign nature of cavernous hemangioma, the case that we report had suffered from long-term intermittent pain and discomfort. Hence, we chose to perform space-occupying lesion resection under laparoscopy. Here, we also performed a literature review of the diagnosis and treatment of retroperitoneal cavernous hemangioma and retroperitoneal lymphatic cyst in recent years and made a comparison between them.

CASE PRESENTATION

Chief complaints

A 43-year-old woman of Tibetan ethnicity was admitted to the hospital with a 3-year history of pain and discomfort in the left lower abdomen and aggravation in recent 1 year.

History of present illness

The pain was not obvious at the time of admission examination 3 years ago, so the patient did not receive further treatment.

History of past illness

The patient underwent tubal ligation 20 years ago, with no history of acute pancreatitis, abdominal trauma, chronic diseases, or infectious diseases.

Personal and family history

The patient denied any family history of malignant tumours.

Physical examination

Admission physical examination revealed: Normal body temperature, respiration, and blood pressure,

and a pulse rate of 65 beats/min; good nutritional status; flat abdomen, no intestinal pattern and peristaltic waves, and no varicose veins in the abdominal wall; soft abdomen, with left lower abdominal tenderness, no muscle tension and rebound pain, no palpable liver or spleen under the ribs, no abnormal masses palpated in the abdomen, and Murphys' sign negativity; no shifting dullness, with normal bowel sounds, and no air-over-water sounds or abnormal vascular murmurs.

Laboratory examinations

Laboratory tests after admission indicated that routine blood indexes, tumor markers, and biochemical indicators were all within normal range.

Imaging examinations

On ultrasonography, a retroperitoneal cystic mass with a size of 88 mm × 62 mm was seen, with clear borders and internal separation, and no significant blood flow signal was observed on color Doppler flow image. Enhanced CT showed multiple retroperitoneal cystic hypo-intense shadows, partially fused into a mass of approximately 46 mm × 35 mm in size, with no significant enhancement on enhancement (Figure 1). MRI showed multiple irregular masses of long T1 and long T2 signal shadows above the pancreas, within which linear short T2 signal shadows were seen. The diffusion-weighted imaging sequence showed hypo-signal shadows (Figure 2). The retroperitoneal layer was clear without obviously enlarged lymph nodes. These imaging findings did not suggest evidence of mass invading surrounding organs or retroperitoneal lymphadenopathy.

FINAL DIAGNOSIS

The preoperative diagnosis was retroperitoneal lymphatic duct cyst. Laparoscopic resection of the retroperitoneal mass was performed after surgical evaluation.

TREATMENT

During the operation, a soft cystic tumor above and posterior to the pancreas was found, which was closely connected with the inferior vena cava. The size of the tumor was about 6 cm × 7 cm, and it was prone to bleeding when touched. Reddish fluid was seen in the cyst, and the tumor envelope was intact (Figure 3). Microscopically, blood vessels with different lumen sizes and uneven wall thickness were seen in the tissues, and coagulation was seen in some vessels, conforming to the morphology of cavernous hemangioma, and a small amount of adipose tissue was seen around (Figure 4). Immunohistochemistry revealed CD34 (+), CD31 (+), SMA (+), and Desmin (partially +).

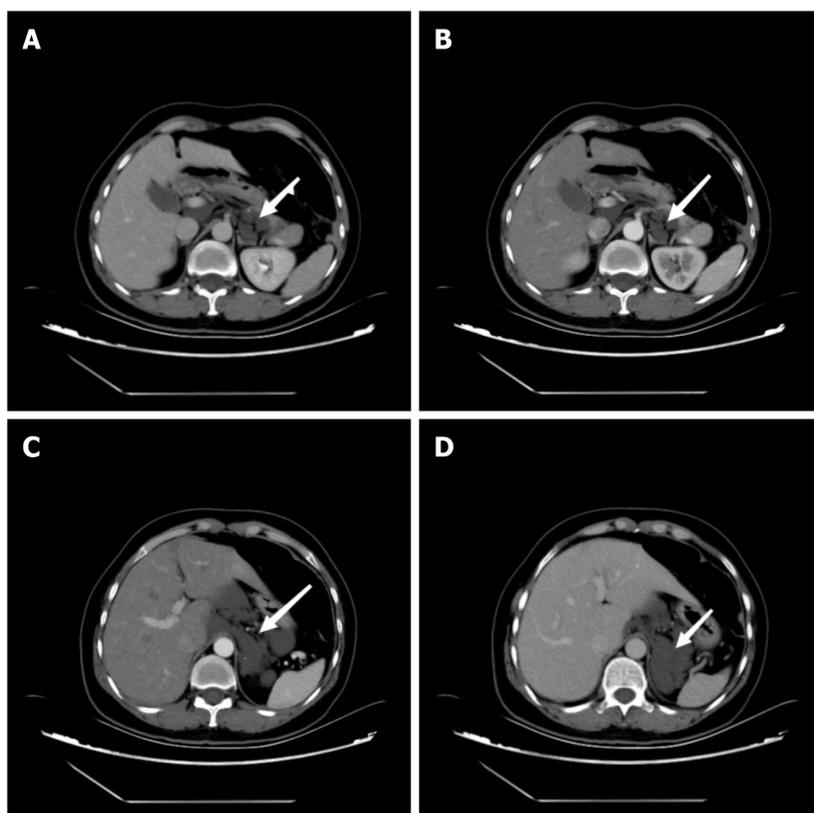
OUTCOME AND FOLLOW-UP

The histopathology confirmed the diagnosis of retroperitoneal cavernous hemangioma.

DISCUSSION

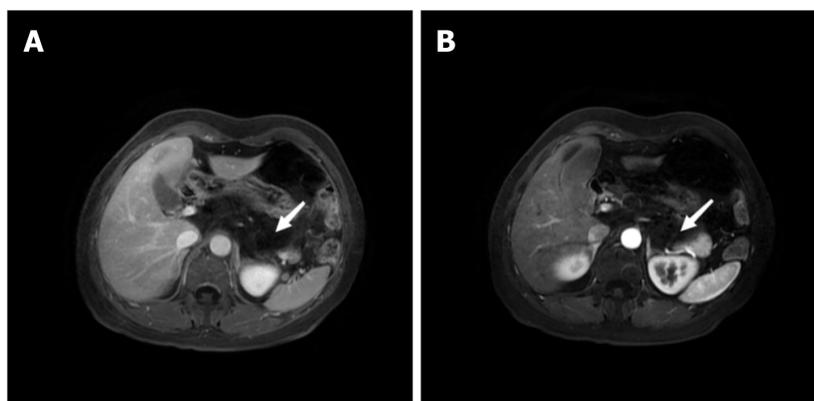
In this report, we describe a case of retroperitoneal cavernous hemangioma misdiagnosed as retroperitoneal lymphangioid cyst preoperatively. Retroperitoneal tumors are extremely rare, accounting for less than 0.2% of all tumor types[10]. This disease mainly originates from the fat tissue, loose connective tissue, and fascia of the abdominal cavity. Hemangiomas located in the mesentery and gastrointestinal tract can invade outwardly the peritoneal cavity[11,12]. Liposarcoma is the most common tumor located in the retroperitoneal space. Among the malignant tumors of the retroperitoneum, liposarcoma and leiomyosarcoma are most frequently seen, while teratomas, cysts, and neuromas are the common benign tumors[3,13-14]. Vascular tumors include lymphangiomas, hemangiomas, lymphangioidomas, hemangiotheliomas, and hemangioblastomas, all of which are benign lesions with slow growth and few malignant changes. These lesions are commonly found in the skin, eyes, liver, brain, spleen, and other organs, while cavernous hemangioma is extremely rare[15].

The majority of cavernous hemangiomas often do not exhibit obvious clinical symptoms in the early stage due to the small volume of the space-occupying lesion. Non-specific symptoms, such as abdominal pain and anemia, present only when oppressive symptoms or invasion of adjacent tissues occur[16]. A few patients may be admitted due to lower back pain or gastrointestinal bleeding[17]. In the literature that we review, there are 6 cases of cavernous hemangiomas primarily originating from the adrenal gland[18-23], 12 cases of cavernous hemangiomas primarily originating from the pancreas



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Figure 1 Computed tomography images. A: Computed tomography showed multiple retroperitoneal cystoid hypodensity shadows (arrow); B-D: No significant enhancement was observed in the arterial phase, balance phase, and venous phase (arrows).



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Figure 2 Magnetic resonance imaging. A, B: The retroperitoneal layer was clear without obviously enlarged lymph nodes (arrows).

[24-32], 3 cases of primary retroperitoneal cavernous hemangioma[33-35], 7 cases unspecified[14,36-41], and 1 case originating from the psoas major muscle[42]. Detailed clinical data are shown in Table 1.

The imaging manifestations of cavernous hemangioma are also varied. On ultrasound, cavernous hemangioma is often manifested as internal hypo-echo or irregular mixed echo, usually with no or low blood flow signal, so it is difficult to determine the nature of the lesion. CT and MRI are extensively used for preoperative diagnosis of cavernous hemangioma. Usually, hemangiomas can be significantly enhanced in the arterial phase on CT. However, pancreatic cavernous hemangioma is a cystic tumor that usually contains neurovascular components, with an arteriovenous shunt, and lacks larger supply vessels, which may result in the absence of enhancement in the arterial phase[32]. Also, the different proportions of cystic and solid components in the tumor may lead to different degrees of enhancement in the arterial phase[43]. Only a small number of patients show characteristic changes, and it is usually difficult to make a diagnosis by imaging means. CT scan can locate the tumor and distinguish the density changes of the tumor, but origin determination and qualitative diagnosis can hardly be realized.

Table 1 Clinical data of patients

No.	Gender	Age	Ultrasonic	CT	MRI	Positive sign	Preoperative diagnosis	Postoperative pathology	Ref.
1	Male	20	Small amount of low-velocity blood flow signal in the tumor with heterogeneous echogenicity	No	No	No	Retroperitoneal cavernous hemangioma	Cavernous hemangioma (lumbar major muscle origin)	[44]
2	Female	51	No	The edges of the mass are clear and smooth, with heterogeneous density, mild enhancement at the edge of enhancement, and nodular significant enhancement seen internally	The lesion shows delayed marked enhancement	Pain in the right lower abdomen, numbness and pain in the right lower extremity	Neurogenic tumor?	Cavernous hemangioma	[37]
3	Female	16	No	No	No	Fresh blood stools, black stools, severe anemia	Hemorrhoids? Colonic vasodilatation?	Cavernous hemangioma	[38]
4	Male	35	No	Uniform density mass. Homogeneous enhancement is seen	Smooth margins of the mass and uniform internal signal	Abdominal pain	Adrenal tumors; tumors of vascular origin; tumors of neurological origin; adrenal pheochromocytoma?	Cavernous hemangioma	[39]
5	Male	57	No	Isointense-slightly hypointense shadow of the right adrenal gland with clear margins, lobulated, heterogeneous density, nodular enhancement	No	Elevated blood pressure	Cortical cancer?	(Right adrenal gland) cavernous hemangioma	[19]
6	Male	14	Clearly defined mass with internal cystic mass and no significant blood flow signal	Hematoma in the right adrenal region	No	No	Tumor with hemorrhage	Hemangioma (left adrenal gland) with hemorrhagic necrosis	[20]
7	Male	57	No	Uneven density masses	No	Left upper abdominal pain	Left adrenocortical carcinoma, pheochromocytoma? Retroperitoneal mesenchymal-derived tumor?	Hemangioma (left adrenal gland) with hemorrhagic necrosis	[21]
8	Female	70	No	Clearly defined mass with heterogeneous density and scattered foci of speckled calcification at the margins; enhanced lesions with heterogeneous enhancement and multiple foci of nodular hyperenhancement at the periphery	No	Abdominal pain	Giant retroperitoneal occupancy with consideration of benign tumor of adrenal origin	(Left adrenal) cavernous hemangioma; immunohistochemistry: Vim (+)	[22]
9	Female	47	No	The density of the mass is uneven, and vascular shadow is seen in enhancement	T1W1 low signal, T2W1 heterogeneous high signal, lesion is heterogeneously reinforced, vascular shadow is seen	Abdominal distension, abdominal pain	Malignant tumors	Cavernous hemangioma with bleeding; Immunohistochemistry: CD31 (+); CD34 (+), CD2-40 (+), ERG (+), SMA (+)	[40]

10	Female	71	Irregular hypoechoic solid occupying lesion of the pancreas	Heterogeneous hypodense lesion in the pancreatic neck with clear borders and marked enhancement	No		Abdominal pain	No	Pancreatic cavernous hemangioma; immunohistochemistry: CD31 (+), CD34 (+), CD2-40 (-)	[25]
11	Female	37	Cystic solid mass	No	No		Abdominal pain	No	Pancreatic cavernous hemangioma	[26]
12	Female	63	Cystic solid mass	No	No		Abdominal pain	Pancreatic cancer	Pancreatic cavernous hemangioma with calcification and cyst formation	[26]
13	Male	79	Cystic solid mass in the head of the pancreas with clear borders, uneven internal echogenicity, scattered multiple echogenic areas, and poor blood flow signal	Occupation of the head of the pancreas with lamellar enhancement and increased delayed enhancement, with multiple microvascular shadows seen within	No		Abdominal pain, abdominal distension	No	Pancreatic cavernous hemangioma	[27]
14	Male	55	Cystic predominant mass in the tail of the pancreas with clear borders. Irregular shape, no obvious blood flow signal	Hypodense lesion in the body of the pancreas	Cystic occupying lesion in the body of the pancreas		Abdominal pain	Cystadenoma, cyst?	Pancreatic cavernous hemangioma	[28]
15	Male	49	No	A round-like hypodense lesion is seen in the neck of the pancreas, and enhancement is seen	Pancreatic neck occupancy with clear margins and homogeneous signal; enhancement is not obvious		Pain in the lower back and lower right abdomen	Pancreatic cancer?	Pancreatic cavernous Hemangioma	[29]
16	Female	58	No	Plain scan shows a round-like hypodense mass in the tail of the pancreas with well-defined borders, and moderate enhancement in the arterial phase and "thin thread-like" moderate enhancement in the venous and delayed phases	A round-like lesion was seen in the tail of the pancreas, with uniform low signal on T1WI and high signal on T2WI, and a little bit of slightly low signal was seen inside (DW1 showed slightly high signal, and mild to moderate patchy enhancement was seen inside the lesion)		Abdominal distension, abdominal pain, jaundice	Cystadenoma of the pancreas	Pancreatic cavernous hemangioma	[30]
17	Male	66	The envelope was intact and continuous, and multiple irregular echogenic areas were visible inside, separated by hyperechoic structures	A large round-like hypodense mass in the right upper abdomen with clear borders and nodular irregular enhancement around the mass in the arterial phase	A round abdominal abnormal signal shadow with well-defined borders, T1-weighted moderately low signal and nodular low signal in the periphery; T2-weighted mass is predominantly high signal, with striated low signal inside. After enhancement, T1-weighted nodular enhancement was seen in the periphery, and the enhancement became more obvious on delayed scan and expanded inward. There was no central enhancement on both scans		Symptomless	Cavernous hemangioma	Adrenal cavernous hemangioma	[31]
18	Female	54	Strongly echogenic mass in the right posterior lobe of the liver with clear boundaries and uneven internal echogenicity	A rounded hypointense shadow is seen in the right adrenal area, and a more hypointense area with clear borders is visible within it; the periphery of the lesion is mildly enhanced	The oval-shaped mass between the liver and the right kidney is hyposignal on T1WI, with an eccentric more hyposignal area within it; it is high-signal on T2WI, with a well-defined border and a non-enhancing hyposignal area within it		Abdominal pain	Retroperitoneal space-occupying lesions (malignant)	Primary retroperitoneal cavernous hemangioma	[34]

19	Female	60	Cystic	Slightly hypointense shadow with inconspicuous enhancement	No	Abdominal pain, fever	Cystic lesion of the tail of the pancreas. Cystic adenoma? Cystic adenocarcinoma?	Pancreatic cavernous hemangioma	[32]
20	Female	41	Cystic solid	No	No	Medical examination	Cystic dominant cystadenoma or islet cell carcinoma in the body of the pancreas	Pancreatic cavernous hemangioma	[32]
21	Female	30	Cystic solid	No	No	Abdominal pain, choking on food	Cystic solid occupancy of the head of the pancreas is likely to be benign	Pancreatic cavernous hemangioma	[32]
22	Female	57	Uneven hypoechoic	Uneven reinforcement	No	No	Gastrointestinal mesenchymal tumors (GISTs), carcinoid tumors, neurogenic tumors, metastatic lymphadenopathy, or other rare tumors	Retroperitoneal cavernous hemangioma	[41]
23	Male*	63	Mixed echogenicity within the indistinct contour of the head of the pancreas, CDFI shows hyperechoic and visible blood flow within the separation	A well-defined cystic mass in the head of the pancreas with foci of internal calcification	No	Abdominal pain, constipation		Pancreatic cavernous hemangioma	[33]
24	Male	36	Uneven echoes	Tumor between the dorsal head of the pancreas and right kidney without significant enhancement	Lower signal in the tumor, some relatively high intensity areas	Abdominal pain	Retroperitoneal sarcoma	Cavernous hemangioma; Immunohistochemistry: CD31 (+), CD34 (+), CD2-40 (partially +)	[42]
25	Male	35	No	Well-defined, cyst-like masses, no enhancement of the mass wall in the arterial and portal phases, persistent hypodensity of the mass	No	Abdominal pain	Benign tumors such as lymphangioma cysts, GISTs, or primitive retroperitoneal benign tumors	Primary retroperitoneal cavernous hemangioma; Immunohistochemistry: CD31 (+), CD34 (+), DOG1 (-), CD117 (-)	[35]
26	Female	71	No	Low density, no enhancement	No	Lower limb edema, dyspnea, abdominal distention, anemia		Retroperitoneal cavernous hemangioma; Immunohistochemistry: CD34 (+)	[43]
27	Male	70	No	Uniform density, surrounding calcification	(a) Molecular water diffusion limitation within the lesion on T2 fat-saturated weighted image; (b) high signal intensity intra-lesion region on T1 fat-saturated weighted image; (c) high signal region on enhanced T1 fat-saturated weighted image; (d) inhomogeneous enhancement	Physical examination		Adrenal cavernous hemangioma	[23]
28	Male	67	No	Clearly defined mass with significant peripheral enhancement in the early arterial phase	No	Back pain	Adrenal tumor, pheochromocytoma	Adrenal cavernous hemangioma	[24]
29	Male	38	No	Cyst-like, well-enveloped tumor	No	Abdominal pain		Primary retroperitoneal	[36]

with mildly enhancing walls in the portal phase

cavernous hemangioma;
Immunohistochemistry: CD31
(+), Vim (+)

CT: Computed tomography; MRI: Magnetic resonance imaging.

It is also because cavernous hemangioma lacks enough characteristic imaging manifestations that it is easy to be misdiagnosed.

Retroperitoneal lymphatic cyst usually presents as a large, thin-walled, multi-septal cystic mass, with fluid content varying from fluid to fat attenuation. The elongated shape and crossover from the retroperitoneal compartment to the adjacent area are the characteristics of the mass. Few cystic lymphangiomas have wall calcification[44], and there is no gold standard for diagnosis.

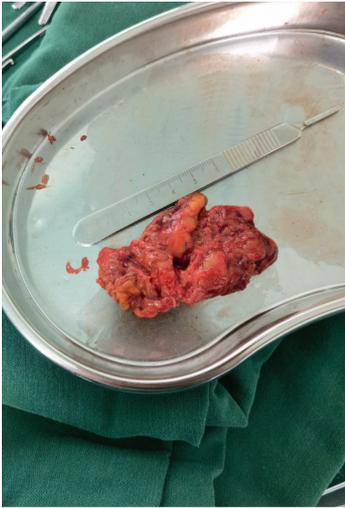
Currently, the gold standard for the diagnosis of retroperitoneal cavernous hemangioma is still pathological examination. Microscopically, cavernous hemangioma is mainly composed of dilated abnormal sinuses lined with monolayer vascular endothelial cells and incompletely spaced fibrous tissues within the sinuses, forming a sponge-like structure[45,46]. Depending on the size of the vascular spaces, they can be capillary or spongy. In immunohistochemistry, CD31- and CD34-positivity suggests benign vascular tumors such as hemangiomas and lymphangiomas, and D2-40-negativity excludes lymphangiomas.

There is no need for further treatment or surgical intervention when retroperitoneal hemangioma is asymptomatic, but further surgical treatment is needed when the tumor grows rapidly, oppresses adjacent organs, or develops non-specific symptoms[47]. Unlike the polycythemia exhibited by hemangiomas originating from other regions, retroperitoneal cavernous hemangioma is mostly ischemic. It is generally considered to be associated with asymptomatic episodes[48]. Due to the difficulty in diagnosing retroperitoneal cavernous hemangioma, it is necessary to fully understand the imaging features of retroperitoneal hemangioma and combine them with relevant clinical and laboratory tests to exclude the possibility of malignant tumor, thereby reducing the surgical or postoperative complications. The surgical modality should be selected according to the location of the tumor and its proximity to the adjacent tissues. If the tumor is large or invades the adjacent organs, interventional treatment can be chosen accordingly; relatively, in the presence of symptoms of gastrointestinal or biliary tract obstruction, pancreaticoduodenectomy, gastrojejunostomy, or choledochojejunostomy can be performed to relieve the obstruction if necessary.

Overall, although retroperitoneal cavernous hemangioma is a benign lesion, it is difficult to make a diagnosis preoperatively. Surgical resection may be the only treatment, which not only allows histopathological confirmation as a diagnostic purpose and excludes any risk of malignancy, but also avoids invasion of adjacent tissues, oppression, and other complications as a therapeutic goal.

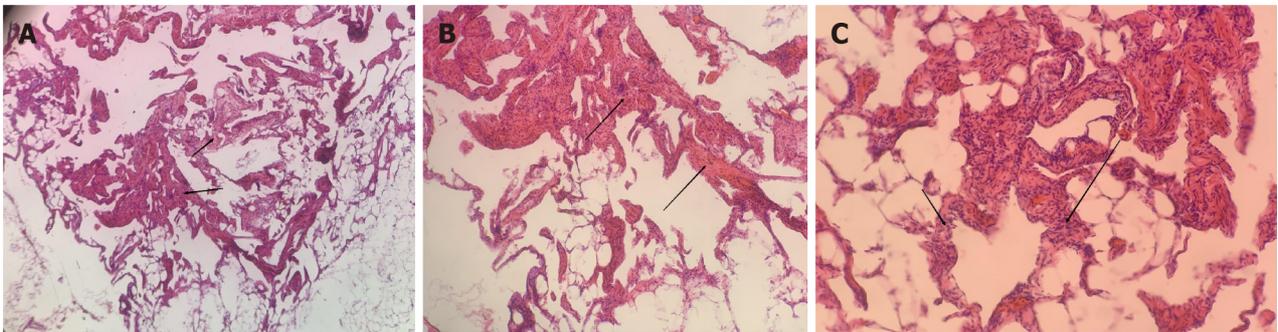
CONCLUSION

Overall, although retroperitoneal cavernous hemangioma is a benign lesion, it is difficult to make a diagnosis preoperatively. Surgical resection may be the only treatment, which not only allows



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Figure 3 Resected tumor lesion.



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Figure 4 Pathological examination. A-C: Blood vessels with different lumen sizes and uneven wall thickness were seen in the tissues (arrows). A: 50×; B: 100×; C: 150×.

histopathological confirmation as a diagnostic purpose and excludes any risk of malignancy, but also avoids invasion of adjacent tissues, oppression, and other complications as a therapeutic goal.

FOOTNOTES

Author contributions: Hou XF contributed to manuscript writing and editing, and data collection; Zhao ZX contributed to data analysis; Liu LX contributed to conceptualization and supervision; all authors have read and approved the final manuscript.

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