

# World Journal of *Clinical Cases*

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# Successful resection of a huge retroperitoneal venous hemangioma: A case report

Yan Qin, Peng Qiao, Xing Guan, Song Zeng, Xiao-Peng Hu, Biao Wang

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## Abstract

### BACKGROUND

Venous hemangioma is a benign and non-invasive type of tumor, which is rarely identified due to the absence of clinical manifestations. A retroperitoneal benign tumor is comparatively rare, and hemangioma is exceptional. Because of the different types and locations of hemangioma, presentations are varied; thus, establishing an accurate diagnosis before surgery is challenging.

### CASE SUMMARY

A 45-year-old female patient visited our hospital with the complaint of a retroperitoneal mass without symptoms discovered during a medical examination. An abdominal and pelvic computed tomography (CT) revealed a giant hypodense mass that extended from the lower edge of the liver down to the right groin and showed no marked enhancement in the arterial phase of the enhanced CT. On magnetic resonance imaging, the retroperitoneal mass was hyperintense on the T2-weighted image and hypointense on the T1-weighted image. The mass was completely resected and confirmed as a venous hemangioma by pathology.

### CONCLUSION

Venous hemangioma is rare in adults, and an accurate diagnosis before surgery is challenging. Surgery is the curative treatment for venous hemangioma, and the definitive diagnosis relies on pathology.

**Key Words:** Retroperitoneal hemangioma; Venous hemangioma; Diagnosis; Case report

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**Core Tip:** Venous hemangioma is a benign disease which is rarely reported as it lacks clinical manifestations. However, many cases have presented with abdominal symptoms. An accurate diagnosis before surgery is challenging due to non-specific manifestations and low incidence. Surgery is the first-line treatment for venous hemangioma, and the definitive diagnosis depends on pathology.

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## INTRODUCTION

Venous hemangioma is defined as a benign tumor consisting of vein-like vessels, and is a rare but clinically significant disease. The lesion may occur in various sites but is rarely found in the retroperitoneum[1,2]. Retroperitoneal hemangioma is extremely rare and accounts for less than 5% of all retroperitoneal tumors in adults[3]. Data indicate that abdominal distension and pain are the two most common manifestations. To the best of our knowledge, only a few isolated cases have been reported. Due to the different locations and varied manifestations of venous hemangioma, establishing an accurate diagnosis before surgery is challenging. We here report a case of giant venous hemangioma in the retroperitoneum and review the literature concerning the clinical and pathological features of this disease.

## CASE PRESENTATION

### Chief complaints

A 45-year-old female patient visited our hospital with the complaint of a retroperitoneal mass without symptoms discovered during a medical examination.

### History of present illness

The patient was found to have a retroperitoneal mass during a physical examination at another hospital 5 mo previously, and ultrasonography revealed a large anechoic mass that occupied almost all of the right side of the peritoneal cavity.

### Physical examination

A physical examination revealed no mass and tenderness in the abdomen.

### Laboratory examinations

Laboratory tests showed no abnormalities.

### Imaging examinations

Abdominal computed tomography (CT) (Figure 1A and B) revealed an enormous hypodense mass extending from the lower edge of the liver down to the right groin, with a regular margin and without marked enhancement in the arterial phase of the enhanced CT, which suggested a benign tumor. Magnetic resonance imaging (MRI) (Figure 1C and D) revealed a retroperitoneal mass with low signal intensity on the T1-weighted image and high signal intensity on the T2-weighted image. A benign cyst in the right retroperitoneum was diagnosed based on these findings.

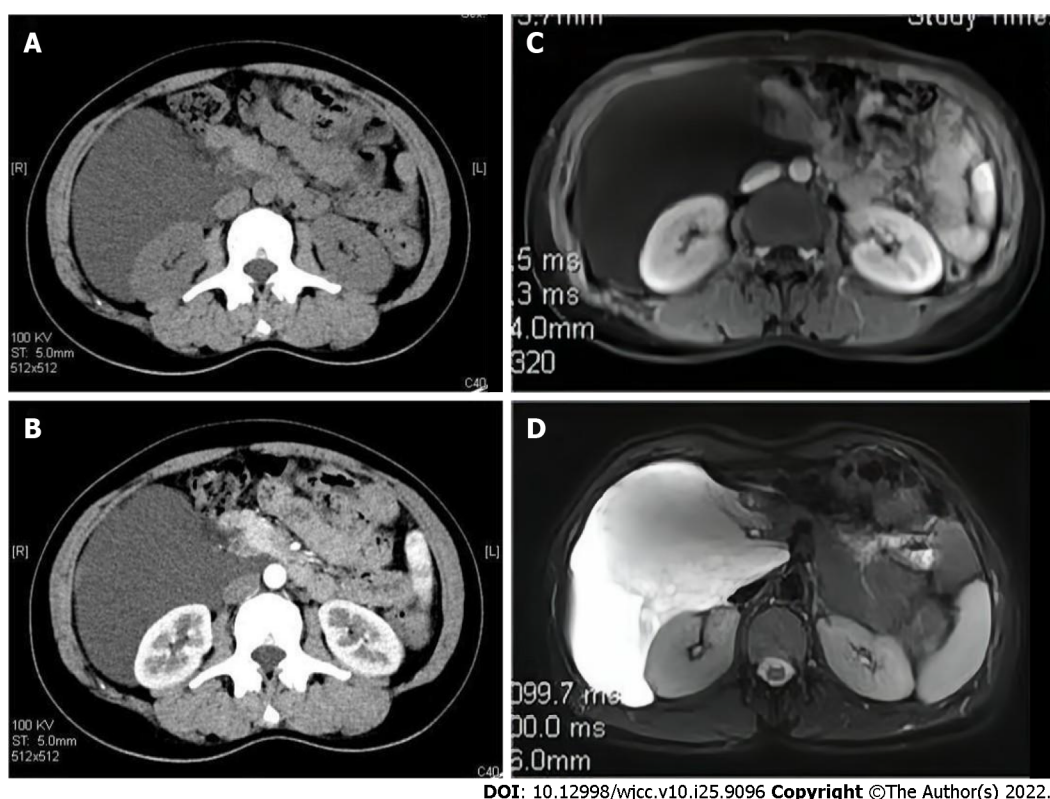
## FINAL DIAGNOSIS

Retroperitoneal venous hemangioma.

## TREATMENT

A laparoscopic exploration and cystectomy were performed. During the laparoscopic surgery, it was found that a milky white cystic neoplasm measuring approximately 20 cm was loosely attached to the





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**Figure 1** Imaging findings of a retroperitoneal mass. A: CT scans of the abdomen demonstrates a hypodense mass in the right retroperitoneum; B: The enhanced CT shows the mass without remarkable enhancement; C: On MRI, T1-weighted image shows the mass as low signal intensity without enhancement; D: T2-weighted image shows the mass with high signal intensity. CT: Computed tomography; MRI: Magnetic resonance imaging.

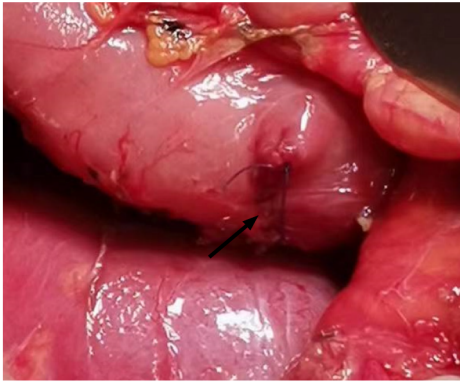
muscular tissue. The tumor displaced the intestine and caused compression of the inferior vena cava, right kidney, as well as the bladder, without signs of infiltration. Moreover, the mass protruded from the retroperitoneal space into the duodenal wall, and was firmly attached to the wall of the second part of the duodenum and the head of the pancreas. Therefore, we chose to separate the loosely attached capsule wall initially, and milky liquid was noted during the separation process. The fluid was immediately taken for laboratory testing, and the results indicated that amylase was not elevated. About 2 L of milky white liquid was drained intraoperatively. Then to protect the duodenum and pancreas, the cystic wall which was tightly adhered to the duodenum was removed by laparotomy. There was no significant blood loss, and the entire capsule wall was completely resected (Figure 2). A 8 cm grayish specimen and a 5 cm fawn cystic specimen were obtained for pathological examination. Microscopic examination showed that the cystic wall was lined by a single layer of flattened cells, with smooth muscle cells and adipose tissue located around this layer. The immunohistochemical study showed that these cells stained positive for smooth muscle actin (SMA) and vascular marker CD34, and some endothelial cells were positive for CD31 and D2-40. No atypical cells with hyperproliferation or mitotic division were observed, and the proliferation marker (Ki-67 antigen) expression was less than 5 (Figure 3). In brief, these findings indicated that the retroperitoneal mass, which was derived from the vein, was a venous hemangioma.

## OUTCOME AND FOLLOW-UP

The patient had an uneventful recovery, and no residual mass in the retroperitoneum was verified on CT scan 1 mo postoperatively. At the 6-mo follow-up evaluation, the patient was doing well without recurrence of hemangioma.

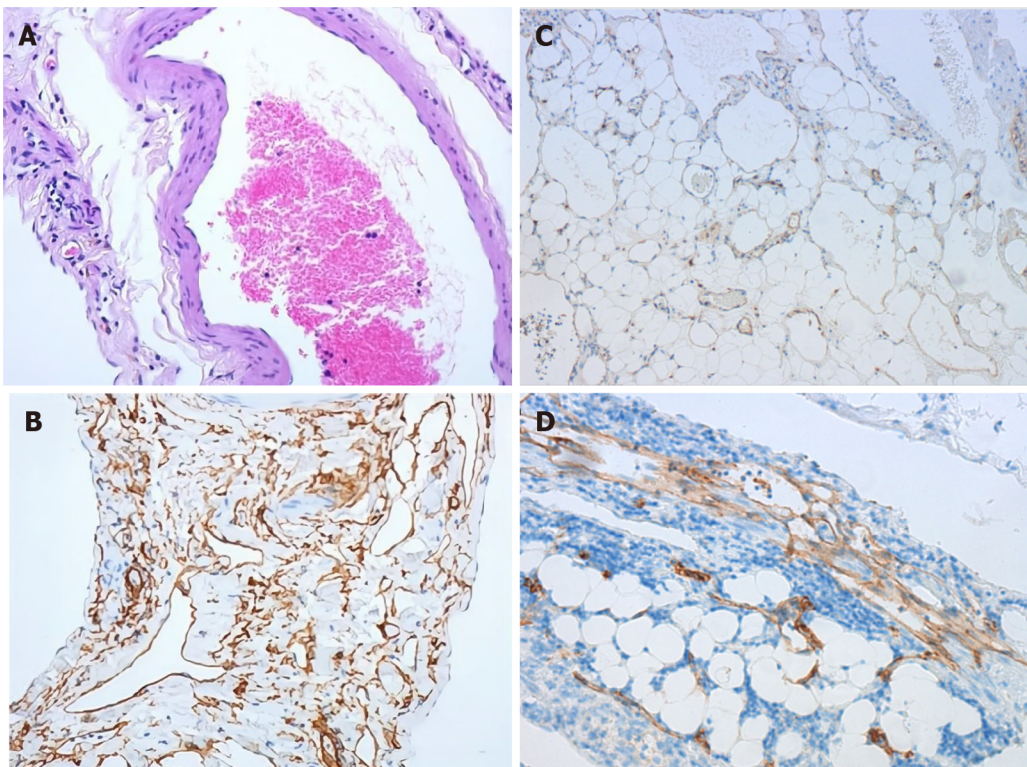
## DISCUSSION

Venous hemangioma is a benign and non-invasive type of tumor, and is rarely identified due to the absence of clinical manifestations. A retroperitoneal benign tumor is comparatively rare, and hemangiomas are exceptional. Most hemangiomas are present at birth, some regress spontaneously, and others proliferate during puberty or pregnancy or following local trauma[4]. Hemangiomas may occur



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**Figure 2** During operation, the cystic wall with duodenal adhesion was completely removed by laparotomy (as shown by the arrow).



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**Figure 3** Pathologic findings of the venous hemangioma. A: The specimen tissues are composed of various-sized cystic cavities with walls consisting of single-layered flattened cells; B: Immunohistochemically, the endothelial cells are positive for CD34; C: Some endothelial cells are positive for CD31; D: Some endothelial cells are positive for D2-40.

in any part of the body, but are extremely unusual in the retroperitoneum[5]. Almost all retroperitoneal hemangiomas are cavernous hemangiomas, and venous hemangioma is very rare[6].

A search was conducted in the PubMed database using the term "retroperitoneal hemangioma." The clinical characteristics of the disease in the relevant studies are shown in Table 1. Abdominal distension and pain were the two most common manifestations. In asymptomatic patients, the retroperitoneal mass was usually an incidental finding during a medical examination or emergent spontaneous bleeding. Tumors were removed entirely by means of laparoscopy or laparotomy in 13 patients who all had an uneventful recovery. However, subtotal resection was performed in one patient due to technical difficulties. Follow-up MRI seven months after surgery showed a reduction in size and no signs of recurrence.

The manifestations of retroperitoneal hemangioma are non-specific, and include abdominal pain, heartburn, dyspepsia, back pain, epigastralgia, as well as abdominal distension[4,5,7-9]. Our patient was found to have a retroperitoneal mass without symptoms during a medical examination. Routine preoperative laboratory examinations were made, including blood, urine and serum amylase tests, and

Table 1 Clinical characteristics of included literature

Ref.	Age	Sex	Complaints	Location	Treatment	Pathological types	Outcome
Powis and Rushton[18]	26	F	Abdominal swelling	Right abdomen	Total resection	Venous hemangioma	Uneventful
Igarashi and Hanazaki [6]	28	M	Hemangioma recurrence	Left lumbar	Total resection	Venous hemangioma	Uneventful
Martin-Fernández <i>et al</i> [5]	39	NM	Heartburn and abdominal pain	Right abdomen	Total resection	Venous hemangioma	Uneventful
Pérez Martín <i>et al</i> [4]	26	F	Dyspepsia and back pain	Left lumbar	Laparoscopy	Venous hemangioma	Uneventful
Tseng <i>et al</i> [19]	61	F	NM	Retroperitoneum	Subtotal resection	Venous hemangioma	Smaller residual
Forbes[11]	75	M	Abdominal and back pain	Left retroperitoneum	Laparotomy	Cavernous hemangioma	Uneventful
Choi and Oh[14]	29	F	Incidentally found	Right abdomen	Laparoscopy	Cavernous hemangioma	Uneventful
Kobayashi <i>et al</i> [13]	54	M	Left flank pain	Left Retroperitoneum	Laparoscopy	Venous hemangioma	Recurrence
He <i>et al</i> [7]	38	M	Dull epigastralgia	Right upper quadrant	Laparotomy	Cavernous hemangioma	Uneventful
Nakatsuka <i>et al</i> [8]	65	F	Abdominal fullness	Right side of uterine	Total resection	Venous malformation	Uneventful
Hanaoka <i>et al</i> [10]	36	M	Right upper quadrant pain	Right retroperitoneum	Laparotomy	Cavernous hemangioma	Uneventful
Amati <i>et al</i> [20]	20	F	Abdominal distention and pain	Retroperitoneum	Laparotomy	Malformation	Uneventful
Chen <i>et al</i> [9]	46	F	Right waist pain	Near renal pelvis	Laparoscopy	Cavernous hemangioma	Uneventful
Laih <i>et al</i> [3]	57	F	Incidentally found	Right retroperitoneum	Laparotomy	Cavernous hemangiomas	Uneventful

NM: Not mentioned; F: Female; M: Male.

all were within the normal ranges, but tumor markers were not tested. We speculated that the mass was not discovered earlier due to the sizeable retroperitoneal space and slow development of the mass. Retroperitoneal hemangioma typically has no initial symptoms, and the appearance of clinical manifestations is usually caused by an increase in tumor size and its compression of other organs or spontaneous bleeding[10,11]. Due to the non-specific presentation and low incidence, an accurate diagnosis before surgery is extremely challenging.

Recurrence of hemangioma is very rare. Only one case has been reported previously[6], in a 28-year-old man who underwent resection of a left lumbar cavernous hemangioma. However, two years later, he was admitted to hospital with recurrence of the left lumbar hemangioma. Abdominal CT revealed a large retroperitoneal tumor with a papillary structure. The tumor was completely resected, and the pathologic diagnosis was retroperitoneal venous hemangioma. Moreover, no cases of malignancy arising from retroperitoneal hemangioma were reported. As a result, more attention should be paid to patients with recurrent hemangioma, and complete resection of the cyst wall of the tumor and postoperative follow-up are necessary.

There are many imaging findings are not specific, but can aid to categorize the lesion as probably benign. CT is the primary imaging method for diagnosing retroperitoneal hemangioma. Although it lacks the specificity of presentation, it is helpful in the initial identification of benign or malignant masses. Abdominal ultrasonography is also an important technique for differential diagnosis. The main imaging features of the disease in the relevant studies are shown in Table 2. Laih *et al*[3] reported a patient whose abdominal ultrasonography showed a heterogeneous hypoechoic mass in the right retroperitoneal cavity. Similarly, the present patient presented to our hospital with the abdominal ultrasonography result of a hypoechoic cystic mass in the right retroperitoneal cavity during a medical examination. Abdominal ultrasonography is conducive to differentiating solid and cystic masses. Furthermore, abdominal CT is a preliminary examination for hemangioma, which may reveal a hypodense mass without or with slight enhancement in the arterial phase[12,13]. For example, Choi and Oh[14] reported a patient in whom a mass with internal multiple small calcifications (phleboliths) was identified by abdominal CT, and the mass showed slightly increased enhancement. We speculated that this phenomenon was probably caused by internal phleboliths of the mass, which can occur with slight or marked enhancement. In our patient, abdominal CT revealed an enormous hypodense mass with a



Table 2 Imaging features of included literature

Ref.	Age	Sex	Location	Imaging studies
Powis and Rushton[18]	26	F	Right abdomen	Intravenous pyelography show a soft-tissue mass compressing and displacing the ureter
Igarashi and Hanazaki[6]	28	M	Left lumbar	CT show a papillary structured mass in the left peritoneum, and slight enhancement in the late phase of the enhanced CT. Ultrasonography show a hyperechoic mass lesion
Martin-Fernández[5]	39	NM	Right abdomen	Ultrasonography and CT show a solid-cystic mass with septations
Pérez Martín[4]	26	F	Left lumbar	Ultrasonography and CT show a retroperitoneal solid cystic polylobulated mass
Tseng <i>et al</i> [19]	61	F	Retroperitoneum	Ultrasonography and CT show heterogeneous mass
Forbes[11]	75	M	Left retroperitoneum	CT show left-sided retroperitoneal hemorrhage
Choi and Oh [14]	29	F	Right abdomen	Ultrasonography show a hypoechogenic mass, and CT show irregular shaped mass with internal multiple small calcifications (phleboliths)
Kobayashi <i>et al</i> [13]	54	M	Left Retroperitoneum	CT show a cystic mass with the thickened wall having a contrast enhancement
He <i>et al</i> [7]	38	M	Right upper quadrant	Ultrasonography showed a giant cystic mass, and CT show a low density mass with mild enhancement
Nakatsuka <i>et al</i> [8]	65	F	Right side of uterine	MRI show a cystic with high intensity on the T2-weighted image
Hanaoka <i>et al</i> [10]	36	M	Right retroperitoneum	CT show a tumor without marked contrast enhancement, MRI show the tumor with low intensity on the T1-weighted image and high intensity on the T2-weighted image
Amati <i>et al</i> [20]	20	F	retroperitoneum	CT show a low density mass with regular borders and contrast-enhanced septation
Chen <i>et al</i> [9]	46	F	Near renal pelvis	CT show an ill-defined soft tissue mass with unevenly enhanced and with calcified margins. MRI show significant and continuous uneven mass, limited diffusion weighted imaging (DWI), and high signal intensity on apparent diffusion coefficient (ADC) map
Laih <i>et al</i> [3]	57	F	Right retroperitoneum	Ultrasonography show a heterogeneous hypoechoic lesion, and CT show heterogeneous enhancement over the right retroperitoneum

CT: Computed tomography; MRI: Magnetic resonance imaging; NM: Not mentioned; F: Female; M: Male.

regular margin and without marked enhancement in the arterial phase of enhanced CT. In addition, MRI is an alternative technique, which is especially helpful in distinguishing the surrounding soft tissue structures. The mass was hypointense on the T1-weighted image, and was hyperintense on the T2-weighted image[10,15]. In our case, it was suggested that the patient should undergo further MRI examination, which revealed the retroperitoneal mass with low signal intensity on the T1-weighted image and high signal intensity on the T2-weighted image. However, accurate preoperative diagnosis of retroperitoneal venous hemangiomas is very difficult. Therefore, it is recommended that preoperative imaging be used to assess both the benignity and malignancy of the tumor and the location and proximity to organs and structures surrounding tissue structures.

Surgical resection is the mainstay of treatment for retroperitoneal venous hemangioma. Even though retroperitoneal hemangioma is a benign tumor, the enlarged tumor may compress the surrounding organs and cause rupture and bleeding. In our patient, a mass approximately 20 cm was found in the right retroperitoneum following laparoscopic exploration. The tumor compressed the adjacent tissues and organs, and most importantly, the mass was firmly attached to both the wall of the second part of the duodenum and the head of the pancreas. Therefore, in order to completely remove the capsule wall of the mass and protect the surrounding organs, we chose to resect the tumor by laparotomy. Consequently, the patient had a successful postoperative recovery and follow-up was uneventful.

The pathological features of a venous hemangioma consist of positivity for the vascular markers CD34 and CD31, and the vascular wall contains smooth muscle and is lined with a single layer of endothelial cells[4,16,17]. In our case, microscopic examination showed that the cystic wall was lined by a single layer of flattened cells, smooth muscle cells, as well as adipose tissue. Immunohistochemistry was positive for SMA, CD34 and CD31. These findings were consistent with a typical venous hemangioma.

Although imaging has provided assistance in the diagnosis of retroperitoneal cysts, we did not focus on adhesion of the mass to the surrounding organs. Moreover, sufficient evidence to demonstrate the superiority of laparoscopy for the treatment of retroperitoneal venous hemangioma is lacking. It is suggested that the appropriate procedure should be chosen according to the patient's condition before surgery.

## CONCLUSION

Venous hemangioma is a rare benign lesion in adults, and an accurate diagnosis before surgery is challenging due to non-specific manifestations, imaging features and low incidence. Retroperitoneal venous hemangiomas essentially involve no malignancy, but venous hemangiomas may grow and cause symptoms of compression as well as adhering to surrounding tissues. When symptomatic massive hemangiomas are present, surgery is considered. In addition, surgery is the curative treatment for venous hemangioma, and the definitive diagnosis relies on pathology. Attention should be paid to lesion residues after resection, and CT should be reviewed periodically during the follow-up period.

## FOOTNOTES

**Author contributions:** Qin Y and Qiao P participated in the management of this case, collected the material of this case, and drafted the manuscript; Guan X and Zeng S contributed to the conception and design of the article; Hu XP and Wang B participated in the management of this case and were in charge of revising the manuscript; all authors read and approved the final manuscript.

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