

# Retroperitoneal ovarian venous hemangioma: A case report

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**Abstract.** Retroperitoneal venous hemangioma is a rare vascular malformation. The present study reports a unique case of a 68-year-old female with a retroperitoneal ovarian venous hemangioma that was surgically resected. Computed tomography and magnetic resonance imaging showed a retroperitoneal mass anterior to the left kidney. Enhanced imaging revealed a close relationship between the mass and surrounding vessels, suggesting a retroperitoneal vascular neoplasm. Histopathological examination of the tissue after complete surgical resection confirmed the diagnosis of a retroperitoneal ovarian venous hemangioma. This case provides crucial insights for clinicians to enhance their understanding of ovarian venous hemangioma through the integration of multimodal imaging characteristics and pathological evidence, thereby contributing to the reduction of misdiagnosis in clinical practice.

## Introduction

Retroperitoneal venous hemangioma is a rare disease and involvement of the ovaries is rarer. To date, no cases involving the ovaries have been reported in the literature, to the best of our knowledge. The disease usually has a subtle onset, with non-specific clinical symptoms and imaging findings, making it difficult to diagnose without a pathological specimen (1). Reports on retroperitoneal hemangiomas show that most are cavernous hemangiomas (2). On imaging, they typically appear with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images (3). They also tend to be closely attached to surrounding muscles, occasionally causing local muscle signal disruption (2,4). These imaging characteristics are important for diagnosing the disease. The present case, through the comprehensive

multimodal imaging techniques including CT and MRI, provides a detailed analysis of the lesion's morphological characteristics, vascular supply features, and its relationship with surrounding anatomical structures. Combined with postoperative histopathological examination and immunohistochemical staining, a final diagnosis of a rare ovarian-origin retroperitoneal venous hemangioma was established. The present study provides a valuable reference for ovarian venous hemangiomas to reduce misdiagnosis.

## Case report

A 68-year-old female patient presented to Xiangyang No. 1 People's Hospital (Xiangyang, China) in June 2024 with a three-month history of persistent dull abdominal pain. The pain was mainly located around the umbilicus, with a more pronounced left-sided component; it was not accompanied by nausea, vomiting, diarrhea, constipation or hematochezia. There was no obvious trigger for the pain. The patient's overall condition was good, with a history of breast cancer surgery and no family history of genetic diseases. Physical examination revealed a soft abdomen with tenderness on the left side and a palpable mass in the left lower quadrant. The initial outpatient diagnosis was 'abdominal mass', and the patient was admitted for further evaluation and treatment. Laboratory tests showed no abnormalities and hormone levels were within the normal range. Computed tomography (CT) revealed a heterogeneous, enhancing retroperitoneal mass in the left retroperitoneal space, adjacent to the spine and anterior to the left kidney. The tumor extended upward to the level of the left renal vein and downward to the lower pole of the left kidney (Fig. 1A). The lesion measured ~29x34x58 mm and had an uneven density. On contrast-enhanced scans, the tumor showed a progressive enhancement pattern in the arterial, venous and delayed phases (Fig. 1B-D). Coronal, sagittal and three-dimensional CT reconstructions showed that the tumor was connected to blood vessels at both poles (Fig. 1E-H). Magnetic resonance imaging (MRI) showed heterogeneous high signal intensity on T2-weighted images and low signal intensity on T1-weighted images, with significant enhancement after contrast administration (Fig. 2A-D). Diffusion-weighted imaging showed slightly higher signal intensity (Fig. 2E and F). No evidence of fat components was found on in-phase and out-of-phase imaging (Fig. 2G and H). Based on these imaging findings, the lesion was highly suspected to be of vascular origin. The

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patient subsequently underwent surgery in July 2024, during which the tumor was found to be connected to the left ovarian vein and to drain into the left renal vein (Fig. 3A). The tumor appeared grayish-red and irregular, measuring 50x20x20 mm, with a partial capsule. The tumor section was grayish-white and grayish-red, soft and solid, with some areas appearing mucoid and gelatinous (data not shown). Specimens were fixed in 10% neutral buffered formalin at room temperature for 24-48 h, embedded in paraffin, and sectioned at 4-5  $\mu$ m. Mayer's hematoxylin was applied at room temperature for 5-8 min, followed by alcoholic eosin at room temperature for 30-60 sec. For immunohistochemistry, sections were deparaffinized in xylene, rehydrated through a descending alcohol series, and subjected to heat-induced antigen retrieval in Tris-EDTA buffer (pH 9.0) at 95-100°C for 20 min. Endogenous peroxidase activity was blocked with 3% hydrogen peroxide at room temperature for 10-15 min, and nonspecific binding was blocked with 5% normal goat serum (Leica Biosystems) at room temperature for 1 h. Sections were then incubated with specific primary antibodies from Leica Biosystems (1:200) at room temperature for 1 h or overnight at 4°C. Secondary antibody incubation was performed using the Leica Bond Polymer Refine Detection System (cat. no. DS9800), an HRP-conjugated polymer, at room temperature for 30 min. DAB was used as the chromogen, and sections were counterstained with hematoxylin at room temperature for 30-60 sec. All sections were observed under a light microscope. Histopathological and immunohistochemical analysis indicated the following: Vimentin (+), CD31 (+), CD34 (+), ETS-related gene (ERG) (+), Friend leukemia integration 1 transcription factor (Fli-1) (+),  $\alpha$ -inhibin (partially weak +), D2-40 (podoplanin) (-), smooth muscle actin (SMA) (+), STAT6 (cytoplasmic +), S-100 protein (scattered +), CD56 (-), pan-cytokeratin (PCK) (-), synaptophysin (Syn) (-), chromogranin A (CgA) (-), GATA binding protein 3 (GATA-3) (-), Melan-A (weak +), human melanoma black 45 (HMB45) (-), Desmin (+), Ki-67 (hot spots ~20%); rare mitotic figures were observed, with a count of 0-1 per 10 high-power fields (Fig. 3B-D). The endothelial cells were strongly stained for the CD31, CD34 (Fig. 3C and D) and ERG markers (Fig. 4B), confirming the characteristic of vascular lesions (5-7). Based on the immunohistochemical findings (Fig. 4A-O), the final diagnosis was ovarian venous hemangioma. Post-surgical treatment, the patient achieved excellent therapeutic outcomes with an excellent prognosis. After one year of follow-up, the patient reported no discomfort and imaging studies showed no recurrence.

## Discussion

Vascular anomalies encompass a variety of diseases. According to the classification of the International Society for the Study of Vascular Anomalies (ISSVA), they are divided into two primary types: Vascular tumors, characterized by proliferative endothelial changes, and vascular malformations, resulting from structural anomalies of blood vessels. Vascular malformations are further classified based on the predominant type of anomalous vessel involved, including capillary malformations, lymphatic malformations, venous malformations and arteriovenous malformations. Venous malformations are historically known as 'cavernous hemangiomas' (8). Prior

to the establishment of the ISSVA classification system in 1996, the terminology used to describe vascular lesions was inconsistent and imprecise, with the term 'hemangioma' being widely used to describe various vascular lesions (9). Even after the widespread adoption of the ISSVA classification, studies indicate that traditional terminology persists in both the literature and clinical practice. A systematic evaluation by Steele *et al* (10) from 2019 of 195 articles published in 2015 and 2016 found that 64.6% of publications used incorrect terminology for vascular anomalies according to the 2014 ISSVA classification, and this figure remained at 60.5% when using the 2018 ISSVA classification. Boulogeorgou *et al* (11) noted that despite the latest ISSVA update in 2018, traditional terminology remains prevalent in clinical and pathological practice, which explains the use of the conventional diagnosis in the pathological report of this case.

The majority of reported vascular tumors are categorized as benign cavernous hemangiomas (5). Primary retroperitoneal ovarian venous hemangioma is a rare disease with atypical clinical features. Its imaging characteristics differ from those of previously reported retroperitoneal venous hemangiomas. Previous reports of retroperitoneal hemangiomas have mainly described their distribution adjacent to the aorta (12,13), paraspinal region (14), perirenal tissue (15,16), adrenal glands (17-20) and the intestines (21). The number of reported cases is ~30 (5,22). The case of the current study represents the first reported case of an ovarian venous hemangioma, to the best of our knowledge. The diagnosis was established based on critical surgical and pathological correlations: The tumor was contiguous with the wall of the left ovarian vein and drained into the renal venous system via the same vessel. This anatomical relationship strongly supports an origin from the vascular components of the ovarian vein wall itself, classifying it as a primary venous vascular neoplasm. Its growth pattern was characterized by localized, expansile growth along the venous axis, distinct from a tumor arising from the ovarian parenchyma with secondary vascular invasion or metastasis. This represents the first reported case of a hemangioma unequivocally originating from the ovarian vein, with unique clinicopathological and anatomical significance.

Enhanced CT and MRI scans are key for preoperative diagnosis. The most prominent feature is an enhancing retroperitoneal mass directly connected to the ovarian vein, with progressive enhancement on contrast-enhanced scans. It needs to be distinguished from other vascular diseases, such as vascular sarcoma and vascular endothelial sarcomas (5). Histopathological examination and immunohistochemical analysis are key for definitive diagnosis and classification (3). Surgical resection remains the primary treatment, particularly for symptomatic patients or cases suspected of being malignant. Most patients achieve clinical cure following surgical intervention (23). Meissner *et al* (24) proposed the Symptoms-Varices-Pathophysiology classification for pelvic venous disorders, noting that in patients with left ovarian vein incompetence, the left renal hilum serves as an important venous reservoir. In the present case, the left ovarian vein was notably dilated, raising the possibility that the retroperitoneal mass may represent secondary collateral pathways resulting from ovarian vein incompetence. However, based on intraoperative findings-the tumor was contiguous with the ovarian

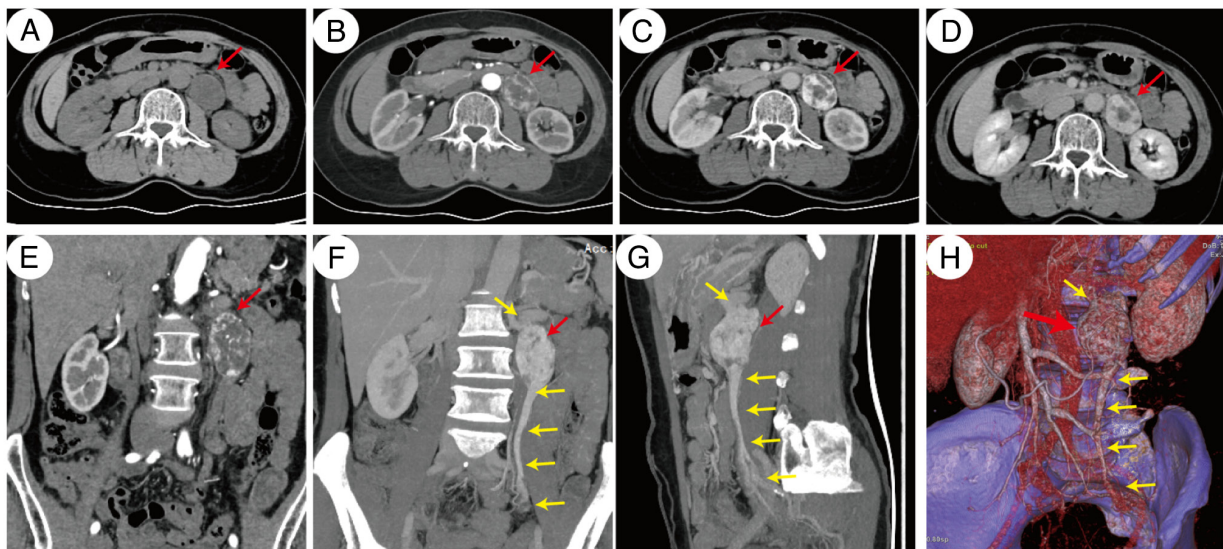


Figure 1. CT images of the retroperitoneal mass. (A) Plain scan. (B) Arterial phase. (C) Venous phase. (D) Delayed phase. (E) CT enhanced coronal image shows the range of the mass and its relationship with adjacent structures (red arrow). (F) CT enhanced coronal image showing the association between the mass (red arrow) and the ovarian vein (yellow arrow). (G) Sagittal maximum intensity projection image showing the association between the mass (red arrow) and the ovarian vein (yellow arrow). (H) 3D reconstructed image shows the mass (red arrow) connecting the left ovarian vein (yellow arrow) at its upper and lower poles, joining the left renal vein.

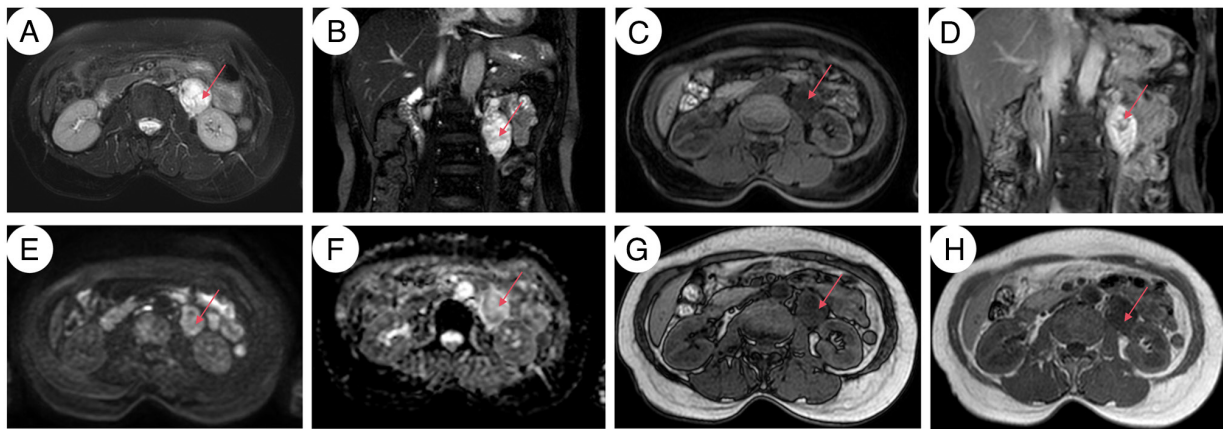


Figure 2. MRI of the retroperitoneal mass. (A) MRI T2 transverse. (B) MRI coronal images show the signal characteristics of the mass (red arrow) and adjacent structures. (C) MRI T1 transverse image. (D) MRI enhanced coronal image shows significant non-uniform enhancement. (E) Diffusion-weighted imaging demonstrated a mild high signal. (F) Apparent Diffusion Coefficient: Mild low signal, indicating mild diffusion restriction. (G) Out-of-phase: No signal drop vs. in-phase. (H) In-phase: Anatomical reference indicates no fat component.

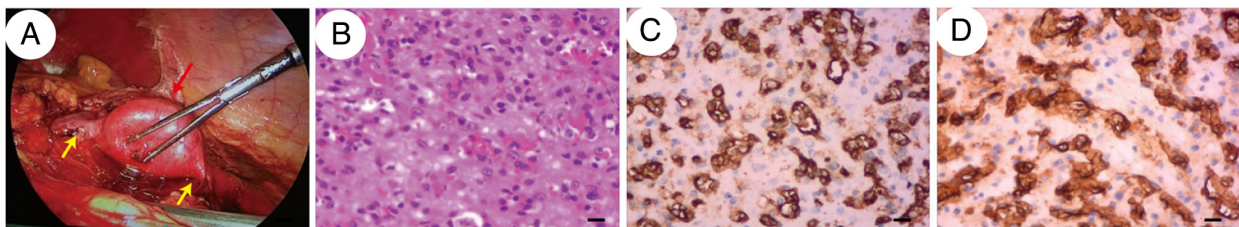


Figure 3. Intraoperative and pathological features of the retroperitoneal mass. (A) Intraoperative gross anatomy shows the ovarian vein (yellow arrow) connecting the mass (red arrow) and joining the left renal vein. (B) Hematoxylin and eosin staining revealed thick-walled vascular channels and disorganized smooth muscle bundles. (C) Immunohistochemical staining shows positive CD31 staining [(SH28.1) scale bar=50 μm; magnification, x200]. (D) Immunohistochemical staining shows positive CD34 staining [(SH28.1) scale bar=50 μm; magnification, x200].

vein wall and demonstrated expansile growth along the venous axis- and histopathological confirmation of thick-walled vascular channels with smooth muscle (features typical of a

primary venous malformation rather than a simple varicosity), it is likely that the lesion is more consistent with a primary venous malformation arising from the vein wall.

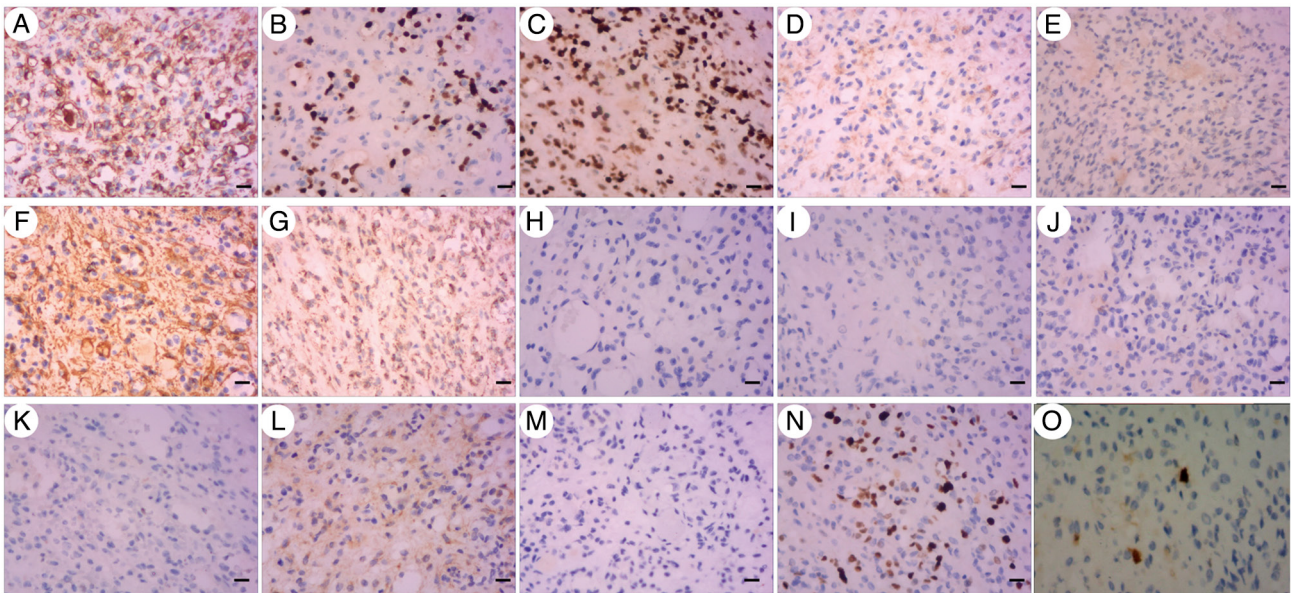


Figure 4. Pathological immunohistochemical images. Immunohistochemical staining for (A) Vimentin, (B) ETS-related gene), (C) Fli-1 (Friend leukemia virus integration site 1), (D)  $\alpha$ -inhibin, (E) D2-40 (podoplanin), (F) SMA (smooth muscle actin), (G) STAT6, (H) PCK (pan-cytokeratin), (I) Syn (synaptophysin), (J) CgA (chromogranin A), (K) GATA binding protein 3), (L) Melan-A, (M) Desmin and (N) Ki-67 (magnification, x200). (O) S-100 (scale bar=50  $\mu$ m; magnification, x40)

The final diagnosis in this case was ‘ovarian venous hemangioma’. This diagnosis was established based on characteristic histological morphology (thick-walled vascular channels) and an immunohistochemical profile (positive for CD31, CD34 and ERG), supporting its vascular origin. First, STAT6 showed cytoplasmic positivity with nuclear negativity, which is distinct from the characteristic nuclear-positive pattern of solitary fibrous tumor (SFT). Therefore, the observed cytoplasmic staining was considered nonspecific, effectively ruling out retroperitoneal SFT (25). Second, although the Ki-67 proliferation index reached up to 20% in localized ‘hotspot’ areas, the positive signals were predominantly confined to vascular endothelial cells and the mitotic count was very low throughout the specimen (0-1 per 10 high-power fields). Combined with the clear tumor boundaries, lack of significant cellular atypia and absence of multilayered endothelial cell arrangements, these features are more consistent with a cellular benign hemangioma and do not support a diagnosis of low-grade angiosarcoma. Finally, the focal weak positivity for  $\alpha$ -inhibin is a noteworthy but nonspecific finding. This marker is typically associated with sex cord-stromal tumors but is also known to be expressed in various mesenchymal cells, including pericytes (26). In this case, its expression may reflect activated myofibroblasts or pericytic components within the tumor stroma. Given the extensive positivity for vascular endothelial markers and the clinical context, this finding does not support a diagnosis of a sex cord-stromal tumor or other  $\alpha$ -inhibin-specific neoplasms.

Here, the patient presented with a chronic clinical course and a mass-like lesion in front of the left kidney. The initial diagnosis by the radiologist considered the possibility of a renal tumor. Subsequently, based on the analysis of enhanced imaging features, the lesion was highly suspected to be of vascular origin. The patient then underwent surgical treatment, during which the lesion was found to be directly connected

to the left ovarian vein. The final diagnosis, confirmed by pathological analysis using immunohistochemistry, was a hemangioma originating from the ovarian vein. In this study, a discrepancy was observed between the preoperative CT and postoperative gross specimen measurements in three dimensions. The CT measurements reflected the tumor in its *in vivo*, distended state, where the imaging boundaries might include peritumoral edematous or inflammatory zones, and the tumor shape could be influenced by compression from surrounding organs. By contrast, the surgical specimen, after excision, undergoes changes in morphology and volume reduction due to interruption of blood flow, tissue collapse and formalin fixation. Pathological measurements also tend to focus more on the solid tumor region. These factors are the primary reasons for the dimensional discrepancies (27). However, the reported maximum tumor diameter (CT: 58 mm; pathology: 50 mm) was consistent between the two modalities. This confirms the reliability of imaging assessment for tumor size, with the main differences attributed to morphological changes in non-principal dimensions post-excision.

In conclusion, ovarian vein hemangioma behind the peritoneum is a rare condition and its diagnosis can be highly challenging. Multimodal imaging, particularly enhanced scanning, is crucial for the diagnosis of this condition. This case will help radiologists and clinicians increase their awareness of the disease and reduce the likelihood of misdiagnosis.

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### Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

### Authors' contributions

JL conceived and designed the study. JT and SC analyzed and interpreted data. All authors read and approved the final manuscript. JT and JL confirm the authenticity of all the raw data. JT, HH, and SC performed the literature review.

### Ethics approval and consent to participate

Written informed consent was obtained from the patient for participation in the present study. The present study was conducted in accordance with the World Medical Association's Declaration of Helsinki (2013 revision).

### Patient consent for publication

The patient provided written informed consent for the publication of the present case report, including clinical information and images.

### Competing interests

The authors declare that they have no competing interests.

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