

World Journal of *Clinical Cases*

World J Clin Cases 2020 October 26; 8(20): 4688-5069



MINIREVIEWS

- 4688 Relationship between non-alcoholic fatty liver disease and coronary heart disease
Arslan U, Yenercağ M

ORIGINAL ARTICLE

Retrospective Cohort Study

- 4700 Remission of hepatotoxicity in chronic pulmonary aspergillosis patients after lowering trough concentration of voriconazole
Teng GJ, Bai XR, Zhang L, Liu HJ, Nie XH

Retrospective Study

- 4708 Endoscopic submucosal dissection as alternative to surgery for complicated gastric heterotopic pancreas
Noh JH, Kim DH, Kim SW, Park YS, Na HK, Ahn JY, Jung KW, Lee JH, Choi KD, Song HJ, Lee GH, Jung HY
- 4719 Observation of the effects of three methods for reducing perineal swelling in children with developmental hip dislocation
Wang L, Wang N, He M, Liu H, Wang XQ
- 4726 Predictive value of serum cystatin C for risk of mortality in severe and critically ill patients with COVID-19
Li Y, Yang S, Peng D, Zhu HM, Li BY, Yang X, Sun XL, Zhang M
- 4735 Sleep quality of patients with postoperative glioma at home
Huang Y, Jiang ZJ, Deng J, Qi YJ
- 4743 Early complications of preoperative external traction fixation in the staged treatment of tibial fractures: A series of 402 cases
Yang JZ, Zhu WB, Li LB, Dong QR
- 4753 Retroperitoneal vs transperitoneal laparoscopic lithotripsy of 20-40 mm renal stones within horseshoe kidneys
Chen X, Wang Y, Gao L, Song J, Wang JY, Wang DD, Ma JX, Zhang ZQ, Bi LK, Xie DD, Yu DX
- 4763 Undifferentiated embryonal sarcoma of the liver: Clinical characteristics and outcomes
Zhang C, Jia CJ, Xu C, Sheng QJ, Dou XG, Ding Y
- 4773 Cerebral infarct secondary to traumatic internal carotid artery dissection
Wang GM, Xue H, Guo ZJ, Yu JL
- 4785 Home-based nursing for improvement of quality of life and depression in patients with postpartum depression
Zhuang CY, Lin SY, Cheng CJ, Chen XJ, Shi HL, Sun H, Zhang HY, Fu MA

Observational Study

- 4793** Cost-effectiveness of lutetium (^{177}Lu) oxodotreotide *vs* everolimus in gastroenteropancreatic neuroendocrine tumors in Norway and Sweden
Palmer J, Leeuwenkamp OR
- 4807** Factors related to improved American Spinal Injury Association grade of acute traumatic spinal cord injury
Tian C, Lv Y, Li S, Wang DD, Bai Y, Zhou F, Ma QB
- 4816** Intraoperative systemic vascular resistance is associated with postoperative nausea and vomiting after laparoscopic hysterectomy
Qu MD, Zhang MY, Wang GM, Wang Z, Wang X

META-ANALYSIS

- 4826** Underwater *vs* conventional endoscopic mucosal resection in treatment of colorectal polyps: A meta-analysis
Ni DQ, Lu YP, Liu XQ, Gao LY, Huang X

CASE REPORT

- 4838** Dehydrated patient without clinically evident cause: A case report
Palladino F, Fedele MC, Casertano M, Liguori L, Esposito T, Guarino S, Miraglia del Giudice E, Marzuillo P
- 4844** Intracranial malignant solitary fibrous tumor metastasized to the chest wall: A case report and review of literature
Usuda D, Yamada S, Izumida T, Sangen R, Higashikawa T, Nakagawa K, Iguchi M, Kasamaki Y
- 4853** End-of-life home care of an interstitial pneumonia patient supported by high-flow nasal cannula therapy: A case report
Goda K, Kenzaka T, Kuriyama K, Hoshijima M, Akita H
- 4858** Rupture of carotid artery pseudoaneurysm in the modern era of definitive chemoradiation for head and neck cancer: Two case reports
Kim M, Hong JH, Park SK, Kim SJ, Lee JH, Byun J, Ko YH
- 4866** Unremitting diarrhoea in a girl diagnosed anti-N-methyl-D-aspartate-receptor encephalitis: A case report
Onpoaree N, Veeravigrom M, Sanpavat A, Suratannon N, Sintusek P
- 4876** Paliperidone palmitate-induced facial angioedema: A case report
Srifuengfung M, Sukakul T, Liangcheep C, Viravan N
- 4883** Improvement of lenvatinib-induced nephrotic syndrome after adaptation to sorafenib in thyroid cancer: A case report
Yang CH, Chen KT, Lin YS, Hsu CY, Ou YC, Tung MC
- 4895** Adult metaplastic hutch diverticulum with robotic-assisted diverticulectomy and reconstruction: A case report
Yang CH, Lin YS, Ou YC, Weng WC, Huang LH, Lu CH, Hsu CY, Tung MC

- 4902** Thrombus straddling a patent foramen ovale and pulmonary embolism: A case report
Huang YX, Chen Y, Cao Y, Qiu YG, Zheng JY, Li TC
- 4908** Therapeutic experience of an 89-year-old high-risk patient with incarcerated cholecystolithiasis: A case report and literature review
Zhang ZM, Zhang C, Liu Z, Liu LM, Zhu MW, Zhao Y, Wan BJ, Deng H, Yang HY, Liao JH, Zhu HY, Wen X, Liu LL, Wang M, Ma XT, Zhang MM, Liu JJ, Liu TT, Huang NN, Yuan PY, Gao YJ, Zhao J, Guo XA, Liao F, Li FY, Wang XT, Yuan RJ, Wu F
- 4917** Woven coronary artery: A case report
Wei W, Zhang Q, Gao LM
- 4922** Idiopathic multicentric Castleman disease with pulmonary and cutaneous lesions treated with tocilizumab: A case report
Han PY, Chi HH, Su YT
- 4930** Perianorectal abscesses and fistula due to ingested jujube pit in infant: Two case reports
Liu YH, Lv ZB, Liu JB, Sheng QF
- 4938** Forniceal deep brain stimulation in severe Alzheimer's disease: A case report
Lin W, Bao WQ, Ge JJ, Yang LK, Ling ZP, Xu X, Jiang JH, Zuo CT, Wang YH
- 4946** Systemic autoimmune abnormalities complicated by cytomegalovirus-induced hemophagocytic lymphohistiocytosis: A case report
Miao SX, Wu ZQ, Xu HG
- 4953** Nasal mucosa pyoderma vegetans associated with ulcerative colitis: A case report
Yu SX, Cheng XK, Li B, Hao JH
- 4958** Amiodarone-induced hepatotoxicity — quantitative measurement of iodine density in the liver using dual-energy computed tomography: Three case reports
Lv HJ, Zhao HW
- 4966** Multisystem involvement Langerhans cell histiocytosis in an adult: A case report
Wang BB, Ye JR, Li YL, Jin Y, Chen ZW, Li JM, Li YP
- 4975** New mutation in EPCAM for congenital tufting enteropathy: A case report
Zhou YQ, Wu GS, Kong YM, Zhang XY, Wang CL
- 4981** Catastrophic vertebral artery and subclavian artery pseudoaneurysms caused by a fishbone: A case report
Huang W, Zhang GQ, Wu JJ, Li B, Han SG, Chao M, Jin K
- 4986** Anastomosing hemangioma arising from the left renal vein: A case report
Zheng LP, Shen WA, Wang CH, Hu CD, Chen XJ, Shen YY, Wang J
- 4993** Bladder perforation caused by long-term catheterization misdiagnosed as digestive tract perforation: A case report
Wu B, Wang J, Chen XJ, Zhou ZC, Zhu MY, Shen YY, Zhong ZX

- 4999** Primary pulmonary plasmacytoma accompanied by overlap syndrome: A case report and review of the literature
Zhou Y, Wang XH, Meng SS, Wang HC, Li YX, Xu R, Lin XH
- 5007** Gastrointestinal stromal tumor metastasis at the site of a totally implantable venous access port insertion: A rare case report
Yin XN, Yin Y, Wang J, Shen CY, Chen X, Zhao Z, Cai ZL, Zhang B
- 5013** Massive gastrointestinal bleeding caused by a Dieulafoy's lesion in a duodenal diverticulum: A case report
He ZW, Zhong L, Xu H, Shi H, Wang YM, Liu XC
- 5019** Plastic bronchitis associated with *Botrytis cinerea* infection in a child: A case report
Liu YR, Ai T
- 5025** Chest, pericardium, abdomen, and thigh penetrating injury by a steel rebar: A case report
Yang XW, Wang WT
- 5030** Monocular posterior scleritis presenting as acute conjunctivitis: A case report
Li YZ, Qin XH, Lu JM, Wang YP
- 5036** Choriocarcinoma with lumbar muscle metastases: A case report
Pang L, Ma XX
- 5042** Primary chondrosarcoma of the liver: A case report
Liu ZY, Jin XM, Yan GH, Jin GY
- 5049** Successful management of a tooth with endodontic-periodontal lesion: A case report
Alshawwa H, Wang JF, Liu M, Sun SF
- 5057** Rare imaging findings of hypersensitivity pneumonitis: A case report
Wang HJ, Chen XJ, Fan LX, Qi QL, Chen QZ
- 5062** Effective administration of cranial drilling therapy in the treatment of fourth degree temporal, facial and upper limb burns at high altitude: A case report
Shen CM, Li Y, Liu Z, Qi YZ

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Anastomosing hemangioma arising from the left renal vein: A case report

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Abstract

BACKGROUND

Anastomosing hemangioma (AH) is a rare subtype of benign hemangioma that is most commonly found in the genitourinary tract. Due to the lack of specific clinical and radiologic manifestations, it is easily misdiagnosed preoperatively. Here, we report a case of AH arising from the left renal vein that was discovered incidentally and confirmed pathologically, and then describe its imaging characteristics from a radiologic point of view and review its clinicopathologic features and treatment.

CASE SUMMARY

A 74-year-old woman was admitted to our department for a left retroperitoneal neoplasm measuring 2.6 cm × 2.0 cm. Her laboratory data showed no significant abnormalities. A non-contrast-enhanced computed tomography (CT) scan showed a heterogeneous density in the neoplasm. Non-contrast-enhanced magnetic resonance imaging (MRI) revealed a heterogeneous hypointensity on T1-weighted images and a heterogeneous hyperintensity on T2-weighted images. On contrast-enhanced CT and MRI scans, the neoplasm presented marked septal enhancement in the arterial phase and persistent enhancement in the portal phase, and its boundary with the left renal vein was ill-defined. Based on these clinical and radiological manifestations, the neoplasm was initially considered to be a neurogenic neoplasm in the left retroperitoneum. Finally, the neoplasm was completely resected and pathologically diagnosed as AH.

CONCLUSION

AH is an uncommon benign hemangioma. Preoperative misdiagnoses are

Checklist (2016).

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common not only because of a lack of specific clinical and radiologic manifestations but also because clinicians lack vigilance and diagnostic experience in identifying AH. AH is not exclusive to the urogenital parenchyma. We report the first case of this neoplasm in the left renal vein. Recognition of this entity in the left renal vein can be helpful in its diagnosis and distinction from other neoplasms.

Key Words: Anastomosing hemangioma; Angiosarcoma; Computed tomography; Magnetic resonance imaging; Case report; Pathology

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Core Tip: Anastomosing hemangioma (AH) is difficult to recognize because of its rarity and atypia. Herein, we report a case of AH in an old woman that was discovered incidentally and confirmed pathologically. To the best of our knowledge, this is the first case of AH reported in the left renal vein. Recognizing that AH may occur in the left renal vein will help improve doctors' vigilance and reduce the probability of misdiagnosis.

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INTRODUCTION

Hemangiomas are most common in the skin or subcutaneous soft tissue, while few occur in the liver, kidney, and other parenchymal organs. Morphologically, most are classic capillary hemangiomas or venous hemangiomas. In 2009, Montgomery and Epstein^[1] reported a unique type of capillary hemangioma occurring in the kidney and testis. Its morphology consists of an ethmoid, sinusoid, anastomosing vascular pattern lined with hobnailed endothelial cells, which is similar to the red pulp of the spleen, and the mass was first named "anastomosing hemangioma" (AH). Subsequently, a few cases of AH were reported in the liver, gastrointestinal tract, retroperitoneum, ovary, bladder, adrenal gland, nasal cavity, and intracranial space^[2-9].

Here we report the case of a 74-year-old woman who presented without any symptoms and was accidentally found to have a left retroperitoneal neoplasm by imaging examination. After surgical resection, the neoplasm was pathologically diagnosed as AH.

CASE PRESENTATION

Chief complaints

A 74-year-old woman was admitted to our department for a left retroperitoneal neoplasm that was found 1 mo prior during a routine checkup.

History of present illness

One month prior, abdominal non-contrast-enhanced computed tomography (CT) at a local hospital revealed a left retroperitoneal neoplasm. However, the patient had no symptoms.

History of past illness

The patient had a history of hypertension for 20 years and had been taking 0.15 g irbesartan once a day to control blood pressure to within the normal range. In addition, the patient had a history of auricular fibrillation for 1 mo and had been taking 0.1 g aspirin each night for anticoagulant therapy and 47.5 milligrams succinic

metoprolol once a day to control the heart rate to approximately 90 beats/min.

Physical examination

The patient had no superficial lymph node enlargement. An abdominal physical examination showed that her abdomen was flat and soft without tenderness.

Laboratory examinations

Routine blood tests, biochemical function, coagulation function, and tumor markers, as well as the levels of cortisol (19.13 µg/dL, reference range 6.24-18 µg/dL), aldosterone (105.04 ng/L, reference range 50-313 ng/L), the three hypertension items [plasma renin activity, 1.03 µg/L/h, reference range 1.45-5 µg/L/h; angiotensin I, 0.96 µg/L; angiotensin II, 50.19 ng/L, reference range 32-90 ng/L] were all within normal ranges.

Imaging examinations

A CT scan of the abdomen was performed in our hospital, on which the neoplasm presented as a circular heterogeneous hypodense soft-tissue shadow (Figure 1A). On contrast-enhanced CT, the neoplasm presented heterogeneous septal enhancement in the arterial phase (Figure 1B) and persistent enhancement in the portal phase, and its boundary with the left renal vein was ill-defined (Figure 1C).

To further clarify the diagnosis, we also performed a contrast-enhanced magnetic resonance imaging (MRI) scan. On T1-weighted images, the neoplasm showed a circular heterogeneous hypointensity (Figure 2A). On T2-weighted images and diffusion-weighted images, the neoplasm showed a circular heterogeneous hyperintensity (Figure 2B and C). On arterial phase post-contrast T1-weighted images, the neoplasm showed obvious septal enhancement (Figure 2D). On axial and coronal portal venous phase post-contrast T1-weighted images, the neoplasm showed persistent enhancement and its boundary with the left renal vein was ill-defined (Figure 2E and F).

FINAL DIAGNOSIS

Based on these clinical and radiological manifestations, the neoplasm was initially diagnosed as a neurogenic neoplasm of the left retroperitoneum.

TREATMENT

The patient planned to undergo laparoscopic resection of the left retroperitoneal neoplasm, but due to the ill-defined boundary between the neoplasm and the left renal vein, massive bleeding occurred during the surgical separation, so the patient was transferred to open surgery. Finally, the neoplasm was completely resected in the case of renal vascular occlusion, and we reconstructed the left renal vein.

OUTCOME AND FOLLOW-UP

The patient recovered smoothly without any complications. *In situ*, the neoplasm was located in front of the left renal vein, and the boundary between them was ill-defined (Figure 3A). Macroscopically, the neoplasm presented as a mahogany brown lesion, with no capsule (Figure 3B). Microscopically, the neoplasm was composed of a clear ethmoid, sinusoid, and anastomosing vascular pattern, which was separated by fibers into lobules and had the appearance of the red pulp of the spleen. The vascular endothelial cells of the neoplasm looked like a hobnail, and the local vascular endothelial nuclei were enlarged, hyperchromatic, and slightly abnormal, but no mitotic figure was seen (Figure 3C and D). On immunohistochemical examination, the cytoplasm of the vascular endothelial cells were positive for CD31 (Figure 3E) and CD34 (Figure 3F), and the nuclei of the vascular endothelial cells were positive for ERG (Figure 3G), while only a few endothelial cells were positive for Ki 67 with a positive rate of 7% (Figure 3H). These characteristics were consistent with a diagnosis of AH. There were no signs of postoperative recurrence during the 2-mo CT follow-up.

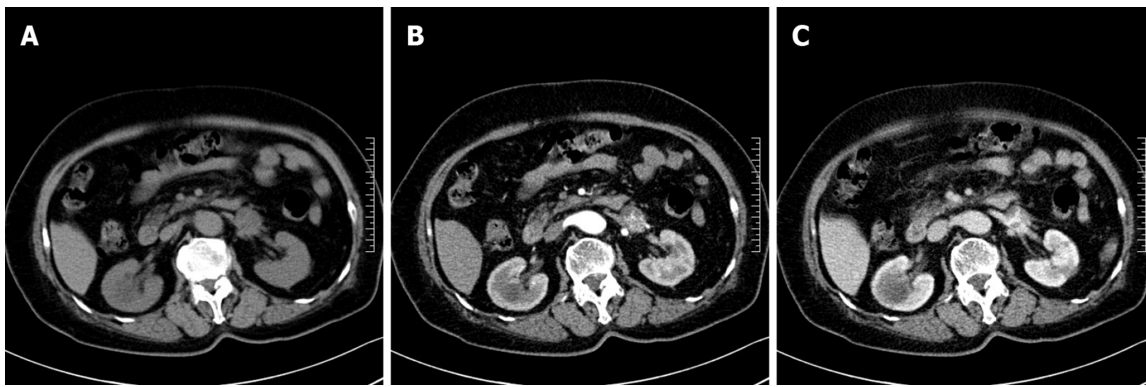


Figure 1 Anastomosing hemangioma of left renal vein on computed tomography. A: Non-contrast-enhanced scan; B: Arterial phase of contrast-enhanced scan; C: Portal phase of contrast-enhanced scan.

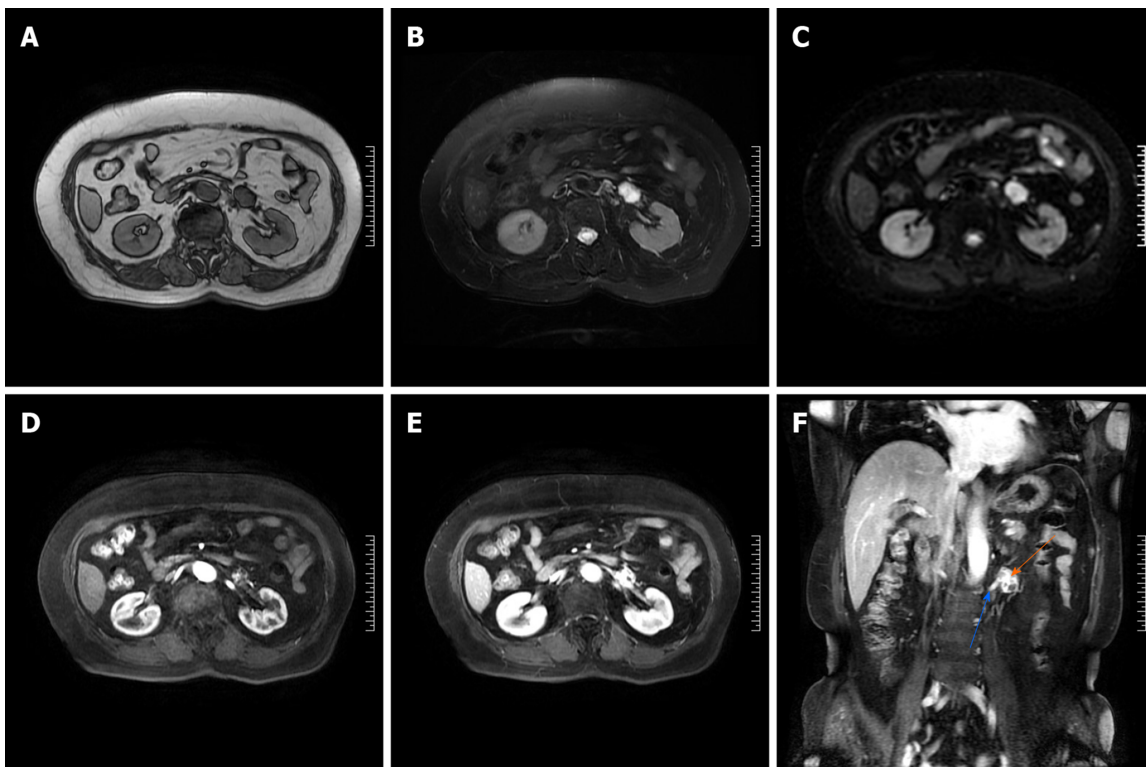


Figure 2 Anastomosing hemangioma of left renal vein on magnetic resonance imaging. A: T1-weighted image; B: T2-weighted image; C: Diffusion-weighted image; D: Arterial phase post-contrast T1-weighted image; E: Portal venous phase post-contrast T1-weighted image; F: Coronal portal venous phase post-contrast T1-weighted image. Orange arrow marks the anastomosing hemangioma, and blue arrow marks the left renal vein.

DISCUSSION

AH is extremely rare, and its etiology and pathogenesis remain unclear. It mainly occurs in the genitourinary tract; however, several cases have been reported in the liver, gastrointestinal tract, retroperitoneum, nasal cavity, and even the intracranial space^[3,4,8,9]. Although AH involving the left renal vein branches has also been reported^[10], we presented the first case of isolated anastomotic hemangioma arising from the left renal vein rather than the renal parenchyma.

AH is more common in middle-aged and elderly people, with a slight male predilection^[11]. Generally, its diameter ranges from 0.1 cm to 6.0 cm^[12]. AH has no special clinical symptoms or laboratory indicators, which is often found on accidental imaging examination. However, the imaging findings of AH show no specificity and are similar to most benign space-occupying lesions. On non-contrast-enhanced CT, the AH showed lobulated lesions, with soft-tissue attenuation, and on contrast-enhanced CT, it presented as a heterogeneous solid lesion with persistent enhancement^[11,13]. On

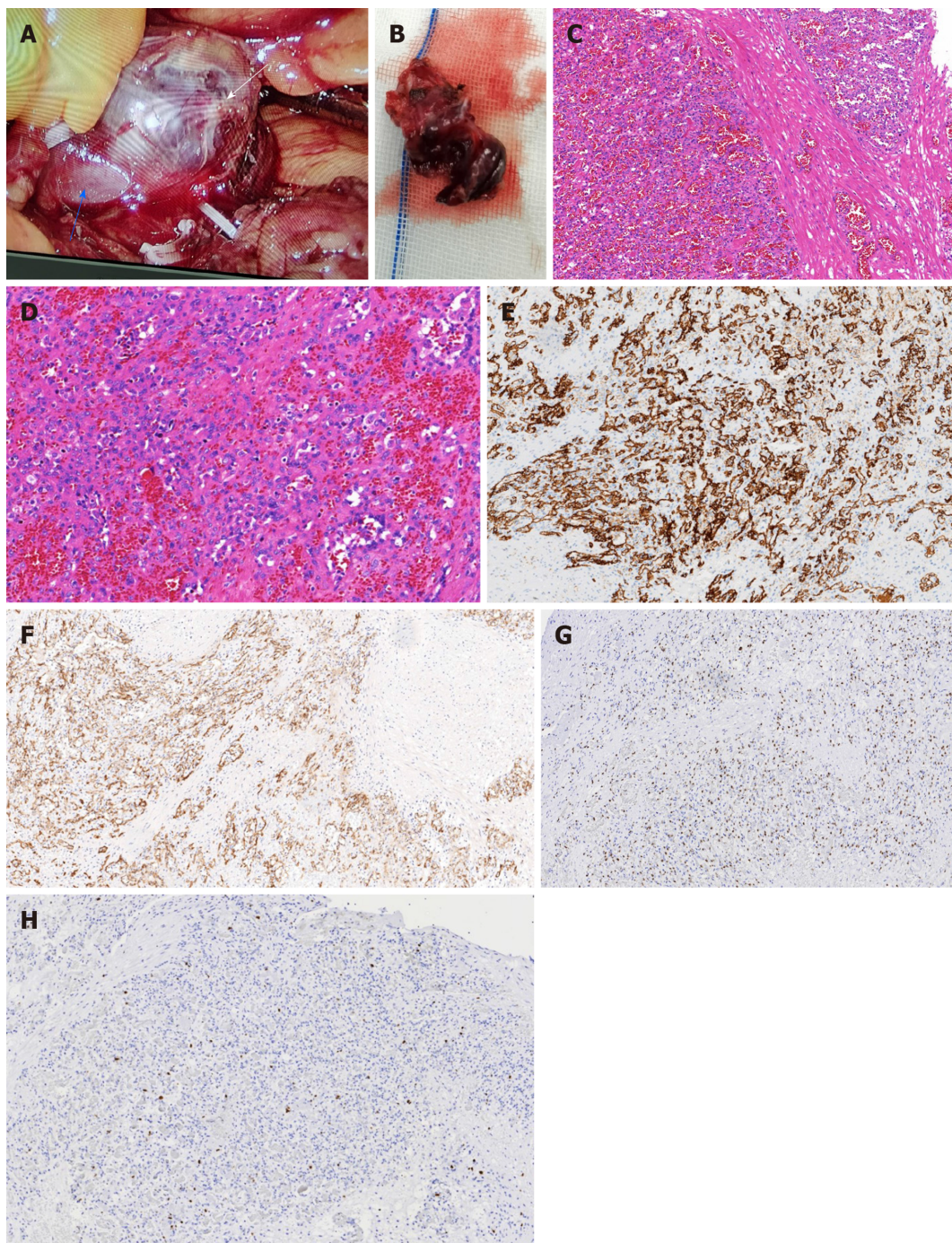


Figure 3 Histopathological findings of anastomosing hemangioma. A: *In situ* anastomosing hemangioma (AH, white arrow) and the left renal vein (blue arrow); B: Cut surface of the AH; C: Hematoxylin and eosin (H&E) staining (magnification: 100 ×); D: H&E staining (magnification: 200 ×); E-H: Immunohistochemical staining (magnification: 100 ×) for CD31 (E), CD34 (F), ERG (G), and Ki 67 (H).

non-contrast-enhanced MRI, the AH presented as a round, well-defined T1-hypointense and T2-hyperintense lesion, and on contrast-enhanced MRI, it presented with avid peripheral enhancement in the arterial phase, which persisted in the delayed phase without central enhancement^[14]. However, Merritt *et al*^[15] also described the characteristics of their AH on MRI. In their report, the lesion presented as homogenous enhancement in the arterial phase, both peripherally and centrally, which persisted in the delayed phase. In our case, the lesion showed heterogeneous septal enhancement in the arterial phase, which persisted in the portal phase.

The diagnosis of AH mainly depends on histopathological examination. Macroscopically, AH is usually a spongy neoplasm with no capsule, but with a clear boundary and mahogany-brown in color^[11,12,16]. Microscopically, AH is characterized by tightly packed capillary channels lined with hobnailed endothelial cells, which are similar in appearance to the red pulp of the spleen, have extramedullary

hematopoiesis, and lack endothelial atypia^[11,17]. Immunohistochemical staining revealed strong and diffuse CD31, CD34, and EGR positivity^[17,18]. It is important to note that there is no mitotic activity, no or slight cellular atypia, and a low Ki-67 index^[11,12,18].

AH should be mainly differentiated from angiosarcoma^[19]. Angiosarcoma is a rare, invasive, malignant tumor and radiologic examination is unable to differentiate it from AH. Histologically, it also presents with hobnailed endothelial cells and can mimic AH^[11]. However, angiosarcoma is characterized by high-grade cell atypia, multiple layers of endothelial cells, and obvious mitotic activity, none of which were present in our case. Therefore, based on these characteristic histopathological findings, our case was diagnosed as AH.

The treatment of AH is controversial because a diagnosis cannot be made from preoperative radiologic examination. When biopsy results are available, the treatment may vary depending on the lesion location, lesion size, and presence of symptoms, such as observation, embolization or radiofrequency ablation, and local or radical resection, to avoid overtreatment. However, some scholars are wary of percutaneous biopsies because of the risk of bleeding in some cases.

AH runs a benign clinical course. Previous studies have shown that there is no propensity for disease recurrence^[11,20]. Yet recent research suggests that a recurrence of AH is indeed possible^[21] and recurrent GNAQ mutations in the pathogenesis of AH^[22]. Although other capillary hemangiomas, especially congenital hemangioma, also have GNAQ mutations, the clinical setting (that is, the age and location of the patient) makes AH unique within this group^[22]. Moreover, GNAQ mutations are not found in angiosarcoma, which may play an important role in distinguishing AH from angiosarcoma. Currently, due to the rarity of AH, there are no established guidelines for its follow-up.

CONCLUSION

We report a case of AH arising from the left renal vein, which, as far as we know, has never been reported before. In light of the lack of specific clinical and radiologic manifestations, AH is easily misdiagnosed preoperatively. Awareness of this entity occurring in the left renal vein will not only help improve doctors' vigilance and reduce the probability of misdiagnosis but also can aid in determining the most appropriate treatment for the patient.

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