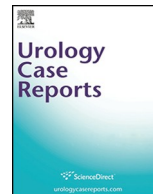




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## Oncology

## Incidental finding of bilateral renal and adrenal anastomosing hemangiomas: A rare case report

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## ARTICLE INFO

## ABSTRACT

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Anastomosing hemangiomas are rare variants of vascular tumors found in adrenal, hepatic, and gastrointestinal tissue. Frequently, renal anastomosing hemangiomas are misdiagnosed on computed tomography (CT) as kidney cancers, resulting in unnecessary workups and detrimental treatments. We present a rare case of bilateral renal and adrenal anastomosing hemangioma found incidentally on renal biopsy. Patient is a 39 year-old African American male on hemodialysis with a history of end-stage renal disease secondary to lupus who presented with acute pericarditis and worsening renal insufficiency.

## Introduction

Vascular tumors of the urinary system are exceptionally rare diagnoses.<sup>1</sup> Anastomosing hemangiomas are a variant of vascular tumors with complex pathologic architecture characterized by anastomosing vessels, rare hobnailing, and benign features.<sup>2</sup> Although anastomosing hemangioma is predominately observed in the genitourinary system, it has been reported in the adrenal glands, liver and gastrointestinal tract.<sup>3</sup> In this case report, we present a patient with incidental findings of renal and adrenal anastomosing hemangiomas during workup for glomerulonephritis.

## Case presentation

A 39 year-old African American male with a history of glomerulonephritis secondary to systemic lupus erythematosus presented to the emergency department with chest pain. Past medical history is significant for hypertension, hyperlipidemia, and anemia. Denies family history of kidney disease or renal malignancy. Patient is a lifetime nonsmoker. During hospital course, patient was diagnosed with acute pericarditis and worsening chronic kidney disease. Renal biopsy was performed showing segmentally sclerotic foci, and glomerular basement membrane thickening, and extensive subepithelial spikes in addition to moth-eaten appearance to the basement membrane. Granular capillary wall staining was positive for IgA, IgG, IgM, kappa light chains, C3, and C1q. Based on these microscopy and immunofluorescence findings, patient was

diagnosed with membranous lupus nephritis, class V. Interestingly, a section of renal tissue contained a neoplasm comprising irregular vascular channels lined by benign appearing endothelial cells without evidence of mitotic activity or cellular atypia (Fig. 1). Patient was diagnosed with incidental finding of benign renal anastomosing hemangioma. Two years later upon annual renal CT monitoring, bilateral hypervascular renal masses measuring 9 mm × 9 mm and 13 mm × 15 mm were discovered (Fig. 2). Of note, radiologist noted a 10 mm enhancing right adrenal lesion concerning for metastatic disease. In the follow years, patient was dependent on peritoneal dialysis, underwent bilateral nephrectomy, and enlisted for kidney transplant. Surgical specimen showed non-encapsulated multicystic mass measuring up to 0.6 cm with vascular prominence suggesting a hemangioma. The cysts were lined by a single layer of cuboidal oncotic cells (Fig. 3a and b). Both kidneys in addition to the right adrenal gland were noted to have features of anastomosing hemangiomas. Patient successfully underwent kidney transplant and is doing well with adequate follow up.

## Discussion

With increasing utility of imaging modalities and biopsy techniques, incidental findings have become more prevalent. Uncommonly, primary benign vascular lesions are discovered on CT scans and surgical pathology specimens. Often, these vascular lesions can mimic malignancies, and thus, requiring unnecessary workup and surgical interventions.

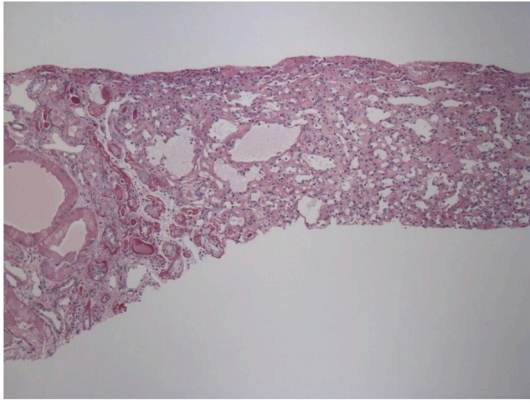
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**Fig. 1. Needle Core Biopsy of Renal Parenchyma.** A lesion transected by the core biopsy is composed of anastomosing capillary sized vessels lined by benign appearing endothelium. PAS stain, 200x.

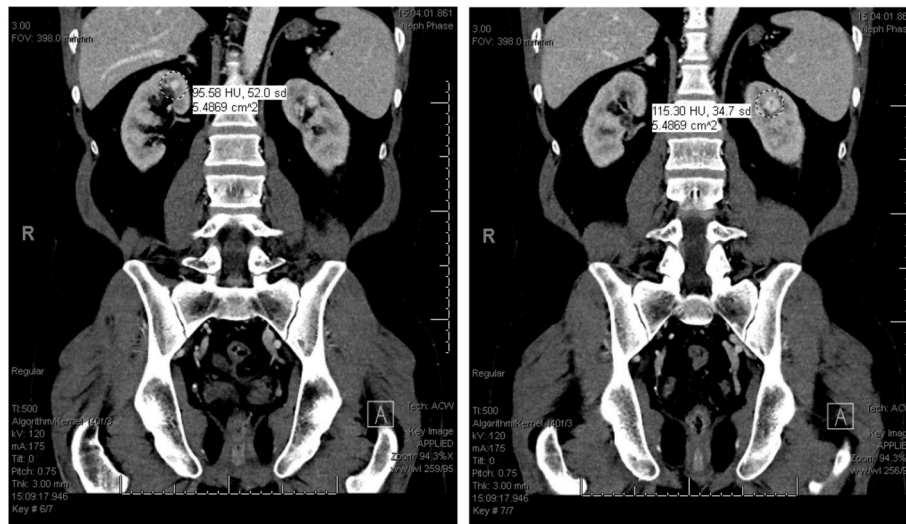
Anastomosing hemangiomas are a rare variant of capillary hemangiomas. These hemangiomas were initially documented by Montgomery and Epstein who described six cases of anastomosing hemangiomas of the kidneys, perinephric adipose tissue, and testicles.<sup>2</sup> Consequently, more cases of this vascular tumor have been reported in literature involving tissues from the genitourinary system, adrenal glands, liver, and gastrointestinal tract.<sup>3</sup> Approximately 60% of

anastomosing hemangiomas are asymptomatic. Patient with renal anastomosing hemangiomas typically present with nonspecific findings such as hematuria, flank pain, abdominal pain, hematoma, and/or dysuria. Non-renal anastomosing hemangiomas may present with local or back pain, neurologic deficits, or pleural effusion.<sup>4</sup> Most patients are diagnosed in their fifth to sixth decade of life. This tumor is more common in males with a male:female ratio of 11:8.<sup>5</sup>

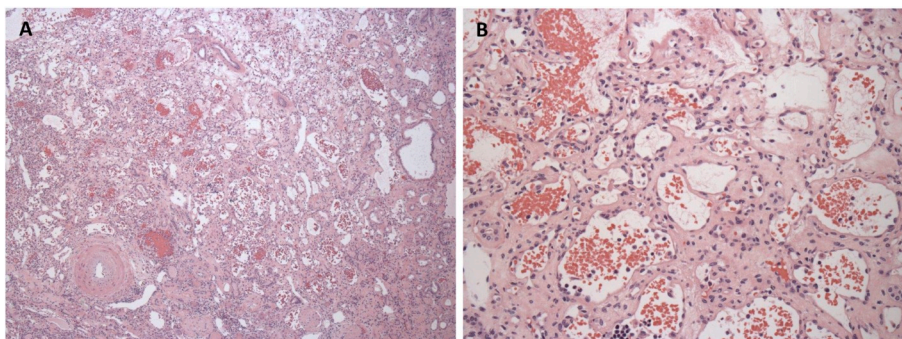
Although anastomosing hemangiomas share similar radiological features to various other tumors, certain features can help radiologist consider this diagnosis. A renal mass marked with T2 hyperintensity, similar to a cyst, is often characteristic of renal anastomosing hemangiomas. Filing pattern of contrast from peripherally to centrally on dynamic CT or MRI should help guide clinical decision making to pursue percutaneous renal biopsy rather than nephrectomy.<sup>3</sup>

Histologically, anastomosing hemangiomas can be confused for angiosarcomas due to the presence of hyaline globules, anastomosing vascular patterns, and hob-nailing endothelial cells. However, unlike these malignant tumors, anastomosing hemangiomas lack mitotic activity, multilayering, and atypia of endothelial cells.<sup>3</sup> Microscopically, these vascular tumors demonstrate loosely lobulated architectures with alternating cellular and hypocellular areas. Of note, the cellular region contains significant capillary vasculature proliferation in an anastomosing pattern.<sup>1</sup>

Several syndromes, including Sturge-Weber and Klippel-Trenaunay syndromes, are associated with hemangioma, particularly in the bladder. However, there is no evidence for the association of renal anastomosing hemangiomas with any syndromic condition. Of the reported cases, about 40% of patients have an underlying diagnosis of



**Fig. 2. Computed tomography of abdomen and pelvis showing bilateral renal masses.**



**Fig. 3. Surgical Pathology Specimen from Bilateral Radical Nephrectomy.** A. Hematoxylin and eosin stain at 100X objective showing multiple well-defined lesions composed of anastomosing sinusoidal capillary sized vessels. B. Hematoxylin and eosin stain at 400X objective showing capillary sized vessels with mild endothelial cell nuclear variability and scattered endothelial cells with hobnail shapes. Mitotic figures are absent on slide.

ESRD and 60% of cases were incidentally found during evaluation of ESRD.<sup>1</sup> The pathogenesis of anastomosing hemangiomas in patients with severe chronic kidney disease still remains unclear.

Although pathologically this tumor is benign, 90% of renal anastomosing hemangiomas are treated with total nephrectomy. In contrast, more conservative treatment is reported for non-renal anastomosing hemangiomas.<sup>4</sup> This disparity in treatment for renal and non-renal involvement is most likely due to misdiagnosis on imaging for renal malignancy. Prognosis for this diagnosis is good. Of the 31 case reports, only one patient died of unrelated disease; no patients developed disease recurrence or metastatic disease.<sup>1</sup> Hence, more imaging studies are necessary to better analyze radiologic findings for renal anastomosing hemangiomas to prompt appropriate clinical decision making.

## Conclusion

In conclusion, we presented a case of incidental diagnosis of renal and adrenal anastomosing hemangiomas, a rare form of vascular tumors. Since anastomosing hemangiomas are benign tumors, less aggressive treatment, such as active surveillance, may be sufficient to monitor these individuals. However, given that these tumors are frequently misdiagnosed as more aggressive diseases, conservative options are often not considered. Better understanding of this condition and radiographic indicators can reduce misdiagnoses to ensure patients receive optimal treatment.

## Statement of ethics

This manuscript has not been published in whole or part elsewhere nor currently being considered for publication in another journal. All authors have been personally and actively involved in the publication of this paper and will hold themselves jointly and individually responsible for its content. Personal and identifying information regarding the case has been retracted for confidentiality purposes.

## Disclosure statement

The authors declare that there are no conflicts of interest regarding the publication of this paper.

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## Author contributions

Sagar Patel, Obafunbi Abimbola, and Tiaggpaul Bhamber drafted the manuscript and researched the current literature. Carol Weida was the pathologist who read the tissue specimen. Ornob Roy supervised the project and provided edits to manuscript.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.eucr.2019.100912>.

## References

1. Tao L-L, Dai Y, Yin W, Chen J. A case report of a renal anastomosing hemangioma and a literature review: an unusual variant histologically mimicking angiosarcoma. *Diagn Pathol.* 2014;9(1) <https://doi.org/10.1186/s13000-014-0159-y>.
2. Montgomery E, Epstein JI. Anastomosing hemangioma of the genitourinary tract. *Am J Surg Pathol.* 2009;33(9):1364–1369. <https://doi.org/10.1097/pas.0b013e3181ad30a7>.
3. Cheon PM, Rebello R, Naqvi A, Popovic S, Bonert M, Kapoor A. Anastomosing hemangioma of the kidney: radiologic and pathologic distinctions of a kidney cancer mimic. *Curr Oncol.* 2018 Jun;25(3):e220.
4. Perdiki M, Datseri G, Liapis G, et al. Anastomosing hemangioma: report of two renal cases and analysis of the literature. *Diagn Pathol.* 2017;12(1) <https://doi.org/10.1186/s13000-017-0597-4>.
5. Zhao M, Li C, Zheng J, Sun K. Anastomosing hemangioma of the kidney: a case report of a rare subtype of hemangioma mimicking angiosarcoma and review of the literature. *Int J Clin Exp Pathol.* 2013;6(4):757–765.