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An Unusual Case of Renal Angiomyolipoma Hemorrhage: Renal Angiomyolipoma Endowed with a Vascular Supply That Defied Tradition, Originating Directly from The Abdominal Aorta

Olağandışı Bir Renal Anjiyomiyolipom Kanaması Vakası: Geleneklere Aykırı Damarsal Kaynağa Sahip Olan, Doğrudan Abdominal Aorttan Kaynak Alan Renal Anjiyomiyolipom

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ABSTRACT

Renal angiomyolipomas typically receive their blood supply from the renal arteries. However we present a rare case of spontaneous renal angiomyolipoma hemorrhage, where the angiomyolipoma's arterial supply originated directly from the abdominal aorta rather than the renal artery. This case report outlines the clinical presentation, diagnosis, and successful management of this unique anomaly through coil embolization. To the best of our knowledge, this is the first reported case in the literature of a renal angiomyolipoma with abdominal aortic blood supply stabilized using coil embolization.

Keywords: Renal angiomyolipoma, hemorrhage, coil embolization, aorta

ÖZ

Renal anjiyomiyolipomlar genellikle kanlanmalarını renal arterlerden alırlar. Ancak burada anjiyomiyolipomun arteriyel kanlanmasının renal arter yerine doğrudan abdominal aorttan kaynaklandığı nadir bir spontan renal anjiyomiyolipom kanaması olgusunu sunuyoruz. Bu olgu sunumu, bu benzersiz anomalinin klinik tablosunu, tanısını ve koil embolizasyonu ile başarılı tedavisini özetlemektedir. Bildiğimiz kadarıyla, direkt abdominal aorttan kanlanmaya sahip renal anjiyomiyolipomun koil embolizasyonu ile stabilize edildiği literatürde bildirilen ilk olgudur.

Anahtar Sözcükler: Renal anjiyomiyolipom, hemoraji, koil embolizasyonu, aort

INTRODUCTION

Renal angiomyolipomas are rare benign tumors consisting of three components: smooth muscle, vascular structures, and fatty components (1). They are most commonly diagnosed. However, they may be associated with tuberous sclerosis complex and sporadic lymphangioleiomyomatosis (2). These benign tumors often present no cause for concern. However, although they are often asymptomatic and detected incidentally, they can cause gross hematuria, flank pain, palpable tender mass, and retroperitoneal hemorrhage (3). Renal angiomyolipomas are the most common cause of retroperitoneal bleeding of non-traumatic renal origin (1). Various methods such as active surveillance, ablation, selective arterial embolization, and open or laparoscopic surgery are used in the treatment of renal angiomyolipomas, depending on their pathological and clinical features (4). The prevalence of sporadic renal angiomyolipomas in the general population was found to be 0.44% in a study of 61,400 patients, and increases with females (5). The diagnosis of renal angiomyolipomas is made more easily using ultrasonography, computed tomography (CT), magnetic resonance imaging, and biopsy (2).

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They are conventionally acknowledged as receiving their vascular nourishment from the intricate network of branches originating from the renal artery. In this exceptional instance, we chronicle the extraordinary revelation of a renal angiomyolipoma that defied convention by deriving its blood supply not from the renal arterial tributaries but directly from the abdominal aorta. This unprecedented vascular anomaly precipitated a cascade of events, culminating in a spontaneous and potentially life-threatening angiomyolipoma hemorrhage. That set the stage for an intricate clinical conundrum. As we delve into this remarkable case, it becomes evident that this phenomenon represents a medical enigma of the highest rarity, with no precedent or documentation in the annals of medical literature to illuminate the path we were about to traverse.

CASE REPORT

A 45-year-old female patient presented to the emergency department with complaints of weakness, right-sided abdominal pain three days, and hematuria occurring three days prior. The patient was free of comorbidities, had no substantial medical history, and had never had surgery. On physical examination, the patient had right costovertebral angle tenderness and no fever. Other system examinations were normal. Laboratory tests revealed a progressive drop in hemoglobin levels from 11.4 g/dL to 9.4 g/dL, to 7.2 g/dL, within one to two days. A dynamic renal ultrasonography confirmed the presence of an angiomyolipoma and a retroperitoneal hematoma. Abdominal CT scan, revealed a 132x92 mm cyst with thickened walls and partial contrast enhancement in the lower pole of the right kidney, suspected to be an angiomyolipoma. Additionally, a hematoma was observed within and around the angiomyolipoma, extending into the retroperitoneal space (Figure 1). The patient was hospitalized, stabilized with six units of erythrocyte suspension, and subsequently evaluated by interventional radiology.

Treatment and Management

Coil embolization was chosen as the treatment modality after evaluation. The aberrant vessel was responsible for the angiomyolipoma's hemorrhage. It was found to originate from the abdominal aorta and was successfully embolized using coils (Figure 2). Subsequent monitoring revealed the cessation of bleeding into the angiomyolipoma, and the patient remained stable (Figure 3). She was discharged in a stable condition. Follow-up exams at 1 and 3 months after discharge showed no sign of a recurrence of the angiomyolipoma hemorrhage, which was also supported by laboratory testing and CT scans.

DISCUSSION

Although renal angiomyolipomas are usually diagnosed incidentally, in some cases they may present with hemorrhage (6). Renal angiomyolipomas have long been associated with a consistent vascular supply, predominantly arising from branches stemming from the renal artery. However, our presented case challenges the very essence of this conventional understanding by unveiling a remarkable and exceedingly rare anomaly: a renal angiomyolipoma endowed with a vascular supply that defied tradition, originating directly from the abdominal aorta. This aberration, while perplexing, is not without precedent in the broader realm of vascular anomalies; nevertheless, its occurrence within the context of a renal angiomyolipoma, coupled with the consequential life-threatening spontaneous hemorrhage, elevates it to unparalleled uniqueness. Spontaneous retroperitoneal hemorrhage originating from the kidney was first described by Bonnet in 1700, and this rarely observed form of hemorrhage was given its name by Wunderlich in 1856 (7).

The case presented here highlights the intricate nature of renal vascular anatomy. While the majority of renal angiomyolipomas indeed derive their blood supply from the renal artery, aberrant

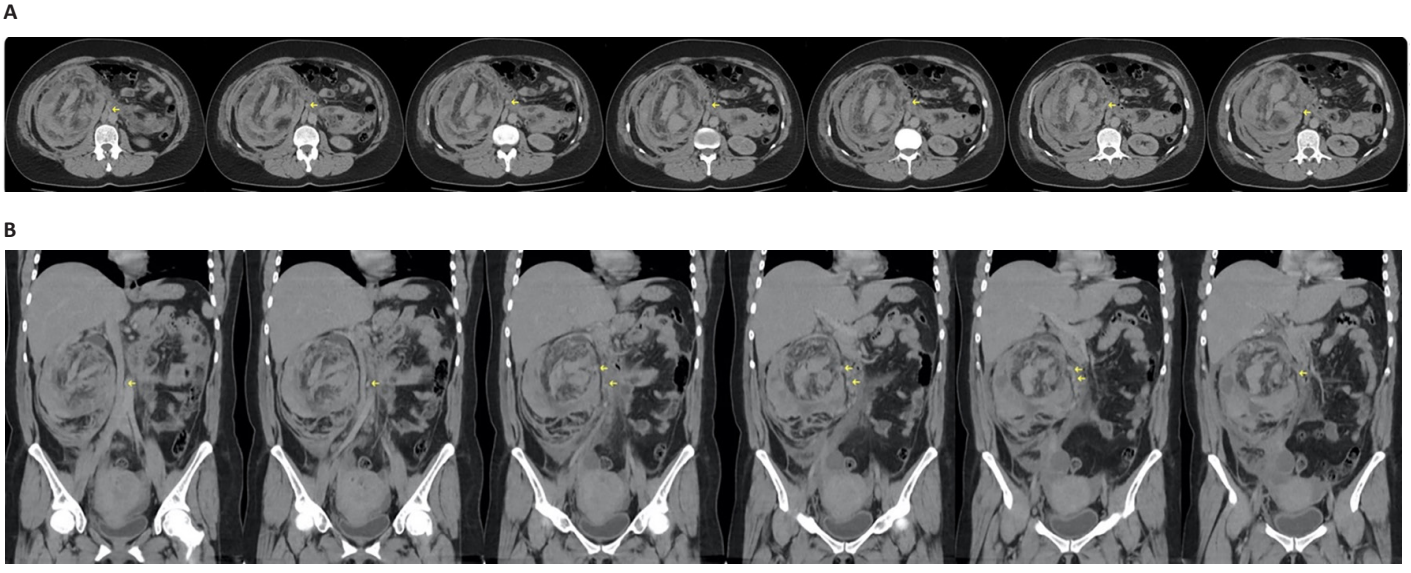

Figure 1. (A) Hemorrhagic renal angiomyolipoma and artery from the aorta on CECT image (Axial). B) Hemorrhagic renal angiomyolipoma and artery from the aorta on CECT image (Coronal).

CECT: Contrast-enhanced computed tomography

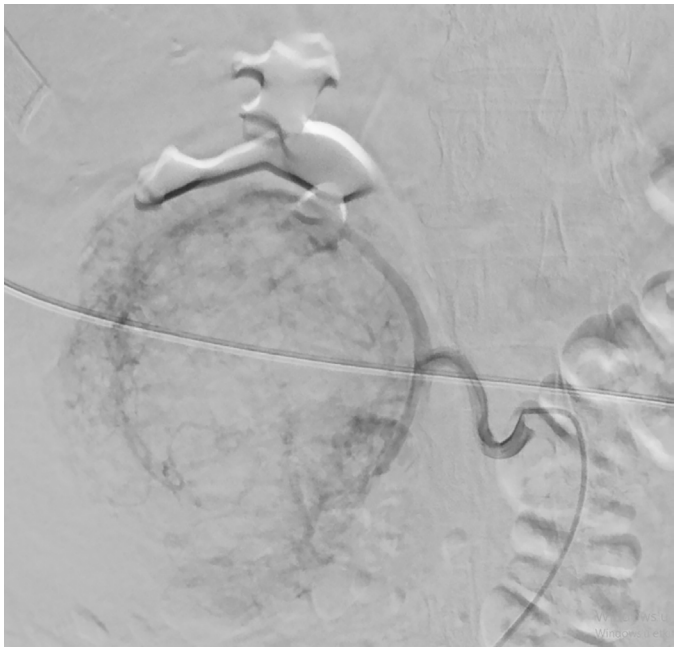

Figure 2. Coil embolization image of a renal angiomyolipoma artery originating from the abdominal aorta.

vascular patterns are not unheard of in the vascular landscape. Anomalous arterial origins have been previously documented in various anatomical contexts, such as renal vascular anomalies and abdominal aortic variants. However, the emergence of such an aberrant vascular supply in the context of a renal angiomyolipoma is an exceedingly rare occurrence. Its presentation as spontaneous hemorrhage is a clinical conundrum that demands meticulous exploration.

The precise craftsmanship of coil embolization is a hallmark of a well-known procedure in the field of interventional radiology. Also, it is crucial for non-invasive interventions, such as in this unusual instance. It has been shown by Kothary et al. (8) and Blakeley and Thiagalingham (9) that selective arterial embolization is an effective method for controlling hemorrhage resulting from benign renal lesions. As in our case, Wang et al. (10) successfully applied arterial embolization as a non-surgical treatment method to stop hemorrhage in the presence of spontaneous retroperitoneal hemorrhage caused by renal angiomyolipomas. We were able to occlude this aberrant vessel rooted from the abdominal aorta. Then, we effectively halted hemorrhage within the angiomyolipoma, and then we prevented the further bleeding. This intervention not only proved instrumental in stabilizing the patient but also paved the way for her complete recovery.

The successful outcome of this case serves as a testament to the crucial role of interventional radiology in managing intricate vascular anomalies. Coil embolization, with its precision and efficacy, stands as a valuable tool in the armamentarium of clinicians faced with such challenging scenarios. Our experience adds to the growing body of evidence supporting the utility of this intervention in managing not only renal angiomyolipoma hemorrhage but also other vascular anomalies with aberrant origins.

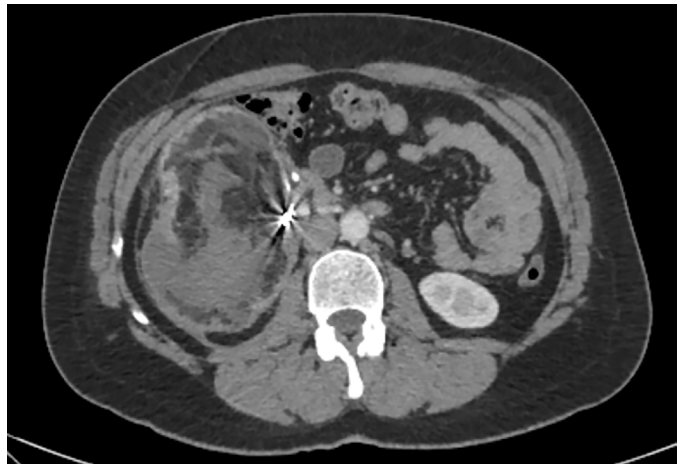

Figure 3. CECT image after coil embolization.

CECT: Contrast-enhanced computed tomography

While aberrant vascular supply to renal angiomyolipomas is a rarity, it is not entirely unprecedented in the broader context of vascular anomalies. Previous reports have highlighted variations in renal vascular anatomy, such as accessory renal arteries and cases where renal angiomyolipomas received their blood supply from unusual sources like the inferior mesenteric artery. However, the specific scenario of a renal angiomyolipoma directly supplied by the abdominal aorta is rarely sparse in the literature.

Our case report contributes to the existing knowledge base by showcasing the successful application of coil embolization in managing this exceptional anomaly. To the best of our knowledge, this is the first documented case where a renal angiomyolipoma with an abdominal aortic blood supply was stabilized and hemorrhage controlled using this method. The absence of precedent highlights the rarity of such cases and underscores the importance of sharing our experience.

CONCLUSION

This case highlights the importance of considering aberrant vascular anatomy when managing renal angiomyolipoma hemorrhage. While renal artery supply is the norm, anomalous cases such as this one should be kept in mind. Coil embolization emerged as an effective intervention in this instance, preventing recurrent hemorrhage and ensuring the patient's stability. Further research and reporting of similar cases are necessary to broaden our understanding of such rare anomalies.

Ethics

Informed Consent: Patient consent it was obtained.

Footnotes

Authorship Contributions

Concept: C.Y., A.Y., Design: C.Y., A.Y., Data Collection or Processing: C.Y., A.Y., Analysis or Interpretation: C.Y., A.Y., Literature Search: C.Y., A.Y., Writing: C.Y., A.Y., Critical Review: C.Y., A.Y.

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