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Adrenal Anastomosing Hemangioma: A Rare Incidentaloma Mimicking Malignancy

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ABSTRACT

Anastomosing hemangioma is a rare benign vascular tumor most commonly described in the genitourinary tract, with occasional involvement of the adrenal gland.

Due to its nonspecific imaging features, adrenal anastomosing hemangioma may mimic malignant adrenal tumors, potentially leading to misdiagnosis and unnecessary surgical resection.

Here we report a case of a 74-year-old man with an incidental 4-cm right adrenal mass discovered during evaluation for elevated prostate-specific antigen levels. Imaging findings were indeterminate, with moderate Computed Tomography (CT) attenuation, marked T2 hyperintensity and low 18F-Fluorodeoxyglucose (FDG) uptake, while hormonal evaluation excluded a functional adrenal tumor. Laparoscopic adrenalectomy was performed. Histopathological examination confirmed the diagnosis of adrenal anastomosing hemangioma.

This case highlights the diagnostic challenge of adrenal anastomosing hemangioma, both radiologically and histologically. Increased recognition of this entity may help refine management strategies and reduce overtreatment.

Keywords: Anastomosing hemangioma, Adrenal gland, Vascular tumor

Abbreviations: MRI: Magnetic Resonance Imaging; CT: Computed Tomography; HU: Hounsfield Units; FDG: ^{18}F-Fluorodeoxyglucose; EMA: Epithelial Membrane Antigen; HMB-45: Human Melanoma Black-45

1. Introduction

Anastomosing hemangioma is a rare, benign vascular tumor that can arise in various anatomic locations. It typically occurs in the genitourinary system but also affects other organs such as soft tissue and bone, gastrointestinal tract and adrenal gland[1-3]. This vascular neoplasm was originally reported in 2009[4]. The first adrenal gland case was described in 2012[5]. To our knowledge, approximately twenty cases of anastomosing hemangioma with adrenal involvement have been reported[5-17].

Anastomosing hemangiomas of the adrenal gland are typically known as asymptomatic tumors and are most often identified as incidentalomas. While adrenal anastomosing hemangioma usually presents as a solitary lesion, multifocal presentations have also been reported[15,16]. The absence of pathognomonic

imaging features frequently raises suspicion of malignancy, which may subsequently lead to surgical overtreatment. We report a case of adrenal anastomosing hemangioma incidentally discovered on Magnetic Resonance Imaging (MRI) performed for evaluation of elevated prostate-specific antigen.

2. Case Presentation

A 74-year-old male patient with a history of benign prostatic hyperplasia underwent pelvic Magnetic Resonance Imaging (MRI) for evaluation of elevated prostate-specific antigen. The MRI revealed two focal PI-RADS 3 lesions and an incidental 4-cm mass of the right adrenal gland.

Further evaluation with abdominal Computed Tomography (CT) demonstrated a right adrenal oval lesion measuring 4.0 x 3.7 x 3.2 cm with an unenhanced CT value of 15 Hounsfield Units (HU) (Figure 1). Absolute and relative washout values were 26% and 21%, respectively. Based on these findings, pheochromocytoma or adenoma were considered in the differential diagnosis. T2-weighted MRI sequences revealed a markedly hyperintense lesion, consistent with the "light-bulb" sign classically associated with pheochromocytoma (Figure 2). The lesion appeared to contain multiple septations, without any fatty component and demonstrated peripheral enhancement (Figure 3). The mass abutted the VII segment of hepatic parenchyma and local invasion could not be excluded. A complementary ^{18}F-Fluorodeoxyglucose (FDG) PET-CT showed low radiotracer uptake (SUVmax 2.3) and no evidence of other hypermetabolic lesions.

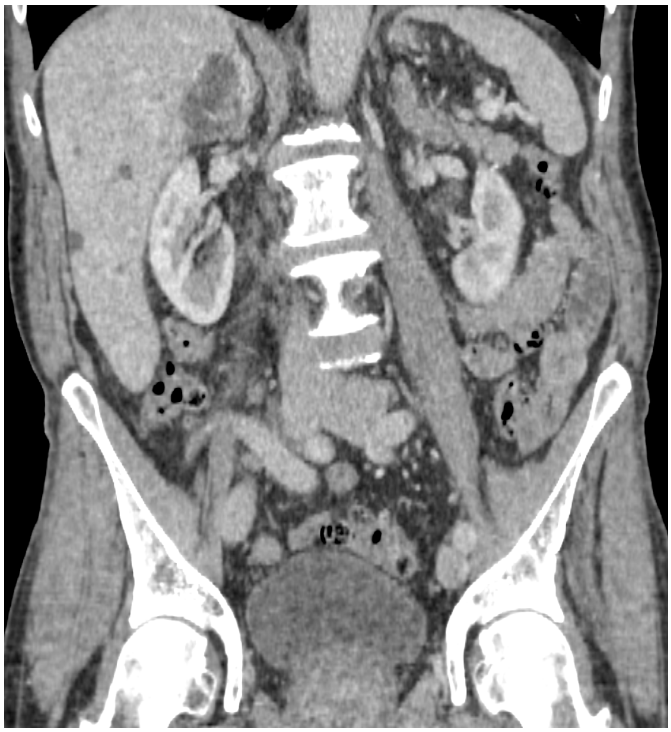

Figure 1: Coronal unenhanced CT image showing an oval-shaped lesion of the right adrenal gland.

The patient had a past medical history of suspected hypertension for several months, confirmed only by home blood pressure monitoring, although no antihypertensive treatment had been initiated. He reported none of the symptoms consistent with the classic triad of pheochromocytoma (headache, sweating and palpitations). Physical examination revealed no clinical signs of Cushing's syndrome.

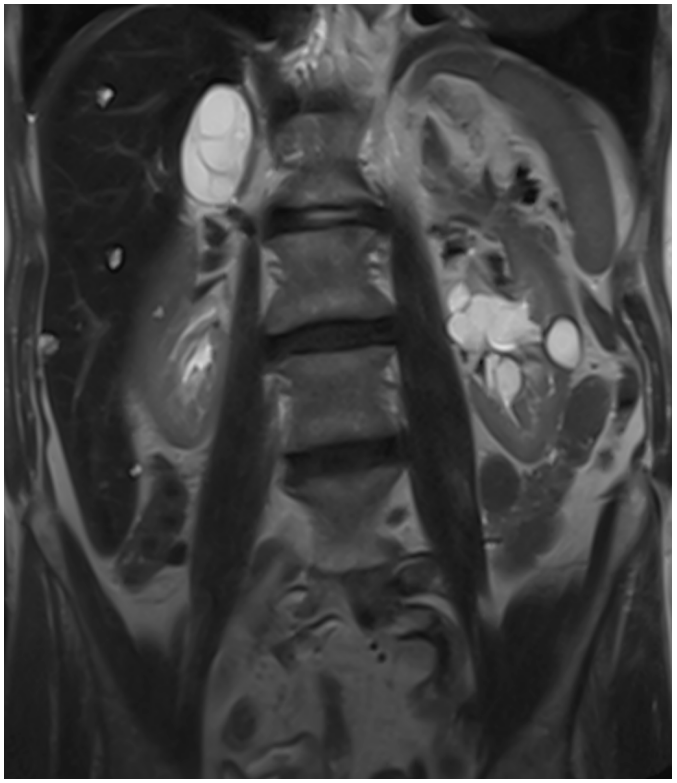

Figure 2: Coronal T2-weighted HASTE MRI demonstrating a well-circumscribed right adrenal lesion with marked hyperintensity ("light-bulb sign").

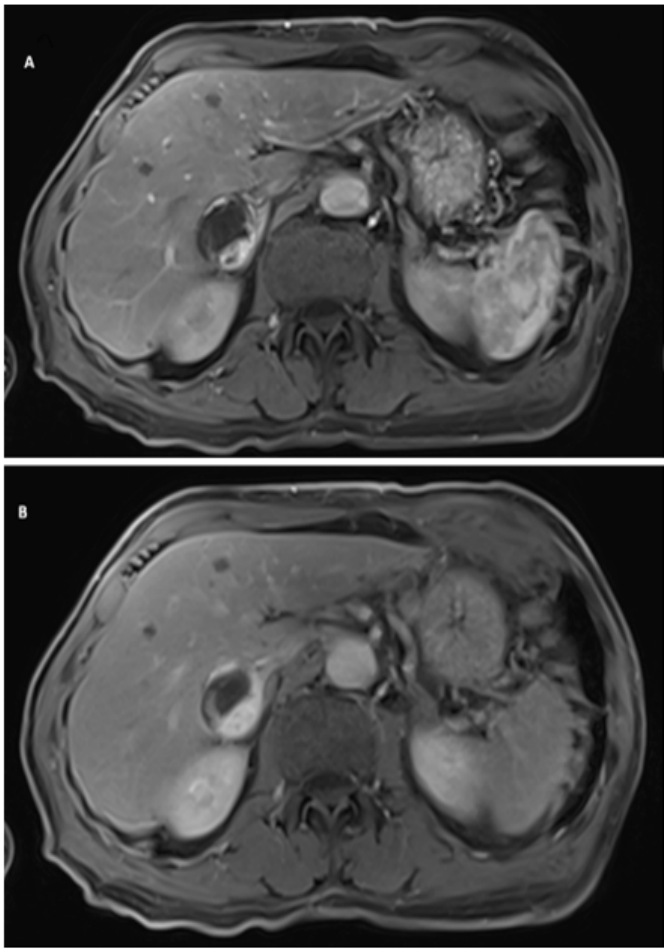

Figure 3: Arterial-phase (A) and delayed-phase (B) contrast-enhanced MRI demonstrating peripheral enhancement of the right adrenal lesion with progressive centripetal filling.

Endocrine workup demonstrated plasma metanephrine and normetanephrine levels within normal limits, with no biochemical evidence of pheochromocytoma. Normal salivary cortisol levels together with a borderline dexamethasone suppression test result made Cushing's syndrome unlikely. Dehydroepiandrosterone and steroid precursor concentrations were also within the normal range. The plasma aldosterone-to-renin ratio was within normal limits, making primary aldosteronism unlikely.

Four months after the initial detection, the patient was diagnosed with symptomatic bilateral pulmonary embolism, with no identifiable thromboembolic risk factors. Anticoagulation was initiated and an inferior vena cava filter was placed three days before surgery. Following appropriate preoperative anticoagulation management, a right laparoscopic adrenalectomy was successfully performed.

Macroscopic examination revealed a 3.2 × 2.8 × 2.4 cm cystic, encapsulated nodule containing mucoid and spongy material. Microscopically, the tumor consisted of thin-walled, anastomosing, capillary-sized vascular channels lined by a single layer of prominent hobnail endothelial cells separated by paucicellular stroma. Additional histological findings included small thrombi and rare hematopoietic cells. No significant mitotic activity or tumor necrosis was observed. Immunohistochemistry demonstrated CD31 and CD34 positivity, while MYC protein expression was negative. Immunohistochemical markers for epithelioid tumors, including Epithelial Membrane Antigen (EMA) and Human Melanoma Black-45 (HMB-45), pancytokeratin and Melan-A showed negative staining. The MIB-1 proliferation index was less than 1%.

The postoperative course was uneventful. The patient was discharged on postoperative day 2 while receiving anticoagulation therapy.

3. Discussion

Although renal anastomosing hemangioma may present with nonspecific symptoms such as abdominal or flank pain, hematuria or dysuria[18], adrenal anastomosing hemangiomas are typically clinically silent and are often identified incidentally on imaging performed for unrelated indications. These tumors show a male predominance, similar to renal anastomosing hemangioma[18]. Although age at presentation varies, most patients are diagnosed in the fifth decade of life or later.

Endocrine laboratory findings are typically normal, as adrenal anastomosing hemangioma is a nonfunctioning tumor. Functional tumors, such as pheochromocytoma and aldosterone- or cortisol-producing adenomas should be excluded through appropriate biochemical testing[19-21]. Urine steroid metabolomics, in combination with imaging characteristics, may enable highly accurate detection of adrenocortical carcinoma[22]. In the present case, a comprehensive hormonal workup was unremarkable, consistent with previously reported cases.

Radiological evaluation of adrenal anastomosing hemangioma should be interpreted in accordance with European Society of Endocrinology guidelines for newly discovered adrenal incidentalomas. These recommendations rely primarily on unenhanced CT attenuation values (<10 HU, 11-20 HU or >20 HU) and lesion size (≤4 cm or >4 cm), which are key determinants of risk stratification and subsequent management[19]. However, as illustrated by this case, adrenal anastomosing hemangiomas may fall into indeterminate categories. The typical imaging appearance of adrenal anastomosing hemangioma includes a round or oval, well-circumscribed solid or cystic lesion with low to mild density and, in most cases, a filling pattern of peripheral enhancement during the arterial phase, similar to anastomosing hemangiomas at other sites. Moreover, MRI typically demonstrates low signal intensity on T1-weighted images and high signal intensity on T2-weighted images with marked arterial-phase enhancement and a centripetal filling pattern during the portal phase. These findings may overlap with those of pheochromocytoma, representing a significant source of diagnostic uncertainty. ^{18}F-FDG PET-CT typically demonstrates low to mild FDG uptake with correspondingly low standardized uptake values (SUVmax)[8] and may provide complementary evidence supporting a benign process.

Histologically, anastomosing hemangioma is a well-demarcated lesion characterized by a lobulated architecture composed of sinusoidal, capillary-like vessels arranged in an anastomosing pattern and lined by hobnail endothelial cells.

Angiosarcoma represents a key histological mimicker of anastomosing hemangioma and typically exhibits an infiltrative growth pattern, in contrast to the well-circumscribed nature of anastomosing hemangioma. Hallmarks of malignancy, including cytological atypia, endothelial multilayering and increased mitotic activity, are characteristically absent in anastomosing hemangioma. Immunohistochemically, tumor cells show positivity for endothelial markers, including CD31, CD34, ERG, FLI-1 and factor VIII-related antigen[7,8,10,11]. The proliferation index is usually low[7,10,11]. Other diagnoses considered include Kaposi sarcoma, which demonstrates strong HHV-8 expression[8,12,15] and angiomyolipoma, which expresses melanocytic markers such as HMB-45 and Melan-A[8,15].

This case highlights the limitations of current diagnostic approaches to adrenal incidentalomas. Despite non-functional status and low metabolic activity on PET imaging, surgical resection was pursued due to persistent diagnostic uncertainty. This underlines the need for improved preoperative characterization of rare benign vascular tumors to avoid unnecessary surgical intervention.

4. Conclusion

Adrenal anastomosing hemangioma is a rare benign vascular neoplasm for which unnecessary or aggressive treatment should be avoided. Accurate preoperative diagnosis remains challenging and most cases are managed surgically due to suspicion of malignancy. The lack of specific radiological features in adrenal anastomosing hemangioma complicates early diagnosis through imaging. Conservative strategies, including monitoring, may be considered in selected patients with indeterminate but stable lesions and reassuring metabolic features. Greater awareness of the clinical presentation and imaging features of this entity may improve clinical decision-making.

5. Declarations

5.1. Ethical Approval and Consent

Ethical approval was not required for this case report. Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

5.2. Conflict of Interest

The authors declare no conflicts of interest.

5.3. Funding

This research received no external funding.

5.4. Author Contributions

RB drafted the manuscript. SM selected and interpreted the radiologic images. SM and IF critically revised the manuscript. All authors approved the final version of the manuscript.

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