Retroperitoneal hematoma secondary to inferior adrenal artery spontaneous hemorrhage

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Spontaneous retroperitoneal hematoma (SRH) is a rare, potentially lethal entity that can arise from a variety of etiologies. We present a case of SRH secondary to hemorrhage from the right inferior adrenal artery in a 22-year-old woman. The patient presented to the emergency room with significant right flank pain, and computed tomography (CT) demonstrated a large, right-sided retroperitoneal hematoma with no identifiable etiology. Renal angiography revealed active extravasation from the right inferior adrenal artery. The patient was definitively treated with endovascular coiling of the ruptured artery, and long term follow up demonstrated resolution of the retroperitoneal hematoma.

Key Words: adrenal artery hemorrhage, spontaneous retroperitoneal hemorrhage, renal angiography

Introduction

Nontraumatic retroperitoneal hemorrhage is an uncommon but potentially life threatening condition with a variety of causes ranging from vascular rupture to renal and adrenal malignancy, benign lesions such as renal cysts, and angiomyolipomas.[1] Spontaneous adrenal artery hemorrhage in an uncommon etiology of spontaneous retroperitoneal hematoma (SRH) and has been more frequently described in patients on dialysis or anticoagulation.[2] Clinical instability secondary to refractory bleeding for an SRH is a feared progression of the condition and can be fatal if not aggressively managed. Computed tomography (CT) imaging remains the gold standard for diagnosis in the setting of SRH, and renal angiography and endovascular coiling are vitals tools in stabilizing an actively bleeding patient with minimal long term complications.

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Case report

A healthy, 22-year-old woman presented to the emergency department (ED) with right-sided flank pain increasing over a 24 hour period. She denied any history of flank trauma or nephrolithiasis. She denied regular use of any prescription medications. She also denied any personal or familial history of any connective tissue disorders. The patient denied fevers, dysuria, hematuria, or signs of systemic infection. Laboratory testing revealed anemia (hgb 9.6 g/dL) and a mildly elevated white blood cell count (13,200/µL). CT scan with intravenous contrast revealed a large, right-sided retroperitoneal hematoma with no identifiable etiology, including adrenal or renal malignancy, Figures 1a and 1b. The patient was admitted to the hospital for pain control and observation. Her anemia remained refractory to multiple blood transfusions. The computed tomography (CT) scans were reviewed with the radiology team, and in the absence of an identifiable renal or adrenal mass, the decision was made to perform angiography to identify the possible source of bleeding. Renal angiography revealed active extravasation of the right inferior adrenal artery, Figure 2a.

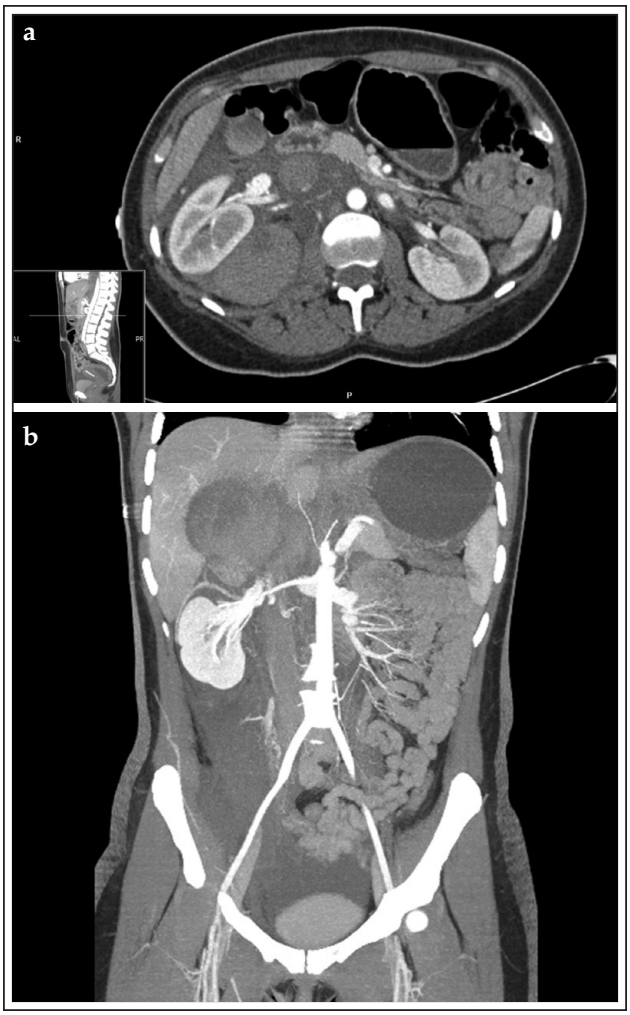

Figure 1. CT with contrast demonstrates a hyperdense fluid collection in the retroperitoneum in the area of the right adrenal gland displacing the kidney anteriorly and inferiorly as seen in both the axial **(a)** and coronal **(b)** views.

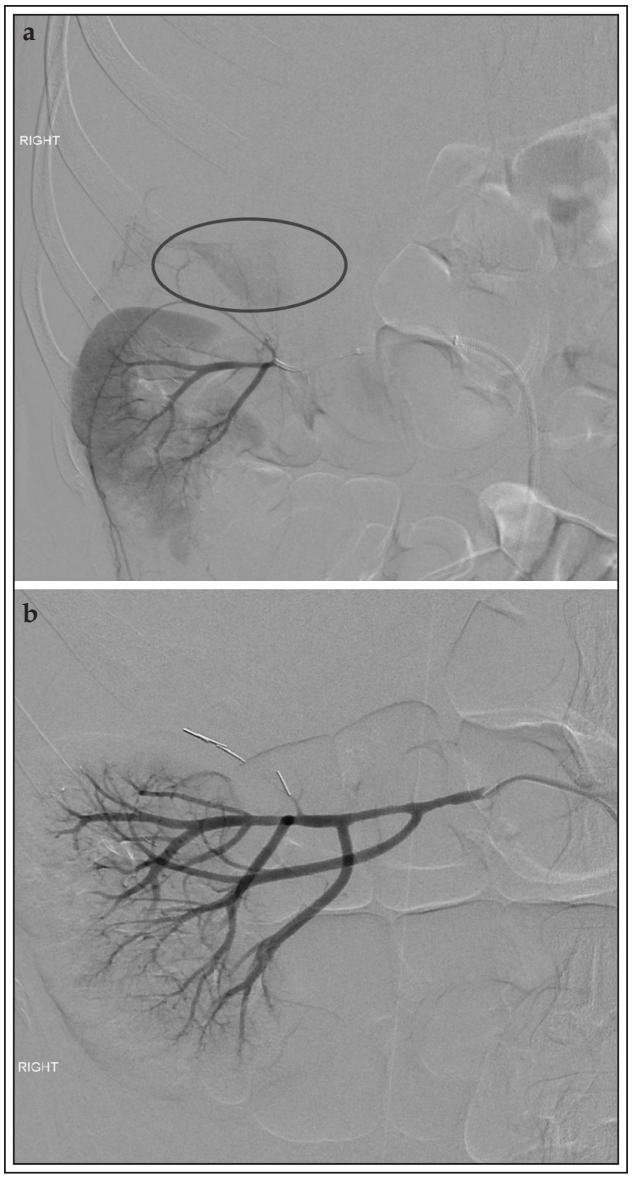

Figure 2. Angiography performed revealing active extravasation consistent with bleeding from the inferior adrenal artery **(a)**. After successful embolization with endovascular coils **(b)**, no more extravasation is noted with continued perfusion of the right kidney.

Coil embolization was performed on the right inferior adrenal artery with preservation of the renal vasculature, Figure 2b. The patient stabilized following coil embolization and was discharged after 3 days of observation. Repeat abdominal imaging with CT scans was performed at 3 to 6 month intervals over the next 24 months revealing continued resolution of the retroperitoneal hematoma, Figure 3. The patient remains asymptomatic 2 years later.

Discussion

SRH is a rare, yet potentially lethal condition with a higher prevalence in elderly patients and those on anticoagulation therapies or dialysis.[2] In 89 patients with SRH, Sunga et al noted a median age at diagnosis of 72 years old with 66.3% of the patients taking anticoagulation therapy.[3] Hemorrhage from malignancy is another well-known cause of SRH. In another series of patients with SRH, renal or adrenal malignancy was noted in a quarter of cases, highlighting the importance of excluding malignancy as a cause of SRH.[4] Spontaneous rupture of the adrenal artery is a rare cause of SRH.[5] Adrenal artery aneurysms are proposed to be an etiology for spontaneous rupture and

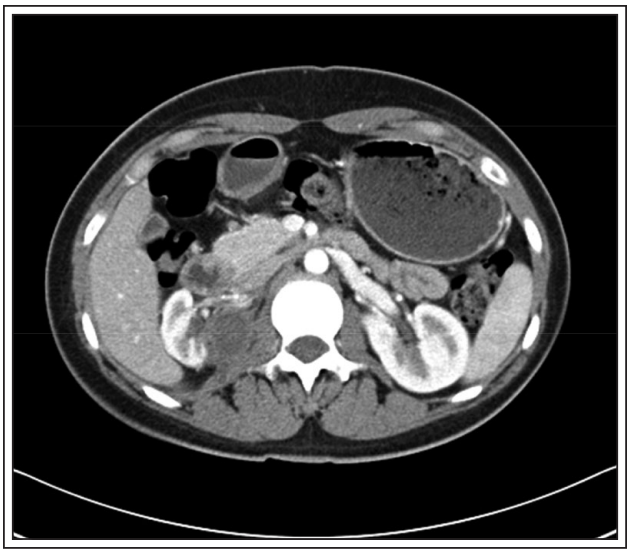

Figure 3. CT with contrast revealing a resolving hematoma in the area of the right adrenal gland 2 years later with normal kidney perfusion.

have been described in 10 cases previously.[6] Although the exact etiology of rupture is not known, congenital malformation of the artery or disease processes that impair the integrity of the arterial wall have been implicated such as atherosclerosis, connective tissue disorders, inflammation of nearby viscera, and infective conditions. One condition associated with spontaneous arterial hemorrhage includes patients with Ehler-Danlos syndrome (EDS), which consists of a heterogenous group of connective tissue disorders. The arterial complications of type IV EDS involve spontaneous rupture of major arteries, aneurysm formation with subsequent rupture, and dissection. The vessels most commonly affected include the aorta, and the carotid, vertebral, popliteal, iliac, and femoral arteries. Spontaneous rupture of nonaneurysmal arteries is a classic vascular complication in this syndrome and occurs more frequently than aneurysm formation or dissection. Although no family history is noted by our patient, patients with type IV EDS are frequently unaware of the disorder until a catastrophic rupture of an artery or the bowel occurs, and one should have a high clinical suspicion as the diagnosis is often a clinical one.[7]

Most patients with SRH will present with abdominal or flank pain as a presenting symptom.[3] Other signs and symptoms include hip pain, leg pain, leg paresthesias, nausea, vomiting, syncope, pallor, or altered mental status. CT imaging of the abdomen and pelvis has become the gold standard of diagnosis with extremely high sensitivity and specificity.[3,4,8] It is important to have a high index of suspicion when working up a patient with SRH, as the mortality can be as high as 5% within 7 days, 10% at 30 days, and 19% at 6 months.[3] These mortality statistics may be better reflective of an older, more infirmed population unlike the young woman we present above; however, extreme care and vigilance should remain standard for any patient with a SRH, given the tenuous nature of such a bleed. Most often, these patients can be managed conservatively with observation or endovascular intervention, as surgical exploration can lead to worsening hemorrhage and visceral injury.[9] Urgent embolization should be performed in cases of arterial bleeding or contained vascular injuries supplying the retroperitoneal hematoma. Surgical intervention should be reserved for cases refractory to initial measures of stabilization.

Conclusion

SRH is a rare yet well-recognized clinical entity with a variety of known etiologies. Spontaneous adrenal artery rupture is a rare but possible etiology of SRH. In patients refractory to standard resuscitative efforts, interventional angiography should be utilized to aid in both the diagnosis and treatment of SRH secondary to vascular hemorrhage. Follow up imaging should be performed to rule out malignancy as a potential cause of hemorrhage. □

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