Case Reports in Women's Health 32 (2021) e00357

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

Hematuria in pregnancy due to renal arteriovenous malformation: A case report

Ashie Kapoor *, Leo Yamaguchi, Sherwin Azad, Yi McWhorter

HCA Healthcare, MountainView Hospital, 3100 N Tenaya Way, Las Vegas, NV, USA

ARTICLE INFO

Keywords:
Renal arteriovenous malformations
Pregnancy
Hematuria
Digital subtraction angiography (DSA)
Emergent cystoscopy
Coil embolization

ABSTRACT

A 31-year-old pregnant woman at 22 weeks and 2 days of gestation presented to the emergency room with complaints of painless hematuria and passage of clots. Initial computed tomography angiography (CTA) of the abdomen and pelvis performed after ultrasound revealed evidence of blood products in the bladder. However, the CTA did not reveal any source of bleeding. Given hemodynamic instability and persistent pain, the patient was taken to the operating room for a cystoscopy, which revealed bleeding from the left renal unit, giving rise to suspicion of a renal arteriovenous malformation (AVM). The patient then underwent left renal digital subtraction angiography (DSA), which produced no evidence of active bleeding. Due to high clinical suspicion and ongoing symptomatic hematuria, she underwent DSA a second time, which did demonstrate renal AVM bleeding, and embolization was performed. This case highlights the importance of cystoscopy in diagnosing a renal AVM in a pregnant patient despite the risks of general anesthesia during pregnancy.

1. Introduction

Renal arteriovenous malformations (AVMs) are abnormal communications between the intrarenal arterial and venous systems with a vascular nidus. They are typically congenital and have an estimated incidence of 0.04% in the general population and less than 0.04% in pregnant patients [1,2]. Most occur in the first trimester of gestation and present in the third to fourth decade of life. Overall, there is a threefold greater incidence in women than in men, and right kidneys are more commonly affected than left [3].

Although the most common presenting symptom is hematuria [4], life-threatening bleeding can persist even in small, peripherally located AVMs due to rupture of the high-pressure, thin-walled veins that drain into the collecting system. Among modalities to evaluate AVMs, digital subtraction angiography (DSA) with iodinated contrast is the gold standard [5]. During pregnancy, though the preferred diagnostic modalities are Doppler ultrasonography (US) and magnetic resonance imaging (MRI), radiographic studies and associated contrast should not be withheld from a pregnant patient if clinically indicated, according to the American College of Obstetricians and Gynecologists (ACOG) [6].

Here we present a difficult and rare case of renal AVM in a second-trimester pregnant patient whose clinical course was dependent on multiple diagnostic studies and an operating room procedure that led to accurate diagnosis and definitive treatment.

2. Case presentation

A 31-year-old pregnant woman (G2P0010) presented at 22 weeks and 2 days of gestation to the emergency room with a complaint of painless hematuria and passage of clots for one day. She was previously healthy, without any past medical or surgical history. In the emergency department, she was afebrile with blood pressure 101/66 mmHg, pulse rate of 109 beats/min, and oxygen saturation of 97%. Her initial physical exam revealed bladder distention without flank tenderness. A temporary indwelling urinary catheter was promptly inserted and drained 800 mL of urine with gross hematuria and large clots. A renal US scan showed significant bladder distention with echogenic material within the urinary bladder, likely indicating blood products. Her initial laboratory finding was significant for hemoglobin of 11.0 g/dL, which subsequently dropped to 8.7 g/dL after a few hours. An emergent

Abbreviations: ACOG, American College of Obstetricians and Gynecologists; AVM, arteriovenous malformation; CTA, computed tomography angiography; DSA, digital subtraction angiography; ICU, intensive care unit; IR, interventional radiology; MRA, magnetic resonance angiography; MRI, magnetic resonance imaging; US, ultrasound.

* Corresponding author at: 2880 N Tenaya Way, 2nd Floor, Las Vegas, NV 89128, USA.
E-mail address: kapoorashie@gmail.com (A. Kapoor).

https://doi.org/10.1016/j.crwh.2021.e00357
Received 9 September 2021; Received in revised form 12 September 2021; Accepted 13 September 2021
Available online 16 September 2021

urology consultation was obtained from the emergency department, and the patient was admitted to the intensive care unit (ICU) for further evaluation and management.

In the ICU, the patient developed severe abdominal pain. Her diagnostic study was changed from magnetic resonance angiography (MRA) to emergent computed tomography angiography (CTA) of the abdomen and pelvis as she was unable to lay still due to severe pain. This exam again noted a large (11x9x9cm) intraluminal hyperdense mass in the urinary bladder, consistent with hemorrhagic products and clot, yet no other abnormality or source of bleeding was identified. At this time, intravenous opiate medications were insufficient to control her pain caused by severe bladder distention and retained blood clots. The urologist performed manual irrigation and evacuation at the bedside, followed by continuous bladder irrigation. Even though there was substantial pain relief, hematuria persisted, with ongoing tachycardia and hypotension. The patient was resuscitated with intravenous fluids and packed red blood cells, and taken to the operating room for emergent cystoscopy under general anesthesia.

Intraoperatively, intermittent bleeding was noted from the left renal unit, although no source of bleeding was identified. The urologist suspected that a renal AVM could be the culprit of bleeding and consulted interventional radiology (IR) for diagnostic angiography with possible embolization. The patient underwent left renal digital subtraction arteriography (DSA), but no active bleeding, AVM, or other treatable lesions were identified. During the next 48 h, she continued to have transfusion-dependent anemia and was scheduled for MRA of the abdomen and pelvis. However, after an episode of frank hematuria, emergent left renal DSA was performed again, and it demonstrated a small focal vascular malformation characterized by an abnormal nidus of vessels in close relation to a left upper pole interlobar artery (Fig. 1). There was no obvious premature filling of the venous system that would be typical of an AVM. Coil embolization was carried out using 5 mm, 3 mm, and 2 mm detachable platinum coils, which were delivered via a microcatheter, beginning with embolization of the anterior superior segmental branch of the renal artery. Post-embolization angiography demonstrated no filling within the nidus and resolution of contrast extravasation (Fig. 2).

After embolization, the patient did not experience any further episodes of frank hematuria, though old clots continued to clear from the left renal collecting system. Her hemoglobin remained stable, and she did not require any further blood transfusions. She recovered well and was discharged on hospital day seven. The patient returned at 37 weeks and 5 days of gestational age for a normal spontaneous vaginal delivery of a healthy newborn weighing 2950 g (69th percentile) with APGAR scores of 8 and 9 at 1 and 5 min, respectively.

3. Discussion

Idiopathic renal AVMs in pregnancy represent rare and challenging cases for the multidisciplinary team. As the primary ICU team who coordinated this patient's care, we encountered numerous obstacles when collaborating with other services during her rapidly changing clinical course. First, we considered an MRA of the abdomen and pelvis as the initial diagnostic test to detect the area of bleeding. However, as the blood clots accumulated in her bladder, she became progressively more agitated due to severe pelvic pain and was unable to lay still. In addition, she had labile vital signs, which made prolonged remote monitoring in MRA difficult and potentially unsafe. We then proceeded with CTA, with the lowest allowable intravenous contrast. It was unclear whether the low contrast dose or intermittent bleeding was the cause of the negative study in this case. According to the ACOG 2017 update, US and MRI are preferred over CT as the latter exposes the fetus to ionizing radiation. The American College of Radiology also states that MRIs are safe in all trimesters, with gadolinium being the preferred type of contrast [6,7]. We, therefore, considered MRA over CTA for our pregnant patient. However, when her clinical condition changed, we carefully balanced the benefit of quick diagnostic yield against potential teratogenicity in the context of ongoing hemodynamic instability.

Next, the decision to proceed with operative management is necessary if a surgical disease is identified. In this case, cystoscopy retrospectively offered invaluable information in the patient's care. Emergent cystoscopy under general anesthesia during pregnancy carries additional risks to the mother and fetus. One retrospective study demonstrated that non-obstetric surgeries and general anesthesia during pregnancy were more likely to lead to preterm births and low birth weight, though causation cannot be concluded [8]. Though the second trimester is generally deemed the safest period for receiving general anesthesia, longer, more frequent, and higher-dosage anesthetics are considered to increase the risk of fetal neurotoxicity [9]. Despite these potential harms, life-saving procedures for pregnant patients with surgical disease should not be delayed based on recommendations from the US Food and Drug Administration and ACOG [10]. In this patient, cystoscopy was paramount in identifying the source of bleeding in her left kidney, and the next appropriate referral to IR was made to pursue further diagnostic imaging and definitive treatment. Thorough and informative consultation with the patient regarding the risks and benefits of cystoscopy under general anesthesia during pregnancy is required in patient-centered care.

Eventually, we referred her a second time for DSA based on highly suspicious renal AVM bleeding from her left kidney despite the initial negative DSA. We propose several technical factors which were intended to reduce radiation exposure but which may have contributed to the initial negative DSA: contrast injection rate, injection volume, injection bolus timing, fluoroscopic views (oblique vs. orthogonal), image

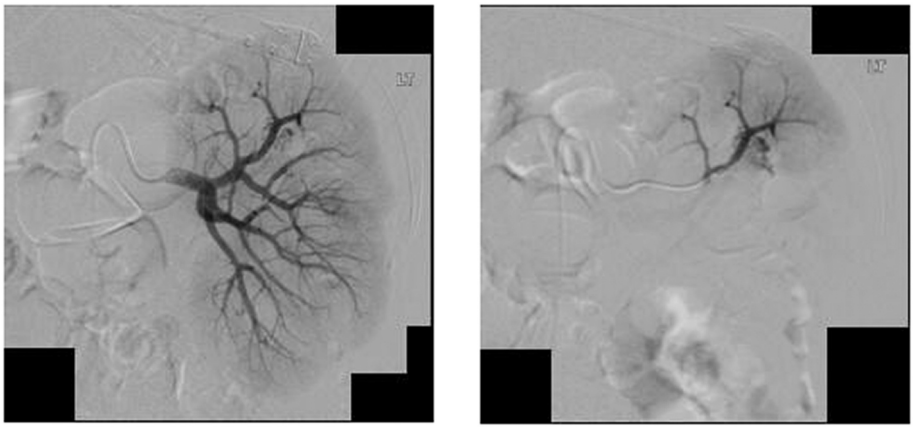

Fig. 1. A. Selective catheterization and digital subtraction angiography of the left renal artery, angiogram in arterial phase, demonstrates a renal arteriovenous malformation arising from the anterior superior segmental branch characterized by a nidus of small vessels projecting inferiorly. **B.** Superselective catheterization and digital subtraction angiography. This angiogram re-demonstrates the arteriovenous malformation and confirms arterial inflow via feeders arising from the anterior superior segmental branch of the left renal artery. Prominent contrast blush and extravasation are seen, consistent with active hemorrhage.

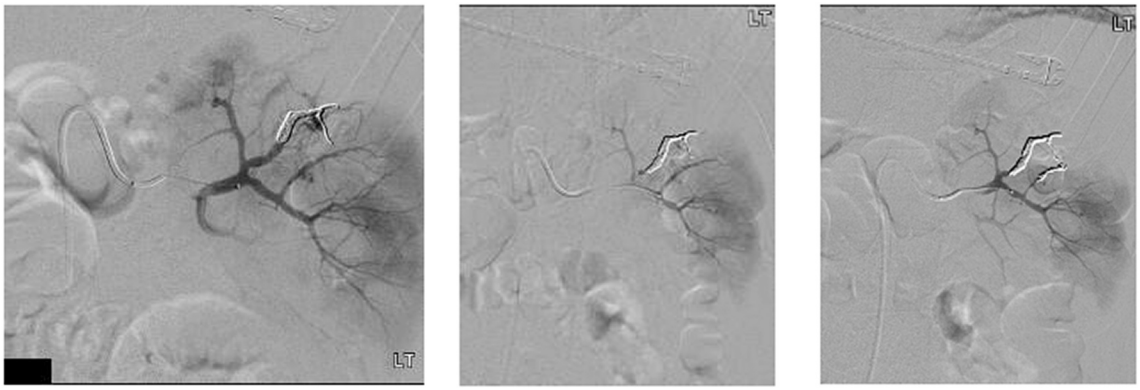

Fig. 2. A. Coil embolization of the anterior superior segmental branch of the left renal artery was performed utilizing 5 mm and 3 mm detachable platinum microcoils. **B.** Super-selective catheterization and digital subtraction angiography of the anterior inferior segmental branch demonstrates several small-caliber arterial feeders arising from an interlobar artery supplying the nidus. **C.** Coil embolization of the interlobar artery was performed utilizing 2 mm detachable platinum microcoils. Follow-up super-selective digital subtraction angiography of the anterior inferior segmental branch following embolization demonstrates no residual filling within the nidus. Extravasated contrast is seen outlining an associated renal calyx.

processing, magnification, and exposure. Patient- or disease-specific factors may also have contributed, including the presence of active hemorrhage, degree of vasodilation, and small size of the lesion. We chose to monitor the patient in the ICU setting due to a high index of suspicion for intermittent bleeding from renal AVM. We planned for follow-up MRA 48 h after her initial bleeding; however, it changed to repeat DSA after a multidisciplinary discussion was held when she had sudden clinical decline with another episode of aggressive hematuria. Bleeding was identified during the repeat DSA, likely due to the enhanced rate and volume of hemorrhage. We learned to adjust management strategies based on the rapidly changing clinical course in this challenging case of idiopathic renal AVM. Cystoscopy proved to be an essential component in this case as it supported a high suspicion for the presence of renal AVM despite the negative initial DSA, thus, justifying repeat DSA as the necessary life-saving procedure.

In conclusion, renal AVMs are exceedingly rare in the obstetric population. Accurate diagnosis and prompt treatment are crucial as recurrent, life-threatening hematuria represents a surgical disease that may lead to maternal and fetal endangerment. It is essential to maintain open and clear communication with patients, families, and consultants to coordinate care expediently and effectively. Clinicians need to continuously balance the benefits of prompt maternal treatments against potential fetal harm in challenging cases where surgeries and radiation exposures may be necessary and life-saving.

Contributors

Ashie Kapoor contributed to the case report's conception and design, and drafting and editing of the manuscript.

Leo Yamaguchi contributed to the case report's conception and design, and drafting and editing of the manuscript.

Sherwin Azad contributed to the drafting of the manuscript, and was directly involved in the patient's care.

Yi McWhorter contributed to the case report's conception and design, critically revised the manuscript for important intellectual content, and was directly involved in the patient's care.

All authors saw and approved the final draft of the report.

Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

No funding from an external source supported the publication of this case report.

Patient consent

Obtained.

Provenance and peer review

This article was peer reviewed.

Acknowledgements

The authors would like to acknowledge the involvement of Dr. Victor E. Grigoriev in the clinical care of this patient.

This report was supported (in whole or in part) by HCA Healthcare and/or an HCA Healthcare affiliated entity. The views expressed in this publication represent those of the authors and do not necessarily represent the official views of HCA Healthcare or any of its affiliated entities.

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