AACE Clinical Case Rep. 7 (2021) 72-74

Contents lists available at ScienceDirect

AACE Clinical Case Reports

journal homepage: www.aaceclinicalcasereports.com

Case Report

Vanishing Adrenal Mass in Pregnancy

Sheila R. Gupta, MD¹, Hans K. Ghayee, DO^{2,*}

¹ Division of Endocrinology, Diabetes, & Metabolism, Department of Internal Medicine, Florida State University College of Medicine, Tallhassee, Florida
² Division of Endocrinology, Diabetes, & Metabolism, Department of Internal Medicine, University of Florida College of Medicine and the Malcom Randall VA Medical Center, Department of Internal Medicinology, Diabetes & Metabolism, Gainesville, Florida

ARTICLE INFO

Article history: Available online 27 November 2020

Key words: adrenal pregnancy hemorrhage mass

ABSTRACT

Objectives: The evaluation of an adrenal mass is challenging. We present the case of a 33-year-old pregnant woman who was found to have an adrenal incidentaloma. Four months after the initial imaging, the mass vanished.

Methods: We described the case of a pregnant woman with hypertension and an incidentally found right adrenal mass.

Results: A magnetic resonance imaging scan showed a right adrenal mass measuring $7.9 \times 3.9 \times 3.0$ cm with a multilobulated appearance. Initial biochemical testing was concerning for a pheochromocytoma with positive metanephrines during hospitalization while being treated for an infection. Repeat outpatient adrenal hormone results, including metanephrines, were negative. Four months after her initial magnetic resonance imaging scan, the right adrenal mass was no longer present.

Conclusion: A 33-year-old pregnant woman was found to have a right adrenal mass that later vanished as a result of the resolution of a unilateral adrenal hemorrhage. Predisposing factors to adrenal hemorrhage in the presented case include pregnancy, infection, and hypertension.

Published by Elsevier Inc. on behalf of the AACE. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Adrenal hemorrhage is an uncommon but potentially serious complication during pregnancy. The clinical presentation of adrenal hemorrhage may include abdominal pain, tenderness, fever, hypotension, and shock. Abdominal pain during pregnancy is a common symptom, and a high suspicion of adrenal hemorrhage is crucial. Appropriate imaging techniques are useful for establishing a timely diagnosis. The differential diagnosis and evaluation of adrenal hemorrhage during pregnancy are discussed in the case presented.

Case Report

A 33-year-old primigravida female at 12 weeks' gestation presented to the emergency room with right flank pain radiating to the

E-mail address: hans.ghayee@medicine.ufl.edu (H.K. Ghayee).

right lower quadrant, nausea, and vomiting. She had a history of hypertension diagnosed at age 18, which was well controlled on lisinopril before pregnancy. During pregnancy, her medication was changed to labetalol 200 mg daily, with an excellent response, along with aspirin 81 mg daily. She was afebrile with a white blood cell count of 12×10^3 /mL and a normal platelet count. Urinalysis confirmed a urinary tract infection. Abdominal ultrasound showed no inflammation of the appendix and a normal gallbladder. She was started on intravenous antibiotics initially and then discharged home on oral antibiotics. Over the next few days, her right flank pain continued to intensify, now accompanied by back and chest pain. Her obstetrician ordered a magnetic resonance imaging (MRI) scan of the abdomen and pelvis, which showed a large right adrenal mass measuring $7.9 \times 3.9 \times 3.0$ cm with a multilobulated appearance (Fig. A). The mass demonstrated mild heterogeneous T2 hyperintensity and was grossly isointense relative to the renal parenchyma on T1-weighted images. There was a surrounding T2 signal extending superiorly and posteriorly to the dome of the liver and inferiorly along the retroperitoneum at the midportion of the right kidney, consistent with surrounding inflammation. Due to a rising white blood cell count of 23×10^3 /mL and temperature of 101.0 °F, she was presumptively diagnosed with pyelonephritis, started on intravenous ceftriaxone, and transferred to a tertiary care center for work-up of the adrenal mass.

https://doi.org/10.1016/j.aace.2020.11.020







Abbreviations: ICU, intensive care unit; MRI, magnetic resonance imaging. * Address correspondence and reprint requests to Dr Hans Ghayee, Division of Endocrinology, Diabetes, & Metabolism, Department of Internal Medicine, University of Florida College of Medicine and the Malcom Randall VA Medical Center, 1600 SW Archer RD, Suite H-2, Gainesville, FL 32608.

^{2376-0605/}Published by Elsevier Inc. on behalf of the AACE. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/ 4.0/).



Fig. A, Red arrow showing right adrenal mass and white arrow showing normal left adrenal gland. B, red arrow showing resolution of the mass and normal right adrenal gland and white arrow showing normal left adrenal gland.

Upon admission to the intensive care unit (ICU), she had a temperature of 99 °F, a pulse of 79 bpm, and blood pressure of 147/81 to 166/97 mm Hg. Several days later, her systolic blood pressure reduced to the 130s. Laboratory evaluation showed a potassium level of 3.4 mmol/L, creatinine of 0.47 mg/dL, aspartate aminotransferase of 12 IU/L, alanine aminotransferase of 8 IU/L, and alkaline phosphatase of 55 IU/L. consistent with normal renal and liver function. The aldosterone level was 9.1 ng/dL (nonpregnant reference, <31.0 ng/dL), plasma renin activity was 2.9 ng/mL/h (normal, 0.5-4.0 ng/ml/h), and random cortisol level was 17.2 µg/dL (normal, 7.0-23.0 µg/dL). A 24-hour urine free cortisol was 43.6 µg/ 24 h (normal, <45.0 µg/24 h). Plasma metanephrine was 0.82 nmol/ L (normal, 0.00-0.49 nmol/L), and plasma normetanephrine was 1.50 nmol/L (normal, 0.00-0.89 nmol/L). Her 24-hour urine metanephrine was 656 μ g/d (normal, 39-143 μ g/d) and 24-hour urine normetanephrine was 1031 µg/d (normal, 109-393 µg/d). After several days of ceftriaxone in the hospital, the patient was discharged on cefdinir. Her pyelonephritis subsequently resolved. She was scheduled with endocrinology and general surgery for possible surgical removal of the right adrenal mass pending a repeat biochemical work-up as an outpatient. Given that the imaging findings were inconsistent with a typical adrenal adenoma, the differential diagnosis included pheochromocytoma, adrenal cortical carcinoma, or adrenal hemorrhage.

Her outpatient work-up showed normal 24-hour urine fractionated metanephrines: 24-hour urine metanephrine was 85 µg/d (normal, 36-190 µg/day), 24-hour urine normetanephrine was 415 µg/day (normal, 35-482 µg/day), and 24-hour urine total metanephrines were 500 µg/day (normal, 115-695 µg/day). Plasma fractionated metanephrines showed free metanephrine of <25 pg/mL (normal, <57 pg/mL) and plasma-free normetanephrine of 49 pg/mL (normal, <148 pg/mL). Her androstenedione level was 160 ng/dL (normal 51-213 ng/dL), chromogranin A was 31 ng/mL (normal, 21-106 ng/mL), and 11-deoxycortisol level was 29 ng/dL (normal, <46 ng/dL). A repeat MRI 1 month after initial imaging showed a significantly decreased size of the right adrenal mass, measuring 3.3×1.9 cm. The multilobulated appearance was no longer present, and the adrenal mass had a smooth fusiform shape. The extensive edema in the right retroperitoneum had resolved. The mass had a significantly increased T1 signal, consistent with an evolving adrenal hemorrhage.

MRI repeated four months after initial imaging showed no masses on the adrenal glands. There was complete resolution of the previously seen T1 and T2 hyperintense mass within the right adrenal gland (Fig. *B*). Although the patient's blood pressure was well controlled throughout the majority of her pregnancy, after 25 weeks' gestation, her blood pressure remained high despite escalating doses of blood pressure medications. She delivered a baby girl by cesarean birth at 26 weeks due to eclampsia. The patient continued on labetalol 600 mg twice a day and nifedipine 90 mg daily after delivery. The baby girl was born at 1 pound 6 ounces and discharged home after 10 weeks in the neonatal ICU.

Discussion

A case of a 33-year-old pregnant woman found to have a right adrenal mass is presented. She developed this unilateral adrenal mass as a result of adrenal hemorrhage. The hemorrhage may have been caused by a combination of factors, such as pregnancy and infection. Unilateral adrenal hemorrhage in pregnancy is uncommon but has been recognized and reported in the literature.¹⁻⁷ The exact incidence rate of adrenal hemorrhage in pregnancy is unknown and often involves the right adrenal gland.^{7,8} Adrenal hemorrhage may be spontaneous or result from contributing factors, such as pregnancy, infection, anticoagulation, hypertension, tumor, or trauma.⁷ The vascular supply of the adrenal gland makes it particularly susceptible to hemorrhage. Any increase in adrenal perfusion pressure or adrenal venous pressure may cause hemorrhage into the gland. In addition, hemodynamic changes during pregnancy contribute to vulnerability toward adrenal hemorrhage. Increased cardiac output and blood volume, along with decreased vascular resistance, increase the blood flow into the adrenal glands.^{6,8} Several reported cases of unilateral adrenal hemorrhage during pregnancy involved the right adrenal gland,⁴⁻⁷ which may be due to the anatomical proximity of the right adrenal gland to the inferior vena cava.

The patient's elevated plasma and urine-fractionated metanephrines while hospitalized were due to the body's stress response to either the acute infection with pyelonephritis or the adrenal hemorrhage itself. Plasma metanephrine and normetanephrine can be elevated in stressful situations, like an ICU admission, and therefore, results should be interpreted with caution. Although the patient's hypertension was well controlled prior to her acute illness, her elevated blood pressure at the time of presentation also contributed to her risk of adrenal hemorrhage by increasing arterial flow to the adrenal glands. An underlying adrenal tumor is a relatively common etiology of adrenal hemorrhage. Benign lesions, including myelolipomas and hemangiomas larger than 10 cm, may spontaneously bleed.⁹ Pheochromocytoma and adrenal cortical carcinoma are also associated with adrenal hemorrhage.

Anticoagulation is also associated with nontraumatic adrenal hemorrhage. The patient was taking baby aspirin, consistent with the current American College of Obstetrics and Gynecology guidelines.¹⁰ While all anticoagulants can increase bleeding risk. the classic anticoagulant linked to adrenal hemorrhage is heparin. There have been several reports of bilateral massive adrenal hemorrhage in the setting of heparin-associated thrombocytopenia.¹¹ Adrenal hemorrhage also occurs due to trauma.¹² Possible mechanisms for traumatic injury to adrenal glands are direct compression of the adrenal gland between the spine and liver or spleen or a rise in venous pressure due to compression of the inferior vena cava. Imaging modalities to detect adrenal hemorrhage include ultrasound, computed tomography, and MRI. Our patient's initial MRI showed findings of an isointense T1-weighted image and hyperintense T2-weighted image. Initially, in the first week of a bleed, an adrenal hematoma will appear isointense on T1 images and hypointense on T2 images due to a high concentration of intracellular deoxyhemoglobin. As hemoglobin oxidizes, the hematoma becomes hyperintense on T1- and T2-weighted images. After a week, high signal intensity starts at the periphery and then fills in over weeks. A chronic adrenal hemorrhage will have a hypointense rim on T1-and T2-weighted images due to hemosiderin deposition.¹³ Our patient's MRI showed that the T1 signal progressed from isointense on the first image to hyperintense on the second image, with a significant decrease in size of the mass. consistent with adrenal hemorrhage. Four months after her initial presentation, the patient's MRI showed complete resolution of the hematoma.

The presentation of adrenal hemorrhage can vary from an incidental finding by abdominal imaging to hemorrhagic shock and adrenal insufficiency. A conservative approach with supportive care is generally favored when possible. In more severe cases, surgical intervention, including arterial embolization, adrenal repair, adrenalectomy, or early delivery, has been reported.⁹ Assessment of adrenal function and replacement with steroids are paramount in cases of adrenal hemorrhage.

Conclusion

Unilateral adrenal hemorrhage during pregnancy is uncommon. Predisposing factors to adrenal hemorrhage in our pregnant patient included acute stress from infection and hypertension. MRI is beneficial in diagnosis and guidance in the management of adrenal hemorrhage. Screening for adrenal hormone function is important in the work-up of adrenal hemorrhage due to the potential for adrenal insufficiency. Early diagnosis and treatment with steroid replacement therapy (if needed) improves survival.

Disclosure

The authors have no multiplicity of interest to disclose.

References

- 1. Gupta A, Minhas R, Quant HS. Spontaneous adrenal hemorrhage in pregnancy: a case series. *Case Rep Obstet Gynecol*. 2017;2017:3167273.
- Kadhem S, Ebrahem R, Munguti C, Mortada R. Spontaneous unilateral adrenal hemorrhage in pregnancy. *Cureus*. 2017;9(1):e977.
- Yang L, Zhu YC, Liu RB. Spontaneous adrenal hematoma in pregnancy. A case report. *Medicine (Baltimore)*. 2018;97(47), e13329.
- Arnold G, Richer AG, Lepore JJ. Suprarenal hemorrhage in pregnancy; report of a case with a review of the literature. N Engl J Med. 1949;240(26):1040–1043.
- Shen JZ, Stranks SN, Ho JT. Spontaneous adrenal haemorrhage in pregnancy. Intern Med J. 2011;4(41):362–363.
- Anagnostopoulos A, Sharma S. Spontaneous adrenal haemorrhage in pregnancy. BMJ Case Rep. 2011;2011, bcr0720114496.
- Gavrilova-Jordan LP, Edmister WB, Farrell MA, Watson WJ. Spontaneous adrenal hemorrhage during pregnancy: a review of the literature and a case report of successful conservative management. Obstet Gynecol Surv. 2005;60(3):191–195.
- Rafi J, Khalil H. Maternal morbidity and mortality associated with retroperitoneal haematomas in pregnancy. JRSM Open. 2018;9(1):1–9.
- Simon DR, Palese MA. Clinical update on the management of adrenal hemorrhage. Curr Urol Rep. 2009;10(1):78–83.
- ACOG Committee Opinion No. 743: low-dose aspirin use during pregnancy. Obstet Gynecol. 2018;132(1):e44–e52.
- Vella A, Nippoldt TB, Morris III JC. Adrenal hemorrhage: a 25-year experience at the Mayo Clinic. Mayo Clin Proc. 2001;76(2):161–168.
- Lehrberg A, Kharbutli B. Isolated unilateral adrenal gland hemorrhage following motor vehicle collision: a case report and review of the literature. *J Med Case Rep.* 2017;11(1):358.
- Kawashima A, Sandler CM, Ernst RD, et al. Imaging of nontraumatic hemorrhage of the adrenal gland. *Radiographics*. 1999;19(4):949–963.