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Case Report

Idiopathic unilateral adrenal hemorrhage in a term pregnant primigravida female

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ABSTRACT

Idiopathic or spontaneous adrenal hemorrhage is a rare entity and a serious medical condition which is usually underestimated but can result in multiorgan failure, hemodynamic instability and death. It is usually diagnosed postmortem, as it has a nonspecific presentation in view of some other concurrent illness. Pregnancy-induced adrenal hemorrhage in itself is quite rare and a poorly understood disorder. We report here a case of 24-year-old primigravida female at 38 + 6 weeks of gestation who presented with right loin pain associated with fever and chills. Abdominal ultrasound and magnetic resonance imaging were performed which revealed presence of hematoma in right suprarenal location. A diagnosis of idiopathic right adrenal hemorrhage was made. The patient was managed conservatively and was stable post delivery. As this condition can have complications which can potentially be life-threatening, clinicians should keep a high suspicion for this disease when a pregnant female presents with acute abdominal pain.

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Introduction

Abdominal pain is one of the common symptoms in pregnancy which can have variable causes; adrenal hemorrhage is a rare cause which is usually not kept high in the list of differential diagnoses. Blunt abdominal trauma and adrenal tumors are among the common etiological factors of unilateral adrenal hemorrhage. Idiopathic/spontaneous adrenal hemorrhage (SAH) is an uncommon cause of acute abdominal pain in pregnancy. It occurs with negative history of trauma or with

the use of anticoagulants. Autopsy reports have revealed incidence of adrenal hemorrhage between 0.03% and 1.8% in unselected cases, however the exact incidence among pregnant women is unknown [1].

The common presentation of adrenal hemorrhage are non-specific pain located in the flanks/back and fever, while some patients may be asymptomatic [2]. Mostly it is the bilateral idiopathic adrenal hemorrhage which leads to adrenal crisis and shock which can necessitate emergency adrenalectomy [2,3]. We present a case report in which the patient comes to obstetric department with an acute abdominal pain; imaging

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showed SAH involving the right adrenal gland. The case was managed conservatively.

Case report

A 24-year-old primigravida female at 38 + 6 weeks of gestation, married for 1 year with spontaneous conception, presented with 12-hour history of right loin pain associated with right shoulder pain, 24-hour history of inability to void urine and 1 day history of fever with chills. The pain was constant and radiating to back. Past medical/surgical history did not reveal anything significant. Antenatal ultrasound scans were normal with no gross congenital malformations. An examination revealed increased blood pressure (156/90 mm Hg) and high pulse rate (96 beats per minute). Fever was low grade (100–101°C). Rest of the vitals were normal. No previous history of raised blood pressure records was there during the entire pregnancy and the patient was not on any anti-hypertensive medication.

Complete blood count of the patient was done (Hemoglobin – 8.9 g/dl, Total Leucocyte Count – 16,200/ml, Platelet count – 194,000/ml). The kidney function tests (Urea – 41 mg/dl, Creatinine – 1.2 mg/dl), liver function tests (bilirubin – 0.5 mg/dl, ALT – 25.7U, AST – 43U) and comprehensive metabolic panel (Na⁺ – 134 mEq/L, K⁺ – 4.4 mmol/L, Cl⁻ – 109 mEq/L) were normal. The 24-hour urine albumin was raised (2+). Arterial blood gas parameters of the patient were within the normal range. Urine and blood cultures were sterile. Coagulation parameters of the patient were normal (APTT – 28.3 seconds, PT – 13 seconds, PTI – 100%, INR – 0.95). An abdominal ultrasound was performed which revealed 10 x 8 x 6 cm heteroechoic fluid collection in right perinephric space extending to right suprarenal location. Diagnostic tapping under ultrasound guidance from this collection revealed thick clotted blood, so a possibility of right perinephric/adrenal hematoma was kept. Because of gestation, the patient underwent noncontrast abdominal magnetic resonance imaging (MRI) to confirm the diagnosis/rule out other possibilities and to know the extent of the lesion. MRI revealed a well-defined 11 x 9.4 x 7.3 cm T1 heterogeneously hyperintense, T2 iso to hyperintense lesion with T2 hypointense rim in right perinephric location extending to right suprarenal location. The right adrenal gland is not separately visualized from the lesion (Fig. 1). The lesion is compressing the right kidney and displacing it medially and inferiorly.

She underwent caesarian delivery on the second day of her hospitalization due to raised blood pressure records and adrenal hemorrhage. Intraoperatively, hematoma was seen in retroperitoneal space, but no active intervention for hematoma was done after taking urology consultation.

In the postoperative period the patient had normal general condition. Patient still complained of abdominal pain till 3 days following delivery. Fever subsided post delivery. Baby was healthy with normal birth weight (3400 g) and normal APGAR scores. The morning nonstress cortisol level was sent on third day following delivery, which was mildly elevated (24.7 µg/dL). The adrenal/perirenal hematoma was stable on follow up ultrasound scans (Fig. 2). She did not require steroids or

any other active intervention for the hematoma and was discharged in stable condition.

Discussion

Idiopathic adrenal hemorrhage is a rare condition which has a nonspecific presentation; severe cases may lead to acute adrenal crisis resulting in shock and death, usually only if both the adrenal glands are affected [4]. The pathophysiology of adrenal hemorrhage in nontraumatic cases is unclear. The usual hypothesis suggested is that in pregnancy, there is associated adrenal cortical hyperplasia, which in turn may predispose to venous congestion and subsequently to hemorrhage [5].

Adrenal hemorrhage occurring during pregnancy can have various causes like spontaneous abortion, twisted ovarian cyst, toxemia of pregnancy, antepartum hemorrhage and coagulation disorders (eg, APLA); these usually cause bilateral adrenal hemorrhage. Adrenal tumors (primary or metastatic) and blunt abdominal trauma are the usual causes of unilateral adrenal hemorrhage. Few rare causes of unilateral adrenal hemorrhage include uncomplicated pregnancy, chronic nonsteroidal anti-inflammatory drug (NSAID) use or NF-1. Only few reports of spontaneous/idiopathic unilateral adrenal hemorrhage have been described, which is even more uncommon during pregnancy.

The patients of adrenal hemorrhage commonly present with fever, abdominal pain and in severe cases hypotension and shock. Ultrasound is the primary imaging modality of choice during pregnancy, while MRI has high sensitivity and specificity for hemorrhage and also to rule out underlying causes like adrenal tumors [6]. Contrast MRI is required to rule out any underlying adrenal mass. Clinical differential diagnosis includes ectopic pregnancy, uterine rupture, abruption placentae, and many other pregnancy unrelated causes (pancreatitis, appendicitis etc). Unilateral adrenal hemorrhage usually does not cause adrenal insufficiency but cases of bilateral adrenal hemorrhage may present with acute adrenal crisis which occurs only when 90% of the adrenal cortex is destroyed. In pregnant patients for diagnosing adrenal insufficiency, the requirements are—an early morning cortisol level of less than 3 µ/dL in the nonstressed state and presentation with typical symptoms [7].

We reviewed 5 case reports [8–12] and a case series (with 2 cases) [13] for SAH in pregnancy. Most of these presented in third trimester. Most of these presented with acute symptoms in the form of severe abdominal pain for few days. On imaging evaluation, most of these cases had unilateral adrenal hemorrhage. Cortisol level for diagnosing adrenal insufficiency was available in 6 out of 7 of these cases; insufficiency was diagnosed in all cases of bilateral adrenal hemorrhage and one case of unilateral adrenal hemorrhage. Coagulation parameters were done in all of these cases and none revealed any coagulopathy.

Patients are usually conservatively managed in the form of steroid therapy, fluid resuscitation and correction of underlying coagulopathies if present. Preterm delivery and emergency adrenalectomy may be warranted in hemodynamically

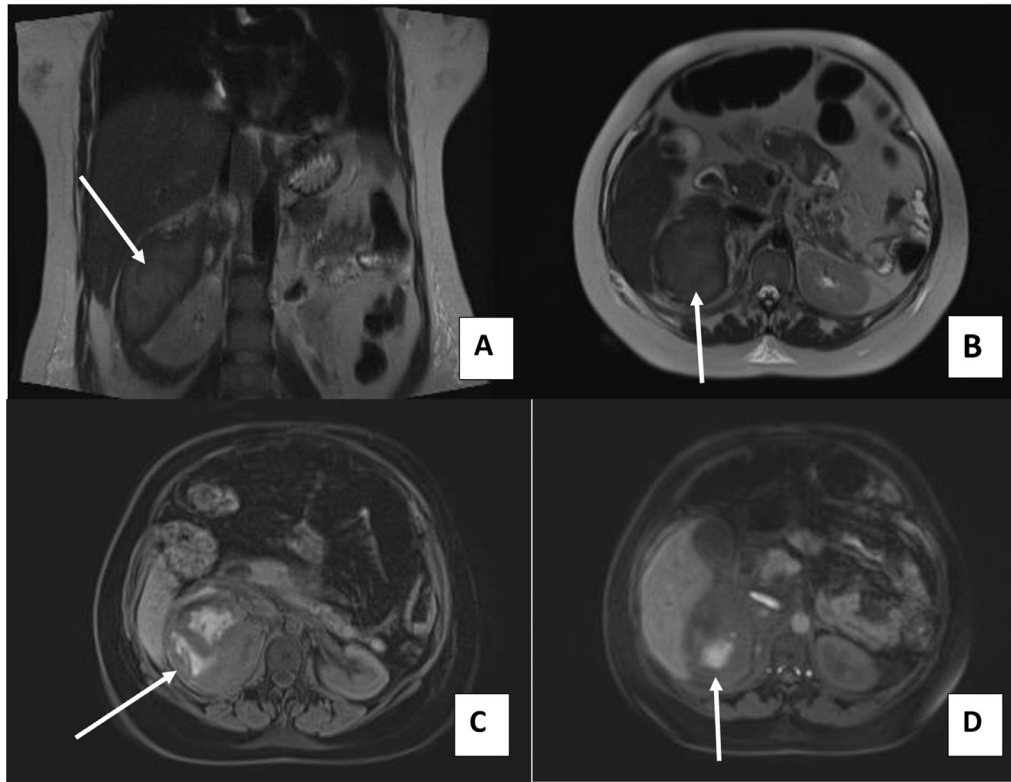


Fig. 1 – Noncontrast MRI of the patient. (A) T2 weighted coronal and (B) T2 weighted axial images showing iso to hyperintense lesion with hypointense rim in right perinephric location extending to right suprarenal location (as depicted by the solid arrow), (C) and (D) T1 weighted axial images showing heterogeneously hyperintense lesion in right suprarenal location (as depicted by the solid arrow). Right adrenal gland is not separately visualized.

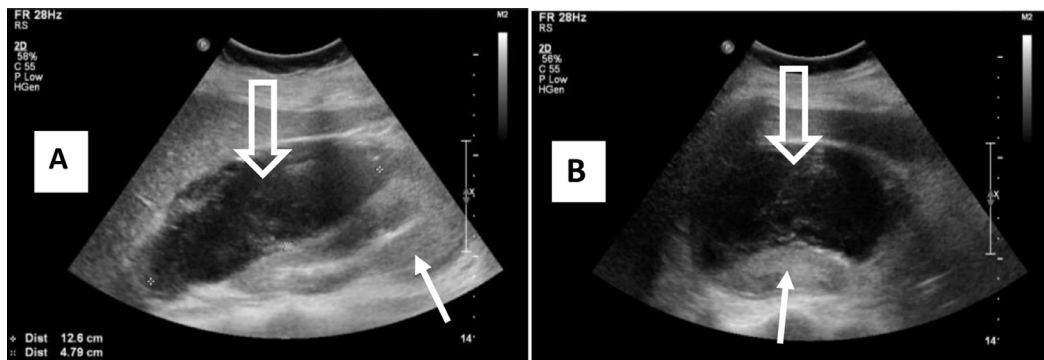


Fig. 2 – Follow up ultrasound images of this case. (A) Image in longitudinal plane showing a well-defined 12.6 x 4.8cm heterogeneously hypoechoic lesion (transparent white arrow) seen in right suprarenal location suggestive of residual hematoma (solid white arrow representing right kidney), (B) The same findings in transverse plane image.

unstable patients with massive hemorrhage. To prevent circulatory collapse, adrenal insufficiency should be addressed promptly [9]. Arterial embolization of adrenals may be needed as a bridging therapy for subsequent surgery in severe hemorrhage cases. In our reviewed 7 case reports [8–13], all the cases were managed conservatively; none required surgery for adrenal hemorrhage. The cases with adrenal insufficiency were managed with steroids. Perinatal mortality was not reported in any case.

Conclusion

Idiopathic unilateral adrenal hemorrhage is an unusual complication during pregnancy which can have serious consequences. Clinicians should have high suspicion for this entity in pregnant patients presenting with features of abdominal or flank pain and fever. Conservative management and steroid replacement usually suffices in most cases.

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