BRITISH JOURNAL OF HOSPITAL MEDICINE

CASE REPORT

A Rare Condition: Nonhemorrhagic Infarct of Adrenal Gland in Pregnancy

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Abstract

Adrenal infarction can be hemorrhagic or non-hemorrhagic, while the latter is much less common. Non-hemorrhagic adrenal infarction (NHAI) is a rare but potentially serious cause of acute abdominal pain in pregnancy that can lead to significant adverse outcomes for both mother and baby if not treated promptly and appropriately. Diagnosis of this condition is challenging due to the non-specific nature of clinical and laboratory findings. Ultrasound (US) has limited utility in diagnosing retroperitoneal pathology, while the use of computed tomography (CT) is restricted due to concerns about fetal radiation exposure. Diagnosis is typically based on magnetic resonance imaging (MRI) findings, with other potential etiological causes of abdominal pain being systematically ruled out. Treatment primarily consists of analgesics and anticoagulants. In this case report, we present a pregnant patient who arrived at the emergency department with an acute abdomen. Various MR images are provided, illustrating the diagnostic process. The patient was successfully treated with anticoagulants and antibiotherapy. Through this case, we aim to highlight the role of imaging in the differential diagnosis, management, and follow-up of pregnant patients presenting with acute abdominal conditions.

Key words: infarction; adrenal gland; pregnancy; case report

Submitted: 19 October 2024 Revised: 4 March 2025 Accepted: 26 March 2025

Introduction

Acute abdominal pain during pregnancy may have uncommon but clinically significant causes, such as non-hemorrhagic adrenal infarction (NHAI). The overlapping symptoms and non-specific clinical presentation in pregnant women make diagnosing NHAI particularly challenging.

Both hemorrhagic adrenal infarction (HAI) and NHAI are believed to result primarily from adrenal vein thrombosis, with adrenal hemorrhage occurring during the reperfusion phase of necrotic or injured arteries. According to a recently published retrospective analysis, the prevalence of adrenal vein thrombosis during pregnancy is estimated at 1.5 per 10,000 live births (Descargues et al, 2019).

Unilateral non-hemorrhagic adrenal infarction is a rare condition, especially in pregnant women. While bilateral adrenal infarction typically leads to adrenal insufficiency, which can result in shock, sepsis, and even death, unilateral adrenal infarction often presents with acute abdominal pain that is refractory to analgesia, along with nausea and vomiting. Its symptoms can mimic a variety of other causes of acute abdominal pain, further complicating diagnosis (Fei et al, 2019). Although

How to cite this article:
Ocal ZA, Demirci Y. A Rare Condition: Nonhemorrhagic Infarct of Adrenal Gland in Pregnancy. Br J Hosp Med. 2025. https://doi.org/10.12968/hmed.2024.0783

the exact etiology remains unclear, this condition is thought to be associated with hypercoagulable states and the hemodynamic changes that occur during pregnancy (Fei et al, 2019; Struble et al, 2015).

Adrenal infarction occurs when there is a disruption in the blood supply to the adrenal gland, leading to tissue damage without significant bleeding (Fei et al, 2019). Pregnancy is a hypercoagulable state in which the homeostatic balance shifts toward coagulation, increasing the risk of thrombosis and other maternal complications (Struble et al, 2015).

Fox's landmark article provides the most comprehensive description of the pathophysiology of adrenal infarction. According to his findings, the majority of adrenal infarction cases are venous in origin, resulting from clots in the intra-adrenal veins, venous sinuses, parietal sinuses, capsular veins, and primary extra-adrenal veins. His theory suggests that capsular arteries may undergo spasms in response to adrenal vein thrombosis, leading to cortical necrosis without hemorrhage. This mechanism could explain the absence of adrenal hemorrhage in our patient (Fox, 1976).

Diagnosis is challenging due to the non-specific clinical presentation and overlap of symptoms with other common conditions. Additionally, pregnancy can make it difficult to locate the source of pain because of the shifting intra-abdominal organ positions caused by the expanding uterus.

When a pregnant woman experiences abdominal pain, ultrasound is the first diagnostic procedure to be used. If additional non-contrast magnetic resonance imaging (MRI) examination is required, it can be used to rule out other more common pathologies and confirm the presence of adrenal findings on MRI, which allows for the diagnosis of adrenal infarction.

Because of the difficulty in detecting non-hemorrhagic adrenal infarction, clinicians should be aware of this situation and approach it carefully in order to provide the best possible treatment and achieve the best outcome for both the mother and the fetus. Because it does not cross the placenta and is safe for the fetus, low-molecular-weight heparin such as enoxaparin is recommended as the first choice for anticoagulation in pregnancy.

In addition to the most common etiologies of pain such as appendicitis, cholecystitis, and renal colic in a pregnant woman presenting with acute unilateral abdominal pain, we wanted to contribute to the literature by presenting a rare case of non-hemorrhagic adrenal infarction and control imaging findings.

Case Report

A 33-year-old woman in her 25th week of pregnancy (gravida 4, parity 2) presented to the emergency department with severe right upper quadrant pain. Physical examination revealed no rebound tenderness or discomfort in the right subcostal area. She reported persistent pain symptoms. Her initial vital signs, including temperature, oxygen saturation, blood pressure, and pulse rate, were within normal limits. Laboratory results showed an elevated white blood cell count of 20,000/μL (normal range: 4000–10,000/μL), with 91.9% neutrophils (normal range: 50–70%)

and 4.9% lymphocytes (normal range: 20–40%). Platelet count was 380,000/micrL (normal range: 100–300 $\times$ 10^3 cells/μL). Urea was measured at 13 mg/dL (normal range: 16.6–48.5 mg/dL), creatinine at 0.35 mg/dL (normal range: 0.5–0.9 mg/dL), and C-reactive protein (CRP) at 23.04 mg/dL (normal range: 0–5 mg/dL). Because of the right upper quadrant pain and elevated CRP levels, which suggested acute cholecystitis, an abdominal ultrasound was performed. The CARE checklist has been attached as **Supplementary Material** associated with this article.

The ultrasound did not identify any conditions that could explain the abdominal pain, including cholecystitis, pancreatitis, urolithiasis, or appendicitis. The only notable finding was a minimal amount of fluid adjacent to the upper pole of the right kidney, with no other abnormalities detected. Due to persistent, severe discomfort localized in the right upper quadrant, the patient underwent a contrast-free MRI. MRI revealed T2 hyperintense and T1 isointense areas, measuring 21 $\times$ 9 mm at the body and 14 $\times$ 9 mm at the medial crus of the right adrenal gland, with no signs of bleeding. Additionally, a fluid collection was observed adjacent to the right adrenal gland and the upper pole of the right kidney (Fig. 1). Increased signal intensity in the same regions as T2 was also noted in diffusion and apparent diffusion coefficient (ADC) scans (Fig. 2). The patient had no personal or family history of coagulation disorders. With these findings, unilateral non-hemorrhagic adrenal infarction stood out as the main differential diagnosis to be considered.

After the diagnosis, the patient received intravenous fluids, antibiotics (because of the increase in CRP), analgesics, and was started on subcutaneous enoxaparin 80 mg two times per day. The prenatal examination was completely normal, and there were no laboratory findings such as hyponatremia, hyperkalemia and hypoglycaemia, which would otherwise be suggestive of adrenal insufficiency.

The patient was prescribed a therapeutic dose of enoxaparine (80 mg subcutaneously twice a day) continuing through the 6th postpartum week and discharged on the fourth day of hospitalization, exhibiting a decrease in CRP level and resolution of abdominal pain.

The patient did not experience a recurrence of abdominal pain during pregnancy follow-up or within four to six months after delivery. She was advised to undergo a follow-up MRI to assess her adrenal glands. The MRI performed four months postpartum showed that the adrenal abnormalities had completely resolved (Fig. 3).

Discussion

Unilateral non-hemorrhagic adrenal infarction is an extremely rare condition and is not typically considered among the primary causes of acute abdominal pain in pregnant women. Unilateral adrenal infarction may present with acute abdominal pain refractory to analgesia, nausea and vomiting, and may mimic a variety of other causes of acute abdominal pain, whereas bilateral adrenal infarction usually presents with adrenal insufficiency leading to shock, sepsis and death (Fei et al, 2019).

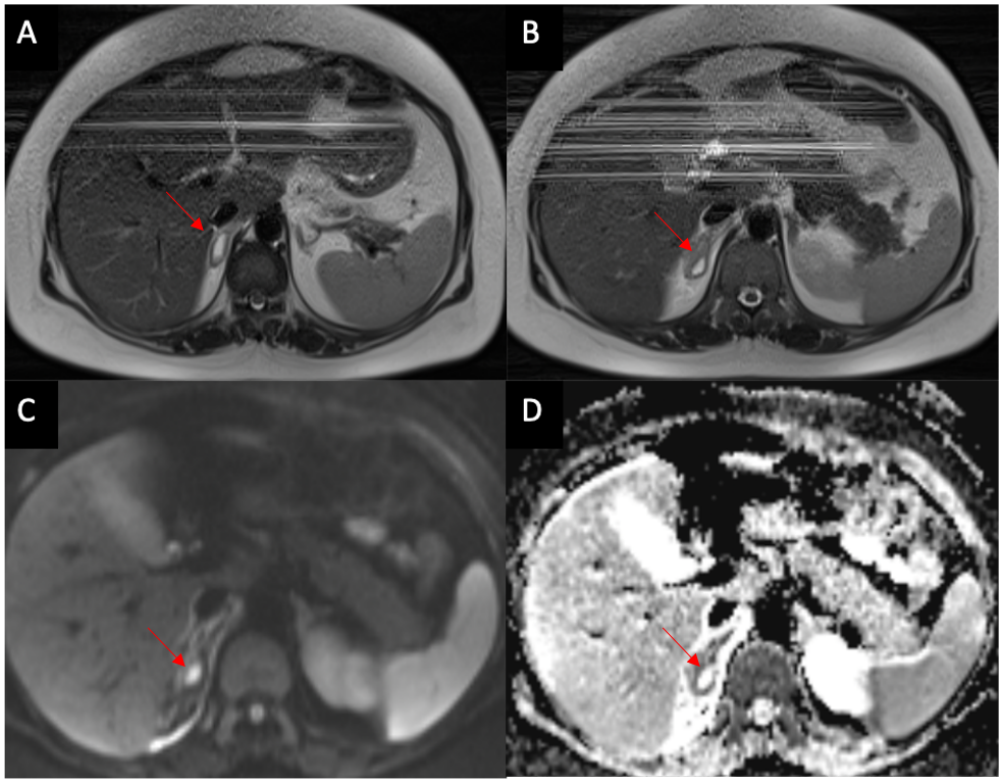

Fig. 1. The abnormalities within the adrenal gland (marked with red arrows). The right adrenal gland shows T2 hyperintense T1 isointense areas measuring 21 × 9 mm at body level (A) and 14 × 9 mm at the medial crus (B). This area shows high signal intensity on diffusion-weighted imaging (DWI) (C) and apparent diffusion coefficient (ADC) (D), indicating the absence of diffusion restriction.

Upon literature review, we found that the majority of reported cases of non-hemorrhagic adrenal infarction in pregnant women are unilateral as in our case, which involved the right adrenal gland, with the incidence of bilateral non-hemorrhagic adrenal infarction being only 8.3% (Tschuertz et al, 2023). The predominance of right adrenal involvement in these cases may be due to compression of the inferior vena cava by the expanding uterus during pregnancy. Given that the right adrenal vein drains directly into the inferior vena cava and that it's shorter and thinner than the left, the risk of venous congestion in the right adrenal gland is greater than in the left (Riddell and Khalili, 2004; Tschuertz et al, 2023).

Diagnosis is challenging due to the non-specific clinical presentation and overlapping symptoms with those of others. Patients may present with a complaint of severe right upper quadrant pain, which may mimic the symptoms of cholecystitis, renal stones or other abdominal problems. The absence of classic symptoms such as nausea, vomiting, fever and dysuria may further hinder the diagnosis.

In unilateral non-hemorrhagic adrenal infarction, the patient's serum cortisol (in our case 42.8 µg/dL) levels and responses to the adrenocorticotropic hormone (ACTH) stimulation test are typically normal because the contralateral adrenal gland is functional. During pregnancy, cortisol levels are elevated due to multifactorial mechanisms, and serum cortisol levels were also elevated in our case (Shah et al, 2022).

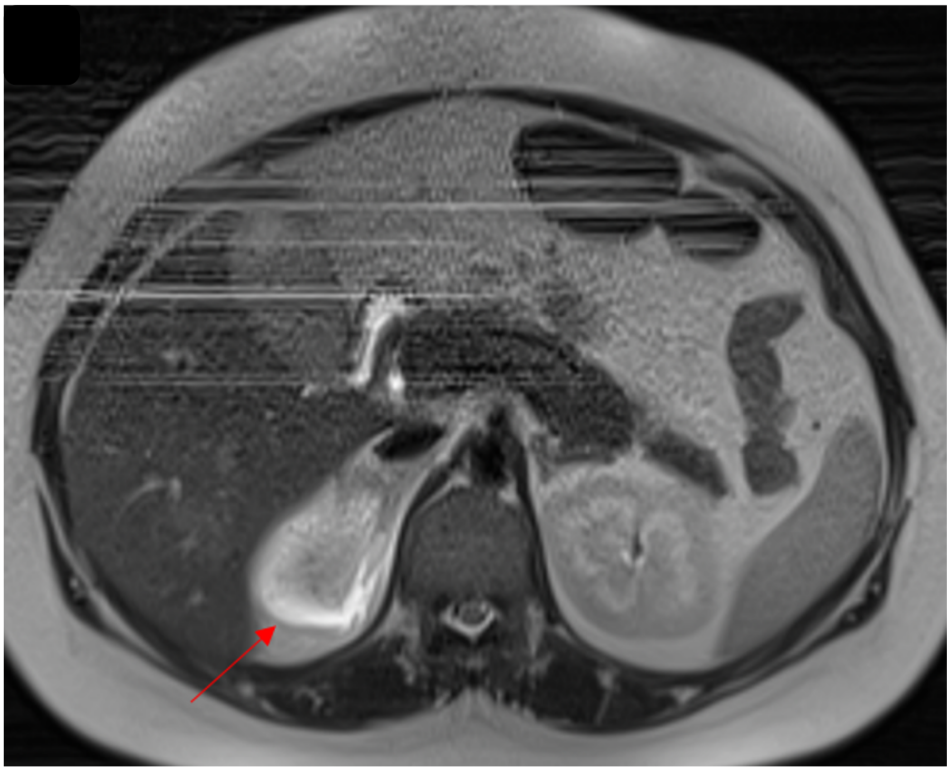

Fig. 2. The fluid beside the adrenal gland. There is a small amount of free fluid next to the upper pole of the right kidney shown with the red arrow.

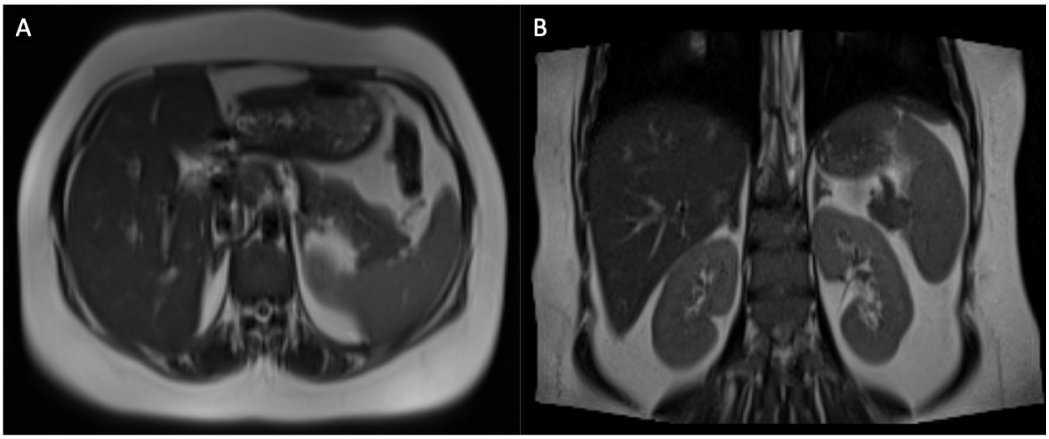

Fig. 3. Normal appearance of the adrenal glands at four months after delivery. At the previous examination, the T2 signal changes in the right adrenal gland (Fig. 1) and the fluid collection in the perirenal region (Fig. 2) had completely resolved and returned to normal on the axial image (A) and coronal image (B).

Imaging techniques play a crucial role at this stage. Ultrasound is usually the first radiological examination to be performed in a pregnant woman, and in the absence of an obvious diagnosis on ultrasound, a second choice of imaging must be performed (Sidibe et al, 2021). Unlike CT, MRI is non-invasive and does not use ionising radiation. It is, therefore, safer for both the mother and the fetus (Sidibe et al, 2021). It provides excellent soft-tissue contrast, allowing accurate visualisation

of retroperitoneal pathology that may not be apparent on ultrasound. In addition, MRI can differentiate between hemorrhagic and non-hemorrhagic infarcts, providing important information for treatment planning.

The infarcted adrenal gland is enlarged and the signal intensity is increased on T2-weighted magnetic resonance (MR) images with surrounding oedema, and T1 hyperintensity may also be seen due to acute ischemic injury (Glomski et al, 2018; Chagué et al, 2021). The signal abnormalities may be homogeneous or heterogeneous with multiple foci, especially on T2 and diffusion-weighted imaging (Molière et al, 2017). The ADC value of the whole adrenal gland may not be significantly reduced due to the predominance of oedema over ischaemia (Molière et al, 2017).

The retroperitoneal oedema centred on the adrenal gland seen on MRI is likely to reflect local reactive processes around the infarcted adrenal gland and the thrombosed adrenal vein (Glomski et al, 2018).

Contrary to case reports of uncomplicated perinatal outcomes without anticoagulation. However, there are studies recommending continued anticoagulation for 6–8 weeks postpartum parallel to our study due to the risk of thromboembolic events which are higher after delivery than they are during pregnancy. However, some of these women have been reported to have been readmitted to the hospital during pregnancy with similar symptoms (Gavrilova-Jordan et al, 2005).

The duration of anticoagulation therapy after the end of pregnancy was not always reported in the various case reports. In our case, therapeutic anticoagulation therapy was continued through the end of 6th postpartum week. This aspect of treatment should be discussed with a haematologist, especially in cases of underlying thrombotic disease (Chasseloup et al, 2019).

Since the MR results in our case in point were consistent with the literature, we were confident in our diagnosis and prescribed therapeutic anticoagulation through the fourth month of pregnancy. In our case, the acute phase was managed with analgesics, fluids, and antibiotics to address the possibility of a superimposed infection. Following the acute event, follow-up adrenal imaging is recommended, as patients may develop adrenal hemorrhage due to revascularization or adrenal gland atrophy (Shah et al, 2022). In our patient, follow-up MRI findings showed complete resolution of the adrenal abnormalities.

Conclusion

In conclusion, pregnancy-related adrenal infarction is a rare condition that may be associated with systemic coagulation disorders. However, the hypercoagulable state of pregnancy itself can also be a contributing factor. Acute abdominal pain is often the first symptom of adrenal infarction, and initial diagnostic tools such as ultrasound are often inconclusive. Therefore, when the cause of pain remains unclear, MRI should be considered as a crucial imaging modality to identify the underlying pathology.

Learning Points

- Adrenal infarction is not a diagnosis that initially comes to mind, and it is typically identified through a process of exclusion after ruling out more common causes of abdominal pain.
- MRI is the preferred imaging modality secondary to ultrasound for pregnant women due to its safety and reliability. The infarcted adrenal gland, surrounded by edema and lacking T1 hyperintensity and typically exhibiting high T2 signal intensity.
- Given the heightened risk of hypercoagulability extending up to the sixth week postpartum, anticoagulant therapy at a therapeutic dose serves as a protective measure to prevent potential complications.

Availability of Data and Materials

The datasets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

Author Contributions

ZAO and YD contributed significantly to the manuscript's conception. ZAO wrote the first draft and both authors worked together to revise the manuscript critically. The final manuscript was read and approved by both writers. Both authors agreed to take responsibility for all sections of the work and contributed substantially.

Ethics Approval and Consent to Participate

The patient gave permission for her case to be anonymously reported in the medical literature. The authors affirm that the protocols were carried out in compliance with the guidelines set forth in the World Medical Association Declaration of Helsinki.

Acknowledgement

Not applicable.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

Supplementary Material

Supplementary material associated with this article can be found, in the online version, at https://www.magonlinelibrary.com/doi/suppl/10.12968/hmed.2024.0783.

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