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CASE REPORT

RARE UNILATERAL ADRENAL INFARCTION DURING PREGNANCY: A CASE REPORT

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Abstract

1. Introduction:-

Adrenal infarction during pregnancy is a rare pathology.Only a few cases have been reported in the literature[1], most of them occurring during pregnancy[2]and report a bilateral damage. Pregnancy is a known state of acquired hypercoagulability[3] which, in association with the compression by the gravid uterus, constitutes a favorable environment for the occurrence of thrombosis. We report here a case of spontaneous unilateral infarction during pregnancy and review the literature to describe this clinical entity.

2. Case presentation:

A 25-year-old female patient, body mass index of 33, presented to the obstetric emergency room at 34 weeks and 5 days of her first gestation for severe, sharp, and acute abdominal pain in the right hypochondrium which radiated to the back, associated with nausea and vomiting. She had gestationaldiabetes controlled by dietary measures.During this pregnancy,she was hospitalized and treated at 18 weeks and 4 days of gestation for premature rupture of membranes and was discharged 4 days later. She had a history of cholecystectomy for biliary lithiasis. Her medication regimen included iron and prenatal vitamin supplementation.

On presentation to the hospital, she was tachycardic with a heart rate of 113/min, her blood pressure was normal. Abdominal exam revealed tenderness in the right flank. Her obstetrical examination reported a uterine height of 33cm, present and regular foetalheartbeat, a mid-length non dilated cervix with a softened consistency, applied cephalic presentation and clear amniotic fluid. The fetal heart rate monitoring wasnormal. The standard biological workup did not show any inflammatory syndrome or signs of renal or hepatic impairment. Obstetrical ultrasound reported good fetal vitality and the placenta didn't show abnormalities. Abdominal ultrasound did not reveal any hepato-biliary, renal, or urinary tract abnormality. The patient was initially put on analgesics level 1 and 2 then a titration of morphine was initiated. As the pain did not improve with morphine, an abdominal-pelvic computed tomography (CT) scan with injection of contrast agent was performed and showed an increase in volume of the right adrenal gland with no enhancement after injection, and infiltration of the peri-renal fat.The left adrenal gland enhanced normally (Figure 1). The diagnosis of right adrenal infarction was made and anticoagulant treatment with low molecular weight heparin (LMWH); enoxaparin 60 mg subcutaneously twice daily was started to prevent further venous thrombosis, which she continued for a total of 6 months.

Biological tests showed hypercortisolemia at 1264 nmol/l. Treatment with hydrocortisone 30mg/d was prescribed. The cortisol level returned to normal at 600nmol/l and then 445nmol/l respectively at day 4 and day 6. The ACTH level measured at day 7 of diagnosis was 93.4 pg/ml (for a normal <48). Other tests that included factor V Leiden,

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prothrombin mutation, protein C, protein S, antithrombin III, factor VIII activity revealed no thrombophilia. Antiphospholipid antibody test was also negative. The pain disappeared at day 10. The patient was induced at 38 weeks and 2 of gestation for macrosomia after a therapeutic window of LMWH. She then delivered by cesarean section for stagnation of dilation, a baby of 4205g.
An ACTH stimulation test was performed six months later. Her baseline and stimulated cortisol levels were normal.

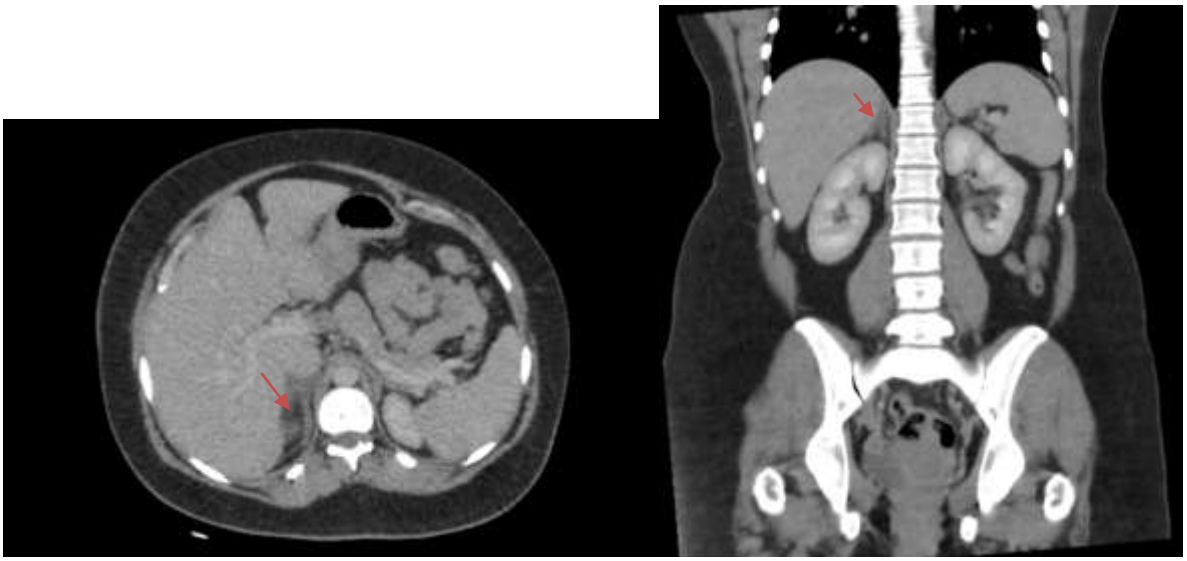

Figure 1:- Injected thoraco-abdomino-pelvic CT scan showing thrombosis of the right adrenal vein associated with edema.

3. Discussion:-

Adrenal infarction is a very rare cause of abdominal pain in pregnancy. Few cases have been described in the literature describing this non-hemorrhagic form of adrenal infarction[1, 4]probably because of the under-diagnosis of this pathology.

It is described mainly on the right side given the gravid uterine dextroposition and the anatomy of the right adrenal vein flowing into the inferior vena cava without collateral circulation[5]. The arterial supply is rich but venous drainage is limited to a single vein, and the localization of thrombi in the adrenal vein may result in local blood stasis, leading to edema and necrosis of the gland. Most adrenal infarcts described in the literature are bilateral [6] and are often accompanied by hemorrhage that occurs during reperfusion of the necrotic or damaged vessels [7].

Pregnancy represents a state of hypercoagulability that aims to decrease the risk of bleeding during delivery[3], but in return it increases the thrombogenic risk by 5 times. However, adrenal infarction may reveal an underlying coagulopathy such as a factor V Leiden mutation [8] or antiphospholipid syndrome[9].
- Clinically,unilateral adrenal infarction may present as an acute abdomen with severe unilateral abdominal pain of acute onset, associated with nausea and vomiting.
- Biological signs of adrenal insufficiency are often absent.
- Radiologically, as the presentation is more suggestive of more common surgical emergencies such as acute cholecystitis, acute pancreatitis, acute appendicitis, ureteral calculus or even ovarian torsion, ultrasound is the first radiological examination performed in pregnant women[10]. It has limited sensitivity and specificity for the diagnosis of venous or arterial adrenal thrombosis due to the technically difficult image acquisition, secondary to the gravid uterus. CT with contrast injection is considered the reference imaging modality to exclude adrenal ischemia in an emergency setting. The imaging signs of acute adrenal ischemia consist of adrenal hypertrophy with absent or low enhancement of the adrenal gland, perilesional fatty inflammatory changes, and the "capsular sign": a subtle hyperdense peripheral line around a hypodense adrenal gland and, in some cases, vein thrombosis (CT article). Ideally, during pregnancy, MRI should be performed because of the safety of this examination with respect to radiation, but it should be performed without gadolinium injection because of the risk of fetal toxicity[11, 12]. The characteristics of adrenal infarction on magnetic resonance imaging include an increased T2 signal suggesting edema in the gland. The gland may be slightly enlarged. There may be adjacent free fluid surrounding the adrenal

gland in the retroperitoneum. If gadolinium is used, the affected gland will likely be hypoechoic compared with the normal contralateral adrenal gland [13].

- Therapeutically, once the diagnosis of adrenal infarction is made, anticoagulation is usually initiated[14]. This prevents recurrent thrombosis, especially in the physiologically hypercoagulable state of pregnancy. Low-molecular-weight heparin such as enoxaparin is recommended as the first choice for anticoagulation during pregnancy, as it does not cross the placenta and is safe for the fetus[15]. The duration of anticoagulant therapy after the end of pregnancy is not always indicated in the various cases reported in the literature[1]. This aspect of treatment should be discussed with a hematologist, particularly in the case of underlying thrombotic pathologies. When a subsequent pregnancy is planned, preventive anticoagulant treatment should be discussed with a specialist to avoid the possible development of contralateral adrenal thrombosis and thus expose the patient to the risk of acute adrenal insufficiency.

Since it can be life-threatening,our case highlights that obstetricians and midwives should include adrenal infarctionas a differential diagnosis when faced with severe resistant abdominal pain during pregnancy, even without anyidentified thrombophilic factors.

This case report has been reported in line with the SCARE Criteria[16]

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