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Endovascular management of renal artery aneurysm rupture in pregnancy – A case report

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ABSTRACT

INTRODUCTION: Renal artery aneurysm rupture is an extremely rare cause of acute abdominal pain and haemodynamic instability in pregnancy. Due to its rarity, the diagnosis may not be immediately considered, and therefore there is a high associated mortality rate for both mother and fetus.
PRESENTATION OF CASE: We present a case of a 41-year old primigravida who presented to the obstetricians at 22 weeks' gestation with severe abdominal pain, shock and fetal loss. A bleeding renal artery aneurysm was discovered at laparotomy and radiologically coiled with sacrifice of the left kidney. Treatment of a contralateral aneurysm by autotransplantation of the remaining kidney allowed for preservation of residual renal function.
DISCUSSION: Surgical acute abdominal presentations can be difficult to interpret in pregnant patients. Pregnancy is known to be a contributing risk factor for spontaneous rupture of renal artery aneurysms, an otherwise rare mode of aneurysm presentation. Prompt use of imaging to diagnose and treat non-obstetric causes of the acute abdomen should not be delayed because of perceived risks to the fetus. Endovascular arrest of aneurysmal haemorrhage may be more effect in the context of a gravid uterus than surgical management.
CONCLUSION: In the shocked pregnant patient with an acute abdominal presentation, visceral artery aneurysm rupture may be comparatively more common, and should be considered in the absence of other localizing symptoms. Prompt interventional radiological treatment may be lifesaving in such cases.

1. Introduction

Exsanguinating haemorrhage as a presentation of renal artery aneurysm is documented but rare. We present a rare case of bilateral renal artery aneurysms in a 41-year old primigravida who presented with rupture of a left-sided renal artery aneurysm at 22 weeks' gestation and subsequent fetal loss. Treatment of a large asymptomatic contralateral renal artery aneurysm was successfully performed by kidney autotransplantation, allowing for preservation of the patient's remaining kidney function and giving the patient and her husband the option to conceive again.

2. Presentation of case

2.1. History and examination

A previously well 41-year old primigravida, who had conceived through IVF due to endometriosis, presented acutely to the Obstetric Emergency Unit at 22 weeks' gestation with sudden-onset severe left-sided abdominal pain, which had appeared 90 min earlier whilst sitting. She was nauseous and dizzy, but there were no urological or gastrointestinal symptoms and she was aware of normal fetal movements. She had hypermobile wrist joints, but no other features of Ehlers–Danlos Syndrome. There was no personal or family history of connective tissue disorders or other illnesses relevant to subsequent findings.

On arrival she was afebrile but tachycardic, with local peritonism in the left renal angle and left iliac fossa despite large amounts of opioid analgesia. She was of slim body habitus. Her uterus was soft and non-tender to palpation. Vaginal examination demonstrated a closed os with no abnormal discharge or blood, but left adnexal stimulation was severely tender. There were no palpable abdominal masses and bowel sounds were normal. Arterial blood gas analysis showed a deep metabolic and lactic acidosis with respiratory compensation, partially responsive to fluid resuscitation. Urinalysis showed no haematuria to suggest ureteric lithiasis. Bedside transabdominal ultrasound showed no signs of fetal distress or intrauterine growth retardation and normal kidneys and ovaries. Her initial haemoglobin and renal function were normal.

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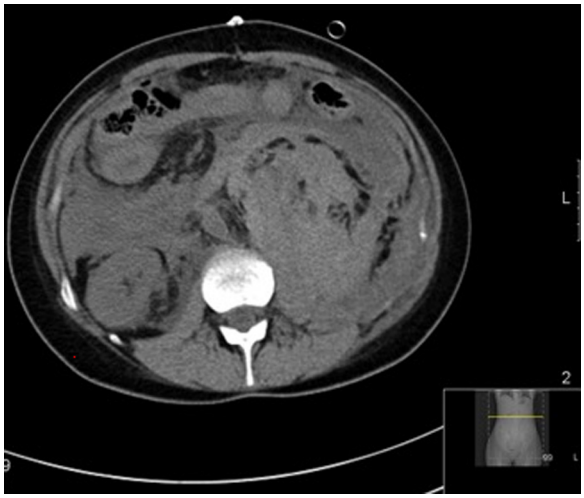

Fig. 1. Unenhanced CT demonstrating a large retroperitoneal haematoma surrounding the left renal artery.

2.2. Emergency management and diagnosis

Laparoscopy was precluded given the pregnancy's gestation, and out-of-hours MRI imaging was not rapidly available. Before a low-dose abdominal CT scan could be performed, the patient became haemodynamically unstable despite fluid resuscitation with signs of fetal distress. A midline laparotomy was immediately performed at which the uterus was found to be displaced to the right by a large left retroperitoneal haematoma, which was not disturbed. The deceased fetus was delivered by Caesarian section and the abdomen was closed.

An immediate abdominal CT (Fig. 1) and emergency abdominal angiography was performed under the same general anaesthetic. Ongoing brisk bleeding was seen to be coming from a large saccular left-sided renal artery aneurysm, close to the aortic origin of the main renal artery stem (Fig. 2). A symmetrical unruptured aneurysm was also seen on the contralateral renal artery. Coil embolisation of the left main renal artery main stemmed the haemorrhage but caused infarction of the left kidney (Fig. 3).

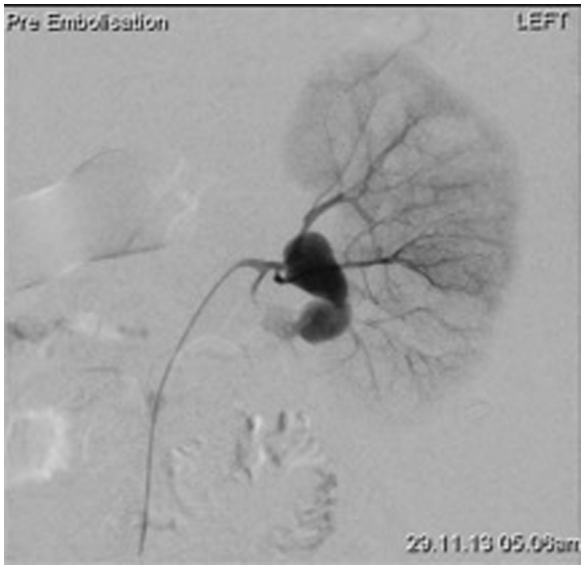

Fig. 2. Pre-embolisation angiography demonstrating a bleeding left renal artery aneurysm.

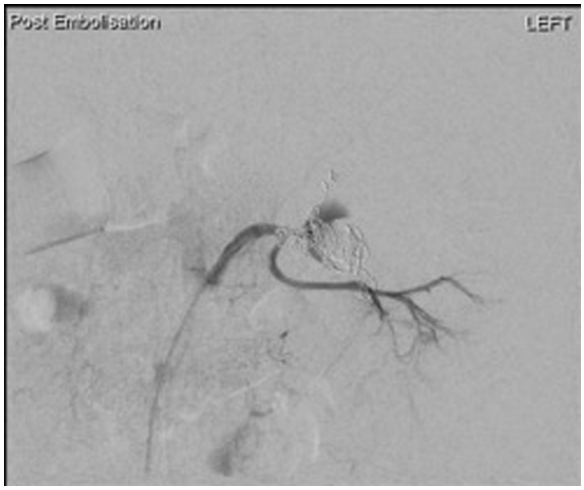

Fig. 3. Post-embolisation angiography of left renal artery demonstrating aneurysm securement and renal sacrifice.

2.3. Outcome and management of contralateral side

The patient made a good recovery after her laparotomy in the intensive care unit, despite a pulmonary embolism for which she received Rivaroxaban. Definitive management of the large contralateral unruptured aneurysm was deemed necessary to prevent future rupture and loss of her remaining renal function, particularly as the patient and her husband wished for future pregnancies. As endovascular stenting was unsuitable due to the aneurysm's proximal position, she underwent successful right kidney autotransplantation. No further aneurysms or vascular tortuosities were identified on imaging, and COL3A1 gene sequencing was normal. She has recovered well with a return to work and normal daily activities, and has conceived again without IVF.

3. Discussion

Renal artery aneurysms are uncommon, affecting less than 1 in 1000 in the general population [1]. They are usually an incidental discovery following investigation of potential renal causes of hypertension. Although spontaneous rupture as a mode of presentation occurs in less than 2% of cases, pregnancy is a well-reported risk factor. This could be due to hormonal influences on connective tissues, increased intraabdominal pressures or the hyperdynamic state of pregnancy [2]. There does not appear to be a link to increasing maternal age or number of previous pregnancies [3].

Prognosis following aneurysmal rupture is poor, with nearly universal loss of the affected kidney in the emergency scenario [4]. There is over 85% fetal (and around 50% maternal) mortality [5], although case reports of aneurysm rupture with subsequent fetal survival following Caesarian delivery have been reported at later gestational ages [6]. This may be due in part to delays in diagnosis whilst more common obstetric causes of abdominal pain and shock are considered. Although obstetric causes of abdominal pain had been excluded in this patient by the time of surgical referral, the list of differential diagnoses was still relatively broad, including ovarian, general surgical, urological and vascular pathologies. Where less profound maternal cardiovascular instability is present, urgent imaging may afford a pre-operative diagnosis [7,8].

In most cases, there is at least microscopic haematuria on urinalysis [2], but the decision to perform radiation-heavy investigations may be delayed in the context of pregnancy, particularly in the presence of normal fetal physiology. Ultrasound imaging may not elucidate the retroperitoneal pathology and MRI imaging is not suitable for haemodynamically unstable patients. A low-dose

CT scan is sensitive and low-risk in pregnancy (delivering radiation equivalents of around 6 abdominal x-rays) [9], and should be considered early. Laparoscopy is possible during pregnancy, but trochar placement becomes more dangerous from the beginning of the second trimester onwards, due to the increasing size of the gravid uterus [10]. In cases of postulated vascular pathology and where there is rapid access to interventional radiology, angiography is a better first choice than MRI/CT for delineating operative anatomy, with the option of proceeding to therapeutic interventions on confirmation of the diagnosis. Coil embolisation of ruptured RAAs can lead to preservation of renal function in distal aneurysms, however proximal aneurysms remain difficult to manage without occlusion of the renal artery trunk [11].

Treatment of incidental contralateral aneurysms is not often required, given that serial surveillance has been shown to be safe in the majority of cases [12]. However, the threshold for treatment is much lower in women of child-bearing age due to the excess risk for renal artery aneurysm growth and rupture in pregnancy [13]. Autotransplantation is employed in cases of proximal renal aneurysms not amenable to endoluminal stenting or coiling [14].

4. Conclusions

Rapid diagnosis of non-obstetric causes of abdominal pain can often prove challenging in the critically ill pregnant patient. Given their propensity to grow and rupture in pregnancy, a possible diagnosis of visceral abdominal aneurysm should be considered in shocked patients with abdominal pain in the absence of other symptoms. The risk of radiation to the fetus is irrelevant in cases of maternal instability, and low-dose CT imaging should be considered early. Early involvement of interventional radiology services, where possible, is of great help for both diagnosis and therapeutic treatment where a gravid uterus may hamper surgical efforts to control visceral haemorrhage.

Conflict of interest

None Declared.

Sources of funding

Nothing to declare.

Ethical approval

Nothing to declare.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief on request.

Author's contribution

Elizabeth Maughan – preparation of manuscript and literature review.

C. Webster – preparation of manuscript and consent of patient.

T. Konig – primary review of manuscript.

I. Renfrew – procedure and review of manuscript.

Guarantor

Elizabeth Maughan MRCS (ENT), MSC, MBBS, MA (CaNtab).

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