CASE REPORT

Renal arteriovenous fistula revealed by severe hypertension during pregnancy

Morgane Perrin,¹ Ruben Lousquy,¹ Mathias Rossignol,² Philippe Bonnin³¹Department of Obstetric Gynecology, Lariboisiere Hospital, Paris, France²Anesthesiology and Critical Care, Lariboisiere Hospital, Paris, France³Department of Physiologie Clinique Explorations Fonctionnelles, Lariboisiere Hospital, Paris, France**Correspondence to**Dr Ruben Lousquy,
rubenlousquy@gmail.com**SUMMARY**

A 35-year-old woman developed severe hypertension resistant to antihypertensive treatment during the second trimester of pregnancy at 24 weeks gestation. Doppler ultrasonography achieved the diagnosis of idiopathic renal arteriovenous fistula in the left kidney associated with parenchymal hypoperfusion. A Caesarean section was performed 6 days after the diagnosis because of severe pre-eclampsia. After delivery, the symptoms disappeared. Fistula persisted after follow-up for over 1 year but with a dramatic decrease in its blood flow and normalisation of the left kidney hemodynamics. Nevertheless, embolisation was performed without complications to prevent recurrence during the next pregnancy expected by the patient.

BACKGROUND

Renal arteriovenous lesions are rare and can be congenital, acquired or idiopathic.^{1 2} Idiopathic renal arteriovenous fistulas (AVF) represent 3%–5% of renal arteriovenous lesions³ and are mostly discovered during the occurrence of cardiovascular symptoms. We found only 13 published cases of kidney arteriovenous lesions, four fistulas and nine malformations, during pregnancy including only two cases of idiopathic renal AVF.^{4 5} Idiopathic renal AVF can induce hypertension and prematurity during pregnancy. As we did not find a lot of articles in literature on this topic, this brief report is original and informative to manage this situation. How to make the diagnosis? How to manage the medical supervision, the delivery, the induced prematurity and the postpartum?

CASE PRESENTATION

A 35-year-old black woman initially presented uncontrolled hypertension discovered during the second trimester of pregnancy. She had no medical history. She initially experienced a first trimester miscarriage. At 24 weeks gestation, she developed severe hypertension resistant to three antihypertensive drugs taken orally. Indeed, high blood pressure (160/90 mm Hg) was observed with nicardipine 50 mg (calcium blocker) three times a day, labetalol 200 mg (β blocker) four times a day and molsidomine 2 mg (vasodilator) four times a day. Initially pre-eclampsia was suspected but we did not notice proteinuria. We decided to search for another cause of hypertension. Renal ultrasonography was normal and the two kidneys were of normal size. Blood samples did not evidence alteration of the renal function. Colour-coded and pulsed Doppler ultrasound (US) imaging evidenced an actual renal

AVF in the lower medullar part of the left kidney, near the hilum, consisting of a large dilatation (over 1 cm) of a unique lobular vein within very high blood flow velocities and turbulent flow (figure 1A–C). Upstream from the lesion, the left renal artery evidenced increased blood flow velocities; downstream from the lesion, the left renal vein was dilated and evidenced increased blood flow velocities exhibiting an arterial modulation. The vascular arteriovenous lesion was associated with decreased blood flow velocities in all lobular arteries of the left kidney compared to the right (figure 2A–D). Ultrasonographer thus evoked a hemodynamic steal syndrome produced by the medullar AVF at the expense of the renal parenchyma. RMI confirmed the diagnosis (figure 3). Because of the secondary occurrence of severe pre-eclampsia with HELLP syndrome, the patient was then transferred to a tertiary referral centre. A Caesarean section was performed at 25 weeks gestation, six days after discovering renal AVF.

OUTCOME AND FOLLOW-UP

One week after delivery, blood pressure became normal with only one antihypertensive drug (calcium blocker). One year after delivery, blood pressure was normalised without any medication and US imaging evidenced normalisation of all blood flow velocities in the lobular arteries of the left kidney compared to the right, whenever renal AVF persisted but with dramatically decreased velocities. Urotomography of the lower part of the left kidney displayed the persistence of the AVF measured at 18 mm of diameter. We did not find any other aetiology of pre-eclampsia. Different possible managements including surgery, embolisation or medical supervision were evaluated and we finally decided to perform an embolisation of the AVF. The purpose was then to prevent recurrence of severe hypertension and pre-eclampsia during the next pregnancy expected by the patient. The procedure was performed without any complications.

DISCUSSION

More than 200 cases of fistulas or other renal vascular malformations have been reported. Nevertheless, only 13 cases of kidney arteriovenous lesions, 4 fistulas and 9 malformations were published during or at the waning of pregnancy.^{4–7} Two of the four cases with well-known causes of acquired AVF were unmasked during pregnancy. The first one, induced by a renal traumatism prior to pregnancy, was discovered during the first trimester; the second one was induced by a

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Figure 1 Arteriovenous fistula to the inferior pole of the left kidney. Two-dimensional ultrasound imaging revealed a large dilatation (over 1 cm) of a unique lobular vein near the hilum of the left kidney (A) Colour-coded, ultrasound imaging evidenced a dramatic increase in blood flow velocities with aliasing (B) and pulsed, Doppler a high speed turbulent flow within the lesion (C).

percutaneous renal biopsy before pregnancy and discovered during the third trimester. Two of the other four cases with idiopathic fistulas were discovered after delivery because of persistence of severe cardiovascular symptoms, the first was associated to fibromuscular dysplasia of the renal artery and to severe hypertension, the second was associated with severe heart failure. Here, we discovered AVF during the second trimester of pregnancy because of the occurrence of a severe resistant hypertension. Two mechanisms were assumed to explain why symptoms were unmasked during pregnancy. We first hypothesised that pregnancy promotes the development of arteriovenous lesions because it submits lesions to increased levels of vasoactive agents and angiogenic factors such as vascular endothelial growth factor (VEGF).⁸ Kearse *et al*³ suggested increased renal blood flow may have promoted the erosion of a vein's wall by aneurysmatic intra renal arteries, causing the opening of an AV communication.

Here, our pregnant woman presented hypertension² as the main symptom. Mechanism of this hypertension may be related to the relative hypoperfusion of renal parenchyma downstream from the fistula. During pregnancy, Sundrani⁹ showed that maternal plasma VEGF and placental growth factor (vascular growth factors synthesised in the foetoplacental unit and released in the maternal circulation) increased during the first and the second trimester suggesting that pre-existing fistula can dramatically develop consecutively in the impregnation of these factors. This development can induce an actual 'haemodynamic steal syndrome' at the expense of the renal parenchymal network. Induced cortical renal hypoperfusion and induced putative decrease in arterial input pressure may have caused secondary activation of the renin-angiotensin-aldosterone system, resulting in progressive increase in systemic blood pressure and poor response to antihypertensive drugs.² As a complication of the severe arterial hypertension, cardiomegaly and congestive

Figure 2 Pulsed Doppler ultrasonography of the right (A) and left (B) renal arteries, and of the lobular arteries in the right (C) and left (D) kidneys. The left renal artery exhibited elevated peak systolic and end-diastolic blood flow velocities compared to the right artery. One of the left kidney median lobular arteries, downstream from the arteriovenous fistula (AVF) evidenced decreased peak systolic and end-diastolic blood flow velocities compared to those measured in the right kidney. Moreover, resistance index was decreased in the lobular artery in the left kidney (0.52) compared to that measured in the lobular artery in the right kidney (0.62), which evoked vasodilation downstream from the AVF.

Figure 3 Three-dimensional T1-weighted angio-MR image at the arterial phase (12 mm thick coronal maximal intensity projection reconstruction) slicing through the renal hilums shows the early contrast media enhanced of the dilated left renal vein in comparison with the right one.

heart failure can occur and has been previously described.² After delivery, levels in the vascular growth factors and blood flow in the fistula decreased involving the amendment of the renal parenchymal hemodynamic steal syndrome with normalisation of the renal parenchymal perfusion and with inactivation of the renin-angiotensin-aldosterone system.

Arteriography remains the gold standard to identify and treat renal AVF or malformations. However, this procedure is invasive and radiations need to be avoided during pregnancy as the first-line diagnosis. Doppler US imaging, angio-magnetic resonance imaging (MRI) can contribute to diagnosis.^{2–5} In that case, Doppler US imaging achieved the diagnosis of aneurysmal anomaly with a large communication between an artery and a vein in opposition to cirroid anomaly with a tangle of tortuous vessels of small diameter, with multiple varix-like communications between the artery and the vein.¹⁰

Treatment consists of embolisation or surgery. The goals of AVF treatment are the preservation of a functional renal parenchyma, eradication of symptoms and restoration of normal intrarenal hemodynamics.² Here, after 1 year of follow-up, symptoms disappeared after delivery while intrarenal hemodynamics were restored whenever AVF persisted but with dramatically lowered blood flow. Nevertheless, we decided to treat and to prevent severe hypertension, pre-eclampsia and prematurity during the eventual next pregnancy. We chose embolisation which is the most effective and less invasive treatment after delivery.^{11–12} The risks of this treatment are reflux of obliterating agents into proximal vessels such as normal renal parenchyma or bowel, pulmonary embolism or postembolisation syndrome with pain and fever.² The alternative treatment is surgery with

excision of the fistula, ligation of the feeding vessels, partial or total nephrectomy if embolisation fails or is not possible because of anatomic characteristics of the fistula.¹³

To conclude, idiopathic renal AVF is a rare renal arteriovenous lesion, which can induce hypertension and prematurity during pregnancy. Doppler ultrasonography must be preferred to arteriography for the diagnosis to avoid fetal radiation. Embolisation rather than surgery seems the treatment of choice to amend symptoms after delivery to prevent recurrence of hypertension during the next pregnancy.

Learning points

- ▶ Rare disease during pregnancy.
- ▶ Multidisciplinary management.
- ▶ Difficult diagnosis.
- ▶ Particularity during pregnancy.
- ▶ Treatment in order to prevent recurrence.

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