Rupture of renal artery aneurysms in pregnancy

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Introduction

We present a case of a 35-year-old female who presented at ten weeks gestation with lower abdominal and right flank pain and gross haematuria. A preoperative diagnosis of ruptured right renal artery aneurysm was made with colour Doppler ultrasound and is the first reported case in a pregnant female.

The high maternal and fetal mortality of ruptured renal artery aneurysm in pregnancy can partly be attributed to an incorrect preoperative diagnosis. In only one reported case was a diagnosis made preoperatively using computer tomography (CT) scanning.[1] Most of the patients were managed as obstetric emergencies with abruptio placentae being the commonest preoperative diagnosis. In addition, hypertension and haematuria during pregnancy add to the diagnostic dilemma.[2] Recent advances in ultrasound technology have meant that colour Doppler ultrasound can now be used as a quick non-invasive method for assessing the kidneys, renal arteries and retroperitoneal spaces.

Case report

A 35-year-old African female, gravida IV, para III, was referred by a general practitioner with a two week history of lower abdominal pain, and frank haematuria. The patient was 10 weeks pregnant. There was no significant past medical history and her previous pregnancies were all normal.

Examination revealed a blood pressure of 150/100 mmHg and a pulse of 82 beats/min. The patient had suprapubic and right flank pain on palpation of the abdomen. A bruit was audible on auscultation of the epigastrium. Catheterisation of the bladder demonstrated frank haematuria with clots. Haemoglobin was 8.4 g/dl and haematocrit was 24.3%. An emergency ultrasound examination demonstrated a normal, viable intrauterine pregnancy and blood clots were noted in the bladder. The left kidney was normal. Examination of the right kidney demonstrated a large 7.3 cm by 5.6 cm hypoechoic lesion in the lower pole (Figure 1a). Colour Doppler and duplex Doppler demonstrated extensive blood flow within the lesion (Figure 1b). A diagnosis of right renal artery aneurysm was made and arrangements were made to perform an emergency angiogram. The patient was counselled and agreed to

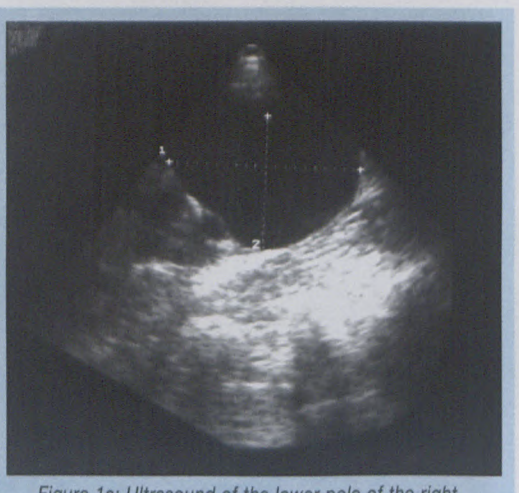

Figure 1a: Ultrasound of the lower pole of the right kidney demonstrating a 7.3 cm by 5.6 cm hypoechoic lesion

to page 11

from page 10

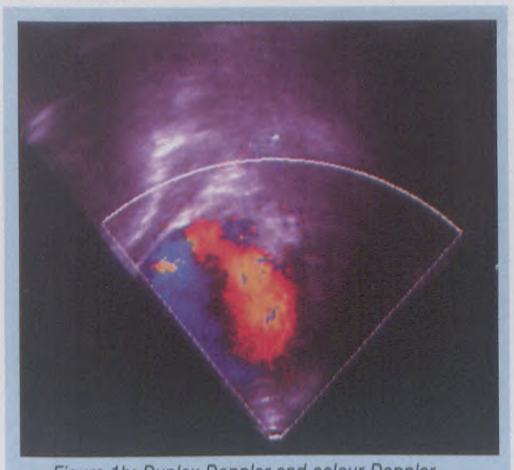

Figure 1b: Duplex Doppler and colour Doppler ultrasound demonstrating extensive blood flow within the lesion

have a termination of pregnancy. At angiography, a large saccular aneurysm arising from the lower primary division was detected (Figure 2). There did not appear to be any other feeding vessels. In addition, a fistula tract into the right renal vein was noted.

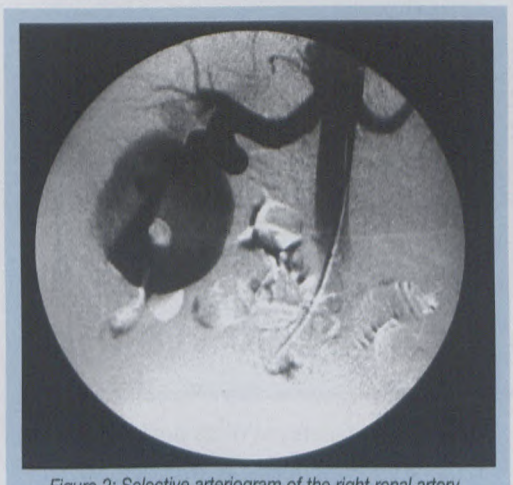

Figure 2: Selective arteriogram of the right renal artery demonstrating a large saccular aneurysm arising from the lower primary division and early venous filling of the renal vein and inferior vena cava

An attempt was made to embolise the feeding artery after discussion with the vascular surgeon. This however, was unsuccessful as the diameter of the artery proved to be much larger than the diameter of the endovascular coil. The coil entered the inferior vena cava via the arteriovenous fistula and lodged in the right lung. The patient was taken to theatre where a right eleventh rib resection was performed and the kidney and its vascular pedicle were explored. The feeding artery was mobilised and ligated with silk suture and this resulted in collapse of the aneurysm and disappearance of the bruit. The postoperative period and recovery were uneventful.

The patient had a termination of the pregnancy and a follow-up angiogram performed two weeks later demonstrated no evidence of the aneurysm or arteriovenous fistula.

Discussion

Incidence

The true incidence of renal artery aneurysms is still unknown. The incidence based on autopsy studies is 0.01%.[3] However, angiographic studies suggest a much higher incidence (9.7%).[4,5] In addition, Harrison *et al*[6] reported that 1.5% of all potential kidney donors who underwent angiographic evaluation had renal artery aneurysms. Only 24 cases of renal artery aneurysm rupture have been reported in pregnancy (Table I) and a further three cases[7,8,9] have been diagnosed post partum.

Aetiology

The formation and rupture of a renal artery aneurysm in pregnancy is still not fully understood, but is thought to be multifactorial. The aneurysm may be classified either as true or false. True aneurysms may be congenital or acquired and are either saccular or fusiform. False aneurysms usually arise secondary to trauma. The combination of hormonal and haemodynamic changes that occur during pregnancy are considered to play a major role in the development and rupture of renal artery aneurysms in pregnancy.

Wexler[10] has described the various changes that occur in the arterial wall of breeder rats during successive pregnancies and Manalo-Estrella and Baker[11] have documented connective tissue changes in the aortic media of pregnant females in 16 autopsy specimens. Intimal thickening has also been seen in the arteries of rats treated with synthetic steroids.[12]

Histopathological examinations were conducted on 12 of the 24 cases of renal artery aneurysm that ruptured during pregnancy. Atherosclerosis was demonstrated in three cases, fibromuscular dysplasia in three cases and neurofibromatosis in one case (Table I). In five cases no specific changes of atherosclerosis or fibromuscular dysplasia were seen. These findings are in contrast to Lacombe's study where 90% of his 123 patients operated for renal artery aneurysms had evidence of fibromuscular dysplasia.[13] The haemodynamic factors include an increased cardiac output and hence increased renal blood flow that occurs during pregnancy and compression of the aorta by the gravid uterus.[14]

Patient profile and clinical presentation

Review of the 24 previously published cases indicates that there is no relationship between maternal age or parity and the formation and rupture of a renal artery aneurysm in pregnancy. Only one of the patients was being managed for essential hypertension prior to her pregnancy[15] and none of the other patients had any significant medical history.

to page 12

from page 11

This is the second reported case of rupture in the first trimester of pregnancy[16] and there appears to be no correlation between fetal gestational age and rupture in this review.[17]

Patients with unruptured renal artery aneurysms are usually asymptomatic.

Abdominal bruits, murmurs or abnormal pulsations may occasionally be detected.

Calcification is noted in 30 to 40% of saccular aneurysms[4,18] and may be detected on abdominal x-ray. In the presence of a rupture however, acute abdominal and unilateral flank pain together with hypovolaemic shock, appear to be the commonest presenting symptoms. It is interesting to note that gross haematuria, indicating rupture into the renal pelvis, was the presenting symptom in only two other cases.[15,19]

Diagnosis

The preoperative diagnosis of a ruptured renal artery aneurysm was made in only one of the 24 previously reported cases.[1] The presence of haemorrhagic shock in the pregnant patient is usually presumed to be due to an obstetric cause such as an ectopic pregnancy, abruptio placentae and ruptured uterus. This is confirmed by the fact that abruptio placentae was the commonest preoperative diagnosis in the cases reviewed.

Rupture of a splenic artery aneurysm[20] and rupture of a thoracic aorta aneurysm[21] may also present with spontaneous retroperitoneal and intra-abdominal bleeding and mimic an obstetric emergency.

The use of pulsed Doppler and colour Doppler ultrasound to diagnose renal artery aneurysms is well documented.[22,23] Colour Doppler ultrasound provides a quick non-invasive method of assessing the kidneys, renal arteries and retroperitoneal spaces and should be used more routinely in the pregnant female who presents in haemorrhagic shock.

Table I: Reported cases of ruptured renal artery aneurysm in pregnancy

Case no	Authors	Year published	Preoperative diagnosis	Histology
1	Chisholm AE [30]	1926	abruptio placentae	none
2	Ostling K [31]	1938	not stated	non specific
3	Lennie & Sheehan [32]	1942	not stated	atherosclerosis
4	Lennie & Sheehan [32]	1942	not stated	none
5	Low DM [33]	1944	ruptured uterus	none
6	Kenny & Doniach [34]	1945	not stated	none
7	Zummo *et al* [20]	1952	abruptio placentae	none
8	Hack RW [35]	1953	left pyelonephritis	atherosclerosis
9	Ward & Martins [36]	1955	not stated	none
10	Burt RL *et al* [37]	1956	not stated	atherosclerosis
11	Tapp & Hickling [38]	1968	ruptured aortic artery aneurysm	neurofibromatosis
12	Thomas & Gillis [39]	1970	ruptured uterine artery	none
13	Cohen SG *et al* [19]	1972	not stated	non specific
14	Patterson WM [40]	1973	abruptio placentae	non specific
15	Saleh & McLead [41]	1977	not stated dysplasia	fibromuscular
16	Love WK *et al* [24]	1981	abruptio placentae	none
17	Barrett JM *et al* [42]	1981	?ruptured renal artery aneurysm	non specific
18	Hidai H *et al* [43]	1985	ectopic pregnancy dysplasia	fibromuscular
19	Cohen & Shamash [44]	1987	abruptio placentae	none stated
20	Schoon IM *et al* [25]	1988	abruptio placentae	non specific
21	Dayton B *et al* [1]	1990	ruptured Rt renal artery aneurysm post arteriography	none
22	Murakami M [45]	1993	not stated	
23	Whiteley MS *et al* [16]	1994	ruptured ectopic pregnancy	none
24	Rijbroek A *et al* [15]	1994	not stated dysplasia	fibromuscular
25	Current study	1997	ruptured Rt renal artery aneurysm	none

Treatment

Because of the late diagnosis of rupture in the antepartum period, 10 nephrectomies were performed in the 24 cases we reviewed. In only four cases was repair of the renal artery performed.[1,16,24,25]

The management of an unruptured renal artery aneurysm in pregnancy or in the woman of child-bearing age who may become pregnant is elective surgery because of the increased risk of rupture with potentially fatal consequences. The management differs however, in the nonpregnant patient. Conservative management for calcified renal artery aneurysms less than 1.5 cm in diameter in the asymptomatic, nonhypertensive patient has been

to page 13

from page 12

recommended.[26,27,28] Indications for surgery include renovascular hypertension, flank pain attributable to the aneurysm, haematuria, aneurysm more than 2 cm in size (with or without calcification),renal infarction and lack of calcification in an aneurysm.[27,29] When elective surgery is performed, kidney sparing procedures are recommended. These would include excision of the aneurysm and primary or patch closure. Autotransplantation and bypass grafting are other options that are available.

Conclusion

Since Chisholm[30] reported the first case of rupture of renal artery aneurysm in pregnancy, 23 other cases have been published. The high maternal and fetal mortality rates have mainly been due to incorrect preoperative diagnosis, with most cases being managed as obstetric emergencies. We have provided the first case in which colour Doppler ultrasound was used to make a preoperative diagnosis and recommend its routine use in assessing the pregnant patient who presents with gross haematuria, acute flank pain and haemorrhagic shock.

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