

Intra-Parenchymal renal artery aneurysm in a young pregnant patient

Shariful Islam, Devin Hosein, Dave Harnanan, Dilip Dan

ABSTRACT

Introduction: Renal artery aneurysms are rare clinical entities with an estimated incidence of less than 1% with 10% of these aneurysms being associated with intraparenchymal aneurysms of the renal artery. The presentation of these aneurysms is highly variable. The consequences of missing the diagnosis can be catastrophic. Pregnancy increases the risk of rupture with these aneurysms and presents a dilemma for the managing physician. There are several methods of managing a renal artery aneurysm although no established guidelines exist. Surgical approaches range from endovascular techniques to open procedures such as ex-vivo aneurysmal repair with auto-transplantation, with a partial or total nephrectomy being reserved as techniques of last resort.

Case Report: We present a case of a 36-year-old primigravid female with a left renal artery aneurysm unsuitable for endovascular therapy. Attempt was made for laparoscopic donor nephrectomy, ex-vivo aneurysm repair with auto transplantation; however laparoscopic left nephrectomy was performed because of the intraparenchymal location of the aneurysm.

Conclusion: Renal artery aneurysms should always be considered as a differential in patients with secondary hypertension. Close follow up is required if detected in pregnant patients as these increased the risk of fatal outcomes. Attempt should be taken to preserve the kidney function. A multi-disciplinary approach is required to decrease morbidity and mortality in these patients.



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CASE REPORT

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donar nephrectomy, ex-vivo aneurysm repair with auto transplantation; however laparoscopic left nephrectomy was performed because of the intraparenchymal location of the aneurysm. **Conclusion:** Renal artery aneurysms should always be considered as a differential in patients with secondary hypertension. Close follow up is required if detected in pregnant patients as these increased the risk of fatal outcomes. Attempt should be taken to preserve the kidney function. A multi-disciplinary approach is required to decrease morbidity and mortality in these patients.

Keywords: Ex-vivo repair of renal artery aneurysm, Hypertension in pregnancy, Intraparenchymal renal artery aneurysm, Laparoscopic donar nephrectomy, Renal artery aneurysm

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INTRODUCTION

Renal artery aneurysms are an uncommon clinical entity with an estimated incidence of less than 1% in the general population, with intraparenchymal renal artery aneurysms being rarer, noted to occur with 10% of renal artery aneurysms [1–2]. The presentation of these aneurysms are highly variable and nonspecific with hypertension being the most common sign. As a result, they are usually detected incidentally while investigating

other unrelated abdominal pathologies. Treatment requires a multidisciplinary approach with the goal of preserving kidney function where possible. Pregnancy complicates management as there is an increased risk of rupture and associated mortality, therefore, elective termination could be offered in such circumstances. Treatments described include endovascular modalities, ex-vivo aneurysmal repair with auto transplantation, partial or total nephrectomy. We report a case of a 36-year-old primigravida with a left sided intraparenchymal renal artery aneurysm.

CASE REPORT

A 32-year-old primigravida at eight weeks gestation presented to the obstetric service with a three-day history of left loin, lower abdominal and back pain. She is a known hypertensive and has a history of chronic back pain for five years which worsened with pregnancy. She had no associated PV bleeding, hematuria or dysuria. The rest of the history and examination were unremarkable. Urinalysis showed no evidence of hematuria or other abnormalities, a full blood count and serum creatinine were within normal limits. An ultrasound performed revealed a 3.5x2.0x4.5 left renal artery aneurysm. After lengthy discussion with the patient, the pregnancy was terminated on advice from the obstetrician. The patient was then referred to the urology and vascular surgery service and underwent a CT angiogram and DTPA renal scan.

Computed tomography angiogram (Figure 1) revealed a 2.5 cm (AP) x 2.7 cm (TS) x 5.3 cm saccular aneurysm occupying most of the left hilum and arising for the outer branch of the upper pole of the left inter-lobar artery with three 4–7 mm saccular aneurysms in the upper inter-lobular artery. There was global dilation of the left renal artery. There was no stenosis, thrombosis or active extravasation noted. The DTPA scan of the kidneys showed the function of the left and right kidneys at 60% and 40%, respectively.

The case was discussed at a multidisciplinary team meeting involving urologists, interventional radiologists, vascular and general surgeons and it was decided that the aneurysm was not suitable for stenting or embolization. The decision was made for an elective laparoscopic nephrectomy with ex-vivo aneurysmal repair and autotransplantation. Unfortunately on exploration, the aneurysm was found to be entirely intraparenchymal (Figure 2). Hence a laparoscopic left nephrectomy was performed, which was uneventful. The patient was discharged on day-2 postoperatively.

At 2 years follow-up our patient became normotensive without the need of further medication there was also resolution of her back pain. The patient is currently planning to become pregnant again.



Figure 1: Computed tomography angiogram with 3D reconstruction demonstrating large left intraparenchymal artery aneurysm (Inter-lobar artery) with multiple 4–7 mm aneurysm of upper inter-lobular artery.



Figure 2: Left nephrectomy specimen dissected to show the large intraparenchymal aneurysm.

DISCUSSION

Aneurysms of the renal vasculature are rare. The first case was reported by Rouppe in 1770 where he described a sailor who fell on his right flank resulting in rupture of a renal artery aneurysm to which he later succumbed [3]. The incidence of these aneurysms is approximately

0.01%–0.9% on autopsy studies [4]. This however, is on the rise, with the more frequent use of diagnostic imaging, as was shown in this case, with an estimated incidence of 2.5% in hypertensive patients undergoing angiography [2]. The average age of diagnosis is 46 with a female preponderance of 68% [5]. The right kidney is more likely to be involved accounting for 60% of cases with bilateral involvement found in 7% of cases. Intraparenchymal aneurysms account for less than 10% of renal artery aneurysms [2].

Renal artery aneurysms can be classified as true or false aneurysms. True aneurysms involve all layers of the artery and are usually fusiform or saccular with 90% being extra parenchymal [6]. They are usually caused by connective tissue disorders such as Ehlers-Danlos syndrome or other diseases like fibromuscular dysplasia [7–9]. Pseudoaneurysms on the other hand do not include all layers of the vessel wall. They tend to be saccular with causes including trauma such as blunt abdominal or iatrogenic during endovascular procedures. Other causes include infection resulting in mycotic aneurysms, Kawasaki disease, arterial dissections and finally some aneurysms have no identifiable causative factor [10–14].

The presentation of patients with renal artery aneurysms can be highly variable and non-specific and their symptoms may be attributed to other common conditions. The majority of patients tend to be asymptomatic with the aneurysms being discovered incidentally while the patient is undergoing diagnostic imaging for an unrelated condition [7]. Of the patients who are symptomatic, 90% have hypertension. The cause of the hypertension is a source of controversy, some authors state it is a product of anatomical kinking of the aneurysm resulting in decrease renal perfusion other theories include co-existing stenosis or emboli from a thrombus in the aneurysm resulting in infarction and decreased perfusion. Arterial deformation of the aneurysm is also a postulated cause for secondary hypertension [8]. All theories, however, ultimately result in activation of the renin angiotensin aldosterone system which leads to systemic hypertension. Our patient was hypertensive but there was no evidence of stenosis or distal infarction to suggest thromboembolism on CT scan. Other documented symptoms include flank pain and hematuria [7].

The decision to repair an aneurysm is controversial as no well established guidelines exist. The risk of rupture of a renal artery aneurysm is small. However, there is increased risk of rupture during pregnancy with an associated higher mortality [1, 15–17]. Based on review of available literature, repair should be considered under the following conditions [6, 7, 18, 19]:

- I. Asymptomatic patients with an aneurysm diameter > 2.0 cm
- II. Selected symptomatic patients such as hypertension refractory to medical management, evidence of stenosis or thromboembolism

III. Patients wishing to become pregnant

IV. Any ruptured aneurysm

Many surgical techniques have been described to treat renal artery aneurysms with the ultimate goal of preservation of renal function. The choice of technique will be affected by the type of aneurysm, extent of involvement, kidney function, whether the aneurysm is ruptured and the available expertise. Options include endovascular or open surgery. Endovascular techniques include coil or liquid embolization or placement of a stent graft which has the added advantage of treating both stenosis and an aneurysmal segment [20–24]. Some aneurysms may not be amenable to endovascular treatments, such as large aneurysms with intraparenchymal extension or the expertise or equipment may not be available. In such cases, open surgical repair is performed. Such repairs include aneurysmectomy with patch repair or excision of the aneurysmal segment with graft placement. There have even been cases where extracorporeal repair was performed with autotransplantation [25].

Unfortunately, there are instances where the preservation of the kidney is technically impossible. In such cases, a partial or even a total nephrectomy must be performed. Therefore, every patient must be counseled and consented for the possibility of a total nephrectomy. Total nephrectomies should be considered in patients with multiple or a large intraparenchymal aneurysm not amenable to partial nephrectomy, as in our case, a non-functioning kidney or a hemodynamically unstable patient secondary to a renal artery aneurysm rupture.

Follow-up of these patients have shown in selected patients resolution of hypertension with variable rates of 50–100% [18, 19, 26, 27]. There is no data on recurrence of these aneurysms after repair. As a result, more data is required to determine a standard of care for these patients. Ultimately, the decision for treatment will involve a multidisciplinary approach with appreciation of the risk factors and involvement of the patient at each step.

Learning points

- Intraparenchymal renal artery aneurysm (IPRAA) is very rare with widely variable presentation.
- May be a cause of secondary hypertension.
- Pregnancy is associated with high risk of rupture
- Treatment is multidisciplinary with the goal of preserving kidney function.
- Endovascular techniques, ex-vivo aneurysmal repair with auto-transplantation, partial or total nephrectomy can be performed in selected patients.

CONCLUSION

Renal artery aneurysms seldom make the differential list of a managing physician due to its rarity. However, it

should always be considered in patients with secondary hypertension as treatment of the aneurysm will result in a decrease or normalization of blood pressure. Also, if detected in pregnant patients, close follow-up is required as these patients are at increased risk of adverse outcomes. There are many surgical techniques currently available for treatment of these aneurysms with the ultimate goal of preserving kidney function. It should be highlighted however, that a multi-disciplinary approach is required as early as possible to decrease morbidity and mortality in these patients.

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Written consent was obtained from the patient for publication of this case.

Author Contributions

Shariful Islam – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Devin Hosein – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Dave Harnanan – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Dilip Dan – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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