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Case Report (Open Access)

A CASE REPORT OF A TREATED SPONTANEOUS RENAL ARTERIOVENOUS FISTULA

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ABSTRACT
We report a case of a 54 year old woman with mild hypertension, who presented with sudden onset of painless hematuria.
Preliminary ultrasonography revealed, unilateral obstructed kidney with blood clots within the pelvicalyceal system and ureter.
On contrast enhanced CT imaging, an arterio-venous fistula was diagnosed. This was treated with embolization with coils in the IR suite, with successful outcome. The etiology in our cases was narrowed down to idiopathic or congenital.
The clinical presentation of sudden spontaneous painless hematuria raises an alarm to exclude a malignancy and/ or to look for an underlying obstructive / non- obstructive lesion of the renal parenchyma/ collecting system and ureter. However, we need to remember, to review hilar and intra renal vessels on imaging to evaluate the possibility of a vascular lesion / malformation.

Keywords: *Arteriovenous Fistula, Coil Embolization, Spontaneous Hematuria, Dense Persistent Nephrogram, Unilateral Renal Obstruction*

INTRODUCTION
Arteriovenous fistula is defined as abnormal communication between artery and vein, resulting in shunting of blood from artery into the vein. Renal arteriovenous fistula is a rare entity with overall prevalence of approximately 0.04% in the general population. It is two times more common in females as compared to males (Dönmez *et al.*, 2008) and is seen between the age group of 30 to 40 years. Right kidney is more frequently involved as compared to the left kidney (Dönmez *et al.*, 2008).
Most cases of renal arteriovenous fistulas are clinically asymptomatic, therefore the true prevalence is further masked (Rhee *et al*, 2019). Symptomatic patients commonly present with flank pain, sudden onset of gross or microscopic hematuria, hypertension and symptoms of congestive heart failure. Occasionally, palpable thrill and auscultable bruit are noted in the flank region on physical examination (Miyuki, *et al*. 2016, Rhee *et al*, 2019). Arterial hypertension occurs secondary to decreased renal arterial flow, causing renin release and thus activating renin-angiotensin pathway. High output heart failure is also a common manifestation associated with high flow arteriovenous fistula as a result of increased venous return from the fistula.
Based on the etiology, renal arteriovenous fistula are classified as congenital (14– 27%), acquired (70–80%) or idiopathic (3%) (Rhee *et al*, 2019). The congenital arteriovenous fistula originates during development and maturation of the arterial and venous systems between 4th and 8th weeks of embryonic development. The causes of acquired AVF include iatrogenic (nephrostomy, renal biopsy, renal surgery, malignancy) or traumatic (blunt or penetrating trauma).

CASE REPORT
A 54 year old lady presented with sudden onset painless hematuria. She had mild tachycardia with elevated blood pressure of 160/100 mm Hg.

A preliminary USG revealed a moderately hydronephrotic right kidney with moderate hydroureter. Echogenic blood clots were seen within the pelvicalyceal system, ureter as well as the bladder. No mass lesion was found in the renal parenchyma or pelvicalyceal system.
On further evaluation with contrast-enhanced CT study, a fistulous communication was identified between the interpolar division of the right renal artery and draining right renal vein on the arterial phase, which was seen as a focal intense blush of contrast. This was suggestive of an arteriovenous fistula. No evident features of atherosclerosis/vessel wall disease were identified on the arterial phase. On venous images, rest of the renal vasculature was normal on either side.
A dense persistent nephrogram was seen of the right kidney with delayed excretion of contrast, which is suggestive of acute renal obstruction. The presence of large blood clots within the proximal ureter as well as distal segment of the ureter explained the hydronephrosis.
The left kidney was normal. The urinary bladder also showed luminal blood clots, however, wall thickness and distensibility was normal. There was no other abnormality in the abdomen and pelvis on the CT imaging.
The patient's baseline cardiac evaluation with 2D-ECHO was suggestive of changes associated with hypertensive disease, which were probably due to long standing duration of essential hypertension. ECG was within normal limits. No clinical features of congestive cardiac failure were seen.
With the diagnosis of right renal arteriovenous fistula on the CT study, the patient was posted as a candidate for post-DSA embolization of the arteriovenous fistula. DJ stent was inserted to retrieve blood clots and obstruction. On catheter renal angiography, the lesion was confirmed. Coil embolization was performed under fluoroscopic guidance.
The procedure was uneventful. Post procedure, the patient was monitored, mild hypertensive state persisted, which was followed up on medical management at the institution.
The hematuria ceased, the blood clots and the urinary tract dilatation improved over time. Patient was followed up at 1 month, 3 months and 6 months. USG did not reveal obstruction of the right kidney. No aliasing of colour flow / shunt was seen on the colour doppler. The outcome of the intervention was successful.

DISCUSSION

Arteriovenous fistula is defined as abnormal communication between artery and vein, resulting in shunting of blood from artery into the vein. Renal arteriovenous fistula is a rare entity with overall prevalence of approximately 0.04% in the general population. It is two times more common in females as compared to males (Dönmez *et al.,* 2008) and is seen between the age group of 30 to 40 years. Right kidney is more frequently involved as compared to the left kidney (Dönmez *et al.,* 2008).
Most cases of renal arteriovenous fistulas are clinically asymptomatic, therefore the true prevalence is further masked (Rhee *et al*, 2019). Symptomatic patients commonly present with flank pain, sudden onset of gross or microscopic hematuria, hypertension and symptoms of congestive heart failure. Occasionally, palpable thrill and auscultable bruit are noted in the flank region on physical examination (Rhee *et al*, 2019, Miyuki *et al*., 2016). Arterial hypertension occurs secondary to decreased renal arterial flow, causing renin release and thus activating renin-angiotensin pathway. High output heart failure is also a common manifestation associated with high flow arteriovenous fistula as a result of increased venous return from the fistula.
Based on the etiology, renal arteriovenous fistula are classified as congenital (14– 27%), acquired (70–80%) or idiopathic (3%) (Rhee *et al*, 2019). The congenital arteriovenous fistula originates during development and maturation of the arterial and venous systems between 4th and 8th weeks of embryonic development. The causes of acquired AVF include iatrogenic (nephrostomy, renal biopsy, renal surgery, malignancy) or traumatic (blunt or penetrating trauma).
Based on the angiographic imaging findings, renal AVFS are classified as: - (Miyuki *et al*. 2016)
a) Traumatic: Direct fistulous formation between a single artery and a single draining vein.
b) Non-traumatic:
- Type I: Single or a few arteries shunting to a dilated single draining vein.
- Type II: Multiple arterioles shunting to a single dilated draining vein.

- Type III: Multiple shunts between the arterioles and venules, forming a complex vascular network.

The congenital renal arteriovenous fistulas mostly show knotted, cricoid structure associated with multiple intercommunicating fistulae between the feeding artery and the draining vein. The congenital AVFs present with hematuria, as it is present just beneath the mucosa of urothelial of collecting system. However, the acquired or idiopathic renal AVFs show single communication between the feeding artery and the draining vein (Rhee *et al.*, 2019, Dönmez *et al.*, 2008). In our case described above, as the patient presented with sudden onset of painless hematuria and hypertension, the etiology can be either congenital or idiopathic.

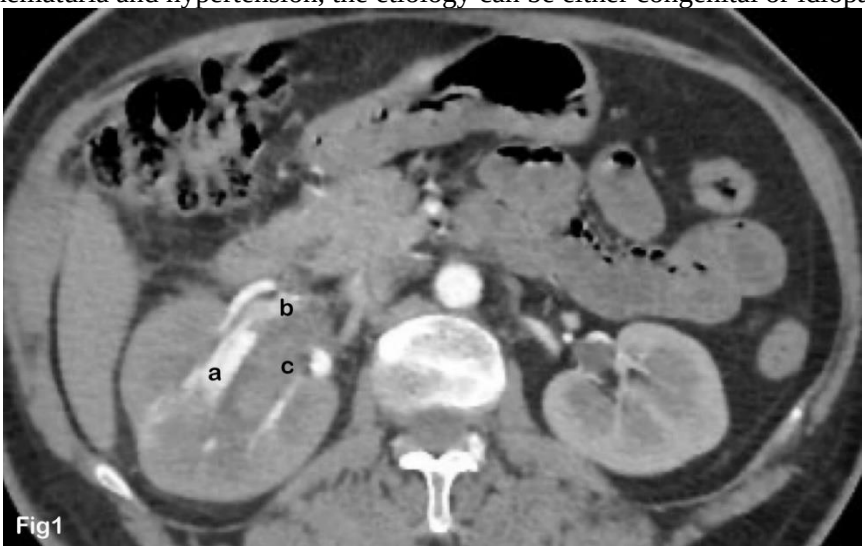

Figure1: A 54 year old female with spontaneous renal arterio-venous fistula.
Axial contrast enhanced CT image in the arterial phase showing prominent intra renal arteries (a) of the right kidney with dense contrast and the draining vein(b) showing faint contrast. Hydronephrosis and blood clot is seen within the renal pelvis in this section (c).

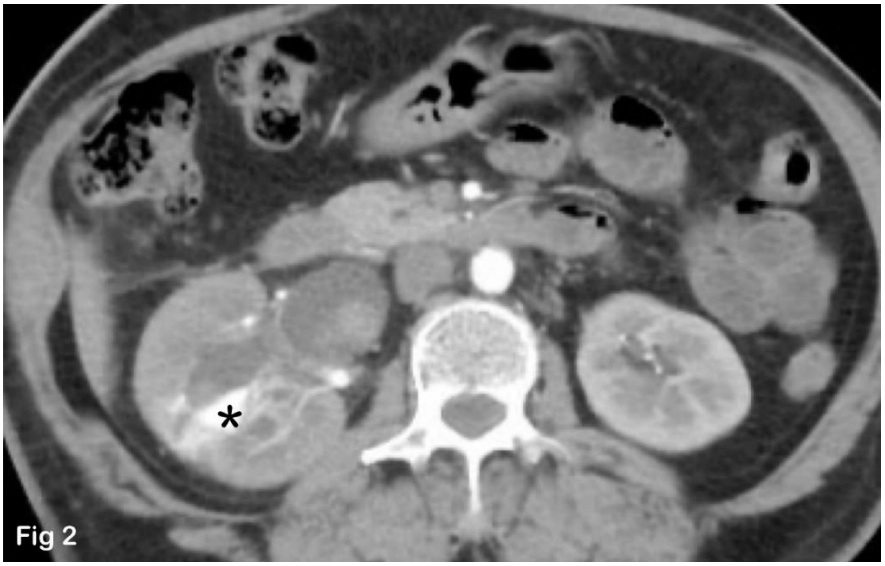

Figure 2: A 54 year old female with spontaneous renal arterio-venous fistula.
Arterial axial CT image showing focal intensely enhancing vascular blush, which is the site of arterio-venous communication and represents an AVF (*) between the intra renal branch of the right renal artery and the draining right renal vein.

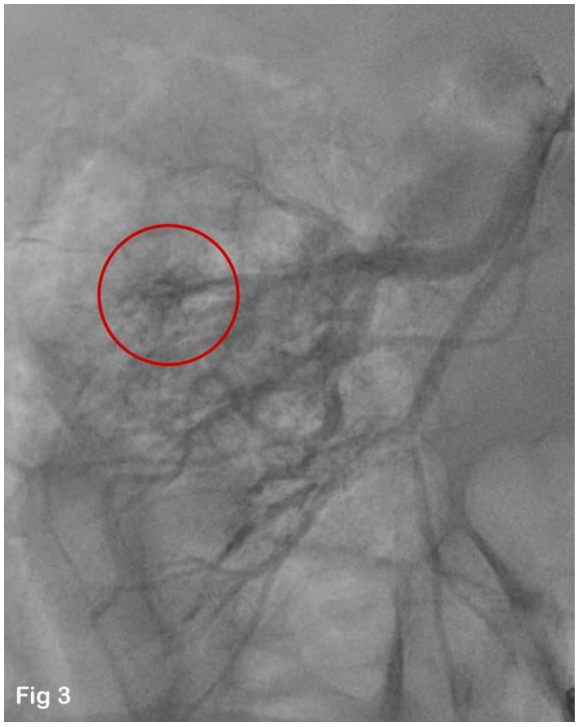

Figure3: A 54 year old female with spontaneous renal arterio-venous fistula
Renal angiogram confirmed the site of the right renal AVF between the right intrarenal artery in the right interpolate cortex and the draining vein.

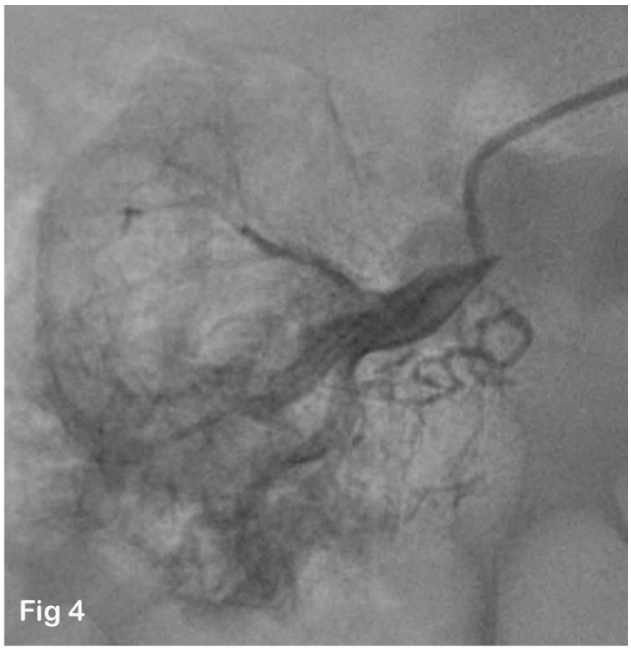

Figure 4: A 54 year old female with spontaneous renal arterio-venous fistula
Renal angiogram of the right renal AVF post coil embolization, with no residual lesion.

The first line modality for imaging in the patient with suspected renal arteriovenous fistula is USG colour doppler. The gray scale USG shows a relatively well defined hypoechoic cystic lesion. This is can be easily misdiagnosed as a renal cyst. On colour doppler study, mosaic pattern and perivascular soft tissue colour speckling is seen. Spectral analysis demonstrates decreased arterial resistance, increased flow velocity and arterial wave forms in the outflow vein. Digital subtraction angiography (DSA) is the gold standard in the diagnosis of renal AVFs. It can demonstrate small feeding vessels and intralesional feeding vessels, which are important in the planning of interventional procedure. Contrast enhanced CT and MR imaging techniques can be used for follow up studies (Dönmez *et al.,* 2008). However, in our patient, due to the presence of large blood clots within the pelvicalyceal system, the lesion was not detected on Doppler ultrasonography. The lesion was identified and diagnosed on contrast-enhanced CT. Evaluation of the lesion on cross sectional study helps to exclude a lesion in the contra lateral kidney, multiplicity of lesion as well as the presence of an occult neoplasm. Transcatheter intra-arterial occlusion procedures have drastically replaced conventional surgical methods of treatment of the renal arteriovenous fistula (Rhee *et a*, 2019, Baleato *et al.,* 2009.). Several embolic materials are used for the embolization of renal AV fistulas, including particles (gelatin sponge particles and polyvinyl alcohol [PVA] particles), coils (pushable and detachable coils), vascular plugs, detachable balloons, and liquid materials (absolute ethanol, NBCA, and ethylene vinyl alcohol copolymer [Onyx]) (Miyuki, *et al.,* 2016). The material used for the procedure depends upon the type of the AVFs. Surgery is considered as the standard treatment. Nephrectomy is the most common surgical procedure for the treatment of large aneurysmal and cirsoid lesions (Crotty *et al.,* 1993). Spontaneous resolution of the arteriovenous fistulas are reported in few cases, usually the post-traumatic case (Rhee *et al.,* 2019).
Our patient, showed good response to the embolization procedure performed, with a successful outcome.

CONCLUSION

The clinical presentation of sudden spontaneous painless hematuria raises an alarm to exclude a malignancy and/ or to look for an underlying obstructive / non- obstructive lesion of the renal parenchyma/ collecting system and ureter. We need to remember, to review hilar and intra renal vessels on imaging to evaluate the possibility of a vascular lesion / malformation. The use of colour Doppler in a case of hematuria is also relevant, as a renal AVF can mimic a renal cyst in echotexture on ultrasonography.

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