**Complication after embolization of a complex renal vascular malformation**

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**Introduction**

Renal arteriovenous fistulae (AVFs) and associated aneurysmal malformations, located either intra- or extrarenally [1], are rare and characterized by a single cavernous communication between the artery and the venous outflow [2]. Clinically they may present with flank bruit and pain, haematuria, uncontrolled hypertension [3] and high-output cardiac failure. Whereas compromise in renal and cardiac function in cases of large shunt volumes is caused by AVFs, rupture with severe bleeding is most feared as a complication of renal aneurysms.

We describe the diagnostic, therapeutic and follow-up radiological findings in an unusual case, highlighting a possible complication after embolization therapy in a patient with von Willebrand factor (vWF) syndrome.

**Case**

At the end of 2003, a 57-year-old woman was referred for control of a renal artery aneurysm diagnosed incidentally in 1991 as she had felt palpitations in the left flank for 2 weeks. Concomitant morbidities were varicosis, depression and a vWF syndrome type I. The patient was asymptomatic for the aneurysm in 1991. Contrast-enhanced axial–spiral computer tomographic (CT) measurements from 1991 showed a 2 cm diameter arterial aneurysm, an AVF in the lower pole of the left kidney and an associated 5 cm aneurysm of the renal vein (Figure 1a). An intravenous angiography mirrored this finding.

At the end of 2003, magnetic resonance angiography of the renal arteries was performed and revealed a 2 × 2.5 × 3 cm renal artery aneurysm, a following AVF and a venous aneurysm of up to 4.5 cm maximum (Figure 1b). Angiography confirmed the huge aneurysmal renal AVF in the left hilum with high flow evidenced by nearly simultaneous arterial and venous opacification and a progression of the arterial aneurysmal part of the malformation. The arterial neck actually measured 2 cm, growing up to a maximum of 3.5–4.0 cm; the venous aneurysm behind the AVF was 5.0 cm. We observed poor filling of the intrarenal branches and nearly no nephrogram. There was no marked dilatation of the suprarenal vena cava. With duplex ultrasound, a shunted blood volume of >500 ml/min could be calculated. Together with the vascular surgeons and nephrologists, a decision was made to proceed with minimally invasive therapy and the patient was electively embolized in February 2004 after premedication with Minirin (desmopressin-acetate) in the Department of Radiology.

Via a left common femoral approach, a Cordis 7French (F) renal double curve (RDC) guiding catheter was introduced in the saccular part of the aneurysm using a 0.035 inch wire (Figure 2a). A 5F Cordis Cobra catheter was subsequently used for the imaging of the whole aneurysm. Contrast-enhanced axial–spiral computer tomographic measurements from 1991 showed a 2 cm diameter arterial aneurysm, an AVF in the lower pole of the left kidney and an associated 5 cm aneurysm of the renal vein (Figure 1a). An intravenous angiography mirrored this finding.

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the renal artery was superselectively catheterized with a Cordis Maastransit Microcatheter via the 0.018 inch wire and coiling was performed for the renal artery aneurysm through the stent struts of the previously implanted PRECISE stent. In total, 27 fibred complex helical platinum coils from Boston Scientific, with 3 and 4 mm in the helical configuration and an original length of 30 mm were used to achieve complete stasis in the arterial aneurysm. Thereafter, 0.8 ml of histoacryl (tissue adhesive substance enbucrilat, n-butyl-cyanoacrylate) and lipiodol (oily contrast media) in a mixture of 1:2 was administered into the coiled area. After a control angiogram with a 7F Cordis Prostar catheter and the application of a closer system (Abbott, USA), the procedure ended without any complications (Figure 2b).

Unfortunately, the patient complained about pain in the left flank 3 days later, accompanied by an elevation of C-reactive protein up to 6.7 mg/dl and creatine kinase up to 334 U/l max. Therefore, a contrast-enhanced spiral CT scan was done, revealing a thrombosis in the left renal vein reaching into the inferior vena cava (Figure 3a). An incipient infarction of the left kidney involving ~20–30% of the parenchyma could be seen. Despite the increased bleeding risk due to the vWF syndrome, the anticoagulation with enoxaparine, 2 × 0.7 ml subcutaneously was prolonged. On the evening of the same day, the patient...
developed fever. Uroculture was not performed but the patient was placed on antibiotics (clindamycin and a third generation cephalosporin) for another 3 days until the fever had broken and no more infection signs were seen in the blood work-up. Kidney function parameters were stable and the patient did not show any signs of dehydration. A spiral CT performed 2 weeks after the intervention showed an absence of thrombus in the inferior vena cava and a residual thrombosis in the renal vein with re-established flow. Parenchymal infarction was constant with 20–30% of the kidney volume without any functional deterioration. Three days later, heparin was discontinued. The next control follow-up CT scan of this patient obtained 3 months after the intervention showed a partially organized thrombus in the left renal vein and a parenchymal retraction in the left kidney as a correlate of infarction. Eight months after the procedure, the last control CT scan revealed almost completely normal blood flow in the renal vein with a minimal residual stenosis and the absence of new thrombus material (Figure 3b).

Discussion

AVFs develop secondary to a trauma, renal surgery, percutaneous needle biopsy, bacterial endocarditis, vasculitis and renal cell carcinomas; they constitute ~75% of AV communications. They often (~70%) heal spontaneously.

In contrast, a possible aetiology for renal arteriovenous malformations (AVMs) is thought to be a pre-existing renal arterial aneurysm, which has eroded into an adjacent vein. AVMs are classified as congenital or idiopathic [3,4].

Congenital AVMs constitute ~20% of the total AV communications. They are often located near the collecting system and are described as cirsoid with multiple AV communications; they commonly present clinically with gross haematuria. Idiopathic malformations as in our case are rare, with only ~5% of all renal AV communications. They are characterized as single cavernous channels with defined aneurysmal arterial and venous elements. Their clinical appearance is often associated with abdominal bruit, hypertension and/or high-output cardiac failure.

A useful diagnostic tool is ultrasound with colour Doppler imaging and contrast-enhanced CT or magnetic resonance imaging (MRI), whereas arteriography best demonstrates the detailed anatomy of the feeding and draining vessels and is necessary for the accurate planning of the intervention.

The indication for the treatment of such lesions is the presence of clinical symptoms such as flank bruit and pain, haematuria, uncontrolled hypertension and high-output cardiac failure or the prevention of vessel rupture. Respectively, treatment would consist of an occlusion of the fistula if problems are primarily volume associated or in a switch-off of the arterial aneurysmal part to prevent eventual rupture. Risk of rupture in AVMs is supposed to be slight, but a rupture rate of up to 24% is described for single arterial aneurysms larger than 2 cm in diameter [5]. Dzsinich et al. recommend not to treat small non-symptomatic renal arterial aneurysms <1.5 cm or those with circumferential calcification [1]. However, asymptomatic ones of >2.5 cm in diameter [1], or symptomatic ones as well as those aneurysms with documented expansion, peripheral renal parenchymal embolization and those in women anticipating pregnancy should be treated to prevent rupture.

As our patient did not have volume-associated problems, such as cardiac compromise but presented with a documented expansion of size of the arterial aneurysm beyond the critical margin, it was decided to embolize only the arterial aneurysmal part of the malformation without damaging the renal parenchyma. The aim was to prevent a possible rupture and not to occlude the fistula, which, by increasing the pressure in the arterial part, would have impaired the risk of rupture. Additionally, endovascular embolization...
therapy was thought to minimize the risk of post-operative inadequate coagulation/bleeding especially due to the vWF syndrome of the patient relative to an alternative surgical approach.

Successful endovascular repair and embolization has been described in some case reports for renal AVMs and renal artery aneurysms [2,3,6,7]. In terms of embolization, there are a number of options available. Some AVMs have been treated successfully with superselective embolization therapy, for example in pregnant women [3], with, for example, metallic coils. However, there are few reports on successful endovascular treatment of renal artery aneurysms such as, for example, in wide neck aneurysms with a TrisPan coil (neck-bridge device developed by Boston Scientific Target) and Guglielmi detachable coils [6] or in a patient with fibromuscular dysplasia treated with a Jostent stent-graft [7]. In this patient, satisfying long-term patency results and a successful aneurysm exclusion were seen at 16 months after stenting with a Jostent [7], suggesting long-term validity.

In our case, we used a non-covered stent in order not to compromise blood flow via the side branches visualized in the angiography. In order to obtain stasis in the aneurysmal blood flow, we had to embolize the saccular part through the stent struts. We believe that particulate embolus alone, such as polyvinyl alcohol, absorbable gelatin sponges and gelatin spheres, are not suitable for embolization of large aneurysms unless used in combination with other occlusive devices such as metallic coils. We also believe that use of particles would not be likely to result in occlusion of such aneurysms, even with a tight coil nest, and would have the potential for embolization to the lungs. We thus preferred commercial platinum coils to begin a coil nest.

We required deployment of 27 complex helical 3 and 4 mm platinum coils and the administration of 0.8 ml of histoacryl and lipiodol in a 1:2 mixture to decrease the aneurysmal flow significantly. Thrombin could probably have passed through the coil nest and the stent struts and may have caused serious systemic thromboembolic complications such as pulmonary embolization or renal infarction by embolization material displacement. Potentially, coils could also serve as a nidus for pulmonary embolism [8], but the stent struts were thought to serve as a barrier and would prevent this complication.

In our case, we unfortunately saw an extensive thrombosis detected by CT as a useful tool for general diagnostic screening and the detection of vascular patency. Fever and infection signs were thought to have occurred mainly secondary to the parenchymal infarction and thrombosis and not through primary renal infection where the development of a thrombus is generally also possible as a complication [9]. Angiography and local thrombolysis would have been considered, especially if the stented arterial part were occluded, but the possible effects of a local thrombolysis were feared considering the patient’s coagulation disorder. Probably there was thrombus development starting in the arterial embolized part of the malformation, appositionally growing through the AVF into the venous part of the malformation. Thrombus formation might have been started because of a possible partial protrusion of several coils through the mesh of the stent, the thrombogenicity of the embolization material and the relative post-interventional stasis and changed haemodynamics. Additionally, the low dose and not full dose post-interventional heparinization together with probable vWF-related alterations in coagulation might have played a significant role. Generally we believe that sufficient post-interventional anticoagulation and an accurate selection of embolic agents/particles should be mandatory. It is surely advantageous to use as few embolic agents/particles as possible to prevent thrombus formation, and probably longer coils could have secured better positioning inside the aneurysm to prevent an eventual partial protrusion as an accelerator for local thrombosis. In the literature, inferior vena cava thrombosis after embolization therapy is extremely rare. Harada et al. reported a case of a 50-year-old woman with renal AVM treated with transarterial embolization who developed ipsilateral flank pain and haematuria 12 months after treatment. As malignancy could not be ruled out, a nephrectomy was performed. During nephrectomy, an extensive AVM and inferior vena cava thrombus was palpable and an additional thrombectomy was performed [10].

Nevertheless, while considering these points, we believe that catheter angiography should be the first choice of intervention, not only for the occlusion of AVMs but also in the occlusion of dangerous aneurysms.

Conflict of interest statement. None declared.

References

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