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Case Report

Recurrent urinary retention due to clots caused by a congenital renal arteriovenous malformation that forms a complex vascular network: Report of two cases

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Abbreviations & Acronyms
AVM = arteriovenous malformation
CECT = contrast-enhanced computed tomography
NBCA = *n*-butyl 2-cyanoacrylate

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Introduction: Repeated urinary retention due to clots caused by congenital renal arteriovenous malformation is rare.
Case presentation: A 40-year-old woman (case 1) and a 66-year-old man (case 2) experienced recurrent urinary retention due to clots. Neither patient had a history of renal trauma nor was taking any medications. Contrast-enhanced abdominal computed tomography revealed a large hematoma in the renal pelvis and ureter with no masses, stones, or vascular lesions and only dilated blood vessels in the arterial phase in case 1. Angiography of the kidney in both patients revealed an arteriovenous malformation, and embolization of the arteries feeding the arteriovenous malformation was performed. Soon after embolization, hematuria disappeared completely with no recurrence for 5 years in case 1 and 2 years in case 2.
Conclusion: Congenital renal arteriovenous malformation should be considered as a cause of recurrent clot retention, and angiographic embolization of the feeding arteries can be an effective treatment.

Key words: angiography, embolization, gross hematuria, renal arteriovenous malformation, urinary retention.

Keynote message

We report two cases of clot retention caused by congenital renal arteriovenous malformation (AVM). This type of AVM should be considered as a cause of repeated clot retention, and angiography can be valuable for diagnosis and treatment in suspicious cases.

Introduction

Congenital renal AVMs are rare, with a prevalence of approximately 0.04% in the general population.[1] However, they may cause flank pain, massive hematuria, and high-output heart failure, and hence, urgent treatment is required in some cases. Renal AVMs are usually diagnosed by ultrasonography or abdominal CECT, but in some cases, especially in Type III AVMs, in which multiple shunts are present between the arterioles and venules that form a complex vascular network, diagnosis may be difficult. We report two cases of renal AVM that were difficult to diagnose by CECT but could be diagnosed and treated with the aid of angiography.

Case presentations

Case 1

A 40-year-old woman presented to the emergency department with gross hematuria leading to urinary retention caused by acute clots. She had no history of renal trauma, urolithiasis, or medications. Vital signs were stable, and blood and urine tests showed no evidence of infection. Her biochemical and coagulation parameters were within normal limits. Urinary cytology

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was negative. Cystoscopy showed a large number of blood clots in her bladder and bleeding from the right ureteral orifice but no obvious tumor or other source of bleeding. CECT revealed a large number of blood clots in the right renal pelvis and ureter. However, no sources of bleeding such as from urolithiasis or tumorous lesions were found. Therefore, we diagnosed her as having idiopathic renal bleeding and expected that spontaneous hemostasis would occur following conservative treatment with a hemostatic agent. However, urinary retention with clots recurred again, and her anemia worsened. To investigate the cause of the renal bleeding, reexamination of the CECT images with a radiologist revealed dilated blood vessels in the arterial phase (Fig. 1a), which were considered to indicate an arteriovenous fistula. We suspected a congenital renal AVM and performed angiography for both diagnosis and treatment. Angiography of the right renal artery revealed an arteriovenous fistula with a complex vascular network in a branch of the right middle pole renal artery, which we diagnosed as a Type III renal AVM (Fig. 1b). Selective renal artery embolization using absolute ethanol was performed, and we confirmed that the abnormal blood vessels had disappeared (Fig. 1c). Gross hematuria completely disappeared soon after the embolization, and there has been no recurrence for 5 years.

Case 2

A 66-year-old man presented to the emergency department with complaints of gross hematuria with urinary retention. He also had no history of renal trauma, urolithiasis, or medications. Vital signs were stable, and blood tests showed only slight anemia. Urinary cytology was negative. CECT showed a hematoma in the right renal pelvis but no urolithiasis or tumorous lesions (Fig. 2a). Cystoscopy showed no obvious tumor or sites of bleeding in the bladder. Retrograde pyelography revealed a shadow defect suspected to be a hematoma in the right renal pelvis (Fig. 2b). Right renal pelvis cytology was negative. Although we initially diagnosed idiopathic renal bleeding, urinary clot retention recurred, and spontaneous hemostasis would not be expected during follow-up. Reexamination of CECT images in this case showed no obvious findings, but based on our experience in case 1, angiography was performed to rule out a congenital renal AVM. In fact, angiography revealed an arteriovenous fistula with a complex vascular network in a branch of the right upper pole renal artery, and the patient was diagnosed as having a Type III congenital renal AVM (Fig. 2c). He underwent successful selective renal artery embolization with absolute ethanol (Fig. 2d), and his hematuria disappeared immediately thereafter. He was discharged 5 days after embolization, and there has been no recurrence of hematuria for 2 years to date.

Discussion

Renal AVMs are classified into three types according to angiographic morphology[1]: type I, a single or few arteries (<4) shunt into a single draining vein; type II, multiple arterioles shunt into a single draining vein; and type III, multiple shunts are present between the arterioles and venules that form a complex vascular network (Fig. 3). In types I and II, pre-existing aneurysms are considered to have formed the arteriovenous shunts. In type III, there are no capillaries, and arteries and veins form shunts through abnormal blood vessels to create an abnormal vascular network. Types I and II account about 75% and type III for about 25% of all AVMs. Each type has its own characteristic angiographic appearance. In types I and II, a large aneurysm is formed, whereas in type III, an abnormal vascular network is formed between a large number of meandering and dilated feeding arteries and draining veins. The formation of large aneurysms as in types I and II has a large effect on the cardiovascular system; therefore, clinical symptoms such as abdominal bruits, hypertension, and heart failure are often observed.[2] In type III, hematuria is the most common clinical symptom because the abnormal vascular network is located just below the urothelium.[2] In fact, hematuria has been reported in 72% of type III AVMs compared to 21% of type I AVMs.[2] Both of the present

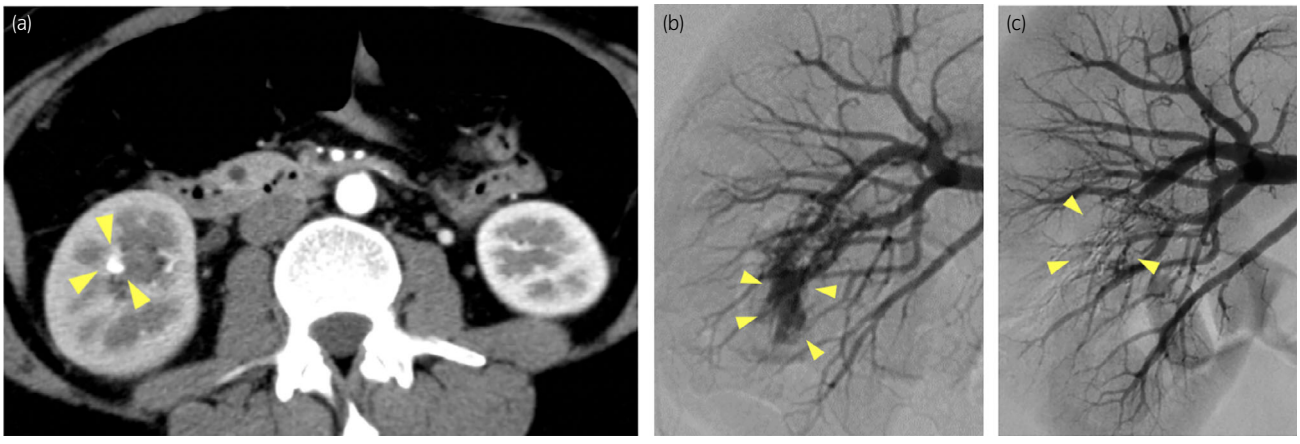

Fig. 1 Contrast-enhanced computed tomography (a) and angiography (b, c) in Case 1. (a) The yellow triangles indicate dilated blood vessels in the arterial phase. (b) The yellow triangles indicate an abnormal complex arteriovenous network in a branch of the right middle pole renal artery. (c) The abnormal blood vessel network disappeared after selective renal artery embolization.

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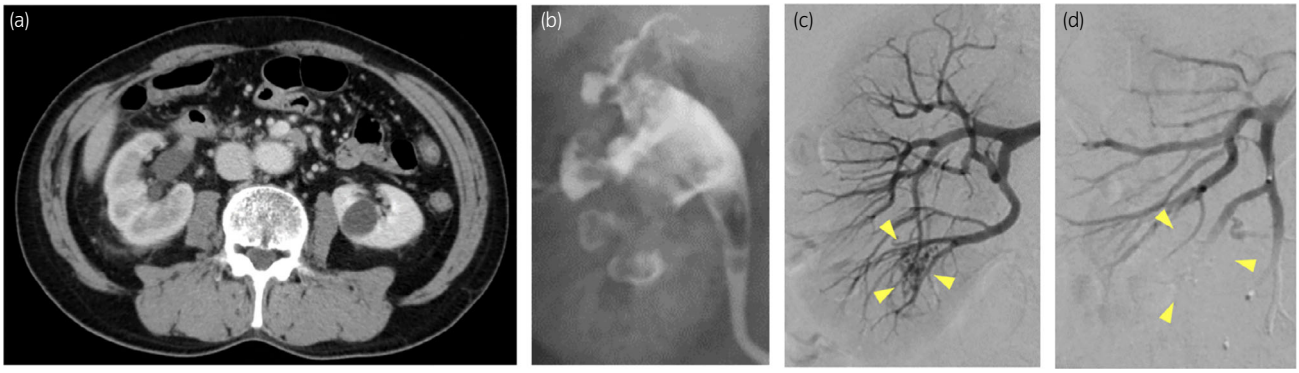

Fig. 2 Contrast-enhanced computed tomography (CECT) (a), retrograde pyelography (b), and angiography (c, d) in Case 2. (a) CECT showed no obvious findings. (b) Retrograde pyelography only revealed a shadow defect suspected of being a hematoma in the right renal pelvis. (c) Angiography revealed an arteriovenous fistula with a complex vascular network in a branch of the right upper pole renal artery. (d) The abnormal blood vessel network disappeared after selective renal artery embolization.

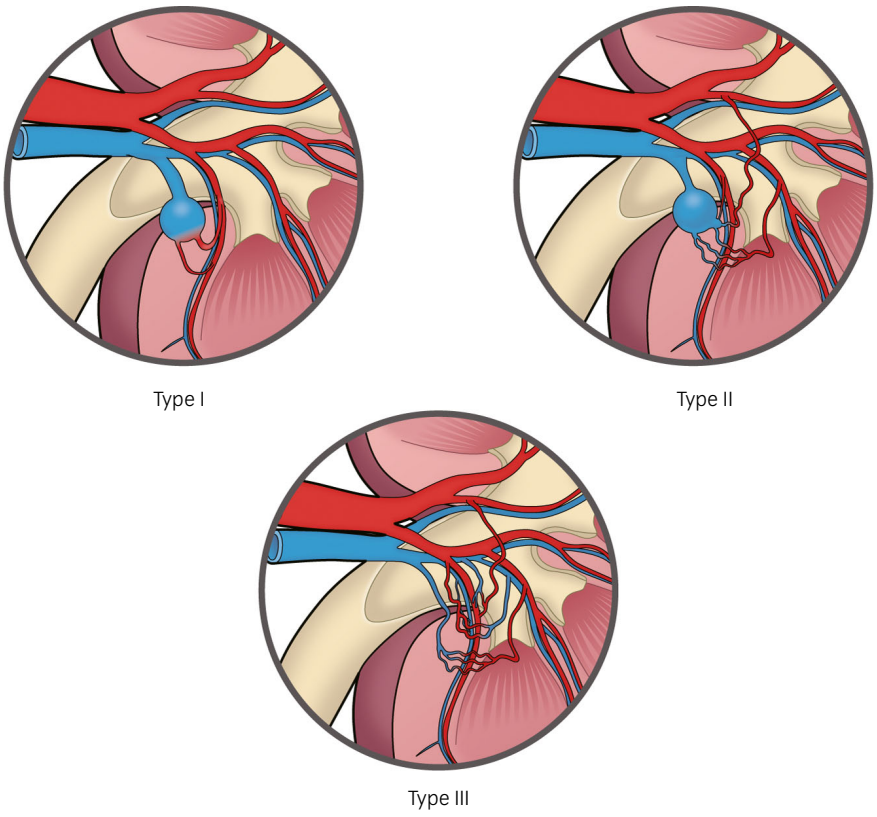

Fig. 3 Illustration of the three types of renal arteriovenous (AV) shunts. Type I: direct AV fistulas with single or a few separate feeding arteries shunting directly to a single draining vein with a large venous sac. Type II: multiple arterioles shunting to a single dilated draining vein forming a large venous sac into which multiple feeders converge. Type III: multiple shunts between arterioles and venules with a complex vascular network (or nidus). This type usually consists of multiple feeding arteries and multiple draining veins.

patients had type III AVMs in which no large aneurysms are formed; therefore, it was difficult to make a diagnosis with CECT. Several cases of renal AVMs have been reported that could not be diagnosed by CT examination or ultrasonography but were diagnosed by angiography, and most of them were caused by an abnormal vascular network corresponding to a type III AVM.[3–6] Color Doppler ultrasonography is inexpensive and the preferred initial diagnostic method for evaluation of the kidneys, and specific findings such as colored mosaic patterns can lead to the diagnosis of type I and II AVMs.[7,8] However, type III is difficult to diagnose with ultrasonography, likely because the complex, tiny, abnormal vascular networks are difficult to recognize. To our knowledge, there are no case reports of type III AVMs diagnosed with ultrasonography. We concluded that congenital renal AVMs should be considered as a cause of recurrent clot

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retention, and angiography can be valuable for diagnosis and treatment in suspicious cases, even though angiography is highly invasive compared to ultrasonography and CT.

Absolute ethanol was used for embolization in both patients according to the surgeon's experience and preference. Although absolute ethanol is effective in the treatment of AVMs and results in a low rate of recurrence, serious complications and recurrence can occur, so careful follow-up is essential.[9,10] One report mentioned that complete obliteration of type III shunts can be obtained with NBCA by following some key points: use a relatively low concentration of NBCA (20%–33%), control flow of all feeding arteries by balloon occlusion of the renal arterial stem, administer a slow and prolonged injection, and wedge the catheter tip deeply into the proper feeding artery.[1]

Author contributions

Makoto Ishii contributed to acquisition of subjects and/or data, data analysis, interpretation, and preparation of manuscript. Wataru Nakata contributed to study concept and design, acquisition of subjects and/or data, data analysis and interpretation, and critical review of the manuscript. Yuki Horibe, Go Tsujimura, Yuichi Tsujimoto, Mikio Nin, and Masao Tsujihata contributed to acquisition of subjects and/or data.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an institutional reviewer board

This study was approved by the Ethics Committee of Osaka Rosai Hospital (approval number: 2020-137).

Informed consent

Informed consent was obtained from the patients included in this study.

Registry and the registration no. of the study/trial

Not applicable.

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Editorial Comment

Editorial Comment from Dr. Kato to Recurrent urinary retention due to clots caused by a congenital renal arteriovenous malformation that forms a complex vascular network: Report of two cases

Renal arteriovenous malformations (AVMs) are a rare disease that is characterized by anastomotic and abnormal communications between renal arterial and venous systems.[1] Among them, congenital renal AVMs are rare renal vascular abnormalities which is present in approximately 20% of all renal AVM cases with a prevalence of 0.04% in general population and usually remain asymptomatic until the age of 30 or 40.[2] These are a rare but sometimes fatal cause of hematuria because about 30% of patients may present with signs of congestive heart failure from high-output fistulas, and up to 50% with cardiomegaly and hypertension.[3] So far, congenital AVMs are subdivided into three categories; type I, a single or few arteries (<4) shunt into a single draining vein; type II, multiple arterioles shunt into a single draining vein; and type III, multiple shunts are present between the arterioles and venules that form a complex vascular network.[4] Especially, type III renal AVMs pose a diagnostic challenge in clinical settings.

In the present study, Ishii et al. reported two renal congenital renal AVM cases that were adequately diagnosed by renal angiography.[5] Importantly, a contrast enhanced computed tomography was not enough to make a diagnosis of these AVM cases since type III AVMs consists of an arteriovenous fistula with a complex vascular network in a small branch of

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