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CASE REPORT

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Renal arteriovenous fistula discovered ~2 years after renal biopsy: A case report

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Key Clinical Message

Although percutaneous renal biopsy is considered safe, this invasive procedure has complications such as renal arteriovenous fistula (RAVF). Even if complications such as RAVFs are not observed early after renal biopsy, considering the possibility of delayed renal hemorrhage, follow-up with ultrasound after renal biopsy even in asymptomatic cases could be important.

Abstract

Although percutaneous renal biopsy is considered safe, this invasive procedure can lead to complications such as renal arteriovenous fistula (RAVF). RAVF occurs when some arteries and veins communicate in the absence of capillaries in the renal hilum or renal parenchyma. It was previously thought to be relatively rare; however, with advances in imaging diagnostics, it is sometimes found asymptomatically. In addition, renal biopsy is the most common cause of acquired RAVF. In this case, RAVF was discovered 2 years after renal biopsy. Late-onset RAVF is scarce. This case highlights that even if complications such as RAVFs are not observed early after renal biopsy, considering the possibility of delayed RAVF, follow-up with ultrasound could be important.

KEYWORDS

children, renal arteriovenous fistula, renal biopsy

1 | INTRODUCTION

Percutaneous renal biopsy (PRB) provides important information for diagnosis, management, and prognosis of patients with renal diseases. Complications after renal biopsy have decreased because of improvements in imaging techniques and biopsy needles.

Major adverse events are noticed in 5%–6% of the patients, whereas deaths and nephrectomy rates are around 0.9%–1%.

Of these major complications, ~60% were identified 4 h after biopsy, ~70% after 8 h, ~90% after 24 h, and approximately 10% after 24 h.[1,2] Although serious complications beyond 24 h after PRB are considered rare, occurrences after 24 h are rarely reported.[3]

Renal arteriovenous fistula (RAVF) occurs when some arteries and veins communicate in the absence of capillaries in the renal hilum or renal parenchyma. It was previously thought to be relatively rare; however, with advances in imaging diagnostics, it is sometimes found

asymptomatically.[4] In addition, renal biopsy is the most common cause of acquired RAVF.[5]

We diagnosed a late-onset RAVF post PRB when hematuria relapsed. The literature on late-onset RAVF is scarce. Herein, we report and discussed a case of late-onset RAVF.

2 | CASE

A 9-year-old Japanese boy was admitted to our hospital for microhematuria investigations. His medical history included the development of abdominal pain and purpura in his lower extremities at the age of 6 years. Thereafter, he had urinary protein 3+ and hematuria; thus, prednisolone (PSL) 1 mg/kg/day was administered for suspected Henoch–Schönlein purpura nephritis (HSPN). However, nephrotic-level urinary protein and hypoalbuminemia did not improve, so he was referred to our hospital. No abnormalities were observed in prothrombin time (PT), activated partial thromboplastin time (APTT), bleeding time, and ultrasound (US) before renal biopsy.

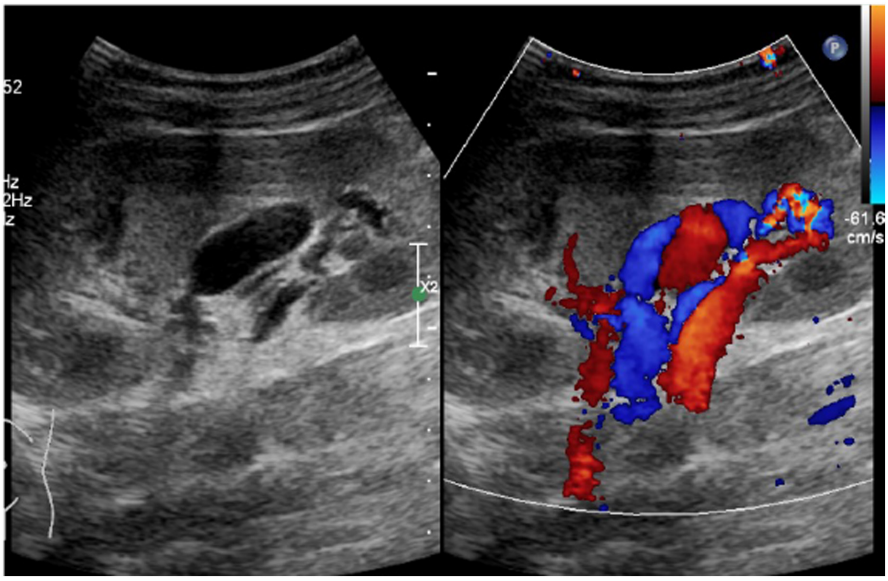

FIGURE 1 Abdominal ultrasonography. An echogenic luminal structure was observed in the right kidney. A blood flow signal was observed at the same site, suggesting vascular malformation. The most dilated vessel diameter was 11 mm.

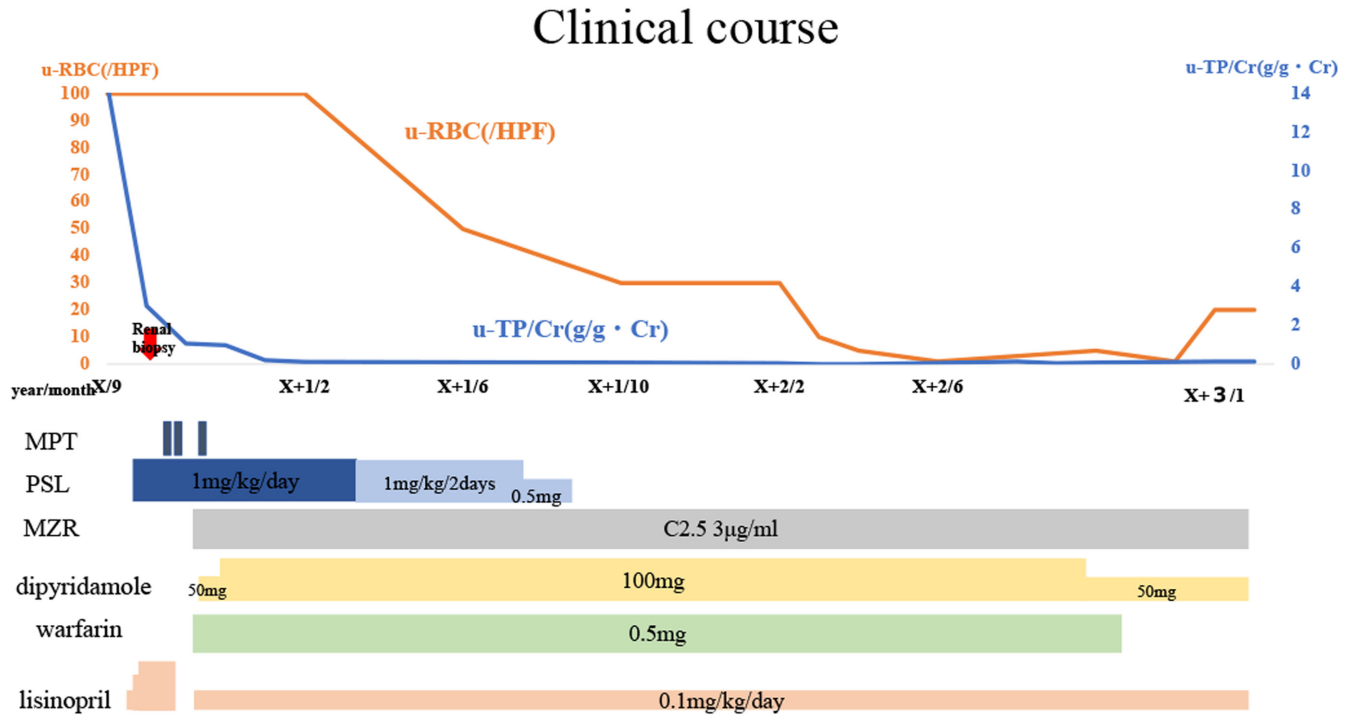

FIGURE 2 Clinical course. Urinary protein–creatinine ratio (u-TP/Cr), urinary red blood cells (u-RBC), and the treatment up to embolization.

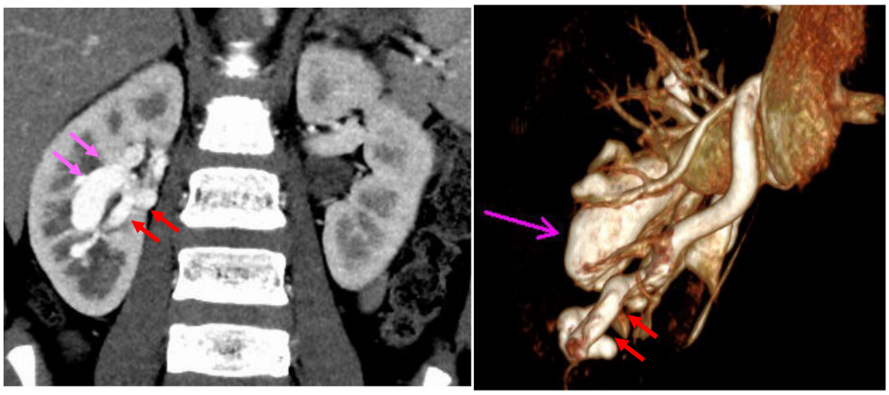

FIGURE 3 Abdominal contrast-enhanced computed tomography. Dilated (pink arrow) and tortuous (red arrow) vasculature observed at the right renal hilum.

In the same year, PRB was performed from the lower pole of the right kidney, using an automated device with a 16-gage and 16-mm (stroke length) needle under real-time US guidance by pediatricians, and a specimen was obtained. No gross hematuria or hematoma was noted after renal biopsy. The patient stayed for ~3 days in strict bed rest after renal biopsy according to our hospital renal biopsy protocol.

After renal biopsy, he was administered carbazochrome sodium sulfonate hydrate (50 mg/day) and tranexamic acid (250 mg/day) for 3 days. He did not complain of subjective symptoms such as back pain.

The pathological diagnosis was HSPN (International Study of Kidney Disease in Children Classification grade IIIa), and three courses of methylprednisolone (90 mg/kg/3 days) pulse were administered. Combination therapy with PSL (2 mg/kg/day), mizoribine (100 mg/day), dipyridamole (100 mg/kg/day), warfarin (0.5 mg/day), and lisinopril (0.1 mg/kg/day) were started based on the Guidelines for the treatment of childhood IgA nephropathy,[6] which yielded a good response: urinary protein reported negative 5 months later and hematuria disappeared 21 months later.

Hematuria relapsed 6 months after complete remission was achieved: without any other symptoms, so an US was performed showing turbulent blood flow with aliasing in Doppler (Figure 1). The patient was hospitalized for vascular embolization under general anesthesia. Figure 2 shows the clinical course.

On admission, the patient was 135.5 cm (0.5 SD) tall and weighed 31 kg (0.2 SD), with a blood pressure of 94/52 mmHg, heart rate of 78/min, and temperature of 36.8°C. No peripheral edema, purpura, neuropathy, or arthritis was observed.

Laboratory tests revealed white blood cell count of 5000/μL, hemoglobin of 12.9 g/dL, and platelet count of 28.8×10^4/μL. In addition, his serum albumin level was 4.5 g/dL; blood urea nitrogen, 14.9 mg/dL; creatinine, 0.33 mg/dL; Cre-estimated glomerular filtration rate (e-GFR) (Creatinine-based equation to estimate the glomerular filtration rate in Japanese children), 145.98 mL/min/1.73 m^2; aspartate aminotransferase, 27 IU/L; alanine aminotransferase, 21 IU/L; and C-reactive protein, 0.01 mg/dL. His activated partial thromboplastin time and prothrombin time were 28 s (normal 27–40 s) and 13.5 s (normal 10–13 s), respectively. His urinary protein–creatinine ratio was 0.09 g/day. Sediments contained 11–30 erythrocytes per high-power field, and dysmorphic red blood cells were detected.

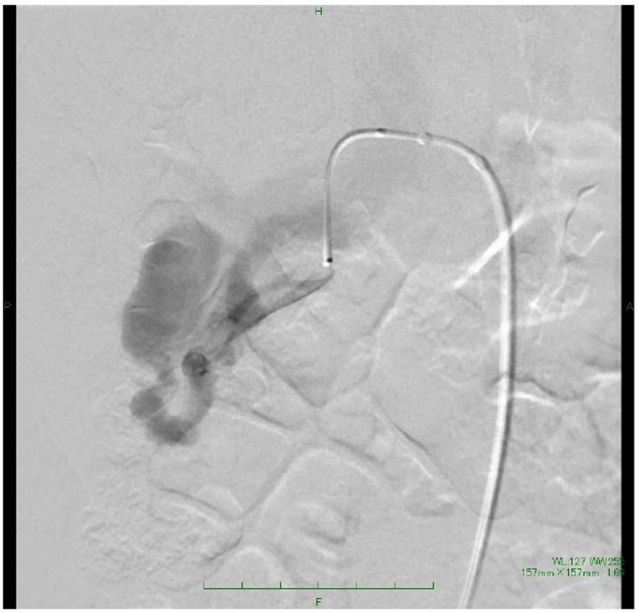

FIGURE 4 Arterial angiography before TAE. Dilated inflow and outflow vessels observed in the right lower pole of the kidney.

Abdominal contrast-enhanced computed tomography showed dilated and tortuous vasculature at the right renal hilum, suggesting a RAVF. Morphologically, it was judged to be of an aneurysmal type (Figure 3). Arterial angiography revealed an RAVF and dilated inflow and outflow vessels in the right lower pole of the kidney (Figure 4). Transcatheter renal arterial embolization (TAE) was performed using a micro-coil (diameter, 5 mm; length, 5 cm; Penumbra Inc.),

three packing coils (diameter, 1 mm; Penumbra), and four coils (diameter, 4 mm; length, 10 cm; AZUR, Terumo). After TAE, the dilated draining renal vein and pseudoaneurysm were no longer detected (Figure 5). After the RAVF was embolized, the macroscopic hematuria subsided. Two months after the embolization, ultrasonography showed no dilated blood vessels and a coil in the lower pole (Figure 6).

In radioisotope (99mTc-DTPA/MAG3 renal scintigraphy), the e-GFR of the right, left, and both kidneys were 64.9, 74.7, and 144.5 mL/min (219.6 mL/min/1.73 m^2), respectively; the right renal function was slightly lower than left renal function (Figure 7). Figure 8 shows the clinical course.

3 | DISCUSSION

Renal biopsy is a very useful tool for diagnosis, prognostic determination, and treatment guidance. Although percutaneous renal biopsy is considered safe, this invasive procedure has complications such as RAVF. Despite being rare, it occurs in 3%–5% of native kidneys and 10%–16% of the transplanted kidneys. If biopsies are performed with real-time ultrasound guidance and automatic needles, the incidence of RAVF can be reduced to <0.1%.[7–9] In addition to obtaining sufficient material for diagnosis, the procedure is considered successful if few adverse outcomes occur.[10]

RAVF is an abnormal connection between the arterial and venous systems. RAVFs have three types: congenital, idiopathic, and acquired. Acquired RAVFs are caused by trauma, inflammation, surgery, tumor, atherosclerosis, or percutaneous biopsy and accounts for 70%–80% of RAVF.[7,9,11] Of these, the most common is RAVF associated with PRB.[12] Idiopathic RAVF develops at some point in life but has no definite etiology.[13]

More than 95% of RAVFs after renal biopsy resolve spontaneously within 2 years.[14] Risk factors for RAVFs include hypertension, nephrosclerosis, renal fibrosis, arteriosclerosis, large needle (14 G), and non-echo-guided renal biopsy.[15] Although this case had no risk factors, the patient had IgA vasculitis. When hematuria was observed again, the patient was taking dipyridamole orally, so he had a predisposition to bleed easily.

The clinical diagnosis of RAVF can be difficult. Signs and symptoms include microscopic and macroscopic hematuria, refractory arterial hypertension, flank pain, and audible renal artery sounds caused by turbulent blood flow.[16,17] Moreover, 80% of RAVFs are asymptomatic and may not be diagnosed until symptoms appear. A follow-up US examination of the kidney soon after renal biopsy and 6 months later is recommended for early detection of non-resolving AVFs to prevent further complications.[18]

The goal of RAVF treatment is to eradicate symptoms and hemodynamic effects (arterial hypertension and heart failure) and ensure maximal preservation of the functioning of the renal parenchyma.[9,11,19]

Invasive options that may be required in refractory cases with severe hematuria or hemodynamic instability include coil embolization (~85% success rate in patients with acquired fistulas) and surgical nephrectomy.[20,21]

As for treatment, if the patient is asymptomatic, follow-up is possible, and if hematuria is mild immediately after the injury, conservative treatments such as rest or administration of hemostatic agents can be considered.

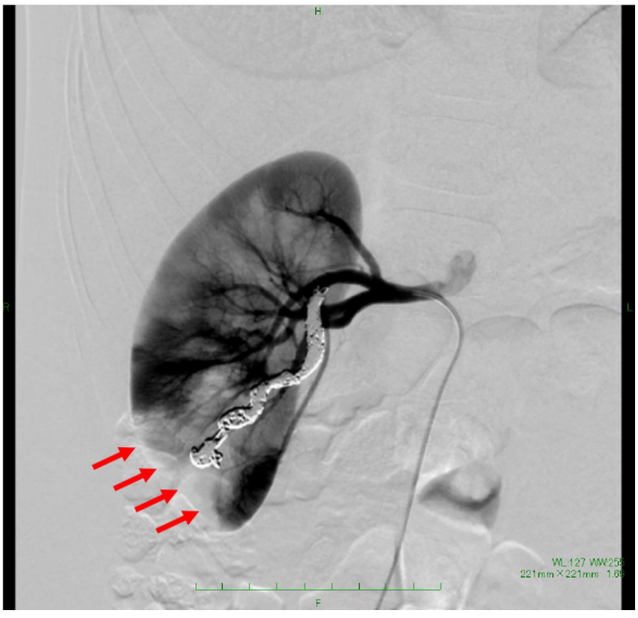

FIGURE 5 Arterial angiography After TAE. Dilated draining renal vein and pseudoaneurysm were no longer detected. An infarction site was found in about 1/8 of the right kidney (red arrow).

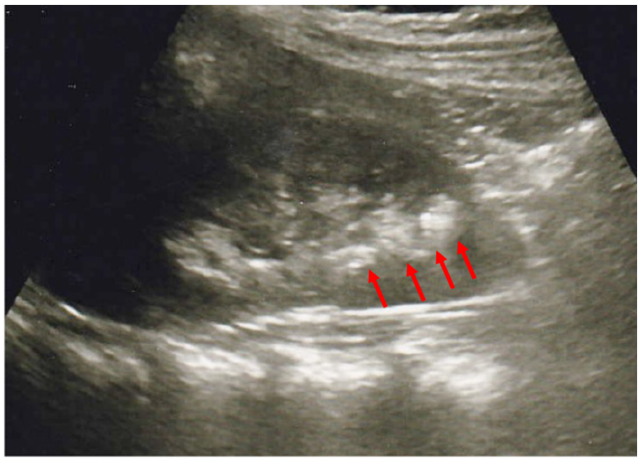

FIGURE 6 Abdominal ultrasonography. Two months after the embolization, ultrasonography showed no dilated blood vessels and a coil in the lower pole (red arrow).

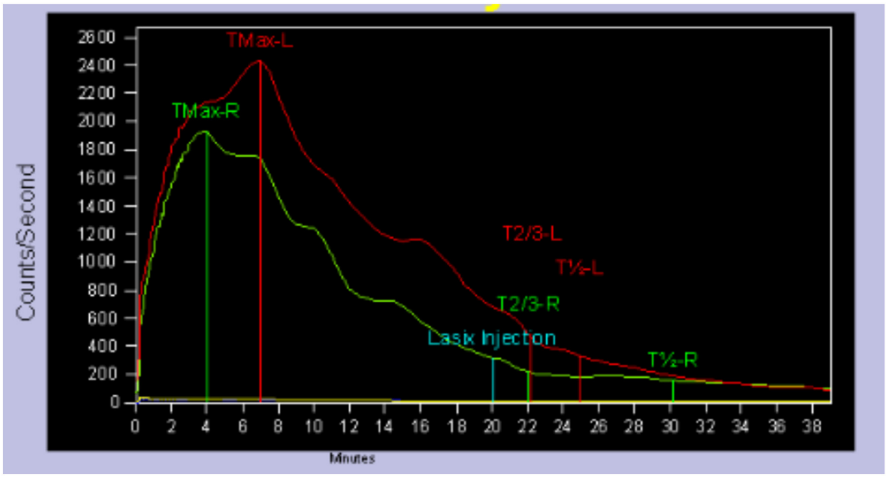

FIGURE 7 99mTc-DTPA/MAG3 renal scintigraphy. The GFR of the right, left, and both kidneys were 64.9, 74.7, and 144.5 mL/min (219.6 mL/min/1.73 m^2), respectively; the right renal function was slightly lower than left renal function.

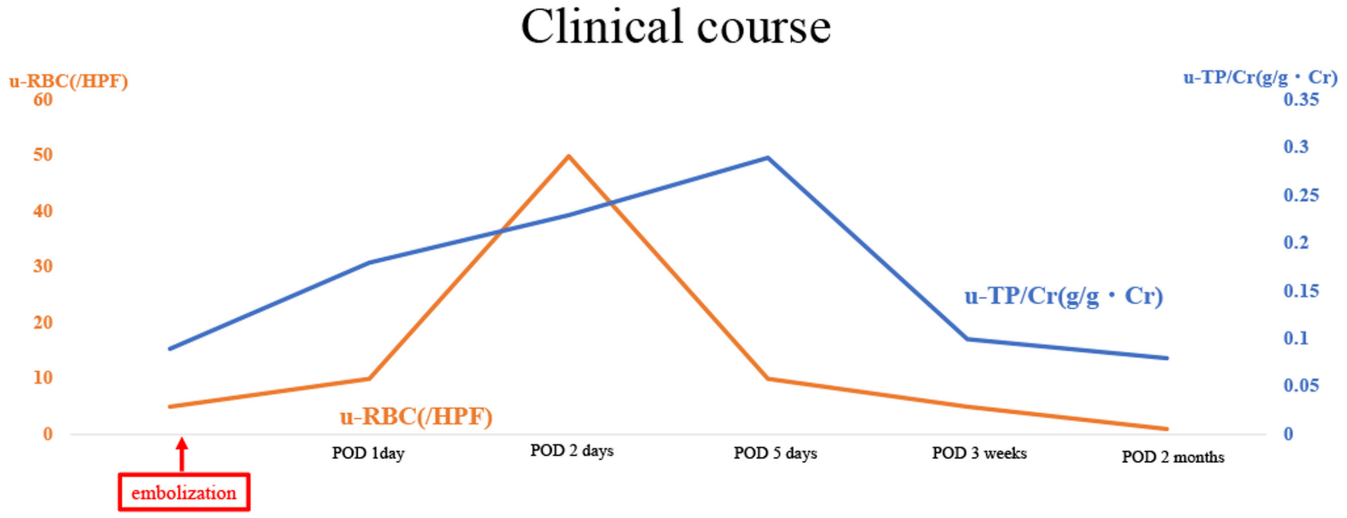

FIGURE 8 Clinical course. Clinical course with urinary protein–creatinine ratio (u-TP/Cr), urinary red blood cells (u-RBC), after embolization.

However, congestive heart failure, hypertension, persistent gross hematuria, and aneurysm formation with a risk of arteriovenous fistula rupture require surgery or endovascular treatment.

Renal arteriovenous fistula with aneurysm formation generally results in a large fistula, and treatment is indicated even in asymptomatic cases to prevent cardiovascular complications due to high shunt blood flow.[22]

Although this patient had no other symptoms, US examination was performed, which revealed RAVF because of microhematuria.

Table 1 summarizes the clinical features, pathological findings, and treatment details of the eight cases.[4,15,18,23] Patient age, sex, and duration of discovery varied. The underlying disease was mostly mesangial proliferative nephritis. Symptoms vary, but one patient was asymptomatic. Most of the cases were treated with embolization. One patient had undergone nephrectomy. All patients had a good prognosis.

In this case, no RAVF was detected by US in the early period after renal biopsy; however, it is possible that it was not detected appropriately and that the fistula was small at that time. Since the hematuria had improved once, there may have some kind of an episode such as exacerbation of the RAVF when hematuria was observed again. However, US follow-up was not performed, and the time of onset was unknown.

Severe renal bleeding leading to hematoma has been reported to usually occur within 24 h after renal biopsy. Considering the possibility of delayed RAVF, regular follow-up US after renal biopsy could be important even in asymptomatic cases.

4 | CONCLUSION

We experienced a case of RAVF, which was discovered with recurrence of hematuria 2 years and 3 months after

TABLE 1 Cases showing a long period of time from renal biopsy to RAVF discovery.

Age (years)	Sex	Primary disease	Time from renal biopsy to discovery RAVF	Symptoms	Treatment	Outcome	Ref. No
9	Male	Purpura nephritis	27 months	Microscopic hematuria	Embolization	Symptom improvement	This case
17	Female	Mesangial proliferative glomerulonephritis	36 months	Asymptomatic	Embolization	Symptom improvement	15 Matsuoka et al. (2018)
33	Female	IgA nephropathy	17 months	Proteinuria, decreased renal function	Embolization	Symptom improvement	18 Yang et al. (2008)
46	Female	IgA nephropathy	61 months	Gross hematuria, proteinuria, decreased renal function	Embolization	Symptom improvement	18 Yang et al. (2008)
48	Female	Chronic glomerulonephritis.	27 years	Decreased renal function, proteinuria, Vascular murmur	Embolization	Symptom improvement	4 Suzuki et al. (2016)
56	Male	IgA nephropathy	11 years	Palpitations, tachycardia, vascular murmurs	Nephrectomy	Symptom improvement	23 Yamaguchi et al. (1998)
66	Female	IgA nephropathy	13 years	Gross hematuria, decreased renal function	Embolization	Symptom improvement	4 Suzuki et al. (2016)

renal biopsy. Embolization was performed, the hematuria improved, but renal function has not deteriorated. Even if complications such as RAVFs are not observed early after renal biopsy, considering the possibility of delayed RAVF, regular follow-up with US after renal biopsy even in asymptomatic cases could be important.

AUTHOR CONTRIBUTIONS

Maki Oyama: Data curation; writing – original draft. **Hiroshi Tamura:** Data curation; writing – original draft. **Yuko Hidaka:** Data curation. **Keishiro Furuie:** Data curation. **Shohei Kuraoka:** Data curation.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

ETHICS STATEMENT

All the procedures performed in studies involving human participants were in accordance with the ethical standards of the Institutional Committee and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards (64th WMA General Assembly, Fortaleza, Brazil, October 2013). Informed consent for examinations and to publish their cases, including images, was obtained from patients and/or their family members.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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