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Case Report

Arteriovenous malformation in the kidney allograft: A rare cause of hematuria in the post-transplant patient☆,☆☆

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ABSTRACT

Gross hematuria is one of the most common complications in postrenal transplant patients, accounting for 12% of all renal recipients. The management plan in these cases varies depending on different entities, including infection, renal cell carcinoma, chronic graft rejection, kidney calculus, or recurrence of primary disease. On the other hand, vascular malformation like arteriovenous malformation was less likely to be mentioned due to a lack of consensus in the natural history, pathogenesis, and current management. In this article, we report a 62-year-old man presenting with spontaneous hematuria for a week and 2 days of anuria after 3 years of renal transplantation. Abdominal ultrasound and abdominopelvic computed tomography noted an obstruction of the renal pelvis due to blood clots without signs of vascular injuries. An emergency operation was performed to remove blood clots in the renal pelvis, but after that, hematuria was still recurrence. A digital renal graft subtraction angiography (DSA) revealed an arteriovenous malformation (AVM)in the kidney allograft. This lesion was then successfully selective embolized with glue. Given the high accu-

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racy of DSA, our case highlights the potential role of this imaging modality in diagnosing and treating AVM after failure with other modalities.

Introduction

Arteriovenous malformation (AVM) is the abnormal communication between artery and vein without bridging the capillary bed. Nidus is formed by a cluster of tortuous enlarging vascular networks, which is the significant difference between AVM and arteriovenous fistula. Congenital renal AVM is rare, less than 1% among the general population [1]. Post-renal transplanted AVMs are far less common. They can result from both congenital and acquired conditions like infection, trauma, or post-biopsy procedure [1,2]. Hematuria occurs due to the rupture of abnormal vessels into the urinary system and can immediately threaten hemodynamic shock and graft loss. Here, we aimed to describe a case of hematuria due to arteriovenous malformation in the kidney allograft, which was initially obscured in clinical practice. The vascular malformation was detected and successfully embolized using digital subtraction angiography (DSA).

Case presentation

A 62-year-old man presented to our hospital due to hematuria for a week and anuria for 2 days. His medical history included hypertension, type II diabetes mellitus, gout, and cirrhosis for many years relating to chronic hepatitis C infection, which were managed in a local hospital. Three years ago, the patient underwent a living–donor kidney transplantation due to end-stage renal disease. No abnormality of the donor's kidney was documented. The post-transplantation examination was uneventful. He had no renal biopsy history or other interventions in the kidney allograft. On clinical examination, he was conscious and had a mild fever (37.5°C–38 °C). His heart rate was 80 beats per minute, and his blood pressure was 120/80 mmHg. Laboratory tests revealed a high creatinine level (596 μmol/l) and uremia (22.75 mmol/l). The other tests encompass hyperglycemia (17,29 mmol/l), a slight increase in white blood cells (10.95 G/l) and neutrophils (81% of total), and a thrombocytopenia of 47 G/l. The coagulation tests were within the normal limits.

The abdominal ultrasound found signs of cirrhotic liver with irregular margins and heterogeneous echotexture, splenomegaly, and portal trunk dilatation of 15 mm in diameter. Bilateral native kidneys were hypotrophies with reduced cortical thickness and increasing renal cortical echogenicity. Renal allograft ultrasound highlighted renal pelvicalyceal dilatation with multiple high echogenicity structures inside, suggesting to occlusive blood clots. Doppler renal assessment detected no abnormality with a resistive index value of 0.86. Contrast-enhanced abdominopelvic computed tomography (CT) was performed to investigate the etiology of hematuria. It revealed that a high-density component (approximately 65 Hounsfield units) obstructed the pelvis and upper ureter of the kidney allograft. No renal mass or calculus was detected on CT images. The arteries of kidney allograft were not dilated on the arterial phase images, and neither pseudoaneurysm nor vascular malformation was identified (Fig. 1).

Due to the anuria, an emergency operation was performed to remove the renal pelvic blood clots and double J-stent placement. No abnormal vascularity or any causes of hematuria were found during the operation. Results of blood and urinary bacterial cultures, mycobacteria growth indicator tube

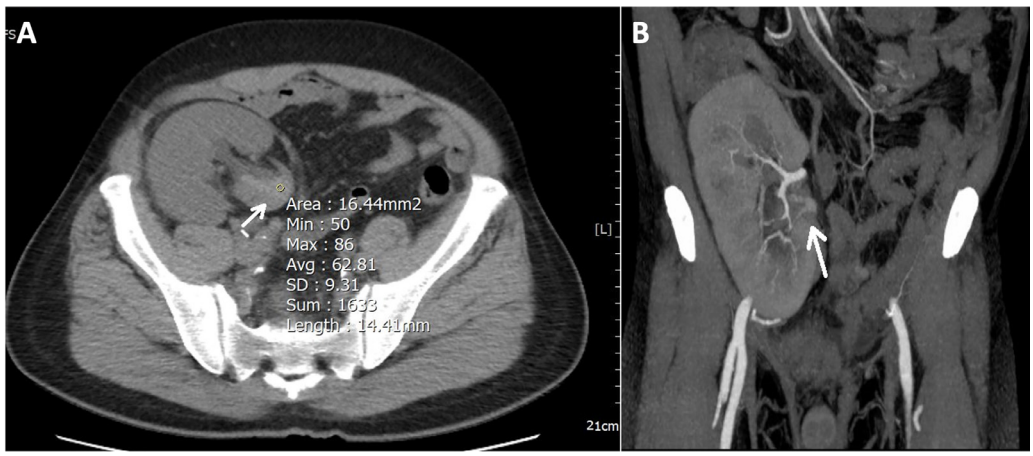

Fig. 1 – The pre- and post-contrast abdominal computed tomography images. (A) The pre-contrast image on the axial plane showed the dilation of the pelvis of the allograft kidney with clotting blood inside (arrow). (B) The post-contrast image on the coronal plane showed no visible vascular malformation with vague early vein enhancement on the arterial phase (arrow).

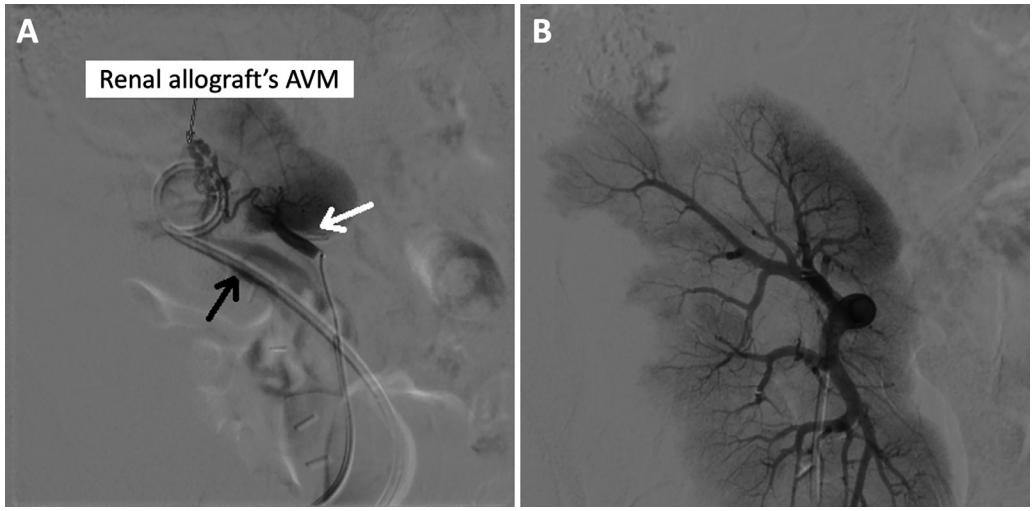

Fig. 2 – (A) The selective digital subtraction angiography showed a small nidus on the superior pole of the allograft kidney. The feeding artery (white arrow) and drainage vein (black arrow) were clearly demonstrated. (B) Postembolization image showed no residue of vascular malformation.

culture, and tests for cytomegalovirus and adenovirus were negative. Post-operative follow-up, however, the hematuria still recured without improvement; DSA was then indicated. On selective arteriography, A small AVM at the upper pole of the renal allograft was found on selective arteriography. Eventually, the AVM was embolized by a mixture of n-butyl-2-cyanoacrylate and lipiodol (1:4 ratio) (Fig. 2). Final angiography demonstrated that the AVM was eliminated, and no other abnormality was seen. The gross hematuria sign was reduced after several days, and the creatinine level decreased remarkably. The patient was discharged after ten days of hospitalization. At 1 month follow-up, the hematuria was resolved; the CT scan and renal creatinine detected no abnormality.

Discussion

Gross hematuria in renal transplant patients may result from a wide range of abnormalities, including infection, malignancy, chronic graft rejection, kidney calculus, or recurrent primary disease [3]. Jong-Hoon Lee et al. reported gross hematuria was in 126/1060 renal transplant recipients [2]. Among the hematuria population, fifteen of them had urolithiasis, 2 patients had bladder cancer, and only 1 was found to have renal cell carcinoma [2]. Another prospective study found that 37% of patients developed urinary tract infections after transplantation [4]. The isolated bacteria were Escherichia Coli (31.5%), Candida albicans (21.0%), and Enterococcus spp. (10.5%), which are typically responsible for hemorrhagic cystitis [4]. On the other hand, due to the daily use of immunosuppressive agents, adenovirus, cytomegalovirus, and herpes virus were frequent causes of hematuria. These etiologies, however, often present as microscopic hematuria [5]. The confirmed diagnosis requires a serum screen test, and a renal biopsy may need to be performed. The classical histopathology characteristics are tubular-interstitial necrosis with viral inclusion and lymphocytic infiltration. Anti-viral agents should be inserted to prevent irreversible damage to the allograft.

Renal cell carcinoma comprises 1.37% of renal transplant patients [6]. The incidence of developing malignancy in renal recipients is 15-fold higher than in the general population [6]. It can be explained by the causative of deoxyribonucleic acid damage and the interference of immunosuppressive drugs with the deoxyribonucleic acid repair mechanism [7]. Interestingly, the native kidneys are far more likely to be affected than the transplanted kidney (4.2% and 0.07%, respectively) [8]. This fact highlights the role of other pre-existing factors in the renal carcinogenesis process, like renal failure and long-term dialysis. There was no evidence of reducing renal cell carcinoma mortality, but routine screening should not be neglected in all cases [9]. Despite our patient showing no abnormality history on routine screening, abdominal ultrasound and CT scanner detected no urologic masses.

Besides that, thrombocytopenia due to cirrhosis also raises questions about the patient's hematuria condition. However, the absence of systemic hemorrhages like skin bruises, gum bleeding, and normal coagulating test results logically ruled out this idea. Other scenarios that may contribute to hematuria in allograft kidneys, like chronic rejection (commonly present with microscopic hematuria), renal hemorrhagic cyst rupture, and calculus, are all excluded.

AVM is a vascular abnormality that can be congenital or acquired. Acquired AVM consists of more than 70% of all AVM, which often occurs due to inflammation, trauma, or percutaneous procedures (e.g., renal biopsy) [1,10–14]. Conversely, congenital AVM accounts for the remaining 20% of cases; the vast majority of them are unknown [11]. They can be further classified into cirsoid, angiomatous, and aneurysmal. The cirsoid type has multiple feeding arteries and draining veins, and the aneurysmal type has aneurysm dilation, as it is named [11,12]. Furthermore, angiomatous AVM is usually characterized by a single connection between an artery and a vein

without a nidus [10]. The vast majority of victims suffer from hematuria, flank pain, hypertension, or even being asymptomatic. Ultrasonography is frequently used as the first-line modality in these cases with several serpiginous hypoechoic structures and mosaic high-flow patterns on Doppler. Even though the early renal venous filling on the arterial phase of an abdominal CT scan is a classical finding of AVM, the role of this modality in detecting AVM is still unreliable. In the case of small-nidus with low flow (like in our case), a very faint early venous enhancement can easily be missed. Magnetic resonance imaging can also be considered based on its high-resolution advantage, but the thick slide may make it remarkably uncertain.

DSA remains the modality of choice in establishing renal AVM diagnosis and embolization [14,15]. Selective embolization effectively treats AVM and interferes less with the renal parenchyma. As such, it should be considered the first choice of therapy whenever the urgency of these cases permits.

Conclusion

Gross hematuria resulting due to ruptured AVM in renal transplant is a rare condition and can lead to transplant dysfunction. Clinical diagnosis may be obscure due to vague presentation. Angiographic embolization is an effective method for detecting and treating renal AVM.

Author contributions

Conceptualization: all authors. Data curation: TVS, NQD. Formal analysis: LTD, DDH. Methodology: DDH. Project administration: all authors. Visualization: PHK, LTD. Writing–original draft: NQD, TVS. Writing–review & editing: all authors.

Ethics approval

Our Institutional Review Board approved this report (Ref: 07.2023.NCVD dated December 25, 2023).

Patient consent

Informed consent of patient and his relationship was obtained.

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