Renal artery embolization in the treatment of urinary fistula after renal duplication: A case report and review of literature

Tao Yang, Jun Wen, Tan-Tan Xu, Wen-Jing Cui, Jian Xu

ORCID number: Yang Tao 0000-0003-1448-3640; Jun Wen 0000-0003-2990-5092; Tan-Tan Xu 0000-0003-2015-7462; Wen-Jing Cui 0000-0002-1941-4056; Jian Xu 0000-0001-7101-7844.

Author contributions: Xu J was guarantor of integrity of the entire study and edited the manuscript; Yang T prepared and edited the manuscript; Wen J, Xu TT, and Cui WJ performed the literature research, data analysis, and text proofreading.

Informed consent statement: All study participants, or their legal guardian, provided informed written consent prior to study enrollment.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Abstract

BACKGROUND
Duplicate renal malformation is a congenital disease of the urinary system, with an incidence rate of 0.8%. Surgical treatment is suitable for symptomatic patients. Urinary fistula is one of the complications of heminephrectomy. Long-term urinary fistula has a great impact on patients' lives.

CASE SUMMARY
This article mainly reports on a 47-year-old man with duplication of kidney deformity, long urinary fistula after partial nephrectomy, and no improvement after conservative treatment. We have achieved positive results in the arterial embolization treatment of the residual renal artery, indicating that selective arterial embolization is a good way to treat urinary fistula after partial nephrectomy. It is worth noting that this patient violated the Weigert-Meyer law, which also gave us more consideration.

CONCLUSION
Renal artery embolization may be a simple and safe method to treat urinary fistula inefficacy with conservative treatment.

Key Words: Renal artery embolization; Urinary leakage; Urinary fistula; Duplicate renal malformation; Selective arterial embolization; Case report

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Core Tip: Urine leakage is one of the complications of heminephrectomy, and it leads to a serious decline in the patient’s quality of life. For patients who fail conservative treatment, they may also face the risk of reoperation. We successfully treated a patient with urine leakage after heminephrectomy through selective renal artery embolization. After 3 years of follow-up observation, the patient's renal function was normal and no urinary fistula recurred.

INTRODUCTION
Duplicate renal malformation is a rare congenital disease of the urinary system, with an incidence rate of 0.8%. There is no significant difference in the incidence of bilateral kidneys. The incidence of women is twice that of men. There is genetic susceptibility, with about 1/8 of patients’ parents or siblings having similar conditions. Asymptomatic patients did not need treatment. Surgical treatment is suitable for patients with hydronephrosis, repeated urinary tract infections, and (or) urinary incontinence[1].

CASE PRESENTATION
Chief complaints
A 47-year-old patient suffered a urinary fistula for up to 14 mo after nephrectomy.

History of present illness
Within 1 wk after heminephrectomy, the patient’s daily drainage tube drained about 30-50 mL of light red liquid. From the 10th day after surgery, the patient’s wound began to seep light yellow transparent fluid, up to 350 mL in large quantities. A computed tomography (CT) scan showed a large amount of fluid around the right kidney 2 mo after the surgery. After repeated local puncture and drainage, the wound still did not heal. The symptoms had lasted for 14 mo.

History of past illness
The 47-year-old patient suffered from symptoms of recurrent right sided flank pain and discomfort. At the local hospital for the first time, CT examination results confirmed duplicate kidney on the right side, severe hydrops, and ureteral stones in the right kidney. Next, lithotripsy was performed. After 4 mo, she went to a senior hospital again because of persistent symptoms. CT was performed again and showed that the upper 1/3 renal pelvis and ureter were normal and the lower 2/3 renal pelvis was severely hydrolyzed without ureter display (Figure 1). After doctor-patient consultation, the right ureteroscopy was performed, and a double J tube was released into the right side of the normal renal pelvis and ureter. One month later, she underwent "laparoscopic right side repeated nephrectomy" under general anesthesia (Figure 2). Visible in the surgery were hydrops in the right lower kidney and normal upper right kidney. The blood supply of the upper and lower parts of the right kidney was each supplied by the renal artery and vein, and the upper and lower parts had independent ureters. The ureter belonging to the lower right kidney was significantly dilated. An ultrasonic knife was used to free completely the junction of the hydronephrosis kidney and the normal kidney to remove the diseased kidney. The ureter was cut about 15 cm away from the renal pelvis. A perinephric drainage tube was placed that led out through the puncture hole above the iliac crest. Postoperative anti-infection and hemostasis treatment was performed. Postoperative pathology proved a duplicate kidney.
Figure 1 Duplicate renal malformation confirmed by computerized tomography urography. A and B: The patient has a duplication of renal malformation on the right side, with independent ureters in the upper (orange arrows) and lower parts (orange triangle), and the lower kidney is a malformed kidney with hydrops.

Figure 2 Perinephric effusion after heminephrectomy of computed tomography. A and B: Shown is the perinephric effusion (orange arrows) in the lower pole of the right kidney after the operation; C and D: Shown is puncture drainage of right perinephric effusion.

**Physical examination**
The patient’s temperature was 36.2 °C, heart rate was 76 bpm, respiratory rate was 18 breaths/min, and blood pressure was 127/74 mmHg.

**Laboratory examinations**
Re-admitted to hospital on December 16, 2016, the drainage fluid biochemical examination showed creatinine 225 μmol/L, urea 9.0 mmol/L, and serum creatinine 68 μmol/L.

**Imaging examinations**
Renal CT angiography showed that a branch artery of the lower pole of the right kidney ran along the lower part of the right kidney (Figure 3).
Further diagnostic work-up

Fistulography showed that the fistula was located in the lower part of the right kidney, which was a limited residual cavity without entering the abdominal cavity.

FINAL DIAGNOSIS

The final diagnosis of the presented case was urinary fistula.

TREATMENT

The right renal digital subtraction angiography (DSA) and renal artery embolization were performed based on the patient’s informed consent. DSA showed that the right inferior renal artery branched to the residual kidney tissue of the right kidney (Figure 3). Polyvinyl alcohol (PVA) particles (100-300 μm) embolized the blood supply artery of the residual kidney by super-selective intubation. After embolization, DSA showed only the main trunk of the residual renal artery. One day after the operation, the drainage tube fluid was significantly reduced.

OUTCOME AND FOLLOW-UP

One month after the operation, the drainage tube was removed. The wound was completely healed 3 mo after the operation. On January 8, 2018, the abdominal CT
angiography (CTA) showed that only the main trunk of the right renal residual artery remained (Figure 4). After 3 years of follow-up, the patient did not leak urine again and her renal function was normal. After 3 years of follow-up observation, the patient's renal function was normal, and no urinary fistula recurred.

**DISCUSSION**

This patient’s kidney has two sets of ureters. The lower kidney was accompanied by obvious ureteral dilation and hydronephrosis. CT urography suggested that the upper kidney had normal secretory function and urinary system. The lower kidney has no urinary function. This case does not conform to the Meyer-Weigert rule; the Meyer-Weigert rule predicts the double ureteral drainage pattern of bipolar renal repetition. The upper pole is generally considered to be ectopic, so it is hypoplastic resulting in obstruction[14]. Although the Meyer-Weigert rule applies to most duplication kidney cases, some case reports in recent years have pointed out exceptions to this rule[17,18]; this is true for our case.

According to the Meyer-Weigert rule, the clinical symptoms of duplication of kidney deformity are often related to the superior kidney. The suitable surgical treatment is heminephrectomy[19]. Since the right upper kidney of this patient had normal function, heminephrectomy was performed for the right lower kidney. Partial nephrectomy is widely used in the resection of early renal cancer due to its lower overall mortality and cardiovascular morbidity compared to radical resection[21,22]. However, the complication rate of urinary fistula after heminephrectomy is between 0.8% and 5.2%[23-25]. After heminephrectomy, if the remaining nephrons can still secrete urine, the urine will extravasate through the damaged collecting system. The extravasated urine will gather in the perinephric space and be wrapped by fibrous tissue to form a urinoma[26,27]. The patient’s drainage tube extravasated liquid contained urine, which was proved by biochemical verification after the operation. It met the diagnostic criteria for urinary fistula. Most heminephrectomy-related urinary fistula can be resolved by conservative treatment (e.g., percutaneous nephrostomy or percutaneous drainage)[25]. However, when conservative treatment is not effective, we should consider changing the treatment plan.

Conservative treatment achieved a therapeutic effect by continuous drainage to reduce the pressure on the renal pelvis to close the damaged collection system. However, a small number of patients require re-surgical removal of the residual kidney[26]. Meeks et al[17] reported 21 cases of urinary fistula after heminephrectomy, with an average continuous urinary fistula time of up to 53 d. Li et al[29] reported a case of ureteral stent implantation in the treatment of urinoma; the urinoma gradually disappeared after 5 mo. Re-surgical treatment may cause more serious complications because of difficulty in separating the residual kidney and blood vessel due to tissue adhesion. As far as we know, as early as the 1980s, scholars reported that four patients with urinary fistula after heminephrectomy were cured by the residual renal artery embolization[25,26]. In 1993, Japan’s Ogawa et al[32] reported that renal artery embolization was used to treat a severely impaired renal function patient with kidney urinary fistula and achieved good therapeutic effect. Wang et al[33] used arterial embolization to treat urinary fistula after repeated renal surgery and got satisfactory effect. Since then, Yamamoto et al[34] and Nohara et al[35] reported three cases of super-selective renal artery embolization in the treatment of renal carcinoma heminephrectomy with urinary fistula. The curative effect was satisfactory, and no obvious complications occurred[31,32].

In our patient, we observed the remnant renal arteries through preoperative CTA, which clarified the relationship between renal artery lesions and surrounding tissues and organs and provided a reliable basis for the choice of surgical methods. Previous studies on CTA, as a minimally invasive vascular imaging technique, have demonstrated that CTA can clearly show the renal artery and its branches, and the diagnostic accuracy of the anatomical variation of the renal artery is 100%[9,11]. DSA not only confirmed the residual renal artery but also realized the precise embolization of the residual renal artery, achieving the purpose of treatment while protecting the renal function. Permanent embolic agents have been used in previous cases, including coils, N-butyl-cyanoacrylate (NBCA) glue, and PVA particle embolic agents. We used 3F microcatheter to enter super-selectively the residual renal artery and embolized with 100-300 μm PVA granules. After embolization, only the stump of the artery remained. PVA particles are made from a polyvinyl alcohol foam sheet. They are a permanent embolic agent that can activate inflammatory reaction and focal angionecrosis, with
Figure 4 Abdominal computed tomography angiography 3 yr after surgery. A and B: The patient was admitted to the hospital in January 2018, and the computed tomography showed that the right perinephric effusion had disappeared.

vessel fibrosis developing over time\(^{28}\). Complications of renal artery embolism mainly include fever, nausea, abdominal and (or) waist pain, ectopic embolism, infection, etc. The incidence rate is less than 10% in the literature\(^{29}\). In previous reports, there have been many successful cases of using fibrin glue, coils, or NBCA glue for percutaneous obliteration of urinary fistula\(^{30-37}\). However, the operation can be cumbersome sometimes, and multiple uses of materials could also lead to excessive hospitalization costs. For instance, Nouri et al\(^{30}\) used percutaneous puncture coils combined with NBCA glue to seal the urinary fistula tract and urinoma in 10 patients, and the highest case used 18 coils to seal the urinary fistula. In our case, residual renal artery embolization was used to treat urinary fistula, and a significant effect was achieved.

CONCLUSION

Although it is currently difficult to report large samples of arterial embolization for the treatment of urinary fistula, with the increase in the number of case reports and the development of interventional techniques, super-selective renal artery embolization is a minimally invasive, safe, effective and maximize retention renal function method to treat urinary fistula after heminephrectomy.

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