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Unusual Bilateral Renal Parenchymal Urine Leak After Pediatric En Bloc Kidney Transplantation: First Case Study Report

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Abstract. Kidney transplantation is usually the best course of treatment for patients with end-stage renal disease. En bloc kidney transplantation (EBKT) is a surgical treatment option that increases available donor organs with excellent graft survival for patients with end-stage renal disease. Herein, we report a case of an unusual bilateral renal parenchymal urine leak after EBKT leading to removal of both moieties of the EBKT. This unfortunate complication after EBKT, to our knowledge, is the only reported of its kind. We explore the possible causes of the bilateral parenchymal urine leaks and suggest preventive strategies to avoid urological complications after EBKT.

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Kidney transplantation is the most effective treatment option for patients with end-stage renal disease (ESRD).¹ Unfortunately, the demand and supply mismatch between patients with ESRD awaiting kidney transplantation and available donor organs is on the rise.² Transplantation of kidneys from pediatric donors either as single kidney transplantation or en bloc kidney transplantation (EBKT) is an attractive option to meet this increasing demand for organs.³⁻⁷ Traditionally, EBKT has been believed to maximize graft function, whereas single kidney transplantation maximizes resource availability as each donor produces 2 recipient grafts.^{8,9} The short- and long-term patient and

graft survivals after EBKT are comparable to standard criteria deceased donor kidney and living donor kidney transplantation.^{3,10,11}

The use of EBKT has not been universally accepted due to the risk of technical difficulties (vascular and urological) and medical concerns related to inadequate nephron mass and hyperfiltration injury.¹¹ The incidence of urinary leak after EBKT may vary from 0% to 11%.^{3,7,10,12} The most common and the only reported site of urine leak in literature is at the ureteroneocystostomy.^{5,6} We report an unusual case of renal parenchymal urine leak after EBKT that ultimately led to sequential removal of both renal moieties. To the best of our knowledge, this is the first such case report in literature.

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CASE DESCRIPTION

We transplanted pediatric en bloc deceased donor kidneys to a 49-year old African American male with ESRD secondary to hypertension who had been on peritoneal dialysis (PD) for 1.5 years. The preoperative cytotoxic and flow crossmatch were negative. The en bloc pediatric kidneys were procured from a brain dead, 2-month-old donor who weighed 4.6 kg. The kidneys were transported in cold University of Wisconsin solution, and no procurement injury was reported. The right kidney measured 4.5 × 2.5 × 1.5 cm and the left kidney measured 5.0 × 2.0 × 1.5 cm. Both donor kidneys were free of any plaque, infarcted areas, capsule tear, cysts, subcapsular hematoma, or any signs of trauma during procurement. Kidneys were procured with adequate lengths of aorta and inferior vena cava. During back-table preparation special attention was paid to careful ligation of small aortic and caval lumbar vessels. Care was taken not to skeletonize the renal pedicle especially the ureters. The suprarenal aorta and the vena cava were then closed with running polypropylene 6-0 suture, taking care not to occlude or kink the renal vascular orifices.

In the recipient, the right external iliac artery and vein were exposed via a lower quadrant extraperitoneal approach and mobilized. Heparin (35 units/kg) was administered intravenously. The caudal ends of the donor inferior vena cava and aorta were anastomosed to the recipient's external iliac vein and artery, respectively, in an end-to-side fashion using polypropylene 6-0 sutures in a continuous fashion. The donor ureters were spatulated and then their posterior walls conjoined using a polydioxanone 6-0 absorbable suture. The conjoined ureters were transplanted into a single urinary bladder opening in an extravescicular fashion, over 2 pediatric (4.5 Fr) ureteric stents. The kidneys were carefully positioned and fixed to the lateral pelvic wall using the perinephric fat. The final orientation of the paired reperfused kidneys was medial-lateral to each other (Figure 1). A drain was placed. The cold ischemia time was 19 hours and 20 minutes, whereas the warm ischemia time was 28 minutes.

The recipient received rabbit antithymocyte globulin (1.5 mg/kg per day (thymoglobulin; Genzyme Corp, Cambridge, MA) from postoperative day (POD) 0 to 3. Maintenance immunosuppression consisted of tacrolimus, mycophenolate mofetil, and a tapering dose of steroids. No immediate posttransplant anticoagulation was instituted. Aspirin (81 mg/d) was started on POD 1. Hypertension was controlled to maintain systolic blood pressures less than 110 mm Hg.

On POD 6, there was an increased output of clear fluid in the wound drain. The drain fluid creatinine level was 55 mg/dL compared with the serum creatinine level of 9.9 mg/dL indicating a urine leak. A CT-cystogram was performed at this time which showed mild caliectases of the both

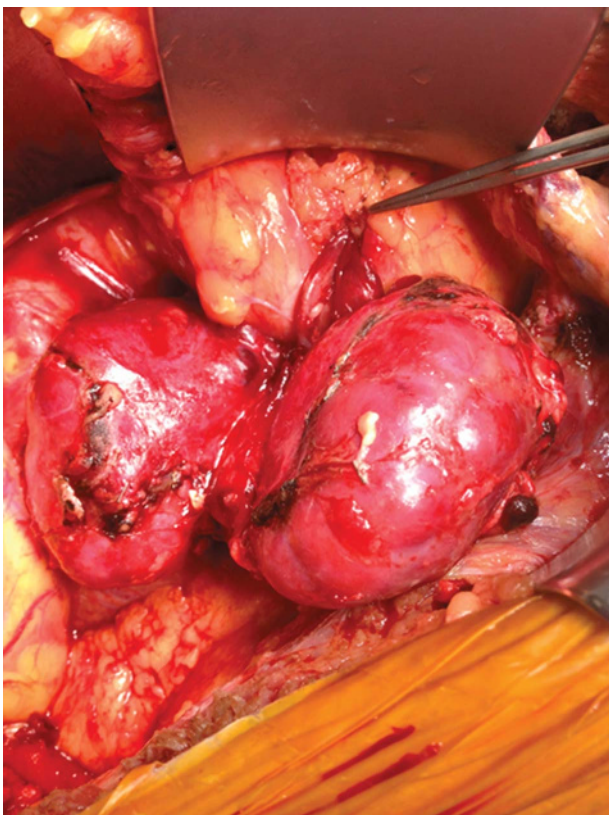


FIGURE 1. Pediatric en bloc kidneys after transplantation. The forceps points at the ureteroneocystostomy.

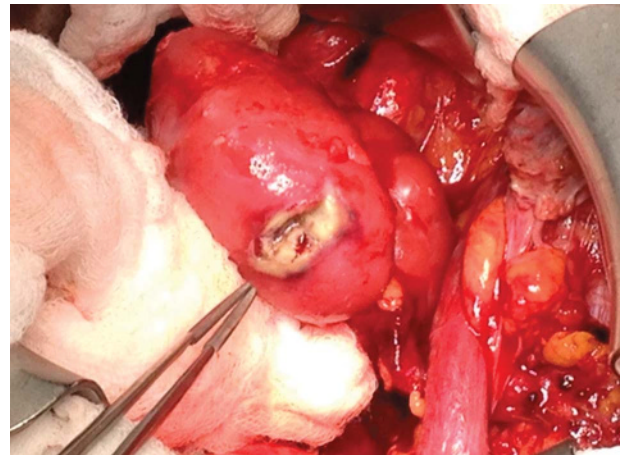


FIGURE 2. Site of urine leak from the necrotic renal parenchyma at lower pole of the lateral pediatric kidney.

transplant kidneys but no evidence of anastomotic bladder leak. The Foley catheter was replaced.

As the drain output increased, a surgical reexploration on POD 8 revealed a necrotic area on the inferolateral pole of the lateral kidney actively leaking urine (Figure 2). There was no evidence of any vascular compromise on intraoperative handheld Doppler examination. The parenchymal leakage was repaired with interrupted 4.0 chromic catgut sutures with a vein patch as pledget and surrounding fat as buttress. The bladder was distended with antibiotic solution, and no leak was noted from the ureteroneocystostomy. The drain output remained high ranging from 400 to 1600 mL/d. Recipient developed delayed graft function requiring hemodialysis. Patient was discharged home with the Foley catheter and wound drain in place. There was no evidence of infection after surgical reexploration.

On POD 31 from the EBKT, patient was readmitted for abdominal and groin pain. Immediate renal scan with furosemide revealed tracer accumulation originating from the lower pole of lateral kidney confirming persistence of urine leak (Figure 3). Exploratory laparotomy was performed on POD 32 due to increasing abdominal pain. Exploration

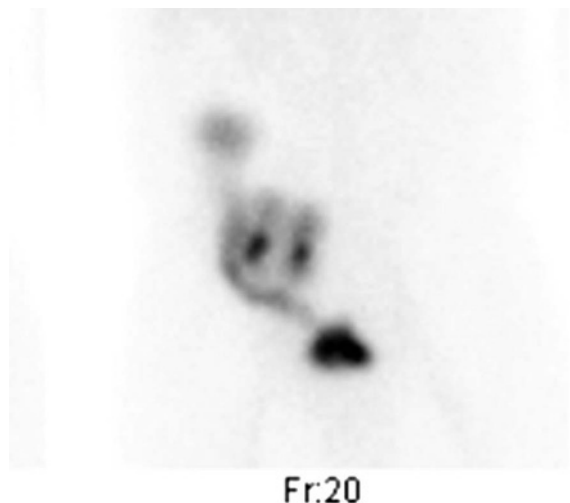


FIGURE 3. Renal scan with furosemide after en bloc kidney transplantation showing tracer originating from the lower pole of lateral kidney.

TABLE 1.
Strategies to prevent complications after en bloc pediatric kidney transplantation

Strategies to prevent complications after EBKT

- Avoid very small pediatric donors with history of severe hypotension, prolonged resuscitation or history of hypercoagulable disorders
- Organ procurement and transplantation by experienced surgeons
- Ensure good flushing of pediatric kidneys in donor
- Avoid any preimplantation or postimplantation biopsies
- Avoid hilar dissection to prevent vascular injuries
- Preserve perinephric and periureteric fat
- Avoid thermal damage to renal cortex during implantation
- Use pediatric ureteric stents of appropriate length and diameter
- Avoid recipients with uncontrolled hypertension
- Consider perioperative anticoagulation

confirmed recurrent lateral kidney urine leak at the site of previous repair but the other kidney also showed a necrotic area at the lower pole which leaked when the urinary bladder was distended with saline. The hilum of the EBKT was then clamped, and both the kidneys were removed because the patient had been symptomatic with abdominal pain. Patient was subsequently relisted for kidney transplantation and is currently undergoing renal replacement therapy. The explant pathology showed focal areas of cortical necrosis measuring $1.0 \times 1.0 \times 0.8$ cm and $1.0 \times 0.5 \times 0.5$ cm on the lateral and medial kidneys, respectively.

DISCUSSION

We present an unusual case of parenchymal urine leak after pediatric en bloc kidney transplantation. We discuss the possible mechanisms that may have likely lead to this rare complication that resulted in sequential loss of both renal moieties.

In most published reports, the site of urine leak is the new ureter to bladder anastomoses and the leak rates are higher in some series where ureteric stents were not used.⁸ At our center, a majority of the ureteral anastomoses in EBKT is performed using the “double barrel” technique with an extravesical ureteroneocystostomy over 2 separate internal pediatric ureteral stents.¹³ From a recent study of 20 EBKT performed at our institution, 1 patient had urine leak (5% leak rate).³ This was the only patient where the Bladder Patch Technique was used and failed secondary to necrosis of the bladder patch. However, others have reported acceptable outcomes with this technique.¹⁴

The current patient with EBKT developed the urine leaks from the lateral and medial kidneys about a week and a month, respectively, after transplantation. The etiology of these leaks remains unclear to us. In retrospect, there was no hydronephrosis immediately after the EBKT. This implies that there was no distal obstruction at the ureteral anastomoses (and the internal stents were still in place). A thorough review of the patients' preoperative work-up did not reveal any signs or symptoms benign prostatic hyperplasia or urethral strictures. Moreover, the patient had voided uneventfully after removal of his Foley catheter, thereby ruling out any urethral obstruction. The early onset of urine leak (within 1 week) from a necrotic area on the lateral renal moiety, points toward vascular or ischemic injury to the affected area. Interestingly, the

pediatric recipient of the liver from the same donor as these en bloc kidneys developed portal vein thrombosis, although this may or may not be related. It is possible that both en bloc kidneys in our recipient may have suffered lower pole devascularization or cold preservation injury or poor perfusion in the donor leading to necrosis and parenchymal urine leak in the recipient.

Another possible mechanism of urine leak could be thermal injury if Argon beam coagulation is used for superficial hemostasis on the thin cortex of these pediatric kidneys after reperfusion. In our case, Argon beam was possibly used for hemostasis over the perinephric fat. We therefore recommend that any energy device should be avoided close to the thin renal parenchyma of these small kidneys. We recommend that in EBKT, the perinephric fat should be preserved to avoid decapsularization and renal parenchymal trauma to these small kidneys.³ The excess perinephric tissue is also used to pexy the kidneys and thus prevent torsion and vascular thrombosis at the hilum. We used 4.5-Fr ureteric stents that may be considered large for the ureters of these infant kidneys. A snug fit of the stent tips in the pelvicalyceal system could have potentially caused pressure necrosis of the thin parenchyma and resulted in bilateral urine leaks. We therefore recommend that ureteric stents of appropriate length and diameter (3 Fr) should be used when transplanting these small kidneys. Parenchymal urine leak can also result from postoperative severe hypertension leading to renal parenchymal rupture (authors' unpublished observation in a patient with single pediatric kidney transplant).

It could be speculated that prolonged surgical drainage along with bladder decompression could have salvaged these en bloc kidneys. However, this patient continued to have abdominal pain despite the presence of surgical drain. The second kidney was removed chiefly due to concern for impending infection at the vascular anastomosis. Although unfortunate, removing the EBKT enabled this patient to be relisted earlier for a retransplant. Our strategies to avoid complications after EBKT are summarized in Table 1.

To our knowledge, this is the first reported case of bilateral parenchymal urine leaks from after pediatric EBKT. The exact etiology of this unusual complication in these en bloc pediatric kidneys still remains unclear. We conclude that en bloc pediatric kidney transplantation is a challenging procedure that may be associated with unusual complications even at experienced centers.

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REFERENCES

1. Wolfe RA, Ashby VB, Milford EL, et al. Comparison of mortality in all patients on dialysis, patients on dialysis awaiting transplantation, and recipients of a first cadaveric transplant. *N Engl J Med.* 1999;341:1725–1730.
2. Axelrod DA, McCullough KP, Brewer ED, et al. Kidney and pancreas transplantation in the United States, 1999–2008: the changing face of living donation. *Am J Transplant.* 2010;10:987–1002.
3. Sharma A, Fisher RA, Cotterell AH, et al. En bloc kidney transplantation from pediatric donors: comparable outcomes with kidney transplantation. *Transplantation.* 2011;92:564–569.
4. Sharma A, Ramanathan R, Behnke M, et al. Single pediatric kidney transplantation in adult recipients: comparable outcomes with standard-criteria deceased-donor kidney transplantation. *Transplantation.* 2013;95:1354–1359.

5. Sanchez-Fructuoso AI, Prats D, Perez-Contin MJ, et al. Increasing the donor pool using en bloc pediatric kidneys for transplant. *Transplantation*. 2003;76:1180–1184.
6. Bhayana S, Kuo YF, Madan P, et al. Pediatric en bloc kidney transplantation to adult recipients: more than suboptimal? *Transplantation*. 2010;90:248–254.
7. Mahdavi R, Arab D, Taghavi R, et al. En bloc kidney transplantation from pediatric cadaveric donors to adult recipients. *Urol J*. 2006;3:82–86.
8. Satterthwaite R, Aswad S, Sunga V, et al. Outcome of en bloc and single kidney transplantation from very young cadaveric donors. *Transplantation*. 1997;63:1405–1410.
9. Lam VW, Laurence JM, Robertson P, et al. En bloc paediatric kidney transplant: is this the best use of a scarce resource? *ANZ J Surg*. 2009;79:27–32.
10. Beltran S, Kanter J, Plaza A, et al. One-year follow-up of en bloc renal transplants from pediatric donors in adult recipients. *Transplant Proc*. 2010;42:2841–2844.
11. Sureshkumar KK, Reddy CS, Nghiem DD, et al. Superiority of pediatric en bloc renal allografts over living donor kidneys: a long-term functional study. *Transplantation*. 2006;82:348–353.
12. Memel DS, Dodd GD 3rd, Shah AN, et al. Imaging of en bloc renal transplants: normal and abnormal postoperative findings. *AJR Am J Roentgenol*. 1993;160:75–81.
13. Nghiem DD. Single tunnel extravesical ureterocystostomy in pediatric en-bloc kidney transplantation. *Transpl Int*. 1991;4:250.
14. Kato T, Selvaggi G, Burke G, et al. Partial bladder transplantation with en bloc kidney transplant—the first case report of a 'bladder patch technique' in a human. *Am J Transplant*. 2008;8:1060–1063.