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Kidney transplantation in a patient with absent right common iliac artery and congenital renal abnormalities

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ABSTRACT

INTRODUCTION: Congenital atresia of the common and external iliac arteries is a rare vascular anomaly that may be associated with congenital renal or genitourinary malformations. In ESRD patients, its presence may pose potential problems during renal transplantation.
CASE PRESENTATION: We report a rare case of kidney transplantation in a patient with VACTERL syndrome who was found to have absent right common and external iliac arteries during pre-operative imaging. Vascular supply to the right lower limb is derived from an anomalous branch from the left internal iliac artery which takes on a convoluted course across the pelvis. Kidney transplantation was performed successfully with implantation performed on the left side.
DISCUSSION: Isolated cases of congenital iliac artery atresia have been described in association with urological abnormalities but no clear association has yet been established. However, we feel that it may be useful to perform routine angiographic evaluation for ESRD patients with congenital genitourinary abnormalities being planned for kidney transplantation. While most cases of congenital iliac artery anomalies are symptomatic with claudication, some remain asymptomatic with normal physical examination findings. There is some evidence in literature suggesting the usefulness of routine pre-operative CT in a selective group of patients.
CONCLUSION: Kidney transplantation in such cases is safe and we recommend routine pre-operative imaging of patients known to have congenital genitourniary abnormalities. The kidney should be implanted heterotopically to the contralateral side of the vascular anomaly and care must be taken to preserve vascular supply to the lower limbs.

1. Introduction

Renal transplantation has become the standard of care for end stage renal disease (ESRD), including cases secondary to congenital genitourinary malformations. However, the literature on kidney transplantation in patients with associated congenital malformations of the iliac vessels is limited. Congenital vascular malformations of the ilio-femoral arteries are less common than those in the thoracic and abdominal aorta and usually discovered incidentally or by the presence of chronic lower limb ischemia [1,2]. Congenital hypoplasia or atresia are the most commonly reported malformations of the iliac arteries, and may be associated with a persistent sciatic artery [3,4]. Also, the majority of vascular malformations are unilateral, usually on the right side [1]. We herein report a very rare case of kidney transplantation in a patient with of absent right common iliac artery and VACTERL (vertebral anomalies, anal atresia, cardiac defects, tracheo-esophageal fistula/atresia, renal and radial anomalies and limb defects) association.

2. Case presentation

A 24 year old male underwent an elective living-related kidney transplant (LRRT) for ESRD secondary to neurogenic bladder and vesico-ureteric reflux (VUR). He has a background history of VACTERL association with imperforate anus (previous colostomy creation and subsequent closure), sacral hemivertebrae (neuro-

Abbreviations: CT, computed tomography; ESRD, end stage renal disease; LRRT, living related renal transplant; MAG-3, mercaptoacetyltriglycine-3; VACTERL, vertebral anomalies, anal atresia, cardiac defects, tracheo-esophageal fistula/atresia, renal and radial anomalies and limb defects; VUR, vesico-ureteric reflux.

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Table 1
Timeline.

Timeline of previous surgical interventions	
1991	Right colostomy creation and subsequent closure (for imperforate anus)
1991	Re-implantation of right ureter
1994	Re-implantation of left ureter
June 2013	Initiated on hemodialysis
July 2013	Colonic bladder augmentation and Mitrofanoff creation
December 2013	Nephrectomy of cross-fused kidney

logically normal) and renal/urological defects: (a) bilateral grade 5 VUR with previous bilateral ureteral re-implantation, (b) neurogenic bladdder status post-colonic bladder augmentation and mitrofanoff creation, (c) crossed fused ectopia of the left with right kidney with prior nephrectomy in preparation for LRRT (Table 1).

In view of his multiple congenital anomalies and previous surgeries, computed tomography (CT) of the abdomen and pelvis was done for pre-operative planning. The CT revealed aberrant iliac vasculature – the right common and external iliac arteries are not visualized as the abdominal aorta ends with the left common iliac artery bifurcating into the left internal and external iliac arteries (Fig. 1). A branch arising from the right distal abdominal aorta superior to the bifurcation gives off a lumbar branch before passing posterior to the right psoas muscle into the pelvis. It gives off several pelvic branches before continuing anteriorly along the pelvic side wall to receive a large anomalous branch from the left internal iliac artery, just proximal to the femoral canal. The large anomalous branch, which is the main vascular supply to the right lower limb, demonstrates a convoluted course across the pelvis from left to right. The combined vessel then continued distally as the right common femoral artery (Fig. 2).

Despite the vascular abnormalities detected on radiological imaging, the patient was asymptomatic. He had no evidence of limb ischemia and pulses were symmetrical bilaterally in the lower limbs. He subsequently underwent LRRT with implantation of kidney performed in the left iliac fossa. Intra-operatively, dissection of the left iliac vessels clearly demonstrated the anomalous vessel branching off the left internal iliac artery and this was carefully preserved (Fig. 3). The renal artery and vein were anastomosed end-to side to the left external iliac artery and vein, respectively, a significant distance distal to the origin of the anomalous vessel. Ureteral anastomosis was performed to the augmented colonic cystoplasty. Good perfusion to the kidney was observed after vascular clamps were released and bilateral dorsalis pedis pulses were palpable intra-operatively. Post-operatively, the patient did not develop any

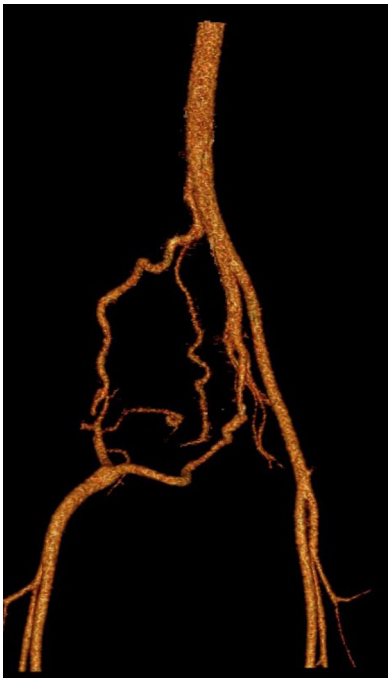

Fig. 2. 3D reconstruction of CT showing a missing right common iliac artery and the anomalous branch from the left internal iliac artery crossing the pelvis. A branch arising from the right side of the distal abdominal aorta, superior to the bifurcation, gives off a few pelvic branches before continuing along the pelvic side wall to receive the anomalous branch from the left internal iliac artery just proximal to the femoral canal. This then continues distally into the right lower limb as the right common femoral artery.

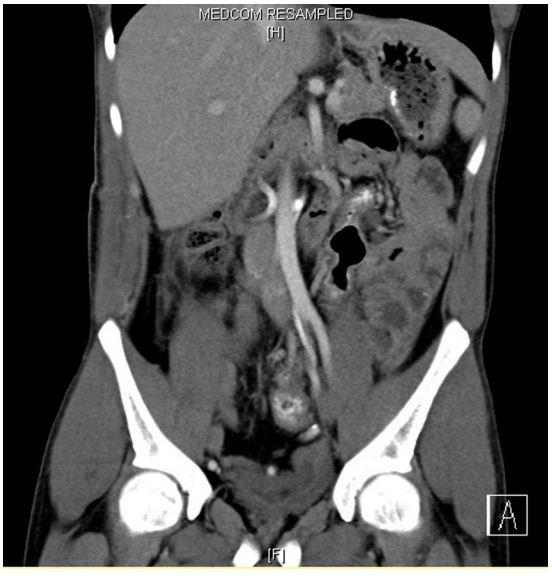

Fig. 1. Coronal section of CT showing the abdominal aorta continuing as the common left iliac artery which bifurcates into the left internal and external iliac arteries. The right common iliac artery is absent.

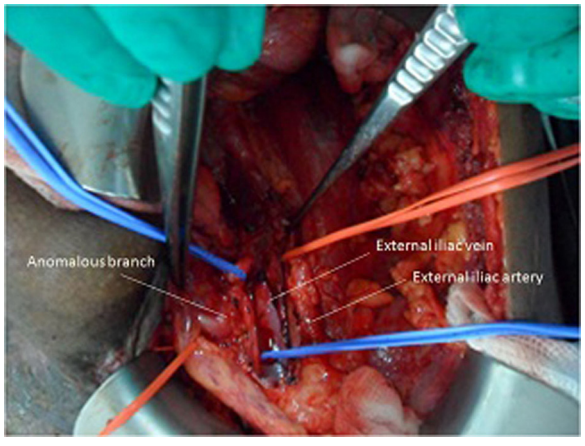

Fig. 3. Intra-operative image of the left external and internal iliac arteries, as well as the preserved anomalous vessel branching off the internal iliac artery.

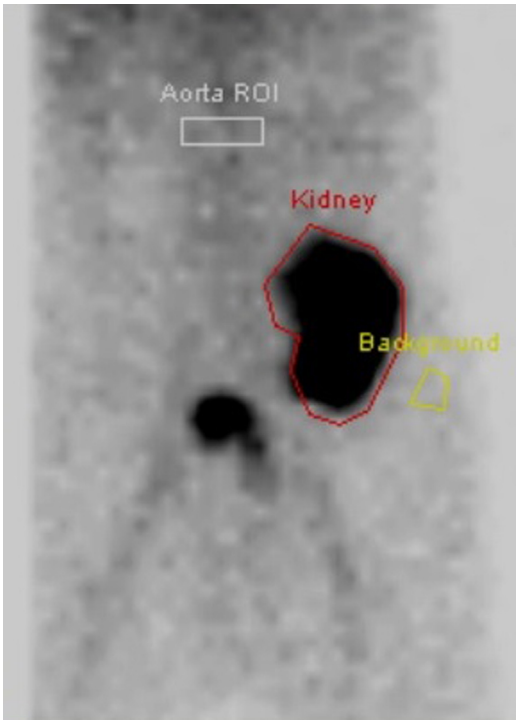

Fig. 4. MAG 3 scan depicting perfusion to the transplanted kidney in the left iliac fossa.

signs or symptoms of limb ischemia and was able to ambulate without difficulty. Mercaptoacetyltriglycine-3 (MAG-3) and ultrasound scans showed prompt perfusion to the transplant kidney with normal flow velocities and Doppler waveforms (Fig. 4). There was immediate graft function and renal function recovered quickly with good urine output. The patient made an uneventful recovery and was discharged home on post-operative day 12 with a creatinine of 103 μmol/L. Subsequent outpatient follow-up within the first post-operative year did not reveal any features of lower limb ischemia. His graft function also remained stable on an immunosuppression regime consisting of Prednisolone, Azathioprine and Tacrolimus. He did require a reduction in Tacrolimus dose due to an episode primary *Cytomegalovirus* infection that was treated successfully with valgancyclovir.

3. Discussion

Congenital anomalies of the iliofemoral arteries are uncommon, with only 6 cases reported in a series of 8000 angiograms by Greeb et al. [5]. Hypoplasia/atresia of the external iliac artery was the most commonly observed anomaly, present in 5 out of the 6 cases of this series [5]. Other reports in the literature include, bilateral aplasia of internal iliac or external iliac arteries [6,1], agenesis of external iliac artery [4], congenital absence of a common iliac artery [2,7], absence of unilateral common and external iliac arteries [8], abnormal external iliac artery arising from the ipsilateral renal artery [9], and quadrifurcation of the terminal aorta where all four iliac arteries arose separately from the aorta [5]. A report by Tamisier et al. classified congenital malformation of the external iliac artery into three groups: (1) anomalies in origin or course of the artery, (2) hypoplasia or atresia with a persistent sciatic artery and (3) isolated hypoplasia or atresia [3].

Congenital absence/atresia of the common iliac artery has only been described sparingly in a few isolated case reports. Similar to our patient, the cases described by Llauger et al. and Midani et al. both demonstrated an absent right common iliac artery with perfusion to the right lower limb supplied by an anomalous branch from the left internal iliac artery, which was seen to be continuous with the right external iliac artery after a convoluted course horizontally across the pelvis. Inferior gluteal and pudendal branches could be identified arising from the aberrant vessel before its ending in the right external iliac artery [2,7]. Despite the similarities, our patient is unique in several ways – such a vascular anomaly, to our knowledge, has never been described in a patient with VACTERL association. This is also only the second reported case of kidney transplantation in an ESRD patient with congenital absence of the common iliac artery.

As in our patient, the case described by Midani et al. also possessed a congenital renal abnormality with an absent right kidney [7]. Isolated cases of congenital iliac artery atresia have previously been described in association with renal, ureteric, renovascular, and other pelvic abnormalities [2]. However, no clear association between congenital iliac vasculature and renal abnormalities has yet been established. In particular, congenital atresia of iliac arteries has not been reported in the presence of crossed fusion renal ectopia. Nonetheless, it should be recommended that routine angiographic evaluation be performed for ESRD patients with congenital genitourinary abnormalities who are being planned for kidney transplantation. Careful palpation of the femoral arteries should be an important step in clinical examination of the potential recipient and weak or unequal pulsations should be further evaluated by imaging. While most cases of congenital iliac artery anomalies are symptomatic with claudication, some remain asymptomatic with normal physical examination findings like our patient. Pre-operative CT angiography is currently not performed routinely in patients with normal physical findings. However, a recent retrospective review by Smith et al. to define the value of routine CT angiography found that 22.9% of recipients had a meaningful alteration in treatment based on angiographic findings [10]. Furthermore, contrast-induced nephropathy was avoided with the use of moderate-dose nonionic iodinated contrast and adequate hydration as there was no change in renal function or need for emergent dialysis in the pre-dialysis cohort [10]. Although the cost was substantial, predictors of a study that would alter care include chronic infection, low patient weight and ventral torso surgical scarring [10]. We feel that congenital renal abnormalities should be added to the list of predictive factors, and CT angiography with 3D reconstruction may be performed to evaluate for underlying vascular anomalies that may complicate surgery. If a similar unilateral vascular abnormality is found, the kidney should be implanted on the contralateral normal side, with vascular anastomosis performed distal to the origin of anomalous vessel and careful preservation of the vessel.

Our experience from this case has shown that it is safe to perform kidney transplantation in such cases with the appropriate pre-operative imaging and surgical planning. There appears to be no evidence of lower limb ischemia or steal phenomenon and no compromise of graft function. The presence of an absent common iliac artery is therefore, not a contraindication to transplantation. However, the presence of such a vascular abnormality may pose a potential problem in the future with the development of atherosclerotic disease or thrombosis. These patients should be kept on close regular follow-up in terms of the vascular status of their lower limbs.

4. Conclusion

Kidney transplantation in such cases is safe and we recommend routine pre-operative imaging of patients known to have congenital genitourniary abnormalities. The kidney should be implanted heterotopically to the contralateral side of the vascular anomaly

and care must be taken to preserve vascular supply to the lower limbs.

Conflict of interest

The authors of this manuscript have no conflicts of interest to disclose.

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Author contribution

C.M. Tay, T.K. Ng and H.Y. Tiong were part of the surgical team that performed the operation and contributed in designing the report; E.P.Y. Siew reported the pre-operative scans and performed the 3-D reconstruction of images; A. Vathsala has been responsible for the posttransplant care of the patient and is his Principal Renal Transplant physician. C.M. Tay wrote the manuscript; T.K. Ng and H.Y. Tiong helped with revision of the manuscript.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. All patient identifiers have been removed to ensure the patient confidentiality.

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