Re: Lowrey et al: Bladder Agenesis and Associated Pelvic Arterial Anomaly in 2 Female Pediatric Patients (Urology 2019;123:227-229)

TO THE EDITOR:

In their recently published case report, Lowrey et al presented 2 patients with bladder agenesis and concomitant pelvic arterial anomaly and suggested the 2 anatomical variants are causally related.1 Bladder agenesis is extremely rare with only 25 live births recorded in the literature.2 With so few patients to study, much of what is known is through case reports such as Lowery's.

We herein would like to present a case of bladder agenesis who recently received a living donor kidney transplant 3 weeks ago and was found to have normal pelvic vascular anatomy both on preoperative imaging and intraoperatively. These findings contradict Lowery’s interpretation that bladder agenesis may be causally related to aberrant pelvic arterial anatomy.

CASE

Our patient is an 8-year-old female with a history of left multicystic dysplastic kidney, ectopic pelvic right kidney with reflux nephropathy and bladder agenesis. On presentation the patient had complete incontinence with urine draining at her vaginal vestibule. In October of 2015 the patient underwent a left nephrectomy and creation of an ileocolonic neobladder with an appendicovesicostomy. No aberrant vascular anatomy was noted during that operation. Over the next few years the patient’s kidney function continued to deteriorate with recurrent bouts of urinary tract infections.

In January of 2019, the patient received a living donor kidney transplant. Preoperatively an MRI was performed which did not identify any aberrant pelvic vascular anatomy. During the surgery a left lower quadrant incision was made. The donor kidney’s vein was anastomosed end-to-side to the left common iliac vein and the renal arteries (donor kidney had 2 arteries) were anastomosed end-to-side to the left common iliac artery. This was a joint case with the Pediatric Transplant team and the Pediatric Urologist who performed the prior neobladder surgery. During the procedure no aberrant pelvic vasculature was noted.

Our patient confirms previous studies which have shown that bladder agenesis is associated with other kidney abnormalities such as renal ectopy and multicystic dysplastic kidneys.3 However, we did not find any pelvic vascular anomalies as was the case with Lowery’s 2 patients. Lowery also cited a third case in the literature of pelvic vascular anomaly in a patient with bladder agenesis.4 This association of aberrant pelvic vascular anatomy and bladder agenesis has not been widely studied and may be just that, an association, and not a causal relationship seeing as it is not present in all patients with bladder agenesis.

References


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Author Reply to “Letter to the Editor on: Bladder Agenesis and Associated Pelvic Arterial Anomaly in 2 Female Pediatric Patients” (#URL-D-19-00427)

Since bladder agenesis is exceedingly rare, it is likely that we will never know the etiology of this malformation, or whether it could be the end result shared by more than one mechanism of maldevelopment. We agree with the authors of the “Letter to the Editor” that most published bladder agenesis cases do not comment on presence or absence of anomalous pelvic arterial anatomy. Thus, the authors are likely correct that the vascular malformation is not the root cause in most bladder agenesis cases. This point is theoretical and less important. The main take-home lesson of our publication is to “See what you look for and look for what you know.” Look for such variant arterial supply preoperatively, since it can greatly impact
leg and/or renal transplant function and possibly prevent catastrophic surgical misadventure.

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