Bilateral ureteroceles progressing to reversible acute renal failure in an adult

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We present a case of an adult male who was admitted with acute renal failure. In evaluating the potential causes of renal failure, workup discovered bilateral ureteroceles. Surgical treatments of the ureteroceles lead to reversal of his acute renal failure. This is the first time that treatment of ureteroceles has been reported to correct acute renal failure in the English literature.

Key Words: adult ureteroceles, acute renal failure, bilateral ureteral obstruction, surgery

Case presentation and management

A 52-year-old male was referred with a chief complaint of acute renal failure. He had a history of recently diagnosed hypertension and slowly progressive renal failure over 5 months. Due to the rising creatinine level, his primary care physician reduced his dose of lisinopril, which he was taking for hypertension. On admission to our hospital his creatinine level was 5.1 mg/dL (normal range of 0.8-1.5 mg/dL). He denied any history of nocturia, difficulty in urination, hesitancy, urgency, fever, abdominal pain or hematuria. His physical examination was unremarkable including prostate examination.

On laboratory testing, his urine analysis was normal. His serum prostatic specific antigen (PSA) level was 0.7 ng/ml (normal < 4.0ng/ml). At

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admission, his blood urea nitrogen (BUN) was 73 mg/dL (normal 9 mg/dL-20 mg/dL), and the serum bicarbonate was low at 19 mmol/L (normal 22 mmol/L-30 mmol/L). The serum potassium level was normal at 4.3 mmol/L (normal 3.5 mmol/L-5.0 mmol/L).

At the time of admission, his angiotensin converting enzyme (ACE) inhibitor medication was discontinued. The creatinine continued to climb to a peak of 6.1 mg/dL, without any ACE inhibitor use.

Renal ultrasound, Figure 1 showed left hydronephrosis, with an atrophic right kidney.

CT scan of the abdomen and pelvis without contrast was obtained. It confirmed the findings from the ultrasound. No stones were seen. The left kidney was measured at 12.2 cm, with mild to moderate hydronephrosis. The right kidney was measured at 8.6 cm in length, with mild hydronephrosis and a patulous extrarenal pelvis. Magnetic resonance angiography (MRA) noted no evidence of renal artery stenosis on either side. These initial studies had focused exclusively upon the kidneys and upper tracts. On the basis of these radiographic findings, we proceeded to exclude distal ureter or lower urinary tract obstruction as the cause of his renal failure.

Cystoscopy revealed a non-obstructed prostatic urethra. The ureteric orifices exhibited cystic dilations bilaterally around each orifice. The diagnosis of bilateral intravesical ureteroceles was made. A single ureteric orifice was seen on each side. Left retrograde pyelogram showed a dilated left pelvicalyceal system with hydronephrosis down to the bladder level. A double-J ureteric stent was inserted into the left side. We decided to observe the result of stenting for empirical evidence that ureteral obstruction from the ureteroceles was responsible for his acute renal failure. A progressive decrease in the serum creatinine level was noted after stenting the left side. Four days after stenting, his serum creatinine had declined to 2.6 mg/dL.

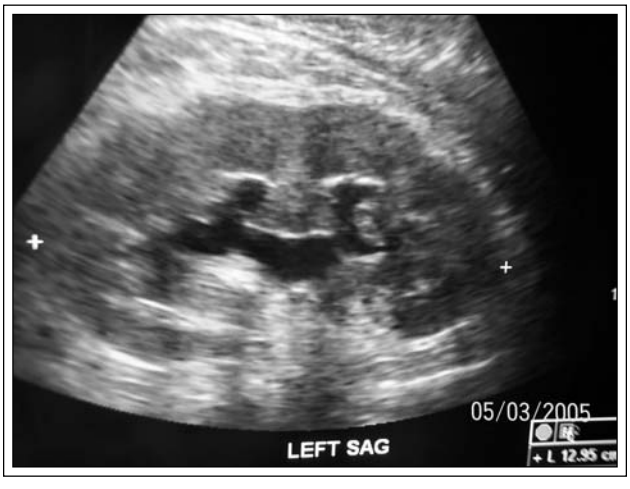

Figure 1. Renal ultrasound showing hydronephrosis of the left kidney. Renal parenchyma is well preserved in this kidney. No stones are evident. This ultrasound was obtained soon after the patient's admission for acute renal failure.

As definitive treatment, we unroofed the ureteroceles bilaterally. During the procedure, both ureters were stented. We then used a resectoscope with a Collins' knife to incise the ureteral orifices. The incisions were carried about 1 cm proximally to unroof the ureterocele without affecting the submucosal passage of the ureter. The rationale for unroofing along the course of the ureter was that the presence of the stent prevented inadvertent perforation of the bladder wall during this procedure. The stents were kept in place for 7 days in order to allow complete epithelialization of the incisional edges. When the patient returned for cystoscopy and stent removal, large open ureteric orifices were noted. His serum creatinine subsequently stabilized at 1.7 mg/dL. His electrolytes were normal. His metabolic acidosis resolved.

Postoperatively, his hypertension persisted. He was restarted on lisinopril without showing any rise in his creatinine level. A Tc99m labeled diethylenetriaminepenta-acetic acid (DTPA) scan was performed 4 months after the bilateral ureteroceles had been incised. The total glomerular filtration rate (GFR) was 40.7 ml/min (normal 95 ml/min-135 ml/min). Split differential function revealed 75.6% of overall function to be on the left side, and 24.4% to be from the right kidney.

Several lines of reasoning led us to believe that the patient's renal failure was due to progressive bilateral ureteral obstruction. His hypertension was of recent onset, less than 1 year ago. The ACE inhibitor used to treat his hypertension was discontinued entirely, yet the serum creatinine continued to climb. A review of his other medications did not reveal any nephrotoxic drugs such as non-steroidal anti-inflammatory drugs (NSAIDS). There was no radiographic evidence that the patient had pyelonephritis, or urinary tract calculi. The patient had no voiding symptoms suggestive of bladder outlet obstruction. There was no evidence of dehydration or low cardiac output to account for prerenal azotemia. The MRA ruled out any renal arterial stenosis. Ultimately, the rapid drop in serum creatinine after left ureteric stent placement showed that ureteric obstruction from the ureteroceles was the

cause of his renal failure. This was followed by incision of the ureteroceles bilaterally as definitive therapy.

Discussion

An ureterocele is a congenital cystic dilation of the submucosal portion of the intravesical ureter.[1,2] In the pediatric age group the incidence of ureterocele has been reported to be between 1 in 5000 and 1 in 12000 pediatric admissions.[3] The female to male ratio is 4-6:1.[4] In children ureteroceles are most commonly associated with the upper pole of a duplex system (80%) with an orifice located ectopically (60%) in the urethra, while in adults they are usually part of an orthotopic single system. When found in adults, they are usually intravesical, associated with a single collecting system, and are less likely to alter the function of the involved kidney.[5] In adults ureteroceles mostly present with flank pain, fever, urinary frequency, urgency and dysuria.

A Medline review of the literature concerning ureteroceles in the English language found 370 citations dating back to 1965.[6] However, only 73 of these citations (approximately 20%) dealt with adult ureteroceles. Very few of these papers deal with renal failure in the context of ureteroceles. Thilagarajah et al[7] reported on a case of a 36 year old man who was found to have bilateral ureteroceles while being investigated for hypertension. The patient had bilateral hydroureteronephrosis and renal cortical thinning. Horizontal endoscopic incision of the ureterocele did not improve his hypertension or progressive rise in serum creatinine. This case is the most similar to ours, although the outcome is different.

Andrews et al,[8] examined the ultrasonographic criteria for diagnosis of ureteroceles. Two patients (out of six) in their series had renal failure and ureteroceles. However, they did not report whether these ureteroceles were ever treated, and if the renal failure of these patients improved as a result of such treatment. Abrahamsson et al,[9] from Denmark reported upon their 18 year experience with 28 adults having ureteroceles. No patients with renal failure were in their series. However, they did have one patient with severe Charcot-Marie-Tooth disease for 20 years, with a unilateral non-functioning hydronephrotic kidney with an ureterocele. Unroofing of the ureterocele did not improve the renal function on the affected side. A series of 56 adult ureteroceles in 52 patients followed for up to 25 years was presented by Madsen et al.[10] In their series, no renal failure was observed. However, it should be noted that 11 patients were lost, or died before the follow up study was performed. This series is the one with the longest follow up period.

For intravesical ureteroceles, endoscopic incision is definitive in the majority of cases. Although there is a risk of vesicoureteric reflux (VUR) following endoscopic incision, in an adult male, reflux may not be of any consequence. Roper et al, in 1965[6] noted secondary VUR and renal deterioration due to recurrent pyelonephritis only in two adult females (out of 15 patients) following ureterocele meatotomy. No injury occurred to the males with VUR following an ureterocele meatotomy. Abrahamsson et al,[9] used the same longitudinal incisional technique that we used. Out of 16 patients in whom a follow up voiding cystourethrogram (VCUG) was available, only two exhibited VUR. Both were asymptomatic adult males with sterile urine.

In the pediatric literature, secondary VUR is a concern. Ben Meir et al[11] reported upon 12 children with ectopic ureteroceles. These children were treated with an endoscopic transverse incision at the ureterocele base. They reported a decompression success rate of 84%, and an improved renal function rate of 41.6%. However, 7/12 of the children had VUR, five of who subsequently required surgery to correct this because of recurrent urinary tract infections. In their review of the literature, they noted that regardless of whether or not the ureterocele was deflated using a puncture, longitudinal or transverse incision, there were "no major differences in the development of new onset vesicoureteral reflux or decompression success rate".[11]

In summary, we present an unusual case of an adult male with bilateral ureteroceles. He presented to the hospital with acute renal failure. We managed to restore his baseline renal function by relieving the obstruction from the ureteroceles. This is the first time that such an outcome has been reported in the English literature. □

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