Duplex renal collecting system presenting with hydroureteronephrosis following pelvic organ prolapse and sacrocolpopexy in an adult female

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ABSTRACT

Duplex collecting systems are common congenital abnormalities of the urinary tract but are infrequently reported in adult populations. This abnormality can present with hydroureteronephrosis secondary to urinary tract obstruction or concomitant vesicoureteral reflux (VUR), recurrent urinary tract infections (UTIs), and urinary incontinence. Options for surgical management include common-sheath ureteral reimplantation, ureterostomy, pyelostomy, and heminephroureterectomy. We report the case of a 39-year-old female with a duplex kidney who presented with severe hydroureteronephrosis following a sacrocolpopexy.

1. Introduction

A duplicated renal collecting system is a congenital abnormality of the urinary tract affecting 0.7–4% of the population, with females more commonly affected than males. 1 While many duplex systems cause no symptoms, upper pole hydroureteronephrosis (secondary to ectopic insertion or an obstructive ureteroceles) and lower pole vesicoureteral reflux (VUR) are commonly reported findings. 2 Duplex collecting systems are usually diagnosed perinatally or among pediatric patients but occasionally do not present until adulthood. These adult patients often remain asymptomatic, but can also present with hydroureteronephrosis, recurrent urinary tract infections (UTIs), or urinary incontinence. 3 We report the case of a 39-year-old patient with known duplex collecting system presenting with renal colic and new hydroureteronephrosis following sacrocolpopexy.

2. Case presentation

Our patient was a 39-year-old female who presented to the emergency department with acute, right flank pain. She reported diaphoresis but no fever, nausea, vomiting, or other urinary symptoms. Past medical history was remarkable for chronic urinary incontinence, endometriosis treated with hysterectomy more than 10 years prior, and pelvic organ prolapse (POP) treated with sacrocolpopexy two years prior. She denied a history of UTIs. Laboratory analysis showed mild leukocytosis to 11,500/μl with neutrophilic predominance. Urinalysis was significant for the presence of leukocytes but was nitrite- and culture-negative. Computed tomography (CT) showed a cystic structure arising from the atrophic upper pole of the right kidney with a tubular structure suggestive of a ureter extending into the lower pelvis (Fig. 1). These findings were consistent with a duplex collecting system complicated by severe hydroureteronephrosis of the right upper moiety. CT imaging from three years prior showed an atrophic right upper pole but no evidence of hydroureteronephrosis (Fig. 2). The patient remained afebrile and hemodynamically stable and was discharged with a seven-day course of ciprofloxacin and pain medication.

The patient was evaluated in clinic. We believed it unlikely for the upper moiety to be significantly functional given several years of imaging demonstrating an atrophic right upper pole. Therefore, we deferred renal functional testing. Due to refractory pain and concerns for pyonephrosis, we recommended surgical intervention. The patient underwent robotic heminephroureterectomy of the right upper moiety. The megaureter from the upper pole emptied into a large cavity in the pelvis posterior to the bladder, which we left patent. This pelvic cavity did not communicate with the bladder or vagina. Intraoperative retrograde pyelogram showed a ureter of normal caliber leading to the right lower pole without connection to the upper pole system. There were no intraoperative complications, and the postoperative course was
unremarkable.

3. Discussion

Duplex collecting systems are common congenital abnormalities of the urinary system. During normal renal development, the ureteric bud arises from the mesonephric duct to meet the metanephros. The metanephros differentiates into nephrons while the ureteric bud bifurcates sequentially to form the branches of the collecting system. An error in this process can result in a duplex kidney with incomplete or complete duplication.\textsuperscript{1}

The relationship between the upper and lower pole ureters is described by the Weigert-Meyer rule, which states that the lower pole ureter inserts orthotopically in the bladder while the upper pole ureter inserts inferomedial to the lower pole ureter. The lower pole ureter is more likely to experience VUR, while the upper pole ureter is more likely to be associated with ectopic insertion or a ureterocele with obstruction.\textsuperscript{3} Ectopic ureters may insert to the bladder neck or urethra. In
females, ectopic ureter insertion can occur to Müllerian structures, such as the vagina or uterus, resulting in continuous urine leakage. In males, ectopic ureters always insert proximal to the external sphincter and do not present with incontinence.\textsuperscript{1}

Duplex systems are commonly detected antenatally or in early childhood. However, they can occasionally be diagnosed in adulthood upon incidental imaging or when they present with symptoms. The most common presenting symptoms are flank pain, recurrent UTIs, and urinary incontinence.\textsuperscript{1} On CT imaging, patients with an upper pole ectopic ureter or ureterocele may demonstrate hydronephrosis and dilatation of the upper pole moiety.\textsuperscript{2} Symptomatic duplex collecting systems in adults are managed with heminephrectomy of the affected pole with ureterectomy if necessary. While there is only a small cohort of data investigating heminephrectomy for duplex kidneys in adults, the procedure is associated with complete resolution of symptoms in most patients with a low risk of complications.\textsuperscript{4}

Our patient presented with flank pain and severe hydronephrosis of the right upper pole moiety, which had not been present on imaging from three years prior. The right upper pole ureter appeared to extend toward the prior location of the uterus. This positioning suggests that this ureter may have ectopically inserted to the vagina, a common site of insertion. One would expect the patient to have constant urine leakage with insertion at this site. While she did not report long-standing leakage, it is possible that the atrophic right upper moiety was only producing minimal urine leading to a tolerable, small degree of leakage.

The patient underwent a sacrocolpopexy for POP at an outside facility approximately one year after her previous CT scan. It is unknown whether the duplicated collecting system was recognized at the time of surgery, and the sacrocolpopexy itself may have created an iatrogenic obstruction of the distal upper pole ureter leading to upstream hydronephrosis. One possibility is that attachment of mesh to the vaginal wall during the sacrocolpopexy obstructed the insertion of the upper pole ureter into the vagina. While obstruction in a duplex system associated with POP has been reported previously,\textsuperscript{7} to our knowledge, this is the first presentation of secondary obstruction of an ectopic upper pole ureter following sacrocolpopexy.

4. Conclusion

While newly symptomatic duplicated collecting systems are uncommon in adulthood, the diagnosis of a hydronephrotic duplex collecting system should be considered for patients presenting with flank pain and a cystic lesion in the upper kidney. Patients with refractory pain, UTI, and a nonfunctioning moiety can be managed with heminephroureterectomy of the affected moiety.

Author contributions

TDS – Investigation, Visualization, Writing-original draft; HHS – Writing-review and editing; AF – Writing-review and editing; LLW – Supervision, Writing-review and editing; KE – Conceptualization, Writing-review and editing.

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Declaration of competing interest

The authors declare no conflicts of interest.

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