CASE REPORT

Urinary incontinence in a dog with a duplex renal system and extramural ectopic ureter

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A 1-year-old female spayed French Bulldog was referred to the Interventional Radiology and Endoscopy Service for evaluation of urinary incontinence with partial response to medical management (phenylpropanolamine). Cystourethroscopy and retrograde ureteropyelogram were performed and revealed multiple congenital abnormalities including; a duplex renal system of the left kidney with one normal ureter with an intravesicular ureteral orifice and one extramural ectopic ureter with an associated branching and ureteral diverticulum. A dual vagina was also noted. Left neoureterocystostomy of the extramural ectopic ureter was performed and a temporary ureteral stent was concurrently placed and removed cystoscopically one month later, leaving the patient with two patent left ureters. Thirty-four months later the patient is mildly incontinent with no other comorbidities.

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INTRODUCTION

This case report highlights a duplex renal system and extramural ectopic ureter in a young female dog. A duplex renal system is a term used to describe a single kidney with two distinct pelvicalyceal systems, referred to as moieties. Each moiety, commonly called upper and lower moiety, drains into a distinct ureter which either fuses distally (partial duplication), or never fuses (true duplication) (Whitten & Wilcox 2001). In human medicine, ectopic ureters are almost always associated with a duplex renal collection system (Smith *et al.* 1989, Decter 1997, Geavlete *et al.* 2016, Merguerian & Rowe 2018). Based on the literature search, this phenomenon has only been described in dogs five other times in veterinary medicine (Esterline *et al.* 2005, Novellas *et al.* 2013, Newman & Landon 2014, Kopp *et al.* 2020, Sprocatti *et al.* 2020).

The patient had a standard work-up for urinary incontinence in a young dog including urine testing, blood work and abdominal ultrasound. Given only partial response to medical management with phenylpropanolamine (PPA), the decision was made to refer for specialty care. This report demonstrates the value of utilising endoscopic evaluation in conjunction with fluoroscopy; it enabled identification of the duplex renal system and its association with an extramural ectopic ureter.

CASE HISTORY

History

A 1-year-old 8.7-kg (19 lb) female spayed French Bulldog was presented to the primary care veterinarian. On examination, the patient was found to be dribbling urine and rectal exam revealed a urethral thickening near the pelvic brim. At the time of the visit, it was mentioned that the patient had been having urinary accidents (spotting, urination with excitation) that were presumed to be secondary to poor house training. Urinalysis and culture were normal with no evidence of microbial growth. One month later, the patient was re-presented as the urinary accidents had continued. An abdominal ultrasound revealed a questionable abnormality associated with the distal aspect of the left ureter; the left ureter did not make its normal curve as it approached the trigone. Aside from a prominent uterine stump, the abdominal ultrasound was otherwise unremarkable.

Advanced imaging and work-up

The patient was referred to the interventional radiology/interventional endoscopy department at the same hospital for further evaluation. The continence score of this dog at the time of examination, while on PPA (1.38 mg/kg PO, q 12 h), was rated as a 7.5 out of 10 (Appendix). This was an improvement from 5

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out of 10 before starting PPA. Based on the patient's clinical history and abdominal ultrasound findings, the decision was made to perform cystourethroscopy and a ureteropyelogram to further evaluate the urogenital tract.

Routine cystourethroscopy was performed with a 2.7-mm non-integrated 30° rigid cystoscope (Richard Wolf, Vernon Hills, IL, USA). At the vestibulovaginal junction there was a persistent paramesonephric remnant (PPMR) visualised extending to the cervix, diagnosing this as a dual vagina (Fig 1C). Just distal to the trigone, a tunnel with a ureteral orifice was noted on the left side from which urine was flowing (Fig 1B). A retrograde ureteropyelogram of this opening was performed using 3 mL of iohexol (240 mg/mL) by advancing an angle-tipped hydrophilic guidewire (Weasel Wire 0.025-in., Infiniti Medical LLC, Menlo Park, CA, USA) up the ureter, and an open-ended ureteral catheter (4 Fr., Cook Medical Inc, Bloomington, IN, USA) over that guidewire. The guidewire was removed and 1 to 3 mL of 100% of contrast was injected up the ureter to fill the ureter and the renal pelvis while recording under fluoroscopic guidance using digital subtraction angiography. This ureter was found to be ectopic and extramural in location and connected to the cranial aspect of the left renal pelvis (Fig 2A-E). A second ureteral orifice was identified in a normal position within the urinary bladder on the left side (Fig 1A). Retrograde ureteropyelogram of this ureteral orifice revealed that it connected to the caudal pole of the left kidney and did not communicate with the left ectopic ureter nor the cranial renal pelvis (Fig 2D, E). Taken together, these findings diagnosed a duplex collecting system of the left kidney. Additionally, a branching and ureteral diverticulum extending off of the ectopic ureter were identified (Fig 2C, E). Ureteropyelogram of the right ureter confirmed it to be normal in location, diameter and connect to a singular renal pelvis (Fig 2F). Next, an abdominal CT with intravenous contrast was performed under the same anaesthetic event. Ultimately, CT did not reveal any abnormalities not already identified.

Treatment and outcome

Approximately 6 weeks later, a neoureterocystostomy of the left ectopic ureter was performed with concurrent cystourethroscopy. Two weeks before surgery, urine culture was performed and was negative for microbial growth. Each left ureter was cannulated and the ectopic ureter stented (2.5 Fr ureteral stent, Infiniti Medical LLC, Menlo Park, CA, USA). Fluoroscopy with contrast was used to confirm placement in the renal pelvis (Fig 3A). The diverticulum associated with the ectopic ureter was identified, ligated and resected with 4-0 polydioxanone. The ectopic ureter was easily visualised with the guidewire present and it was isolated with careful peri-ureteral dissection. The ectopic ureter was standardly re-implanted at the ventral apex of the bladder using 8-0 polyglactin 910. A guidewire was maintained to ensure ureteral lumen patency during re-implantation. Once the ureteral re-implantation was complete, a 2.5 Fr ureteral stent (Infiniti Medical LLC) was placed in the re-implanted ectopic ureter (Fig 3B). The bladder was then closed using 4-0 poliglecaprone 25 in a simple continuous pattern and leak tested.

Four weeks later, cystourethroscopy was performed and the stent (Infiniti Medical LLC) was removed using a 3-prong grasper (Cook Medical Inc) through the working channel of the endoscope. A contrast study was performed confirming patency of the transplanted ureter (Fig 3C). Within the urinary bladder and urethra there was a large amount of inflammatory debris. Consequently, laser ablation of the PPMR to address the dual vagina was not performed, due to concern for the presence of infection. Urine was collected for urinalysis and culture and the stent was submitted for culture and sensitivity testing. The patient was started on cefpodoxime (12 mg/kg PO q24h, 2-week course) while the culture was pending. The urine culture showed no microbial growth but the stent cultured a *Staphylococcus pseudintermedius*. Fortunately, this bacteria was found to be sensitive to cefpodoxime.

The patient was continent at the time of last evaluation 21 days after ureteral stent removal while still on PPA. After that time, the patient was lost to follow-up. The initial post-operative plan included follow-up ultrasound to evaluate the re-implanted ureter as well as cystoscopy for possible laser ablation of PPMR. The owners were contacted 34 months post-surgery, at which time the patient was reported as having recurrence of mild urinary incontinence.

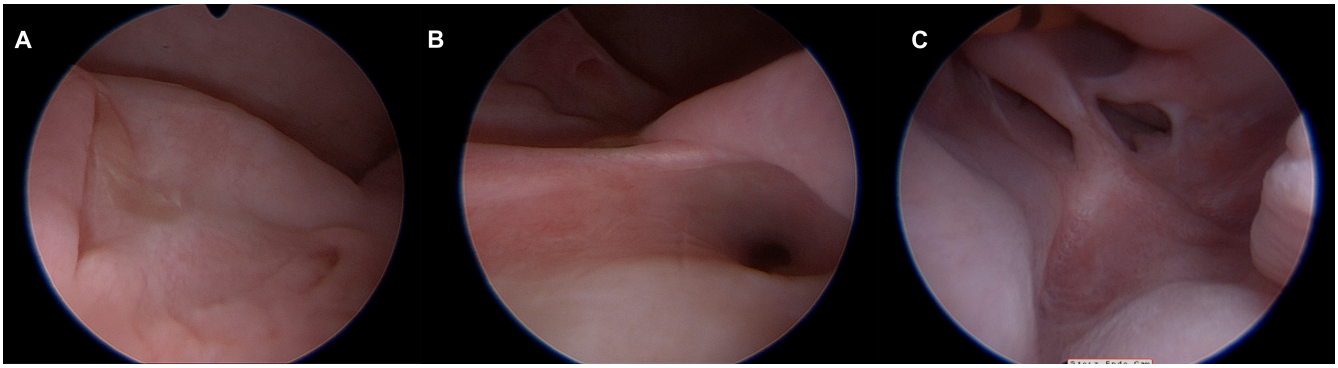

FIG 1. Endoscopic images taken during initial evaluation and diagnosis. Dog is in dorsal recumbency. (A) endoscopic image from the urinary bladder trigone documenting a normal left and right ureteral orifice, located approximately 1 cm cranial to the urethrovesicular junction. (B) endoscopic image of the left ectopic ureteral orifice in the proximal urethra, caudal to the bladder trigone. (C) persistent paramesonephric remnant at the vestibulovaginal junction extending to the cervix (dual vagina)

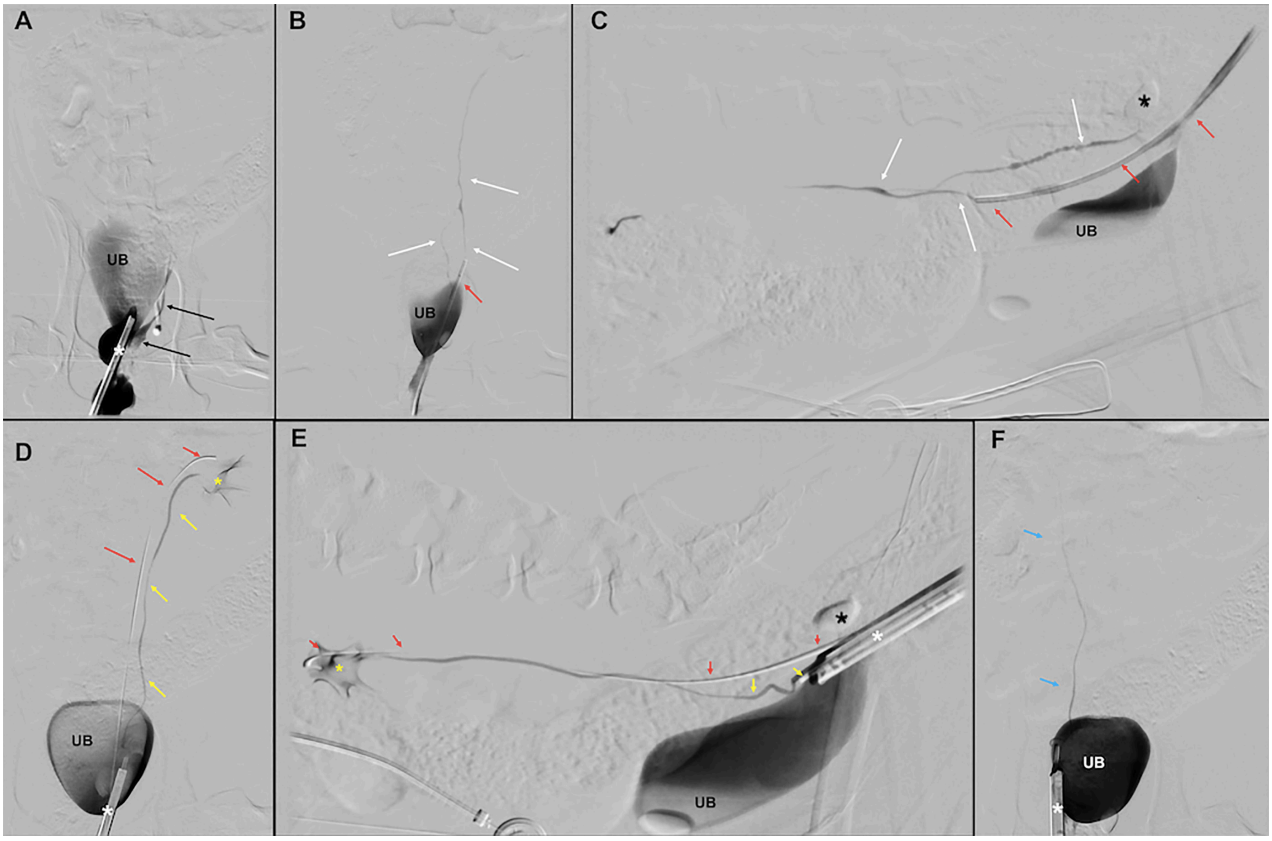

FIG 2. Fluoroscopic images taken during initial evaluation and diagnosis. All images are taken under digital subtractions angiography (DSA) setting on the fluoroscopy machine using iohexol contrast material (240 mg/mL). (A) A cystourethrogram documenting contrast in the urinary bladder (UB), the urethra, and the left extramural ectopic ureter (black arrows) coming off just caudal to the bladder trigone. The 2.7-mm 30° lens cystoscope[a] is identified with a white asterisk. (B) and (C) are ventral-dorsal (B) and lateral views (C) during a retrograde ureteropyelogram of the left extramural ectopic ureter. A 4 Fr. open-ended ureteral catheter[c] (red arrow) was advanced up the ureter and a contrast study is documenting the ureteral lumen (white arrows) to the renal pelvis. Notice, as contrast fills the ectopic ureter, there is a caudal branching of this ureter that leads to a diverticulum (black asterisk). (D) and (E) In the ventral-dorsal (D) and lateral (E) projections there is a 0.025″ angle tipped hydrophilic guidewire[b] (red arrows) in the extramural left ectopic ureteral segment that ends into the cranial pole of the left kidney. Concurrent retrograde ureteropyelogram of the normal left ureteral orifice (yellow arrows) documents a separate ureteral lumen connecting to the caudal pole of the left kidney. These two poles are not connected. (F) A retrograde ureteropyelogram of the normal right ureteral lumen (blue arrows)

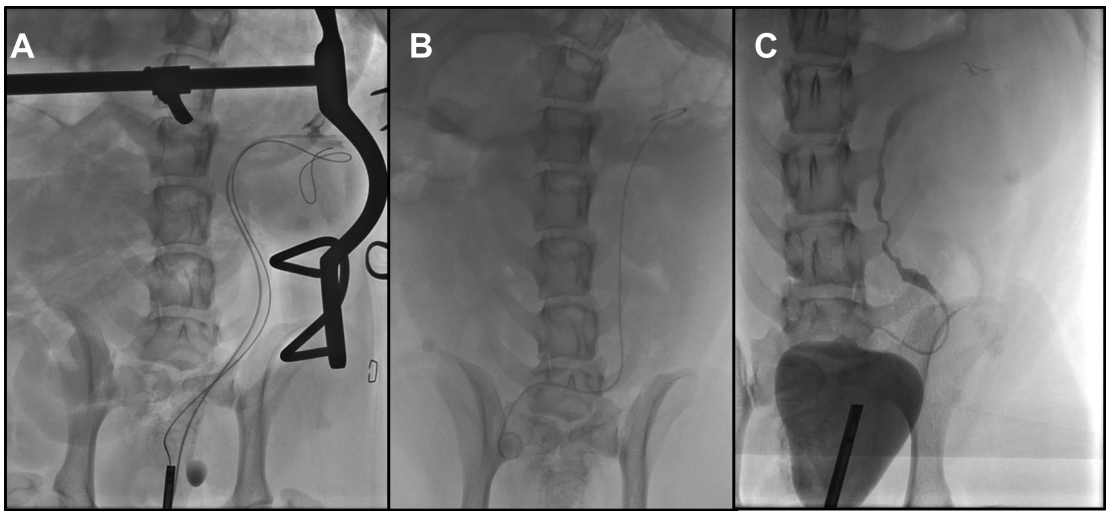

FIG 3. Fluoroscopic images taken during the neoureterocystostomy of the ectopic ureter as well as immediately after ureteral stent[d] removal. The dog is positioned in dorsal recumbency. (A) Fluoroscopic image of the left kidney prior to ureteral re-implantation with a 0.025″ angle-tipped hydrophilic guidewire[b] within the normal left ureter, coiled in the caudal pole of the left renal pelvis, and an open-ended ureteral catheter[c] in the left ectopic ureteral lumen advanced to the cranial pole of the left renal pelvis. (B) Fluoroscopic image after left extramural ectopic ureter correction by ureteral reimplantation with a 2.5 Fr double pigtail ureteral stent[d] left in situ. (C) Fluoroscopic image captured after ureteral stent[d] removed 4 weeks following re-implantation. Iohexol contrast material (240 mg/mL) fills the ureteral lumen extending to the cranial renal pelvis confirming ureteral patency

DISCUSSION

The following databases (PubMed, ScienceDirect, BioOne, Google Scholar) have been searched with the following keywords; ectopic ureter, ureteral ectopic, duplex renal system, duplex kidney, urinary incontinence (1966-present); the following textbooks have been consulted (Avery's Diseases of the Newborn 10th edition and the Handbook of Endourology). The following reports of duplex renal systems have been found doing these searches (Esterline *et al*. 2005, Ghantous & Crawford 2006, Novellas *et al*. 2013, Newman & Landon 2014, Kopp *et al*. 2020, Sprocatti *et al*. 2020).

The novelty of this case is in the identification of a duplex renal system in a dog and the way in which it was diagnosed and managed. Based on the literature search, a duplex renal system has been documented in veterinary medicine in dogs only five other times and interestingly each time it has been associated with an ectopic ureter, amongst other congenital abnormalities (Esterline *et al*. 2005, Novellas *et al*. 2013, Newman & Landon 2014, Kopp *et al*. 2020, Sprocatti *et al*. 2020). This phenomenon has been documented in cats only once (Ghantous & Crawford 2006). Imaging modalities utilised in these reports included ultrasound, pneumocystography, intravenous ureterography, vaginoscopy and CT. Management options included ureteronephrectomy, ureterectomy and neoureterocystostomy. In contrast to these reports, our patient's diagnosis was aided via cystoscopy with retrograde ureteropyelogram and was managed via neoureterocystostomy with temporary ureteral stenting.

Ectopic ureters are more common in both canine and human females compared to their male counterparts (Brannan & Henry 1973, Hayes 1984, McLoughlin & Chew 2000, Owen 2019). In humans, ectopic ureters are associated with a duplex renal system in 80% to 90% of cases, similarly with more females being affected than males (Decter 1997, Geavlete *et al*. 2016, Merguerian & Rowe 2018). In contrast, in the previously reported veterinary cases documenting duplex kidneys with ectopic ureters in dogs, four out of five were male (Esterline *et al*. 2005, Novellas *et al*. 2013, Newman & Landon 2014, Kopp *et al*. 2020, Sprocatti *et al*. 2020). It is often the upper moiety that connects to an ectopic ureter, parallel to the case described here (Brannan & Henry 1973, Decter 1997, Whitten & Wilcox 2001). However, the presence of a blunt ended diverticulum has not been reported in previous studies in veterinary medicine. There is no consensus on optimal management of these cases and evaluating individual moiety function is crucial to long-term treatment plans. Ultrasound combined with ^{99m}technetium-dimercaptosuccinic acid (^{99m}Tc-DMSA) is a contrast imaging modality that allows for a minimally invasive dynamic evaluation of the urogenital tract and assessment of the function and contribution of each draining ureter of the duplex kidney (Pattaras *et al*. 1999, Whitten & Wilcox 2001). Hemine-phrectomy, neoureterocystostomy and uretero-ureterostomy are all potential treatment options depending on individual moiety function (Mandell *et al*. 1981, Whitten & Wilcox 2001, Leavitt *et al*. 2012, Sander *et al*. 2015, Michaud & Akhavan 2017). Endoscopic evaluation and retrograde pyelography are possible, although less common in children compared to dogs.

In veterinary medicine, cystourethroscopy has come into favour as the gold-standard imaging technique with the highest sensitivity to diagnose ectopic ureters in both male and female dogs (Smith *et al*. 2010, Berent *et al*. 2012, Owen 2019). Cystoscopy has also gained favour as it allows simultaneous diagnosis and management of intramural ectopic ureters via cystoscopic-guided laser ablation of ectopic ureters (CLA-EU) (Berent *et al*. 2012). Cystoscopic-guided laser ablation of ectopic ureters involves using a laser to ablate the medial aspect of the ectopic ureteral wall in order to reposition the ureteral orifice into the urinary bladder (Berent *et al*. 2012, Owen 2019). For extramural ectopic ureters, traditional surgical correction is currently the treatment of choice (Holt & Moore 1995, McLoughlin & Chew 2000, Owen 2019).

The authors of this paper exclusively perform cystoscopy in conjunction with fluoroscopy when evaluating for ectopic ureters. A retrograde ureteropyelogram outlines the anatomy of the ectopic ureter and can confirm ureteral patency. In this case, it also demonstrated two separate renal pelvises, a ureteral diverticulum, and the presence of one ureter being extramural, necessitating the need for surgical correction. If cystoscopy was performed without concurrent fluoroscopic evaluation, the extramural ectopic ureter would have been incorrectly diagnosed as intramural as it initially tunnelled within the urethral wall. As such, CLA-EU of this ureter could have resulted in a life-threatening uroabdomen. Alternatively, had this patient only had a surgical exploration, the duplex renal system could have been missed. The ureteral lumen of both the left normal and ectopic ureter were <1 to 1.5 mm in diameter and were difficult to visualise without cannulation of the ureters. Had either of the left ureters been ligated and resected, then 50% of the associated renal draining system would have been compromised.

Our patient's incontinence recurred post-operatively. The cause of her ongoing incontinence is unclear as repeat imaging and subsequent urine cultures after surgery were never pursued as planned. The owners were not interested in future interventions such as laser ablation of the PPMR or collagen injections for the ongoing incontinence and were happy with her outcome.

Ethics statement

Anonymity and confidentiality of the patient and family have been maintained.

Conflict of interest

None of the authors of this article has a financial or personal relationship with other people or organisations that could inappropriately influence or bias the content of the paper.

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APPENDIX

Incontinence score: 10 = fully continent with no leakage; 7.5 = leakage only when resting and recumbent; 5 = leakage mostly when resting and recumbent, but also when walking with a full bladder between urinations; and 1 = completely incontinent with constant leakage and never urinating a typical-size urine puddle during voluntary micturition (Currao *et al*. 2013, Shea *et al*. 2019).