ECTOPIC URETEROCELE IN A MALE DOG WITH ASSOCIATED HYDRONEPHROSIS: A CASE REPORT

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Introduction

A ureterocele is a rare congenital anomaly characterized by a cystic dilation in the distal portion of the ureter, potentially causing ureteral obstruction.[3,8,14,16] Only 18 cases of ureteroceles have been documented in the English-language veterinary literature from 1971-2006, one of which was in a cat.[2,3,13,14,15] Females compose 89-95% of cases and most present with urinary incontinence during the first year of life. Rare occurrences have been reported in males, who tend to present later in life.[14] Ureteroceles commonly occur in conjunction with other congenital abnormalities associated with the urinary tract, such as an ectopic ureter, which is the leading cause of congenital urinary incontinence in dogs.[1,14] Ureteroceles can be intravesicular, which enter the bladder in the normal anatomical position, or ectopic, which are associated with ectopic ureters and have an orifice within or distal to the neck of the bladder.[8] Clinical signs vary depending on the size of the ureterocele and damage to the renal system. These can include stranguria, pollakiuria, incontinence, dysuria, uremic signs, recurrent urinary tract infections and abdominal pain and distension.[3,8,13,14] The primary etiology is relatively unknown; however many theories support a disruption in normal embryogenesis.[6,12,14] Surgical management can include neoureterostomy, neoureterocystostomy, nephroureterectomy and ureterocelectomy.[3,14] The purpose of this report is to describe the presentation, diagnostic plan, surgical procedure and outcome in an older male dog who presented with gastrointestinal signs and urinary incontinence associated with a right-sided ectopic ureterocele, hydroureter and severe hydronephrosis.

History and Presentation

An 8-year-old, male neutered schnauzer mix dog presented to the Animal Health Center of Mississippi State University for an acute episode of vomiting, straining to defecate and lethargy. Three days prior the owner reported the dog had consumed parts of a lunchbox, and a

foreign body was suspected. The dog had mild incontinence since adoption 3.5 years ago, which was managed on phenylpropanolamine. Upon presentation, the patient was quiet, alert and responsive. His abdomen was tense and distended with a potbellied appearance. He was historically sensitive upon abdominal palpation.

Due to the possible gastrointestinal obstruction, abdominal radiographs were performed. A large, ovoid, smoothly marginated, soft tissue mass in the mid to right abdomen was seen measuring 19.6 cm x 13.1 cm. This mass was causing caudal and leftward displacement of the intestines. On abdominal ultrasound, the mass was found to be a hydronephrotic right kidney. The kidney was extremely distended with anechoic fluid and no cortical or medullary tissue was seen. The right ureter was dilated at least 9mm. A thin-walled, anechoic fluid-filled structure was noted at the trigone of the bladder. Severe hydronephrosis, likely secondary to an ectopic ureter and concurrent ureterocele were diagnosed. A complete blood count and serum chemistry were within normal limits. A urinalysis revealed mildly decreased urine specific gravity at 1.010.

The patient was scheduled for surgery the following. Upon exploring the abdomen, right hydronephrosis and hydroureter were confirmed, and a right nephroureterectomy was performed. The right ureter was ligated at the point of contact with the bladder, leaving the intramural and ectopic portions of the ureter and ureterocele intact. The right ureter and kidney were submitted for histopathology, and findings were consistent with hydronephrosis with marked dilation of the renal pelvis and approximately 2mm of renal parenchyma remaining. The left kidney and urinary bladder appeared grossly normal. A urine sample was collected during surgery for culture and sensitivity which was negative. The patient recovered well in the intensive care unit over the next 24 hours and was discharged. The owner reported mild incontinence, which was thought to be due to permanent urethral sphincter damage.

Forty-four days post-operatively, the patient was noted to be straining to urinate. He appeared to involuntarily void a large quantity of urine in the house on three occasions that day. Urinalysis revealed a mild elevation in protein (1+) and trace SSA. A chemistry panel was within normal limits. Due to the possibility of a urinary tract infection, he was sent home on Baytril

One week later, now 54 days post-operatively, he presented with dysuria, stranguria, vomiting and straining to defecate. A large bladder was palpated and a red rubber was passed removing 360 ml of urine. Complete blood count was within normal limits and a serum chemistry revealed an elevation in ALT (121 U/L, ref 10-90). Abdominal radiographs revealed an enlarged bladder measuring 6.2 cm causing dorsolateral deviation of the descending colon. Abdominal ultrasound revealed a thin-walled, round, fluid-filled structure protruding into the right urinary bladder neck and proximal urethra which was considered the persistent ureterocele. CT scan was performed and 5ml of contrast was injected retrograde into the urethra. Contrast was seen filling the urethra, urinary bladder and left kidney. A large, non-contrast enhancing, fluid-filled sac, measuring 4.2 cm x 4.7 cm x 7.7 cm, was noted causing ventral deviation and compression of the bladder, and dorsal displacement of multiple intestinal loops and it was assumed to be the previously described ureterocele. Surgery was scheduled for a ureterostomy. The abdomen was opened and the fluid-filled structure was 2.5 times the size of the bladder. Fluid was collected and determined to be urine. The bladder was closed using 3-0 PDS in a simple continuous pattern. A leak check revealed leakage, so further sutures were used. A second leak check revealed adequate closure. The patient returned to the intensive care unit with a urinary catheter for 48 hours until discharge. Urinary incontinence worsened after surgery and was assumed to be related to the cystectomy.

Eight days after the second surgery, the patient was acutely lethargic and vomiting. He was brought to the emergency service for suspect uroabdomen. On fast scan, no free fluid was found and the bladder appeared intact. The patient was discharged as bloodwork was pending. Bloodwork results later revealed mild azotemia with an elevated BUN (27 mg/dl, ref 8-24) and Creatinine (1.49 mg/dl, ref 0.5-1.4). The client returned for 200 ml of subcutaneous fluids.

The following morning, the patient was anxious, panting heavily and was painful. Free fluid was found on ultrasound and an abdominocentesis was performed. A fluid creatine and potassium ratio was 3:1 (fluid creatinine 4.8 mg/dl, serum creatinine 1.61 mg/dl, ref 0.5-1.4), which is consistent with uroabdomen. BUN was also elevated (27mg/dl, ref 8-24). A positive contrast cystogram revealed a small leak from the dorsal aspect of the bladder, likely due to dehiscence of the bladder incision, and poor serosal detail consistent with peritoneal effusion. A urinary catheter was placed and IV fluids given for 48 hours until renal values normalized. A second cystogram was performed 10 days later, and no urine leakage was noted. The urinary catheter was then removed and the patient was discharged.

Pathophysiology

A ureterocele is the result of a dilation in the distal ureter and is commonly associated with ectopic ureters.[8,12] The development of this anomaly remains controversial; thus, a thorough understanding of renal anatomy and growth is necessary for diagnostic and treatment plans. Many theories suggest a disruption in the development of the urinary system during fetal growth.

The ureter, the tubular structure which connects the kidney to the urinary bladder, consists of an intravesicular aspect, which is composed of intramural and submucosal segments. The intramural segment is surrounded on all sides by the detrusor muscle, while the submucosal

segment, the terminal portion of the ureter, courses under the bladder mucosa into the submucosa, and is connected to the detrusor muscle dorsally.[8,12] Ureteroceles can be located in two positions: within the bladder wall at a normal anatomical position (intravesical or orthotopic ureterocele), or associated with an ectopic ureter (ectopic ureterocele), and thus, connected to the urethra (extramural) or tunneling through the submucosa and terminating distally as an intramural ectopic ureterocele.[8,10,16] Many intravesicular ureteroceles are small in size and usually asymptomatic.[13] Ectopic ureteroceles are more severe and commonly cause urinary outflow obstruction and clinical signs associated with uremia and hydronephrosis.[8,13]

During the development of an embryo, the ureter, termed the metanephric duct or ureteric bud during early stages of development, arises from an outgrowth on the mesonephric duct. The orifice of the ureteric bud is caudal relative to the orifice of the mesonephric duct.[8] The mesonephric duct develops, in both males and females, into the trigone of the urinary bladder, but with further development and introduction of hormones, will differentiate into the efferent ducts of the testis and prostate in males, and become vestigial in females.[14] The ureteric bud and mesonephric duct share a common excretory duct and opening into the bladder as it first forms.[12] As the bladder grows, the common duct is absorbed and the mesonephric duct and ureter begin to migrate.[8,14] The mesonephric duct rotates over the ureteric bud, exchanging locations so that the orifice of the ureter is established at the tip of the trigone.[8,12] The mesonephric duct continues to become the trigone of the bladder and an orifice in males is situated in the urethra.[12] Problems can arise if the developing ureter originates more cranially than normal on the mesonephric duct. The ureteric bud will be carried caudally with the mesonephric duct and will develop within the trigone of the bladder, thus preventing the ureter from establishing an orifice in the bladder cranially but resting caudally at the neck of the bladder or further distally at the urethra.[14] This is

how an ectopic ureter is formed. The higher the ureteric bud is displaced, the more ectopic the ureter will become.[8] If the ureteric bud begins to expand as it is connected to the mesonephric duct, this can cause a cystic-like dilation to form. As it moves through the bladder in an abnormal position and begins to dilate further, it can contribute to the formation of a ureterocele.[8] Other contributing factors to the ureterocele formation are stenotic ureteral opening, arrested myogenesis of the ureter and weakness of the connective tissue within the ureter.[14] The processes of developing a ureterocele are not fully understood and require further studies.

Many complications can arise with the development of a ureterocele. One of the most important is hydronephrosis.[5,6,12] Ureteral dilation occurs and urine begins to leave the ureter too slowly causing further damage to ureteral peristalsis. As the dilation continues, peristalsis becomes weaker, and eventually walls are unable to tighten with each contraction. Then the ureter is ineffective in delivering urine to the bladder.[5] With ureteral outflow obstruction and progressive dilation of the upper urinary tract, damage to the renal parenchyma and loss of renal function may occur.[7] Pressure within the kidney causes dilation within the renal pelvis, and ischemia within the kidney as it is compressed against the renal capsule.[7,11,12] If the contralateral kidney remains functionally normal, clinical signs of uremia are not recognized.[7] Azotemia requires a 70% loss of the functional renal unit.[13] The sooner the obstruction is resolved, the higher the likelihood the kidney can return to normal function. No permanent damage will occur if the obstruction is corrected within one week. If the obstruction remains for four weeks, 25% of renal function may return. However, if the obstruction is not corrected by this time, complete loss of renal function may occur.[11] The degree of hydronephrosis can progress such that the renal tissue is merely a pouch for urine storage. If no clinical signs are recognized, the only abnormal physical examination finding may be abdominal distension.[7]

An additional common complication with ureterocele formation is the recurrence of urinary tract infections.[5,12,13,14] This is a frequent result from the stagnant urine within the ureterocele and dilated urinary tract, leading to an increase in bacteria formation.[5,6,12]

Diagnostic Approach/Considerations

A thorough history and physical exam should be performed to discover signs of urinary incontinence, abdominal distension or pain, and clinical signs associated with uremia such as nausea, vomiting and lethargy. Complete blood cell count and serum chemistry are generally within normal limits, except when renal function has been compromised. In this case, an elevation in blood urea nitrogen (BUN) and creatinine may occur along with a decreased glomerular filtration rate (GFR). A urinalysis may reveal a urinary tract infection as these occur in 64% of cases of ectopic ureters.[14]

The size, shape, and location of the kidneys and bladder can be assessed with plain abdominal radiographs.[13] Intravenous urography is the method of choice in diagnosing ectopic ureters and pneumocystography can increase the sensitivity, however this is an involved process and may not be feasible. Contrast radiography, such as retrograde urethrography and vaginocystography, can locate an ectopic ureter.[14]

Ultrasonography allows visualization of the urinary tract and may provide evidence of megaureter, hydronephrosis, decreased urine flow, and possibly the orifice of an ectopic ureter. Hydronephrotic kidneys will present hyperechoic with abnormal renal structure such as dilated renal pelvis and loss of renal tissue. Ureteroceles are identified as smooth cystic structures located in the bladder wall or at the distal aspect of the ureter.[14] Computed tomography with contrast is an excellent procedure to identify ectopic ureters in dogs. Dilations in the bladder

wall, ureter and kidney are visible with a CT scan.[14] When available, cystoscopy can provide exact locations of the orifice of ectopic ureters and other congenital abnormalities in the ureter, bladder, urethra and vagina.[14]

Treatment and Management

Surgery is the treatment of choice in cases with an ectopic ureter and for ureteroceles.[15] Medical management alone is inadequate for treating ectopic ureteroceles and has not been proved to be successful in any diagnosed cases.[8,15] The choice of surgical technique depends on the extent of damage to the urinary system and should be catered to each patient.[6] The goals of surgery should be to alleviate any clinical signs, and to improve or maintain renal function if possible.[6,7] Surgery should be aimed at correcting the ectopic ureter and removing the ureterocele.[6] Surgical options include neoureterostomy with urethral-trigonal reconstruction, neoureterocystostomy, nephroureterectomy and ureterocelectomy.[6,15] The choice of technique(s) depends on a full assessment of both the upper and lower urinary tracts.[15] Neoureterostomy and neoureterocystostomy are surgical procedures to correct ectopic ureters. Nephroureterectomy will remove the affected kidney and ipsilateral ureter and a ureterocelectomy is required in cases of ureteroceles to dissect the tissue, removing all connections with the urinary system.[3,6,15]

Neoureterostomy with urethral trigonal reconstruction is a procedure used to repair intramural ectopic ureters. The distal portion of the submucosal ureter should be removed and the ureter repositioned into a normal location.[15] Neoureterocystostomy is the ureteral reimplantation of an extramural ectopic ureter.[15] Nephroureterectomy, a salvage procedure, is the removal of the affected kidney and ipsilateral ureter. This is indicated when the kidney is non-functional and the contralateral kidney has the ability to compensate.[15]

Once the ectopic ureter is corrected, the final step is to manage the distal hydroureter and ureterocele.[8] Ureterocelectomy is performed by dissection of the mucosa covering the cystic structure.[3,6,8,10] The ureterocele should be completely excised to remove the communication between it and the bladder.[6,8]

Medical management post-operatively may be necessary as some urinary incontinence may not resolve. This is especially true in cases of dogs who presented later in life and are more likely to have acquired urethral sphincter mechanism incompetence.[15] Alpha-adrenergic agonists such as phenylpropanolamine have been proved to reduce signs of urinary incontinence.[15]

Case Outcome

The patient in this report had a complex, right-sided ureterocele associated with an intramural ectopic ureter with concurrent hydronephrosis. Many treatment modalities were required and the patient recovered well following the episode of peritonitis, now one year ago. At the time of this report, normal renal function has been maintained by the unaffected kidney. The patient remains mildly incontinent, which is similar to his presentation before the original surgery. He has been managed well on phenylpropanolamine. In this case, the best management of the ureterocele was a nephroureterectomy, since the affected kidney was unviable from hydronephrotic atrophy, and concurrent ureterostomy to remove the compressing cyst-like structure.

Conclusion

Treatment of ectopic ureters and ureteroceles remains controversial. Renal function and severity of renal damage must be taken into consideration for each case. Reimplantation of an ectopic ureter should restore a connection between the kidney and the bladder, however, if a

ureterocele is present, it must also be managed. Failure to remove the ureterocele may result in persistent urinary tract infections because of the stagnant urine and can interfere with normal micturition. In cases of severe renal atrophy, a nephrectomy is indicated. A classification system has been proposed to differentiate treatment options and is in the following chart:[13]

Grade	Urinary System damage
Grade 1	Ureterocele with no evidence of upper urinary tract disease
Grade 2	Ureterocele with ipsilateral hydroureter and hydronephrosis
Grade 3	Ureterocele with bilateral hydroureter, hydronephrosis or chronic renal disease

This chart does not take into account the variability in size of the ureterocele or the location of the ureter such as ectopic or intravesicular. Further studies are required to create specialized treatment plans and a better understanding on the development of this congenital abnormality.

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