Four-dimensional CT excretory urography imaging findings, cystoscopy and exploratory surgery for the diagnosis of a vesicovaginal fistula in a young dog

Vierdimensionele excretoire urografie CT, cystoscopie en exploratieve chirurgie voor de diagnose van een vesiculovaginale fistel bij een jonge hond

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ABSTRACT

An eight-month-old, entire female German Shepherd was referred for investigation of continuous urinary incontinence not responding to medical therapy. A four-dimensional excretory urography (4D-CTEU) abdominal CT scan revealed a hypoattenuating fluid-filled structure dorsally to the urinary bladder, extending cranially, continuing as the uterine horns, and ending blindly caudally. On post-contrast images, during all contrast phases, a continuity of the urinary bladder and cranial vaginal walls was underlined by a hyperattenuating mucosal surface with the creation of a hypoattenuating fistulous tract. Cystoscopy and exploratory surgery confirmed the presence of a vesicovaginal fistula and revealed that the distal vagina and urethra consisted of one structure.

SAMENVATTING

Een acht maanden oude, vrouwelijke, intacte Duitse herder werd doorverwezen voor verder onderzoek naar de oorzaak van continue urinaire incontinentie die niet reageerde op medicamenteuze therapie. Een vierdimensionele excretoire urografie (4D-CTEU) computertomografische scan (CT) toonde de aanwezigheid van een vochtgevulde structuur dorsaal van de urineblaas. Deze structuur mondde naar craniaal toe uit in de uterushoornen terwijl het caudaal blind eindigde. Een continuïteit van de wand van de vagina en de urineblaas werd vermoed op basis van een contrast-capterend mucosaal oppervlak met de vorming van een hypoattenuerende fistelgang tussen beide structuren. Cystoscopie en de daaropvolgende chirurgie bevestigden de aanwezigheid van een vesicovaginale fistel en toonden dat de distale vagina en urethra uit één structuur bestonden.

INTRODUCTION

Vesicovaginal fistulas are female urogenital anomalies in which a direct communication is present between the vagina and the urinary bladder with involuntary urine passage into the vaginal vault resulting in continuous urine incontinence. In veterinary patients, vesicovaginal fistulas have rarely been described and can be acquired or congenital (Shea at al., 2019). In this case report, the computed tomographic (CT) findings are described of a congenital vesicovaginal fistula in a female entire dog that was confirmed with cystoscopy and exploratory surgery.

CASE DESCRIPTION

An eight-month-old, female entire German Shepherd was referred for investigation of urinary incontinence which had been present since the dog was a

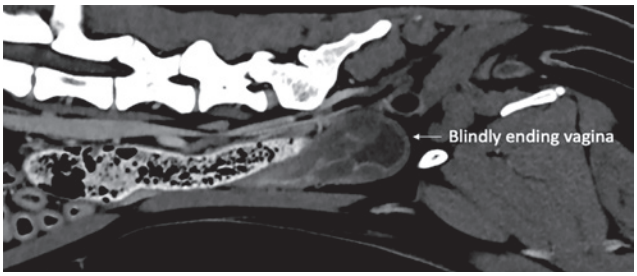

Figure 1. Post-contrast sagittal image showing a diffusely thickened (arrows) vaginal wall with an irregularly outlined inner aspect facing the lumen. Histopathological examination of the urogenital tract revealed a marked chronic lymphoplasmacytic cellular inflammation and ulceration, consistent with chronic vaginitis presumably due to urinary stasis.

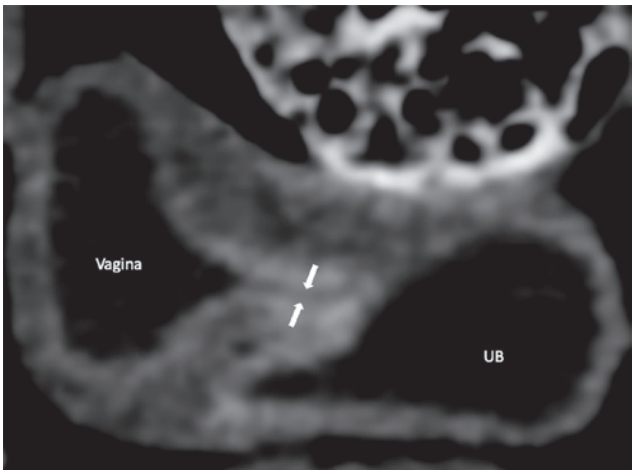

Figure 2. Post-contrast transverse image showing millimetric hypoattenuating vesicovaginal fistula outlined by continuous contrast-enhanced vaginal and urinary bladder (UB) walls (depicted by arrows).

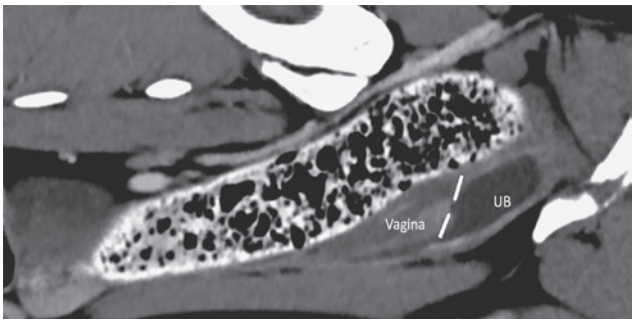

Figure 3. Post-contrast sagittal image showing a hypoattenuating indentation within the urinary bladder (UB) wall suspected to be a point of communication between the vagina and urinary bladder (depicted by arrows).

puppy. The dog was diagnosed by the referring veterinarian with bacterial cystitis and was treated with amoxicillin/clavulanic acid (Synulox, Zoetis, Belgium) 12.5 mg/kg orally, twice daily for ten days). Subsequently, the dog received desmopressin 0.1 mg orally three times daily for seven days, with no improvement. At the time of presentation, the dog did not receive any medication.

Physical examination was unremarkable, besides the presence of a small umbilical herniation. The external urogenital tract inspection did not reveal any anatomical abnormalities. A CT scan under general anesthesia was recommended to investigate the urinary incontinence. Prior to the CT scan, an ultrasound-guided cystocentesis was performed for culture and sensitivity testing.

The dog was premedicated with dexmedetomidine (Dexdomitor, Zoetis, Belgium; 5 µg/kg, IV) and butorphanol (Dolorex, MSD, Belgium; 0.02mg/kg, IV). Propofol (PropoFlo; Zoetis, Belgium; to effect) was given intravenously to induce general anesthesia. The dog was intubated, and anesthesia was maintained with isoflurane vaporized in oxygen.

A four-dimensional CT-excretory urography (4D-CTEU) abdominal CT scan (320-slice single-source CT scanner, Aquilion ONE ViSION, Canon Medical Systems, Belgium) was performed. A 4D-CTEU protocol was used, as previously described (Schwarz et al. 2021). Briefly, a pre-contrast helical scan of the abdomen, a 40s post-contrast injection (parenchymal phase) helical scan of the abdomen, a 120s post-contrast injection helical scan of the UVJ, a 180s post-contrast conventional excretory CT urography (CTEU) were performed, ROI was positioned at the ureter and an automatic peak of 100 HU triggering the 4D-CTEU series was established. Contrast was injected intravenously via the cephalic vein (700 mg/kg) with a power injector (OptiVantage, LF, Belgium). The images were acquired with the following scan settings: 120KV, 200 mA, slice thickness 0.5mm, matrix 512x512, reconstruction with low pass filter and spiral pitch factor 0.638. The images were viewed using a soft-tissue window (window width (WW) 350, window level (WL) 50) on a computer workstation (Apple Mac, Apple) with DICOM viewer software (Osirix v.13.0.1). A maximal intensity projection was applied to some imaging series (MIP).

The CT revealed a large tubular, smoothly marginated soft-tissue structure mildly distended at its caudal aspect with hypoattenuating fluid, located adjacent and dorsally to the urinary bladder. Its wall was diffusely thick with an irregularly marginated surface towards the lumen (Figure 1). The structure extended cranially to the urinary bladder and continued as mildly dilated uterine horns until the level of the ovaries. Caudally, it terminated blindly at the level of the first caudal vertebrae. On post-contrast CT images, homogeneous enhancement of the structure's wall was noted with increased contrast uptake of the inner wall lining. The urinary bladder wall was diffusely moderately thickened with a similar enhancement pattern. At the level of the first caudal vertebrae, a connection between the lumen of the fluid-filled structure and the lumen of the urinary bladder was consistently appreciated on multiple series as a millimetric hypoattenuating fistulous tract outlined by the continuous

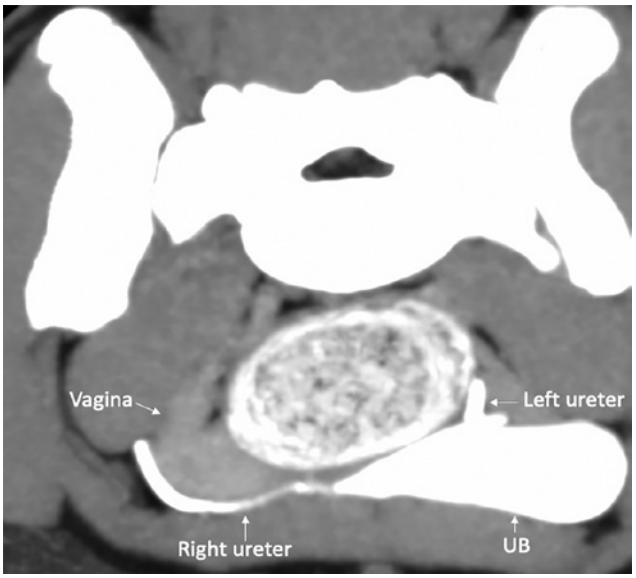

Figure 4. Post-contrast transverse image showing fluid filled vagina, moderately distended urinary bladder (UB), right ureter wrapped around the vagina and left ureter.

hyperattenuating inner wall layer of the urinary bladder and vagina (Figures 2 and 3). During all phases, no enhancement of the structure's fluid-filled cavity was seen. A small amount of contrast pooling was noted during the last series of the CT (early excretory phase) within the dependent cranial portion of the urinary bladder lumen. Both ureters were visible with no signs of ectopia. The right ureter was running adjacent to the fluid-filled structure and terminated caudally to the left ureter (Figure 4). There was faint mineralization within the urinary bladder lumen appreciated on its dependent wall. The urethra was mildly distended with hypoattenuating fluid during all CT series (pre- and post-contrast) with diffusely and uniformly enhancing walls. Both kidneys and ovaries were within normal limits.

The imaging findings were compatible with a blindly terminated, dilated vagina with suspected communication with the urinary bladder and the presence of some mineralized urinary sediment. A diffusely thickened and irregular vaginal wall most likely represents chronic vaginitis due to urinary stasis.

Urine culture was positive for *Proteus mirabilis* and amoxicillin/clavulanic acid was initiated based on sensitivity testing (Synulox, Zoetis, Belgium; 12.5 mg/kg per os, twice daily), which had to be continued until exploratory surgery three weeks later. Prior to surgery, a cystoscopy was performed which revealed a distended vagina, which ended into the urinary bladder. No separate urethral opening, neither a normal cervix could be identified. Subsequently, the dog underwent exploratory surgery. Abdominal inspection showed a severely enlarged proximal vagina which was fused to the dorsal part of the urinary bladder and formed a vaginal pocket. A fistula was present between this vaginal pocket and the urinary bladder at the level of the trigonum. The left ureteral opening ended in a normal position in the trigonum, whereas the right ureter was wrapped around the vagina and ended slightly more distal to the right ureter in the bladder (Figure 5). The urethra was very wide, measuring approximately 2 cm in diameter, and could not be distinguished from the distal part of the vagina. The uterine horns were macroscopically normal. Ovariohysterectomy and partial vaginectomy were performed by removing the vaginal pocket. The edges of the vesiculovaginal fistula were resected and the defect was closed in two layers with poliglecaprone 4/0 (Monocryl, Ethicon, Belgium).

Histopathological examination of the urogenital tract revealed a marked chronic lymphoplasmacytic cellular inflammation and ulceration, consistent with chronic vaginitis presumably due to urinary stasis. The endometrium was normal and under hormonal influence, which matched the active follicles observed in both ovaries.

DISCUSSSION

To the authors' knowledge, this is the first reported case of persistent urinary incontinence due to a congenital vesicovaginal fistula with a vaginal fluid-filled

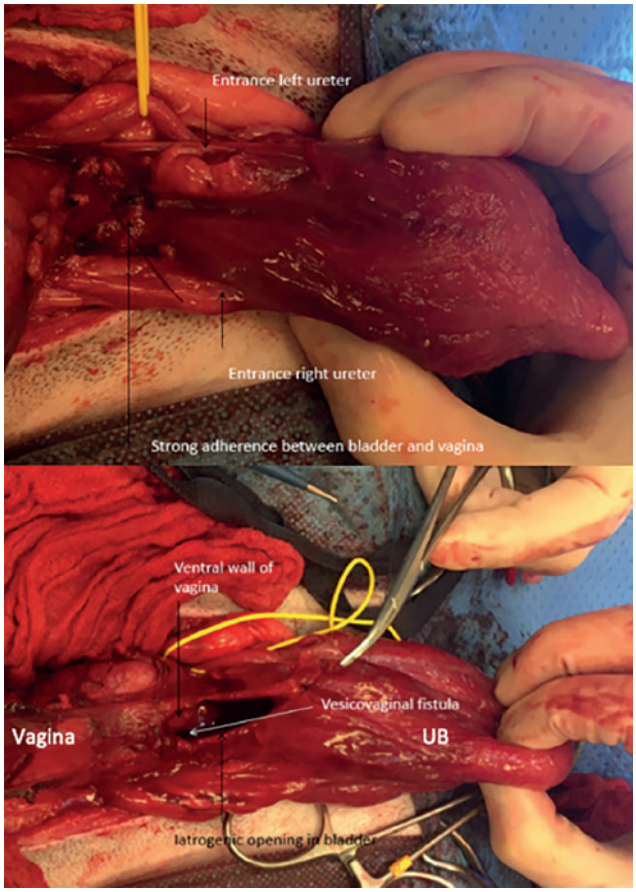

Figure 5. Intraoperative images showing the vesicovaginal fistula and the entrance of both ureters into the urinary bladder (UB), which is flipped caudally.

pocket in an entire female dog diagnosed on CT and confirmed with cystoscopy and exploratory surgery.

Urinary incontinence is a challenging pathology for owners and veterinarians. In intact canine patients, its prevalence has been determined to be below 0.5% and between 3-5% in spayed dogs (Holt and Thrusfield, 1993). Common causes of urinary incontinence in young female intact dogs are congenital anomalies, such as ectopic ureters, hypoplastic urethra, pelvic urinary bladder displacement, hooded vulva, or urogenital sinus malformation (Byron, 2017).

A congenital vesicovaginal fistula is the persistent communication between the vagina and urinary bladder via a small orifice. There is limited information and agreement regarding the etiology, diagnostics and therapeutic approach in the human literature (Martínez-Escoriza et al., 2014).

In small animals, only one case of a congenital vesicovaginal fistula in a five-year-old, female, spayed Maltese cross breed has been reported so far. In that case, definite diagnosis was reached via vaginoscopy after abdominal ultrasound and cystourethrography had failed to identify the presence of the vesicovaginal fistula. Whereas the urethra and vagina were normally developed, the congenital vesicovaginal fistula was associated with congenital aplasia of the left kidney and ureter (Shea et al., 2019). This contrasts with the here presented case, which displayed in addition to the vesicovaginal fistula, a maldevelopment of the vagina and the urinary bladder/urethra resulting in recurrent urinary tract infection.

Recurrent urinary tract infections are known to be linked with urovaginal fistula in small animals (Holt, 1990; Gregory and Trower, 1997; Connery and Spotswood, 2012; Agut et. al., 2016; Shea at al., 2019).

The urovaginal fistula, and in this case, the subtype of the vesicovaginal fistula, is a very rare subtype of the already uncommon urogenital fistulas in veterinary medicine, more particularly in small animals. The following different subtypes of the urovaginal fistulas have been reported: vestibuloperineal, urethrovaginal, ureterovaginal and urethrorectal fistulae (Holt, 1990; Gregory and Trower, 1997; Connery and Spotswood, 2012; Agut et. al., 2016; Shea at al., 2019).

Unlike humans, canines, as in the demonstrated case, tend to get less likely acquired than congenital urogenital fistulas, which are often associated with concurrent urogenital abnormalities and diagnosed in immature patients (Gregory and Trower, 1997; Connery and Spotswood, 2012).

The authors suspect that the cause of the congenital vesicovaginal fistula in the present case was an incorrect attachment of the vaginal plate to the urogenital sinus during the fetal development of the dog. This resulted in the proximal vagina connecting to the urinary bladder and a single distal urogenital tract combining the urethra and distal vagina. Secondary to the fistula, the proximal vagina turned in a urine filled pocket.

Acquired urogenital fistulas, although even more rare than congenital ones in dogs, have been reported in association with surgical complications (Ewers and Holt, 1992; Sokol et al. 2004), or as in one case report of a one-year-old, mixed, intact, female dog, secondary to the vaginal migrating grass awn (Agut et al., 2016).

Vaginal foreign bodies rarely cause urogenital fistulas. They tend to be undiagnosed for a long time resulting in chronic infectious inflammatory changes of the urogenital tract, and affected dogs present with hemorrhagic/ purulent vaginal discharge (Snead et al., 2010).

4D-CTEU combines temporal and spatial cross-sectional assessment of both ureters with ability of multiplanar viewing, which allows the observer to follow the ureteral course; in addition, it is considered advantageous over the traditional CTEU protocol, where data are not acquired in the temporary continuous manner (Schwarz, 2021). 4D-CTEU has 97% sensitivity and 94.6% specificity for ectopic ureters versus 73% sensitivity and 90.2% specificity for CT excretory urography (Schwarz, 2021). In the presen case, 4D-CTEU was helpful in tracking the right ureter, which was challenging due to its abnormal course; however, the vesicovaginal fistula had a similar appearance on all CT series. It was hypothesized that acquiring an additional late excretory phase or delayed contrast could be beneficial and perhaps underline the vesicovaginal fistula better. Additional CT series would not require much more time nor create additional costs for the clients. However, the authors believe that complete pooling of the contrast in the urinary bladder and vagina would be of limited value as proven in other veterinary case reports.

In the present case, 4D-CTEU was beneficial for ruling out ectopic ureters, although it was not crucial for the diagnosis of the vesicovaginal fistula. A consistent communication between the vaginal and urinary bladder wall was appreciated on all post-contrast CT series, giving strong suspicion of vesicovaginal fistula; therefore, further imaging was not performed. A retrograde urethrogram could have been performed to further enhance the visibility of the fistulous tract; however, this would have involved additional cost.

As an alternative diagnostic modality allowing visualization of the vesicovaginal fistula, a retrograde urethrography under fluoroscopy could be considered. It successfully identified a fistula between the proximal urethra and the remnant uterus and vagina in an English cocker spaniel with XX sex reversal (Yoon, 2018). However, fluoroscopic images can be difficult to interpret due to poor anatomical details and tissue superimposition (Schwarz, 2021). Additionally, fluoroscopy increases radiation exposure of the operator and prolongs surgical/anesthesia time.

MRI diagnosis of urogenital fistula in small animals has not been investigated; however, there is a study in the human literature, which showed that only

8 of 15 urinary bladder fistula were diagnosed, appreciated by MRI on post-contrast images (Semelka et al., 1997). The major drawbacks of MRI are the long acquisition and costs.

Although retrograde urethrography under fluoroscopy, vaginoscopy, retrograde urethrogram have been successful in diagnosing urogenital fistulas, they are less valuable in detecting other urinary tract pathologies than cross-sectional imaging studies. CT is considered a superior non-invasive diagnostic imaging modality allowing detailed and reliable assessment of the urogenital tract, and it can help to form an adequate surgical plan as it can get a better overview of the urogenital abnormalities and their extend, and exclude other concomitant pathologies. In a study by Kuhlman and Fishman (1990), a definitive diagnosis of urogenital fistula in humans was obtained in 60% of the patients based on delayed contrast-enhanced CT.

Although no standardized guidelines on the diagnosis of urogenital fistulas have been established in human nor in veterinary medicine, advanced imaging combined with endoscopy has been suggested by Agut et. al (2016), as the diagnosis was also performed in the present case . Due to the tissue folds, urogenital fistulas might be overlooked with solely vaginourethrography, cystourethrography or urethrography. A Doppler ultrasound with use of microbubble contrast medium has shown high sensitivity in detecting 11/12 cases of fistula in humans (Yu et al., 2004); hence, it could be considered as an additional non-invasive and safe procedure to confirm urovaginal fistulas following advanced imaging.

Diagnostic imaging, particularly contrast delayed CT urography, and more recently 4D–CTEU, plays a key role in ruling out anatomical pathologies, such as congenital ectopic ureters in persistently urinary incontinent dogs without response to (empirical) medical therapy (Schwarz, 2021). In the present case, a 4D-CTEU was performed due to its recently demonstrated higher diagnostic accuracy than CT excretory urography in diagnosing ectopic ureters, the most common congenital cause of incontinence in immature female dogs. However, identification and confirmation of vesicovaginal fistulas often demand a combination of diagnostic methods involving diagnostic imaging, endoscopy and exploratory surgery.

CONCLUSION

To the authors' knowledge, this is the first case report, in which 4D-CTEU findings of a congenital vesicovaginal fistula in a juvenile female dog are demonstrated and confirmed by cystoscopy and exploratory surgery. Vesicovaginal fistulas are very rare conditions in dogs and a combination of diagnostic techniques is needed to identify them, as shown in this case report. In addition, to direct visualization with endoscopy, CTEU or 4D-CTEU (if available) is the imaging method of choice to achieve an overview and full information of all urogenital abnormalities and their extent associated with a (congenital) vesicovaginal fistula.

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SMEEKBEDE OM EEN PAARD TE GENEZEN IN HET ALLEROUDSTE NEDERLANDS

Een van de alleroudste Nederlandse teksten (eind negende eeuw) betreft een gebed om een kreupel (lam) paard te genezen:

Visc flot aftar themo uuatare
uerbrustun sina uetherun.
tho ghelida ina use druhtin
the seluo druhtin thie thena uisc ghelida
thie gihele that hers theru spurihelti. AMEN

Vertaling

Een vis dreef over het water,
Vermorzeld zijn vinnen.
Toen genas hem onze Heer.
Dezelfde Heer, die de vis genas,
Moge die genezen het paard van deze lamheid. AMEN

Deze aanroeping (incantatie) om genezing is veel ouder dan het zeer bekende 'Hebban olla vogala nestas …' (ca. 1100), tekstje dat trouwens volgens filoloog Luc De Grauwe meest waarschijnlijk niet in het Nederlands, maar in een zuidelijk Engels dialect gesteld is.

Overgenomen van het Internet, waar ook de nodige referenties te vinden zijn.

Luc Devriese