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TITLE OF CASE ***Do not include "a case report"***
Specific treatment and outcome of urethrorectal fistula associated with type 1 atresia ani in a juvenile male dog.
SUMMARY ***Up to 150 words summarising the case presentation and outcome (this will be freely available online)***
A four-month-old, entire male, German wire-haired pointer presented with tenesmus due to type I atresia ani and with urination observed through this stenosed anal opening. A positive contrast retrograde urethrogram demonstrated a urethrorectal fistula and stricture of the penile urethra. Urine culture revealed heavy mixed bacterial growth, which was treated with appropriate antibiotics. Surgical correction of the congenital urethrorectal fistula was performed via a perineal approach with a 3.5 French catheter placed retrograde into the fistula to facilitate its dissection. The anal stenosis was addressed by surgical anoplasty and the urethral stricture via a scrotal urethrostomy. The dog recovered well with the owner reporting complete resolution of the clinical signs and urination via the urethrostomy site at six months postoperatively. To the authors knowledge this is the first reported case of congenital urethrorectal fistula associated with type 1 atresia ani in a male dog.
BACKGROUND ***Why you think this case is important – why did you write it up?***

Congenital urethrorectal fistulas (CUFs) connect the lumens of the urethra and the rectum.[1] CUFs have been associated with anorectal malformations in humans, horses and cats, with atresia ani most frequently.[2-7] Though CUFs and atresia ani have both been rarely reported separately in dogs, to the authors knowledge these conditions have not been reported to occur in combination.[8-17] This lack of evidence is likely, in part, to be due to affected dogs being euthanised or dying at an early age.

CUFs can be ligated via a perineal or ventral approach, with the perineal approach being preferred to avoid the higher morbidity associated with pubic symphysis osteotomy.[13] Type I atresia ani is defined as congenital stenosis of the anus.[18] Treatment options for type I atresia ani include bougienage, balloon dilation or excision of the stenosed portion. Treatment is often successful though reported complications include stenosis formation and faecal incontinence.[18] This case report describes the clinical presentation, diagnostic findings, surgical management and the medium term outcome of a juvenile male dog with CUF and type I atresia ani concomitantly.

CASE PRESENTATION ***Presenting features, clinical and environmental history***

A four-month-old, male entire, German wire-haired pointer, weighing 9.8 kg, presented with a history of tenesmus and dysuria. These signs had been present from the age of eight weeks old.

On physical examination the anus was noted to be smaller than expected and ventrally displaced, with the anal sac ducts opening dorsally (Fig 1). During urination the majority of urine was expelled via this opening, with only a small proportion voided via the penile urethra. No macroscopic abnormalities of the penis were noted, nor were there signs of faecal impaction and the bladder was normal on palpation.

INVESTIGATIONS ***If relevant***

Haematology revealed a leukocytosis (24.28x109/L ref:6-17x109/L) with mild mature neutrophilia (16.61x109/L ref:3-15x109/L), monocytosis (2.51x109/L ref:0.2-1.5x109/L) and lymphocytosis (4.6x109/L ref:1-4.3x109/L). Urinalysis showed pyuria (WBC 6 cells/hpf) with bacteriuria (rods). Urine culture revealed a heavy growth of Eschericha coli, Klebsiella pneumonia and Streptococcus spp., all sensitive to potentiated amoxicillin. Based on this potentiated amoxicillin (Clavaseptin; Vetoquinol) was prescribed orally at 20mg/kg twice daily as treatment.

Abdominal radiography showed that the rectum had a normal path until it deviated ventrally caudal to the ischium and then opened at the perineum. Abdominal ultrasound detected generalised abdominal lymphadenopathy, likely reactive and/or age-related. An anomalous tubular structure with no flow on Doppler was seen exiting the prostate on the left side but the origin/destination could not be confirmed. To further define this structure a positive contrast retrograde urethrogram and double contrast cystogram were performed. This identified a connection between the pelvic urethra and the rectum through which the contrast freely flowed. It also identified a narrowing of the penile urethra overlying the caudal part of the os penis (Fig 2).

Based on these findings type 1 atresia ani with urethrorectal fistula and secondary bacterial cystitis was diagnosed.

DIFFERENTIAL DIAGNOSIS ***If relevant***

N/A

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TREATMENT ***If relevant***

Under general anaesthesia the patient was positioned in dorsal recumbency. Castration and scrotal ablation were performed followed by scrotal urethrostomy.[19] The dog was then placed in sternal recumbency, the rectal opening of the urethrorectal fistula was identified and retrograde catheterised with a 3.5 French Small Animal/Tomcat urinary catheter (MILA International, inc. Kentucky). A 4cm midline perineal incision was made distal to the rectal opening and blunt dissection was performed to identify the catheterised urethrorectal fistula. The catheter was withdrawn and the fistula was double ligated with two encircling 3-0 polypropylene (Prolene; Ethicon) ligatures. The CUF was transected between the two ligatures and both ends of the remnant were oversewn with 3-0 polydioxanone (PDS II; Ethicon). A 6 French silicone Foley urinary catheter (Infusion Concepts. West Yorkshire, UK) was then placed via the urethrostomy into the bladder to check patency of the urethra. The bladder was manually expressed with urine passing around the catheter, confirming smooth flow of urine from the bladder to the urethrostomy, with no leakage observed. The perineal approach to the CUF was then closed with the deep fascia and subcutaneous fascial layers being closed with 3-0 poliglecaprone 25 (Monocryl: Ethicon) in a simple continuous pattern and intradermals placed with 3-0 poliglecaprone 25 (Monocryl: Ethicon). Anoplasty was performed by extending the existing rectal opening dorsally, taking care to preserve the anal sacs and ducts. The mucosa was then sutured to the skin using 4-0 polydioxanone (PDS II, Ethicon) in a simple interrupted pattern to reconstruct the anal opening (Fig 3).[18]

OUTCOME AND FOLLOW-UP

The patient recovered well and within 24 hours was urinating via the urethrostomy stoma. There was no urinary or fecal incontinence and the preoperative dysuria and tenesmus had resolved. Three weeks postoperatively all clinical signs had resolved. The urethrostomy was healed, the anus patent and there was no evidence of stenosis; perineal reflex was present and unremarkable. At six month telephone follow-up the dog was reported to have no further clinical signs.

Ten months after surgery the patient returned due to recurrent urinary tract infection and pyoderma around the urethrostomy site. These had responded well to medical management and at the time of presentation no stranguria or pyoderma were present. The owners reported that defecation was normal, although they noted he took longer to defaecate than his littermate. On clinical examination redundant folds of skin were present either side of the urethrostomy which, when standing, came together over the stoma site (Fig 4). Surgery was performed to resect the redundant skin folds. The tissue was excised with two elliptical incisions either side of and parallel to the urethrostomy site. The incisions were then closed with subcutaneous simple continuous suture pattern using 3-0 poliglecaprone 25 (Monocryl: Ethicon) and intra-dermal sutures of the same material. The patient made a full recovery, the surgical site healed uneventfully and his clinical signs fully resolved. Sixteen months after the initial surgery telephone follow-up revealed the dog was reported to have had no signs of urinary tract infections since the surgery.

DISCUSSION ***Include a very brief review of similar published cases***

This is the first report describing the clinical presentation, surgical treatment and medium term outcome of a male dog with type I atresia ani in conjunction with CUF. The absence of other case reports is unexpected for two reasons. Firstly due to the multiple similar reports in other species and also due to the embryological reliance of both the anus and urethra on appropriate formation of the urogenital fold and rupture of the cloacal membrane. Indeed it has been suggested that failure of one of these processes may be the underlying cause of both CUF formation and atresia ani, and therefore follows that these defects should be seen concomitantly more frequently.[7,17] Abnormalities in these processes may have also affected the caudal urogenital sinus, leading to the urethral stricture described in this case. However the aetiology of the stricture is not clear and could, alternatively, be related to the chronic urinary tract infection.[20-22]

Surgical treatment of both CUFs and type I atresia ani treated individually has been reported to be successful in the majority of cases.[13,17] Surgical management of both conditions simultaneously in this patient was performed due to the perceived contribution of both conditions to the clinical signs described. The urethrostomy was also performed due to concern that the urethral stricture might restrict urinary flow once the CUF was closed. Oversewing of the transected CUF remnants was performed to prevent any continued patency of the CUF. Though this increased the risk of bacterial sequestration at the site, the risk of granuloma/abscess formation was thought to be small in this scenario and outweighed by the risk of continued patency. Positive contrast retrograde urethrocystography could be considered immediately post-operatively to confirm that no persistent leakage of the CUF is present, although this was not performed in this case. Closure of the anoplasty was performed using absorbable sutures to avoid the need for disrupting the surgical site after surgery. However consideration could be given to using permanent sutures as these will cause less suture reaction and guarantee reassessment of the anoplasty at 10-14 days post-operatively.

Resolution of all clinical signs seen after surgery should encourage practitioners to consider treatment of type 1 atresia ani and CUFs where they are seen together.

LEARNING POINTS/TAKE HOME MESSAGES ***3 to 5 bullet points – this is a required field***

• Ensure that cases of atresia ani are thoroughly assessed for other congenital urogenital abnormalities, and vice versa.

• Surgical management of the CUF and type I atresia ani concomitantly led to a successful outcome and good life quality in the case presented here. This should encourage practitioners to consider treatment of these conditions if identified.

• Surgical correction of these conditions may be required prior to maturity, which may lead to the need for revision surgeries as the patient grows.

REFERENCES ***Vancouver style***

1. Kruger JM, Osborne CA, Lulich JP. Inherited and congenital disease of the lower urinary tract. In: Osborne CA, Finco DR, eds. Canine and Feline Nephrology and Urology. 1st ed. Philadelphia: Lea & Febiger, 1995:681–692.

2. Nastos C, Sotirova I, Papatsoris A, et al. Unusual Case of Urethrorectal Fistula in Adolescence in a Patient with a History of Congenital Anorectal Malformation. *J Endourol Case Rep* 2016;**2(1)**:24-26.

3. Le Duc E. Congenital rectourethral fistula: Report of a case without rectal anomaly. *J Urol* 1965;**93**:272–275.

4. Gideon L. Anal agenesis with rectourethral fistula in a colt. A case report. *Vet Med Small Anim Clin* 1977;**72**:238–240.

5. Kingston RS, Park RD. Atresia ani with an associated urogenital tract anomaly in foals. *Equine Practice* 1982;**4**:32–34.

6. Van Den Broek AHM, Else RW, Hunter MS. Atresia ani and urethrorectal fistula in a kitten. *J Small Anim Pract* 1988;**29**:91–94.

7. Chandler JC, MacPhail CM. Congenital Urethrorecal Fistulas. *Compendium* 2001;**23(11)**:995-1002.

8. Osuna DJ, Stone EA, Metcalf MR. A urethrorectal fistula with concurrent urolithiasis in a dog. *J Am Anim Hosp Assoc* 1989;**25**:35–39.

9. Goulden B, Bergman MM, Wyburn RS. Canine urethrorectal fistulae. *J Small Anim Pract* 1973;**14**:143–150.

10. Whitney WO, Schrader LA. Urethrorectal fistulectomy in a dog, using a perineal approach. *J Am Vet Med Assoc* 1988;**193(5)**:568–569.

11. Osborne CA, Engen MH, Yano BL, et al. Congenital urethrorectal fistula in two dogs. *J Am Vet Med Assoc* 1975;**166(10)**:999–1002.

12. Tobias KS, Barbee D. Abnormal micturition and recurrent cystitis associated with multiple congenital anomalies of the urinary tract in a dog. *J Am Vet Med Assoc* 1995;**207(2)**:191–193.

13. Ralphs SC, Kramek BA. Novel perineal approach for repair of a urethrorectal fistula in a bulldog. *Can Vet J* 2003;**44(10)**:822-823.

14. Holt PE, Long SE, Gibbs C. Disorders of urination associated with canine intersexuality. *J Small Anim Pract* 1983;**24(8)**:475-487.

15. Prassinos NN, Papazoglou LG, Adamama-Moraitou KK, et al. Congenital Anorectal abnormalities in six dogs. *Vet Rec* 2010;**153**:81-85.

16. Ellison WG, Papazoglou LG. Long-term results of surgery for atresia ani with or without anogenital malformations in puppies and a kitten: 12 cases (1983–2010). *J Am Vet Med Assoc* 2012;**240(2)**:186-192.

17. Vianna ML, & Tobias KM. Atresia Ani in the Dog: A Retrospective Study. *J Am Anim Hosp Assoc* 2005;**41**:317-322.

18. Baines SJ, Aronson LR. Rectum, Anus and Perineum. In: Johnson SA, Tobias KM, eds. Veterinary Surgery Small Animal. 2nd ed. Missouri: Elsevier, 2018:1793-1794.

19. Tobias KM. Scrotal Urethrostomy. In: Tobias KM, eds. Manual of Soft Tissue Surgery. 1st ed. Iowa: Wiley-Blackwell, 2010:383-388.

20. Vetterlein MW, Weisbach L, Riechardt S et al. Anterior Urethral Strictures in Children: Disease Etiology and Comparative Effectiveness of Endoscopic Treatment vs. Open Surgical Reconstruction. *Front. Pediatr.* 2019;**7**:5.

21. Kaplan GW, Brock JW, Fisch M et al. SIU/ICUD Consultation on Urethral Strictures: Urethral Strictures in Children. *Urology* 2014;**83**:S71-S73.

22. Latini JM, McAninch, JW, Brandes SB et al. SIU/ICUD Consultation on Urethral Strictures: Epidemiology, Etiology, Anatomy, and Nomenclature of Urethral Stenoses, Strictures, and Pelvic Fracture Urethral Disruption Injuries. *Urology* 2014;**83**:S1–S7.

FIGURE/VIDEO CAPTIONS ***figures should NOT be embedded in this document***

Figure 1. Small and ventrally displaced anus, with anal sac ducts (black arrows) dorsal to the opening.

Figure 2. Positive contrast retrograde urethrogram identifying a congenital urethrorectal fistula (black arrow) and urethral stricture (black arrowhead).

Figure 3. Appearance after castration, scrotal urethrostomy, anoplasty and congenital urethrorectal fistula ligation.

Figure 4. Urethrostomy site (left) and redundant skin folds either side covering the urethrostomy site when the legs are in a standing position (right).

OWNER'S PERSPECTIVE ***Optional***

N/A

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