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A rare case of a complex of multiple congenital anomalies diagnosed using computed tomography in a male puppy

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ABSTRACT: A 50-day-old male Maltese puppy was presented with difficulty in defaecation. Based on the clinical signs, and physical and radiographic examinations, including computed tomography, his condition was diagnosed as hypospadias, along with atresia ani and urethrorectal fistula. Hypospadias is a congenital malformation of the urethra that is relatively uncommon in dogs, while atresia ani is a rare congenital anomaly of the rectum and anus. An additional anatomic abnormality that can be associated with atresia ani is a fistula between the urogenital tract and rectum. After appropriate surgical procedures, the puppy recovered and defaecation via the anus was restored.

Keywords: dog; canine; atresia ani; congenital malformation; hypospadias; urethrorectal fistula

Hypospadias is a relatively uncommon congenital malformation of the urethra (Alam et al. 2005), which is characterised by the abnormal location of the urethral opening and imperfect closure of the external urethra. This abnormality occurs more frequently in males, but can also occur in females (Hardy and Kustritz 2005). In dogs, there are five forms of hypospadias, which are classified depending on the location of the urethral opening, as glandular, penile, scrotal, perineal and anal forms (Jurka et al. 2009). In most cases, the aetiology of hypospadias remains unclear, but generally, it is considered to be a congenital deformity that may be caused by inborn factors resulting in defects in androgen metabolism (Uda et al. 2004). Hypospadias has been reported in dogs, sheep, goats, cattle, rats, non-human primates and humans (Veena et al. 2011).

Atresia ani is also a congenital anomaly of the rectum and anus, which results in anal canal blockage and an abnormal path for the faeces; it is rarely reported in dogs and is even more infrequent in cats (Sherding 1994). Atresia ani is caused by failure of the urorectal fold to completely divide the primitive cloaca, or by failure of the anal membrane to perforate after anal genesis (Vianna and Tobias 2005). Four anatomic types of atresia ani have been reported in dogs and cats, and are graded as Types 1 through 4: congenital anal stenosis with perforated anus (Type 1), imperforate anus only (Type 2), a combination with a more cranial termination of the rectum as a blind pouch (Type 3) and discontinuity of the proximal rectum with normal anal and rectal formation (Type 4) (Vianna and Tobias 2005).

An additional anatomic abnormality that can be associated with atresia ani is a fistula between the urogenital tract and rectum. Urethral anomalies are rare and usually occur in association with other congenital malformations. Urethrorectal fistulas may be either congenital or can develop as acquired anomalies in humans and animals (Tobias and Johnston 2011). Although urethrorectal fistulas have been commonly observed in humans, they appear to be rare in dogs (Silverstone and Adams 2001); however, the incidence of urethrorectal fistulas in dogs is unknown, and no breed predisposition has been identified (Goulden and Bergman 1973).

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Here, we report a rare case of a complex of multiple congenital malformations in a male puppy, diagnosed by means of computed tomography (CT) scanning.

Case description

A 50-day-old male Maltese puppy was presented to the Nowon Animal Medical Center, Nowon-Gu, Seoul, Republic of Korea, with difficulty in defaecation. According to the owner, he seemed to have constipation and a watery stool occasionally leaked from around the inguinal region. No other problems were detected by the owner.

On physical examination, there were some apparent abnormalities. Firstly, an incomplete skin fusion along the cranial part of the ventral prepuce and perineum, a small penis with an imperfect penile shape and the absence of a urethral opening in the appropriate position were noted (Figure 1). Furthermore, there appeared to be no anal opening, and the tail was abnormally short. Yellowish watery stools leaked through the perineal skin fissure, but not from the anus. Using a 6-Fr rubber catheter, urethral meatus was identified through this perineal fissure. Blood work values and vital signs, such as body temperature, heart rate and respiratory rate were within normal physiological ranges. Plain abdominal radiography revealed abdominal distension and megacolon secondary to faecal retention (Rahal et al. 2007) (Figure 2). In accordance with the observations and physical examination, this puppy was considered to have a perineal form of hypospadias associated with Type 2 atresia ani, and communication between the rectum and urethra was also suspected.

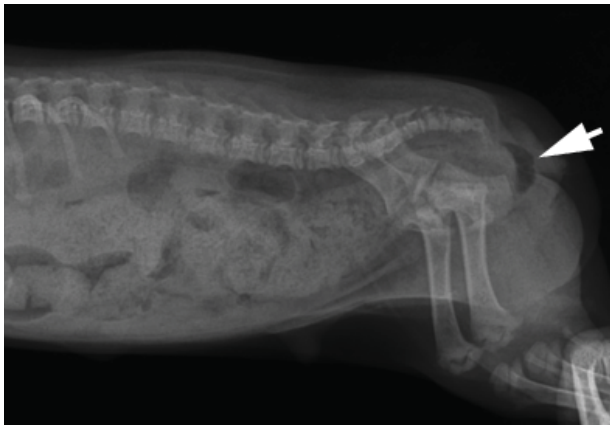

Figure 2. Right lateral plain abdominal radiographic view of Type 2 atresia ani and agenesis of the tail. Note that the colon is completely filled with faeces, and the dead end of the rectum is identifiable (white arrow)

For more detailed investigations, the puppy was anaesthetised and imaged using CT. An ionic contrast medium was administered through the perineal opening in retrograde fashion, using a 6-Fr red rubber catheter. On the CT scan, an abnormal communication between the ventro-distal rectum and perineal urethra was clear. It was diagnosed as a urethrorectal fistula of about 1.6 mm in diameter (Figure 3). Abdominal intestines were also contrast-enhanced via this fistula.

To correct these multiple problems, anoplasty and urethrorectal fistulectomy were performed. Under general anaesthesia using isoflurane, the patient was positioned in sternal recumbency, and the surgical area was clipped and scrubbed with povidone iodine. Briefly, an 18-gauge over-the-needle catheter was placed into the perineal opening for urethral indication. A vertical incision was made in the skin over the anal dimple and the distal blind

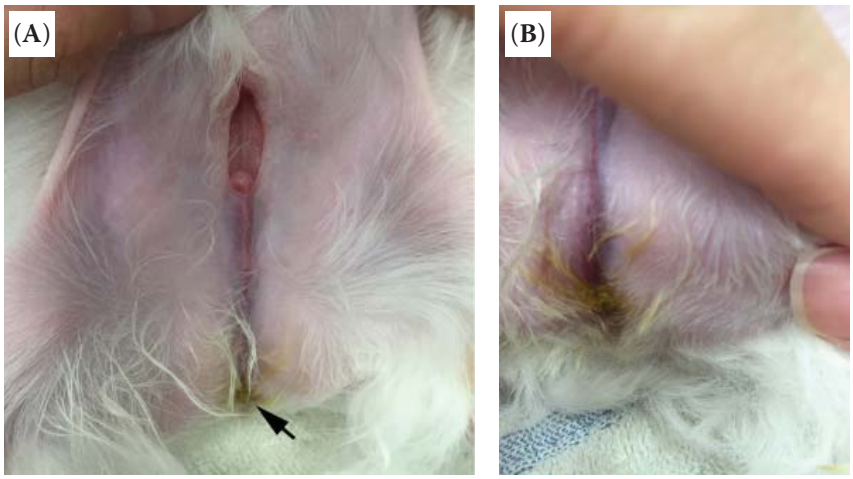

Figure 1. Appearance of the perineal form of hypospadias. Note the non-union prepuce and short penis, and the mispositioning of the urethral orifice. (**A**) Yellow watery faeces leaking through the perineal opening (black arrow); (**B**) close-up photograph of the perineal region

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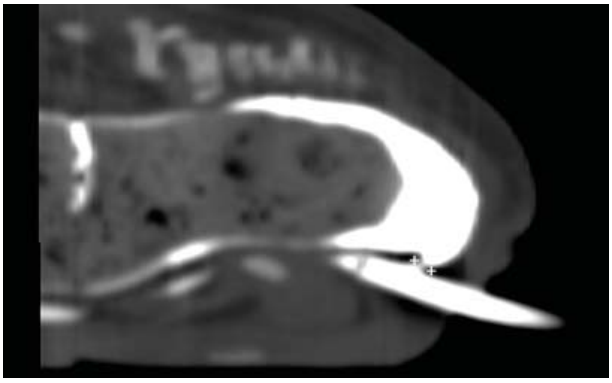

Figure 3. Post-contrast computed tomography image. The contrast medium was administered through the perineal opening in a retrograde fashion. A urethrorectal fistula can be seen between the plus (+) signs. The rectum was also contrast-enhanced via this fistula

rectal pouch was gently dissected away from the surrounding tissue. A stay suture was placed in the identified blind rectal pouch through the surgical opening. The rectal pouch was incised and sutured circumferentially to the nearby subcutaneous tissue and skin using 4-0 absorbable, simple interrupted sutures (Goulden and Bergman 1973) (Figure 4). Next, the fistula communicating with the rectum and urethra was carefully identified, dissected from circumferential tissues and securely double-ligated with 4-0 black silk. Surgical intervention of hypospadias was not attempted due to the presence of a distinct urethral opening at the perineum.

Because of the prolonged constipation, normal defaecation through the anus was not perfectly re-established immediately after surgery. After surgical intervention, no more faecal content leakage was observed through the perineal opening but urination persisted. The puppy recovered well and has been followed up for almost two years after the surgery. He can now defaecate normally through the anus and urinate through the perineal opening without faecal or urinary incontinence. No signs of constipation, megacolon or urinary tract infection were observed. Regular urinalysis was not performed due to the owner's satisfaction with the puppy's current health status.

DISCUSSION AND CONCLUSIONS

The aetiologies of hypospadias, atresia ani and recto-urethral fistula have not been clearly elucidated. It is assumed that failure of the urorectal septum to separate the cloaca completely into an anterior urethrovesical segment and a posterior rectal segment during embryological development is the cause of these congenital anomalies (Tobias and Johnston 2011).

A similar congenital anomaly in a kid was reported by Veena et al. (2011), who described an unusual case of atresia ani concurrent with hypospadias. The authors presumed one cause for the increasing incidence of hypospadias may be environmental pollution, which causes animals to be exposed constantly to oestrogenic compounds, resulting in reproductive problems. In lambs, most animals with hypospadias also have atresia ani, and the co-existence of these two abnormalities has been considered to be due to the origination of both in the urogenital and anal folds from the common cloaca in the hindgut developmental stage (Jurka et al. 2009). In female dogs, recto-vaginal fistulas are commonly present in association with atresia ani Type 2 abnormalities. Partial tail agen-

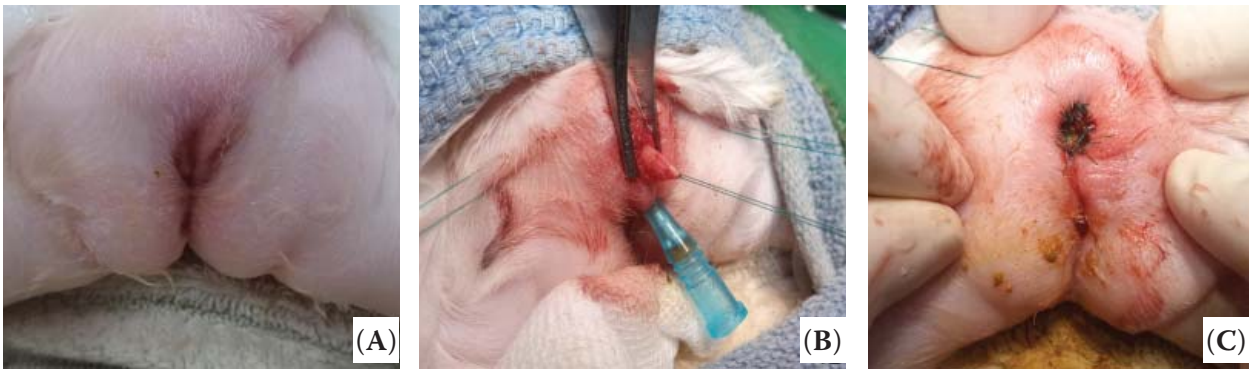

Figure 4. (**A**) Appearance before surgical operation, showing atresia ani. (**B**) A stay suture was placed in the rectal pouch, an 18-gauge over-the-needle catheter was placed to secure the urethra. (**C**) Post-operative appearance after anoplasty

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esis has also been reported in dogs in conjunction with recto-vaginal fistula and atresia ani (Goulden and Bergman 1973).

Thorough physical examination and apparent clinical signs are quite sufficient to establish the diagnosis; therefore, radiographic studies may not be necessary to confirm the abnormalities. Nevertheless, advanced diagnostic imaging modalities, such as CT or magnetic resonance imaging, are considered important for evaluating the precise status of the congenital abnormality. In a case of an English bulldog (Silverstone and Adams 2001) who had a history of recurrent cystitis, a urethrorectal fistula was not diagnosed until a fourth urinary tract contrast study was performed eight years after the patient was first referred to the veterinary hospital. As in this case, a definitive diagnosis of urethrorectal fistula is often difficult to achieve using only plain radiographic studies. CT can provide useful anatomic information and can be used for diagnosis, as well as for evaluating patients pre- and post-operatively.

The true incidence of congenital abnormalities of the genitalia, rectum and anus in dogs is hard to determine, because many puppies may be euthanised before they are diagnosed by veterinary practitioners. Veterinary patients presenting with anorectal abnormalities should be carefully evaluated for other anomalies, because approximately one-third of humans with anorectal abnormalities have anomalies in other body areas (Vianna and Tobias 2005). Surgery should be performed as soon as possible to avoid deterioration of the physical condition, irreversible megacolon and possible infection of the ascending urinary tract. Castration of affected animals is recommended because urethrorectal fistula is considered to be a genetic defect (Rahal et al. 2007).

In conclusion, a complex congenital abnormality of the rectum, anus and genitalia of a male puppy was surgically treated and functionally corrected to some extent. A complex of multiple congenital malformations characterised by hypospadias associated with atresia ani and urethrorectal fistula in one individual has rarely been reported in the veterinary literature, particularly in male puppies.

REFERENCES

Alam MR, Shin SH, Lee HB, Choi IH, Kim NS (2005): Hypospadias in three calves: a case report. Veterinarni Medicina 50, 506–509.

Goulden B, Bergman MM, Wyburn RS (1973): Canine urethra-rectal fistulae. Journal of Small Animal Practice 14, 143–150.

Hardy RM, Kustritz MV (2005): Theriogenology questions of the month. Hypospadias. Journal of the American Veterinary Medical Association 227, 887–888.

Jurka P, Galanty M, Zielinska P, Max A, Sysa P (2009): Hypospadias in six dogs. Veterinary Record 164, 331–333.

Rahal SC, Vicente CS, Mortari AC, Mamprim MJ, Caporlli EH (2007): Rectovaginal fistula with anal atresia in 5 dogs. Canadian Veterinary Journal 48, 827–830.

Sherding R (1994): The Cat: Disease and Clinical Management. 2nd edn. Churchill Livingstone. 1353–1401.

Silverstone AM, Adams WM (2001): Radiographic diagnosis of a rectourethral fistula in a dog. Journal of the American Animal Hospital Association 37, 573–576.

Tobias T, Johnston S (2011): Veterinary Surgery: Small Animals. Elsevier Saunders. 1572 pp.

Uda A, Kojima Y, Hayashi Y, Mizuno K, Asai N, Kohri K (2004): Morphological features of external genitalia in hypospadiac rat model: 3-dimensional analysis. Journal of Urology 171, 1362–1366.

Veena P, Sankar P, Suresh RV, Kokila KS, Lakshmi ND (2011): Congenital absence of tail with atresia ani and hypospadias in a kid: a case report. Journal of Advanced Veterinary Research 1, 76–79.

Vianna ML, Tobias KM (2005): Atresia ani in the dog: a retrospective study. Journal of the American Animal Hospital Association 41, 317–322.

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