

## **Surgical treatment of intra-abdominal vaginal cyst severely adhered to associated organs in a dog**

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### *Abstract*

A twelve-year-old female Shih Tzu was presented for investigation and treatment of a vaginal mass. Partial resection, intracavitary fluid drainage and mucosal lining curettage had been made in the previous laparotomy. Computed tomography (CT) and perioperative gross appearance revealed a large vaginal cyst extended into the pelvic canal that was adhering to the bladder trigone, both ureters, the urethra and the descending colon. The mass was resected completely. The dog recovered uneventfully. To the author's knowledge, this is the first report of a canine intra-abdominal vaginal mass that was preoperatively evaluated using CT and the histological result was most consistent with the Mullerian cyst. This report was also highlighted with intra-pelvic extension and severe adhesion consequences from previous surgeries, making the current surgical treatment much more complicated.

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**Keywords:** dog, intra-abdominal, surgical treatment, vaginal cyst

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## Introduction

A vaginal cyst is an uncommon disease in which the fluid-containing structure is formed within the vaginal wall. The prevalence is approximately less than 1% in humans based on the literature and may originate from either congenital or acquired conditions. Vaginal cysts are classified into different groups based on the location and histological appearance of the lining of epithelial cells. In human patients, a Mullerian cyst is the most common in the congenital group derived from paramesonephric remnants, followed by urogenital sinus and Gartner duct cysts. The most common acquired type is a squamous inclusion cyst which typically is presented post-surgically or post a traumatic event (Heller, 2012; Malpica, 2009). In animal patients, the vaginal cyst locations that have been described in dogs are intra-abdomen and extra-abdomen. There have been a low number of reported cases of an intra-abdominal vaginal cyst in female dogs extending from the urogenital tract at the cervical level into the abdomen cranially, with all such cases being diagnosed as a congenital Gartner duct cyst derived from mesonephric remnants (Cauvin *et al.*, 1995; Fujita *et al.*, 2004; Kim *et al.*, 2007; Siddorn and Mann, 1993; Watanabe *et al.*, 2014). The congenital cyst of mesonephric origin can be associated with other developmental abnormalities of the urinary system (McIntyre *et al.*, 2010), with one report of a multiplex malformation in a canine vaginal cyst (Fujita *et al.*, 2004). However, a Mullerian cyst of the vagina is very rare in animal patients and has only been described in one report of an extra-abdominal vaginal wall cyst in a bitch (Jimenez *et al.*, 2019). In humans, most cases are asymptomatic in the early stage. Treatments would be attended in the major symptomatic cases (Heller, 2012; Eilber and Raz, 2003). Accordingly, clinical signs of a canine vaginal cyst are nonspecific, varying from asymptomatic to severe progressive signs. Frequently, the symptomatic clinical signs of an intra-abdominal vaginal cyst are caused by the progression of the cyst to other abdominal organs, such as dyschezia, tenesmus and dysuria (Cauvin *et al.*, 1995; Kim *et al.*, 2007; Watanabe *et al.*, 2014). Swelling of the perineum has also been reported in one extra-abdominal case (Jimenez *et al.*, 2019). The diagnoses in previously reported cases have been made using abdominal radiography, endoscopy, contrast media radiography and ultrasonography (Cauvin *et al.*, 1995; Fujita *et al.*, 2004; Jimenez *et al.*, 2019; Kim *et al.*, 2007; Watanabe *et al.*, 2014). Computed tomography (CT) is an excellent diagnostic tools to enhance the intra-pelvic view for planning appropriate preoperative treatment (Jimenez *et al.*, 2019). Magnetic resonance imaging (MRI) has also been used in human medicine (Heller, 2012). Treatment of vaginal cyst, marsupialization has been reported in canine cases as an effective delicate procedure in terms of preventing postoperative neurovascular injury (Cauvin *et al.*, 1995; Holt, 1993), postoperative recurrence occurred in one case description (Watanabe *et al.*, 2014). Instead of intracystic lining membrane removal, a complete cystectomy and omentalization of the remnant cystic wall have been reported as a curative treatment in

many intra-abdominal and extra-abdominal vaginal cystic cases (Fujita *et al.*, 2004; Jimenez *et al.*, 2019; Kim *et al.*, 2007; Siddorn and Mann, 1993; Watanabe *et al.*, 2014).

The current case report involved the surgical resection of a vaginal cyst which was recurrent and severely adhered to multiple surrounding organs after the dog had undergone an ovariohysterectomy and partial resection with mucosal lining epithelium curettage.

## Case description

A 12-year-old female Shih Tzu weighing 6.7 kg was referred to the private animal hospital with a month-long history of slow progress dysuria and dyschezia. Prior to the presentation, the dog had undergone an ovariohysterectomy (due to cystic endometrium hyperplasia with a fluid-containing enlarged uterus) at the initial referring hospital 3 months earlier. Information of any intra-abdominal abnormalities was not available. At the private animal hospital, a large anechoic cystic mass was detected located caudodorsally to the bladder trigone based on ultrasonography. The clinical signs transiently decreased after ultrasound-guided fine needle aspiration of the mass and drainage of the light-yellowish mucoid fluid. The laboratory analysis identified a low number of cells and transudate of the fluid that had accumulated inside the mass. Two weeks after the initial presentation, the dog continued to experience the clinical signs of dysuria and dyschezia. Repeated ultrasonography of the mass was performed. The increasing size of the cystic mass with a large volume of fluid inside was noted. The hematological tests revealed that all parameters were within normal ranges. Based on the owner's decision, a second caudal midline laparotomy was performed. The large cystic mass was found to have originated from the lower uterine stump and attached to the caudal of the descending colon, both distal ureters, the dorsal surface of the bladder trigone and the urethra. The fluid was drained using aspiration and partial mass removal was undertaken by proximal blind-end resection, in which the excised part did not adhere to any organs. The dissection was discontinued and a curette was used to remove the lining secretory membrane on intra-cystic wall to confirm that there would not be any fluid production postoperatively. Then, the residual cystic end that had adhered with surrounding organs was sutured. Due to the consideration of the potential risk of the destructive consequence to associated visceral organs following further dissection and the high morbidity to neurovascular structures, the distal part of the tissue was not removed completely. The dog recovered and was monitored closely for clinical problems. The excised tissue was submitted for histopathological examination which revealed a normal, stratified, surface epithelial layer of the endometrium and a dense interlacing of spindle-shaped cells with the smooth muscle of the myometrium, which consisted of uterine tissue without evidence of any parenchymal tumor. One week after the surgery, the dog was presented with a recurrence of dyschezia signs. The

dog was referred to the referral animal hospital for additional diagnosis and treatment.

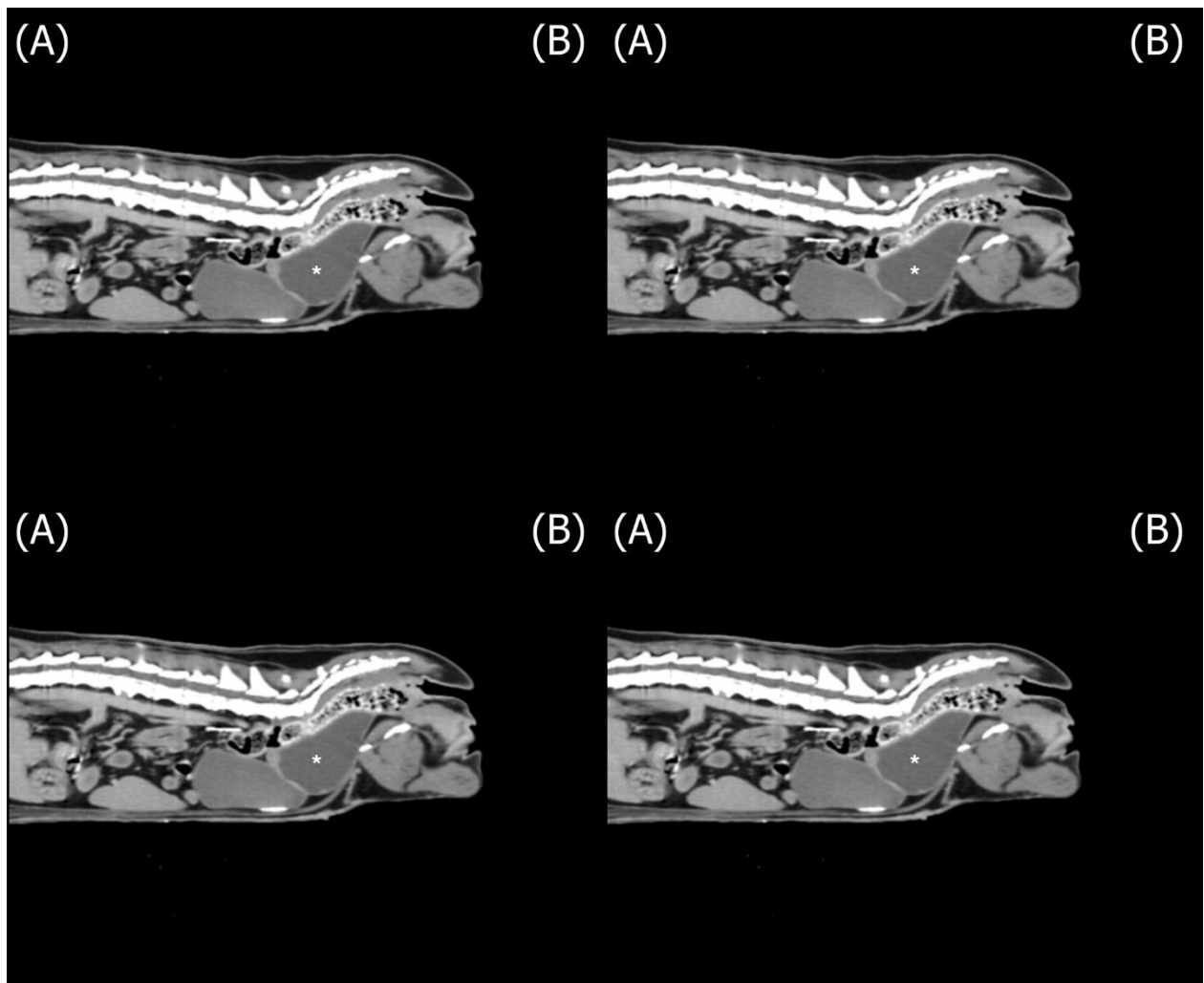
At presentation to the referral animal hospital, a painless smooth mass was detected using caudal abdominal palpation but no other abnormalities were noted during the physical examination. The complete blood count and blood chemistry profile were within the normal limits. Abdominal ultrasonography identification was similar to the previous referral findings. However, the caudal end of the mass was not identified due to extension into the pelvic cavity; however, a cystic uterine stump was suspected. Contrast CT was considered for surgical planning, anatomical localization and evaluation of the potential risk to associated abdominal organs. The images were obtained in the transverse, sagittal and coronal planes under anesthesia using a 64-slice multidetector scanner (Optima 660, General Electric, Tokyo, Japan) with pre- and post-contrast administration of iodinated contrast medium (Omnipaque, GE Healthcare, Shanghai, China). CT images demonstrated a well demarcated, non-contrast, enhanced, tubular, oblong structure oriented in a caudomiddle direction from the caudal abdomen, extending to the pelvic canal and originating along the cranial vaginal level (W 4.1 cm. × L 6.0 cm. × H 3.0 cm.). The cavity content was non-contrast enhancing. This lesion was close to caudodorsal of the urinary bladder, left ureter and descending colon. A vaginal cyst was diagnosed based on the CT scan and surgical excision was performed to remove the vaginal cyst (Fig. 1).

The dog was anesthetized and positioned in dorsal recumbency. A third caudoventral midline laparotomy was made. A large ball-shape single tubular (7 × 5 cm)-cavitary mass was grossly found at the vagina attaching to the ventral border of the caudal descending colon, the dorsal part of the trigone and extending to adhere to most of the full length of the ventral aspect of the urethra into the pelvic canal. In addition, it was found that both ureters were adhered at the ventral border of the mass (Fig. 2). With gentle manipulation, the outer layer of the mass was noticeably adhered to the distal one-third of the left ureter. Other abdominal organs were examined and appeared to be normal. The bladder was retroflex out of the abdomen to approach the mass. The vaginal cystic mass was removed carefully from the associated organs using sharp and blunt dissection. Electrocautery and ligature were used to control the bleeding intraoperatively. First, the mass was dissected from the dorsal surface of the bladder trigone in the

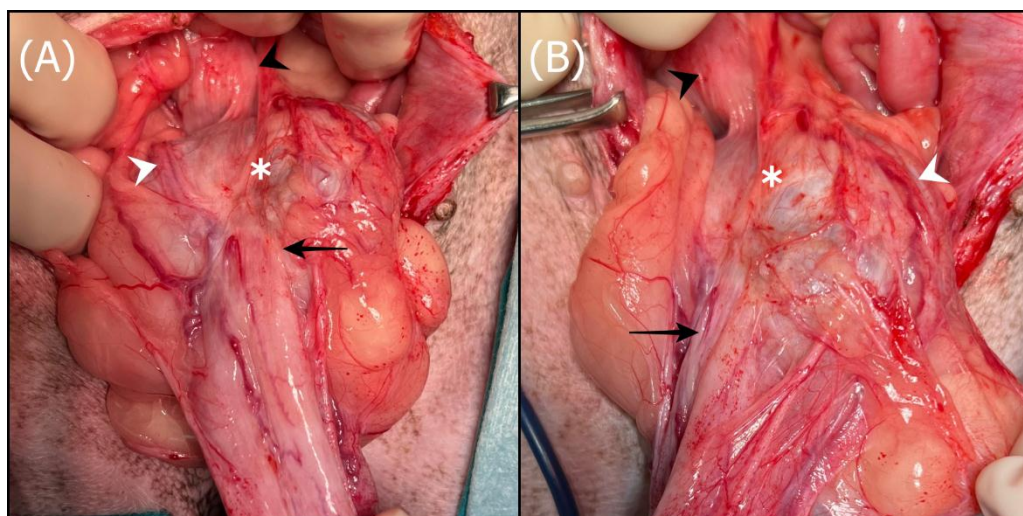
craniocaudal direction. The left ureter was separated from the adhering fibrous area on the cystic wall and was manipulated using a Penrose drain tube to preserve the vasculature and innervation. An 8-French balloon catheter was inserted from the vulva into the urethral opening to the bladder to establish the patency of the urethra during mass resection close to the intrapelvic urethra (Fig. 3A). Attentive dissection of the mass was provided to isolate it from the urethra along the ventral border of the mass. When the cystic mass was freed from the lower urinary tract, the mass was inverted to approach the dorsal surface which adhered to the caudal descending colonic serosa and the contact surface was dissected between them (Fig. 3B). The cystic mass was gently pulled cranially during the dissection along the caudal aspect of the mass to separate the mass from surrounding tissue. Ligation of the vaginal arteries was performed. Finally, the resection was made on the vagina caudal to the distal blind end of the sac and the mass was removed completely (Fig. 4). The remaining vaginal stump was closed using monofilament absorbable suture in a continuous appositional pattern and the abdomen was closed routinely.

The dog had a standard recovery from the surgery and was admitted postoperatively to the Critical Care Unit of the referral animal hospital. Defecation and urination were noted as normal and the dog was discharged one week after surgery. During 6 months of postoperative follow-up at the referral animal hospital, the dog's condition was most satisfactory. There were no other abnormal findings based on physical examination and abdominal ultrasonography. The dog was reported to have normal daily activity during the 4 months of follow-up based on telephone contact with the owner.

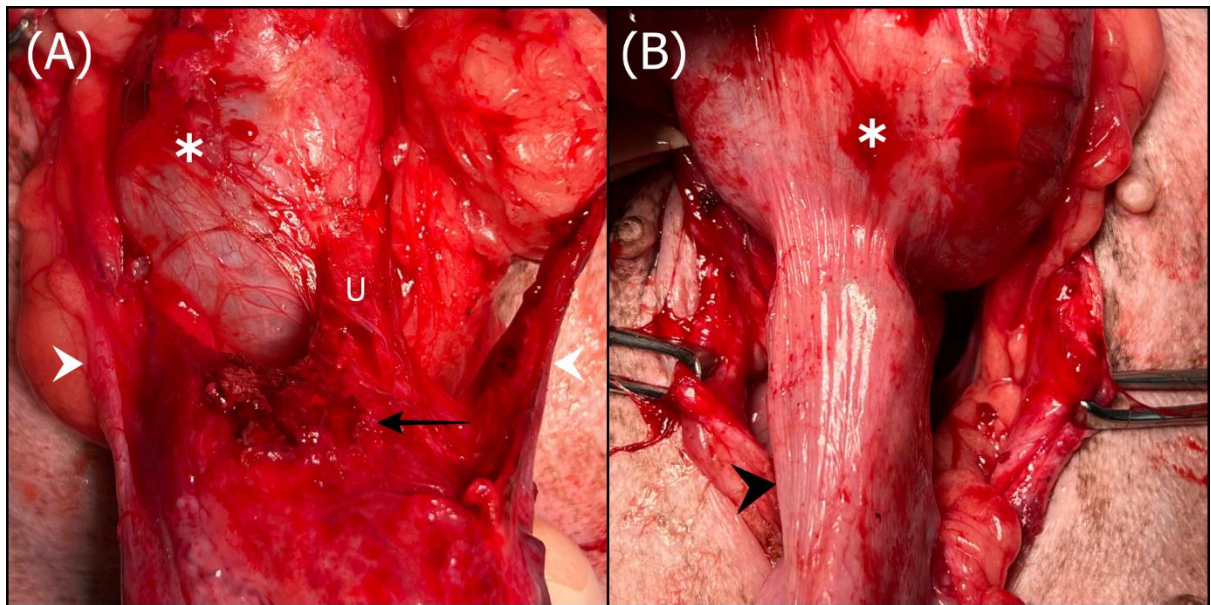
Histologically, the cyst was lined by simple or a stratified of cuboidal-to-low columnar epithelial cells, supported by fibrovascular tissue and an outer thin layer of smooth muscles (Fig. 5). The cells contained a secretory vacuole in the cytoplasm (Fig. 6). The secretion was considered as mucinous material based on Alcian blue staining (Fig. 7). A part of the lining epithelium was attenuated due to compression by the secretion in cystic lumen. Hemorrhage and the accumulation of hematogenous pigment, such as hemosiderin and hematoïdin pigment, were seen in the fibrous stroma. These histological findings were most consistent with the diagnosis of a Müllerian cyst. Additionally, the location and gross appearance also supported this diagnosis.



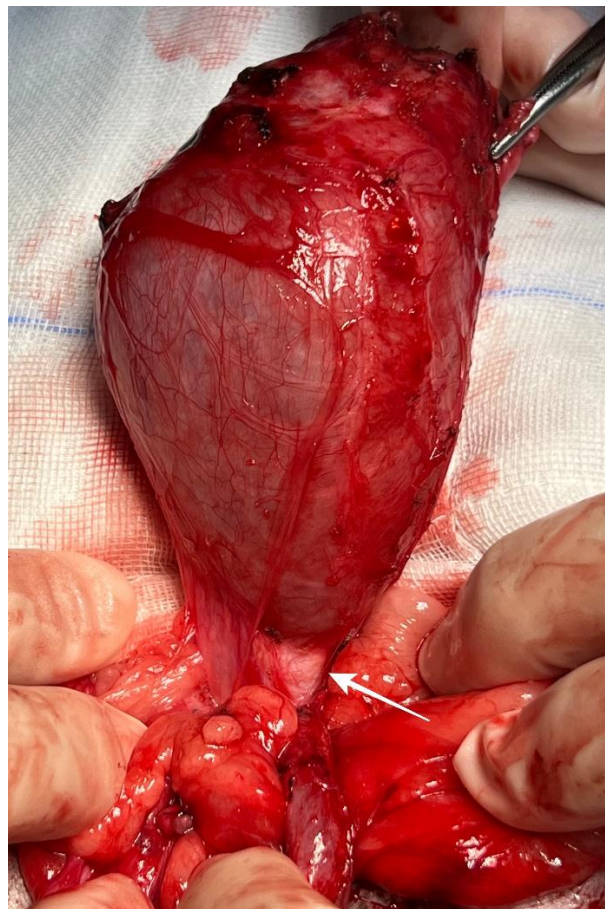
**Figure 1** Sagittal post contrast CT (A) and dorsal post contrast CT (B) of the caudal abdomen through the pelvis showing the tubular structure (arrows) anterior to the pelvic cavity, with the mass extending from the anterior pelvic cavity through the pelvic floor beneath the colon, with left ureter adhering to the outer wall of the tubular structure.



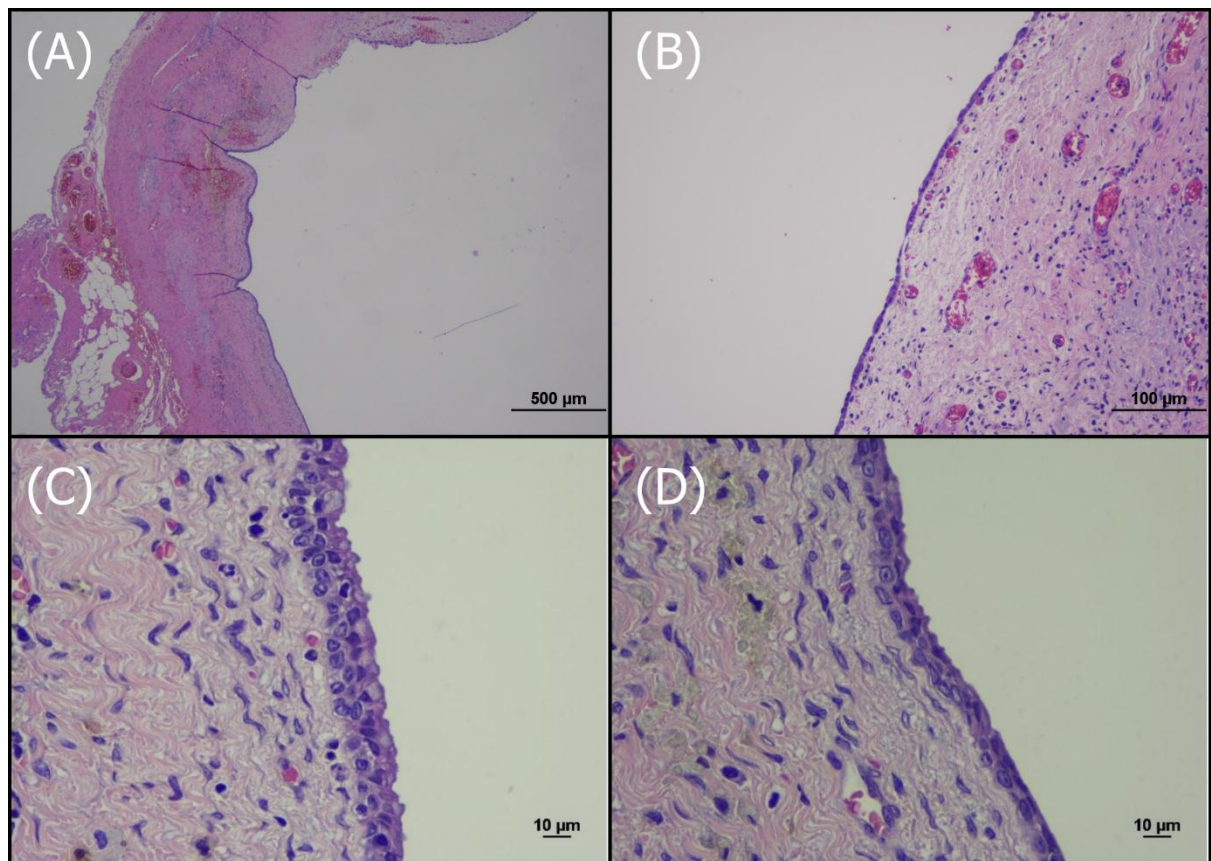
**Figure 2** The identification of large circular shaped vaginal cyst in the third surgery event at the referral animal hospital. The vaginal cyst (asterisk) is elevated and retracted to the left lateral side(A) and the right side (B). The ventral wall is attached to the right and left distal ureters (white arrowheads in A and B, respectively), the descending colon (black arrowhead) and the bladder neck (black arrow).



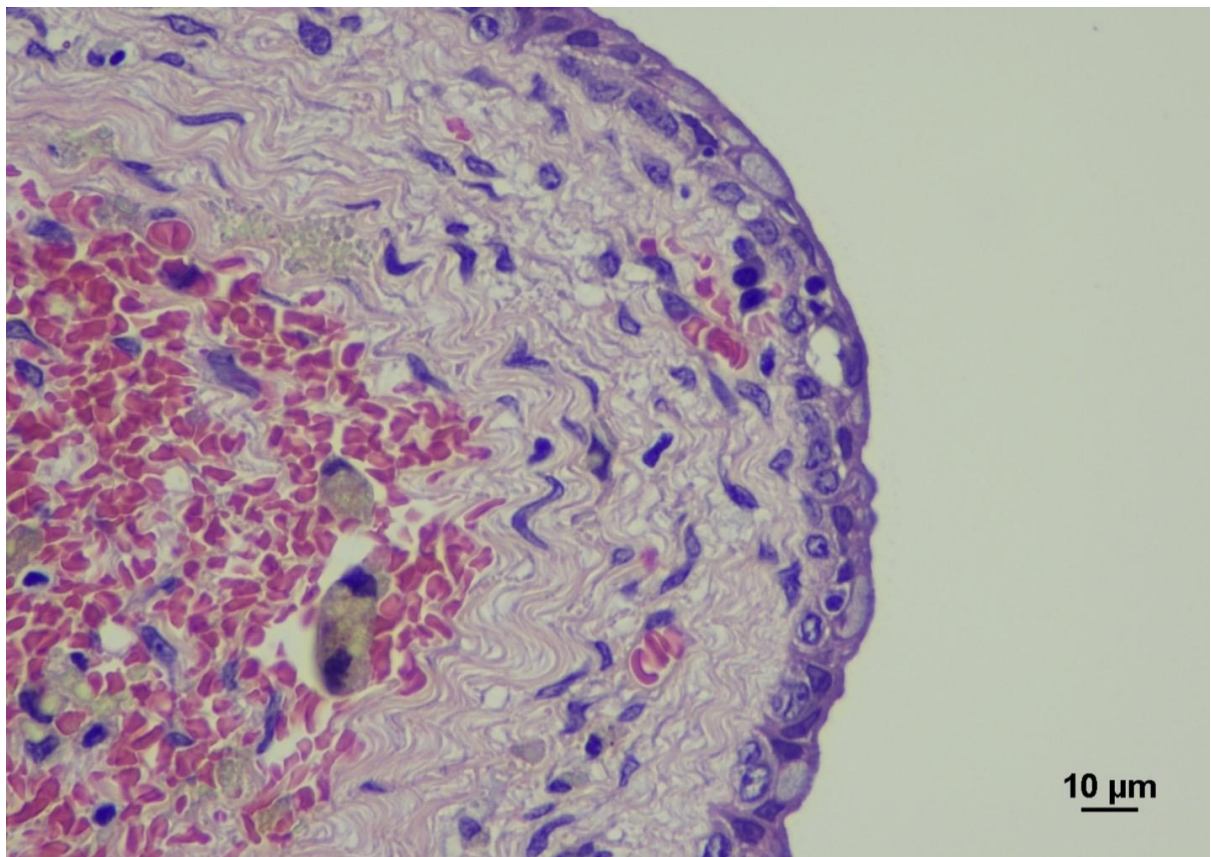
**Figure 3** (A) Dissection of vaginal cyst (asterisk) from the neck of the bladder (black arrow), with the caudal region of the cyst adhered to the urethra (U) and both ureters (white arrowheads) separated from the cyst. (B) Intra-operative photograph of the dorsal wall of vaginal cyst (asterisk) adhering to the descending colon (black arrowhead).



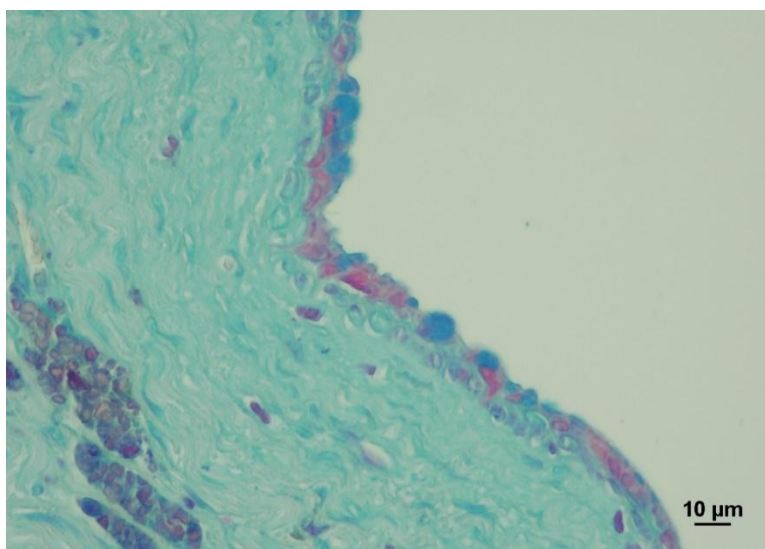
**Figure 4** Vaginal cyst was separated from the surrounding tissue and a complete cystectomy was performed on the vagina caudal to the distal blind end of the sac (white arrow).



**Figure 5** Histological findings of the vaginal cyst. Cystic wall is supported by fibrovascular tissue and a thin outer layer of smooth muscles, 40x (A). Cystic wall inner layer showing lining of thin epithelium and supported by collagenous tissue with numerous vessels, 200x (B). The epithelial lining showing a few layers of cuboidal or columnar cells, 600x (C). Attenuated cuboidal cells (D).



**Figure 6** Histological findings of the vaginal cyst. Few layers of cuboidal epithelial cells presenting secretory vacuoles in the cytoplasm, 600x.



**Figure 7** Histological findings of the vaginal cyst. Alcian blue staining demonstrating intracytoplasmic vacuoles containing mucinous material, 600x.

### Discussion

Due to the lack of information on the vaginal mass from the initial laparotomy from the first referring hospital, neither congenital nor an acquired vaginal cyst was excluded in the differential diagnosis list. The squamous inclusion cyst is the most common acquired type caused by the entrapment of the epithelium at the area of previous surgery or trauma (Eilber and Raz, 2003). An inclusion cyst has also been reported in a human neonate secondary to urogenital sinus development abnormality (Klein *et al.*, 1986). In contrast, the congenital type is caused by an embryonic duct remnant during fetal development. In the female fetus, the pair of paramesonephric ducts will fuse and develop to be the cranial portion of the vagina while the mesonephric ducts regress (Schmidt, 2003). The Müllerian epithelium is replaced by the squamous epithelium. The area which persists with the remnant tissue of the Müllerian epithelium will present as a mucinous cyst which is called a Müllerian cyst or paramesonephric cyst (Töz *et al.*, 2015). The Müllerian cyst may locate in any area of the vaginal wall, whereas the remnant of a mesonephric or Wolffian duct (Gartner duct cyst) is typically localized in the craniolateral vaginal wall (Heller, 2012; Eilber and Raz, 2003). This type was also considered in our case due to its finding in every report of a canine intra-abdominal vaginal cyst. A common histological finding with an inclusion cyst is squamous lining cells and keratinaceous debris. It is difficult to distinguish an inclusion cyst from a Müllerian cyst with squamous metaplasia. Furthermore, a Müllerian cyst may reveal a similar histological pattern to a Gartner duct cyst with cuboidal epithelium. Histochemistry is required to differentiate the mucinous productive lining cell due to the lack of mucin stain in the mesonephric duct remnant (Kondi-Pafiti *et al.*, 2008). There are various types of epithelium lining in a Müllerian cyst, including mucinous endocervial cuboidal epithelium (as was noted in our case), tubal epithelium, squamous epithelium or endometrioid epithelium (Heller, 2012;

Töz *et al.*, 2015). Moreover, the immunohistochemical staining is valuable for determining the cellular origin of the cyst. The epithelial lining will show the positive nuclear staining pattern for the markers such as estrogen receptor (ER), progesterone receptor (PR), Paired box gene (PAX 8) and Wilms tumor protein (WT1) in the Müllerian cyst as reported in human patients (Barona García and Ruiz Maciá, 2021; Fabien-Dupuis *et al.*, 2016; Takebayashi *et al.*, 2021). These expressions can support the tentative diagnosis. However, the molecular techniques were not performed in this case study. Additionally, other types of vaginal cyst and tumor should be listed as a differential diagnosis in this case. Thus, not only the histological appearance (for which there are some gray zones) and immunohistochemical findings of a vaginal cyst but also the location, duration, clinical signs and information of other abnormalities are also required for a definitive diagnosis (Heller, 2012). In our case, the Müllerian cyst was strongly suspected based on the histomorphology and histochemical features.

A vaginal cyst is asymptomatic for most human patients; however, treatment should not be postponed when the clinical signs occur in every patient (Heller, 2012; Töz *et al.*, 2015). A Müllerian cyst is benign but malignant transformation has also been reported in one human patient with an adenocarcinoma diagnosis result (Lee *et al.*, 2005). The case described in the current report was similar regarding the clinical findings reported for previous intra-abdominal cases. The clinical signs, including tenesmus, dyschezia and dysuria, were consistent with intracavitary fluid retention and progressive compression by a large cyst. This could cause a mechanical outlet obstruction and compromise normal defecation and urination.

The diagnostic imaging study in the referral animal hospital was affected by prior investigations by the veterinarians of the private hospital where the appearance and the intra-abdominal location of the mass had been identified. Abdominal ultrasonography was performed prior to the surgery at the private hospital based on mass identification and obtaining

fluid aspiration from the cyst for fluid sample evaluation and to drain the high voluminous cyst to relieve the clinical signs of suffering. The ultrasonographic result from both hospitals were consistent with other published reports, revealing a large anechogenic mass located in the caudodorsal area of the bladder trigone. Ultrasonography is a noninvasive and non-anesthetic modality without radiation exposure; in addition, it also provides initial details of intra-abdominal soft tissue in a variety of image planes without changing patient positions. Nonetheless, the caudal end of the cystic mass in this case extended into the pelvic canal which limited the ultrasonographic evaluation; hence, CT scanning with intravenous contrast was considered as following action. The CT scanning with retrograde vaginourethrography has been performed in reports involving a canine extra-abdominal vaginal cyst (Jimenez *et al.*, 2019). The current report is the first of the CT scanning for preoperative evaluation of an intra-abdominal vaginal cyst in a dog. During the second exploratory surgery, although the cranial part of the vaginal cyst had already been identified, the complications of this mass were lacking between the urinary and gastrointestinal tracts intra-pelvic region due to the potential risk of soft tissue trauma. Consequently, dissection and perioperative lesion examination was terminated just at the edge of the pelvic rim. Furthermore, post-surgical adhesion which was noted by direct visualization in the second surgery was likely to increase the severity and reduce the surgical exposure when the third surgery was performed. Intra-abdominal adhesion can cause pathological complications, such as bowel obstruction (Hassanabad *et al.*, 2021; Hellebrekers *et al.*, 2000) and our case showed clinical signs of dyschezia and dysuria for which the associated adhesion should be a concern. Intra-abdominal adhesions are rarely visible on CT scanning method (Siddorn and Mann, 1993); however, advanced imaging using CT is still required as part of the noninvasive surgical strategic plan for: the precise anatomical localization of a vaginal cyst, organ association, diagnosis of other etiologies that may help identify case symptoms and the detection of adhesion-related complications which provide benefits to preoperative planning and successful surgical procedures. In addition, CT provides the advantage of comprehensive screening of the entire abdomen in senile animal patients.

Surgical treatment is recommended in humans for all symptomatic vaginal cysts. Although various operative techniques have been utilized in treatment, in most cases, a cystectomy is performed as the treatment of choice, whereas marsupialization was the treatment applied with Bartholin duct cysts (Heller, 2012). One published report recommended the vaporization of the inner lining wall to prevent recurrence in an incomplete resection case (Töz *et al.*, 2015). In veterinary medicine, one case report described the excision of the entire cyst lining membrane, leaving the serous membrane intact to preserve the neurovascular adhesion areas. There was no recurrence found after this technique. However, transient incontinence occurred in the long-term postoperative period (Watanabe *et al.*, 2014).

Furthermore, marsupialization was advocated as a less invasive technique in one report (Holt, 1993) but recurrence was found in another case due to the opening having spontaneously closed, after which the author in that case recommended marsupialization in the large breed dog as the patient's anatomy allowed for good operating access to keep the patency of the fluid drainage. Moreover, a vaginal cyst located in the caudal part of the vagina is suitable to be treated with marsupialization (Watanabe *et al.*, 2014). Nonetheless, a cystectomy was still chosen as a surgical technique in one previous case of perineal swelling by extra-abdominal vaginal cyst and a curative outcome was achieved (Jimenez *et al.*, 2019). In the current report, it was not possible to prevent recurrence by undertaking drainage using ultrasound-guided aspiration prior to the surgery. The proximal half of the mass was resected and the fluid was drained during the second exploratory laparotomy. And the distal sac was curetted intra-cystic wall blindly without complete visual access to all part of the cyst. This technique was attempted to prevent the potential risk of neurovascular damage to associated internal organs. The purpose of the curettage was to eliminate secretory epithelial lining that produce the fluid. However, the postoperative recurrence still occurred. A possible explanation might be that this technique that was performed without the full exposure of the cystic mass could not eliminate all secretory cells. Another explanation was the access to the vaginal mass was limited in the cranial region that did not attach to any organs; thus, it was not possible to determine whether the lining membrane was removed completely. However, as part of the histological examination, it was determined that the lining membrane was still intact from the sample collected in the last surgery. These histological findings can explain the fluid production and recurrence of the vaginal cyst after the second surgery of intra-cystic lesion curettage. Additionally, a vaginal cyst is a benign lesion and so a surgical cystectomy may be too radical a treatment. Nonetheless, because the preoperative definitive histopathological examination at the caudal end could not determine the origin and the mass provided a similar presentation, it was not possible to exclude other malignant neoplasms in the differential diagnostic list of this region. Hence, complete excision as performed in this case, should be an election in the final surgery when possible. Thus, our decision to excise the vaginal cyst in this case was influenced by the recurrence after two previous less-invasive treatment procedures and the potential long-term incontinence associated with glandular cell lining membrane removal as was noted in the previous reported case by Watanabe *et al.* (2014). In addition, the CT scanning identified the appearance of the vaginal cystic mass extending from the caudal abdomen to the intra-pelvic area. A pubic symphysiotomy, pubic osteotomy or bilateral pubic and ischial osteotomies are recommend techniques used to improve the exposure access to the pelvic canal in small animal surgery (Baines and Aronson, 2018). Although these techniques were included in our preoperative plan and the owner was informed of the postoperative complications such as sacroiliac luxation, lameness,

neurologic dysfunction, nonunion of the fragment bones, sinus tract, avascular bone infection and sequestration (Baines and Aronson, 2018), neither of these procedures was performed during surgery, as traction of the cyst and dissection of the surrounding soft tissue provided the obligatory area to approach the distal end of the vaginal mass in this case study.

In human medicine, 70–90% of post-surgical abdominal adhesion is associated with previous surgery, with many cases reporting either direct or related disorder to this adhesion, resulting in increased rates of morbidity, mortality and the need for further treatments (Hassanabad *et al.*, 2021). The complications depend on the location and structure of any adhesion. Furthermore, an abdominal adhesion may increase perioperative risks, such as bleeding or perforation, limiting exposure, increasing postoperative complications and prolonging the surgical time during repeat surgeries (Hassanabad *et al.*, 2021; Hellebrekers *et al.*, 2000). Due the post-surgical adhesion of the cyst to the caudal internal organs and the thin appearance of the cystic wall based on the previous surgical curettage with the compression of large voluminous fluid, a complete cystectomy should be performed with caution and using delicate manipulation. In our case, the vaginal cyst was entirely dissected free from its attachment in the caudal abdominal cavity to the cranial pelvic region before completing the excision. This could not avoid completely the trauma of the serosal membrane of all associated organs; however, this did not result in adverse postoperative clinical effects. The vasculature was preserved as well as the innervation of the adhered surface. During 4 months of postoperative follow-up, no further defecation or urination problems were noted in the immediate or long-term postoperative period.

In conclusion, this was the first report of a canine intra-abdominal vaginal cyst that was preoperatively evaluated using CT and Müllerian cyst was strongly suspected in this case. A vaginal cyst should be investigated based on a differential diagnosis in both symptomatic and asymptomatic cases of cystic lesion of the vagina. Symptoms of patient, location and duration of vaginal cyst, imaging tools and histopathology are necessary for tentative diagnosis. Achieving a good outcome in the management of this condition in veterinary patients would have necessitated complete resection of the cystic mass using the images from the preoperative diagnoses to provide the necessary vision of location identification, which would prevent the re-surgery of residual cystic tissue and recurrent signs. Additionally, the surgical excision should be performed carefully with standard delicate skills to prevent the potential morbidity of the other organs.

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