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Rectal duplication in an adult cat: a novel transanal surgical approach

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Abstract

Case summary A 3-year and 8-month-old male entire European domestic shorthair cat was presented with a history of recurrent rectal prolapse, straining and pain when defaecating. Previous non-surgical and surgical treatments had not provided a satisfactory result. Rectal prolapse had recurred within 2 weeks of treatment. Upon clinical examination, an intraluminal mass could be palpated rectally. A CT scan examination revealed the mass was of a cystic nature and the cyst was surgically excised via a transanal approach. On histological evaluation, the cyst walls consisted of three of the layers of normal rectum: mucosa, muscularis of the mucosa and submucosa. These findings led to the definite diagnosis of rectal duplication.

Relevance and novel information Enteric duplication is among the differential diagnoses for straining and rectal prolapse in cats. This condition has previously been discussed in the veterinary literature, with a single case report describing a rectal duplication in a cat. In that particular case, the authors described a perineal surgical approach. Here we present a novel approach whereby the duplicated material was excised transanally in order to limit intra- and postoperative morbidity. The clinical outcome was excellent in our case, with complete resolution of clinical signs and no recurrence 18 months after surgery.

Keywords: Rectal duplication; rare condition; straining; transanal

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Introduction

Digestive tract duplication is a rare condition that has been reported in humans, in 10 dogs,^{1–9} and in eight cats.^{10–17} In humans, the reported incidence is approximately one in 4500 live births.¹⁸ In 1952, the human literature established that, to be classified as such, a duplication must comprise a contact at any point with the alimentary tract, and a smooth muscular coat and mucosa that should resemble that of the digestive tract at any of its levels.¹⁹ This definition implies that the final diagnosis of duplication is histological and not anatomical.²⁰

Duplications are located on the mesenteric side of the native structures and are often singular with tubular or cystic characteristics. They can arise at any level of the digestive tract. In humans, the most common duplication site is the ileum (30–35%), with the colon being the least commonly reported site (7–20%).²¹ These percentages are different in domestic animals, in which colonic

duplications appear to be the most frequent, accounting for up to 41% of reported cases.^{1–3,6–8,14} Digestive tract duplications have also been reported in the small intestines, the stomach and at the level of the oesophagus in the veterinary literature.^{4,5,9–13,16,17}

The exact embryological cause of digestive duplications is unknown; several theories have been suggested for the origin of this malformation.⁶ The ‘aberrant luminal recanalisation theory’ could explain duplications in portions of the gastrointestinal tract that go through a

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'solid stage', including the oesophagus, small bowel and colon, but could not explain duplication at other levels.²² The lumen of the gut is patent in the 6-week-old human embryo. Rapid proliferation of cells leads to the 'solid stage' of development (during which the lumen is not patent). Shortly thereafter 'vacuoles' appear within the solid lumen. In normal circumstances these vacuoles coalesce and the lumen returns to patency. Persistence of a vacuole (that would not coalesce with the others to make the lumen) can result in the development of a cystic or tubular duplication.²³ In humans, the 'split notochord theory' is the most accepted explanation for enteric-lined cysts located in the posterior mediastinum, abdomen or spinal canal, associated with adjacent vertebral anomalies.^{23,24} When it was first presented in 1944, the authors explained this theory by the possible duplication and separation of the notochord during embryo development. The level and the extent of the separation can be variable, leading to different malformations. This separation in the notochord would lead to a gap on the notochord through which the yolk sac or gut analogue endoderm could herniate and adhere to the dorsal ectoderm. This hernia would break and become a fistula, then differential growth of the embryo would tend to close this fistula; the success of this process would determine the extent of the residual lesion.²⁴

Diagnosis of digestive duplications usually takes place at a young age in humans, 85% of which are diagnosed before the age of 2 years.²⁵ Development of clinical signs during adult age have also been reported.^{21,26–28} Both in humans and in domestic animals, the most common clinical signs are related to the location of the duplication. Constipation, tenesmus and rectal prolapse are common signs of duplications of the lower digestive tract.^{1,7,8,14,15} When duplication occurs at the level of the oesophagus, stomach or small intestine, vomiting and anorexia are the most commonly reported clinical signs.^{3,5,9–14,16} Incidental findings of duplications have also been reported both in the human and veterinary literature.^{17,27}

Publications of digestive tract duplications in companion animals are scarce. However, among these publications, numerous concomitant malformations have been described. In one case of colonic duplication in a Poodle, a third pelvic limb was present and linked to the ischium (polymelia).² In two cases, vertebral column malformations were reported in association with lower digestive system duplications.^{6,14} One of the reported cases was a 9-week-old Labrador Retriever that had a colonic duplication and malformation of the fourth and fifth thoracic vertebrae.⁶ The second case was an adult cat with a rectal duplication that presented several misshaped and misaligned vertebrae.¹⁴

The most commonly performed treatment consists of surgical excision by laparotomy.²¹ Endoscopic treatment of colonic duplication cysts has recently been described

in humans, with positive outcomes.^{26,28} The prognosis for digestive duplications is generally excellent if complete resection of the duplication is achieved.^{21,26–29} However, incomplete resection of the duplication leads to recurrence of clinical signs, which has been documented in two cases in the veterinary literature.^{10,15}

The most commonly described surgical technique consists of laparotomy-based procedures: marginal mass resection (debulking), or through enterotomies or enterectomies (en bloc removal).^{1,8,13,15}

We present a new surgical approach, consisting of rectal cyst removal through a transanal approach. This approach has not been previously reported in the veterinary literature. In the only case of rectal duplication previously published in a cat, the authors had described a perineal surgical approach.¹⁴

Case description

History

A 3-year and 8-month-old domestic shorthair cat was referred to our hospital for investigation of recurrent rectal prolapse. Two surgeries had been previously performed by the referring veterinary surgeon to correct the prolapse. The previous procedures consisted of placement of a non-absorbable monofilament purse-string suture at the mucocutaneous junction of the anus, tied loosely to narrow the anal orifice to prevent mucosal prolapse, while still allowing soft faeces to pass through the orifice for 5 days. Recurrence of the prolapse occurred a few months after the first correction and a few weeks after the second one. The owners reported tenesmus, pain when defaecating, rectal prolapse and occasional rectal mucosal bleeding. The cat was on no medication at the time of presentation to our centre. The vaccination and worming status of the cat was up to date.

Clinical examination

On clinical examination the cat was bright, alert and responsive. Mucous membranes were pink and moist, and capillary refill time was >2s. Body condition score was 5/9. Cardiorespiratory auscultation did not reveal any abnormality. The rectal mucosa was prolapsed by 1 cm. Rectal digital examination revealed the presence of a rectal mass approximately 5 cm orally to the anus, as subjectively perceived by palpation. The rest of the clinical examination did not reveal any further abnormalities including absence of megacolon. Routine blood analysis, total blood cell count and biochemistry had been performed by the referring veterinarian and were within reference intervals.

Diagnostic imaging

General anaesthesia was performed. The cat was premedicated with 3.0 µg/kg of medetomidine (Narcostart; Ceva Santé Animale) and 0.2 mg/kg of methadone

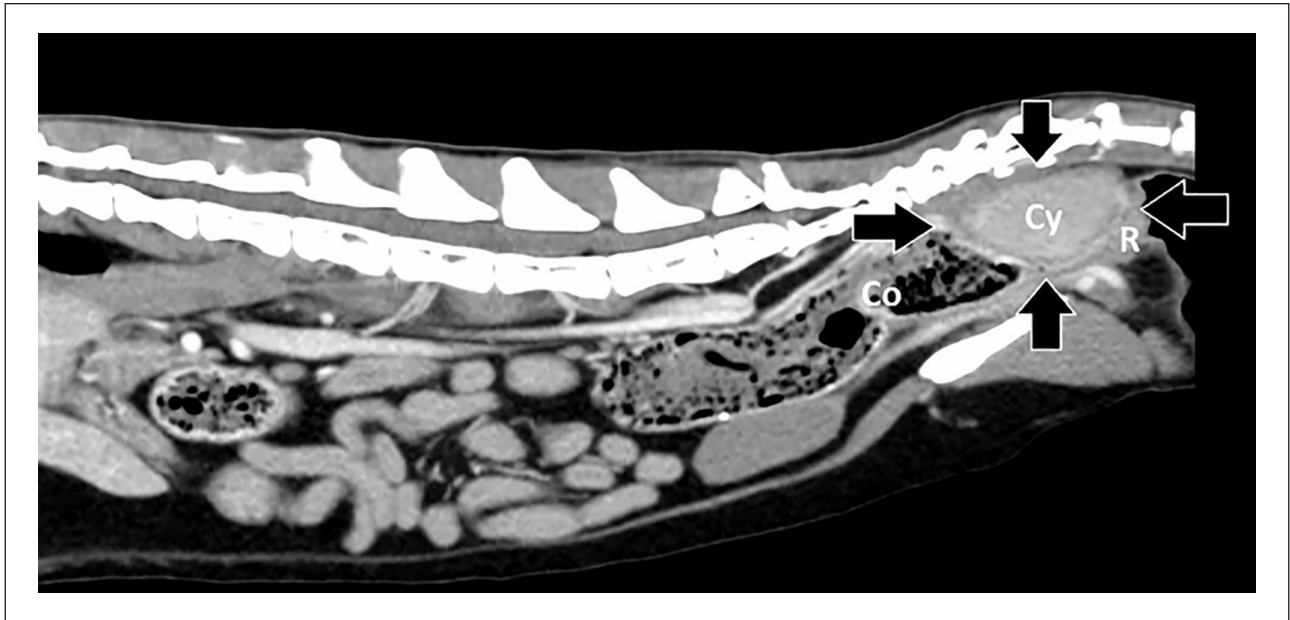


Figure 1 Sagittal CT image of the caudal abdomen and pelvis. The arrows mark the limits of the rectal cyst (Cy). Note the compression of the rectum (R) by the mass, and the presence of faeces in the colon (Co)

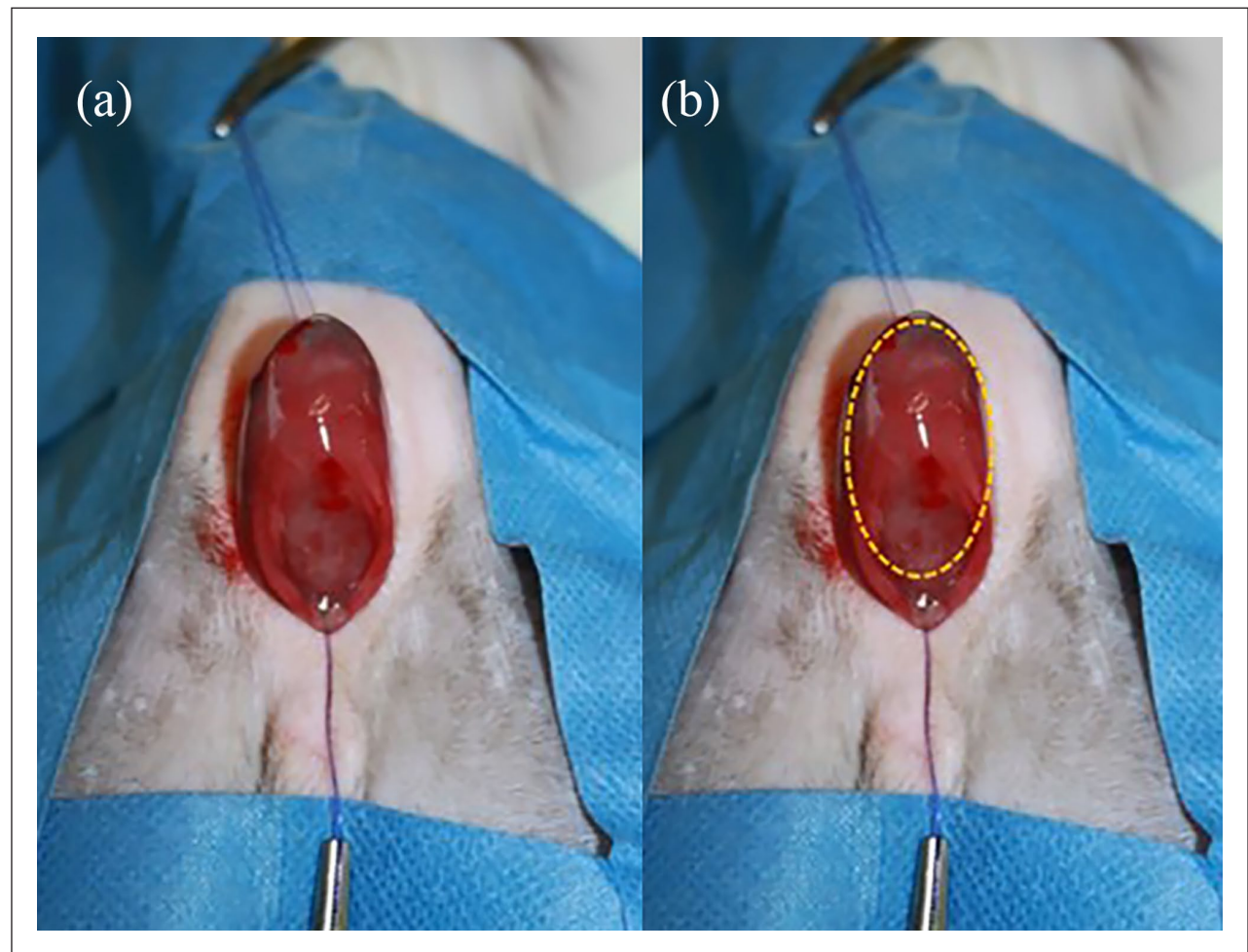


Figure 2 (a) Image of the transanally exteriorised cyst held in place by two stay sutures; one oral (bottom of the image) and one aboral to the cyst (top of the image). (b) Dotted line showing the incision made on the cyst wall



Figure 3 In this image the cyst wall has been removed and the content of the cyst is exposed (dark red mass)



Figure 4 Image showing the defect on the rectal mucosa left after the cyst wall and the cystic content were removed and before suturing with an absorbable monofilament. The finger is in the rectum, the oral margin of the surgical incision is held exteriorised by the stay suture (bottom of the image). The aboral margin is also held by a stay suture (top of the image)

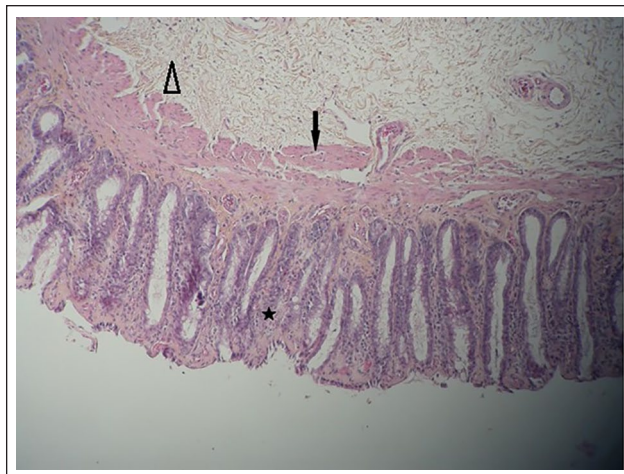


Figure 5 Histological section of the cystic wall. Three layers of normal rectum are present: mucosa (*), muscularis of the mucosa (black arrow) and submucosa (arrowhead)

(Comfortan; Dechra Veterinary Products SAS) were administered intravenously (IV). Five minutes thereafter, the cat was induced with Alfaxalone (Alfaxan; Dechra Veterinary Products SAS) IV to effect. A size 3.5 endotracheal tube was placed, and general anaesthesia was maintained with a mixture of 1.5% isoflurane in 1.5 l/min oxygen.

Abdominal CT images were obtained with the cat placed in sternal recumbency, using a 16-slice CT scanner (GE Brivo CT385; GE Medical Systems) with the following technical settings: 120 kV, 59 mA, slice thickness 0.625 mm, pitch 0.562, matrix of 512×512 pixels, a field of view of 14.5×14.5 cm. Reconstructions were performed using both a bone and a soft tissue algorithm. Contrast medium (Visipaque 320; GE Healthcare) was administered IV at a dose of 640 mgI/kg. Reconstruction with a soft tissue algorithm was used after contrast injection.

The acquired images revealed the presence of a homogeneous and hyper-attenuated pararectal cystic mass lesion in the pelvic canal, dorsal and slightly to the right of the rectum and in direct contact with it. It measured 1.5×1.8 cm and was markedly compressing the rectum on its dorsal aspect (Figure 1). The mass was well demarcated and did not appear to invade surrounding tissue. The filling of the cyst was well contained within the walls; no communication with the intestinal lumen was evident at this level. The size of the medial iliac lymph nodes was within normal limits. These diagnostic imaging findings were strongly suggestive of a rectal duplication. Malformations of the vertebral body of the seventh lumbar vertebra were noticeable: small-sized L7 vertebral body with a convex caudal end plate, and sacralisation of L7 was present with subsequent compression of the cauda equina. These additional findings reinforced

our suspicion of rectal duplication as they are often associated together, according to the existing literature.^{2,6,14}

A new rectal examination was performed during general anaesthesia and the cyst was easily exteriorised through the anus by rectal digital palpation and digital traction. Other working differential diagnoses at the time included benign rectal tumour, rectal diverticulum and rectal cyst of origin other than rectal duplication.

Surgical treatment

Based on the imaging and rectal palpation findings, a transanal approach was elected, as reported in the human literature.³⁰ A warm water enema was performed until only clear fluid was retrieved. The surgical area (perianal area, caudal aspect of the thighs down to the stifles and the entire circumference of the tail and its base) was clipped and aseptically prepared. The cat was placed in ventral recumbency with the pelvic limbs hanging off the edge of the table. The cat was positioned and draped in a way that would allow transformation to a perineal approach if required.

The cyst was easily exteriorised through the anus by rectal digital palpation and digital traction. The absence of communication between the cyst and the rectal lumen was confirmed at this point. Two stay sutures were placed orally and aborally to the mass through the mucosal and submucosal layers (Figure 2a). A circumferential incision of the cyst wall was made with a scalpel blade #11 (Figure 2b). The cyst wall was then removed. A full-thickness incision of the rectal wall was not required as the cyst was intraluminal. Only the cyst wall was incised. Once the cyst wall was removed, the content of the cyst was exposed (Figure 3). The semi-solid consistency of the content allowed en bloc removal by digital traction (seeding out the content as a whole). The two edges of the rectal mucosa (Figure 4) were eventually sutured together by direct apposition with an absorbable monofilament (Polydioxanone size 4/0) suture on a round needle using a simple continuous pattern. Permeability of the rectum was verified by digital rectal palpation. All removed tissues were placed in neutral-buffered 10% formalin solution and submitted for histological examination.

Postoperative period

Recovery from surgery was uneventful. The cat was discharged from hospital 48h after surgery with a 5-day course of meloxicam (Metacam; Boehringer Ingelheim). The use of antibiotics was not deemed necessary in this case, and postoperative antibiotics were not prescribed. Resolution of tenesmus was immediate. The cat passed normal faeces 72 h after surgery. Two weeks postoperatively the cat was re-presented to us for a re-check. Clinical examination was unremarkable. On digital rectal palpation, the patency of the rectum was verified. No evidence of rectal stenosis or mass recurrence was noted.

Eighteen months after surgery the cat was doing very well, according to the owners. No new episode of tenesmus, constipation or difficulty to defaecate were observed.

Histopathological analysis

The content was described histologically as an amorphous eosinophilic content, arranged in concentric lamellae. The cyst wall was similar to normal rectum as it presented a mucosa, muscularis of the mucosa and a submucosa (Figure 5). Histology therefore confirmed our previous suspicion of a rectal duplication cyst.

Discussion

The main presenting complaint in this case was tenesmus and recurrent rectal prolapse. Prolapse in cats usually occurs secondary to tenesmus from gastrointestinal (especially colorectal or anal) or urogenital disease. Predisposing factors for prolapse include: weakness of the perirectal and perianal supporting tissues; uncoordinated peristaltic contractions; excessive straining; inflammation and oedema of the rectal mucosa; gastrointestinal parasitism; typhlitis; colitis; proctitis; colonic duplication; congenital defects; tumours of the colon, rectum or anus; rectal foreign bodies; perineal hernia; anal laxity; cystitis; cystocele; prostatic disease; urolithiasis; dystocia; and anatomical or functional abnormalities, such as an abnormally large anus.

Digestive tract duplication is a rare condition that has been reported in humans, dogs¹⁻⁹ and cats.¹⁰⁻¹⁷ Small intestine³¹ and colonic duplications³² in the human literature have been described. However, to our knowledge no classification of rectal duplication has been published. Also in the human literature, colonic duplications have been classified into type I and type II.³² Partial duplications limited to the colon and rectum (without other abnormalities) are classified as type I. Type II are complete duplication, usually associated with other malformations. Five subtypes are described in type I duplications, mainly based on anatomical features. Type Ia are spherical and non-communicating with the lumen. Type Ib are tubular and non-communicating. Type Ic are tubular and communicating. Type Id are loop-shaped with a separate blood supply. Finally, type Ie corresponds to multiple duplications.²⁰ The duplication reported in the present case report would not fully fall under any of these groups, as the duplication was associated with L7 vertebral malformations (type II), although it was partial and spherical, as it did not include the whole rectum (type Ia). Lumbosacral malformations have also been described in humans with colonic duplications,²⁷ in a dog with a colonic duplication⁶ and a cat with a rectal duplication.¹⁴

Because enteric duplication is a rare condition, it is often misdiagnosed prior to referral. It is not unusual that

some treatments (surgical or non-surgical) have been performed, despite incorrect diagnosis prior to referral.^{1,6,8,15} In the human literature some asymptomatic duplications are described as incidental findings.³³ However, most of them are symptomatic at an early age.³⁴

Digestive system duplication remains a rare condition in companion animals, with only eight cases reported in cats prior to this.^{10–17} Only one case of rectal duplication has been reported in the feline species, with the cat being treated with a perineal excision of the duplicated material.¹⁴ In the present case report, we describe a surgical technique whereby the cyst was removed through a transanal approach instead, with an excellent outcome. This technique allowed us to limit morbidity of the surgery without the risk of complications associated with a perineal approach, such as rectocutaneous fistula formation or inadvertent pelvic plexus damage, for example. We understand that this approach is not applicable to all cases of rectal duplication, as the duplication must be located close enough to the anal margin to allow a transanal exposure without excessive traction on the rectal mucosa, and the cyst needs to be located intra-luminally. This technique has been described in human medicine in a case of rectal duplication cyst in an adult, with an excellent outcome.³⁰ Transanal endoscopic microsurgery (TEM) and laparoscopic intra-abdominal approaches have also been described. TEM allows for more cranially positioned rectal cysts to be transrectally excised.^{35,36} When the duplication can be digitally palpated and exteriorised transrectally, the technique described in the present report is relatively simple and fast to perform. A low morbidity is expected following this procedure, but with luminal obstruction (incorrect suturing or stenosis), rectal wall necrosis or rectal perforation as potential complications.

Conclusions

The present report describes a novel approach for excision of a rectal duplication cyst in a cat. Advanced imaging is essential in determining the characteristics and exact location of the cyst in the rectum. Location is of paramount importance; if the cyst is located too cranial or if the cyst is not intraluminal, the described technique might not provide a successful outcome and may increase complication risks compared with previously described techniques. Surgical removal of a rectal duplication cyst via a transanal approach provided an excellent outcome with no complications in our case.

Conflict of interest The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Ethical approval This work involved the use of non-experimental animals only (including owned or unowned

animals and data from prospective or retrospective studies). Established internationally recognised high standards ('best practice') of individual veterinary clinical patient care were followed. Ethical approval from a committee was therefore not necessarily required.

Informed consent Informed consent (either verbal or written) was obtained from the owner or legal custodian of all animal(s) described in this work (either experimental or non-experimental animals) for the procedure(s) undertaken (either prospective or retrospective studies). No animals or humans are identifiable within this publication, and therefore additional informed consent for publication was not required.

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