

CASE REPORT

Surgical management of a cervical oesophageal duplication cyst with tracheal communication in a dog

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Foregut duplication cysts can arise anywhere in the gastrointestinal system, leading to non-specific clinical signs in small animals. There are few reports of foregut duplication cysts in the literature that have been managed surgically. This report describes a case of multilevel foregut duplication cysts in a dog, including a cervical oesophageal duplication cyst with tracheal communication. Surgical treatment by resection resulted in the resolution of clinical signs. No clinical evidence of recurrence was noted at 12 months post-operatively. Surgical resection may be a feasible option for the management of foregut duplication cysts that cause clinical signs in dogs.

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INTRODUCTION

Foregut duplication cysts (FDCs) are rare congenital anomalies encompassing bronchogenic cysts (BCs), oesophageal duplication cysts (ODCs) and enteric duplication cysts (Kieran et al., 2010). They are a result of errors in embryological development, and multiple theories exist as to why they occur. During development, it is speculated that neural ectoderm and intestinal ectoderm form abnormal connections and/or the developing gut fails to canalise (Carachi & Azmy, 2002; Martin et al., 2007; Parikh & Singh, 2011; Spaulding et al., 1990). Due to the rarity of this anomaly, the prevalence is unknown in animals but has been reported in between one in 4500 and one in 10,000 humans (Phipps et al., 2021). In the human literature, FDCs are rarely reported and less so in the veterinary literature with only eight reports in cats and eight in dogs (Kim et al., 2023). Locations of FDCs reported in dogs include: two oesophageal, one gastric and five small intestinal duplication cysts summarised in Table S1 (Arthur et al., 2003; Asín et al., 2021; de Battisti et al., 2013; Foglia et al., 2018; Jack et al., 2016; Jakowski, 1977; Jung et al., 2009; Landon et al., 2007; Mutascio et al., 2019; Oui et al., 2014; Walling & Arndt, 2015). Due to often subclinical and non-specific clinical signs, the condition is likely under reported. Dogs with cervical ODCs can present with a flocculent cervical mass and signs associated with mass effect and compression on adjacent structures such as difficulty eating, stridor, cough and respiratory compromise. More severe complications

of ODCs have been reported in humans including haemorrhage and perforation (Arham et al., 2023; Foglia et al., 2018; Gabor & Walshaw, 2008; Nasr et al., 2015; Pinn et al., 2015; Sarkar et al., 2008; Snyder et al., 1996).

Multilevel FDCs have been previously reported in humans and one cat (Alsinan & Altokhais, 2024; Jang & Chung, 2021; Kershaw et al., 2008; Mandhan et al., 2014). The presence of a cystotracheal fistula associated with an ODC is unique to this case, based on the authors' review of previously published reports in the English canine and human literature. This was ensured via database search including Embase, Medline (PubMed) and a university library with the keywords duplication cyst, fistula and trachea communication on August 10, 2024, with cross reference to a textbook (Giuffrida & Brown, 2017). This report aims to present the diagnosis, treatment, histopathology and outcome of the dog.

CASE HISTORY

A 4-month-old male entire Newfoundland dog was presented to the hospital with the complaint of an 11-day history of regurgitation when eating, intermittent coughing and a slow-growing ventral cervical swelling. The dog was otherwise healthy with no known medical conditions or abnormalities. The swelling was initially treated with percutaneous drainage and amoxicillin clavulanic acid 200 mg/50 mg (Amoxycrav 250 mg; Apex

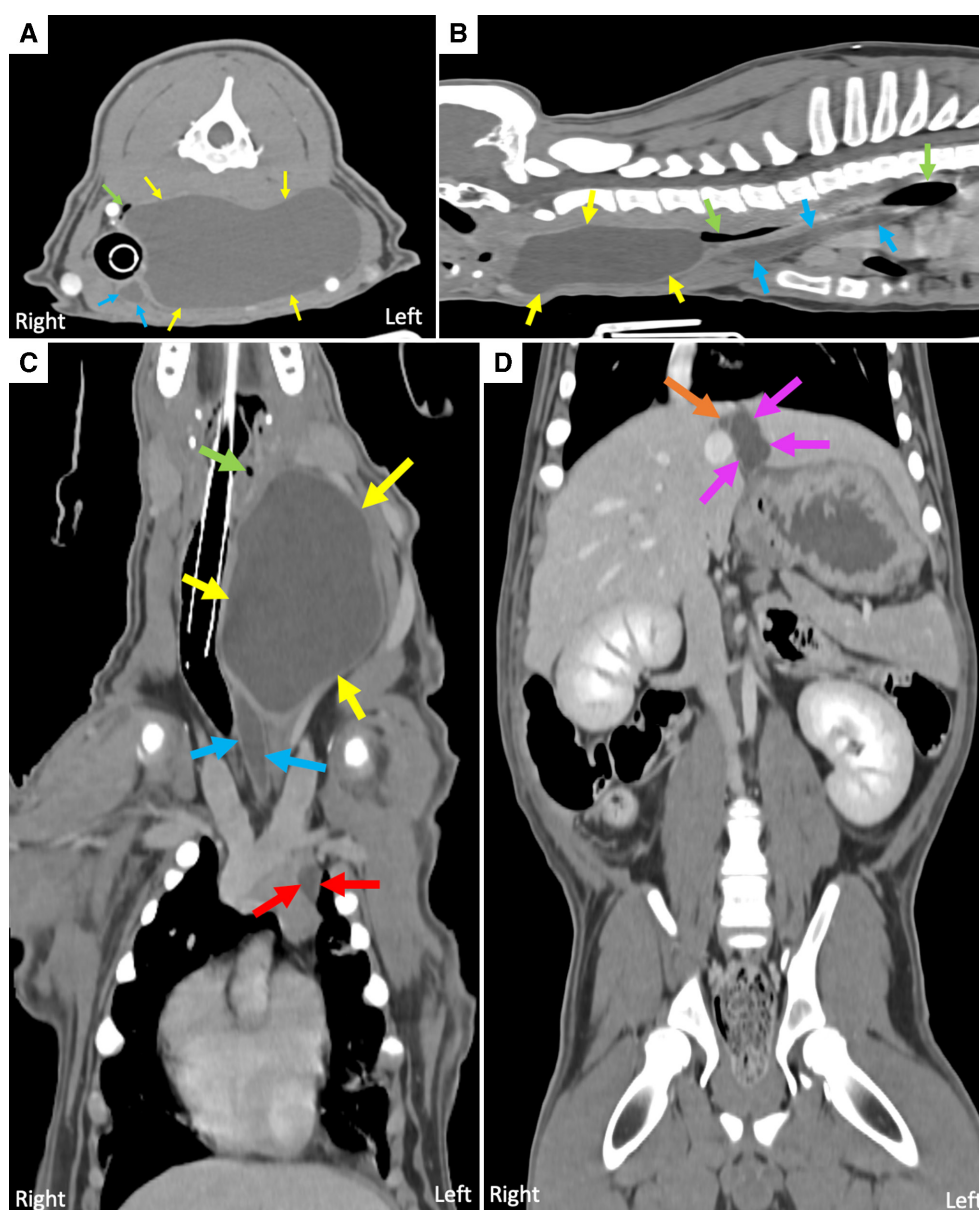
Laboratories Pty Ltd) at 13 mg/kg PO q12h, however, returned to its original size within 24 hours.

On physical examination, the dog had a 10 × 15 cm soft, non-painful mass on the left side of the ventral neck. Thoracic auscultation was unremarkable and no lymphadenomegaly was present. Biochemistry, haematology and blood gases were within normal range.

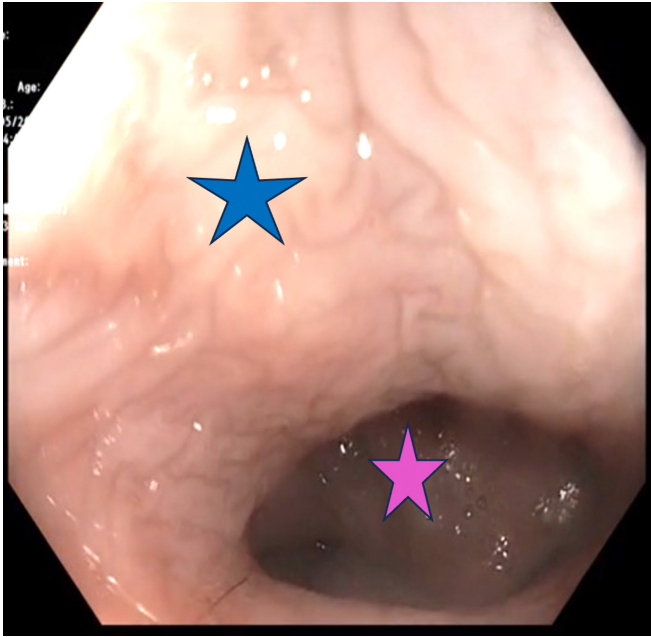
Radiographs showed a large, left-sided mass extending from the base of the skull to the thoracic inlet, displacing the trachea to the right. Ultrasonography was consistent with a fluid-filled mass and 500 mL of yellow, thick tenacious fluid was aspirated. Cytology revealed a light population of unremarkable squamous

epithelial cells, small numbers of neutrophils and small rafts of pleomorphic bacteria free in the background.

Dual-phase computer tomographic (CT) angiography [SOMATOM go.Up (64-slice); Siemens Healthcare] revealed five masses within the ventral neck, thorax and abdomen. All masses shared similar features as shown in Fig 1. The largest mass measured 12.5 × 5.1 × 9.2 cm diameter on CT and was displacing the trachea and larynx to the right, the cervical oesophagus dorsally and to the left, and the left thyroid lobe and left medial retropharyngeal lymph nodes cranially. The right common carotid artery was displaced laterally and ventrally to lie between the mass and the left sternocleidomastoid muscle. Video



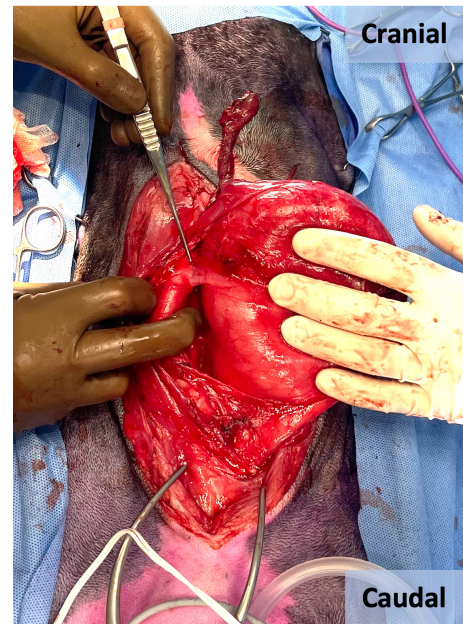
**FIG 1.** Computed tomography (CT) post-contrast delay phase pre-operatively. (A) Axial plane. (B) Sagittal plane. (C, D) Dorsal plane. Five foregut duplication cysts with similar characteristics: thin and smooth, soft tissue attenuating, contrast enhancing walls with a fluid attenuating, non-enhancing centre. Yellow arrows: first circular cyst extending from C1 to C5. Blue arrows: second cyst, tubular structure which communicates with the first through a thin duct and runs medial to the first through the length of the trachea, terminating at the level of the carina. Green arrows: oesophagus. Red arrows: third rounded cyst at the cranioventral mediastinum. Purple arrows: Fourth cyst, short tubular, at the ventral aspect of the gastroesophageal junction. Orange arrows: fifth cyst, small, circular, and possibly communicating via a small duct to the fourth cyst.



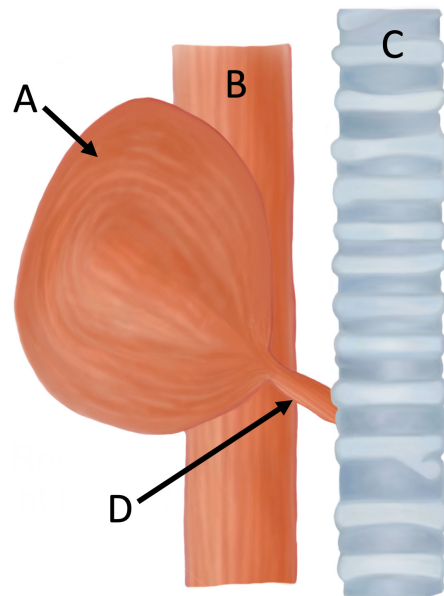
**FIG 2.** Still frame from pre-operative video endoscopy of the oesophagus. Blue star: oesophageal lumen. Pink star: partial luminal obstruction secondary to extraluminal compression by the cervical oesophageal duplication cyst.

esophagoscopy showed normal oesophageal mucosa with an extraluminal mass effect and resultant narrowing of the oesophageal lumen (Fig 2).

Surgical excision of the cervical mass was performed (Fig 3). Methadone hydrochloride (Ilium methadone; Troy Laboratories Pty Ltd) at 0.2mg/kg intramuscularly (im), dexmedetomidine hydrochloride (Dexdomitor; Zoetis Australia Pty Ltd) at 3 µg/kg im, propofol (Propofol Lipuro 1%; B Braun Australia Pty Ltd) at 5 mg/kg intravenously (iv) and cefazolin (Cefazolin-AFT; AFT Pharmaceuticals Pty Ltd) at 22mg/kg iv were administered for analgesia, pre-anaesthetic sedation, anaesthetic induction and antibiotic prophylaxis, respectively. Isoflurane (IsoFlo; Zoetis Australia Pty Ltd) and 100% oxygen via a circular system following endotracheal intubation were used to maintain anaesthesia. The dog was positioned in dorsal recumbency with an oesophageal stethoscope placed within the oesophageal lumen to aid in identification of the oesophagus. A standard ventral midline approach to the neck was made (Rosin, 1973). The mass was identified, and the fascial layers separated from the cyst using Lahey forceps and iris scissors. The left and right vagosympathetic trunks were isolated and retracted with vessel loops (Devon™ Silicone Vessel Loops; Cardinal Health Australia Pty Ltd) to prevent iatrogenic damage. A longitudinal partial thickness incision was made into the muscle layer of the oesophagus and the mass was isolated with blunt dissection. During exteriorisation of the mass, a separate communication was noted between the mass and the trachea. This was confirmed to be a fistula communicating with the cyst lumen by probing with a haemostat intra-operatively, illustrated in Fig 4. The communication was ligated with 2-0 polydioxanone (PDS; Johnson & Johnson International), transfixing and circumferential sutures. The excised mass was fixed in 10% buffered formalin. Excess oesophageal tissue



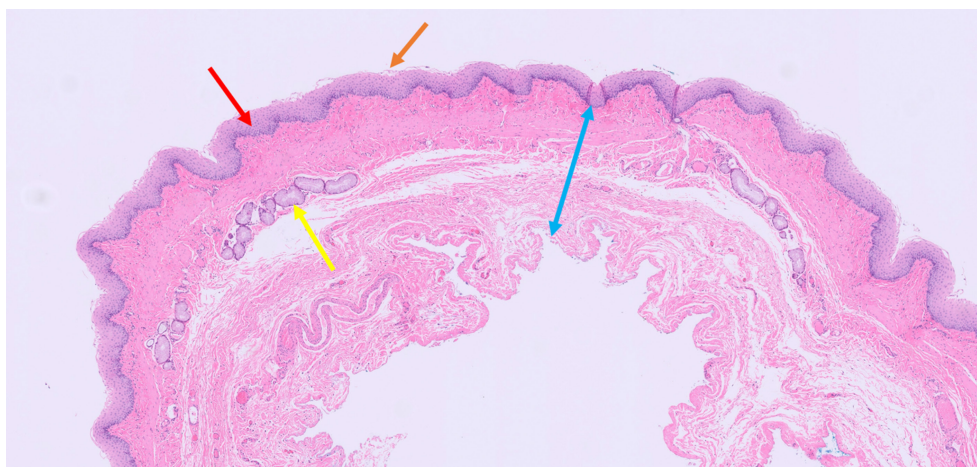
**FIG 3.** Intra-operative photo showing the oesophageal duplication cyst on the right. The trachea is being digitally retracted to show the communicating fistula identified by DeBakey forceps.



**FIG 4.** Illustration of the oesophageal duplication cyst (A), oesophagus (B), trachea (C) and cystotracheal fistula (D). The cyst was contained within the oesophageal wall not communicating with the oesophageal lumen. A cystotracheal fistula was present between the cyst lumen and tracheal lumen. There was no communication between the oesophageal lumen or tracheal lumen.

was resected, and surgical closure was as previously described using 2-0 polydioxanone (PDS; Johnson & Johnson International) in a simple continuous pattern (Rosin, 1973). A Jackson Pratt drain (Jackson-Pratt® Flat Drain 7mm; Cardinal Health Australia Pty Ltd) was placed in the resultant dead space after excision of the cyst and secured with a self-locking suture pattern with 2-0 polyamide monofilament (Dafilon; B Braun Australia Pty Ltd).





**FIG 5.** Haematoxylin and eosin preparation histomicrograph (5× magnification) of the cyst shows non-keratinising stratified squamous epithelium (red arrow) with underlying bundles of smooth muscle (blue arrow) and mucous glands (yellow arrow), with external loose fibrocollagenous adventitia (orange arrow), consistent with an oesophageal duplication cyst. The lack of skeletal muscle on this section reflects the location in the caudal oesophagus.

Histopathological examination of the cyst revealed stratifying squamous mucosa underlying mature fibrocollagenous connective tissue and rare bundles of smooth muscle with rare to infrequent aggregates of submucosal oesophageal glands, thus fulfilling the histologic criterion of an ODC (Kieran et al., 2010). The cystotracheal fistula showed a similar lining; however, squamous mucosa was interspersed with tall columnar pseudostratified ciliated epithelium consistent with respiratory mucosa (Fig 5).

Aerobic and anaerobic cultures were negative after extended incubation with only a few leukocytes observed on Gram stain. Recovery was uneventful and the dog was discharged 4 days following surgery. In the acute post-operative period, opioid analgesia was adjusted to achieve a modified Glasgow composite pain score of four of 24 or lower by a continuous rate infusion of fentanyl citrate (Fentanyl GH; Generic Health Pty Ltd) at 1 to 5 µg/kg/hour. This was discontinued 12 hours post-operatively. Additional medications included: meloxicam (Metacam; Boehringer Ingelheim) at 0.1 mg/kg PO q24h for 5 days, gabapentin (Neurontin; Pfizer) at 10 mg/kg PO q8h for 5 days, cephalexin (Rilexine; Virbac Australia Pty Ltd) at 25 mg/kg PO q12 hours for 14 days. The drain was removed prior to discharge when production was below 0.2 mL/kg/hour over 24 hours.

Minor complications included regurgitation after eating, and the development of a large seroma reported by the owners 7 days post-operatively. Esophagoscopy was repeated at this time and ruled out both oesophageal stricture and cyst recurrence. Clinical signs resolved with placement of a closed-suction drain within the seroma and 6 days of omeprazole (Losec; Pharmaco Australia Ltd) at 0.67 mg/kg PO BID to prevent the development of oesophageal ulceration. The drain (Jackson-Pratt® Flat Drain 7 mm; Cardinal Health Australia Pty Ltd) was placed within the seroma and secured with a self-locking suture pattern with 2-0 polyamide monofilament (Dafilon; B Braun Australia Pty Ltd). The drain was removed after 7 days once output reduced to 0.2 mL/kg/hour over 24 hours.

One year post-operatively, the dog did not have external evidence of recurrence of the excised ODC based on physical

examination or clinical signs. Prior to surgery, the dog regurgitated consistently after eating which has reduced to regurgitation only with rapid, large volume eating. The owners declined repeat imaging at 1 year follow-up and were pleased with the post-operative outcome.

## DISCUSSION

The present report describes a case of multilevel FDCs in a dog with a cervical ODC which was found intra-operatively to have a cystotracheal fistula. In the dog, FDCs have been reported in all levels of the gastrointestinal tract (GIT), most commonly the colon (Arthur et al., 2003; Asín et al., 2021; de Battisti et al., 2013; Gabor & Walshaw, 2008; Jack et al., 2016; Jakowski, 1977; Jung et al., 2009; Landon et al., 2007; Mutascio et al., 2019; Oui et al., 2014; Walling & Arndt, 2015). Similar to a previous report of a cervical ODC in a dog, the current case describes a fluctuant ventral neck mass as the presenting complaint which was subsequently excised with the dog making an uneventful recovery (Gabor & Walshaw, 2008).

Clinical signs of FDCs are non-specific and relate to their size and location, therefore diagnosis requires a combination of advanced imaging and histopathology. In this dog, histopathological evaluation fulfilled the criteria of a FDC which includes: (1) a smooth muscle covering, (2) epithelium derived from the foregut, and (3) attachment to a portion of the foregut. Like ODCs, BCs can be associated with the oesophagus but are distinguished by the lack of hyaline cartilage (Kieran et al., 2010). The authors speculate that a cystotracheal fistula formed as a congenital structure rather than a complication of the cyst. Although communications with the respiratory tract have been reported in cases of ODCs in the human literature, this is limited to congenital fistulas not associated with the cyst and spontaneous fistula formation resulting from complications of the cyst (Arham et al., 2023; Nasr et al., 2015; Pinn et al., 2015; Sarkar et al., 2008; Snyder et al., 1996; Sundaramoorthi et al., 2000).

In contrast to tracheoesophageal fistulas, the ODC in this dog did not communicate with the oesophagus, therefore, although cystotracheal communication was present, there was no communication between the trachea and oesophagus (Fig 4).

Advanced imaging facilitates surgical planning and screening for concurrent congenital abnormalities (Agarwal & Bagdi, 2011; Carachi & Azmy, 2002; Gupta et al., 2010). Congenital anomalies of the urinary tract, genitalia and lower spine have been reported concomitantly with FDCs in humans, while in dogs one case has been reported with concurrent vertebral malformation (Jakowski, 1977; Ricciardolo et al., 2019). Radiography, ultrasonography, CT and MRI can be utilised, showing a fluid filled lesion and often mass effect (Agut et al., 2018; Arthur et al., 2003; Bernardé et al., 2014; Blank et al., 2012; de Battisti et al., 2013; Foglia et al., 2018; Gabor & Walshaw, 2008; Hur et al., 2007; Jung et al., 2009; Landon et al., 2007; Martin et al., 2007; Mutascio et al., 2019; Oui et al., 2014; Phipps et al., 2021; Shinozaki et al., 2000). In the present case, CT imaging identified four other FDCs throughout the GIT which were incidental findings; however, the cystotracheal fistula was not discovered until the time of surgical excision, despite pre-operative complimentary ultrasonography and esophagoscopy. This is not surprising given that CT and tracheobronchoscopy are reported to have low sensitivity for the diagnosis of tracheoesophageal fistulas in dogs (Kaminen et al., 2014; Stogdale et al., 1977).

In humans, surgical management of FDCs is considered urgent due to potential GIT obstruction, perforation, haemorrhage and malignant transformation (Arbona et al., 1984; Schalamon et al., 2000). These sequelae have not been observed in dogs, potentially due to their shorter lifespan. Total mucosectomy, stenting for enteric diversion of cystic contents and resection of the shared wall between the enteric lumen and the cyst have been described in humans (Gonzalez-Urquijo et al., 2022). Therapeutic interventions for this dog were based on addressing clinical signs resulting from the cervical ODC. Although the presence of a cystotracheal fistula was only identified intra-operatively, surgical excision may have prevented additional complications associated with the fistula. These complications might include ascending bacterial migration with subsequent infection of the cyst, and bacterial pneumonia. The human literature reports similar complications in cases with spontaneous fistulisation (Arham et al., 2023; Sundaramoorthi et al., 2000). Excision of the remaining cysts by thoracotomy and celiotomy in either a single session or staged procedure was recommended, however declined by the owners due to financial constraints and potential risk of complications.

Prognosis after surgical removal of FDCs is excellent (Cavar et al., 2006; Spaulding et al., 1990). Complete removal is necessary to prevent continued fluid production and recurrence which has been reported twice in felines after subtotal excision (Bernardé et al., 2014; Kramer et al., 2007). Complete excision was achieved in this dog based on histopathology results, cessation of clinical signs and no clinical evidence of recurrence at 12-month follow-up. Serial follow-up examinations have been recommended to monitor for recurrence and development of clinical signs in association with the remaining cysts.

One limitation of this report is the lack of additional advanced imaging during the long-term follow-up period. Due to the non-specific and sometimes subclinical impacts of FDCs, recurrence may not be detectable on clinical examination alone and therefore cannot be reliably excluded in this dog.

Although rare, FDCs should be a differential diagnosis for masses associated with the GIT. The pursuit of surgical intervention in dogs can be based on the development of clinical signs; however, surgery is the only treatment reported to resolve clinical signs associated with the cyst. Monitoring can be elected for FDCs which do not impact quality of life; however, clinicians should be aware of life-threatening complications reported in other species including haemorrhage, fistulisation, rupture, obstruction, and neoplastic transformation.

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### Author contributions

**S. Costello:** Data curation (equal); investigation (equal); writing – original draft (equal); writing – review and editing (equal). **L. Woolford:** Data curation (equal); resources (equal); validation (equal); writing – original draft (supporting); writing – review and editing (equal). **R. M. Basa:** Conceptualization (equal); data curation (equal); resources (equal); supervision (equal); writing – review and editing (equal).

### Conflict of interest

None of the authors of this article has a financial or personal relationship with other people or organisations that could inappropriately influence or bias the content of the paper.

### Data availability statement

The data that support the findings of this study are available from the corresponding author upon the reasonable request.

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## Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

**Table S1.** Summary of published literature on canine enteric duplication cysts.