

## Surgical treatment of a thoracic oesophageal duplication cyst causing recurrent dysphagia in an adult dog

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**ABSTRACT:** A 7-year-old intact male Rottweiler dog was evaluated for recurrent dysphagia and regurgitation. Physical examination was unremarkable and routine blood works were within normal limits. Computed tomography revealed a defined lesion in the caudal mediastinum arising from the oesophagus. The lesion was excised using intercostal thoracotomy and the histological diagnosis was oesophageal duplication cyst. The dog recovered uneventfully and at a 3-year follow-up no clinical signs were reported. Although extremely rare, oesophageal duplication cysts should be considered in the differential diagnosis in cases of chronic regurgitation and dysphagia associated with evidence of an oesophageal lesion.

**Keywords:** oesophagus; thoracic surgery; foregut anomaly

Oesophageal duplication cysts (ODCs) are congenital embryonic malformations which can manifest as spherical or tubular lesions anywhere along the oesophagus (Carachi and Azmy 2002). They are considered very rare in humans and are infrequently reported in domestic animals (Gabor and Walshaw 2008; Obasi et al. 2011).

Of the foregut duplications described in humans, 75% are located within the abdominal cavity, 20% are completely intra-thoracic and 5% have a thoraco-abdominal location. Rarely, single cases of duplication cysts originating from the tongue, floor of the mouth and hypopharynx have been reported (Sundaramoorthi et al. 2000; Espeso et al. 2007; Obasi et al. 2011). Multiple enteric duplication cysts were recently reported in a human infant (Udiya et al. 2016). To the best of our knowledge, only single lesions have been reported in the dog, in the foregut and in the urinary tract (Oui et al. 2014; Walling and Arndt 2015; Jack et al. 2016; Mutascio et al. 2017; Thierry et al. 2017).

The cysts are often asymptomatic, but in some cases can result in clinical signs. The location and the size of the cysts usually determine the time of presentation and the associated symptoms

(Sundaramoorthi et al. 2000). Complete surgical excision is the treatment of choice in human medicine (Jan et al. 2016).

Only one case of a canine oesophageal duplication cyst located in the cranial cervical tract has been reported in the literature. Moreover, this condition has been recorded in a cat (Doran et al. 2015), in five horses and a cynomolgus monkey (Gabor and Walshaw 2008).

The present paper describes the clinical signs, the diagnostic work up and the surgical treatment of a thoracic oesophageal duplication cyst in a dog. To our knowledge, the treatment of an ODC in the thoracic caudal oesophagus has not been previously reported in the dog.

### Case description

A 7-year-old, intact male Rottweiler dog was presented to the Veterinary Teaching Hospital of Bologna University, with a history of worsening dysphagia and regurgitation from the first year of life. Physical examination was unremarkable and no abnormalities were observed in haematological or

serum biochemistry exams. Thoracic radiographs showed an increased soft tissue opacity in the caudal portion of the thorax (Figure 1).

A further endoscopic examination of the upper gastrointestinal tract showed an extraluminal bulging mass in the right side of the lower third of the oesophagus, without involvement of the overlying mucosa (Figure 2). Distally, the lower oesophageal sphincter appeared opened and multiple radial erythematous lesions suggested reflux esophagitis.

A computed tomography was performed and a well-circumscribed, focal lesion, with a thin wall and a homogenous inner density, was seen in the caudal mediastinum. The lesion arose from the wall of the distal third of the oesophagus, on the right side, and the mean central density was 3.3 Hounsfield units, compatible with a fluid-filled mass, or a low-density solid one (Figure 3).

With a suspicion of either oesophageal cancer, abscess or congenital duplication cyst, exploratory surgery by lateral thoracotomy was scheduled.

After premedication with acetylpromazine (0.02 mg/kg *i.m.*) and methadone (0.2 mg/kg *i.m.*), general anaesthesia was begun with administration of propofol (3 mg/kg *i.v.*), followed by intubation. General anaesthesia was maintained with isoflurane in oxygen and a constant rate infusion of fentanyl (0.003–0.0012 mg/kg *i.v.* as needed) was given for intraoperative analgesia. Lactate Ringer's solution (10 ml/kg/h) was administered. The patient

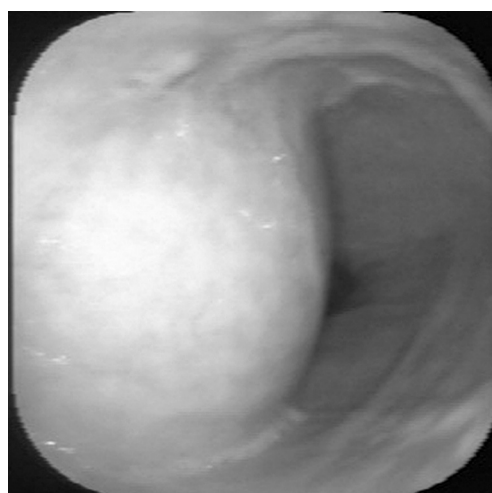


Figure 2. Digital image from endoscopy of the distal part of the oesophagus showing an extra-luminal compression. The oesophageal mucosa appeared intact

was ventilated with intermittent positive pressure at a rate of 12 breaths per minute and continuous monitoring included electrocardiography, pulse oximetry, capnography, indirect blood pressure and recording of temperature. Ampicillin/sulbactam (20 mg/kg *i.v.*) was administered at induction and subsequently every 2 h during surgery.

With the patient in left lateral recumbency, the right side of the thorax was clipped and aseptically prepared and an eighth right intercostal thoracotomy was performed.

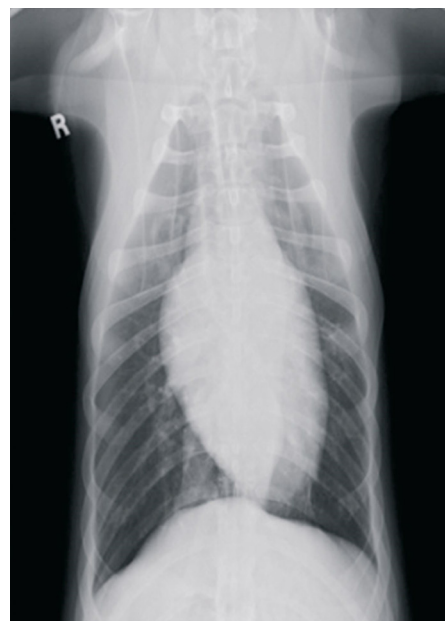


Figure 1. Radiographic aspect of the lesion. Thoracic radiographs show an increased soft tissue opacity in the caudal portion of the thorax

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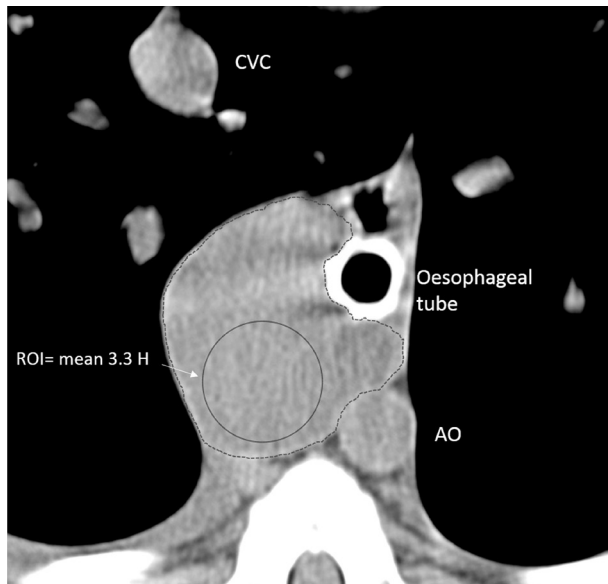


Figure 3. Thoracic CT scan showing a well-circumscribed, round lesion in the caudal mediastinum, which arose from the wall of the distal third of the oesophagus, on the right side. The mass has a thin wall and a homogeneous inner density, and it was compatible with a fluid-filled or well-differentiated solid mass. Transverse view AO = aorta; CVC = caudal vena cava; H = Hounsfield; ROI = region of interest; dashed line = limit of the oesophageal lesion

A 5-cm diameter, well-circumscribed, hollow mass lesion, was identified on the right side of the distal third of the oesophagus. It was fluctuating on palpation and could be felt protruding into the oesophageal lumen with no evidence of obstruction. The mass appeared to be a mural lesion and it seemed to be consistent with a cyst. The mediastinal pleura, oesophageal adventitia and, partially, the muscularis over the cyst were linearly dissected (Figure 4). Electrocautery and blunt dissection were used to remove the suspected cyst after a plane of cleavage was identified. The cyst was separated from oesophageal tissue preserving the muscle layers and vagal nerves and without penetrating the oesophageal mucosa. The incised oesophageal muscle layer was approximated and sutured using one-layer appositional interrupted pattern with 3-0 polydioxanone, engaging the submucosa. A thoracostomy tube connected to a closed continuous suction system was placed at the level of tenth intercostal space and the thoracotomy was routinely closed.

Postoperatively, no complications occurred and omeprazole (1 mg/kg *i.v.* every 24 h) and ranitidine (1 mg/kg *i.v.* every 12 h) were administered as ant-

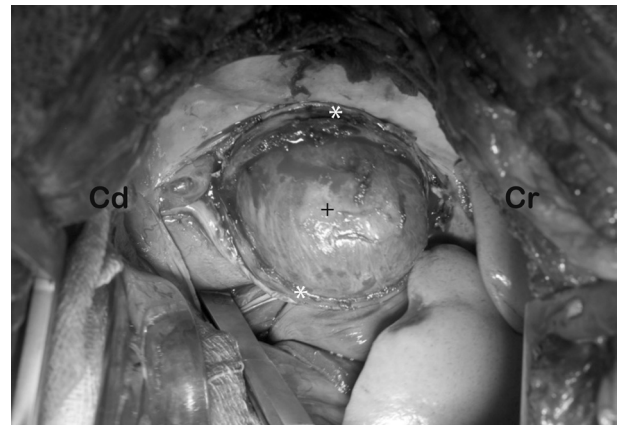


Figure 4. Intraoperative aspect of the cyst after longitudinal incision of the oesophagus. The mediastinal pleura, oesophageal adventitia and partially the muscularis over the cyst were linearly dissected. The cyst was removed by blunt dissection and electrocauterisation, preserving the vagal nerves without penetrating the oesophageal mucosa

+ = outer surface of the duplication cyst; \* = oesophageal adventitia and muscularis; Cd = caudal; Cr = cranial

acids and prokinetic drugs. Sucralfate (1 g orally every 8 h) was administered to treat oesophagitis. A fentanyl CRI (0.002 mg/kg/h *i.v.*) was continued for 12 hours postoperatively for pain management; subsequently, the patient was transitioned to methadone (0.1 mg/kg *i.m.* every 4 h). The dog recovered uneventfully and from one day postoperatively began eating a gastrointestinal canned diet for three weeks. The thoracic drainage was removed on the second day after surgery and the dog was discharged without complications three days postoperatively.

Upon gross examination, the lesion consisted of a 5 × 3 cm unilocular, ovoid cystic mass with a smooth and pink-to-red outer surface. A cut section revealed that the cyst contained abundant, viscous and yellow material which was partly adherent to the cystic wall (Figure 5).

Tissue samples were processed routinely and sections were stained with haematoxylin and eosin. On microscopic examination, the cyst appeared to be lined with cuboidal non-ciliated pseudostratified epithelium and abundant submucosal mucous glands were present. The wall was composed of a double muscle layer, with the outer one being striated. No cartilage or bronchial glands were identified (Figure 6). Based on these findings, the mass was diagnosed as an oesophageal duplication cyst.



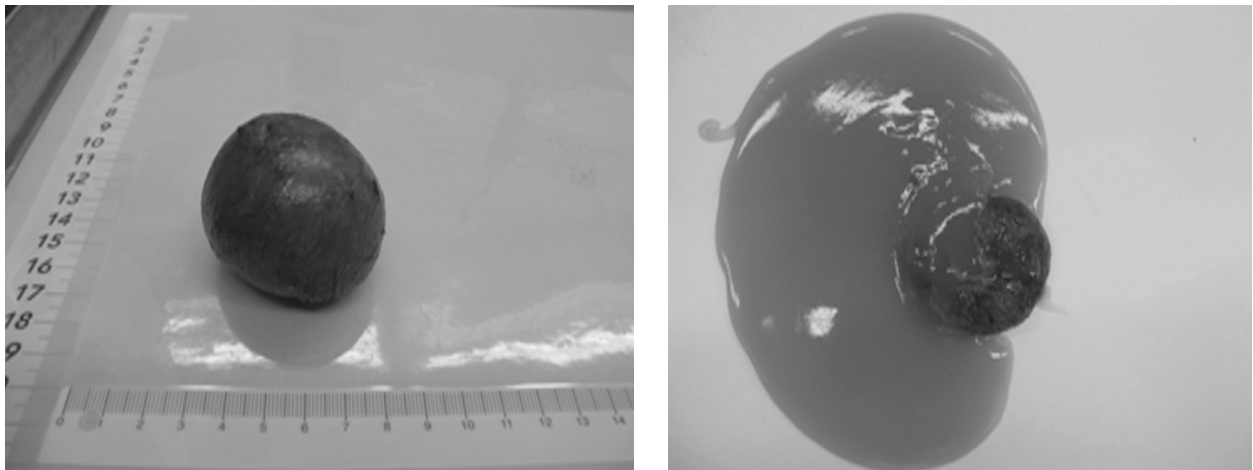


Figure 5. Aspect of the cyst after surgical excision and section. The lesion consisted of a 5 × 3 cm unilocular, ovoid cystic mass with a smooth and pink-to-red outer surface. A cut section revealed that the cyst contained abundant, viscous and yellow material which was partly adherent to the cystic wall

The dog was reevaluated three months after surgery clinically and endoscopically and no abnormalities were found. A further follow-up, three years after surgery, revealed a normal clinical condition.

## DISCUSSION AND CONCLUSIONS

Oesophageal duplication cysts are considered to be a rare condition in humans and result from either incomplete embryologic recanalisation or atypical blastogenesis of the primitive foregut, usually between the fifth and eighth weeks of gestation (Martin et al. 2007).

Sometimes they are associated with other congenital anomalies such as oesophageal atresia distal to duplication, small intestinal duplication, tracheo-oesophageal fistula and spinal abnormalities (Carachi and Azmy 2002; Martin et al. 2007). Hemivertebrae, vertebral fusion, and spina bifida are the most frequent vertebral anomalies in humans with foregut duplication, and they result from a failure of the mesoderm to fuse (Martin et al. 2007). Moreover, cardiac anomalies and pericardial defects are associated congenital disorders (Carachi and Azmy 2002). None of these malformations were found in our case.

The clinical presentation is characterised by signs related to a partial oesophageal obstruction and bronchial or vascular compression, usually

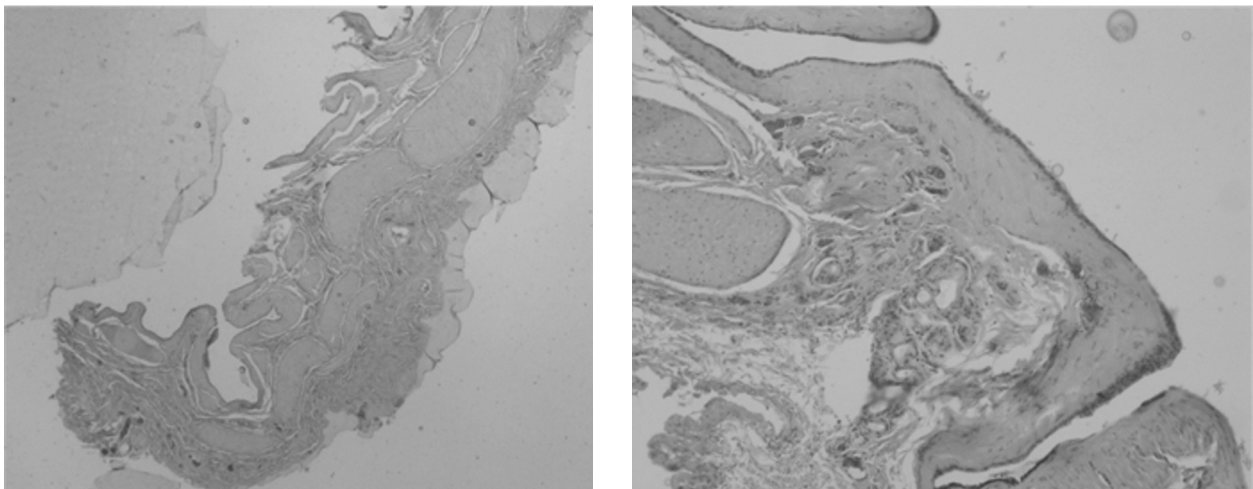


Figure 6. Histological cross-section of oesophageal duplication cyst. Cuboidal non-ciliated pseudostratified epithelium with abundant submucosal mucous glands were present. The wall was composed of a double muscle layer

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as a consequence of enlargement of the structure due to inflammation or haemorrhaging of the cyst (Sundaramoorthi et al. 2000; Carachi and Azmy 2002; Martin et al. 2007). In humans, upper oesophageal cysts usually lead to respiratory distress due to tracheobronchial compression, while middle and lower cysts can lead to gastrointestinal disturbances, such as dysphagia, regurgitation, vomiting and cardiovascular and respiratory symptoms, including arrhythmias and respiratory distress (Sundaramoorthi et al. 2000; Carachi and Azmy 2002). Nonetheless, about 35% of the patients with middle or lower oesophageal cysts are asymptomatic, and, in our patient, only gastrointestinal symptoms were reported (Sundaramoorthi et al. 2000; Jan et al. 2016).

Preoperative diagnosis can be established with standard radiological techniques, computed tomography, magnetic resonance (MR), endoscopy and endoscopic ultrasound (EUS) (Carachi and Azmy 2002; Gupta et al. 2010; Agarwal and Bagdi 2011).

EUS may be helpful in defining the fluid-filled nature of the duplication, and an echogenic inner rim is highly suggestive of a cystic nature of the lesion. Fine needle aspiration of the lesion under EUS guidance can be performed; however, such a procedure could promote infection. An oesophagogram and endoscopic scanning usually show external compression of the oesophagus, without involvement of the mucosa, while computed tomography scans clearly demonstrate the presence of a fluid-filled lesion, its exact anatomical location and the relationship with adjacent structures. In addition, CT also permits simultaneous investigation and evaluation of the spine, thoracic and mediastinal structures, thus enabling the diagnosis of possible complications of the oesophageal anomaly (Carachi and Azmy 2002; Gupta et al. 2010; Agarwal and Bagdi 2011).

In human medicine, a complete surgical excision by thoracotomy or thoracoscopy is the treatment of choice and is always indicated to prevent complications (Sundaramoorthi et al. 2000; Agarwal and Bagdi 2011). Leaving the cyst untreated may eventually lead to significant complications including mediastinitis, due to infection of the mediastinal cyst, life-threatening haemoptysis, broncho-oesophageal fistula or more rarely, malignant transformations (Sundaramoorthi et al. 2000; Carachi and Azmy 2002; Agarwal and Bagdi 2011; Obasi et al.

2011). In addition, surgical excision is also required for a definitive diagnosis based on histological examination of the surgical specimen. The surgical technique must focus on maintenance of the integrity of oesophageal mucosa, the approximation of the muscle layers and preservation of the vagus and phrenic nerves in order to avoid the development of a pseudodiverticulum, as well as cardiopulmonary and gastrointestinal complications (Martin et al. 2007; Kang et al. 2008; Gupta et al. 2010; Agarwal and Bagdi 2011; Obasi et al. 2011).

In humans, open surgical resection achieved using posterolateral thoracotomy is the best surgical option for excision of the cyst, but video-assisted thoracoscopic surgery and robotic surgery have recently become viable surgical options (Obasi et al. 2011). However, the thoracoscopic approach may not always be possible and requires specific equipment and surgical skills (Obasi et al. 2011).

When performing intercostal thoracotomy, the cyst is best approached through the side of the thorax from which it protrudes (Agarwal and Bagdi 2011). The oesophageal muscle should be incised longitudinally to expose the cyst, which can be removed by performing an extramucosal excision, leaving the underlining mucosa intact. Decompression of the cyst may assist in its removal, but the dissection is often aided by leaving the cyst intact for as long as possible (Agarwal and Bagdi 2011).

In our case, the cyst was lying completely within the wall of the oesophagus, was not invading the oesophageal lumen and the removal was completed without opening or draining it.

The overall complication rate of surgical treatment of ODCs is very low in humans and includes pneumonia, oesophageal leaks, pseudodiverticulum, vagus and phrenic nerve paralysis and wound infection (Sundaramoorthi et al. 2000; Carachi and Azmy 2002). None of these complications were observed in our patient.

Oesophageal cysts can be classified as duplications if histological evaluation satisfies three criteria: a lesion adherent to or attached to some segment of the foregut, the presence of two muscular layers in the wall and an internal layer of squamous, columnar, cuboidal, pseudostratified or ciliated epithelium (Gabor and Walshaw 2008). In humans, it is known that duplication cysts may display a variety of epithelial types and are differentiated from bronchogenic cysts by the absence of cartilage or respiratory glands (Martin et al. 2007).

In conclusion, dysphagia is the most frequent clinical sign of lower oesophageal duplication cysts in humans (Carachi and Azmy 2002). Duplication cysts must be considered in the differential diagnosis of oesophageal masses in dogs, despite the rarity of the disease. Therefore, surgical treatment should be recommended, as in humans, immediately after diagnosis, in order to avoid life-threatening complications such as bleeding, aspiration or malignant transformation (Sundaramoorthi et al. 2000; Carachi and Azmy 2002; Martin et al. 2007).

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