pISSN 1598-298X / eISSN 2384-0749
J Vet Clin 35(3) : 114-118 (2018)
http://dx.doi.org/10.17555/jvc.2018.06.35.3.114

Journal of Veterinary Clinics
www.ksvc.or.kr

Multiple Congenital Vascular Anomalies In a Lakeland Terrier: Computed Tomographic Angiographic Evaluation

Moonjung JANG, Sangkyung CHEON, Wanhee KIM, Mincheol CHOI and Junghee YOON[1]

Department of Veterinary Medical Imaging and Research Institute for Veterinary Science, College of Veterinary Medicine, Seoul National University, Seoul 151-742, Korea

(Received: April 25, 2018 / Accepted: June 11, 2018)

Abstract : A 3-month-old intact male Lakeland terrier was presented with recurring regurgitation after removing cervical esophageal foreign body by endoscopy. Blood and urine analysis, radiography, ultrasonography, fluoroscopic esophagography, computed tomographic angiography (CTA) were performed. In radiography and fluoroscopic esophagography, vascular ring anomaly was considered as the primary cause of megaesophagus, and CTA with gas-inflation of the esophagus was performed. Compressed esophagus, persistent right aortic arch (PRAA), aberrant left subclavian artery (LSA), and a venous structure which was confirmed in surgery to be incomplete type persistent left cranial vena cava (PLCVC) connected with the left side azygos vein were observed. Left deviation of the trachea was also revealed in CT, which implies the compression by left ligamentum arteriosum. Therefore, type 3 PRAA with left ligamentum arteriosum and aberrant LSA, was considered as a prior differential diagnosis. Surgical repair was performed and the clinical signs improved. This report describes CTA characteristics of combination of PRAA with aberrant LSA, incomplete PLCVC and Lt. azygos vein in a dog. Although not every vascular anomaly does induce clinical sign, some types can complicate the surgical procedure, and cause clinical signs. Therefore, thorough evaluation of vascular anomalies in the thorax is important, and CTA is a useful method in identifying multiple vascular anomalies in dogs.

***Key words* :** Megaesophagus, aberrant LSA, incomplete PLCVC, Left azygos vein, CTA.

Introduction

Regurgitation in dogs is a principal clinical sign in vascular ring anomaly (VRA) (17). VRA occurs due to the abnormal development of embryonic vessels, as abnormally located vessels or ligaments encircle the trachea and esophagus with complete or partial vascular ring, resulting in megaesophagus (9,13). The most common form of clinical cases of VRA is PRAA which accounts for 95% (4), and PRAA may concur with other VRAs such as aberrant left subclavian artery (LSA), persistent left cranial vena cava (PLCVC), and persistent ductus arteriosus (PDA) (4,9). Classification scheme for various types of VRA was suggested (6). According to the scheme, Type 1 is the most common form consisted of PRAA and left ligamentum arteriosum. Esophageal compression caused solely by aberrant LSA with PRAA and Rt. ligamentum arteriosum is type 2. Combination of PRAA, aberrant LSA and Lt. ligamentum arteriosum is designated as type 3, and double aortic arch is type 4. Although most VRA can be corrected through a left lateral thoracotomy, surgical plan is determined according to certain forms of VRA or existence of vessel that can complicate operative field (9). Therefore, it is important to recognize every vascular anomalies not only the one compressing the esophagus, but also other abnormal vessels or ligaments in the thorax.

Computed tomographic angiography (CTA) is broadly utilized to detect the location and features of the blood vessels, and also used to diagnose various kinds of VRA (2,18,19). The purpose of this study is to present a CT features of unusual combination of VRA in a Lakeland terrier, and to emphasize the utility of CT angiography as an essential diagnostic tool of VRA.

Case

A 40-day-old intact male Lakeland terrier was referred with the chief complaint of regurgitation occurring for the past 2 days. An esophageal foreign body was initially diagnosed by referring veterinarian using positive contrast esophagography. At the time of admission, the dog started weaning and body weight was 0.94 kg. A foreign body was palpated in the cervical region. Serum glucose and globulin levels were mildly low (glucose 60 mg/dL, globulin 1.9 g/dL). On thoracic radiographs, two soft tissue opacity masses, which were suspected to be the foreign body, were located in the cranial cervical esophagus (Fig 1A). Endoscopy was performed the following day, and a soft potato-like material was removed, and the dog was discharged that night.

Two months after the endoscopy, however, the patient visited again. He gained weight (from 0.94 to 2.00 kg), but regurgitation started again 1 month after the removal of the esophageal foreign body, when the patient began to eat solid

[1]Corresponding author.
E-mail : heeyoon@snu.ac.kr

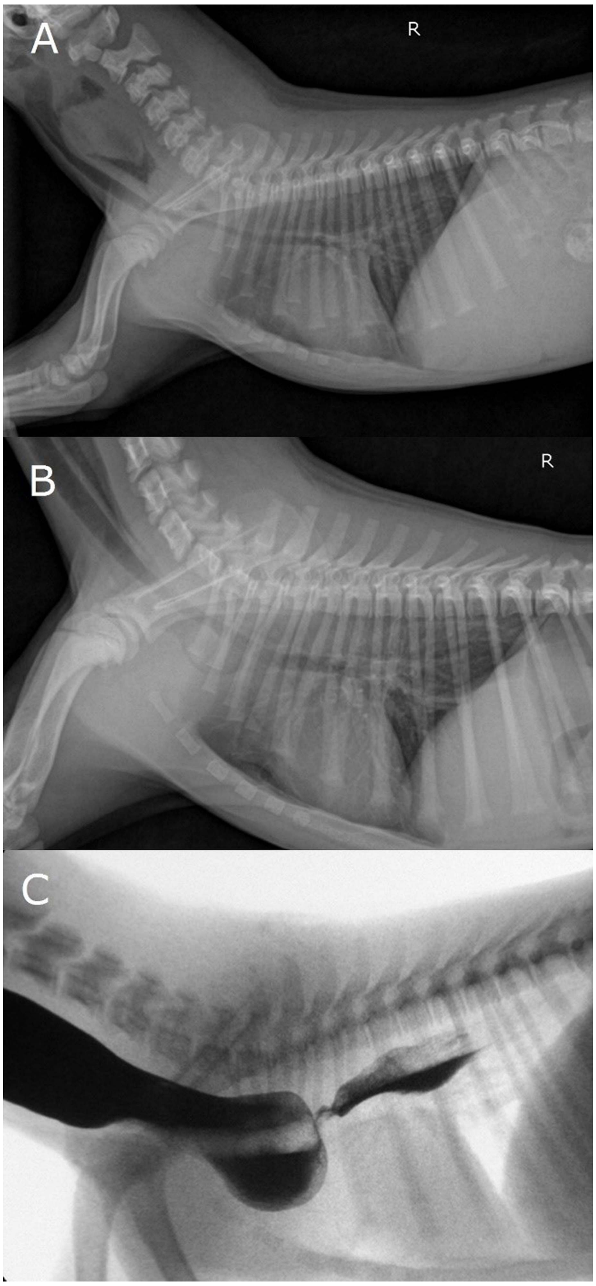

Fig 1. Lateral thoracic radiographs. At the first visit (2 months after the removal of the foreign body (B), and positive contrast fluoroscopic image (C). Soft tissue opacity masses in the cervical esophagus were observed (A). The masses were soft foreign bodies and removed by endoscopy. However regurgitation reoccurred 2 months later, and dilated esophagus with a structure like diverticulum in cranial region of the heart base was detected in the radiography (B). Very narrowed esophageal lumen was discovered by following positive contrast fluoroscopic study (C). Almost all of the contrast media was collected in the cranial esophagus.

foods. Generalized megaesophagus especially in the cervical part and a focal bulging of the esophagus just cranial to the heart base were revealed by radiography (Fig 1B). The tracheal sign, which means focal left side deviation of the trachea and which suggest the presence of a VRA (4), was not clear due to the gas opacity of the megaesophagus (Fig 2A).

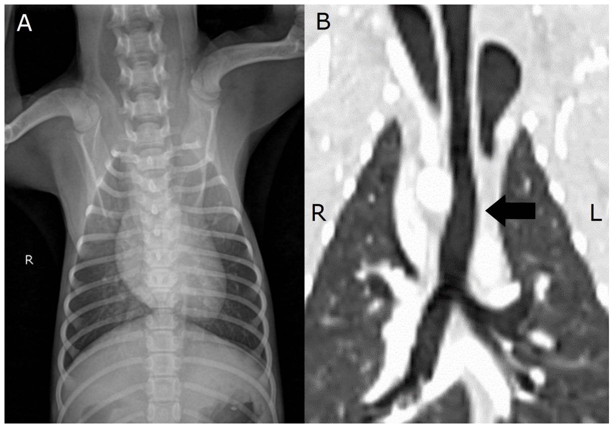

Fig 2. VD view radiograph on the second visit in the hospital (A). Post-contrast enhanced MPR image of CT (B). Megaesophagus in the cranial esophagus with gas was found in the VD view radiography (A). Tracheal margin was indistinct because of the gas opacity in the esophagus and the presence of tracheal sign was ambiguous. In multiplanar reconstruction (MPR) image of CT (B), mild left deviation of the trachea was clearly observed. Gas in the cranial esophagus was also seen, but it could be distinguished from the trachea as the tracheal wall was visualized without superimposition.

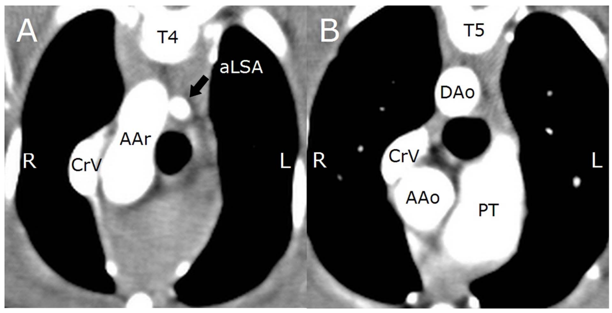

Fig 3. Post-contrast transverse CT images of the thorax at the 4th thoracic vertebra (A), and 5th thoracic vertebra level (B). Aortic arch was located in the Rt. side, and the aberrant LSA was originated from the left dorsolateral surface of the aortic arch (A). Ascending aorta and descending aorta were located on the Rt. side which is consistent with PRAA (B). T4, 4th thoracic vertebra; T5, 5th thoracic vertebra; AAr, aortic arch; aLSA, aberrant left subclavian artery; CrV, cranial vena cava; DAo, descending aorta; PT, pulmonary trunk.

Subsequently, positive contrast esophagography was performed. Most of the contrast media was collected in the severely dilated cervical esophagus, with a very abruptly narrowed lumen located just cranial to the heart base (Fig 1C). These features suggested that the megaesophagus was caused by VRA (4).

CTA was performed using a 64-row MDCT scanner (Aquilion 64®; Toshiba, Tochigi, Japan), to differentiate the types of VRA and to rule out any other causes of megaesophagus. General anesthesia was induced using a combination of midazolam (0.1 mg/kg; IV), ketamine (1 mg/kg; IV), hydromorphone (0.025 mg/kg; IV) and alfaxane (2 mg/kg; IV), and maintained with isoflurane and oxygen. The dog was posi-

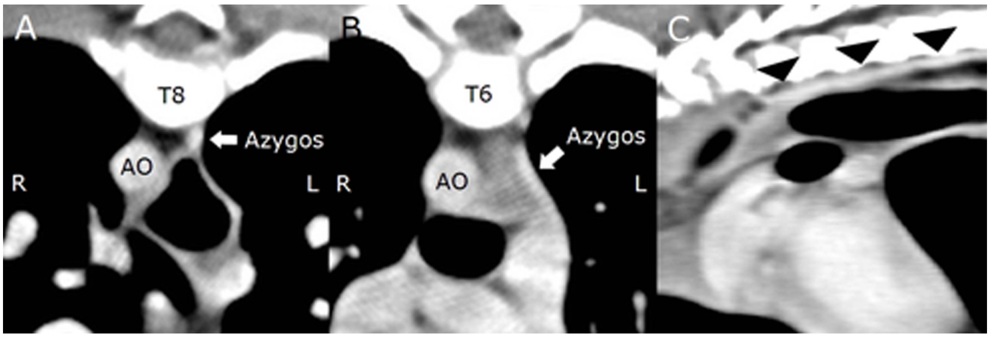

Fig 4. Post-contrast transverse (A, B), and sagittal CT image (C). Azygos vein was coursing on the left side of the aorta from the caudal thorax (A, C). The left azygos vein ran toward the ventral direction at the T6 level to join the PLCVC (B). T8, 8^{th} thoracic vertebra; T6, 6^{th} thoracic vertebra; Ao, aorta; Azygos, azygos vein.

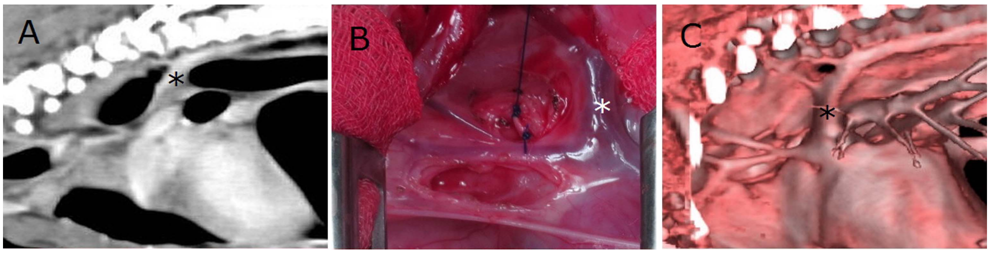

Fig 5. MPR CT image of the costocervical-vertebral trunk (A), and intraoperative photograph. Costocervical-vertebral trunk and left azygos vein (asterisk) were confirmed by left thoracotomy, to meet in a short vein which is the incomplete PLCVC. Reviewed 3D reconstruction image (C) of the preoperative CTA images shows the structure of the incomplete PLCVC.

tioned in sternal recumbency and helical CT scan of the neck and thorax was performed with dual phase angiography (arterial phase in 8 seconds and venous phase in 40 seconds), using total 7 ml of intravenous contrast media, iohexol (Omnipaque 300; GE healthcare, Cork, Ireland) at a rate of 2 ml/s. Air injection in the cervical esophagus was administered manually through a balloon catheter just before prescan. The pre-phase CT scan identified a focal and mild left tracheal deviation at the level of the caudal part of the 3rd thoracic vertebrae (Fig 2B). Also, the gas-filled dilated esophagus had a compressed region from the level of the 3rd to the 6th thoracic vertebrae. In arterial phase, right-sided position of the aortic arch and descending aorta was found, and the right subclavian artery initiated after the bicarotid trunk. The aberrant LSA branched from the left dorsolateral surface of the aorta, after 8.43 mm from the origin of the RSA (Fig 3). The LSA originated from the aorta and coursed cranially along the dorsolateral aspect of the esophagus. The aberrant LSA was moderately compressing the esophagus, and the esophagus cranial to the aberrant LSA was dilated. In venous phase, the azygos vein was identified to course to the left of the aorta, and to enter into the heart (Fig 4). The Rt. and Lt. external jugular veins joined at the level of the thoracic inlet and formed the normal Rt. cranial vena cava. A V-shaped venous trunk was observed at the Lt. cranial region of the heart base (Fig 5A). Based on CT angiography, Rt. sided aortic arch and aberrant LSA were confirmed to consist a vascular ring. Ligamentum arteriosum was not visible, but its existence was suspected because of the tracheal sign. Therefore, among the various types of VRAs, type 3 was our first differential diagnosis.

Surgery to correct the VRA was performed 10 days later. In the left side thoracotomy, the ligamentum arteriosum was revealed and ligation and resection was performed. During the surgical procedure, the costocervical-vertebral trunk and Lt. azygos vein draining into the PLCVC was confirmed (Fig 5B). The V-shaped venous trunk was the left costocervical-vertebral trunk, and the 3D reconstruction review of preoperative CTA images shows the structure in detail (Fig 5C). After post-operative care, the patient was discharged.

No regurgitation occurred with soft food during short term rechecks, but after several weeks, intermittent vomiting recurred when solid food was given. Esophagography was performed again, and dilation of the cervical esophagus and narrowed esophageal lumen at the heart base level was found to be similar to the previous exam. However, we could observe that the motility of the narrowed site improved mildly, and the transition of the contrast media bolus was quite fair in the caudal esophagus. As the symptom persisted, the patient was planned to be checked up regularly and another surgical procedure for esophagus may be considered after the patient become adult.

Discussion

When young dogs have regurgitation and megaesophagus after weaning, a congenital VRA should be included in the differential diagnosis. But in this case, when the patient first visited our hospital, congenital diseases were not considered as the first diagnosis because the esophageal foreign body was obvious in radiography and endoscopy. Furthermore, the location of the foreign body was far cranial from the PRAA site, and the time of weaning, regurgitation, and the presence of a foreign body all overlapped. In spite of the foreign body removal, regurgitation persisted, and on the second visit, various imaging techniques were used to make the exact diagnosis. Esophagography suggested VRA, and type 3 VRA was diagnosed with CTA, and appropriate surgical management was performed.

A previous study suggested that the left ward curvature of the trachea on the radiograph can be used to distinguish PRAA from generalized megaesophagus without performing contrast esophagography (4). The radiograph of this case, however, did not show the tracheal margin as well as the left tracheal deviation. Consequently contrast esophagography was performed and the significant narrowing of esophagus at the base of the heart suggested PRAA. Mild and focal left deviation of the trachea was revealed in the CT images of this dog, even though this sign had not been observed in the radiograph. Superimposition of gas in the enlarged esophageal lumen and cranial mediastinal structures are thought to mask the tracheal wall. If a patient's ligamentum arteriosum is not directly visible in CT images, tracheal deviation can be found using MPR regardless of the any esophageal content or mediastinal structures. Furthermore, tracheal sign in radiograph only can imply the type 1 or type 3 VRA, but when it is applied to CT, state of both aberrant LSA and Lt. ligamentum arteriosum can be recognized. Therefore, the tracheal sign in CT may enable one to differentiate type 1, 2 and 3.

Other findings in this dog are Lt. azygos vein and incomplete PLCVC. In normal development of a dog, the Rt. supracardinal vein becomes azygos vein, but if the Lt. supracardinal vein develops instead, it becomes Lt. azygos vein (3). Previous case report already reported the Lt. azygos vein in a dog with angiographic features, but there is no study reporting CT angiographic feature of it. PLCVC is one of the uncommon congenital venous anomalies in dogs, and is a fetal venous remnant of left cranial cardinal vein, which should normally disappear during development. This anomaly does not induce significant clinical sign, but there are some cases in human in which the PLCVC connected to the left atrium and caused cyanosis (3,14). In veterinary medicine, there is a previous study of angiographic characteristics, classification of complete and incomplete type (3),

and also there is a recent retrospective study describing various CT angiographic features of complete PLCVC (5). Incomplete PLCVC in our patient was observed as a V shaped venous trunk, and confirmed by surgical observation. The incomplete PLCVC was the short proximal portion of the left cranial vena cava which drains the left costocervical-vertebral trunk and the Lt. azygos vein, instead of the Lt. brachiocephalic trunk. The Lt. and Rt. brachiocephalic veins joined into the Rt. cranial vena cava alike normal drainage. A case which is like a mirror image of the incomplete PLCVC was described in the retrospective study (5), but the present report is the first description of CT angiographic features of an incomplete PLCVC in a dog. In human, a recent case report described the coexistence of double superior vena cava and left coursing azygos vein. This report emphasized that awareness of the left superior vena cava and azygos vein is valuable for central venous catheterization and mediastinal operations of large vessels (12), and this would be also valuable equally to veterinary medicine. The imaging characteristics of these anomalies can be unfamiliar because of their rareness. Therefore, care for this kind of anomalies must be taken in evaluation of blood vessels in VRA patients.

Various VRA, resulting in compression of esophagus, occur in dogs. It should be noted that new forms of ring anomaly and concurrent multiple combination of arterial and venous anomalies, are constantly being reported until recently. There was a case report of the fibrous band of PLCVC itself compressing the esophagus (11), and other reports describing hypoplastic aberrant LSA (2), aberrant right subclavian artery (16,20), right patent ductus arteriosus (7), and multiple concurrent venous anomalies (20). Like these cases, unexpected form of VRA, and unusual combination of vascular anomalies are always possible. Therefore, exact identification of all the anomalies should be emphasized for good patient management, and CT angiography can be utilized. Before the CT became a common modality in veterinary medicine, radiography, esophagography, angiography and esophageal endoscopy were used to diagnose VRAs (7,8,10,17). As various cases are present, CT angiography should be used as a superior modality, to obtain direct visualization of the complicated and variant arteries, veins, and adjacent anatomical structures, and also 3D reconstruction.

PRAA, aberrant LSA, incomplete PLCVC and Lt. azygos vein are arterial and venous vascular anomalies found in this case. Several cases of type 3 VRA have been reported in dogs and cats with or without other vascular anomalies (1,10,15), but there are few case reports including CT angiography evaluation (2,19). Furthermore, CTA characteristics of uncommon vascular anomalies make this case worth reporting.

Conclusions

In conclusion, CTA provides information about the type of VRA, and also reveal concurrent vascular anomalies, thereby altering the surgical plan and outcome. Furthermore, identifying new forms of anomalies and accumulating the anatomical data by CTA may be helpful for future studies.

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