ISSN: 1220-3173; E-ISSN: 2457-7618

BUDD-CHIARI-LIKE SYNDROME CAUSED BY A CAUDAL MEDIASTINAL CYST IN A DOG: CASE RAPORT

SINROMUL BUD-CHIARI SECUNDAR UNUI CHIST MEDIASTINAL LA CÂINE: RAPORT DE CAZ

Iulia MELEGA[1),*)], C.P. PEȘTEAN[1)], R. PURDOIU[1)], Lucia Victoria BEL[1)]

ABSTRACT

This report presents a case of intrathoracic caudal vena cava partial obstruction, leading to marked ascites due to a space-occupying cystic lesion in a six-year-old German Shepherd mixed-breed dog. The dog was referred for investigation due to lethargy and progressive abdominal distension persisting for 3 weeks. A thorough examination, including a physical examination, routine blood analysis, abdominal and thoracic ultrasonography, radiography, and computed tomography imaging, was performed. Thoracic radiography revealed a well-delineated, oval-shaped mass measuring 11x12 cm in the caudal mediastinum. The CT scan confirmed the presence of a soft tissue-attenuating space-occupying lesion, a thin-walled, fluid-filled, non-enhancing homogeneous mass attached to the oesophagus but without communication with its lumen. To address the issue, an intercostal thoracotomy was performed to resect a subtotal window from the cystic wall, allowing drainage and healing. Histopathological analysis revealed a fibrous wall, consistent with a cystic structure. Culture and sensitivity tests showed no bacterial growth. The dog recovered uneventfully, and no recurrence of clinical signs was reported during the 6-month follow-up. To the best of the author's knowledge, this is the first reported case of caudal vena cava syndrome caused by a caudal mediastinal cyst in a dog.

Keywords: Budd-Chiari-like syndrome, mediastinum, cyst, thoracoscopy

REZUMAT

Studiul prezent descrie un caz de obstrucție a venei cave caudale intratoracice, care a condus la apariția ascitei, ca urmare a compresiunii cauzate de o leziune chistică la un câine din rasă mixtă ciobănesc german, în vârstă de 6 ani. Pacientul s-a prezentat pentru investigații, pentru letargie și distensie abdominală progresivă de o durată de 3 săptămâni. În vederea stabilirii diagnosticului s-au realizat investigații clinice și paraclinice reprezentate de analize de sânge, ultrasonografia abdominală și toracică, respectiv investigații radiologice și tomografia computerizată. Radiografia toracică a identificat o formațiune de aspect oval, bine delimitată, măsurând 11x12 cm în mediastinul caudal. Scanarea CT a confirmat prezența unei formațiuni cu atenuare de țesut moale, cu pereți subțiri, conținut fluid, ca masă omogenă atașată esofagului, dar fără comunicare cu lumenul acestuia. Tratamentul chirurgical a constat in efectuarea unei toracotomii intercostale cu rezecția subtotală a peretelui formațiunii, permițând drenajul și vindecarea. Testele de cultură și sensibilitate nu au arătat nicio creștere bacteriană. Recuperarea a avut loc fără incidente și nu a fost raportată nicio recidivă a semnelor clinice în timpul urmăririi post-operatorii timp de 6 luni. Din cunoștințele autorului, acesta este primul caz raportat de obstrucție de venă cavă caudală cauzată de un chist mediastinal caudal la câine.

Cuvinte cheie: sindromul Budd Chiari, mediastin, chist, toracoscopie

Mediastinal masses are common occurrences in small animals, constituting a heterogeneous group of both benign and malignant lesions, categorised based on their location and histopathology. Cystic lesions within the mediastinum have been documented in small animals, showing a higher incidence in cranial localization relative to the tissue of origin (12). In human medicine, mediastinal cysts make up 12-18% of all mediastinal masses, found in various compartments of the mediastinum (2). In dogs, previously reported caudal mediastinal cystic lesions originate within the mediastinum, encompassing abscesses (1, 5), mediastinal cysts (5, 6), empyema (7), and congenital duplication cysts (8).

Clinical manifestations of caudal mediastinal lesions are often nonspecific, occasionally reflecting symptoms of oesophageal obstruction. Symptoms secondary to compression of adjacent organs may also present as the primary complaint (7). Caudal vena cava syndrome has been previously reported as the initial manifestation of mediastinal masses, resulting from a direct mass effect significantly impacting the

1) University of Agricultural Sciences and Veterinary Medicine, Faculty of Veterinary Medicine, Cluj Napoca, Romania
*) *Corresponding author: iulia.melega@usamvcluj.ro*

caudal vena cava (14). In veterinary medicine, mechanical obstruction of venous blood flow between the liver and the junction of the caudal vena cava and right atrium is termed Budd-Chiari-like syndrome (BCS) and is characterized by hepatic venous outflow obstruction, passive venous congestion of the liver, marked ascites, and hepatomegaly. Primary BCS results from intraluminal venous lesions, while secondary BCS originates as a consequence of adjacent structures' compression or tumour invasion (1).

This report aims to describe the clinical findings and surgical treatment of a dog presenting with ascites associated with caudal vena cava partial obstruction due to a mediastinal cyst.

CASE DESCRIPTION

A neutered 6-year-old male German Shepherd mix breed dog weighing 39 kg presented with a history of lethargy, intermittent vomiting, progressive abdominal enlargement, and weight loss over the past 3 weeks. Upon examination, the dog had a body condition score of 3/9, and a ballotable fluid wave on abdominal palpation was observed. Vital signs were within normal limits,with a rectal temperature of 37.8°C, a pulse of 90 beats per minute, and a respiratory rate of 30 breaths per minute. Pitting oedema was palpated on both hindlimbs, extending from the extremities to the thigh.

The complete blood cell count revealed mild anaemia (haematocrit = 36.81%, reference interval 35.7-56.7%; red blood cell count = 5.95 x 10^12 /L, reference interval 5.5.-8.5 x 10^12 /L) and leukocytosis (white blood cells = 17.58 x 10^9/L; reference interval: 5-14.1 x 10^9/L). The leukocyte differential showed lymphocytosis (12.49 x 10^9/L, reference interval 0.4-2.9 x 10^9/L), monocytosis (2.7 x 10^9/L, reference intervals 0.1-1.4 x 10^9/L), and neutropenia (2.3 x 10^9/L, reference intervals 2.9 x 10^9-12.5 x 10^9/L).

The serum biochemistry indicated decreased total protein (4 g/dl, reference interval: 5.5-7.5 g/dL) and albumin levels (1.8 g/dL, reference interval: 2.3-3.7g/dL). The initial differential diagnosis considered liver disease, abdominal neoplasia, or congestive heart failure. Ultrasonographic evaluation of the abdominal cavity was impaired by the fluid content, and ultrasound-guided paracentesis was performed, resulting in the removal of three litres of moderate yellow, clear fluid characterised as a modified transudate on the Rivalta test. Cytology analysis of the aspirated fluid revealed a small proportion of lymphocytes, suggesting a mild inflammatory reaction with no bacterial pathogens. Thoracic radiographs displayed an oval-shaped soft tissue density in the caudal mediastinum, measuring approximately 11 x 12 cm (Fig. 1). Native and contrast-enhanced computed tomography (CT) scans of the thorax revealed a well-defined spherical space occupying lesion, fluid attenuating on native CT images but peripherally contrast-enhanced, measuring approximately 86/76 mm in the posterior mediastinum. The lesion involved the oesophageal wall, with no infiltration into other adjacent tissues observed (Fig. 2). These findings were indicative of a fluid-filled mass compatible with a cyst or an abscess. Exploratory lateral thoracoscopy was elected. Before surgery, a cardiology evaluation ruled out any presumed cardiac pathology. After premedication with maropitant (1 mg/kg), morphine (0.4 mg/kg, Morfina 20 mg/ml, Xentiva), and lidocaine (1 mg/kg, Xilina 10 mg/ml, Sicomed), general anaesthesia was induced using diazepam (0.25 mg/kg, Diazepam 5 mg/ml) and ketamine

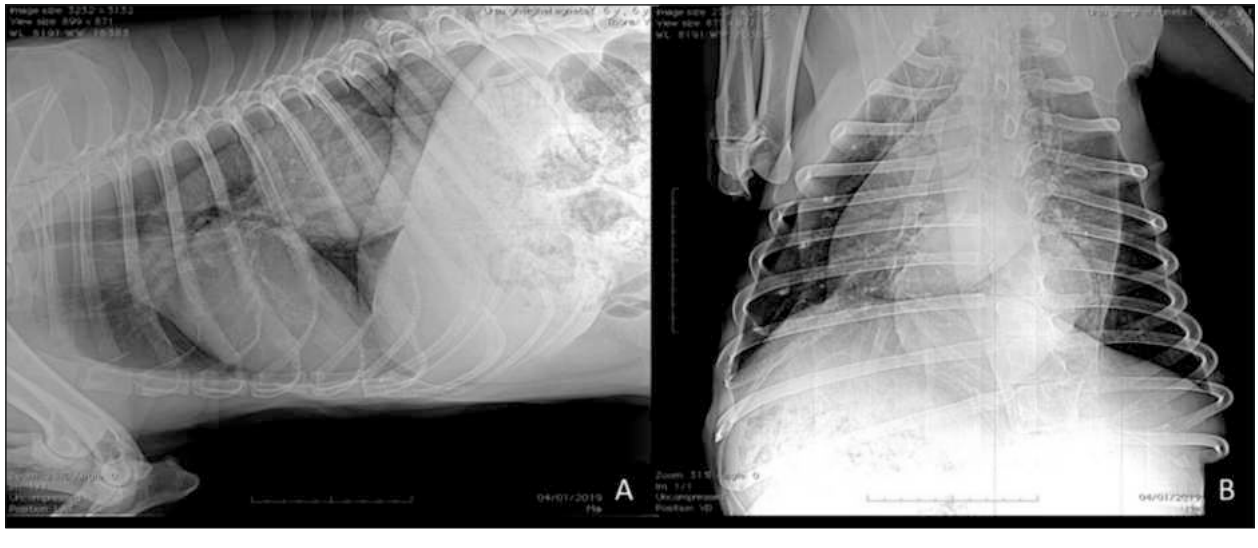

Fig. 1. Two-view thoracic radiography revealing an oval-shaped, well-distinctive soft tissue lesion in the caudal mediastinum (A. Lateral view; B. Dorso-ventral view)

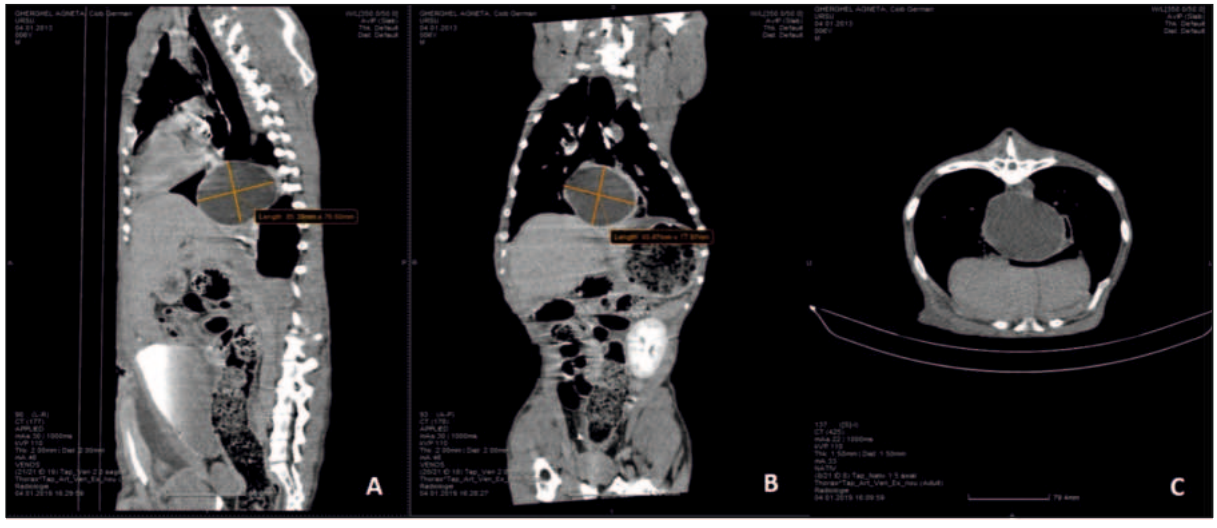

Fig. 2. The CT scan shows a spherical structure, with an isoatenuating aspect, located in the cardiac mediastinum, filled with fluid, thin-walled, well-circumscribed and attached to the oesophagus, but not accompanied by communication with its lumen, with no contrast enhancement within the mass (A. Coronal Plane, B. Sagittal Plane, C. Axial Plane)

(2 mg/kg, Narkamon 100 mg/ml, Bioveta) intravenously. Anaesthesia was maintained with isoflurane (ET 1-1.5%, Isoflutek 1000 mg/g, Laboritorios Karizoo) in oxygen in a semi-closed circuit. Throughout the procedure, a constant rate infusion of morphine (2 mcg/kg/min), ketamine (2 mcg/kg/min), and lidocaine (20 mcg/kg/min) in Ringer's solution was administered. Ventilation was maintained at a tidal volume of 8-10 ml/kg and a frequency of 12-15 breaths per minute using a pressure control ventilator (Drager Fabius Plus XL, Germany). Epidural anaesthesia and intercostal nerve blocks were performed using bupivacaine 0.25% (2 mg/kg, Bupivacaina Infomed 5 mg/ml, Grindeks). The patient was positioned in left lateral recumbency, and the right thorax was aseptically prepared. Monitoring included invasive arterial blood pressure, lead II electrocardiography, respiratory rate, end-tidal CO2, end-tidal isoflurane, oesophageal temperature, oxygen saturation using a multiparameter monitor (Vista Drager,Germany) recorded every 10 minutes.

A 10 mm port was introduced in the middle third of the right ninth intercostal space for the insertion of a 30-degree rigid telescope. The soft tissue mass was visualized in the caudal mediastinum, partially covered by the right diaphragmatic lung lobe, associated with the right side of the oesophagus wall. The lesion consisted of a unilocular, round mass, with the outer surface smooth and red-tan. The video-assisted tho-

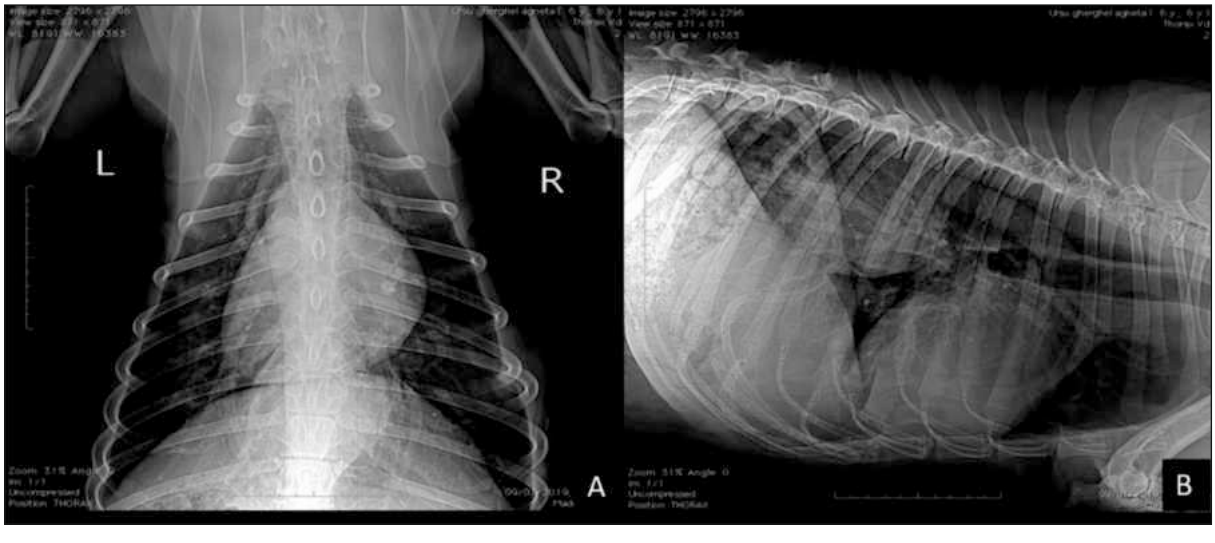

Fig. 3. Post-operative thoracic radiography revealing the reduction in size of the mediastinal mass (A. Dorso-ventral view; B. Lateral view)

racoscopy was converted to lateral thoracotomy due to restricted access. An orogastric tube (12 Fr Nelaton) was inserted for oesophagus identification, and needle aspiration collected serohaemorrhagic fluid from the mass. Active aspiration was performed, and partial resection of the ventrolateral mass wall was done, to allow complete drainage. Total resection was limited by adhesions to the oesophagus and proximity to the caudal vena cava and aorta. The thoracic cavity was irrigated with sterile saline (NaCl 0.9%), and a sample of the mass tissue was sent for histopathology. A thoracic drain was placed before the closure of the thoracotomy incision.

Postoperatively, the dog was hospitalised for seven days. Pain relief was obtained with a constant rate infusion of morphine-ketamine-lidocaine solution for 24 hours, followed by tramadol hydrochloride (4 mg/kg, Tramadol 50 mg, Krka, Slovenia) combined with meloxicam (0.2 mg/kg) for six days. Antibiotic therapy with cephoperazone/sulbactam (25 mg/kg, 1000 mg/1000 mg, Sulcef, Medochemie) and metronidazole (15 mg/kg, 5 mg/ml, Metronidazol Braun intravenously, continued for seven days. The thoracic drain was removed 48 hours postoperatively. Thoracic radiographs after 72 hours showed a reduced mass size with no abnormalities, and abdominal ultrasound revealed no free abdominal fluid (Fig. 3).

Histological examination of the excised tissue revealed a proliferation of mature, fibrous tissue. Additionally, there was a presence of young fibrous tissue proliferation with a lax, edematous appearance, abundant blood vessels of varying calibers, and numerous fibroblasts. This fibrous tissue formed a cavity containing serosanguineous content. The cells showed no atypical characteristics, and no inflammatory aspects were observed. The overall features were suggestive of a cystic capsule. The cytology of the fluid aspirated from the mass did not reveal any pathological agents. Bacterial culture performed on blood agar 5% showed no bacterial growth. At the 6-month follow-up, no recurrence of clinical signs was reported.

DISCUSSIONS

The case report details a successfully treated mediastinal cyst complicated with secondary Budd-Chiari-like syndrome.

The clinical presentation was marked by signs related to partial vascular compression due to the gradual enlargement of the structure. The initial presentation of mediastinal cystic lesions depends on their size and location. In both human and veterinary patients, benign cystic lesions are often incidentally discovered during investigations for unrelated complaints. The affected patients can present with non-specific oesophagus-related symptoms or airway obstruction, lung atelectasis, and arrhythmias as a consequence of compression on mediastinal structures (7, 8). Obstruction of venous blood flow from the liver to the right atrium can occur anywhere along the venous course, from the hepatic venules to the junction of the caudal vena cava with the right heart (1). The clinical picture in affected patients is typically subacute or chronic, with hepatomegaly and protein-rich abdominal effusion being cardinal features. In our patient, the evolution was progressive, featuring anorexia, weight loss, severe abdominal effusion, likely secondary to chronic post-sinusoidal venous obstruction, increased hepatic sinusoidal pressure, and subsequent leakage of high-protein lymph from the liver capsule into the peritoneal cavity. Additionally, the pitting oedema in the rear legs may have resulted from increased pressure in the abdominal cava. Blood biochemistry analysis revealed hypoalbuminemia and decreased total protein content, suggesting chronic protein loss. Previously reported cases of BCS have been associated with various causes, including malignancies and non-malignant causes (11). It's noteworthy to mention the less common presentation of vena cava compression due to mediastinal lesions. One study associated this syndrome with oesophageal leiomyoma, demonstrating that a large oesophageal mass can have a significant hemodynamic effect on caval venous flow (9). The diagnosis of BCS is established based on clinical features, accompanied by imaging findings demonstrating vena cava obstruction (13). Plain radiographic imaging of the thorax revealed the smooth contour of the mass in the caudal mediastinum. In order to better differentiate the origin of the mass (mediastinal vs pulmonary), further investigation using CT was needed (15). Computer tomography imaging revealed a firm attachment between the capsule and the oesophageal wall, with no communication identified with the oesophageal lumen, an no pneumopathy adjacent to the lesion.

The acute clinical course and imaging findings supported the decision for surgical exploration of the mass. Unfortunately, complete resection of the capsule proved difficult due to dense adhesions, but partial resection of the cystic wall allowed for drainage and completely reduced compression of the intrathoracic caudal vena cava. While studies in human patients have reported recurrence following incomplete resection (6), long-term follow-up in dogs with para-esophageal abscesses has shown no recurrence of previous signs (5). Non-surgical clinical remission of a mediastinal abscess was reported by Breheny et al (2023), through an ultrasound-guided placement of a thoracostomy tube into the abscess, allowing drainage to be performed (4).

The cytology analysis of the peritoneal fluid indicated a mild inflammatory reaction with low cellularity. Bacterial culture performed on the fluid sample and ex-

cised tissue failed to identify any growth. Histopathological examination of the capsule revealed fibrous tissue without epithelial lining, consistent with findings in other cases of mediastinal cysts and abscesses in dogs (11). The pathogenesis of the cyst remains incompletely understood, but according to a study by Gendron et al. (2017), it is associated with anatomical location, with lesions resembling fluid-filled pockets in the caudal mediastinal compartment originating from the mediastinal serous cavity. The authors observed a similar appearance of paraoesophageal empyema with the presentation of cysts or abscesses. The development of this lesion is secondary to foreign body penetration, local extension, or hematogenous seeding (10).

The close relationship to the oesophagus could indicate foreign body migration through the oesophagus and contamination of the mediastinum. Contrary to other studies (1, 2), our patient exhibited no neighbouring lesions, further supporting the supposition of an oesophageal origin for the cyst. This observation aligns with three cases reported as inflammatory mediastinal cysts (2) and non-pulmonary caudal mediastinal paraoesophageal abscesses (1).

In Brissot et al.'s (2010) (5) report on caudal abscesses in seven dogs, bacterial agents were successfully isolated in only one case out of seven. Preoperative cytological evaluation revealed intracellular bacteria in two dogs, and in four dogs, the excised tissue showed pyogranulomatous changes consistent with abscessation. Interestingly, Gendron et al. (2017) suggest that the serous exudate aspect, although purulent, could be observed before the development of frank pus with negative bacterial culture (10). In contrast to our findings, Gremilion et al. (2017) described a case of a caudal mediastinal cystic lesion with a positive bacterial culture for Pseudomonas spp., and the definitive aetiology of the cysts in this case was not established (11).

Another potential diagnosis for a thin-walled caudal mediastinal structure is oesophageal congenital cysts, which are malformations resulting from a failure of vacuolization and budding of the primitive oesophagus (8). Gabor and Walshaw (2008) reported the first congenital duplication cyst in the cervical part of the exophages, presenting as a fluctuant mass at the cranioventral aspect of the neck (9). Histopathological examination revealed characteristics like location within the oesophageal wall, double muscle layers in continuation with the muscularis propria, and pseudostratified epithelium with abundant submucosal glands. Despite the cyst being attached to the oesophageal wall in our patient, histological characteristics were inconsistent with sustaining the congenital origin of the lesion.

At the six-month follow-up after surgery, no recurrence of clinical signs was noted, indicating the resolution of BCS through surgical resection. No further blood analyses were performed during this time due to economic constraints.

CONCLUSIONS

Despite the unclear definitive cause of cyst formation, the authors concluded that it likely resulted from a small oesophageal perforation, leading to a chronic fibrous reaction and exudate accumulation, causing caudal vena cava syndrome.While mediastinal cysts are rare, they should be considered as a potential cause of BCS in dogs. To the best of the authors' knowledge, this is the first reported case of caudal vena cava syndrome caused by an oesophageal cyst in a dog.

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