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Case Report

Long-term palliation of right-sided congestive heart failure after stenting a recurrent cor triatriatum dexter in a 10½-year-old pug☆,☆☆

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Abstract A 10½-year-old, male neutered, pug presented with increasing ascites over two months. Echocardiography revealed cor triatriatum dexter with no concurrent cardiovascular anomalies, subsequently confirmed by computed tomography angiography. Balloon dilation of the perforated intra-atrial membrane under fluoroscopic guidance resulted in the transient resolution of all clinical abnormalities, but six months later stenosis and ascites recurred. After repeated balloon dilation, a stent was placed across the membrane. The dog remains asymptomatic fourteen months after the second procedure. One noteworthy feature of this case is the

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onset of congestive heart failure due to a congenital defect only at more than 10 years of age.

Case report

A 10½-year-old, male neutered, pug was referred to the division of cardiology with a two-month history of distended abdomen and apathy. Ascites and dilated caudal vena cava were identified prior to referral, and cor triatriatum dexter (CTD) had already been suspected echocardiographically by the referring veterinarian. The dog was sent for the evaluation of suitability for a catheter-based treatment. Abdominocentesis had not been performed before referral and was not performed at our institution. Upon presentation, the dog was bright, alert, and responsive. Severe abdominal distension with a ballotable fluid wave without jugular venous distension and unremarkable cardiac auscultation were found on physical examination. The body condition score was 3/9, the body weight was 7.2 kg, and the vital parameters were within normal limits (heart rate 90/min, respiratory rate 24/min, temperature 37.8 °C). Strong, synchronous femoral pulses were present bilaterally. Initial diagnostic testing included complete blood count, serum biochemistry profile, and transthoracic echocardiography[e]. The only laboratory abnormality was mild hypoproteinemia (51 g/l, reference >56; with normal albumin, 31 g/l). Two-dimensional echocardiography revealed the marked displacement, respectively, bowing of the interatrial septum into the left atrium and a thick perforated membrane within the right atrium. Upon color Doppler ultrasonography, a narrow jet across the perforated membrane toward the tricuspid valve was visualized (Video 1A & B). The peak velocity of this jet was 3.1 m/s (peak pressure gradient 37 mmHg using simplified Bernoulli equation). The ostium in the membrane was measured as around 2 mm. There was not any additional pathology potentially contributing to right-sided cardiac congestion such as tricuspid valve disease, pulmonic stenosis, or pulmonary hypertension. A large amount of free fluid in the abdomen as well as severely dilated caudal vena cava and hepatic veins were present. An intraluminal obstruction of the caudal vena cava, e.g. by a thrombus or a mass, was not discernible. Hence, the diagnosis of a CTD with right-sided congestive heart failure as cause of the ascites was confirmed.

In order to better characterize the CTD and to rule out any coexisting vascular pathologies potentially overlooked ultrasonographically, a helical computed tomography study[f] of the thorax and cranial aspect of the abdomen was performed, before proceeding to a therapeutic intervention. The dog was premedicated with fentanyl citrate (3 mcg/kg IV) and lidocaine hydrochloride (2 mg/kg IV). General anesthesia was induced with etomidate (0.8 mg/kg IV) and midazolam (0.2 mg/kg IV) and maintained with sevoflurane in oxygen/air via a re-breathing circuit and mechanically ventilated in synchronized intermittent mandatory ventilation-pressure control mode. Fentanyl citrate at 2.5 mcg/kg/h, dexmedetomidine at 0.5 mcg/kg/h and lidocaine hydrochloride at 50 mcg/kg/min continuous rate infusions were administered throughout all anesthesia and cefazolin (20 mg/kg IV) was administered during the procedure. On the ECG-triggered computed tomography angiography study, a thick irregular membrane was visualized dividing the right atrium into a proximal and a distal chamber (Fig. 1). A small perforation of 1.7 mm in the membrane connected the two chambers of the right atrium. There was attenuation of flow after injection into a saphenous vein but not into a cephalic vein. The blood flowed without obstruction from the distal chamber through the tricuspid valve into the right ventricle. The sinus venosus opened ventrally of the pathologic membrane in the distal chamber of the right atrium. The left atrium and the left ventricle were normal. The liver was moderately enlarged and the hepatic veins moderately congested. A moderate amount of free fluid accumulated in the abdomen. The study confirmed not only the presence of a CTD with abdominal congestion but also the absence of any additional pathology.

During the same anesthesia, a 7 Fr introducer[g] was placed into the right femoral vein by cut down which was ligated at the end of the procedure. A 5 Fr multipurpose catheter[h] was advanced to the proximal chamber of the right atrium under

[e] EPIQ 7 ultrasound system, Philips Healthcare, Horgen, Switzerland.

[f] Philips Brilliance CT 16 slice; Philips AG; 8027 Zurich, Switzerland.

[g] 5 and 7 Fr introducer, CookR Medical, CH-6002 Luzern.

[h] 5 Fr multipurpose catheter, CookR Medical, CH-6002 Luzern.

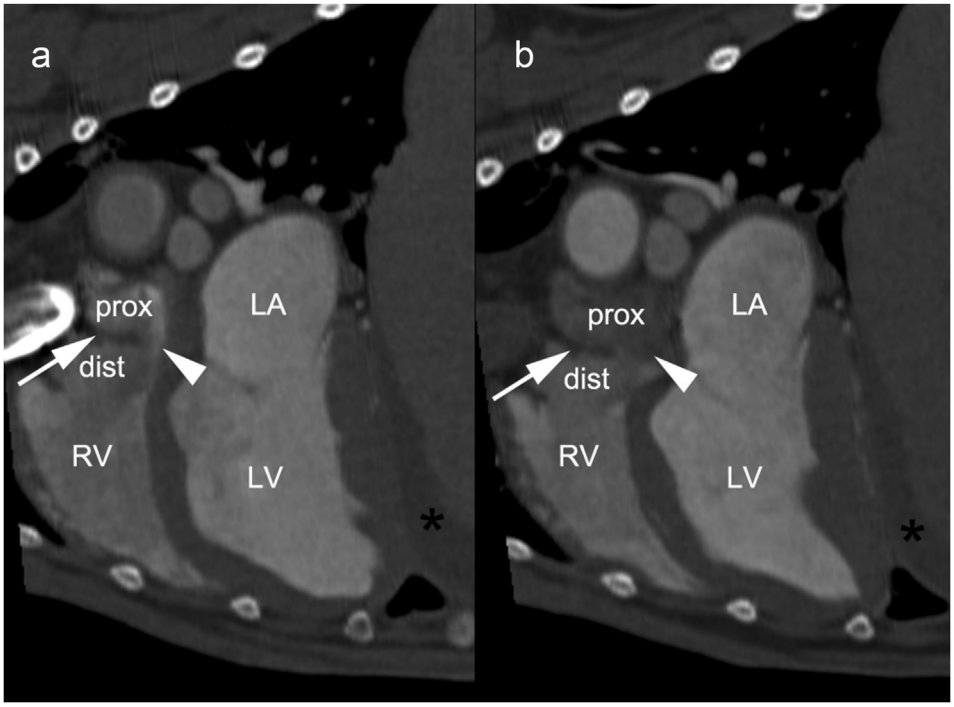

Fig. 1 Reconstruction of the ECG-triggered computed tomography angiography studies of a 10½-year-old pug with a cor triatriatum dexter in a four chamber plane obtained after injection of 5 mL contrast medium[i] diluted with 5 mL saline (a) into the cephalic vein and (b) into the saphenous vein. Monitoring the arrival of the contrast bolus was performed by bolus tracking, and the studies were started with the arrival of the contrast medium in the cranial and caudal vena cava, respectively. A thick irregular membrane (white arrow) divided the right atrium in a proximal and a distal chamber, the latter connecting with the right auricle. A small perforation in the membrane of 1.7 mm diameter (white arrow head) connected the two chambers of the right atrium. The left atrium (LA) and the left ventricle (LV) were normal. A moderate amount of free fluid (black asterisk) accumulated in the abdomen.

fluoroscopic guidance. Manual injection of a 3.5 mL bolus of iodinated, non-ionic contrast medium[i] into the proximal atrial chamber was performed to localize the stenosing membrane (Video 2). A 0.014″ guidewire[j] was advanced across the membrane to the right ventricle, and an 8 mm cutting balloon catheter[k] was placed across the membrane. Subsequently, the dilation was repeated using a 12 mm regular low pressure balloon[l] (Video 3). The waist in the balloon was completely eliminated and the inflation was maintained for around 10 s. Ballooning was repeated once and no waist was visible the second time.

The following day, peak velocity of the jet across the perforated membrane (1.8 m/s, peak pressure gradient 13 mmHg) and size of the proximal chamber had significantly decreased. Medical treatment was started with colchicine (0.5 mg, q24 h PO for 3 months), clopidogrel (18.75 mg, q24h PO for 3 months), and furosemide (20 mg am, 10 mg pm PO for 10 days). All congestive signs resolved, and one week after the intervention, the body weight was 6.6 kg.

The dog remained clinically normal for 6.5 months when gradual abdominal distention again was recognized. Two weeks later, 7 months after the first balloon dilation, the dog was re-presented. His body weight upon admission was 7.6 kg. Two-dimensional, color confirmed restenosis of the balloon-dilated membrane and recurrence of the ascites. The peak velocity of flow across the membrane was 3.1 m/s (Fig. 2a). A repeated balloon dilation but with additional placement of a stent, if deemed necessary, was recommended.

For the second intervention, the dog was premedicated with dexmedetomidine (0.25 mcg/kg IV). General anesthesia was induced with etomidate (0.8 mg/kg IV), fentanyl (1 mcg/kg IV), and midazolam (0.2 mg/kg IV) and maintained with sevoflurane in oxygen/air via a re-breathing circuit and mechanically ventilated in Synchronized Intermittent Mandatory Ventilation-Pressure Control mode. Dexmedetomidine constant rate infusion at

[i] Accupaque 350, Iohexol, GE Healthcare, CH-8152 Glattbrugg.
[j] Hi-torque extra s'port guidewire 0.014″, 190 cm, Abbott Vascular International BVBA, 1831 Diegem, Belgium.
[k] Cutting balloon catheter, 8 mm × 2.0 cm, 90 cm, 90 cm, Boston Scientific corporation, MA, USA.
[l] Veterinary balloon catheter, 12 mm, 3.0 cm, 100 cm length, Tyshak, 4.5ATM, NuMED Inc., Hopkinton, NY 12965.

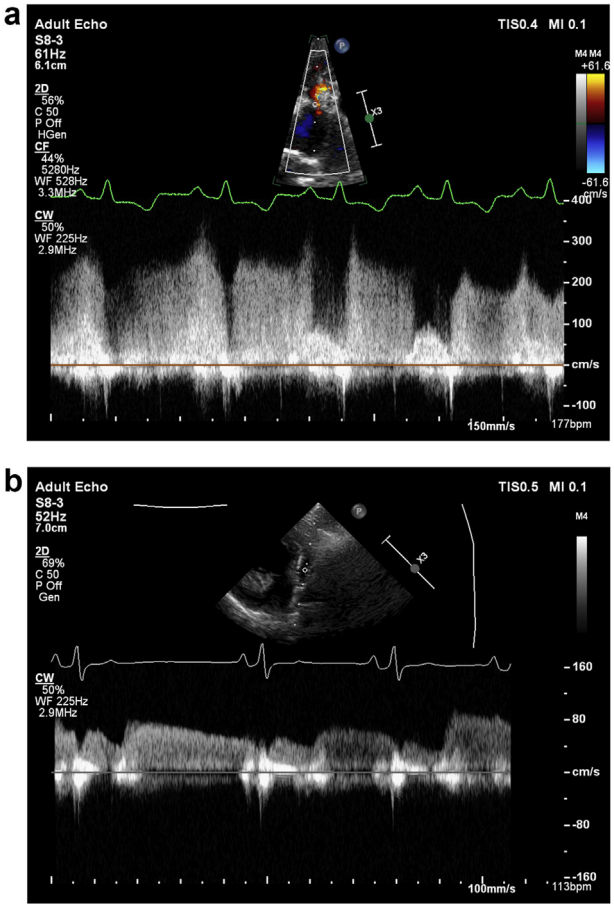

Fig. 2 Continuous wave Doppler image of a 10½-year-old pug with a cor triatriatum dexter 7 months after balloon dilation (a), and 4 months after placing a stent (b). a demonstrates high velocity (V_{max} around 3.1 m/s) multiphasic blood flow in the right atrium toward the right ventricle across a perforated membrane. b shows sustained elimination of the intra-atrial obstruction with a low-velocity blood flow (V_{max} around 0.8 m/s).

0.5 mcg/kg/h, butorphanol constant rate infusion at 0.1 mg/kg/h, and cefazolin (20 mg/kg IV) were administered during the procedure. Dobutamine and noradrenalin constant rate infusion, respectively, at 5 mcg/kg/min and at 0.1 mcg/kg/min were added for blood pressure support.

Through a 7 Fr introducer[g] placed by cut down, now in the left femoral vein ligated at the end of the procedure, a 5 Fr multipurpose catheter[h] was advanced under fluoroscopic guidance to the right heart. Invasive pressure measurements[m] revealed a pressure in the caudal vena cava of 30 mmHg. Passage through the ostium in the intra-atrial membrane proved very challenging at the second intervention. It was not possible to advance a guide wire. By rotating the multipurpose catheter with moderate pressure at the ostium, it was possible to advance the catheter across the membrane into the distal atrial chamber and right ventricle. The right ventricular pressure was 25 mmHg in systole and 0 mmHg in diastole. An 0.035″ Amplatz extrastiff guidewire[n] was placed across the membrane. The stenosis was dilated using a 15 mm regular low pressure balloon[o] and pressure was maintained for around 10 s. Similar to the first intervention, the waist in the balloon was completely eliminated after the first dilation and not waist was visible during one additional inflation. Post-dilation, the pressure in the caudal vena cava was 5 mmHg without any measurable pressure gradient across the right atrial membrane. Upon transesophageal echocardiographic color Doppler interrogation, no turbulence across the membrane was seen, however, the membrane's orifice diameter was measured as only 5 mm. To prevent a second restenosis, additionally a stent was deployed. An 8 mm balloon-expandable stent[p] was advanced to the right atrium and placed across the perforated membrane under fluoroscopic and transesophageal echocardiographic guidance. Subsequently, there was no measurable gradient across the stent post-implantation. A final angiogram revealed appropriate positioning of the stent within the right atrium with unobstructed blood flow through the stent (Video 4). The dog recovered without complications from anesthesia. A non-steroidal-anti-inflammatory drug (Robenacoxib 2 mg/kg IV once), an antibiotic (Cefazolin 20 mg/kg IV q8h for two days), and a platelet inhibitor (Clopidogrel, 18.75 mg q24h PO as continuous therapy) were prescribed. The dog was discharged four days after surgery. At this time, he had lost 1.2 kg of ascites (body weight 6.4 kg).

Upon recheck examinations 1 week and 5 months after stent placement, the dog was asymptomatic. Upon echocardiography, the stent was in situ with no interference with the tricuspid apparatus. Color and CW Doppler examination revealed a broad, laminar, low-velocity jet across the stent (Video 5, Fig. 2b). On follow-up phone interrogations 10, 14 and 16 months after stent placement, the dog was described as continuously clinically well without any signs suspicious for a

[m] Datex Ohmeda S-5 monitor, GE Healthcare, CH-8152 Glattbrugg.

[n] Amplatz extra stiff guide wire, 0.035, 260 cm, Cook Medical, CH-6002 Luzern.

[o] Veterinary balloon catheter, 15 mm, 3.0 cm, 100 cm length, Tyshak II, 3 ATM, NuMED Inc., Hopkinton, NY-12965.

[p] Valeo™ Vascular Stent, diameter 8 mm, length 26 mm, Bard Inc., New Providence, NJ, USA.

relapse. Specifically, the abdominal distention never re-occurred and body weight was stable at 6.5 kg at the last phone interview.

Discussion

In humans, CTD is reported to be an uncommon congenital malformation, but the prevalence may be underestimated because milder forms apparently remain asymptomatic and therefore undetected intra vitam. The underlying pathophysiology is persistence of the right sinus venosus valve forming a fibromuscular membrane and causing partitioning of the right atrium [1—3]. The terminology for describing the chambers in CTD is not consistent in the literature; the authors choose the terms proximal and distal chambers based on Ref. [4].

Since the first report of CTD in a dog in 1993 [5], several case reports and recently a large case series have been published [6—12]. Commonly, CTD was associated with concurrent cardiovascular anomalies. Because CTD is a congenital defect, it may be assumed to cause symptoms and be diagnosed at a young age. However, besides this dog with congestive heart failure only at 10½-years of age, five other dogs were three, six, seven, nine, and twelve years old at the time of congestion respectively diagnosis [5,7,12—14]. Why dogs are asymptomatic for years and become symptomatic with advanced age is not known. Likewise, also people may only become symptomatic at an advanced age [15]. Furthermore, in a recent review in humans with cor triatriatum sinister, the median age at diagnosis was 43 years [16]. Suggested explanations are gradual progressive fibrosis and calcification of the accessory membrane orifice induced by the mechanical stress or age-related remodeling [16]. It is conceivable that the prevalence of CTD (and cor triatriatum sinister) is underestimated also in dogs.

Since Tobias et al. [5] first reported successful resection of the obstructing membrane in a dog with CTD under inflow occlusion, several therapeutic techniques have been reported, including standard balloon dilation [11,12,17,18], cutting balloon dilation [7,13], hybrid procedure [19], direct excision of the membrane via atriotomy with inflow occlusion [9,10,20,21], or with extracorporeal circulation [22] and stent implantation [23]. As opposed to the standard balloon dilation, using a cutting balloon is thought to be a preferable approach with a lower risk of restenosis [13]. Suggested advantages of a cutting balloon compared to a regular balloon are focal rupture with less risk of dissection along an unplanned plane, less endothelial damage, and less arrhythmias [13,24]. As an attempt to minimize scar formation at the rupture site and associated restenosis, colchicine was part of the post-operative treatment in our dog. Colchicine has been used for chronic inflammatory and fibrosing processes around the heart sporadically in dogs and broadly in humans for decades [25,26]. Nevertheless, the balloon-dilated membrane in the dog of this report re-stenosed despite adjuvant colchicine treatment. A possible reason for restenosis in our dog may have been the choice of a relatively small balloon compared to other reports [12,18]. For example, a 22 mm balloon was used for 10—11 kg dogs in one report [17]. Sizing of cutting balloon based on orifice size and the stent based on caudal vena cava diameter have been recommended [27].

Restenosis is a known complication of CTD post standard and cutting ballooning dilation [7,23]. Stent implantation or direct surgical resection of the membrane via atriotomy is considered as second approach, of which the latter is distinctly more invasive.

In conclusion, stent placement is a consideration for adjunctive treatment of CTD to prevent restenosis. Furthermore, patients with a congenital CTD can develop first signs of cardiac congestion only at a very advanced age, possibly due to remodeling of the obstructing fibromuscular membrane.

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Conflicts of Interest Statement

The authors do not have any conflicts of interest to disclose.

Supplementary data

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Video	Title	Description
1	Transthoracic two-dimensional and Doppler echocardiographic images (color compare feature) of a 10½-year old pug with a cor triatriatum dexter.	Right parasternal long (A) and short axis views (B) demonstrate the interatrial septum bowing heavily into the left atrium. There is a thick perforated membrane across the right atrium with a narrow jet of blood flow. The ostium in this intra-atrial membrane measures around 2 mm. (AS = atrial septum; RAprox = proximal right atrial chamber; M, membrane across right atrium; RAdist, distal right atrial chamber; TV = tricuspid valve; LV, left ventricle; LA, left atrium; Ao, Aorta). See Supplemental Fig. IA and IB (available in Supplemental Material on-line) for labeled video still image.
2	Digital subtraction angiography of a cor triatriatum dexter in a 10½-year old pug.	A 5 Fr multipurpose catheter has been introduced into the femoral vein and advanced to the caudal vena cava and right atrium. Contrast medium (3.5 mL) injected at this site fills a proximal part of the right atrium. A perforated membrane in the right atrium markedly blocks the flow further to a distal part of the right atrium. The minimal diameter of the perforation was measured as around 1.7 mm. Subsequently the contrast medium flows into the right ventricle and out through the pulmonary trunk. (CdVC, caudal vena cava; RA, proximal right atrial chamber; M, membrane across right atrium; RAdist, distal right atrial chamber; RAA, right auricle; RV, right ventricle; PA, pulmonary artery). See Supplemental Fig. II (available in Supplemental Material on-line) for labeled video still image.
3	Fluoroscopy demonstrating balloon dilation of a cor triatriatum dexter in a 10½-year old pug.	A 12-mm veterinary valvuloplasty balloon catheter has been introduced into the femoral vein and advanced along a 0.035″ guide wire to and across a membrane in the right atrium. During inflation of the balloon a waist in the balloon caused by the obstructing membrane disappears after complete filling of the balloon.
4	Angiography of a stented cor triatriatum dexter in a 10½-year old pug.	Contrast medium (3.5 mL) injected into the proximal part of the right atrium immediately after stent placement across a balloon-dilated, perforated membrane in the right atrium flows across the stent towards the right ventricle without appreciable obstruction. (CdVC, caudal vena cava; CrVC, cranial vena cava; RAdist, distal right atrial chamber; RAA, right auricle; RV, right ventricle; PA, pulmonary artery). See Supplemental Fig. IIIA (early angiographic frame) and IIIB (late angiographic frame) (available in Supplemental Material on-line) for labeled video still image.
5	Transthoracic color Doppler echocardiography from a right parasternal short axis view in a 10½-year old pug 1 day after balloon dilating and stenting a cor triatriatum dexter.	The stent lies across a thick membrane in the right atrium without interfering with the tricuspid valve. A broad jet of blood flowing across the stent is visualized. RA, right atrium; RV, right ventricle; TV, tricuspid valve; PV, pulmonic valve

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