Arteriovenous malformation of the tongue, resulting in recurrent severe haemorrhage in a young dog

An 8-month-old male entire Beagle was presented to the emergency and critical care service following several severe bleeding episodes from the oral cavity. Oral examination revealed a purple, spongy, pulsatile lesion on the rostral two-thirds of the tongue. Computed tomography angiography revealed a severely distended right linguofacial vein with numerous, tortuous branching vessels within the tongue, consistent with an Arteriovenous (AV) malformation. A cervical surgical approach was performed and the right lingual artery was isolated and catheterised. A direct arteriogram confirmed this was the main feeder artery to the lesion, and it was ligated. Although the bleeding episodes initially resolved, a moderate bleeding episode occurred six days post-operatively and a partial glossectomy was performed. Histopathology was consistent with an AV malformation. The dog had a good recovery from surgery and remains free of clinical signs 13 months later. Following extensive review of the veterinary literature, this is the only reported case of a lingual arteriovenous malformation in the dog. Partial glossectomy resulted in resolution of the clinical signs and was well tolerated. Although rare, AV malformations should be considered as a differential diagnosis for spontaneous oropharyngeal bleeding.

Introduction

Vascular malformations are uncommon in all species and are considered a subcategory of vascular anomalies.[1,2] Several cases of Arteriovenous (AV) malformations and AV fistulae have been described in the veterinary literature.[2,8–17] Treatments reported to date include ligation of the feeding vessel, embolization of the feeding vessel with liquid, glue or a coil, or surgical resection with or without prior embolisation.[2,8–17] The following case report describes an unusual cause of spontaneous oropharyngeal bleeding in a young dog, successfully treated with complete excision of the lesion via partial glossectomy.

Case Report

An 8-month-old, male intact Beagle, weighing 11.4kg presented to our emergency and critical care service, following severe spontaneous bleeding from the oral cavity. A dark area at the rostral edge of his tongue had been noted when he was acquired by the owners at 8 weeks of age. The dog had a 5-month history of minor, spontaneous, self-resolving bleeds from the oral cavity, typically during eating or when chewing on toys. These episodes occurred approximately every 7 to 10 days, would self-resolve within 2 to 3 days and had been assumed to be caused by teething. On the morning of presentation, the dog had three major bleeds from the oral cavity and collapsed. He was taken to his referring veterinarian where initial diagnostics revealed a PCV of 14% and an in-house partial thromboplastin time and activated partial thromboplastin time within normal limits. He was given a 5ml/kg bolus of crystalloid fluids and referred for further stabilisation and investigation. On presentation to our hospital, he was quiet but responsive, with white, tacky mucous membranes. He was tachycardic (heart rate 160 beats per minute) with a grade III/VI systolic murmur. A purple/red lesion was noted at the rostral edge of the tongue. His general physical examination was otherwise unremarkable. Point of care ultrasound of the thorax and

abdomen confirmed no free pleural or abdominal fluid. His PCV was 11% and Total Solids (TS) 48g/l. Venous blood gas analysis revealed a hyperlactatemia (lactate 5.5mmol/l, reference interval: 0.6-2.5mmol/l). Haematology revealed a decreased red blood cell count (1.53 x 10^{12}/l, reference interval: 5.5-8.5 x10^{12}/l), decreased haemoglobin (2.5g/dL, reference interval: 12-18g/dL) and decreased haematocrit (8.7%, reference interval: 37-55%), with a blood smear indicating marked anaemia with strong evidence of regeneration and iron deficiency, consistent with chronic blood loss. Biochemistry revealed a decreased total protein (39.9g/l, reference interval: 54.9-75.3g/l), decreased albumin (18.8g/l, reference interval: 26.3-38.2g/l), decreased globulin (21.1g/l, reference interval: 23.4-42.2g/l) and mildly elevated urea (11.2mmol/l, reference interval: 3.1-10.1mmol/l). Angio Detect test[a] for *Angiostrongylus vasorum* was negative. SNAP 4DX[b] for *Ehrlichia canis, Ehrlichia ewingii, Borrelia burgdorferi, Anaplasma phagocytophilum, Anaplasma platys* and *Dirofilaria immitis* was also negative.

Two 10 mL/kg boluses of a balanced, isotonic crystalloid solution were given over 30 minutes and a transfusion of a single unit of canine packed red blood cells (330mls, 28.9 ml/kg) was administered based on blood type (DEA 1.1 positive). A further profuse bleeding episode occurred in the hospital several hours after admission and a second unit of packed red blood cells (290mls, 25.4ml/kg) was administered, followed by a unit of fresh frozen plasma (120mls, 10.9ml/kg). The following morning the PCV and TS were 14% and 45g/l respectively. The dog was anaesthetised and examination of the oral cavity revealed a purple, spongy, pulsatile lesion on the rostral two-thirds of the tongue, extending further caudally on the right side (Fig. 1). Computed Tomography Angiography (CTA) of the head and neck was

[a] IDEXX, Westbrook, Maine, USA
[b] IDEXX, Westbrook, Maine, USA

74 performed which showed a severely distended right linguofacial vein, measuring 6mm as it
75 branched from the jugular vein, and 13mm at its largest diameter at the caudal aspect of the
76 mandible. In contrast, the right linguofacial vein measured 1.2mm at its largest diameter.
77 Within the tongue, numerous tortuous enlarged vessels were seen (Fig. 2). Lymph nodes
78 were within normal size and no other abnormalities were visualised.
79
80 An arteriovenous malformation was diagnosed on the basis of the CTA and options for
81 further treatment were discussed with the owners. These included ligation or embolization of
82 the main feeder artery to the lesion (right lingual artery), right common carotid artery
83 ligation, or partial glossectomy. Given the caudal extent of the lesion, and the dog's age, the
84 owners elected to attempt ligation of the lingual artery as a first line treatment. An unknown
85 prognosis with this technique was given to the owners.
86
87 The dog was anaesthetised and prepared for surgery. He was placed in dorsal recumbency
88 with a sand bag under the nose and neck. The distended right linguofacial vein was easily
89 visualised sitting just under the skin. A ventral approach was made on the right medial aspect
90 of the mandible, from the mid mandible to the mid cervical region. The approach continued
91 on the medial aspect of the severely distended right linguofacial vein. This vein was carefully
92 dissected towards the caudal aspect of the incision to allow its retraction with a silicone
93 vessel loop. The right common carotid artery was isolated with silicone vessel loops at the
94 rostral and caudal aspects of the right lingual artery branch. A 20G IV catheter was inserted
95 into the lingual artery and contrast fluoroscopy using Iohexol[c] 300mgI/ml was performed,
96 confirming this vessel to be the main feeder artery to the malformation. This vessel (the right

[c] Omnipaque™; GE Healthcare, Illinois, Chicago, USA

lingual artery) was ligated with two 3-0 polypropylene[d] and two 3-0 Polydiaxonone[e] sutures. A routine closure was then performed.

The dog recovered well from surgery and initially no bleeding episodes occurred. He was discharged two days post-operatively with a 5-day course of oral paracetamol (10 mg/kg q12 hr), soft food and monitoring instructions.

Six days post-operatively a mild amount of bleeding occurred when the dog was found to be chewing his bed which quickly resolved, but led to multiple small bleeds throughout that day. Eleven days postoperatively a severe oral bleeding episode occurred and the dog was re-admitted. On presentation his PCV was 10% and TS 48g/l. A transfusion of canine packed red blood cells was administered after a cross match had been performed. After discussion with the owners, a partial glossectomy was performed the following day. The patient was anaesthetised and placed in sternal recumbency with the maxilla suspended between two drip stands. A V-shaped incision was made in the tongue, removing the grossly abnormal areas; this resulted in removal of approximately two thirds of the tongue. The dorsal and ventral mucosal edges were apposed with simple interrupted 3-0 Polyglactin 910[f] sutures (Fig. 3).

The resected tissue was submitted for histological evaluation and the report indicated an extensive vascular proliferation in the subepithelial stroma, with focal epithelial ulceration and adjacent vascular thrombosis. Microscopic examination confirmed the presence of numerous variably sized, tortuous arterioles and venules, admixed with clusters of capillary

[d] Prolene; Ethicon, Sommerville, New Jersey, USA
[e] PDS; Ethicon, Sommerville, New Jersey, USA
[f] Vicryl; Ethicon, Sommerville, New Jersey, USA

vessels, consistent with an arteriovenous malformation or less likely, a haemangioma (Fig. 4).

The dog remained in the hospital for 7 days post-operatively to ensure adaption to eating was sufficient prior to discharge. The partial glossectomy was well tolerated and at the time of discharge he was able to eat soft food balls from a raised bowl and drink from a deep bowl. At re-examination 1 month post-operatively the dog was reported to be doing well. Eating and drinking abilities had improved with continuation of offering from raised bowls. Halitosis had been noted by the owners and some tartar was noted on the teeth. The only abnormality reported by the owners was saliva leaking from the oral cavity. No abnormalities were present on general physical examination and on conscious oral examination the surgical site had healed well. By 3 months post-operatively, the owners reported that the dog was eating and drinking well with self-adaption of throwing balls of food into the back of his mouth and swallowing. He was reported to be bright, lively and back to behaving like a normal puppy, with resolution of the previous saliva leakage. At 13 months post-operatively the owner reported no further bleeding episodes and an excellent quality of life.

Discussion

Vascular malformations are a subcategory of vascular anomalies, with the other major subcategory being neoplastic or neoplastic-like lesions, often called haemangiomas.[1,2] Vascular malformations are always present at birth, grow with the individual and never regress.[3] Conversely, haemangiomas are not present at birth, grow rapidly during a proliferative phase, go through a period of stabilisation and then involute.[3] Vascular malformations are composed of mature endothelium that does not undergo tumour-like

endothelial proliferation. Glucose transporter protein (GLUT)-1 immunostaining has been used in human pathology to differentiate these from haemangiomas.[4]

Several classification systems have been described in the human literature to categorize a variety of documented vascular malformations. The modified Hamburg classification system is thought to be the most accepted system and considered a primary classification based on whether a malformation is capillary, venous, arterial, lymphatic or combined vascular.[5] Sub classification is then established based on embryological characteristics, with the two major categories being extratruncular and truncular.[5] Extratruncular and truncular forms are characterised based on the time at which the defect occurred in embryogenesis; extratruncular lesions arise when developmental arrest occurs in the earlier stage of embryonic life, when the vascular system is in the reticular stage. Truncular forms occur when developmental arrest occurs at the vascular trunk formation period, at a later stage of embryonic development.[5] Arteriovenous malformations that are truncular often result in aplasia or obstruction, whereas extratruncular forms are infiltrating.[5] Another classification scheme complimentary to the endothelial classification system classifies AV malformations based on the identification of their rate of blood flow.[6] In that scheme, vascular malformations are classified into low-flow (venous) and high-flow (arteriovenous) malformations.[6] Although a classification system for vascular malformations in dogs has not been reported, some of the principles of the human classification system can be applied to this case. Based on these, the vascular anomaly seen in the dog described here would be classified as an AV malformation with high flow rates and extratruncular characteristics due to its infiltrative nature. In humans, the importance of classification lies in the ability to help guide clinical decision making; transcatheter embolization or surgical resection is the treatment of choice for high-

flow AV malformations, whereas percutaneous treatment with a sclerosing agent is recommended for low-flow vascular malformations.[7]

Congenital hepatic AV malformations are the most commonly reported AV malformations in the veterinary literature, but acquired cases secondary to trauma (surgical and other forms), and neoplasia have also been reported.[2,8,17,9–16] Treatments reported to date include ligation of the feeding vessel, embolization of the feeding vessel with liquid, glue or a coil, or surgical resection with or without prior embolisation.[2,8–17] Embolization was discussed as a treatment option for this case as it has been successfully performed in veterinary species for treatment of AV malformations.[2,13,16,18] Although transcatheter embolization is the treatment of choice for high flow AV malformations in humans,[7] embolization alone has not been found to be curative in certain cases.[4] A review of the literature regarding lingual vascular malformations in humans suggests that surgical resection with or without prior transcatheter embolization to reduce intra-operative haemorrhage, would be the treatment of choice for the majority of cases.[4]

In the case reported here, ligation of the lingual artery did not result in medium- or long-term resolution of the clinical signs. This was not completely unexpected due to the fact that the right and left lingual arteries anastomose throughout the parenchyma of the tongue muscle. Indeed, disruption of flow through one artery has been shown to have no significant effect on blood flow.[19] However, surgical resection via partial glossectomy is an invasive surgery and can have a significant impact on quality of life. Given the dog's young age, the owners elected to try a less invasive option first. Although this did not resolve the symptoms other than in the very short term, partial glossectomy has resulted in a good long-term outcome to date.

AV malformations of the tongue have not been previously reported in veterinary patients. A single case series describing arteriovenous haemangiomas in two dogs and one cat, reported one case of a lingual haemangioma that was managed by surgical resection.[20] The lesions described in that case series were focal masses seen in mature animals. In contrast, the case described here was seen in a very young animal and was much more locally extensive, suggestive of an AV malformation rather than a haemangioma.

Arteriovenous malformations of the tongue are rarely reported in humans and are often compartmentalised into the context of larger head and neck vascular malformations.[4] A recent case series describes 11 lingual AV malformations in humans, four with evidence of upper neck and mouth involvement and seven focal lingual masses.[4] Presenting clinical signs included sudden enlargement of a previously detected lesion and spontaneous bleeding.[4] Age of presentation for the focal AV malformations ranged from six months to 41 years, with the majority of lesions described as hyperaemic and firm with a clear border.[4] They were found to have a single feeder lingual artery on angiography in the majority of cases, similar to the one described here, and were all treated with surgical resection of the affected tissue, with only one case having embolization of the feeder vessel preoperatively, (in order to reduce intraoperative bleeding).[4] The more complex malformations involving the neck and mouth required extensive surgery for removal of the affected tissue, with a mean operating time of 11 hours.[4] Two out of four cases experienced recurrence within two months, requiring further surgical intervention.[4]

Conclusion

Although rare, AV malformations should be considered as a differential diagnosis for spontaneous oropharyngeal bleeding, especially in younger dogs.

References

1. Lidsky ME, Markovic JN, Miller MJ, et al. Analysis of the treatment of congenital vascular malformations using a multidisciplinary approach. J Vasc Surg 2012;56(5):1355–62.
2. Culp WTN, Glaiberman CB, Pollard RE, et al. Use of ethylene-vinyl alcohol copolymer as a liquid embolic agent to treat a peripheral arteriovenous malformation in a dog. J Am Vet Med Assoc 2014;245(2):216–21.
3. Burrows P, Mulliken J, Fellows K, et al. Childhood hemangiomas and vascular malformations: angiographic differentiation. Am J Roentgenol 1983;141(3):483–8.
4. Richter GT, Suen J, North PE, et al. Arteriovenous malformations of the tongue: A spectrum of disease. Laryngoscope 2007;117(2):328–35.
5. Lee BB, Laredo J, Lee TS, et al. Terminology and classification of congenital vascular malformations. Phlebology 2007;22(6):249–52.
6. Jackson IT, Carreño R, Potparic Z, et al. Hemangiomas, vascular malformations, and lymphovenous malformations: classification and methods of treatment. *Plast* Reconstr Surg 1993;91(7):1216–30.
7. Sadick M, Wohlgemuth WA, Huelse R, et al. Interdisciplinary Management of Head and Neck Vascular Anomalies: Clinical Presentation, Diagnostic Findings and Minimalinvasive Therapies. Eur J Radiol open 2017;4:63–8.
8. Saunders AB, Fabrick C, Achen SE, et al. Coil Embolization of a Congenital Arteriovenous Fistula of the Saphenous Artery in a Dog. J Vet Intern Med 2009;23(3):662–4.
9. Hyndman PS, Worth AJ, Owen MC, et al. Peripheral arteriovenous fistula manifesting as antebrachial dermatopathy in a cat. J Am Vet Med Assoc 2019;254(3):393–8.
10. Aiken SW, Jakovljevic S, Lantz GC, et al. Acquired arteriovenous fistula secondary to

castration in a dog. J Am Vet Med Assoc 1993;202(6):965–7.

11. Butterfield AB, Hix WR, Pickrel JC, et al. Acquired peripheral arteriovenous fistula in a dog. J Am Vet Med Assoc 1980;176(5):445–8.

12. Tobias KM, Cambridge A, Gavin P. Cyanoacrylate occlusion and resection of an arteriovenous fistula in a cat**.** J Am Vet Med Assoc.2001;219(6):785–8, 763.

13. Chanoit G, Kyles AE, Weisse C, et al. Surgical and interventional radiographic treatment of dogs with hepatic arteriovenous fistulae. Vet Surg 2007;36(3):199–209.

14. Easley JC, Carpenter JL. Hepatic arteriovenous fistula in two Saint Bernard pups**.** J Am Vet Med Assoc 1975;166(2):167–71.

15. Sherman A, Kim S, Craft W, et al. Micro-arteriovenous malformation causing spontaneous metacarpal pad hemorrhage in a dog. Can Vet J 2018;59(6):659–62.

16. Eason BD, Hogan DF, Lim C, et al. Use of n-butyl cyanoacrylate to reduce left to right shunting of an abdominal arteriovenous malformation in a dog. J Vet Cardiol 2017;19(4):396–403.

17. Proks P, Stehlik L, Paninarova M, et al. Congenital subcutaneous arteriovenous malformation in a puppy: diagnosis with CT angiography. Vet Dermatol 2015;26(5):384–6.

18. Saunders AB, Fabrick C, Achen SE, et al. Coil Embolization of a Congenital Arteriovenous Fistula of the Saphenous Artery in a Dog. J Vet Intern Med 2009;23(3):662–4.

19. Anderson GM. Soft Tissues of the Oral Cavity. In: Johnstone SA, Tobias KM, eds. Veterinary Surgery. 2nd ed. Missouri: Elsevier, 2018:1637–52.

20. Schöniger S, Tivers MS, Baines SJ, et al. Arteriovenous Haemangioma in Two Dogs and a Cat. J Comp Pathol 2008;139(2–3):130–6.

Figure legends

Figure 1: The dorsal aspect of the tongue showing the lesion on the rostral two-thirds, extending caudally on the right side

Figure 2: A dorsal plane 3D volume-rendered Computed Tomography Angiography (CTA) image of the severely distended right linguofacial vein (*) as it branches from the right external jugular vein (†) and the tortuous branching vessels within the tongue (diamond). The normal left linguofacial vein (open arrowhead) and the internal jugular veins (solid arrowheads) are labelled.

Figure 3: Image showing the dorsal surface of the tongue following glossectomy as the final sutures are being placed

Figure 4: Dorsal aspect of the tongue showing increased numbers of variably-sized, irregular, blood-containing vascular spaces lined by flat endothelium including arterioles (*), venules (†) and capillaries (‡) (H&E stain; bar = 50μm)