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CASE REPORT

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Spinal epidural arteriovenous fistula in 3 cats

Naoko Yayoshi[1] | Karin Yoshida[1] | Masashi Terao[2] | Tsuyoshi Ozawa[2] | Hiroaki Kamishina[2] | Fumitaka Takahashi[3,4] | Yasushi Hara[4] | Yoshihiko Yu[5] | Daisuke Hasegawa[5]

[1]Veterinary Medical Teaching Hospital, Nippon Veterinary and Life Science University, Tokyo, Japan

[2]Kyoto Animal Referral Medical Center, Kyoto, Japan

[3]YPC Tokyo Animal Orthopedic Surgery Hospital, Tokyo, Japan

[4]Laboratory of Veterinary Surgery, Nippon Veterinary and Life Science University, Tokyo, Japan

[5]Laboratory of Veterinary Radiology, Nippon Veterinary and Life Science University, Tokyo, Japan

Correspondence
Daisuke Hasegawa, Laboratory of Veterinary Radiology, Nippon Veterinary and Life Science University, 1-7-1 Kyounan-cho, Musashino, Tokyo 180-8602, Japan.
Email: disk-hsgw@nvlu.ac.jp

Abstract

Three young adult cats with intermittent spinal hyperesthesia and paraparesis and diagnosed with spinal epidural arteriovenous fistula are described. In all 3 cases, magnetic resonance imaging (MRI) showed focal dilatation of the veins in the epidural space of the thoracic spinal cord, whereas computed tomography angiography (CTA) showed dilatation and enhancement from the intercostal vein to the azygos vein at the same site in the arterial phase. Dorsal laminectomy and occlusion of the interarcuate branches running across the dorsal aspect of the spinal cord were performed in all 3 cats to decompress the spinal cord, which resulted in a remission of clinical signs and no recurrence during 14 to 40 months of follow-up after surgery in all cases.

KEYWORDS
CT, CT angiography, MRI, spinal arteriovenous malformation, spinal vascular malformation

1 | INTRODUCTION

In humans, spinal arteriovenous malformations (AVMs) are rare conditions defined as a heterogeneous group of spinal vascular malformations characterized by an abnormal tangle of blood vessels in or around the spinal cord.[1] Cases of vertebral vascular malformations, such as angiomatosis and hamartomas, have been reported sporadically in cats.[2-14] However, spinal AVMs have not been reported in cats.

We report 3 young adult cats with abnormal hindlimb gait and spinal hyperesthesia that were diagnosed by magnetic resonance imaging (MRI) and computed tomography angiography (CTA) to have spinal epidural arteriovenous fistula (AVF), a subtype of AVM.[1] The cats showed clinical improvement after surgical treatment.

Abbreviations: AVF, arteriovenous fistula; AVM, arteriovenous malformation; CT, computed tomography; CTA, CT angiography; MRI, magnetic resonance imaging; NSAIDs, nonsteroidal anti-inflammatory drugs; STIR, short tau inversion recovery; T1WI, T1-weighted images; T2WI, T2-weighted images.

2 | CASE DESCRIPTIONS

Detailed information on the imaging equipment and conditions and hospitals is presented in Table S1, Supporting Information.

2.1 | Case 1

A 14-month-old intact female domestic cat had acute paraparesis and spinal hyperesthesia of 4 days' duration. Despite initial improvement with nonsteroidal anti-inflammatory drugs (NSAIDs), clinical signs worsened. At the time of referral, the cat had nonambulatory paraparesis, and palpation evoked severe hyperesthesia in the dorsal thoracolumbar

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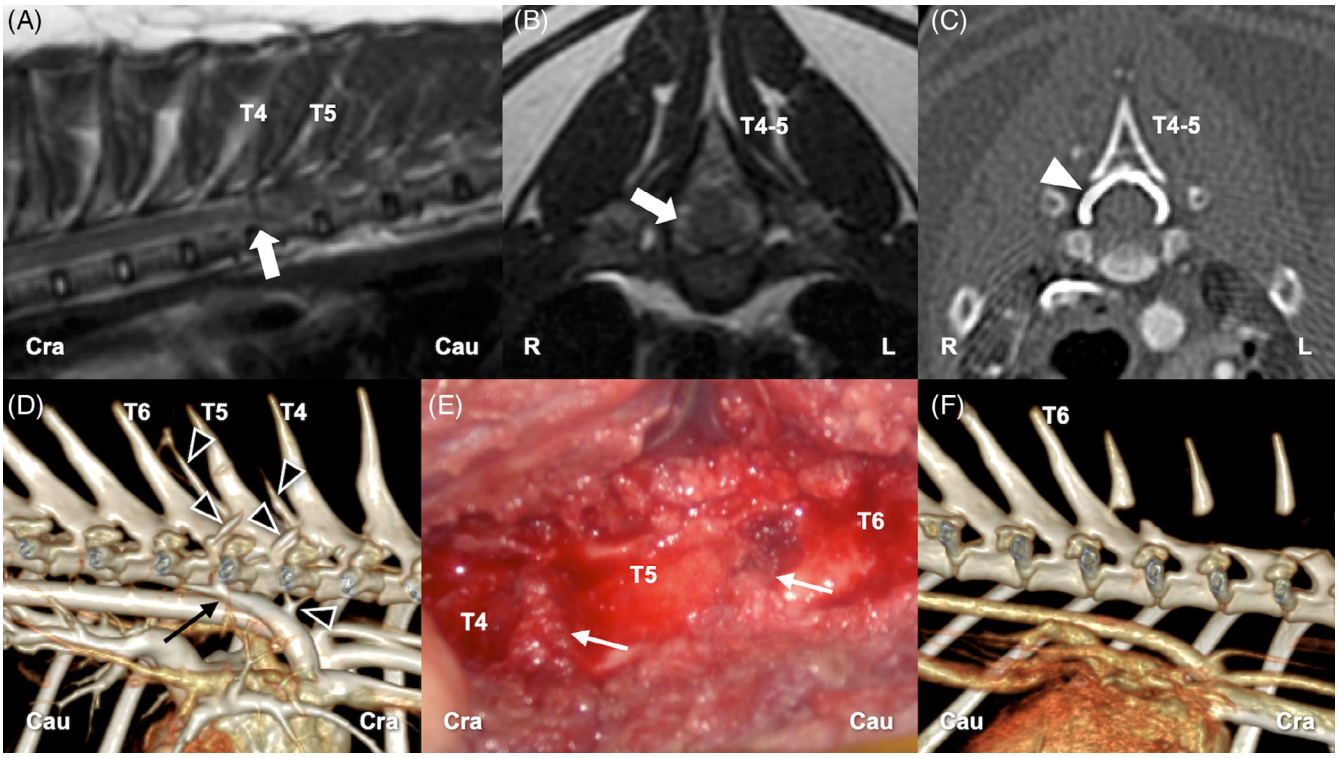

FIGURE 1 Case 1. (A) Sagittal T2-weighted MRI at the level of T4-T5 intervertebral foramen showing an abnormal hypointense extradural linear structure (white arrow) running dorsoventrally across the lateral aspect of the spinal cord. (B) On a transverse T2-weighted MRI at the level of the T4-T5 intervertebral foramen, a slightly hyperintense (relative to the spinal cord parenchyma) and blurred extradural structure (white arrow) surround the spinal cord and the normal circumferential T2 hyperintense CSF/epidural fat signals are disappeared. (C) On the transverse images in the arterial phase of CT angiography, the extradural structure in (B) was recognized as a dilated vessel crossing the dorsal aspect of the spinal cord (white arrowhead). (D) On the right lateral view of the preoperative 3D-rendering CT, the contrast enhancement of veins surrounding the vertebral column (black arrowheads outlined in white) and the azygos vein (black arrow) are seen. (E) During the dorsal laminectomy, two congested blood vessels were found on the dorsal aspect of the spinal cord at the level of T4-T5 and T5-T6 intervertebral foramina. (F) On the postoperative 3D-rendering CT, the contrast enhancement of veins surrounding the vertebral column and the azygos vein seen in the preoperative image (D) has disappeared

region. On neurological examination, conscious proprioception and muscle tone in all 4 limbs seemed to be normal. However, myotatic reflexes were slightly increased with normal flexor reflex in the hindlimbs suggesting a T3-L3 spinal cord lesion. Complete blood count, serum biochemistry, urinalysis, abdominal ultrasonography, and survey radiography (from the cervical to thoracolumbar region) disclosed no abnormalities.

On MRI, the sagittal T2-weighted images (T2WI) showed a slightly T2-hypointense (relative to the spinal cord parenchyma) extradural linear structure running dorsoventrally across the lateral aspect of the spinal cord at the level of the T4-T5 intervertebral foramen (Figure 1A), and the transverse T2WI in the same region showed a slightly T2-hyperintense and blurred extradural structure surrounding the spinal cord that had lost the normal circumferential T2-hyperintensity from cerebrospinal fluid (CSF) or epidural fat (Figure 1B).

Concurrently obtained precontrast computed tomography (CT) showed no morphological abnormalities in the entire vertebral column. However, in the arterial phase of CTA, 2 contrast-enhancing and dilated vessels (suspected to be interarcuate branches) were visible on the left and right lateral and dorsolateral aspects of the spinal cord at the level of the T4-T5 (Figure 1C) and T5-T6 intervertebral foramina. The nearby interspinous, intervertebral, and intercostal veins were similarly contrast enhancing, and contrast medium was flowing from these vessels into the azygos vein at the level of the T6 vertebra (Figure 1D).

Based on these findings, a clinical diagnosis of spinal epidural AVF was made, and we postulated that the interarcuate branches dilated by the shunt caused compression of the spinal cord and led to neurological signs in the cat.

Dorsal laminectomy was performed over the T3-T5 vertebrae to decompress the spinal cord. During the surgery, 2 distended interarcuate branches crossing the dorsal dura of the spinal cord at the T4-T5 and T5-T6 intervertebral regions were identified, and these vessels were ligated and transected (Figure 1E). After surgery, CTA of the arterial phase was performed again. The vascular structures around the vertebral column had disappeared, and no enhancement of the azygos vein was observed (Figure 1F).

Nonambulatory paraparesis began to improve during the first 3 days after surgery, and by the fifth postsurgical day, the cat was able to take several steps on its hindlimbs without hyperesthesia. On the 14th day, the cat was able to jump up onto a bed and couch. During

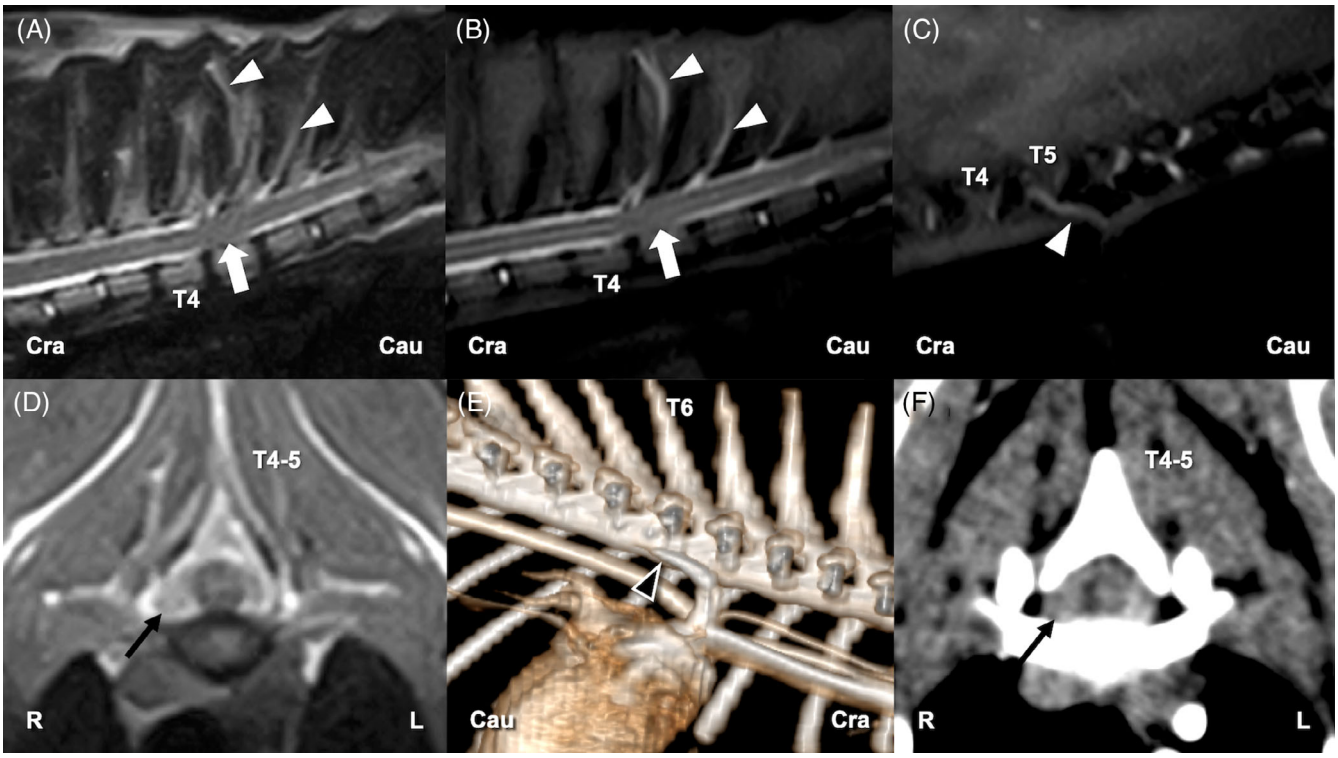

FIGURE 2 Case 2. (A) Midline sagittal T2-weighted and (B) short tau inversion recovery MRI shows the loss of the subarachnoid space from the level of the T4-T5 intervertebral foramen to T5 vertebrae (white arrow) and dilatation of the interspinous veins adjacent to the dorsal spinous processes of T4, T5, and T6 vertebrae (white arrowheads). (C) On the right-sided parasagittal short tau inversion recovery image, a dilated intercostal vein from the T4-T5 intervertebral foramen is also observed (white arrowhead). (D) A transverse plane of postcontrast T1-weighted MRI shows dilatation of the internal vertebral plexus at the level of the T4-T5 intervertebral foramen (black arrow). (E) The right lateral view of the 3D-rendering CT angiography in the arterial phase shows dilatation and contrast enhancement of the azygos vein cranial to T6 vertebrae (black arrowhead outlined in white). (F) A transverse plane of the venous phase of CT angiography at the level of the T4-T5 intervertebral foramen reflects the finding of MRI at the same level (D)

the 40-month postoperative follow-up period, the cat had no difficulty with everyday activities (such as walking and jumping) and no signs of recurrence were observed.

2.2 | Case 2

A 3 years and 11 months old neutered male domestic cat showed a few days' history of decreased activity and appetite, delayed walking speed, and increased vocalizing when stepping on its forelimbs or being held up. Complete blood count, serum biochemistry, survey radiography from the cervical to the lumbar regions, and abdominal ultrasonography showed no abnormalities. No improvement was observed with the use of NSAIDs or prednisolone. At the time of referral, neurological examination identified no abnormalities.

Magnetic resonance imaging showed disappearance of the subarachnoid space surrounding the spinal cord at the level of the T4-T5 intervertebral foramen on sagittal T2WI (Figure 2A) and dilatation of the interspinous veins adjacent to the dorsal spinous processes of T4-T6 vertebrae, as well as dilatation of the fourth intercostal vein on sagittal short tau inversion recovery (STIR) images (Figure 2B,C). Moreover, the transverse T2WI, T1-weighted images (T1WI), and postcontrast T1WI at the level of the T4-T5 intervertebral foramen showed dilatation of the internal vertebral venous plexus (Figure 2D).

Computed tomography angiography showed dilatation and enhancement of the right fourth intercostal vein and azygos vein cranial to the T6 vertebra in the arterial phase (Figure 2E). In addition, the internal vertebral venous plexus at the level of T4-T5 intervertebral foramen was dilated and mildly contrast-enhancing during the venous phase through the delayed phase (Figure 2F). No morphological abnormalities of the vertebral column were found. Based on these findings, congestion of the medial vertebral venous plexus caused by spinal epidural AVF was suspected, and dorsal laminectomy of T5 vertebra was performed. During surgery, as in case 1, a distended interarcuate branch was observed on the dorsal aspect of the spinal cord, which was ligated and transected. Postoperative CTA showed persistent enhancement of the right fourth intercostal vein and the azygos vein and dilatation of the internal vertebral venous plexus. However, no new compressive vascular structures were visible anywhere after ligation of the distended interarcuate branch.

Within 2 weeks after surgery, the cat's walking speed and hyperesthesia improved and it became able to jump to an increased height. No signs of recurrence were observed in the cat during a 16-month postoperative follow-up period.

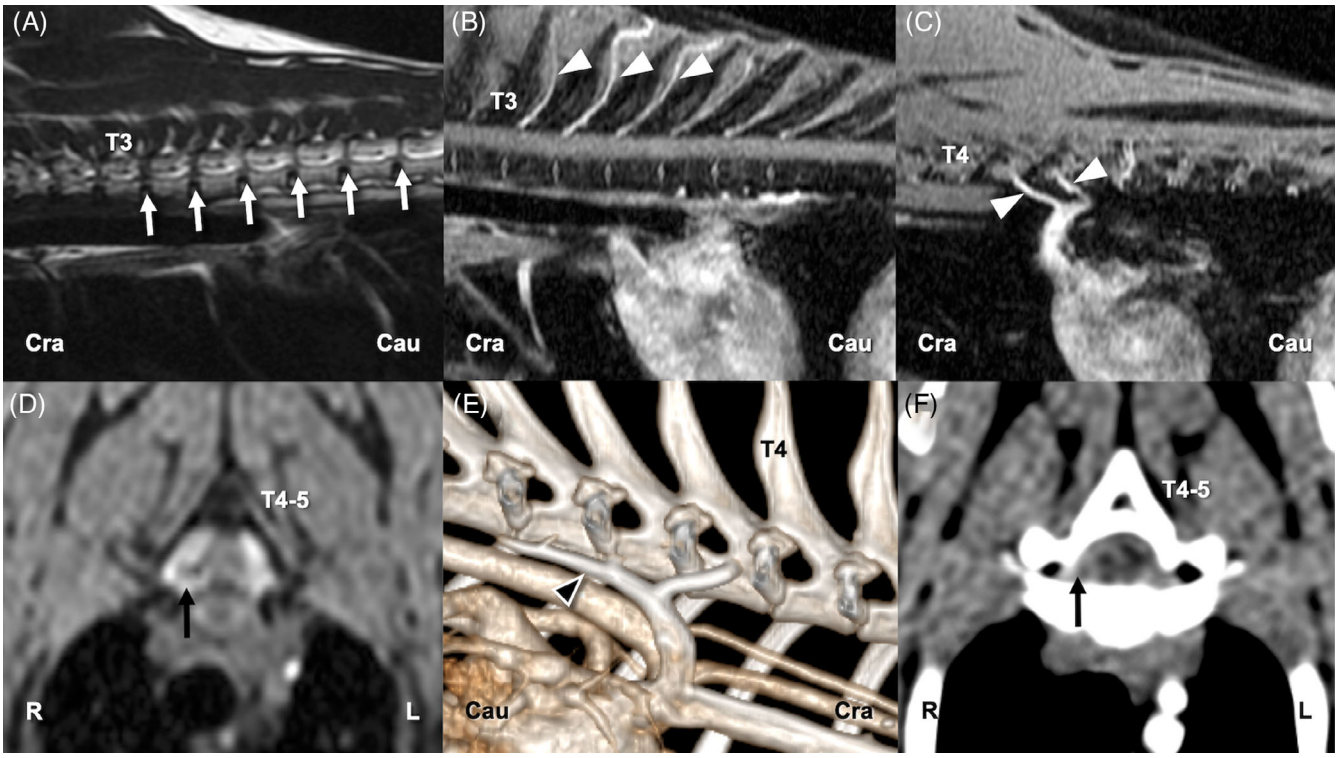

FIGURE 3 Case 3. (A) On a parasagittal T2-weighted image, multiple hypointense dorsoventral linear structures (white arrows) are observed at the level of consecutive intervertebral foramen spaces between T3 and T9 on the lateral aspect of the spinal cord. (B) Midline sagittal and (C) right-sided parasagittal planes of postcontrast fat-suppressed T1-weighted MRI show dilatation of the interspinous veins between T3 and T6 (B; white arrowheads) and the fourth and fifth intercostal veins (C; white arrowheads), respectively. (D) Transverse planes of postcontrast fat-suppressed T1-weighted MRI at the level of the T4-T5 intervertebral foramen reveals a dilated suspected interarcuate branch and internal vertebral venous plexus (black arrow) compressing the spinal cord. (E) The right lateral view of 3D-rendering CT angiography in the arterial phase shows dilatation and contrast enhancement of the fourth intercostal vein and the azygos vein cranial to T7 vertebra (black arrowhead outlined in white). (F) Transverse image of the venous phase of CT angiography also shows dilatation of the internal vertebral venous plexus at the level of the T4-5 intervertebral foramen (black arrow)

2.3 | Case 3

A 2-year-old, neutered, male domestic cat had a 1-month history of spinal hyperesthesia and hindlimb weakness when waking up or starting to move. Survey radiography from the cervical to lumbar regions did not show any abnormalities, and NSAIDs did not provide any improvement. Neurological examination identified no abnormalities, and CBC and serum biochemistry profile also did not show any clinically relevant changes.

On MRI, bilateral parasagittal T2WI disclosed multiple dorsoventral linear extradural structures with T2-hypointensity (relative to the spinal cord parenchyma) at the level of consecutive intervertebral foramina between T3 and T9 vertebrae on the left and right aspects of the spinal cord (Figure 3A). Sagittal postcontrast T1WI with fat suppression showed dilatation of the interspinous and intercostal veins in the range T3 to T6 vertebrae (Figure 3B,C). Additionally, a dilated suspected interarcuate branch and internal vertebral venous plexus compressing the spinal cord were recognized at the level of T4-T5 intervertebral foramen (Figure 3D).

Computed tomography angiography disclosed dilatation and enhancement of vascular structures from the right fourth to sixth intercostal veins to the azygos vein in the arterial phase (Figure 3E). In addition, CTA indicated that the dilated internal vertebral venous plexus at the level of the T4-T5 intervertebral foramen was slightly contrast-enhancing during the venous phase through the delayed phase (Figure 3F).

Based on these findings, spinal epidural AVF and secondary congestion of the medial vertebral venous plexus and interarcuate branch were diagnosed. Dorsal laminectomy of T4 and T5 vertebrae was performed and an interarcuate branch extending dorsal to the spinal cord was recognized, ligated, and transected. Postoperative CTA indicated that contrast enhancement of intercostal veins and the azygos vein in the arterial phase persisted. However, no new compressive vascular structure developed at other sites after transection of the interarcuate branch.

The cat was discharged on the third day after surgery and had no signs of spinal hyperesthesia during the following 2 weeks. No recurrence of clinical signs occurred during a 14-month postoperative follow-up period.

3 | DISCUSSION

In human medicine, several classification systems of spinal AVM have been proposed, with changes resulting from the development of

diagnostic techniques and treatment procedures.[15] Five types of AVM occur in humans according to the latest classification: dural AVFs, intramedullary glomus AVMs, intramedullary juvenile AVMs, perimedullary AVFs, and epidural AVFs.[15]

Epidural AVFs are defined as abnormal direct connections between the segmental artery and epidural venous plexus within the vertebral canal, intervertebral foramen or both.[16] These AVFs can cause neurological deficits because of compression of the spinal cord or spinal nerve roots by the dilated epidural venous plexus, or spinal cord congestion because of backflow into the intradural venous plexus.[16]

All 3 cats in our report were considered to have epidural AVFs according to the AVM classification in humans.[15] A previous case report described a cat with myelopathy secondary to an aortocaval fistula. In that report, a dilated vascular complex in the retroperitoneal space induced the engorged internal vertebral venous plexus that caused extensive bilateral spinal compression.[17] However, no reports of epidural AVFs in cats that have different anatomical and pathological features from the abovementioned report have been published.[17]

In all 3 cats described here, the age of clinical onset was between 1 and 3 years, and the lesions occurred in the upper thoracic vertebral column. According to a systematic review, including 45 cases of epidural AVF in humans,[16] epidural AVFs without reflux into the intradural veins frequently are observed in patients during their thirties, and the cervical and upper thoracic regions of the spinal cord are the most commonly affected areas, which seems similar to our cats. Although the systematic review did not discuss why the cervical spinal cord and upper thoracic spinal cord were frequently involved,[16] the anatomical features of the interarcuate branches may have contributed to the predominance of lesions in the thoracic spinal cord, especially at the level of the T4-5 intervertebral foramen, in all 3 cats in our report. In dogs, the interarcuate branches are veins belonging to the internal vertebral venous plexus, which are best developed in the cervical and thoracic areas, and the left and right interarcuate branches frequently anastomose at the dorsal cranial part of each spinal cord segment within the vertebral column.[18] To the best of our knowledge, the vascular anatomy of the spinal cord in cats has not been described in detail. However, based on the MRI and CT findings in our 3 cases, they seem to have similar anatomy to dogs in the region, and the interarcuate branches anastomose especially in areas around the level of the T4-5 intervertebral foramen. As for the cause of epidural AVF in humans, most cases are considered to be congenital, although some reports suggest an association with surgery.[1,16] In the present report, none of the 3 cats had a history of surgery or trauma. Therefore, we consider that their condition was probably congenital in origin and subclinical at the early stage of their lives. However, a possible hypothesis in young adult cats is that when they become more active or their blood pressure increases, a persistent increase in venous pressure causes gradual dilatation of the interarcuate branches and the internal vertebral venous plexus with compression of the spinal cord and clinical signs.

Interestingly, vertebral angiomatosis and vertebral vascular hamartoma in cats previously reported as vertebral vascular malformations also occurred most commonly in the thoracic area at 1 to 2 years of age,[2-14] which corresponds to the young adult, similar to the age of our cats. Although they were histologically characterized as benign vascular and bony proliferations and the condition was distinct from spinal AVM, spinal vascular malformations in cats may tend to occur in the thoracic vertebral column.

In human medicine, although digital spinal angiography is considered the gold standard for anatomical assessment of spinal AVM,[19] CTA also is considered a valuable diagnostic technique because it detects the feeding arteries and draining veins.[20] In all cases described here, the arterial phase of CTA commonly showed dilatation and contrast enhancement of veins within and surrounding the vertebral column, such as the interarcuate branches; interspinous, intervertebral, and intercostal veins; and the azygos vein cranial to its influx. This finding indicates that the contrast medium flowed from the arteries surrounding the vertebral column directly into the epidural or perivertebral veins, suggesting the presence of spinal epidural AVF. Similar findings have been described in a case report of spinal epidural AVF in a dog, in which the diagnosis of AVF was supported by angiography using an intravascular catheter.[21] Furthermore, in our 3 cases, sagittal T2WI obtained before CTA showed partial loss of CSF signal in the subarachnoid space on the lateral aspect of the spinal cord because of dilated interarcuate branches. Moreover, T2WI, STIR, or postcontrast T1WI with fat suppression showed findings suggestive of dilatation of the surrounding interarcuate branches, internal vertebral venous plexus, or intercostal veins. These findings are consistent with congestion of the venous system secondary to epidural AVF and may be useful to establish the diagnosis.

Epidural AVF in humans is treated by surgery or endovascular embolization (or a combination of both) to occlude the shunt vessel.[19] In all of cats described here, dorsal laminectomy and occlusion of the dilated interarcuate branches were performed to relieve spinal cord compression because CTA could not identify the feeding artery and it was technically difficult to perform endovascular treatment considering the size of cats. On the other hand, because the postoperative CTA in the arterial phase showed the disappearance of the azygos vein in case 1 but not in cases 2 and 3, we considered the shunt vessel to have been occluded in case 1, whereas the shunt remained in cases 2 and 3. However, the clinical signs most likely improved because of the decompression of the spinal cord, and recurrence was not observed in those 2 cases.

It has been reported in humans that the complete occlusion rate by surgical ligation is approximately 75%, which tends to require endovascular treatment or combined treatment with vertebrectomy because epidural AVF without reflux into the intradural vein forms a complex anastomosis with abundant blood flow between the artery and the epidural vein.[16,22] In our cats, dilated veins in the epidural space at the level of multiple thoracic vertebrae were observed, but not all of the feeding arteries and draining veins were identified by CTA. Thus, occlusion of a dilated interarcuate branch in a situation where other shunts may be present could increase the surrounding venous pressure, leading to hemorrhage and dilatation of adjacent interarcuate branches. Informed consent should be obtained from the owner before surgery to perform CT or MRI after surgery and to carry out long-term follow-up.

4 | CONCLUSIONS

We showed that, after diagnosis of spinal epidural AVF in 3 young adult cats with intermittent spinal hyperesthesia and abnormal hindlimb gait, long-term remission of clinical signs was achieved by dorsal laminectomy and occlusion of the dilated abnormal interarcuate branches. The differential diagnosis of spinal AVF should be considered, especially in young cats presenting with clinical signs such as intermittent spinal hyperesthesia and abnormal hindlimb gait.

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CONFLICT OF INTEREST DECLARATION

Authors declare no conflict of interest.

OFF-LABEL ANTIMICROBIAL DECLARATION

Authors declare no off-label use of antimicrobials.

INSTITUTIONAL ANIMAL CARE AND USE COMMITTEE (IACUC) OR OTHER APPROVAL DECLARATION

All owners of animals included in this study gave consent for diagnostic tests, treatments, and educational use of data.

HUMAN ETHICS APPROVAL DECLARATION

Authors declare human ethics approval was not needed for this study.

ORCID

Hiroaki Kamishina https://orcid.org/0000-0001-9929-6654
Yasushi Hara https://orcid.org/0000-0003-4474-4389
Yoshihiko Yu https://orcid.org/0000-0003-3396-6525
Daisuke Hasegawa https://orcid.org/0000-0002-7554-9108

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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