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# Angiographic Detection and Characterization of "Cryptic Venous Anomalies" Associated With Spinal Cord Cavernous Malformations Using Flat-Panel Catheter Angiotomography

**BACKGROUND:** Spinal cord cavernous malformations (CMs) are associated with 2 types of angiographically occult "cryptic venous anomalies," which differ in location with respect to the spinal cord. The anatomic distinction between superficial and intramedullary is important in that the latter heighten the risks of CM resection.

**OBJECTIVE:** To report the observations of both types of cryptic venous anomalies documented during spinal digital subtraction angiography enhanced with flat-panel catheter angiotomography (FPCA).

**METHODS:** Spinal digital subtraction angiography enhanced with FPCA was performed in 2 adult patients with magnetic resonance imaging-documented intramedullary spinal cord CMs and prominent, nonspecific flow voids at the same levels. FPCA was obtained by selective injection of left T4 (case 1) and left T9 (case 2) with 5F Cobra 2 catheters (Terumo, Japan) during a 20-second rotational acquisition. Thirty milliliters of a 75% saline and 25% contrast solution (Omnipaque 300; GE) was administered. The rotational data set was reconstructed on a dedicated workstation (Leonardo; Siemens, Erlangen, Germany) through the use of regular and high-resolution matrixes, 0.4- and 0.1-mm voxel size, respectively.

**RESULTS:** Spinal digital subtraction angiography was unremarkable in both cases. In case 1, FPCA findings indicated an atypical network of prominent posterior perimedullary veins. In case 2, FPCA identified radially oriented channels forming a *caput medusae* pattern collecting into an enlarged intramedullary vein.

**CONCLUSION:** The unique ability of FPCA to image the spinal venous system enables the angiographic detection and characterization of abnormal spinal veins associated with CMs. Differentiating between the types of associated cryptic venous malformations may aid in surgical planning because the intramedullary type is associated with a higher risk of surgical complication.

**KEY WORDS:** Angiographic detection, Cryptic venous anomalies, Developmental venous anomaly, Flat-panel catheter angiotomography (FPCA), Intramedullary spinal cord cavernous malformation, Spinal angiography

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The term cryptic vascular malformations was introduced by Crawford and Russell<sup>1</sup> in 1954 to characterize lesions responsible for "spontaneous" intracranial hemorrhages in

ABBREVIATIONS: AMSV, anterior-median spinal vein; CM, cavernous malformations; DVA, developmental venous anomaly; FPCA, flat-panel catheter angiotomography; SpDSA, spinal digital subtraction angiography young patients. Nowadays, it is generally used to describe angiographically occult intracranial vascular malformations,<sup>2</sup> ie, cavernous malformations (CMs) and capillary telangiectasias, which likely represent extreme forms of a single entity.<sup>3</sup> Cryptic vascular malformations are less common at the spinal level,<sup>4</sup> and the term takes a slightly different meaning because it includes several types of ill-defined venous anomalies in addition to CMs. For example, the cases published by Barnwell et al<sup>5</sup> included an isolated

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venous malformation and 2 venous varices besides CMs. An association between CMs and developmental venous anomalies (DVAs; or venous angiomas) was first suggested at the cerebral level by Rigamonti and Spetzler<sup>6</sup> in 1988. Similarly, Vishteh and coauthors<sup>7</sup> established a link between spinal CMs and cryptic venous malformations, which they divided into 2 types according to their superficial or intramedullary location; these authors also noted a higher surgical risk for the resection of CMs associated with venous anomalies. The association between spinal CMs and venous anomalies was further supported by several small case series, summarized by Gross et al<sup>8</sup> in 2010. In the review of 27 publications including 352 patients, an association was found in 34% of cases (24 of 70 patients in 6 series; range, 0%-94% per series; Table).<sup>4,7-12</sup> Of note, the venous anomalies observed surgically had not been detected in patients investigated by spinal angiography.

We report 2 observations illustrating both types of these so-called cryptic venous anomalies associated with intramedullary CMs that were documented during spinal angiography enhanced with flat-panel catheter angiotomography (FPCA). The striking similarity between the intramedullary form of cryptic venous anomaly and cerebral DVA is discussed, as well as the potential clinical implications of a new imaging technique able to document these anomalies before surgery.

# PATIENTS AND METHODS

# **FPCA Methodology**

FPCA was obtained by selective injection of left T4 (case 1) and left T9 (case 2) with 5F Cobra 2 catheters (Terumo, Japan) during a 20-second rotational acquisition with the patient breathing normally. Thirty milliliters of a 75% saline and 25% contrast solution (Omnipaque 300; GE) was administered (total volume of contrast, 7.5 mL). The rotational data set was reconstructed on a dedicated workstation (Leonardo; Siemens, Erlangen, Germany) using both regular (0.4-mm voxel size) and high-resolution (0.1-mm voxel size) matrixes.

# **CASE DESCRIPTIONS**

#### Case 1

A 52-year-old man presented with a 4-year history of progressively worsening left T10 radiculopathy. Clinical examination and laboratory studies were otherwise unremarkable. Spine magnetic resonance imaging (MRI) showed abnormal intramedullary signal intensity in the posteriorlateral aspect of the spinal cord at the T7 vertebral level, without enhancement or surrounding edema, suggestive of an intramedullary CM (Figure 1A). However, prominent flow voids were noted at the same level, near the right neural foramen (Figure 1B), and spinal digital subtraction angiography (SpDSA) was performed to exclude another type of vascular malformation. SpDSA, venous phase included, was unremarkable, whereas FPCA (DynaCT, Siemens Medical, Erlangen, Germany) documented an atypical network of prominent perimedullary veins located along the posterior aspect of the spinal cord at the level of the CM seen on MRI (Figure 1C-1E).

#### Case 2

A 52-year-old man with a 7-year history of nonprogressing right-hand weakness related to a right brachial plexus neurofibroma presented with

new finger numbness and tingling. An MRI confirmed the plexus lesion but also revealed new focal intramedullary hemosiderin deposition consistent with intramedullary CM and/or hemorrhage at the T1 level (Figure 2A). No change in size or appearance of the lesion was noted in a follow-up study obtained 6 months later, confirming the diagnosis of CM. However, abnormal intramedullary and extramedullary vessels documented at the cervicothoracic level suggested a possible additional vascular malformation, whereas extensive vascular anomalies consistent with superficial venous malformations were noted within the posterior thoracic wall on the right side (Figure 2B; Surgery performed 20 years earlier yielded a nonspecific diagnosis of vascular malformation). The SpDSA was unremarkable. The FPCA documented prominent anteriormedian spinal vein (AMSV) and right posterior-lateral spinal vein, as well as 2 large intramedullary veins, topographically matching the flow voids documented by MRI. The CM was located between the upper and lower intramedullary channels (Figure 2C). No venous structures were seen along the left posterior-lateral aspect of the cord at the corresponding level. The upper intramedullary venous channel extended from C3-C4 to C6, whereas the lower channel could be followed from T1 down to the lower edge of the field of view. The upper channel connected cranially into a vessel that continued upward as a left posterior-lateral spinal vein and caudally into a prominent AMSV through a short connecting segment coursing within the anterior-median sulcus. Only the cranial termination of the lower central channel was seen; after exiting the spinal cord posteriorly, it ran along its right lateral aspect to join the AMSV as well. The AMSV, after collecting these 2 abnormal tributaries, drained into the internal vertebral venous plexus via a prominent anterior radiculomedullary vein at the right T1-T2 level (Figure 2D). Several small, radially oriented channels forming a caput medusae pattern were collecting into the cranial portion of the upper intramedullary vein (Figure 2E and 2F).

Both patients elected for nonoperative management given the absence of previous hemorrhage and associated high-flow vascular malformations.

# DISCUSSION

#### **FPCA** Technique

FPCA is a new angiographic technique in which a 20-second rotational acquisition obtained during selective injection of a vessel of interest is used to generate multiplanar 2-dimensional nonsubtracted angiotomographic reconstructions of the opacified arteries and veins within their anatomic surroundings. This new modality was made possible in large part by the introduction of flat-panel detector technology, with higher dynamic range and digital readout rates that provide higher contrast and spatial resolutions than regular image intensifiers. FPCA has been shown to be a particularly useful complement to spinal angiography, during which a venous phase of diagnostic quality is notoriously difficult to obtain, primarily because of contrast dilution and respiratory or bowel motion artifacts. The lack of susceptibility of FPCA to breathing and bowel motion, combined with its high contrast sensitivity and high resolution capabilities (down to 0.1-mm voxel size), offers a highly detailed depiction of the normal and abnormal perimedullary venous system.<sup>13</sup> Although FPCA obtained during spinal angiography remains an invasive imaging method for which indication must be weighed carefully, it must

Series		Spinal CM	Associated Venous Abnormalities		Location of Venous Abnormality, n		
Author	Journal, Year	Total, n (Location)	Patients, n	Level of Venous Anomaly	Superficial	Deep	Spinal Angiography, n
Vishteh et al <sup>7</sup>	Neurosurgery, 1997	17	16	T6-T7		16	6, Unremarkable
		(8 cervical)		Otherwise not specified			
		(8 thoracic)					
		(1 conus medullaris)					
Cosgrove et al <sup>4</sup>	Journal of Neurosurgery, 1988	5	0				1, Unremarkable
		(1 cervical)					
		(4 thoracic)					
Sandalcioglu et al <sup>10</sup>	Neurosurgical Review, 2003	10	3	C0-C2	3		
		(5 cervical)		C7			
		(5 thoracic)		T1-T2			
Santoro et al <sup>11</sup>	Neurosurgical Review, 2004	10	3	Not specified		3 <sup>b</sup>	1, Unremarkable
		(5 cervical)					
		(5 thoracic)					
Weinzierl et al <sup>12</sup>	Neuroradiology, 2004	12	0				
		(5 cervical)					
0		(7 thoracic)					
Bian et al <sup>9</sup>	Clinical Neurology and Neurosurgery, 2009	16	2	C1-C2	2		
		(9 cervical)		C3-C4			
		(7 thoracic)					
Total		70	24		5	19	8

<sup>a</sup>CM, cavernous malformation. This table summarizes the 6 series analyzed in the review by Gross et al,<sup>8</sup> in which 24 of 70 patients (34%) with a spinal cord CM had an associated cryptic venous anomaly. In the 24 patients, 5 had superficial anomalies and 19 had deep venous anomalies.

<sup>b</sup>Santoro et al observed 3 associated venous anomalies as those defined by Vishteh et al. It is presumed that Santoro et al are referring to deep, rather than superficial, anomalies.



**FIGURE 1.** A 52-year-old patient with 4-year history of worsening T10 radiculopathy. **A**, axial T2-weighted magnetic resonance image (MRI) documenting a intramedullary cavernous malformation at the T7 vertebral body level (ie, at approximately the T10 spinal cord segment). **B**, axial T2-weighted MRI showing prominent flow voids at the right T7-T8 neural foramen level representing a draining vein (arrow). Smaller perimedullary veins are also noted around the spinal cord (and seen in **C**). **C**, axial flat-panel catheter angiotomography (FPCA) reconstruction. Prominent venous structures are seen along the posterior aspect of the spinal cord (arrowhead) and in the vicinity of the right T7-T8 neural foramen (arrow). **D**, sagittal FPCA reconstruction showing enlarged posterior perimedullary veins (arrowheads) and a small anastomotic connection with a segment of anterior-median spinal vein (long arrow), which lies deep to the anterior spinal artery (short arrow). **E**, coronal FPCA reconstruction documenting the same network of dorsal perimedullary veins draining bilaterally but more prominently to the right side. A mild narrowing of the radiculomedullary vein at the junction with the internal vertebral venous plexus represents the antireflux mechanism (arrow).

be noted that, despite its historic reputation, modern spinal angiography carries a very low risk of complications. Neurological events, in particular, were not noted in a recent evaluation of 302 consecutive diagnostic spinal angiograms<sup>14</sup> or in a series of 63 consecutive studies using selective spinal artery FPCA.<sup>13</sup> In both of our reported cases, spinal angiography was performed to exclude the possibility of a spinal cord vascular malformation suggested by the MRI documentation of an intramedullary lesion with abnormal perimedullary vessels.

#### The Venous System of the Spinal Cord

The veins of the spinal cord are divided into an extrinsic or superficial group, consisting mainly of longitudinal channels coursing over the ventral and dorsal surface of the cord, and an intrinsic or deep group found within the cord parenchyma and the median sulci.

The AMSV courses along the anterior-median fissure, deep or lateral to the anterior spinal artery.<sup>15-17</sup> Dorsally, 2 small posterior-lateral veins accompany the posterior spinal arteries, and a larger posterior-median spinal vein lies over the posterior-median fissure.<sup>15,17</sup> The AMSV and posterior-median spinal vein

are relatively constant, in particular at the thoracolumbar and sacral levels.<sup>18</sup> At the mid and upper thoracic levels, spinal veins are more tortuous and assume a networklike appearance.<sup>16,19</sup> The anterior and posterior longitudinal systems are interconnected via numerous superficial anastomoses and through the coronal venous plexus of the pia mater.<sup>15,17</sup> The extrinsic venous system drains into the internal vertebral venous plexus via a variable number of anterior and posterior radiculomedullary veins.<sup>18,20</sup> The direction of flow within these veins is controlled by an antireflux mechanism; a narrowing of the vein as it pierces the dura to reach the internal vertebral venous plexus appears to be the principal mechanism of flow regulation.<sup>18,21,22</sup> The internal vertebral venous plexus, principally via emissary veins coursing along the spinal nerves through the neural foramina.

The intrinsic venous system is divided into central and peripheral components, both draining centrifugally toward the extrinsic venous system. The central component is made of sulcal veins that drain the spinal gray matter at the exception of the dorsal portions of the posterior horns.<sup>17,20,23</sup> As they course ventrally to join the AMSV, the sulcal veins receive small tributaries from the

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**FIGURE 2.** A 52-year-old patient with a 7-year history of upper-extremity weakness. **A**, axial T2-weighted magnetic resonance image (MRI) documenting the intramedullary cavernous malformation (CM). **B**, axial T1-weighted MRI after gadolinium administration documenting a CM within the posterior aspect of the spinal cord (arrowhead), a prominent anterior-median spinal vein (AMSV; long arrow), and a diffuse right posterior thoracic wall venous malformation (short arrows). Also note an enhancing mass within the right C7-T1 neural foramen consistent with a brachial plexus tumor (asterisk). **C**, sagittal flat-panel catheter angiotomography (FPCA) reconstruction matched with 4 axial postgadolinium T1-weighted MRIs at C5 (a), C6-C7 (b), upper T1 (c), and upper T2 (d). At the C5 level (a), FPCA shows a small AMSV and right posterior-lateral spinal vein and a prominent central channel (arrowhead), the intramedullary location of which is confirmed by the corresponding axial MRI. At C6, the intramedullary vein makes a sharp ventral turn to drain into the AMSV. Only the latter can be seen at the C6-C7 level (b). The axial image at the upper T1 level (c) shows the AMSV aiming to the right radiculomedullary vein. The intramedullary CM is documented at the same level. A second longitudinal intramedullary channel is seen at the upper T2 level (double arrowhead), with its caudal end located outside the field of view. The axial MRI at the corresponding level shows the intramedullary vein (long arrow) draining into the AMSV (short arrow), which itself continues as a right medullary vein (arrowhead). The second longitudinal intramedullary vein is indicated by a double arrowhead; the right posterior-median spinal vein, by a double arrow. **E**, coronal FPCA reconstruction, magnified view, detailing the delicate venous network spreading from the central intramedullary vein toward the periphery of the spinal cord. **F**, axial FPCA reconstruction (inferior view) showing the AMSV (arrow) and the right posterior-median spinal vein, by a do

white matter bordering the anterior-median sulcus.<sup>23</sup> Contrary to the strictly unilateral sulcal arteries, sulcal veins collect tributaries from both sides of the spinal cord. The peripheral component includes veins forming near the gray/white junction and draining radially toward the surface of the cord, where they join the coronal venous plexus.<sup>20,23</sup> These veins drain most of the white matter and posterior gray matter, although they can overlap with

the central component, particularly at the cervicothoracic level.<sup>17</sup> The posterior-medial (or posterior septal) veins originate from the base of the posterior horns and from the posterior columns and travel dorsally within the posterior-median septum to join the posterior-median spinal vein.<sup>17,20,23</sup>

Segmental transmedullary anastomoses between anterior and posterior longitudinal veins of the extrinsic venous system are

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established via intramedullary connections between the peripheral and central components of the intrinsic venous system.<sup>19,23,24</sup> During their intramedullary course, these connecting veins usually curve around the central canal<sup>16</sup> and sometimes form a longitudinal loop that parallels the canal over a short distance.<sup>19</sup> Intersegmental anastomoses linking adjacent sulcal or posteriormedial veins also travel longitudinally within the gray matter adjacent to the central canal.<sup>16,17,20,25</sup> These intersegmental anastomoses are longer and more numerous at the thoracic level, where the density of sulcal veins is lower than in the rest of the spinal cord.<sup>17</sup> Therefore, despite the essentially segmental organization of the intrinsic venous system, numerous small intramedullary veins can be found travelling longitudinally in the vicinity of the central canal.

#### The "Cryptic" Spinal Venous Anomalies

We report the angiographic appearance of the 2 types of cryptic venous malformations reported in association with spinal CMs. Our first case illustrates the superficial type, which accounts for 5 of the 24 venous abnormalities noted in the 70 patients reviewed by Gross et al.8 Although not strictly fitting the definition originally proposed by Vishteh and coauthors<sup>7</sup> because of their superficial location, these anomalies are commonly described as a type of cryptic venous malformation in the literature. Our second case documents a far more complex venous anomaly, associated in this instance with an intramedullary CM and a superficial venous malformation. The abnormal intraparenchymal vessels seen in our patient can be described as 2 separate longitudinal veins, 1 superior and 1 inferior, travelling along the central canal, akin to Vishteh and coauthors' second type of abnormal veins, seated deep within the cord substance. These venous channels, connected cranially and caudally with normal longitudinal veins of the extrinsic venous system, likely represent a variation in size and length of the small intersegmental anastomoses normally present along the central canal, acting in this case as a deep or subependymal venous system for the spinal cord. The fact that the extrinsic venous system was partially missing at the corresponding levels strongly suggests a compensatory role for these abnormal channels. Figure 3 illustrates how the venous blood that should have drained centrifugally toward the missing posterior-median or left posterior-lateral veins was redirected centripetally toward the abnormal longitudinal channel, in a fashion reminiscent of the mode of drainage of a DVA, ie, as the compensatory drainage of a superficial territory into the deep venous system. The small tributaries of the longitudinal intramedullary veins even assumed a pattern similar to the typical caput medusae appearance of a DVA.

The characteristics of the structures described in our second case, ie, a deep venous channel participating in the aberrant drainage of a superficial territory via small radial veins forming a *caput medusae* pattern, would be sufficient to diagnose a DVA at the intracranial level. Additional features support this analogy, most significantly the presence of a CM in the immediate vicinity

of the aberrant venous channels. The reported incidence of cerebral CMs associated with DVAs ranges from 8% to  $26\%^{26,27}$ but is likely higher because DVAs too small for diagnosis by current imaging methods may be detected only during microsurgery.<sup>28</sup> A study of 45 patients with brainstem CMs showed a 100% association rate with DVAs, with only 5 identified by preoperative imaging.<sup>29</sup> In the Vishteh et al<sup>7</sup> series, 16 of 17 spinal CMs (94%) were associated with intramedullary venous malformations, defined as abnormal venous channels located deep within the substance of the cord and draining into a larger abnormal venous structure. Finally, our patient also had a superficial venous malformation affecting his thoracic wall at a level corresponding to the spinal cord venous anomaly and CM. Because there is a well-recognized association between superficial venous malformations of the head and neck and intracranial DVAs,<sup>30</sup> this finding may be viewed as additional indirect support of the hypothesis that the reported aberrant venous pathway represents a spinal form of DVA.

## CONCLUSION

The association of abnormal spinal veins with intramedullary CMs has so far been a surgical finding; no imaging technique has been able to document their existence and appearance clearly, including SpDSA. Because of its unique ability to image the spinal venous system, FPCA, a new technique providing high-resolution CT-like tomographic reconstructions of a catheter angiogram,<sup>31,32</sup> is particularly interesting for the field of spinal angiography. In both of our patients, MRI showed some abnormal intramedullary and/or perimedullary vessels but was not specific enough to characterize them as venous anomalies, ie, to differentiate them from other types of vascular malformations. In addition, SpDSA, including a venous phase obtained in each case, was unable to document the abnormal venous structures, which were appreciated only through catheter-based high-resolution FPCA. The angiographic detection and characterization of venous anomalies associated with CMs may have an interesting role to play in the diagnosis and treatment of spinal CMs, in particular for the differentiation between extrinsic and intrinsic types of venous anomalies because the latter is known to carry a higher risk of surgical complication. Finally, the features of the intrinsic venous abnormality present in our second patient strongly suggest that it represents the spinal equivalent of a cerebral DVA.

#### Disclosures

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