Spinal dural and epidural fistulas: role of cone beam CT in diagnosis and treatment

Maksim Shapiro 1, Erez Nossek 2,3, Vera Sharashidze 4,5, Michihiro Tanaka 6, Caleb Rutledge 2,3, Charlotte Chung 4,5, Ayaz Khawaja 4,5, Howard Riina 2, Peter Kim Nelson 4,5, Eytan Raz 2,3

ABSTRACT
Understanding normal spinal arterial and venous anatomy, and spinal vascular disease, is impossible without flow-based methods. Development of practical spinal angiography led to site-specific categorization of spinal vascular conditions, defined by the ‘seat of disease’ in relation to the cord and its covers. This enabled identification of targets for highly successful surgical and endovascular treatments, and guided interpretation of later cross-sectional imaging. Spinal dural and epidural arteriovenous fistulas represent the most common types of spinal shunts. Although etiology is debated, anatomy provides excellent pathophysiologic correlation. A spectrum of fistulas, from foramen magnum to the sacrum, is now well-characterized. Most recently, use of cone beam CT angiography has yielded new insights into normal and pathologic anatomy, including venous outflow. It provides unrivaled visualization of the fistula and its relationship with spinal cord vessels, and is the first practical method to study normal and pathologic spinal veins in vivo—with multiple implications for both safety and efficacy of treatments. We advocate consistent use of cone beam CT imaging in modern spinal fistula evaluation. The role of open surgery is likely to remain undiminished, with increasing availability and use of hybrid operating rooms for practical intraoperative angiography enhancing safety and efficacy of complex surgery.

INTRODUCTION
Understanding spinal vascular disease is a beautiful example of puzzle solving through imaging advances, correlated with meticulous clinical observation. Early case descriptions accompanied (post mortem) findings of congestive myelopathy. Contrast myelography allowed for visualization of dilated perimedullary vasculature in vivo. By the early 1970s, a strong male predominance and venous congestive pathophysiology were identified by Aminoff and Logue. 1,2 In 1977, enabled by early spinal subtraction angiography, Kendall and Logue published the seminal and still perhaps the most important paper on the topic, describing ‘spinal epidural angiomatous malformations’—what we now know as the spinal dural fistula. 3 They established the dural location of these slow flow shunts with consistent presence of a single transdural vein connecting the dilated spinal venous system (figure 1).

Understanding shunt anatomy allowed for development of highly successful targeted surgical division of the culprit vein, leading to post-treatment improvement in about two-thirds of cases and stabilization in the remainder.

Since then, endovascular treatments have been developed by Djindjian and others. 4 5 A range of locations, from cervical to sacral, were documented. The impact of early CT was limited, while MR enabled non-invasive identification of vascular congestion in the majority of cases, leading to earlier diagnoses among more patients. Continued improvement in CT and MR equipment now allows for pinpointing the location of the shunt in some cases, tempered by highly limited availability and interpretive expertise, as well as limited visualization of lower flow shunts and suboptimal assessment of cord supply arteries. Basic subtraction angiography enabled characterization of other spinal vascular shunts—notably, the intradural pial/medullary arteriovenous malformation spectrum lesions as well as complex, multicompartamental, ‘metameric’ shunts. Considerable progress has been made in establishing the underlying genetics and geno-phenotypic correlation of these lesions. The less common spinal epidural fistula was recognized as a unique entity, with similar pathophysiology but different shunt location from that of the spinal dural fistula. 6 7

Important challenges remain. We have no definitive proof of etiology. There are no preventive or reasonable screening options. We are not aware of a genetic predisposition (excepting complex shunts), including why the majority are men aged ≥50 years. We do not know enough about normal distribution of radicular and bridging spinal veins (ex vivo work by Thron probably being the best guide 8), or reasons for their apparent paucity/loss in the majority of symptomatic dural fistulas. They are below the resolution of most commercially available MR sequences, and difficult to image using 2D angiography. However, it is their deficiency, as much as the presence of additional shunt-related inflow, that determines the severity of congestive myelopathy 9 10 (figure 1B). Whether lack of draining veins is due to their pre-existing rarity, acquired fistula-related occlusion, or both, remains unclear. 11

Even after successful occlusion or interruption of the shunt, the spinal venous drainage system remains abnormal due to paucity of these
Spinal transdural drainage pathways, potentially contributing to limitations in post-treatment recovery (figure 1). We currently have no methods for creating additional drainage routes or improving the existing ones, apart from anticoagulation to forestall immediate post-treatment thrombotic catastrophe (the confusion related to Foix-Alajouanine syndrome), and hopefully preserve what remains of the transdural drainage in the long term.

The purpose of this manuscript is to review, in light of the above, our imaging-based anatomical insights into the dural and epidural fistula spectrum (figure 2), emphasizing cone beam CT (CBCT) advances and practical impact on diagnosis and treatment (figure 3).

NORMAL SPINAL ARTERIOVENOUS ANATOMY, DURAL FISTULA ANATOMY, AND PATHOPHYSIOLOGY CORRELATION

The anatomical background is essential for an understanding of spinal dural fistulas, but extensive discussion is beyond the scope of this review. Figures 1 and 2 show the spectrum of normal arterial and venous anatomic variation, relationships between various fistulas, cord arterial supply, and venous drainage.

The various naming systems for spinal vessels can be confusing, and none are ideal. Supply to tissues adjacent to, and within, the spinal canal (nerve root sleeve, epidural space, dura, and cord) is provided by the radiculomeningeal artery or arteries (also known as radiculodural artery), coursing through the neural foramen. Its radicular portions, within the nerve root sleeve, supply the nerve roots. Dorsal and ventral roots are associated with separate radicular arteries—an important point in the relationship between the dural shunts, nerve roots, and spinal cord arterial supply (figures 1 and 2).

Additionally, the radiculomeningeal artery variably contributes to supply of the dura, the epidural spaces, and bone around the spinal canal. Extensive connections between neighboring radiculomeningeal arteries play an important role in fistula angioarchitecture, particularly via the characteristic diamond-shaped dorsal and ventral epidural arcades, along with the lateral dural branches. (figures 1 and 2).

Whenever the (ventral) radiculodural artery, in addition to serving the nerve root, also contributes to supply of the anterior spinal artery, it is called ‘radiculomedullary artery’. Dorsal radiculodural arteries supplying the posterior spinal arteries/network are called ‘radiculopial’. There is tremendous spectrum-like variation present throughout the cord: at cervical and cranial ends the posterior/lateral spinal arteries can be prominent while anterior spinal arteries are correspondingly small. CBCT occasionally shows very prominent posterior spinal arteries over the thoracic region as well—apart from the classic teaching of smaller size thoracic anterior spinal due to lesser cord volume. Finally, there is nothing exclusively ‘medullary’ about the anterior spinal artery or ‘pial’ about the posterior spinal ones. All supply the cord and are in balance with each other.
VENOUS ANATOMY

Venous anatomy is critical in determining fistula-related symptomatology. Normal cord veins, previously impossible to study in vivo, are now consistently visualized with CBCT (figure 3)—a landmark advance which deserves recognition.

Both intrinsic and cord surface veins are highly redundant. Cord outflow must traverse the dura via a limited number of radicular and bridging veins. The former cross the dura at the nerve root sleeve, draining into the foraminal or epidural venous plexi. The latter traverse the dura independently of the nerve roots, allowing for debate as to nomenclature/prevalence of both vein types (figure 1).

These radicular/bridging veins are the ‘weak link’ between highly redundant intradural and extradural venous systems. Their ‘normal’ anatomy—size, number, extent of connection to cord veins—has not been well studied. Perhaps the best source is Armin Thron’s ‘Vascular anatomy of the spinal cord’. He reports their limited number and apparent thrombosis/occlusion of some on post mortem studies in normal subjects, presumably age-related.

ANGIOGRAPHIC AND CBCT IMAGING

Arterial 2D DSA imaging is well described. CBCT provides markedly superior spatial resolution, but presently lacks temporal information. It is particularly helpful in visualizing small, but clinically important, posterior spinal arteries. For acquisition times longer than ~5 s, venous admixture becomes a factor.

Imaging normal and pathologic cord veins is difficult. The anti-reflux mechanism (generally believed to be a kind of functional valve at the transdural portion of the radicular/bridging vein) prevents contrast reflux into cord veins via...
A spinal dural fistula is a dural-based shunt. The majority are located within or near the nerve root sleeve (figures 1 and 2). Arterial supply is derived from dural branches of the radiculodural, radiculomedullary, or radiculopial arteries. Although a number of small arteries are usually present at the fistula site, there does not seem to be a ‘nidus’, and the shunt is relatively simple. The more medially/centrally placed shunts usually recruit progressively greater supply from the lateral dural arcade (figures 2 and 4). Either the dorsal or ventral radiculodural branch is the primary supplier. Exceptionally, a dorsal shunt can be selectively embolized in the presence of an adjacent ventral radiculomedullary artery (figure 5), although most of these cases are better served by surgical disconnection. Endovascular treatment of shunts adjacent to cord supply arteries has also been reported by Adrianto et al.20

The majority of dural shunts are thoracolumbar. However, increasing appreciation of rare cervical (figure 6), as well as foramen magnum (bridging the spectrum of brain dural fistulas) and sacral types (figure 7) is important, underscoring the need for complete angiography.

Venous drainage defines symptomatology, and is in many ways more important than arterial angiography. A single (usually dorsal radicular, sometimes ventral radicular or bridging) draining vein directs the fistulous arterialized flow retrogradely towards the cord, typically into posterior cord veins18 19 21 (figures 1 and 2). The duration and extent of cord congestion are the main determinants of clinical dysfunction. Congestion is principally determined by availability, or lack, of normal regional radicular/bridging veins to drain both the fistula-related inflow and the adjacent cord. Impressive-looking fistulas may be well tolerated when nearby drainage is available.9 Conversely, small and difficult to identify fistulas with no ready outflow produce major morbidity (figure 8)—another setting in which CBCT can be extremely useful. Although a matter of debate, we and others11 believe that either intrinsic deficiency or progressive occlusion of the draining veins by high-flow venopathy and/or thrombosis leads to stepwise disease progression as successive venous outflow routes are eliminated.

In contrast, the far higher flow spinal arteriovenous malformations do not consistently cause venous congestion due to functional venous outlets. If this outflow fails, congestion rather than hemorrhage, may become the principal clinical issue.9 22

**ANATOMY AND IMAGING OF SPINAL DURAL AND EPIDURAL FISTULAS**

**Figure 4** (A–K) Cord venous congestion in dural arteriovenous fistula (dAVF). Mid- (A) and late-(B) phase injections of the Adamkiewicz artery in patient with left T6 level dAVF (C). The late-phase image (B) shows no identifiable cord veins, attesting to their marked congestion. (C) Absence of radicular venous drainage along the majority of thoracolumbar cord—a single functional lumbar vein is present (arrow). (D) Primary fistula supply via left T6 radicular artery. (E) Indirect supply via the lateral dural branch is seen from left T5 level (arrows in D and E). (F) Coronal maximum intensity projection Dyna CT image. (G) n-Butyl cyanoacrylate (nBCA) cast of left T5 level embolization, eliminating the lateral dural branch contribution so as to increase the probability of nBCA penetration into the fistulous vein from subsequent left T6 injection. (H) Lack of wedge position (flow control) as shown by contrast reflux (arrow) reduces probability of successful nBCA injection. (I) Therefore, the headway duo is replaced with a dual lumen Scepter C. Although its final position is more proximal, flow control is ensured by balloon inflation, allowing for successful penetration of nBCA into the foot of the vein (arrow) via several fistulous arteries (dashed arrows). J, K: Post-embolization control injection of the Adamkiewicz artery (J) now visualizes the cord veins (K, arrows), previously parasitized by the fistula.
Spinal Epidural Fistula

A minority of shunts are localized to the epidural, rather than dural, space. Typically, one or more epidural arcade arteries (usually ventral epidural arcade—also known as dorsal somatic arteries) converge on a venous pouch in the epidural space—presumably part of the epidural venous plexus which has somehow become isolated from its multiple connections (figure 2). The pathologic outflow is retrogradely directed via a radicular or bridging vein into the cord venous system (figure 9), resulting in congestion—similar to spinal dural fistulas. The supposed anti-reflux mechanism of the transdural vein must somehow fail in this extradural shunt. The majority are located...
Spine

in lumbar areas, and the characteristic venous pouch is usually appreciated on high-quality MR studies, and always on CBCT. It is possible that asymptomatic epidural fistulas exist by virtue of preserved epidural antegrade outflow away from the cord. There seems to be a wider spectrum of epidural fistula venous drainage (both antegrade epidural and retrograde intradural routes) in comparison with the typical single retrograde vein in dural fistula cases.\textsuperscript{18 25 26} Highly complex epidural fistulas are also encountered, sometimes as part of an underlying genetic disorder (figures 10 and 11).

**CBCT FISTULA IMAGING**

For enhanced safety, superior visualization of shunt angioarchitecture helps to identify anterior and, importantly, posterior spinal artery supply adjacent to the fistula\textsuperscript{27} (figure 12). The posterior spinal arteries, in particular, can be difficult to see on 2D imaging, and may be quite symptomatic if penetrated with liquid embolic agents.\textsuperscript{28} Similarly, CBCT greatly facilitates understanding of collateral routes between adjacent segmental arteries, where contributions to anterior or posterior spinal axes frequently exist—allowing for more informed decision and strategy.\textsuperscript{14} (figure 13). Both cross-sectional and volumetric reconstructions are helpful in identifying optimal projections for microcatheterization.

The protocol for acquisition of high-quality spinal CBCT scans has been reported.\textsuperscript{14 25 29} Success is as much a result of imaging equipment factors as minimizing patient motion, optimal catheter position, and contrast injection. Although sometimes possible to achieve in the awake state, a spinal CBCT scan is best obtained under general anesthesia, with pharmacologic paralysis.

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**Figure 7** (A–F) Foramen magnum and sacral dural arteriovenous fistulas (dAVFs). (A) T2 sagittal view of prominent dorsal cervical cord veins (purple arrows). (B) Foramen magnum fistula supplied by the neuromeningeal trunk (green arrow) of the ascending pharyngeal artery. (C, D) Paucity of radicular drainage along the entire cervicothoracic cord, with several patent lumbar radicular veins (blue arrows) remaining. (E, F) Left S1 sacral dAVF, supplied by left S1 (purple) and filum terminal arteries (orange) via the left S3 (yellow) radicular arteries, with outflow into the filum terminale vein (blue arrows).

**Figure 8** (A–H) Challenging diagnosis of small dural arteriovenous fistula (dAVF) in a patient with three prior nondiagnostic angiograms. (A) Extensive abnormal cord signal with paucity of perimedullary veins. (B, C, D) Small and very slow flow dAVF, with faintly seen perimedullary draining vein (arrows), better shown in the subsequent late phase DSA (E), coronal maximum intensity projection Dyna CT (F), and wedge position microcatheter injection (G, H) Dyna CT scan of n-butyl cyanoacrylate cast, demonstrating permeation into the ‘foot’ of the fistulous vein (arrow). The lateral thoracic dural branch is also filled (dashed arrow).
of skeletal and sometimes smooth muscle (bowel motion), during mechanical breath hold (apnea). Arms may be elevated for best view of thoracic segments. Rotation (and thus injection) times vary depending on imaging equipment. Rates of 1–2 cc/sec for segmental arteries are typical; thus the additional contrast burden is small. A delay of several seconds is useful to achieve steady-state opacification with present-day imaging times. The highest spatial resolution protocol on a well-calibrated machine should be chosen. Secondary reconstructions with smaller fields of view and different algorithms can be very useful. Maximum intensity projection imaging is generally superior to volume rendering at this point, except in the cervical spine. Cervical spine doses are typically 100–200 mGy, while lumbar or sacral doses can be up to 500 mGy. The rotational nature of acquisition mitigates against focal skin exposures typical of complex spinal DSA scans.

**IMAGING IN TREATMENT**

The goal of treatment is elimination of fistula-related venous congestion, allowing cord veins to become the sole user of whatever venous drainage routes remain. Cure requires selective closure of the proximal (with respect to fistula) ‘draining vein’, either by surgical division or endovascular occlusion (usually with a liquid embolic agent), sparing the intrinsic cord venous system.

Pooled studies generally document a higher rate of recurrence and poorer functional outcomes following endovascular therapy compared with surgery, for dural fistulas.30–33 This may be related to insufficient penetration of embolic material into the fistulous vein, on the one hand,33 or its spillage into the longitudinal cord veins, on the other, possibly compromising the already tenuous...
Spine venous drainage. The latter hypothesis is unproven. Others show no difference in outcomes between either modality. Endovascular cure nearly always requires liquid embolic agents, with cyanoacrylates performing better than ethylene vinyl alcohol copolymers (EvOH). However, with the use of dual lumen balloons EvOH reflux, one of its main disadvantages in spinal embolization, can be controlled. In this setting, CBCT can be practically done during injection of EvOH to monitor progress of embolization, particularly when ‘dangerous anastomoses’ are present (figure 13).

**Figure 11**  (A–J). Complex multiple epidural and paraspinous fistulas, in addition to facial arteriovenous malformation and sigmoid dural fistula.  
(A) – Extensive paraspinous flow voids and (B) segmental radicular and low cord congestion.  
(C–D) Selected paraspinous and epidural fistulas, overall involving every segmental artery from T8 to L3. The left T10 origin radiculomedullary artery (arrow) supplying the anterior spinal artery (dashed arrow) is faintly visible on background of a large paraspinous shunt – underscoring the importance of identifying cord supply. The source of cord congestion could not be identified.  
(E) Transvenous coil and Onyx embolization of the epidural (white oval) and paraspinous (black oval) fistulas eventually allowed identification of an epidural pouch (arrows in F,G) unfilled by the embolic material, retrogradely opacifying the right L1 radicular vein (dashed arrows), and demonstrating the culprit cord venous congestion (F).  
(H) n-Butyl cyanoacrylate cast within the epidural venous pouch and the epidural arterial arcade. The embolic material did not reach the foot of the vein. This was followed by recurrence of the fistula and need for surgical disconnection (I), demonstrated by intraoperative angiography (J).

**Figure 12**  (A–F) Importance of comprehensive angiographic evaluation.  
(A,B) Right supreme intercostal origin dural arteriovenous fistula.  
(C,D) Coronal and axial maximum intensity projection Dyna CT images demonstrating possible posterior spinal artery (arrows) associated with the fistula, and confirmed on wedge position microwire injection (E).  
(Lateral cord, ischemic due to liquid embolic penetration can be markedly symptomatic.  
(F) Radial approach intraoperative angiogram demonstrating successful fistula surgical disconnection.
It is important to appreciate that even after successful occlusion or interruption of the shunt, the spinal venous drainage system usually remains deficient due to the paucity of transdural drainage pathways, potentially contributing to limitations in post-treatment recovery (figures 1B and 4). We currently have no methods for creating additional drainage routes or improving the existing ones. Confining embolic material to the radicular vein, without spillage into cord veins where it might become a nidus for thrombosis, is important. Upper and mid-thoracic shunts tend to have shorter radicular veins than lower thoracolumbar/sacral veins, with shorter venous ‘safety’ favoring surgery in our practice.

Post-embolization non-contrast CBCT provides excellent assessment of embolic material penetration (figures 4, 5, 8, 9 and 13) and thus expected treatment durability. It may inform follow-up strategy (or planned surgery) in cases of insufficient venous penetration.

Limitations

Only a minority of laboratories consistently use spinal CBCT. The advantages presented above will hopefully lead to increased use. Nevertheless, literature on the topic is limited. Impact on safety and efficacy is unproven. More systematic studies would probably be needed to effect a broad workflow shift towards routine use of CBCT in the spine. Only the newest machines have acquisition times that are short enough to adequately visualize arteries without venous admixture in normal subjects. Adequate quality selective venous imaging is presently difficult (veins clear too quickly compared with the length of acquisition)—thus veins are only seen together with arteries. Radiation dosage over the thoracolumbar spine, although rotationally distributed, is not negligible.

CONCLUSIONS

Dynamic vascular imaging (angiography) enabled recognition of spinal dural and epidural fistulas as disease entities. Modern imaging enables excellent pathophysiologic correlation. We advocate routine use of cone beam CT as part of spinal dural fistula evaluation to provide better anatomical awareness, with likely safety and efficacy benefits.

Twitter Maksim Shapiro @neuroangio1, Erez Nossek @Enossek, Vera Sharashidze @SharashidzeVera, Michihiro Tanaka @Michihiro Tanaka @Michihiro1966, Charlotte Chung @ChungCharlotte and Eytan Raz @eytanraz

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ORCID iDs
Maksim Shapiro http://orcid.org/0000-0003-1279-5456
Erez Nossek http://orcid.org/0000-0003-2750-421X

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