Title: A rare case of spinal dural arteriovenous fistula with radiculopathy, without myelopathy or spinal edema on magnetic resonance imaging

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Short Title: SDAVF with radiculopathy

Abstract

Background

Spinal dural arteriovenous fistulas (SDAVFs) are the most common type of spinal arteriovenous malformations; they frequently cause progressive myelopathy, including gait disturbances and sensory disorders.

Case Description

We report a rare case of a middle-aged man who experienced right-sided chest pain and Th4 radiculopathy, without any other neurological presentations. Magnetic resonance imaging showed flow void sign on the dorsal aspect of the spinal cord; spinal angiography revealed an arteriovenous shunt between a radicular artery and an intradural vein. Suspecting SDAVF as the cause of the chest pain, we performed surgical resection. Intraoperatively, we observed compression of the rootlet by the draining vein. Right chest pain disappeared completely after obliteration of the SDAVF. The present patient had vascular compression of the spinal nerve rootlet without any venous congestion.

Conclusions

Our experience shows that SDAVF can present not only as a myelopathy but also as a radiculopathy, indicating that radiculopathy may become a main symptom of SDAVF.

Spinal dural arteriovenous fistula (SDAVF) is the most commonly occurring type of spinal vascular malformation.[1,2] Clinical findings of SDAVF include progressive myelopathy induced by a dural shunt and an abnormal connection between the arterial and venous systems that reverses blood flow and increases intradural venous pressure.[3,4] This condition can eventually lead to venous hypertension and venous congestion resulting in myelopathy such as paraparesis, bladder rectal disorder, and sensory disorder.

To our knowledge, only few studies have reported cases of SDAVF with radiculopathy; however, the pathophysiology of the condition remains unclear.[5] Herein, we report a particularly interesting case of SDAVF, which presented with only right-sided chest pain and Th4 radiculopathy. We discuss the possible mechanisms of the radiculopathy based on intraoperative observation of the surgical field. The present case suggests that radiculopathy may become the primary symptom of SDAVF. Magnetic resonance imaging (MRI) or Computed Tomography angiography (CTA) should be considered if the typical symptoms are not observed.

Case Presentation: Written informed consent was obtained from the study patient. No ethical approval was required from our institutional review board since this was a case report.

A 45-year-old man presented with a 4-year history of right-sided continuous lancinating chest pain, which was made worse by exercise such as running or stretching. The pain was not aggravated in a specific position or during the Valsalva maneuver and remained constant over the course of the whole day. Three months ago, he underwent a complete health check-up at a local hospital. He was referred to our hospital for the further examination. Physical examination did not demonstrate motor weakness or hypesthesia. The chest pain was restricted to the right thoracic side (right Th4 area). He had no remarkable history of illness, including traumatic

injury. MRI of the thoracic spine showed prominent dilated vessels on the dorsal surface of the Th3/4 spinal level without spinal cord edema (Fig. 1).

Spinal angiography was performed, including that for all intercostal arteries. Angiography of the right Th3 intercostal artery demonstrated a SDAVF fed by the radicular artery, which drained into the intradural vein. There was no mass lesion in the epidural space (Fig. 2).

Embolization of the right T3 radicular artery was attempted; however, the microcatheter did not advance to the shunt point because of vessel tortuosity. The treatment was shifted to surgical resection. A laminectomy was performed from the lower 2/3 portions of Th3 to the upper 1/3 portions of Th4. Intraoperatively, on opening the dural sac, we noted compression of the nerve rootlet by an abnormally dilated vessel (Fig. 3). We clipped the vein at the point through which it passed through the inner surface of the dura mater. Subsequent intraoperative angiography showed complete resolution of the lesion.

The right-sided chest pain disappeared postoperatively. The patient was eventually discharged after an uneventful course. At postoperative 6-month follow-up, the patient was pain free, without any evidence of recurrence of fistula by MRI.

Discussion: In this report, we present a rare case of SDAVF. The patient presented with radiculopathy, without signs of myelopathy. Additionally, on MRI, flow void was noted in the spinal canal without evidence of spinal cord edema.

This case indicates that SDAVFs can present exclusively with radiculopathy. Pathologically, SDAVFs consist of an abnormal shunt between a dural artery branch and a radicular vein, which leads to increased venous pressure with consecutive venous congestion.[6] Thus, pathogenesis and

clinical manifestation of SDAVFs is often slow; however, progressive symptoms such as myelopathy, gait disturbances, sensory changes, and problems with micturition are noted.[6,7] This case also indicated that symptomatic SDAVFs could manifest only flow void without spinal cord edema on MRI. Cases of SDAVFs are commonly diagnosed by MRI.[6] Venous hypertension results in spinal cord ischemia or edema and manifests as homogenous, longitudinally extensive T2 signal abnormality within the central spinal cord. In addition, an MRI typically shows flow void of the radicular veins and cord enlargement.[8] Subsequent spinal cord angiography provides a definitive diagnosis and localizes the fistula.

A previous report described a case of SDAVFs that presented with similar symptoms.[5] However, the report did not address the pathophysiology of the fistula. In our case, we identified that the right-sided chest pain was manifested by the compression of a rootlet by an enlarged draining vein. Herein, we illustrate the mechanism of the radiculopathy with the schema (Fig. 4). This pathophysiology is similar to that of neurovascular compression syndrome, which presents with hemifacial spasm and trigeminal neuralgia. We successfully performed resection of the draining vein, which enabled complete cessation of the right-sided chest pain postoperatively. This case therefore represents an extremely rare presentation of the early phase of a SDAVF preceding myelopathy. Our experience may help other clinicians recognize SDAVF as the potential cause of radiculopathy, especially in the absence of typical symptoms.

Disclosures:

Conflict(s) of Interest: None

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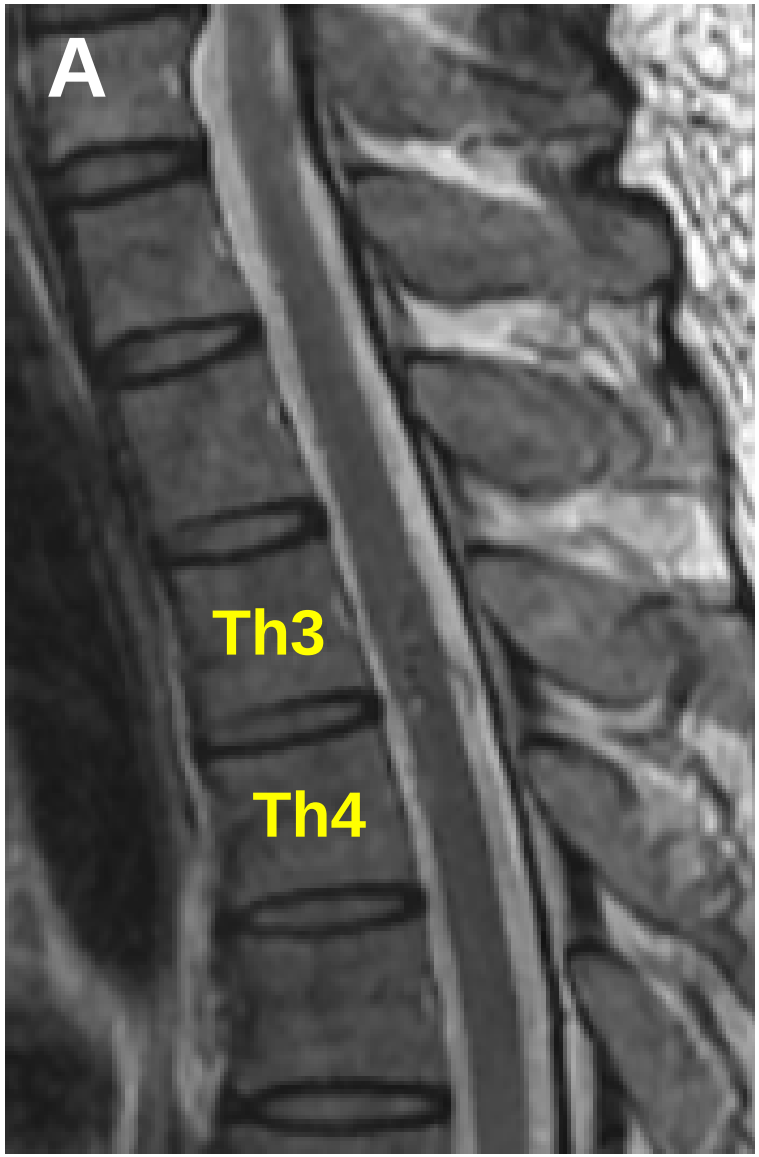
Figure Captions:

Figure 1. Preoperative spinal imaging. (A) Preoperative sagittal T2-weighted image of the thoracic lumbar spine. T2-weighted image shows only the flow void sign on the dorsal aspect of the spinal cord without evidence of signal change or cord swelling. (B) Preoperative coronal computed tomography (CT) angiography shows the feeding artery traveling along the dural sac, and the abnormally dilated intradural veins. (C) Preoperative axial CT angiography at the Th3 level. (D) Preoperative axial CT angiography at the Th4 level.

Figure 2. Digital subtracted angiography. Selective angiography of the right intercostal arteries at Th3 level shows the spinal dural arteriovenous fistulas fed by the Th3 radicular artery (*), making the shunt point (**), and draining into the intradural veins (***).

Figure 3. Operative view of the case. (A) Intraoperative photograph of the spinal dural arteriovenous fistula with an arterialized and dilated vein (*) compressing the right Th4 rootlet (**). (B) After resection of the vein and rootlet, impression of the rootlet is seen. (C) The draining vein penetrates the inner surface of the dural sac.

Figure 4. Schema of the vessel structure. The feeding artery goes through the dura mater from the right Th3. The draining vein passes through the inner surface of the dura mater at the level of Th3/4 and compresses the right Th4 rootlet.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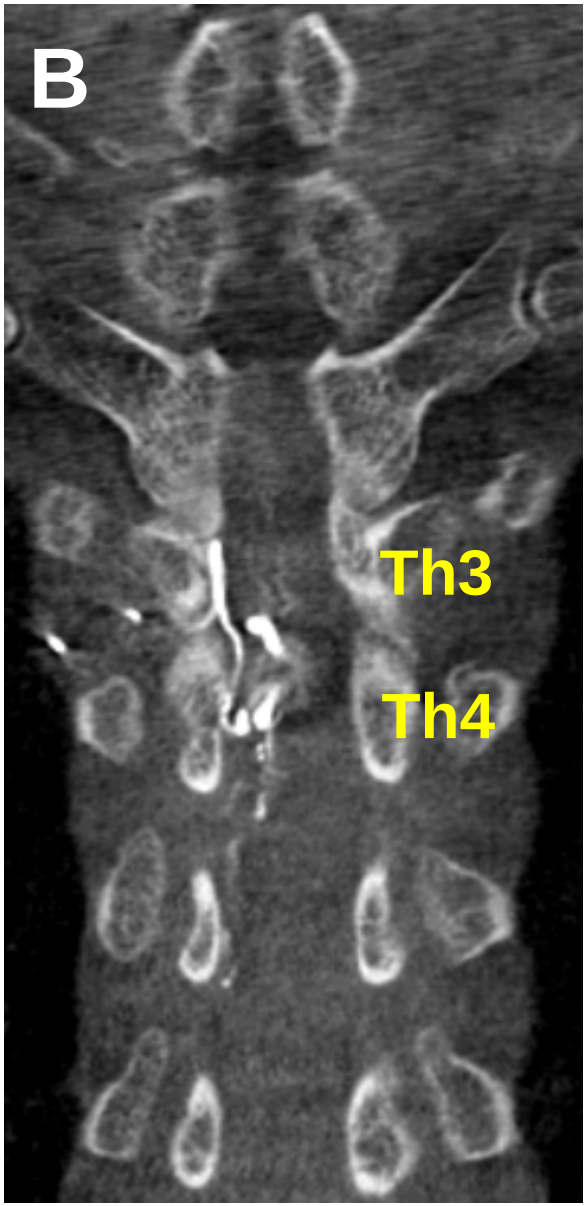
Th3
Th4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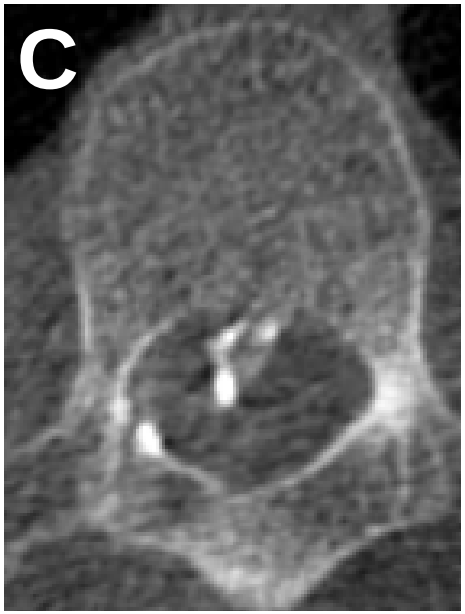
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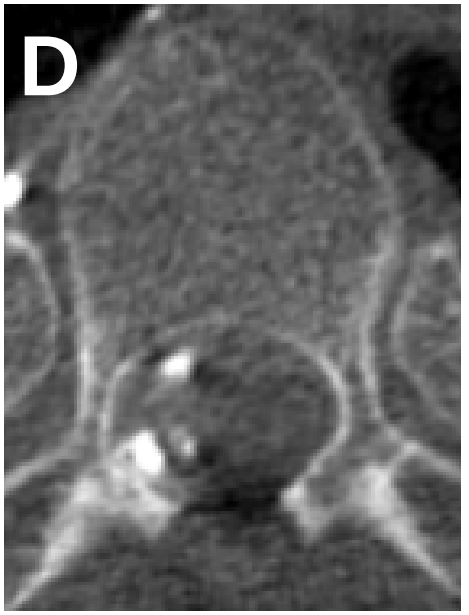
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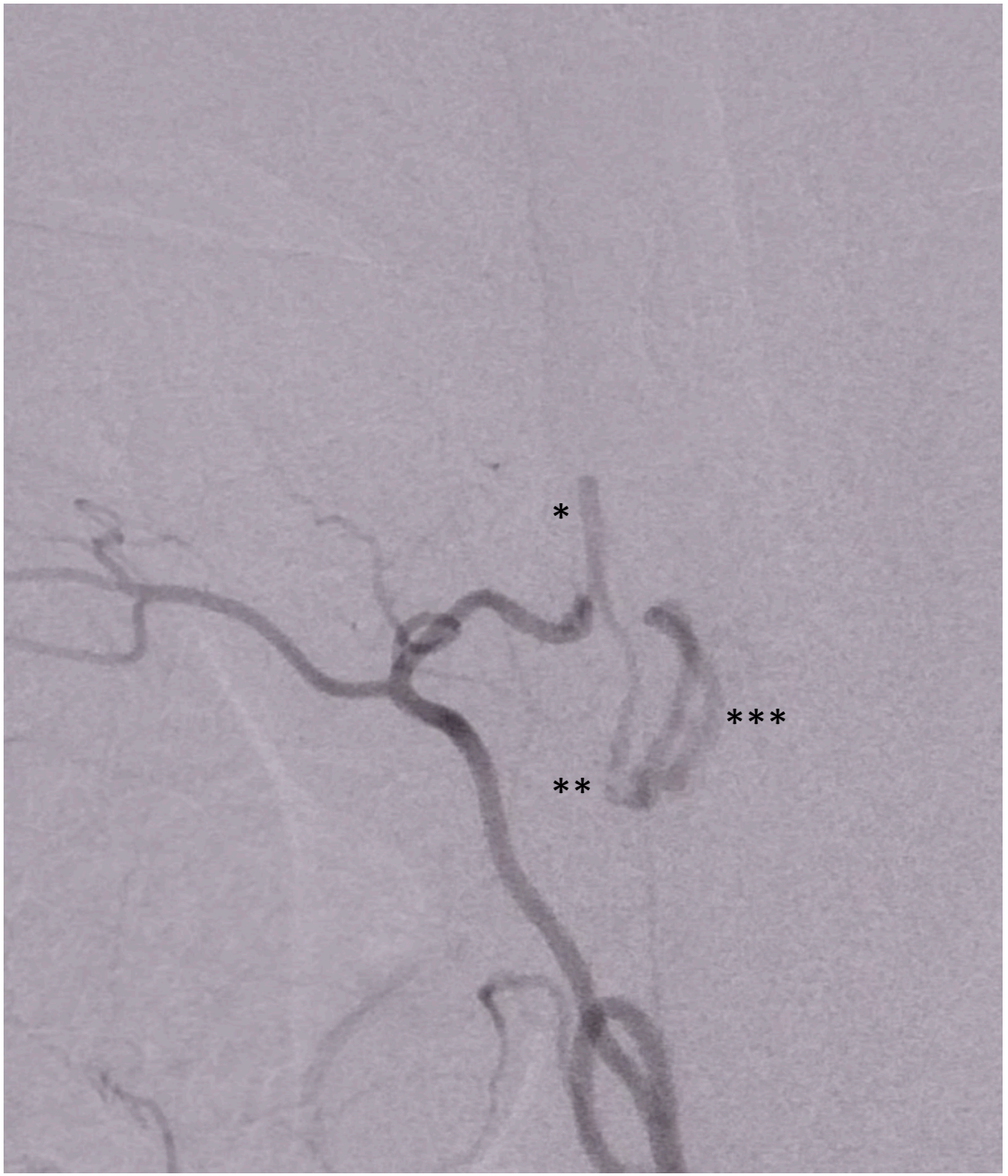

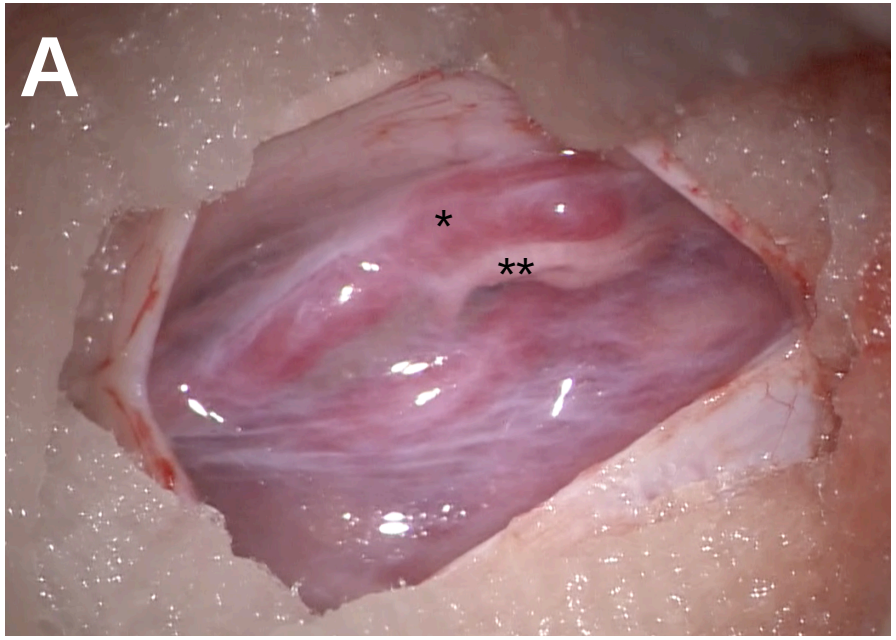
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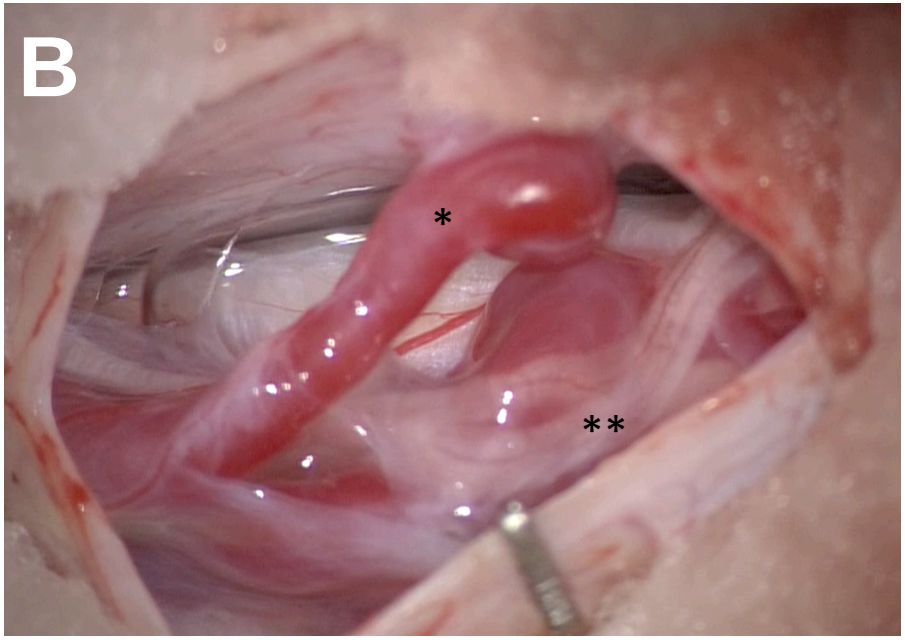
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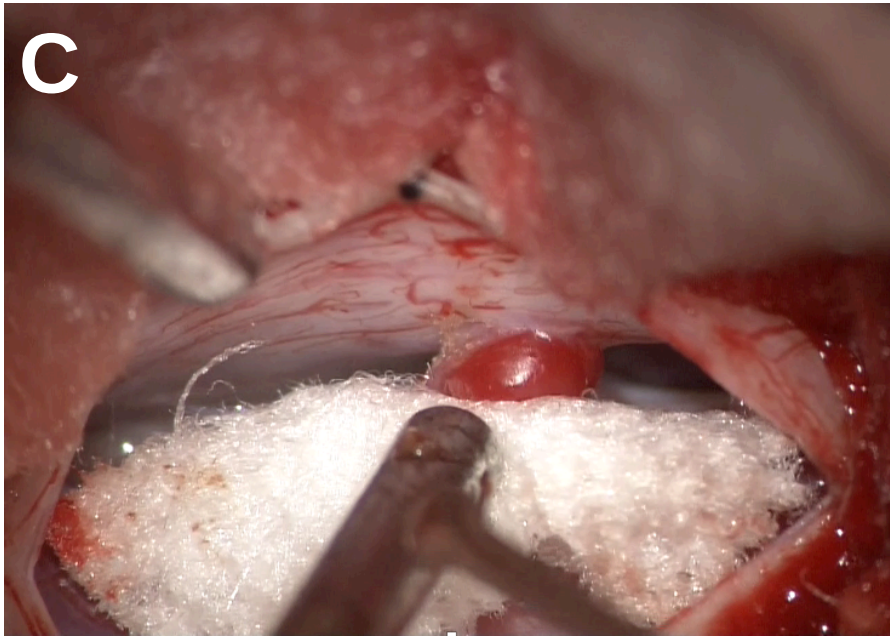
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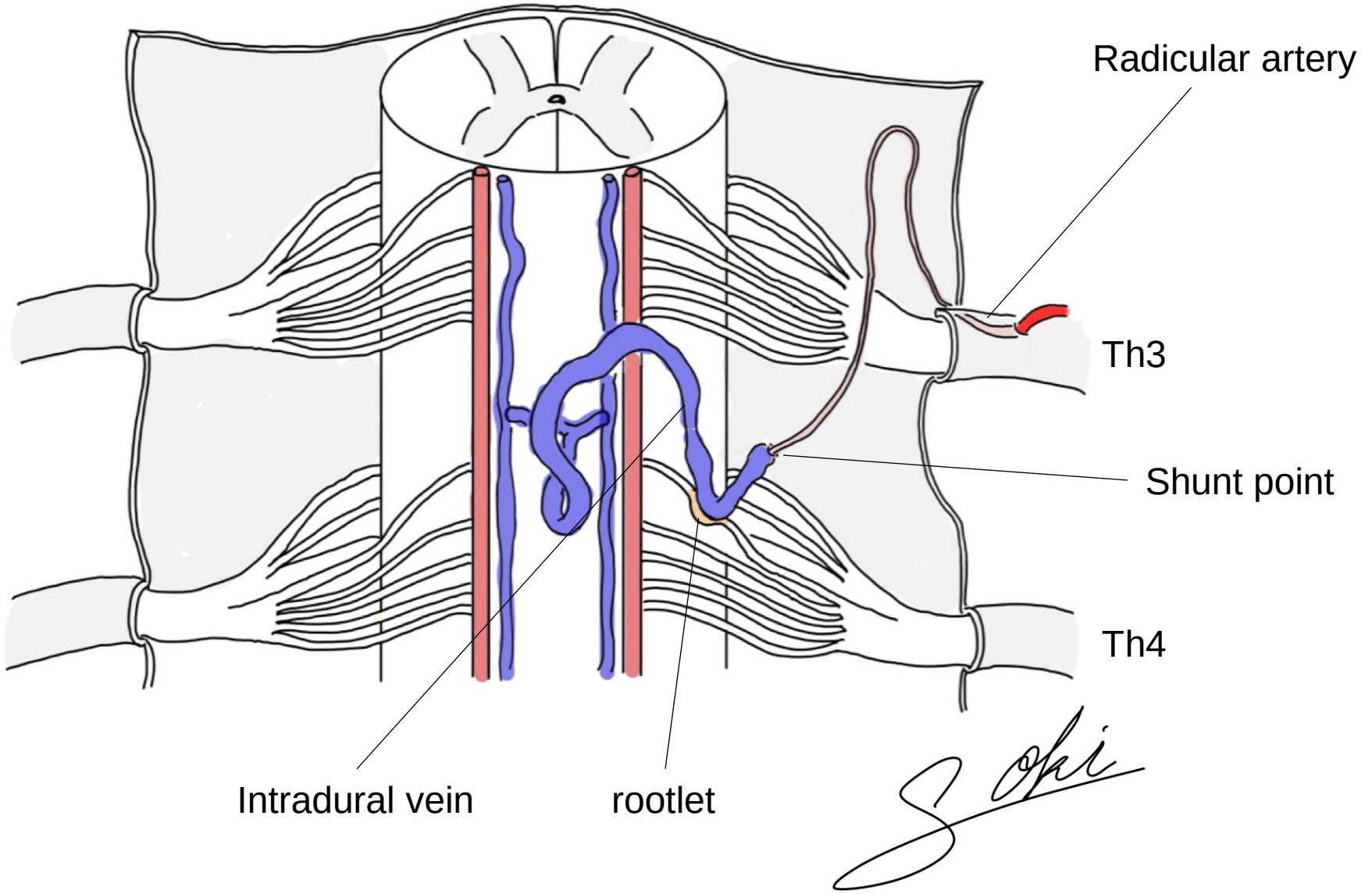
Radicular artery
Th3
Shunt point
Th4
Intradural vein
rootlet