

Case Report

Intramedullary hemorrhage from a thoracolumbar dural arteriovenous fistula

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Abstract

BACKGROUND CONTEXT: Spinal dural arteriovenous fistulas (AVFs) are acquired lesions presenting typically with neurologic deficits secondary to chronic congestive myelopathy. The low-flow and low-volume nature of these lesions makes hemorrhage very unlikely, and intramedullary hemorrhage caused by thoracolumbar dural AVFs is exceedingly rare.

PURPOSE: The purpose of this study was to report a case of intramedullary hemorrhage caused by a thoracolumbar dural AVF.

STUDY DESIGN/SETTING: The study design included a case report and review of literature.

METHODS: A case of intramedullary hemorrhage from a thoracolumbar dural AVF was reported, and the literature regarding hemorrhagic presentations of dural AVF was reviewed.

RESULTS: A 66-year-old woman presented with a sudden onset of abdominal pain, paraplegia, sensory loss below the costal margins, and urinary retention. Magnetic resonance imaging scan showed intramedullary hemorrhage with abnormal flow voids raising suspicion of an intramedullary AV malformation. However, subsequent selective spinal angiography demonstrated a spinal dural AVF fed by the T7 intercostal artery and a varix within the draining vein. Complete obliteration of the dural AVF and the varix was achieved via embolization. As far as we are aware, there are only two other similar cases in the literature. Literature review revealed that presentation of thoracolumbar dural AVFs with hemorrhage is frequently associated with accelerated venous flow and the presence of a venous varix.

CONCLUSIONS: Although very unusual, a spinal dural AVF may present with intramedullary hemorrhage, and hemorrhage in such conditions may be associated with an accelerated venous flow and the presence of a venous varix. © 2015 Elsevier Inc. All rights reserved.

Keywords:

Dural arteriovenous fistula; Intramedullary hemorrhage; Venous varix; Accelerated venous flow; Embolization; Spinal

Introduction

Spinal dural arteriovenous fistulas (AVFs) are acquired lesions presenting typically with neurologic deficits secondary to chronic congestive myelopathy. The low-flow and low-volume nature of these lesions makes hemorrhage very unlikely, but cases reporting subarachnoid

and subdural hemorrhages do exist, albeit predominantly occurring intracranially [1] and within the cervical region. [2–9]. Intramedullary hemorrhage caused by thoracolumbar dural AVFs is exceedingly rare. Here, we report such a case and present a review of the relevant literature.

Case report*History and examination*

A 66-year-old woman with no medical history of note presented with a sudden onset of severe right-sided abdominal pain radiating to the back, immediately followed by

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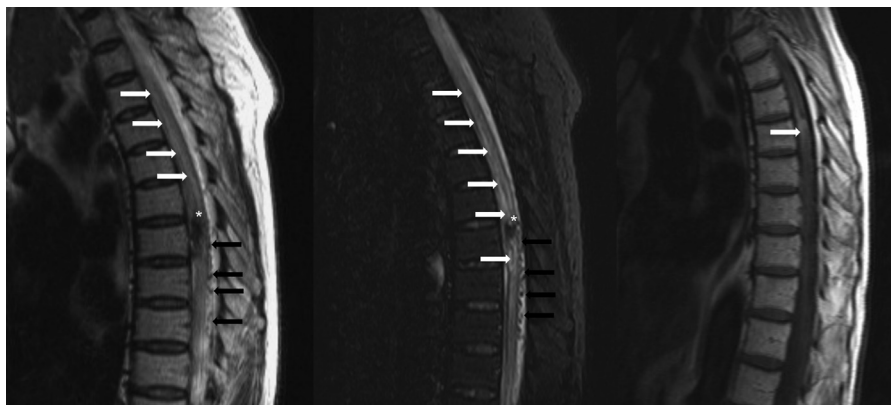


Fig. 1. Preoperative magnetic resonance (MR) images. Sagittal T2 and T2*-weighted MR images of the spine (Left and Middle) show a focal area of low T2 signal at T7 (asterisks), with associated high T2 signal intensity within the spinal cord extending from C7 to T7 (white arrowheads). Flow voids are noted on the posterior aspect of the spinal canal (black arrowheads). T1-weighted MR image (Right) reveals high T1 intensity at T7 (white arrowhead), consistent with subacute hemorrhage.

complete loss of power in lower limbs and immobility. On admission to the local hospital, the pain had become more severe, and neurologic examination revealed a paraplegia, sensory loss below the costal margins, and urinary retention. There were no other symptoms.

Imaging findings

T2- and T2*-weighted magnetic resonance imaging of the whole spine (Fig. 1, Left and Middle) demonstrated a focal area of low T2 signal within the cord (susceptibility artifact) at T7, consistent with intramedullary hemorrhage, and extensive high T2 signal involving the cord extending from C7 to T7 (edema). There were prominent signal voids posterior to the cord extending from cervicothoracic junction down to the conus. Magnetic resonance angiography (Fig. 2) revealed tortuous dilated vessels with a possible nidus in the cord, raising suspicion of an intramedullary AV malformation. Selective spinal angiography (Fig. 3), however, revealed a dural AVF arising from the right T7 intercostal artery with an associated venous varix within the draining radiculospinal vein. The anterior spinal axis

received supply from the right T10 and left L1 intercostal arteries, respectively.

Intervention

Embolization of the dural AVF was performed under general anesthesia. The right T7 intercostal artery was cannulated with a left coronary catheter. Distal access was obtained with a 1.2-F Sonic microcatheter, and 25% Histoacryl (*N*-butyl-2-cyanoacrylate) was injected resulting in the penetration of the origin of the draining vein and the dural AVF. The patient's neurologic deficit remained unchanged after the procedure and at 1 month before discharge for further rehabilitation.

Discussion

Spinal dural AVFs represent the most common type of spinal vascular malformations. These acquired lesions are usually found in the thoracolumbar region, but they can occur at any point along the dura of the spinal canal. A dural

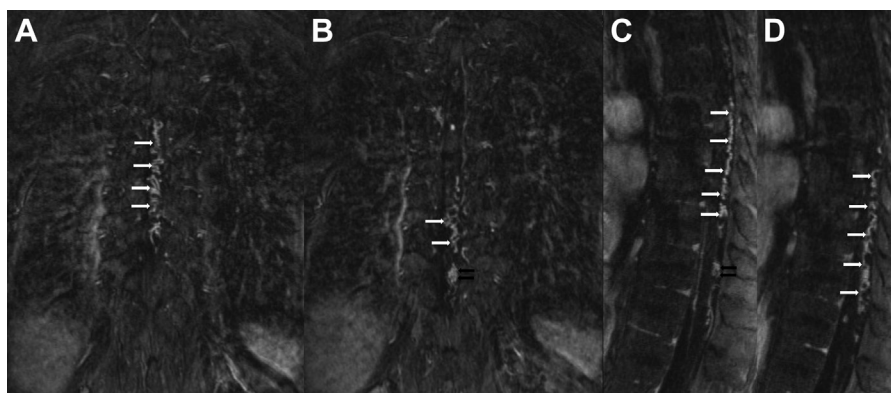


Fig. 2. Preoperative magnetic resonance (MR) angiograms. Coronal (A and B) and sagittal (C and D) images of contrast-enhanced MR angiogram in early (A and C) and late (B and D) phases demonstrate tortuous dilated veins (white arrowheads) with the presence of a venous varix (black arrowheads).

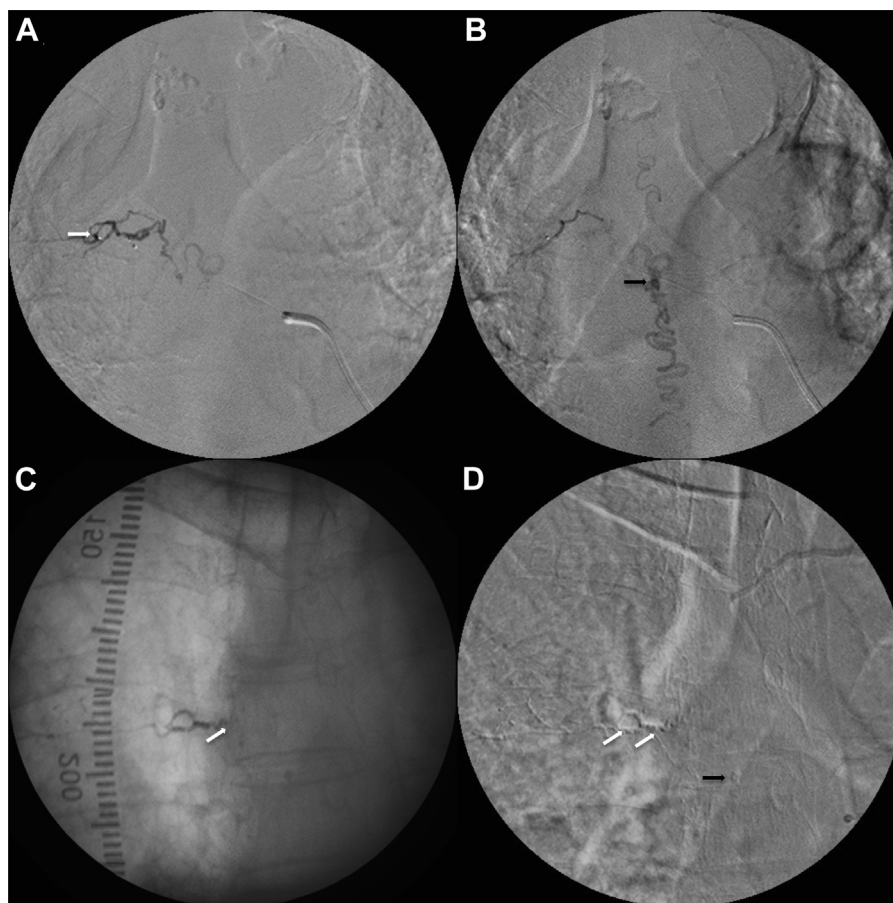


Fig. 3. Preoperative selective angiography images of early (A) and late (B) arterial phases reveal the presence of a dural arteriovenous fistula (white arrowhead) arising from the right T7 intercostal artery and an associated venous varix (black arrowhead) in the draining vein. Postoperative selective angiography images show penetration of the origin of the draining vein (white arrowhead) (C) and visible glue cast within the dural AVF (white arrowhead) and within the venous varix (black arrowhead) (D).

AVF usually consists of a single arterial feeder from a radicular artery that enters the dura at the nerve root sleeve. It is drained by retrograde flow through the medullary vein. Subsequent thrombosis or aging-related fibrosis leads to the engorgement of the coronal venous plexus and intraparenchymal radial veins. The resultant chronic venous hypertension and secondary ischemia give rise to the characteristic progressive myelopathy.

Because of the low-flow and low-volume nature of these lesions, hemorrhage is uncommon [10], but cases reporting subarachnoid and subdural hemorrhages have been reported, albeit predominantly occurring intracranially and within the cervical region with reported incidences ranging from 30% to 68% [1–9,11,12]. A literature search revealed the incidence of hemorrhage in thoracolumbar dural AVFs to be as low as 0.89% (Table), significantly lower than that in the cervical region. In addition, the data regarding intramedullary hemorrhage caused by spinal dural AVFs are scarce; to date, only two cases [18,19] have been reported. As such, alternative diagnoses such as cavernomas or other spinal vascular malformations that present more commonly with hemorrhage (perimedullary AVFs or

AV malformations) [10] should be first considered as differential diagnoses. In our case, selective spinal angiography, which is the gold standard for the diagnosis of spinal dural AVFs, unequivocally demonstrated the presence of the fistula (Fig. 3).

Why hemorrhage is more likely in some cases of spinal dural AVFs compared to others is unclear. Several studies have shown that hemorrhage in these lesions is associated with an accelerated venous flow and the presence of a venous varix. Kinouchi et al. [108] in a study on cervical dural AVFs reported that seven (77.8%) and five (55.6%) out of nine patients presenting with subarachnoid hemorrhage had increased venous flow rate and a venous varix, respectively, whereas none of the patients presenting without hemorrhage had a venous varix. Similarly, Aviv et al. [2] found that a venous varix was present in 7 (35%) of the 20 cervical dural AVF patients presenting with subarachnoid hemorrhage, compared with 1 (5%) in the nonhemorrhage group. These findings have been corroborated in a series of intracranial dural AVFs [109]. In our patient, the rate of arteriovenous transit was faster than that of other patients presenting without hemorrhage (1 vs. 2–3

Table

Summary of cases involving thoracolumbar dural AVF with or without hemorrhage and the presence of a venous varix

Study	Number of thoracolumbar dural AVF cases	Number of cases with hemorrhage (type)	Number of cases with a venous varix
Cases with hemorrhage			
Cho et al. [13]	28	2 (subarachnoid)	0
Dhandapani et al. [14]	21	2 (subarachnoid)	0
Han et al. [15]	1	1 (subdural)	0
Koch et al. [16]	1	1 (subarachnoid)	1
Lee et al. [17]	1	1 (subarachnoid)	1
Mascalchi et al. [18]	1	1 (intramedullary)	1
Minami et al. [19]	1	1 (intramedullary and subarachnoid)	1
Tan et al. [20]	1	1 (subarachnoid)	1
This study	1	1 (intramedullary)	1
Cases with no hemorrhage			
Aadland et al. [21]	12	0	0
Afshar et al. [22]	18	0	0
Aggarwal et al. [23]	1	0	0
Aghakhani et al. [24]	6	0	0
Ahlhelm et al. [25]	1	0	0
Andres et al. [26]	17	0	0
Apostolova et al. [27]	1	0	0
Atkinson et al. [28]	75	0	4
Behrens and Thron [29]	20	0	0
Blackburn et al. [30]	17	0	0
Bléhaut et al. [31]	10	0	0
Bowen et al. [32]	8	0	0
Bradac et al. [33]	13	0	0
Burrows et al. [34]	1	0	0
Cabrera et al. [35]	1	0	0
Cenzato et al. [36]	37	0	0
Cenzato et al. [37]	1	0	0
Cordato et al. [38]	1	0	0
Criscuolo et al. [39]	1	0	0
Deen et al. [40]	1	0	0
Eskandar et al. [41]	18	0	0
Finsterner et al. [42]	1	0	0
Foote et al. [43]	1	0	0
Gilbertson et al. [44]	55	0	0
Hage et al. [45]	5	0	0
Hall et al. [46]	3	0	0
Hanakita et al. [47]	2	0	0
Hassler et al. [48]	18	0	0
Hasuo et al. [49]	2	0	0
Houdart et al. [50]	1	0	0
Huffmann et al. [51]	18	0	0
Hurst et al. [52]	1	0	0
Inci et al. [53]	1	0	1
Isu et al. [54]	2	0	0
Jellema [55]	75	0	0
Kaut et al. [56]	1	0	0
Kirsch et al. [57]	67	0	0
Koch et al. [58]	50	0	0
Koenig et al. [59]	20	0	0
Koerts et al. [60]	1	0	0
Kota et al. [61]	1	0	0
Krings et al. [62]	2	0	0
Lai et al. [63]	7	0	0
Lai et al. [64]	1	0	0
Larsson et al. [65]	3	0	0
Lee et al. [17]	9	0	0
Linden and Berlit [66]	9	0	0
Marquardt et al. [67]	28	0	0
Mascalchi et al. [68]	4	0	0

(Continued)

Table
(Continued)

Study	Number of thoracolumbar dural AVF cases	Number of cases with hemorrhage (type)	Number of cases with a venous varix
Masuo et al. [69]	1	0	0
Matsubara et al. [70]	2	0	0
Meder et al. [71]	1	0	0
Mourier et al. [72]	29	0	0
Muralidharan et al. [73]	1	0	0
Nichols et al. [74]	14	0	0
Niimi et al. [75]	49	0	0
Oldfield et al. [76]	6	0	0
Park et al. [77]	15	0	0
Pierot et al. [78]	2	0	0
Pillai et al. [79]	1	0	0
Prieto et al. [80]	1	0	0
Rizvi et al. [81]	2	0	0
Roccatagliata et al. [82]	1	0	0
Rosenblum et al. [10]	27	0	0
Sakamoto et al. [83]	1	0	0
Saladino et al. [84]	137	0	0
Sato et al. [85]	10	0	0
Shedid and Podichetty [86]	1	0	0
Shiban et al. [87]	1	0	0
Shigematsu et al. [88]	3	0	0
Shimizu et al. [89]	1	0	0
Shinoyama et al. [8]	20	0	0
Sleiman et al. [90]	10	0	0
Song et al. [91]	18	0	0
Stevens et al. [92]	1	0	0
Sugawara et al. [93]	1	0	0
Suzuki et al. [94]	1	0	0
Symon et al. [95]	51	0	0
Takai et al. [96]	28	0	0
Terwey et al. [97]	11	0	0
Touho et al. [98]	3	0	0
van Dijk et al. [99]	2	0	0
Woodall et al. [100]	1	0	0
Wu et al. [101]	1	0	0
Xia et al. [102]	3	0	0
Yamaguchi et al. [103]	8	0	0
Yamaguchi et al. [104]	3	0	0
Yang et al. [105]	32	0	0
Yoshino et al. [106]	1	0	0
Zhao et al. [107]	3	0	0
Total	1,242	11	11

AVF, arteriovenous fistula.

seconds), and the draining vein was associated with a venous varix (Figs. 2 and 3).

Given the lack of data on hemorrhage in thoracolumbar dural AVFs, we performed a literature search comparing cases of thoracolumbar dural AVFs presenting with hemorrhage to those that do not, with regard to the presence of a venous varix (Table). Of the 1,242 reported thoracolumbar dural AVF cases to date, 11 (0.89%) patients presented with hemorrhage and 6 (0.48%) of these were found to have a venous varix within the draining vein. In contrast, only 5 of the 1,231 reported cases of thoracolumbar dural AVFs that presented without hemorrhage (0.41%) were associated with a venous varix. We cannot be sure whether the

description of a venous varix was included in each case because of the rarity of hemorrhage as a presenting symptom or for some other reasons. The formation of a venous varix is probably because of a combination of accelerated venous flow, increased venous pressure, and anatomic changes of the venous vessel wall; these varices may then rupture, causing a hemorrhage. Why these changes occur in some spinal dural AVFs but not in the others remains unknown, but the finding of a venous varix associated with an increased venous flow in a spinal dural AVF is a pointer of future risk of haemorrhage in such lesions.

In cases with no hemorrhage, the outcome of spinal dural AVFs after timely intervention is generally considered

to be very good, with 90% of patients demonstrating stabilization or improvement of symptoms [13,73,110]. There is currently limited data on the outcome of cases with hemorrhage caused by thoracolumbar dural AVFs, which probably reflects the rarity of this presentation. In our literature review, of the 11 patients with thoracolumbar dural AVFs presenting with hemorrhage (including our case), 9 experienced either stabilization (n=6) or improvement (n=3) in function (data not shown). Clinical status before surgery or embolization and early interventions have been shown to be the most important predictors of good outcome [57,111]. Nevertheless, a few reports have shown significant improvements even after advanced neurologic deficits before treatment [24,80] and even after a significant delay in treatment [56].

Conclusions

Although very unusual, a dural AVF may present with intramedullary hemorrhage, and hemorrhage in such conditions may be associated with an accelerated venous flow and the presence of a venous varix.

Acknowledgment

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