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Original Article

Double spinal dural arteriovenous fistulas

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ABSTRACT

Background. – Spinal dural arteriovenous fistulas (SDAVF) are usually solitary lesions. Synchronous and/or metachronous double SDAVF have rarely been reported in the literature. We report on three patients with double SDAVF and present our single center experience in the diagnostic and treatment management in these patients.
Material and methods. – We retrospectively revised our medical database for all patients who were diagnosed and treated in our center due to a SDAVF between 1990 and 2017. All data including demographics, clinical presentations, as well as radiological data were re-evaluated for this study.
Results. – Three (1.4%) of 209 consecutive patients with SDAVF presented double SDAVF with different arterial feeders and venous drainage patterns. All three patients were men. The mean age at time of diagnosis was 67.9 ± 10 years (median; 68, range: 53–82). Myelopathic symptoms were reported in all three cases. All three fistulas were located in the thoracolumbar region between T7 and L2. MRI/CE-MRA showed medullar T2-hyperintensity, intramedullary contrast-enhancement and dilatation of perimedullary veins in various extensions.
Conclusion. – Double SDAVF are extremely rare and were found in 1.4% of patients in our series. The vast majority of the reported double SDAVF in the literature has been detected synchronously within an area of equal or less than three vertebral levels. Thus, whenever the SDAVF is identified, further injections of the fistula-zone neighbored segmental arteries might be recommended. However, due to the extremely low incidence of double SDAVF a complete spinal DSA is not indicated.
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Introduction

SDAVF is the most frequent vascular disease of the spinal cord and its meninges and accounts for about 70% of all spinal arteriovenous (av) pathologies of the spinal cord and its meninges. The disease becomes symptomatic predominantly in elderly men between 40 and 60 years [1,2].

SDAVF are usually solitary lesions and are commonly localized in the thoracolumbar region, They may be encountered from the sacral region, supplied via spinal branches of the iliolumbal arteries or the middle sacral artery, to the level of the foramen magnum supplied via cervical feeders including both vertebral arteries, the thyreocervical and costocervical trunk and the ascending cervical artery [3–5]. The venous drainage in all these spinal arteriovenous shunts occur typically transdurally into the coronal venous plexus producing medullar venous hypertension that leads to swelling and edema within the spinal cord and accompanies the characteristic symptoms of a slowly progressive paraparesis, bowel and bladder dysfunction and sensory disturbances [6].

The fistula-zone itself is predominantly located at or in the nerve root sleeve, supplied by meningoradicular branches of a segmental artery and draining transdurally into the coronal venous plexus [7].

However, multiple SDAVFs have rarely been reported in the literature. (Table 1) In this study we report on the clinical and radiologic features of three patients with double SDAVF and present our single center experience in the diagnostic and treatment management in this patients series.

Material and methods

After obtaining permission from our local ethics board, we retrospectively revised our medical database for all patients who were diagnosed and treated in our center due to a SDAVF between 1990 and 2017.

The initial diagnosis of SDAVF was based on clinical and radiological criteria that comprised:

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Table 1
Overview of all twenty-eight cases of double spinal dural arteriovenous fistulas reported in the literature (including present cases). n.d.: no data.

Author	Age and sex	Timing of diagnosis	Location			Treatment
			Lesion 1	Lesion 1	Lesion 3	
Thiebot et al. [31], 1986	24, F	synchronous	T4 r	T12 l	L4, L5 r	n.d.
Merland et al. [32], 1985	n.d.(4 cases)	n.d.	n.d.	n.d.		n.d.
Barnwell et al. [33], 1991	69, F	metachronous	C1-C2 r	C5-C6 r		surgery
Pierot et al. [14], 1993	47, M	synchronous	T6l	T8r		combined
	64, M	synchronous	T8 r	T9 l		surgery
Chaloupka et al. [34], 1995	48, M	synchronous	L1 l	T9 l		combined
Dam-Hieu et al. [35], 2001	49, M	synchronous	T6 r	L1r		surgery
van Dijk et al. [36], 2002	62, M	metachronous	T5 (n.d.)	T9 (n.d.)		(n.d.)
El-Serwi et al. [37], 2006	47, M	synchronous	T7 r	T5 r		embolization
Sugawara et al. [38], 2005	72, M	metachronous	T6	L1		surgery
Rizvi et al. [39], 2006	50, M	metachronous	T9	L-1		combined/surgery
Shankar et al. [40], 2011	61, M	synchronous	C1,C2 l	C6 l	C3, C6 r	embolization
Ge et al. [41], 2013	30, M	synchronous	T12 l	L1 l		surgery
	62, M	synchronous	T6 l	T3 r		embolization
Oshita et al. [42], 2011	74, M	synchronous	C1	C1		surgery
Cenzato et al. [43], 2007	45, M	synchronous	T5 r	T6 l		surgery
Dagar et al. [44], 2010	58, M	metachronous	T12 l	L1 r		surgery
Hanakita et al. [45], 2012	57, M	synchronous	T7r	T12 l		surgery
Hetts et al. [46], 2013	40, M	synchronous	C5-C6 l	C5-C6 r		surgery
Avecillas-Chasín et al. [47], 2015	72, M	metachronous	T7 r	T12 r		embolization/surgery
	51, M	metachronous	C1 r	C1 l		surgery
Kaku et al. [48], 2017	56, M	metachronous	C1 r	S2 r		surgery
Present cases						
1 [26]	63, M	metachronous	L1 r	L2 l		surgery
2	65, M	synchronous	T8 l	T9 r		surgery
3	51, M	synchronous	T7 l	T7 r		surgery

- clinical sequelae of myelopathy resulting in motor or sensory disturbances with or without vegetative bladder-bowel dysfunctions and;
- MR imaging findings of congestive myelopathy of the spinal cord and/or visibly engorged perimedullary veins.

All suspected cases of SDAVF were verified by DSA. All clinical data including demographics, clinical and neurological presentation, as well as radiological data were assessed by the primary treating physicians and re-evaluated for this study.

Results

We identified 209 consecutive patients with angiographically verified SDAVF. Three (1.4%) of these 209 patients presented double SDAVF and were included in the recent study. (Table 1)

All three patients were men. The mean age at time of diagnosis was 67.9 ± 10 years (median; 68, range: 53–82). All three patients presented with paresis, gait ataxia, and sensory deficits in the lower extremities in various severities. Sphincter dysfunction was present in one of these three patients at time of admission in our center.

All SDAVF were located in the thoracolumbar region between T7 and L2. T2w-hyperintense signal and elongation and/or dilatation of perimedullary veins were present in all three patients. There was intramedullary contrast enhancement in two of these three patients. All three patients underwent a microsurgical interruption of their fistulas. One patient showed postoperatively a space occupying epidural hemorrhage in the post-operative MRA. The hematoma was surgically evacuated without further procedural complications.

Illustrative case

This 51-year-old man complained of a one-year history of weakness and ataxia in the lower extremities, accompanied by cramping pain in both thighs. During the year he had also developed paraesthesia in the dorsal and medial site of the both thighs and feet. Slowly progressive urinary and bowel incontinence followed. At time of admission neurological examination revealed proximal accentuated spastic paraparesis grade 3/5 with gait ataxia and brisk tendon reflexes in the lower extremities. All sensory modalities were reduced in the dorsal site of the both thighs. Magnetic resonance imaging (Fig. 1) revealed a medullar edema of the thoracic spinal cord from T 6 to the conus at L1 vertebral level with centromedullary hyperintensity in T2-weighted images. No medullar contrast enhancement was shown. Enlarged and tortuous perimedullary veins became apparent by flow voids and contrast enhancement, extending from the craniocervical region to T9 vertebral level dorsally to the spinal cord. CE-MRA revealed a pathologic early arterialization of the enlarged perimedullary veins suggesting a SDAVF at the level of T6/T7 intervertebral foramen. Spinal angiography showed a dural arteriovenous shunt at the level of the intervertebral foramen T6/T7 supplied by meningeal branches of the left T7 segmental artery as assumed in the CE-MRA. Further examinations of the ipsilateral and contralateral segmental arteries showed a bi-segmental arterial supply of the left av shunt at T7 via a dural branch from the right T6 segmental artery. The venous drainage of this fistula was ascending to cranicervical region continuing into the left sigmoid sinus. Interestingly, we identified an additional SDAVF localized at the level of the right T6/T7 intervertebral foramen and supplied by the right T7 segmental artery showing a different and less prominent ascending venous drainage pattern. Both fistulae were occluded microsurgically in the same session via laminectomy of T7.

Discussion

Though SDAVFs are considered to be an acquired disease, their precise etiology is still unknown [7]. In the presence of an av shunt in the spinal dura, venous hypertension in the coronal venous plexus causes usually a chronic reduction of the intramedullary av pressure gradient [8]. These hemodynamic changes were considered in the literature to be the most pathophysiological contributing factor of congestive myelopathy [9–11]. Nevertheless, for successful treatment management a sufficient opacification and

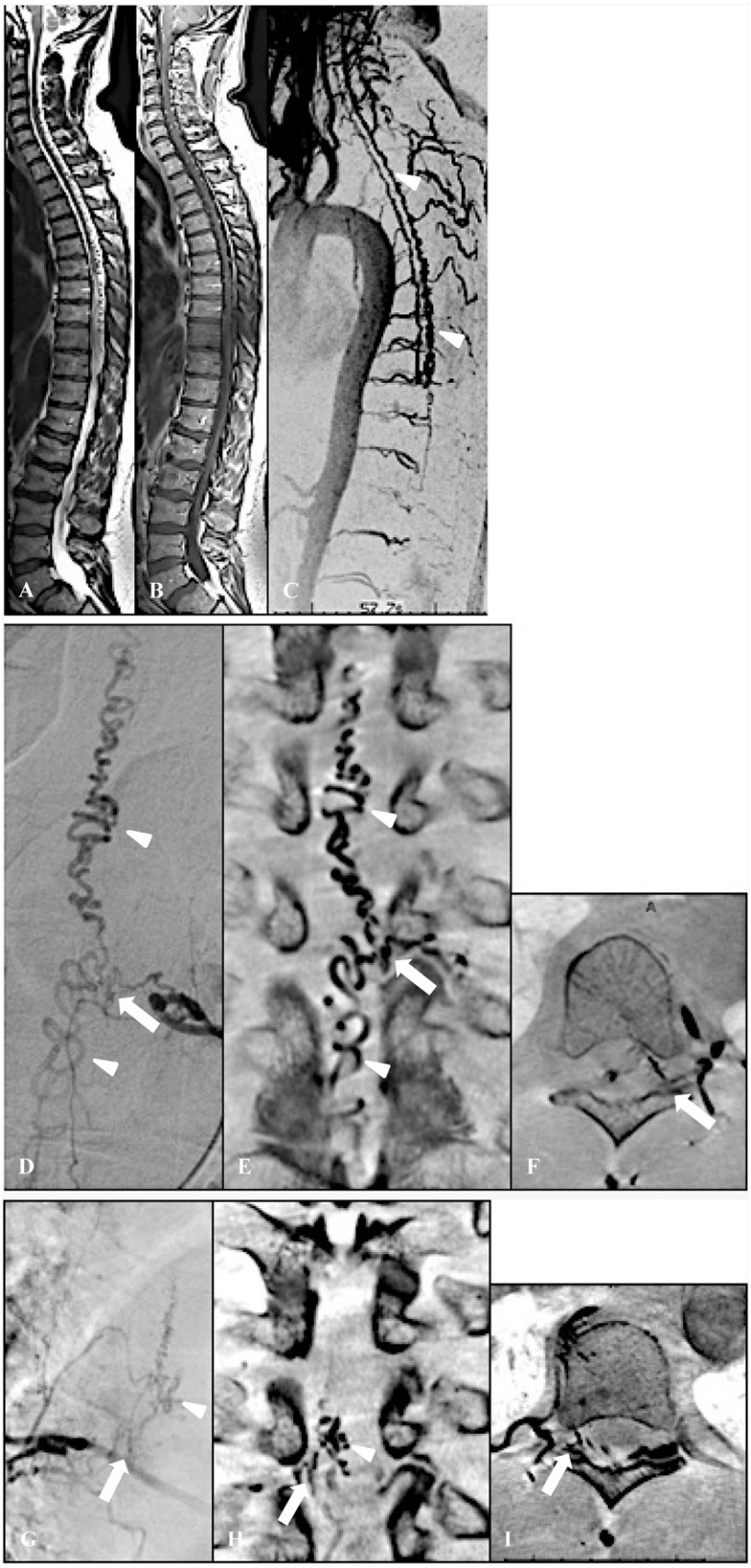

Fig. 1. A–B. Sagittal T2- and contrast-enhanced T1-weighted images show medullar edema in the thoracic region and extensive dilated perimedullary veins. C. Spinal CE-MRA (MIP) reveals abnormal arterialized perimedullary veins in the thoracic and cervical region (white arrowhead). D–F. DSA examinations in pa projection as well as axial and coronar MPR of Dyna-CT of the left T7 segmental artery identify a SDAFV (white arrow) and arterialized perimedullary draining veins (whithe arrowhead). G–I. Further DSA examinations of contralateral segmental artery reveal a second fistula (white arrow) supplied via the right T7 segmental artery and presenting a different and less prominent preimedullary drainage vein (white arrowhead).

profound understanding of the angiomorphology of the fistula via spinal MRI/CE-MRA and DSA are mandatory [12].

Among 209 SDAVF patients who were treated in our institution between 1990 and 2017, only three (1.4%) cases presented a double SDAVF with different arterial feeders, distant fistula zones and various venous drainage patterns.

Multiple SDAVFs have been mentioned in the literature only incidentally in case reports or in few larger case series without further specific descriptions. Table 1 provides an overview about all reported double SDAVF in the literature. An analysis of Merland et al. about 57 patients included four cases with double SDAVFs, estimating an incidence for double SDAVF of 7% in the respective series [13]. However, the authors did not specifically describe the location of neither the fistula zones nor the venous drainage pattern in these four patients. In another series of 50 patients, Pierot et al. reported on two (4%) patients with double SDAVF [14].

Nonetheless, sufficient data about the true incidence of double SDAVF is still missing in the literature. Various factors might hinder the precise estimation of the incidence of double SDAVF. On the one hand, classic SDAVFs are generally a rare entity; Thron et al. assumed an annual incidence for SDAVF of 5–10 cases per million [7]. Due to this low incidence of SDAVF and to the diagnostic difficulties, patients with suspected SDAVF are mainly referred to specialized centers with high expertise in diagnosing and treating this kind of spinal vascular diseases [15]. This might induce, in turn, a referral selection bias in statistical analysis of these patients. On the other hand, the variability of diagnostic, treatment and follow-up strategies in the various referral centers might result also in a discrepancy in the estimated rate of double fistula [16–19].

All three patients in our recent series have been referred to our center due to chronic progressive myelopathic symptoms and radiological findings evident of a SDAVF. Paraparesis and gait ataxia were present in all three patients at time of diagnosis. Case 2 suffered additionally bowel dysfunction at time of admission in our center. MR images in all three cases demonstrated medullar edema and pathologically enlarged perimedullar vessels in various extensions along the thoracolumbar region. Two of these three patients presented centromedullary contrast enhancement in various extensions.

Even though the very small sample size of our recent series does not allow a statistical analysis, we observed no major differences between the clinical presentations and MR findings of this series and those of patients with solitary SDAVF diagnosed in our center [1]. Nonetheless, the typical but non-specific neurological and radiological findings in SDAVF patients contribute, generally, to often misdiagnosing and subsequent delay of the correct diagnosis and treatment [20].

In the last two decades advanced diagnostic tools have been developed to facilitate the diagnosis of spinal vascular malformations [12,21]. Since 2004/2005, we perform initially spinal CE-MRA in SDAVF patients whenever it is possible [12,22]. CE-MRA may serve to detect, localize, and characterize, various types of spinal AV shunts with a high sensitivity [12]. In a previously reported series by our group, CE-MRA could correctly predict the level of the fistula within one vertebral level in nineteen patients with SDAVF [12]. Hence, we usually focus our DSA examinations primarily on the suspected region shown in the prior CE-MRA [12]. All three patients underwent a spinal selective DSA examination in our center for definite diagnosis.

Including the present cases, sufficient data about the timing of the diagnosis was available for twenty-five of all twenty- eight double SDAVF cases. In sixteen (64%) of these twenty-five patients the second fistula was localized in a distance of equal or less than three vertebral levels from the first identified one. Furthermore, in eleven (69%) of these sixteen cases the double SDAVF have been diagnosed synchronously during the same DSA examination.

This frequently close spatial relationship between double SDAVF might be based on common etiological factors. SDAVF were considered to occur secondary to thrombosis and/or outlet disorder of spinal venous system resulting in a so-called medullar venous congestion. Thus it is conceivable that, the presence of a single SDAVF could promote the development of a second fistula due to the subsequently increased medullar venous pressure and the concomitant venous stagnation and thrombosis in the adjacent veins.

Based on our own experience, once a SDAVF is identified further injections of all neighbored ipsi- and contralateral segmental arteries above and below the fistula-zone might be necessary for sufficient visualization of the angiomorphology. Nonetheless due to the:

- extremely rare incidence of double SDAVF and;
- due to the fact that the vast majority of reported double SDAVF were localized in a circumscribed area of the spinal dura and have been often synchronously identified, a routine complete DSA examination of all spinal cords feeding segmental arteries might not be indicated.

It might be otherwise recommended only in cases of discrepancy between CE-MRA findings and the verified fistula location on DSA, such like various suspected arterial feeder and/or different venous drainage patterns.

All three patients in our recent study underwent microsurgical interruption of their SDAVF. Data about treatment strategy of double SDAVF are available for twenty-one of all reported twenty-eight cases. Thirteen (62%) of these twenty-one cases underwent surgical treatment, other five (24%) cases received endovascular and surgical treatment, and the remaining three (14%) patients received solely endovascular embolization.

The main goal of treatment is to disconnect the av shunt and to reduce the venous congestion of the spinal cord by endovascular embolization and/or surgical interruption [16,23–25].

Surgical interruption was the first-line therapy for all SDAVF in our center [17]. Our treatment strategy is in line with various large representative series [19,27]. It is a safe, effective and relatively simple intervention with the exception of sacral fistulae [17]. In a large meta-analysis dealing with sixteen studies that provided information on surgical success rates Steinmetz et al. reported a success rate of 97.9% for surgical treatment regardless of SDAVF localization [19]. In contrast, the success rate of endovascular treatment reported in various studies ranged between 69.2% and 89.5% [28–30].

All included patients in our recent series received a neurological examination in a daily basis and a MRI examination within one week after surgery. We usually recommend, in all SDAVF patients, further clinical and MR follow-up examinations three, six, and twelve months after discharge. Extensive clinical and neuroradiological examinations should be, however, performed in case of progressive deterioration after treatment, persistence of symptomatology and/or pathological MRI findings six months after treatment, or in case of late neurological deterioration after initial improvement with no evidence of other possible contributing pathologies [12]. In those cases, CE-MRA and DSA re-examinations should comprise all spinal cord feedings arteries to exclude local recurrence, second SDAVF, or possible other spinal vascular pathologies.

Limitations

Major limitations of our study are the small sample size and the retrospective approach, both of which might provoke speculative interpretation of our data to some extent.

However, our recent observations may provide an overview of specific diagnostic and treatment strategies of this rare disease, in particular since the incidence of double SDAVF is extremely low and there are very few data concerning this fistuals in the literature.

Conclusion

Double SDAVF are extremely rare and have been yet reported in overall twenty-eight patients in the literature (including the present cases). The rate of double SDAVF in our series accounts for 1.4% of all SDAVF in our own series. The vast majority of the reported cases has been detected synchronously and the multiple fistulas were localized within an area of equal or less than three vertebral levels. Due to these observations, the routine opacification of the SDAVF neighbored ipsi- and contralateral segmental arteries might be recommended in all SDAVF patients, but a complete spinal DSA is not indicated once the dural av shunt is identified. However, if clinical and/or neuroradiological follow-up examinations revealed an evidence of a persisting av shunt, spinal CE-MRA and complete DSA are mandatory.

Disclosure of interest

The authors declare that they have no competing interest.

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