SHORT COMMUNICATION

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Endovascular embolisation as minimally-invasive treatment for spinal dural arteriovenous fistulas — evaluation of long-term results

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ABSTRACT

Aim of study. Spinal dural arteriovenous fistulas (sDAVF) are rare spinal cord lesions formed between a radicular artery and medullary vein leading to venous hypertension resulting in neurological impairment. Endovascular embolisation is a minimally-invasive method aiming to interrupt the shunt between the artery and vein. We report our experience with sDAVF treated endovascularly.

Material and methods. Clinical and procedural data of 16 consecutive patients diagnosed with sDAVF was reviewed. Pre- and post-operative neurological condition was evaluated using both the Aminoff and Logue disability scale and the VAS scale. Rates of complete occlusions, technical difficulties, and procedural complications were noted.

Results. Four of the patients were female and 12 were male; mean age was 62.4 years. Mean interval between symptom onset and treatment was 13.3 months. Complete occlusion was achieved in 88% (14/16 patients). Significant or moderate clinical improvement in long-term follow-up was observed in eight patients (50%). Recurrence was observed in two cases (13%).

Conclusions and clinical implications. While endovascular methods are being refined and thus achieving an increasing percentage of successful occlusions, patients should be closely monitored since this condition is recurrent and the clinical consequences of myelopathy can persist despite complete occlusion of the shunt.

Key words: spinal dural arteriovenous fistulas, endovascular, embolisation, outcome

Introduction

Spinal dural arteriovenous fistulas (sDAVF) are rare spinal cord lesions formed between a radicular artery and medullary vein leading to venous hypertension with subsequent radicular pain and extremity weakness (paresis), as well as loss of bowel and bladder function [1]. They are also the most common vascular malformations found within the spinal canal [2]. Because of the manifold clinical presentations, which resemble much more common diseases such as degenerative disc disease or polyneuropathies, the diagnosis is often delayed [3].

However, due to the fact that sDAVF can result in permanent spinal cord injury if left untreated, prompt and accurate diagnosis is crucial. Initial diagnosis is based on magnetic resonance (MR) findings, with special attention to MR angiography (MRA), but digital subtraction angiography (DSA) is necessary for a thorough understanding of the anatomical condition and planning of the therapeutic strategy [4, 5]. Current medical treatment includes both microsurgical and endovascular methods [6–8]. A recent multicentre study aimed at comparing these two techniques did not find significant differences in overall clinical outcomes, but concluded that patients undergoing embolisation have a higher risk of late

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Table 1. Aminoff and Logue scale of disability

Grade	Characteristics
Gait disturbance	
1	Leg weakness or abnormal gait, no restricted activity
2	Grade 1 with restricted activity
3	Requiring one stick/crutch for walking
4	Requiring two sticks/crutches/walker for walking
5	Unable to stand, confined to bed/wheelchair
Micturition	
1	Hesitancy, frequency, urgency
2	Occasional urinary incontinence or retention
3	Total urinary incontinence or retention

recurrence [9]. However, with constant technological improvements the rate of successful embolisation has been rising and due to the additional advantages of minimally-invasive embolisation (i.e. shorter hospitalisation and a less painful postoperative course), some centres now consider this as the treatment of choice in selected patients [10].

The aim of this study was to report the clinical outcomes, as well as the failure and recurrence rates, of 16 patients with sDAVF treated with endovascular means.

Material and methods

In this single-centre retrospective study, we evaluated the clinical and procedural data of 16 consecutive patients diagnosed with sDAVF and treated with endovascular embolisation between January 2014 and December 2020. All sDAVF were initially diagnosed with MR and MRA and subsequently confirmed with DSA examination. All cases were then reviewed by a multidisciplinary board which consisted of a neurosurgeon and a neuroradiologist. All cases amenable to endovascular treatment were referred for embolisation. Baseline clinical condition was evaluated using both the Aminoff and Logue disability scale and the VAS scale [11]. All patients gave informed consent prior to the procedure (Tab. 1).

All endovascular interventions were performed under biplane angiography unit with 3D rotational angiography and with patients under general anaesthesia. After selective catheterisation of the feeding artery with a microcatheter, definitive embolisation was attempted with N-butyl cyanoacrylate (NBCA; Cordis Microvascular Inc.) or ethylene vinyl alcohol (Onyx, Covidien) depending on the vascular condition of the fistula. Control angiography was performed afterwards.

All patients underwent at least one control DSA and/ /or MR examination as well as a neurological examination 3–6 months after the procedure. In cases of recanalisation/ /incomplete occlusion and no clinical improvement, the possibility of secondary embolisation or surgical intervention was discussed with the patient.

Results

In total, 16 patients met the inclusion criteria. The majority of patients were male (12, 75%) and the mean age on admission was 62.4 years (range 28 to 70). Average time from symptoms onset to endovascular procedure was 13.3 months. In terms of fistulas localisation, the most common on spinal angiography was at the level of T7 (four cases), with 13 fistulas located at the thoracic level and three at the lumbar level. All the malformations were dural spinal arteriovenous fistulas (pathological arteriovenous shunting between the leaflets of the dura), with draining vein on the dorsal surface of the spinal cord, and were classified as type 1 according to the Spetlzer et al. classification [12]. Complete and super-selective occlusion was achieved in 10 patients (63%). Five patients required secondary intervention due to late recurrence (four cases) or the presence of multiple feeders (one case). As far as post-procedural complications were concerned, in one case a transient paralysis of the lower extremities was noted, which resolved within three months of the procedure.

Imaging examination performed at the long-term follow-up (mean duration = 28 months, range 7 to 58) disclosed that complete occlusion of the fistula was achieved in 14 patients (88%). In the two cases in which complete occlusion could not be obtained, one patient was referred for further microsurgical operation and one is being followed-up with incomplete occlusion of the fistula.

In the long term, clinical improvement was observed in eight patients (50%) and stable clinical condition in a further five patients (31%). Deterioration of the patient despite treatment was observed in three cases (19%). Among the patients with clinical improvement or stable neurological condition, average improvement was 1.25 points in the Aminoff and Logue disability scale and 2.1 points in the VAS scale (Fig. 1, Tab. 2).

Discussion

Due to variable clinical presentations, spinal dural arteriovenous fistulas (sDAVF) remain a diagnostic challenge. They are usually diagnosed in middle-aged male patients in the thoracic and lumbar spine, and this was reflected in the findings of our study. Although they might present with a variety of symptoms ranging from mild to severe neurological impairment, accurate diagnosis and effective treatment is crucial as the disease is progressive and leads to disability [13]. Fortunately, recent advances in diagnostic imaging modalities and endovascular treatment techniques have greatly increased the options regarding these lesions.

Magnetic resonance imaging is currently the best tool in the diagnosis of sDAVF. The diagnosis is confirmed by a triad of findings on routine MRI that is present in > 95% of cases: 1) spinal cord oedema (hyperintense signal on T2-weighted images); 2) enlarged veins around the spinal cord (flow voids on T2-weighted images); and 3) disruption of the blood-brain

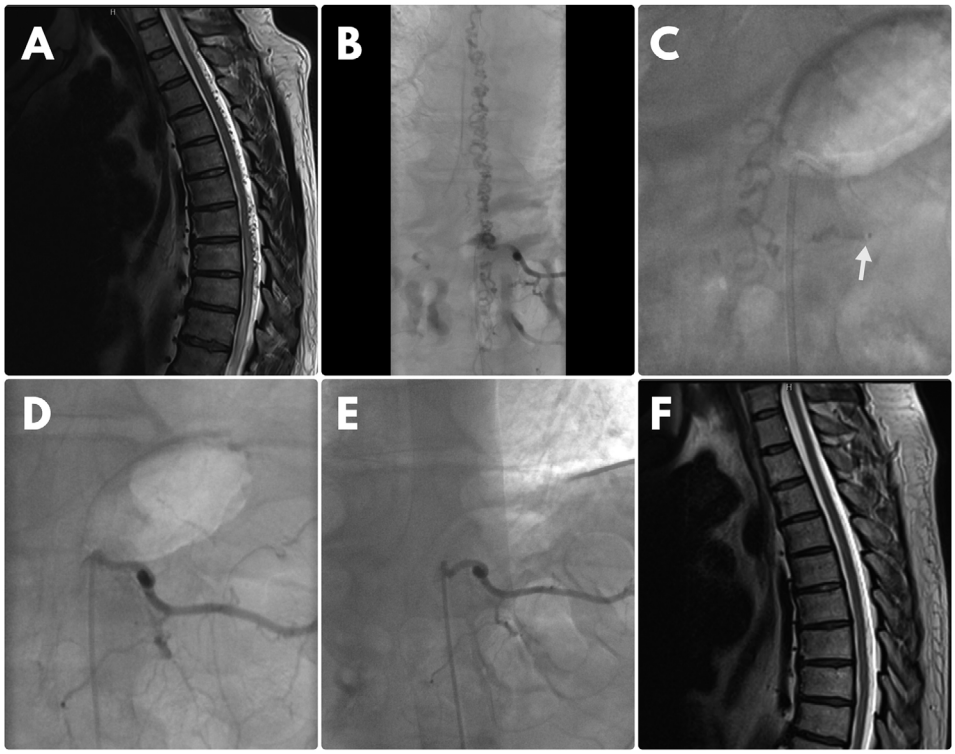

Figure 1. Successful embolisation of a thoracic sDAVF in a 53-year-old male patient who presented with increasing weakness in lower extremities. Initial diagnosis of sDAVF was made after MR examination (**A**) and confirmed in DSA afterwards (**B**). Super-selective catheterisation of feeding artery was performed (**C**), tip of a micro-catheter is pointed with white arrow. Embolisation of fistula was performed with N-butyl cyanoacrylate. Control angiography confirmed successful occlusion (**D**). Control imaging examinations performed six months after procedure (DSA — **E**) and two years after procedure (MR — **F**) showed no signs of recurrence

Table 2. Clinical data of patients and procedural outcomes

Clinical data	
Patients (n)	16
Male/female (n, %)	12 (75%), 4 (25%)
Mean age (years, range)	62.4 (28–70)
Mean time from onset to procedure (months, range)	13.3 (1–39)
Localisation of sDAVF (n, %)	
Thoracic	13 (81%)
Lumbar	3 (19%)
Procedural details	
Occlusion of fistula (n, %)	
Complete	10 (63%)
Incomplete	6 (37%)
Secondary treatment (n, %)	
Endovascular	4 (25%)
Microsurgical	1 (6%)
Complications (n, %)	
Transient paralysis	1 (6%)
Long-term outcome	
Duration of follow-up (months, range)	28 (7–58)
Occlusion of sDAVF (n, %)	
Complete	14 (88%)
Incomplete	2 (12%)
Clinical outcome (n, %)	
Improvement	8 (50%)
Stable condition	5 (31%)
Deterioration	3 (19%)

barrier (cord enhancement on T1-weighted images after contrast injection) [14]. The location of the sDAVF is determined by magnetic resonance angiography (MRA) and/or digital subtraction angiography [5].

The traditional treatment of sDAVF consisted of microsurgical occlusion of the fistulous connection. This involves a hemilaminectomy, opening the dura and coagulation or clipping of the vein [15]. A recent review of the literature published by Maimon et al. [14] described a reported success rate of this surgery ranging from 85–100%. However, being an open surgery, it is associated with several procedural complications including epidural haematoma, CSF leak, wound infection etc. [6, 16].

For this reason, some centres are implementing novel technologies (e.g. microscope-assisted endoscopic techniques in order to reduce the risk of complications) [17].

Minimally invasive endovascular embolisation is an alternative therapeutic approach for patients presenting with sDAVF. During this procedure, liquid embolic material is injected from a microcatheter placed in the proximity of the shunt aiming to permanently occlude the venous side of the fistula [14].

Although the initial experience was less satisfactory compared to the surgery, today with the introduction of modern microcatheters and liquid embolic materials, especially Onyx, the role of minimally invasive endovascular embolisation in

Table 3. Overview of literature on treatment and outcomes of patients with sDAVF

Study	Patients (n)	Treatment	Initial technical success (%)	Complications rate (%)	Overall clinical success (%)
Qi et al. [8]	52	Surgery — 40 Embolisation —12	Surgery — 100% Embolisation — 60%	4%	Improvement to some degree — 100%
Bretonnier et al. [9]	63	Surgery — 23 Embolisation — 40	Surgery — 91% Embolisation — 70%	Surgery — 9% Embolisation — 2%	N/i
Park et al. [10]	18	Embolisation	82%	6%	83%
Saladino et al. [16]	154	Surgery	95%	9%	Improvement — 82% Stability — 14%
Kirsch et al. [18]	78	Surgery — 17 Embolisation — 61	Surgery — 100% Embolisation — 77%	Surgery — 0% Embolisation — 5%	> 75%
Gemmete et al. [19]	33	Surgery — 4 Embolisation — 29	Surgery — 100% Embolisation — 76%	Surgery — 0% Embolisation — 3%	Improvement — 45% Stability — 55%
Sasamori et al. [20]	50	Surgery — 19 Embolisation — 31	Surgery — 95% Embolisation — 71%	Surgery — 11% Embolisation — 13%	Improvement — 66%
Present study	16	Embolisation	63%	6%	Improvement — 50% Stability — 31%

the management of spinal vascular malformations has expanded [8, 10, 14, 19]. In our series, compete occlusion of the fistula in long-term follow-up was achieved in 88% of patients, comparable to the outcomes reported by other authors [19, 20]. Similarly, the rate of complications remains within the range of reported procedural complications described in the above-mentioned articles.

As far as the clinical outcome is concerned, the observed clinical improvement rate was 50%. Similar results have been described in surgical studies [16, 21]. In addition to this, clinical stable condition was achieved in further 31% of monitored patients.

An overview of presented literature on treatment and outcomes of patients with sDAVF is set out in Table 3.

Considering the minimally-invasive nature of endovascular embolisation, and its high success rate combined with low rate of procedural complications, is seems reasonable to refer all sDAVF patients for multidisciplinary board evaluation consisting of a neurosurgeon and a neuroradiologist and to consider embolisation in all amenable patients.

We are aware that our study has some limitations. First and foremost, our small sample size of unrandomised patients limits the validity of the data. Secondly, the absence of a control group treated with surgical methods might be perceived as a potential drawback. Perhaps patients more suitable for microsurgical treatment were referred to other neurosurgical centres, and this preselection bias may have impacted upon the final outcome of our study. Finally, the heterogeneity of the location and morphology of the fistulas might have affected the results. Nonetheless, these drawbacks might be attributed to the rarity of the entity.

In conclusion, the results of this preliminary study suggest that long-term clinical outcomes in patients with sDAVF treated with endovascular embolisation are comparable to those reported in surgical studies, even if the initial success rate is significantly lower compared to microsurgical intervention. Similarly, the rate of procedural complication is comparable with microsurgery.

Clinical implications

Even if microsurgery remains the primary treatment modality for patients with spinal dural arteriovenous fistulas, modern endovascular methods offer a safe and reliable alternative, and should therefore be considered during multidisciplinary evaluation of these patients.

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