Endovascular approach to a type IV-C spinal pial fistula associating coils and glue

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Abstract
Spinal pial arteriovenous fistulas (SPAVFs) are rare superficial intradural vascular malformations consisting of a direct shunt between spinal cord arteries and veins. More recently, the treatment choices for this pathology have gained alternative tools, and endovascular treatment in particular have consistently gained traction as one of the most efficient choices of treatment. Surgery has proven challenging, as the anterior localisation of the shunt predisposes spinal cord manipulation and, therefore, inherent oedema and in some cases ischaemic post-operative changes in the spinal cord tissue. Surgery has increasingly been reserved for cases in which the first option of endovascular treatment is not feasible. We report a case of a type IVc SPAVF that was treated with the association of coils and glue.

Keywords: embolisation, endovascular treatment, coils, spinal pial fistula, glue

Abbreviations
ASA anterior spinal artery
AVF arteriovenous fistula
MRI magnetic resonance imaging
PSA posterior spinal artery
SPAVS spinal pial arteriovenous fistulas

Introduction
Spinal vascular malformations are rare diseases with a wide variety of neurologic presentations. Multiple different classification schemes have been proposed, which causes a considerable amount of confusion as they are based on etiology, location and kind of vascular shunting [1].

Spinal pial arteriovenous fistulas (SPAVFs) are rare superficial intradural vascular malformations consisting of a direct shunt between spinal cord arteries and veins [2]. They account for approximately 10% of all spinal arteriovenous malformations. These fistulas are commonly found anterior to the spine and are
supplied by one or multiple spinal cord arteries (most often predominating the anterior spinal artery) converging into a superficial draining vein via a single fistulous hole [3].

More recently, the treatment choices for this pathology have gained additional tools and endovascular treatment has consistently gained attention as one of the most efficient treatment choices. Surgery has been proven challenging as the anterior localisation of the shunt predisposes spinal cord manipulation and, therefore, inherent oedema in some cases ischaemic post-operative changes in the spinal cord tissue. As a result, surgery has increasingly been reserved for cases in which the first option of endovascular treatment is not feasible. We report a case of a type IVc, SPAVF which was treated with the association of coils and glue, and we perform a systematic review of the literature on this topic.

**Technique**

We report a case of a 15-year-old male patient presenting with progressive weakness in right leg and foot. Investigation with magnetic resonance imaging (MRI) showed dilated perimedullary veins at the thoracolumbar segment of the spinal cord, with radiologic evidence of 2 ecstasies, suggesting arteriovenous spinal malformation. Angiography was performed for further evaluation, demonstrating an arteriovenous pial fistula. Nutrition was provided mainly by the anterior spinal artery (ASA) arising at the left T10 segmental artery and by the right posterior spinal artery (PSA) at the level of the right T11 segment. Venous drainage was observed by one dilated perimedullary vein, as well as 2 venous pouches, one just on the initial portion of the vein and another, two vertebral levels above (Figure 1).

Endovascular embolisation was performed under general anaesthesia. An Excelsior SL10 microcatheter was navigated until the point of the fistula, guided by a Transend 0.014 microguidewire. Because presence of high blood flow at the site of the arteriovenous connection was noted, some coils were introduced inside the venous ectasia to reduce blood flow and prevent migration of the glue distal to the fistulous point. After the embolisation with 10 detachable coils (EV3), a solution of 50% of Glubran and Lipiodol was successfully injected to occlude the fistula (Figure 1). Final angiogram showed complete exclusion of the fistula point filled by the ASA and a discrete residual flow into the venous pouch provided by the right PSA.

On immediate post-operative assessment the patient presented with grade 2 paraparesis, urinary and fecal retention. An MRI was done on the first post-operative day, demonstrating thrombosis of the proximal and distal venous pouches. Corticosteroid treatment was initiated before embolisation, as a means to reduce inflammatory response to the thrombosis of the vein, leading to further inflammation, mass effect and worsening of the initial deficits. On post-operative day 7, the patient reestablished normal control of fecal and urinary sphincter and had progressive improvement of the lower limb deficits. Over 3 months of follow-up, the patient regained his ambulatory capacity, with normal urinary and fecal sphincter control.

**Discussion**

SPAVS are more commonly found at the thoracolumbar region, anteriorly located and fed by the anterior spinal artery, but they can be present at any level of the spinal cord. Venous drainage includes
perimedullary veins over the anterior and posterior surface of the spinal cord, often extending over multiple levels [8].

Spinal shunts are often classified as 1 of 4 types: dural arteriovenous fistula (AVF) (type I), glomus arteriovenous malformations (type II), juvenile metameric arteriovenous malformations (type III) and spinal pial AVF (type IV). Gueguenet et al. [5] developed the first subclassification scheme for type IV lesions. This was later modified and simplified by Anson and Spetzler [6]. The lesions were divided according to number of feeders and size, in small slow flow fistulae (IVa), intermediate fistulae (IVb) and large, high flow fistulae type (IVc). This subclassification has significant clinical and therapeutic implications. Type IVa lesions are more common in adults and symptoms are due to venous congestion. These are generally poor endovascular candidates due to the small caliber of the supplying pial artery. On the other hand, Type IVc AVFs are more frequent in children and presentation can be acute due to haemorrhage. As a result of their large size and high flow and their ventral position related to the spine, these are very challenging from the surgical point of view. Endovascular approach to these lesions is often easy, safe and fast because of the dilated arterial feeders commonly found in these lesions.

The natural history of SPAVs (type IV) is still unknown. Spinal fistulae are presumed to have a relatively malignant natural history with progressive deterioration due to venous hypertension or direct mass effect and acute neurologic events occurring as a result of flow changes, thrombosis or haemorrhage [7]. Gross et al. [9] reported that 93 % of patients presented with a neurological deficit and estimated a 2.5 % annual haemorrhage rate with a higher rate if the initial presentation was due to haemorrhage. As the majority of cases that are diagnosed are already symptomatic, observational treatment is rare.

The diagnosis of the spinal shunt usually requires radiologic investigation, with MRI usually being the initial modality of image assessment, which may show the dilated perimedullary veins and its direct relation with the signal core tissue. Digital angiography is the gold standard for clear anatomical depiction of the lesion and treatment planning. As for any arteriovenous shunt, definitive treatment can be obtained by occlusion of the venous outlet, the shunting zone and distal arterial feeder. Endovascular treatment is becoming the first line of treatment for these lesions, as advances in the field have made it simple to occlude even larger malformations. The embolisation is done mainly with liquid embolic agents; the one most commonly used by previous authors is glue [8, 9] that can be accompanied by coils in some cases. Batista et al. recently reported 2 cases that were satisfactorily treated only with coils. Microparticles appear to have no role in these types of vascular malformations at this point.

The treatment choice in the case presented was the combination of coils and glue because the injection of the liquid embolic agent is difficult to control and eventually glue can migrate distally, inducing the inherent complications of distal vessel obstruction. We decided to fill the venous ectasia with coils, and once the flow at the fistulous point was decreased, glue was injected from the distal arterial feeder. The final result was near total occlusion of the shunt, which maintained blood flow by the posterior right spinal artery, associated with a rather decreased flow.

In selected cases of type IVc SPAVs, the use of coils, with the intention of slowing the high flow at the fistula, can be used previously to injection of liquid embolic agents to achieve a better control of the injection and prevent distal glue migration into the spinal veins.
Conflict of interest
We declare that we have no conflict of interest.

References
Figures

Figure 1 - MRI shows dilated perimedullary veins (a). Angiogram confirms spinal pial fistula (b, c, d). Embolization (e, f).