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Case report

Glue embolization of a pial arteriovenous fistula of the spinal artery

Fabio Martino Doniselli[1], Aldo Paolucci[2], Giorgio Conte[2], Paolo Rampini[3], Antonio Arrichiello[1], Fabio Maria Triulzi[2]

[1] Postgraduation School in Radiodiagnostics, Università degli Studi di Milano, Milan, Italy
[2] Operative Unit of Neuoradiology, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico di Milano, Milan, Italy
[3] Operative Unit of Neurosurgery, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, Milano, Milano, Italy

Summary. There are no clear guidelines about the treatment Pial Arteriovenous Fistulae (PAVF). For high-risk and severally symptomatic fistulae surgery is the first choice of treatment, including feeding artery ligation, surgical resection, radiosurgery and endovascular embolization techniques. We described a case of a patient with a symptomatic PAVF at the craniocervical junction fed by the anterior spinal artery, successfully treated with endovascular approach consisting of glue embolization of the feeding vessel. (www.actabiomedica.it).

Keywords: pial arteriovenous fistula, spinal cord, fistula, embolization glue

Background

Pial arteriovenous fistulae (PAVF) account for less than 2% of all arteriovenous fistulae (AVF) (1). They are fed by parenchyma vessels in contrast with dural AVF which are typical fed by dural arteries (2). The origin of PAVFs is unclear, being reported as congenital, traumatic or iatrogenic, while the location is typically supratentorial. Clinically PAVF can be silent for all life or manifest with haemorrhage, epilepsy, mass effects, cranial nerve deficits or congenital heart failure (3). If untreated, mortality is reported up to 63% (4).

There are no clear guidelines about the treatment; asymptomatic fistulae may heal spontaneously thus conservative approach is recommended. Low-risk minimally symptomatic fistulae should be cautiously monitored. High-risk and severally symptomatic fistulae are usually treated with surgery, including feeding artery ligation, surgical resection, radiosurgery and endovascular embolization techniques.

We described a case of a patient with a symptomatic PAVF at the craniocervical junction fed by the anterior spinal artery, successfully treated with endovascular approach consisting of glue embolization of the feeding vessel.

Case Presentation

A 38 years old man, with no previous history of head trauma, was admitted to our emergency department after the onset of an intense pulsing neck pain associated with nausea and vomiting. Fifteen days before the patient suffered a middle-intensity headache during his usual gym training for which he went to another hospital where clinical examination and brain computed tomography (CT) scan did not detect any abnormality.

Investigations

Upon the admission to our emergency department the patient was alert, without motor and sensory deficits.

Brain CT scan showed haemorrhage in the basal cisterns and in the fourth ventricle (Figure 1). No lesions of the brain parenchyma were found. The ventricle system had normal dimensions. The three-dimensional CT angiography did not show any clear vascular anomaly. A further contrast-enhanced Magnetic Resonance scan confirmed these findings (Figure 1).

The patient underwent angiography under general anaesthesia to further assess any vascular malformation. Through a transfemoral approach with a 6 French (F) introducer, a 6F Envoy catheter (Cordis Endovascular, Miami Lakes, Florida, USA) was guided into the right vertebral artery over 0.035 inch Terumo guidewire (Terumo, Tokyo, Japan) under road-map guidance. The angiography showed a small PAVF fed by one branch of the anterior spinal artery with the venous drainage into the perimedullary veins of the cervicomedullary junction (Figure 2).

Treatment

Considering the unfavourable angle of the right origin of the anterior spinal artery, the 6F Envoy catheter was positioned in the left vertebral artery. A 3x10 millimetres (mm) Hyperglide balloon was positioned and inflate in the basilar artery to reverse the blood flow. A 1.2F Sonic microcatheter (Balt, Montmorency, France) was guided into the right anterior spinal artery over 0.08 inch Mirage microwire (Irvine, California, USA) using the road-map guidance. To reach the feeder of the PAVF 1 mg of Nimodipine (Nimotop, Bayer Healthcare, Germany) was administered from the microcatheter into the anterior spinal artery. After the vasodilatation occurred, the microcatheter was guided into the feeder of the PAVF over the microwire and 0,2 ml of a 50% mixture of glue GLUBRAN 2 (Gem Srl, Italy) and Lipiodol was injected (NBCA-LUF)

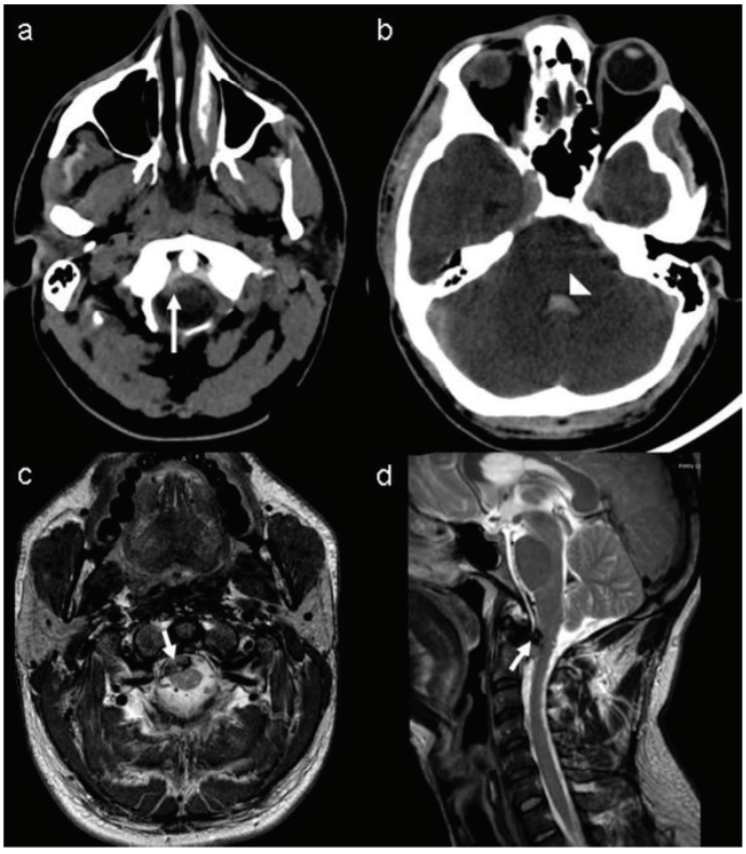

Figure 1. CT and MR scans at admission: axial CT scans (a,b), axial T2-weighted MR image (c), sagittal T2-weighted MR image(d). CT shows subarachnoid haemorrhage in the bulbo-medullary space (long arrows) and in the fourth ventricle (arrowhead). MR shows blood (short arrows) in the subarachnoid space at the front of the bulbo-medullary junction.

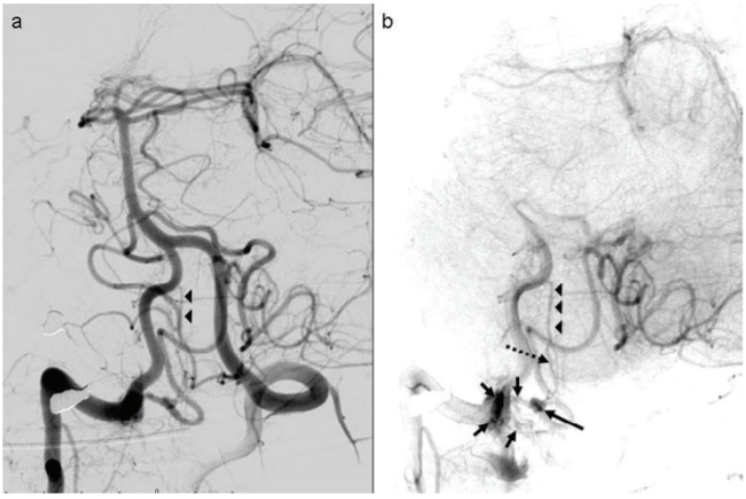

Figure 2. pre-treatment DSA: right oblique view in initial phase (a), right oblique view in late phase (b). DSA shows a branch (dotted arrow) of a prominent right anterior spinal artery (arrowheads) feeding a PAVF with pseudoaneurysmal dilatation (long arrow) in the bulbo-medullary space with drainage into the perimedullary veins (small arrows).

(Figure 3). At the end of the procedure, an angiogram through the catheter did not show any residual PAVF (Figure 4).

Outcome and Follow-up

No procedure-related complications were reported. After 10 days, the patient was discharged without any neurological signs and symptoms. At three-month and one-year follow-up the angiographies excluded any recurrence and clinical examinations confirmed the absence of any neurological deficits (Figure 5).

Discussion

We reported the case of an embolization of a PAVF fed by the anterior spinal artery, located at the level of the cranio-cervical junction and causing a subarachnoid haemorrhage.

The aim of the treatment is to occlude the fistulous connection of the PAVF in order to prevent further bleedings, preserving the blood supply of the spinal cord and avoiding ischemic myelopathy (2). For low-risk minimally symptomatic PAVFs can heal spontaneously thus a conservative approach is recommended, while high-risk or severely symptomatic PAVFs require a immediate treatment, which can be surgical, endovascular or both. The surgical approach, that sometime is guided by intraoperative fluorescence angiography, can occlude the arterial feeder of the PAVF and remove the varicose vein causing mass effect. Surgery is now limited to those cases in which the endovascular procedure is considered dangerous because the arterial feeder is a small branch of a cortical or spinal artery and cannot be occluded (3). The endovascular treatment consists in the embolization of the fistolous connection with coils or liquid embolic agents like n-butyl cyanocrylate (NBCA) or Onyx (3). The positioning of coils can be performed with a better control and is more successful when the fistoulous connection is not so big. Different authors report the use of coils to embolize intracranial PAVFs, in particular Alshekhlee et al. succeeded to embolize a PAVF of the cranio-cervical junction releasing a 1.5 mm by 2 cm coil in the left arterial spinal artery without any complications (2). The use of embolic agents (NBCA or Onyx) is more safe when the size of the feeder vessels allow the distal catheterization, with the superselective catheterization of the feeder vessels having a mortality up to 10% (5). In literature some cases of intracranial PAVFs embolized with NBCA (6,7) or Onyx (8,9) have been reported. Among these cases there is no one

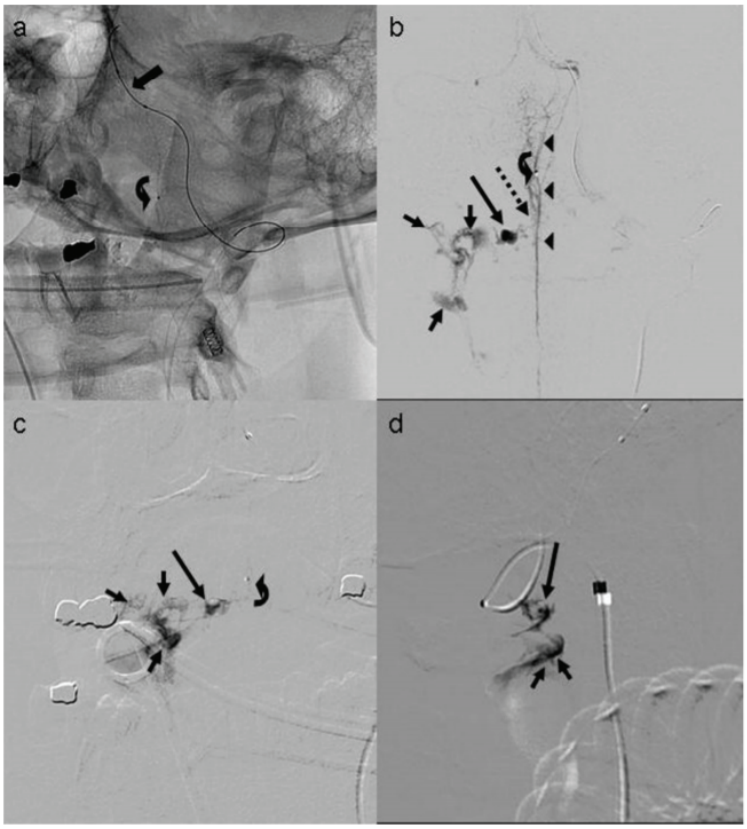

Figure 3. embolization procedure: unsubstracted frontal view (a); frontal view of PAVF after the injection from the microcatheter in the spinal artery (b); frontal view of the superselective catheterism (c) lateral view of the superselective catheterism (d). A balloon Hyperglide 3x10 (thick arrow) was located and inflated in the in the basilar artery to reverse the blood flow and the tip of the microcatheter (curved arrow) was positioned into the anterior spinal artery (arrowheads). Microcatheter injection confirms a branch (dotted arrow) of the anterior spinal artery (arrowheads) feeding a pseudoaneurysmal dilatation (long arrow) with drainage into the perimedullary veins (small arrows).

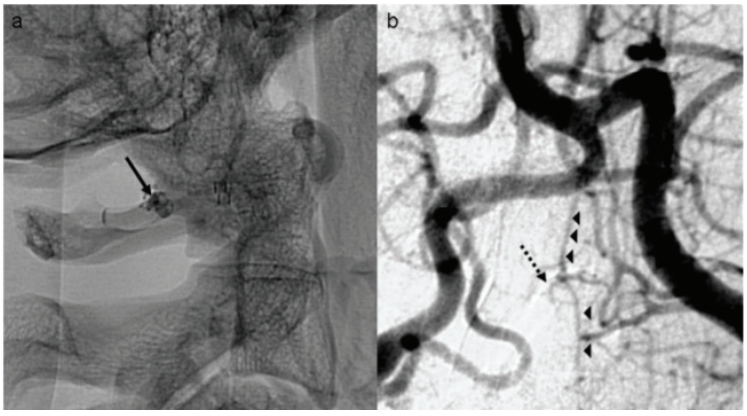

Figure 4. post-treatment DSA: unsubstracted lateral view (a), magnified frontal view (b). The unsubstracted lateral view shows the glue (arrow) within the pseudoaneurysmal dilatation. DSA shows the occlusion of the vessel (dotted arrow) feeding the PAVF and the preservation of the anterior spinal artery (arrowheads).

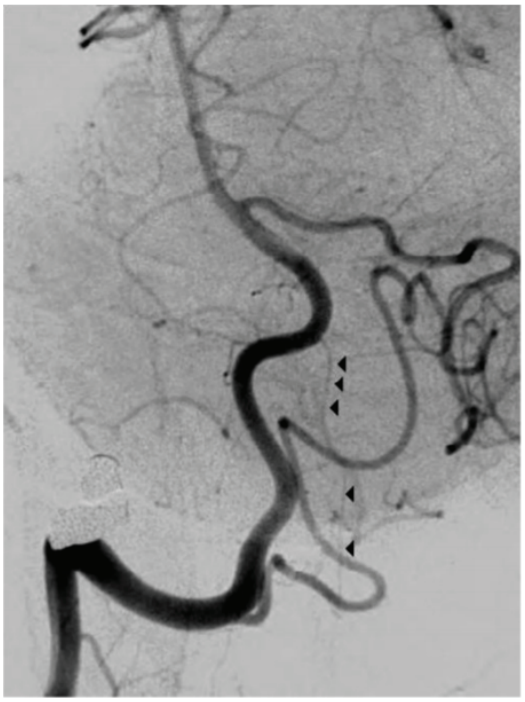

Figure 5. DSA at one year follow-up showing the preservation of the anterior spinal artery (arrowheads) without evidence of the PAVF.

reporting PAVF of the anterior spinal artery located at the cranio-cervical junction.

We decided to obtain a 50% NBCA-LUF to reach the pseudoaneurysmatic dilatation and to reduce the amount of reflux. The use of different embolization materials, as Onyx, was considered unsuitable for the risk of poor distal progression and of reflux. In relation to the small size of the feeder vessel, we avoided coils for the risk of not occlude the pseudoaneurysmatic dilatation and to induce the development of new feeder vessels. The surgical treatment was avoided because of the lack of experience of our neurosurgeons in the treatment of this pathology.

Learning Points/Take Home Messages

- PAVF of the cervicomedullary junction can cause haemorrhage in the basal cisterns and in the fourth ventricle.

- The administration of Nimodipine from the microcatheter into the anterior spinal artery can be useful to navigate the vessel.

- PAVF of the cervicomedullary junction, fed by branches of the anterior spinal artery, could be embolized with Glue.

Informed Consent: Written informed consent to the interventions, CT and the MR exams was obtained from all subjects in this study.

Ethical Approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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Correspondence:
Giorgio Conte MD,
Operative Unit of Neuroradiology, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico di Milano, Milan, Italy.
Via Francesco Sforza 35, 20122, Milano, Italy
E-mail: giorgio.conte@policlinico.mi.it