ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

Romanian
NEUROSURGERY

Vol. XXXV | No. 2 June 2021

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DOI: 10.33962/roneuro-2021-024

Romanian Neurosurgery (2021) XXXV (2): pp. 152-158
DOI: 10.33962/roneuro-2021-024
www.journals.lapub.co.uk/index.php/roneurosurgery

Endovascular treatment options for carotid-cavernous fistulae

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ABSTRACT
A carotid-cavernous fistula is a pathological shunt between the internal or external carotid arteries and the cavernous sinus (CS). The arteriovenous shunt can be direct, between the internal carotid artery and the CS, or indirect, between meningeal branches from the carotid arteries and the dural coverings of the CS. Direct fistulas occur most commonly after craniofacial trauma, while indirect shunts result from various chronic diseases. Signs and symptoms depend on the venous drainage routes. Exophthalmos, chemosis, and diplopia are caused by venous drainage through the superior and inferior ophthalmic veins. If venous egress is mainly through the inferior petrosal sinus, patients complain of pulsatile tinnitus. Cortical venous drainage is the most dangerous route because it can lead to focal neurological deficits and intracerebral haemorrhage. Treatment of carotid-cavernous fistulae can be achieved through different endovascular techniques, using detachable balloons, coils, liquid embolic agents, covered stents, and flow-diverters. This paper aims to report three cases with carotid-cavernous fistulae, which were successfully cured using different transarterial and transvenous modalities.

Keywords
carotid-cavernous fistula, endovascular treatment, transarterial, transvenous

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ISSN online 2344-4959
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First published June 2021 by London Academic Publishing www.lapub.co.uk

INTRODUCTION
Carotid cavernous fistula (CCF) refers to an aberrant arteriovenous communication between the carotid arterial system and the venous compartments of the cavernous sinus (CS). Based on the arterial feeding source, CCF's can be direct or indirect.[1] According to the Barrow classification of CCF's, a direct, high-flow connection between the internal carotid artery (ICA) and the CS is regarded as a "type A" fistula. Indirect CCFs, which typically have a low flow, are classified as type B if they are supplied only by the dural branches of the ICA, type C by dural branches of the external carotid artery (ECA), and type D by both ICA and ECA dural branches. The majority of type A CCF's are more commonly a result of blunt or penetrating trauma, accounting for 75% of all CCF cases.[1] Clinical manifestations of type A fistulas depend on the venous outflow from the CS and include ocular pain, exophthalmos, chemosis, ocular foreign body sensation, visual disturbances, and headaches for an anterior superior ophthalmic venous route. Drainage

to the posterior venous outflow channels can lead to neurologic symptoms, such as aphasia and confusion.[2] Digital subtraction angiography (DSA) is the gold standard in the diagnosis of CCF. It must be performed before any potential intervention. Endovascular treatment is now the first therapeutic option for CCFs because it is associated with high occlusion and low complication rates.[3] Depending on the angioarchitecture of the fistula, endovascular obliteration can be achieved by a combination of coils, detachable balloons, liquid embolic agents, and stents.[4] This paper aims to present a case series of three patients with CCF's occluded using different transarterial and transvenous treatment strategies.

CASE DESCRIPTIONS

Case 1

A 17-year-old female patient was admitted in July 2020 to the emergency department of an outside hospital for craniofacial trauma with multifocal, displaced mandibular fractures, active bleeding in the oral cavity, and marked facial edema after a motor vehicle accident. Urgent maxillofacial surgery was performed to stop the bleeding and treat the mandibular fractures.

After several days the patient complained of left ocular pain. Periorbital edema, chemosis, and exophthalmia developed gradually afterward. CT angiography showed an enlarged left cavernous sinus compared to the contralateral side (Fig. 1, a) and a dilated, engorged superior ophthalmic vein. (Fig. 1, b, c). Five days later, she was transferred to our hospital for further investigations.

Catheter angiography confirmed a high-flow, type A, left carotid-cavernous fistula with a 4.2 mm direct "communication" between the ICA and the CS, located on the horizontal part of the cavernous segment (Fig. 1, d, e, f). Venous drainage was mainly through the superior ophthalmic vein, markedly enlarged, explaining her main signs and symptoms (Fig. 1, e). Additionally, drainage occurred in the pterygoid venous plexus, inferior petrosal sinus, and jugular vein, and through the coronary sinus in the contralateral cavernous sinus (Fig. 1, d). Based on the angioarchitecture of the fistula, a decision was made to occlude the fistula with coils through a transarterial approach using the balloon-remodeling technique.

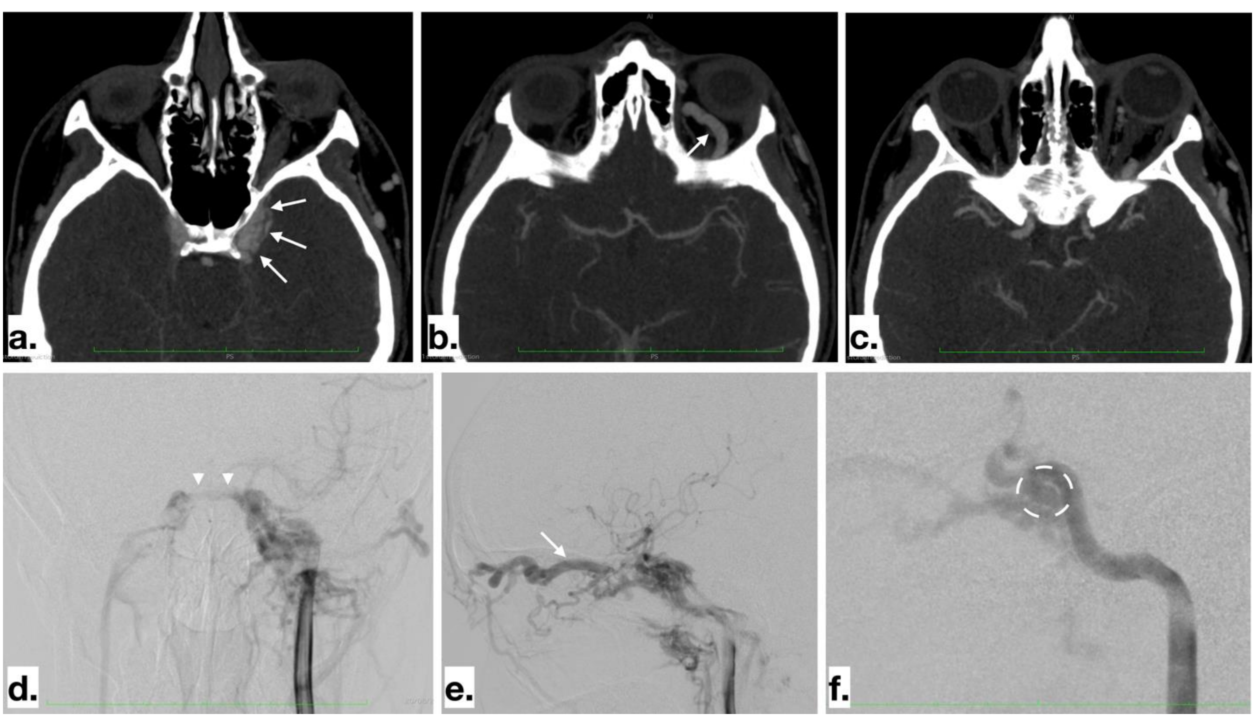

Figure 1. Axial CT angiogram images in **a.** enlarged left CS (white arrows), **b.** engorged left SOV (white arrow), **c.,** and left exophthalmia. DSA images in **d.** frontal projection, the drainage through the coronary sinuses (arrowheads) in the contralateral CS, IPS, and internal jugular vein, **e.** lateral projection, engorged SOV (white arrow), **f.** lateral projection, the "venous pouch" connected to the ICA (white circle). CS=cavernous sinus; SOV=superior ophthalmic vein; DSA=digital subtraction angiogram; IPS=inferior petrosal sinus; ICA=internal carotid artery.

On the following day, under general anesthesia, through a right femoral approach, a 6F guiding catheter was placed in the distal cervical ICA, followed by navigation of a microcatheter in the CS and a hypercompliant balloon in the carotid siphon (Fig. 2, a). While the balloon was inflated over the arterial "tear," six bare platinum, detachable coils were implanted through the microcatheter into the left cavernous sinus (Fig. 2, b). No antiplatelet therapy was used during the procedure. After the first coil was deployed, a bolus of 5000 IU of unfractionated heparin was administered intravenously. After the 6th coil was introduced, complete obliteration of the fistula was obtained (Fig. 2, c, d). There were no periprocedural complications. Immediately after the procedure, the patient was extubated. Her symptoms regressed completely over the following days, and she was discharged four days later. A control angiogram was performed three months later, confirming the total exclusion of the fistula without additional sequelae (Fig. 2, e, f).

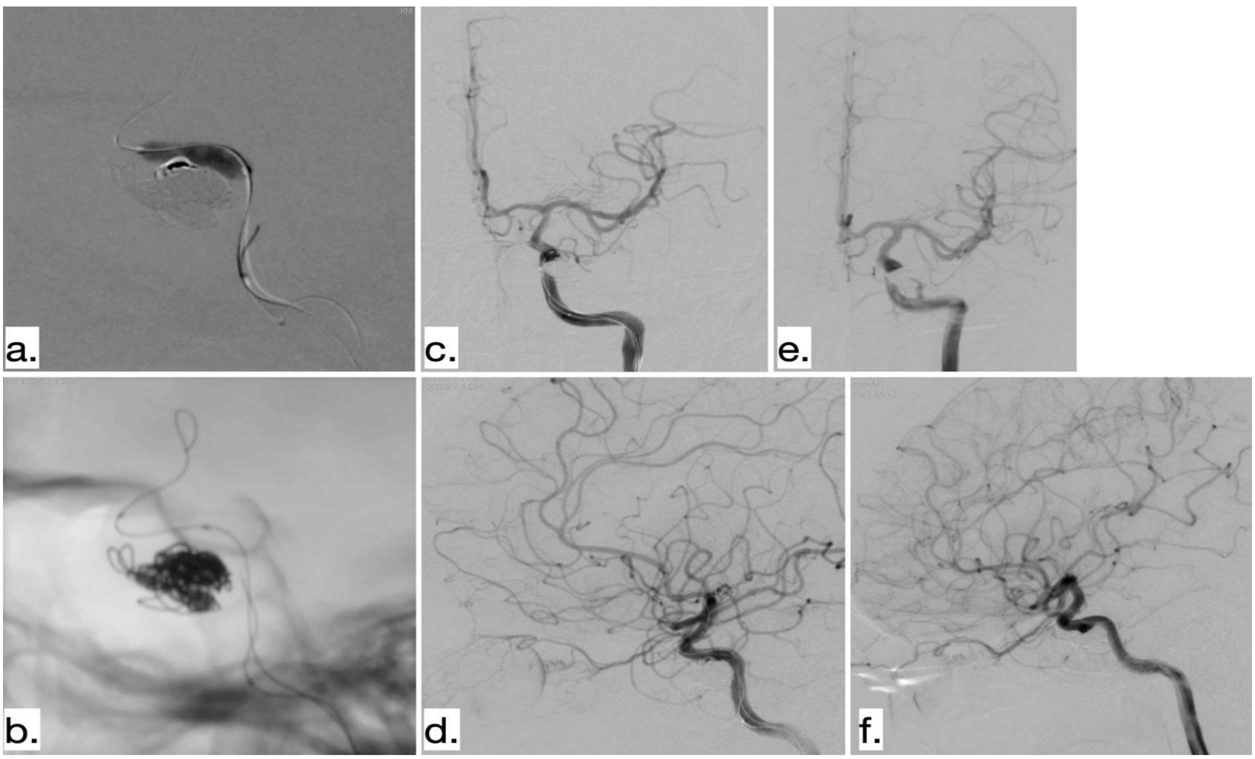

Figure 2. a. blank roadmap image shows the inflated balloon in the ICA, overlying the coil mass; **b.** lateral projection fluoroscopy image: the coil mass is better seen; **c, d.** frontal and lateral projection DSA images immediately after the procedure showing complete obliteration of the fistula; **e, f.** control angiogram after three months demonstrating cure of the fistula. ICA=internal carotid artery, DSA=digital subtraction angiogram.

Case 2

An 8-year-old boy was admitted to our hospital with exophthalmos and chemosis of the left eye, which developed gradually for the last week before admission. Upon neurological examination, the patient was conscious, cooperative, with motor aphasia and right hemiparesis. His neurological deficits resulted from a left hemispherectomy, which he received after significant head trauma during a motor vehicle accident seven months prior. The following day, under general anesthesia, a catheter angiography revealed a left high-flow Barrow type A carotid-cavernous fistula (Fig. 3 a, b). The fistula was also opacified retrogradely from the vertebral artery (Fig. 3 c, d). In addition, due to the high flow, aspiration from the external carotid artery via artery of foramen rotundum was evident (Fig. 3, g).

A decision was made to cure the fistula by sacrificing the left ICA with coils. A 6F guiding catheter was placed in the left ICA, followed by navigation of a microcatheter in the supraclinoid segment. Two Penumbra Occlusion Devices were deployed distal and proximal to the fistula (Fig. 3, e), achieving complete obliteration of the ICA and fistula. The enlarged artery of the foramen rotundum was occluded with a 1mm Target Nano coil (Fig 3. g, h). A cure of the fistula was immediately seen upon contrast injection in the vertebral artery (Fig. 3, i). Chemosis and exophthalmia gradually improved following the procedure, and the patient was discharged three days later.

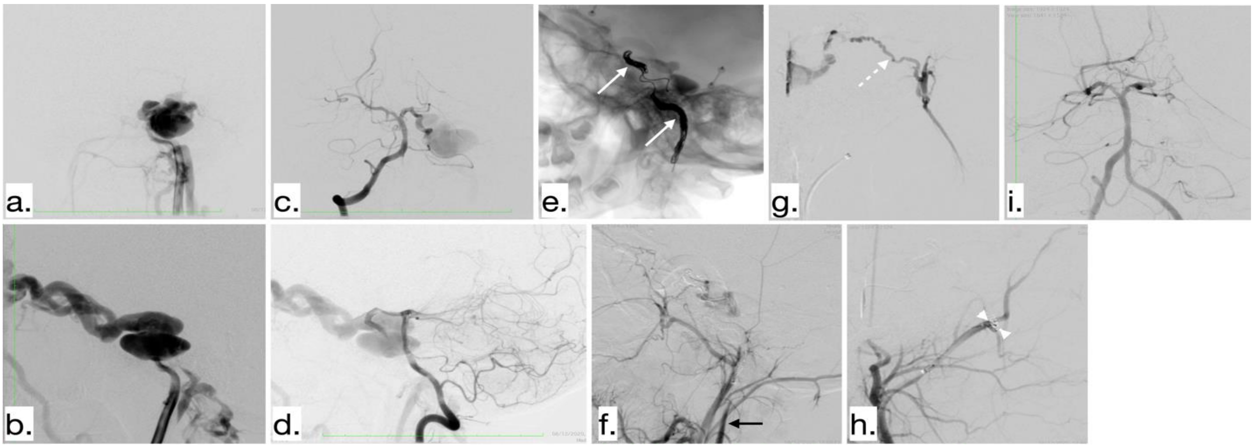

Figure 3. DSA images showing **a, b.** frontal and lateral projections, high-flow fistula with a large venous pouch, draining in the SOV; **c, d.** the fistula is opacified retrogradely through the PCom, during contrast injection in the right VA; **e, f.** Two Penumbra Occlusion Devices (white arrows) were deployed in the ICA distal and proximal to the fistula, with immediate occlusion of the ICA; **g, h.** aspiration through an enlarged artery of the foramen rotundum (dashed arrow) was interrupted by occluding the artery with a 1mm Target Nano coil; **i.** final contrast injection in the VA shows no residual filling of the fistula.
DSA=digital subtraction angiogram; SOV=superior ophthalmic vein; PCom=posterior communicating artery; VA=vertebral artery; ICA=internal carotid artery.

Case 3

A 33 years-old male patient was admitted to our hospital with diplopia, ptosis of the left eyelid, exophthalmia, and chemosis of the left eye. The symptoms had a gradual onset in the last two weeks before admission. MRI performed at another institution revealed enlarged left CS and SOV (not shown). Upon admission, his blood pressure and other laboratory findings were within normal limits. Neurological examination was also normal. The only significant information that he recalls was an abrupt deceleration from 100km/h, without head trauma, while driving a truck, two weeks before symptom onset. Catheter angiogram performed the following day revealed a left, indirect, Barrow type D dural CCF, opacified through meningeal branches from the ICA and ECA (Fig. 4 a, b, c, d). Venous drainage occurred through the SOV and IOV.

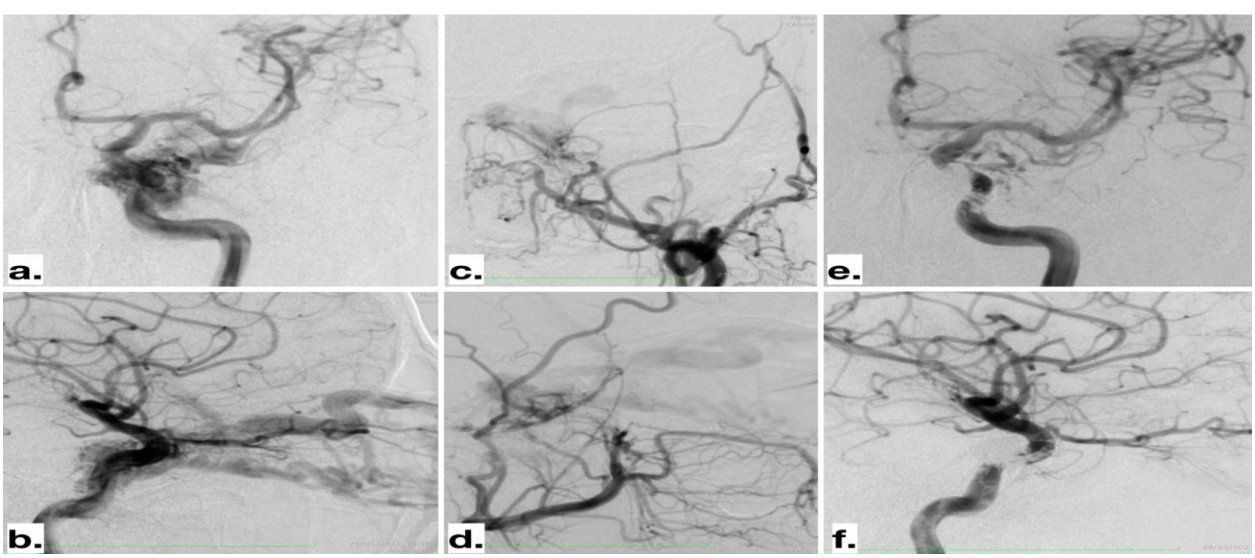

Figure 4. a, b. frontal and lateral projection DSA images showing a left dural, indirect CCF, and venous drainage through the SOV and IOV; **c, d.** the fistula is opacified through meningeal branches from the ECA; **e, f.** complete cure of the fistula obtained with transvenous Onyx injection.
DSA=digital subtraction angiogram; CCF=carotid-cavernous fistula; SOV=superior ophthalmic vein; IOV=inferior ophthalmic vein; ECA=external carotid artery

Under general anesthesia, super-selective catheterization of the ECA and its meningeal branches was performed with a 1.3F Headway Duo microcatheter (Microvention, California, USA) with the goal of occluding the fistula trans arterially (not shown). However, a sufficiently distal position could not be obtained, raising the concern of an incomplete obliteration. Therefore, a combined transarterial and transvenous route was chosen to occlude the fistula by injecting a liquid embolic in the CS while preventing its reflux with a compliant balloon inflated in the ICA.

The left internal jugular vein was punctured under roadmap guidance, followed by the placement of a 6F sheath (Fig. 5, a). A Scepter C compliant balloon (Microvention, California, USA) was navigated in the ICA (Fig. 5, b) and inflated to prevent reflux of the liquid embolic. A 4F diagnostic catheter was placed at the level of the left IPS, which was not visible. After multiple failed attempts to recanalize the IPS with a Terumo 35 guidewire, we managed to cross the IPS and reach the CS with a coronary angioplasty wire, Pilot 14 (Abbott, Chicago, USA) (Fig. 5, c, d) and a Headway Duo microcatheter. Superselective contrast injection confirmed the position inside the CS and the enlarged SOV (Fig. 5, e, f). With the balloon inflated in the cavernous ICA, two vials of Onyx (Medtronic, Dublin, Ireland) were slowly injected through the Headway Duo microcatheter until the fistula was no longer visible (Fig. 4, e, f). No complications occurred during or after the procedure. Symptoms improved almost immediately on the angiographic table. The patient was discharged five days later with residual diplopia.

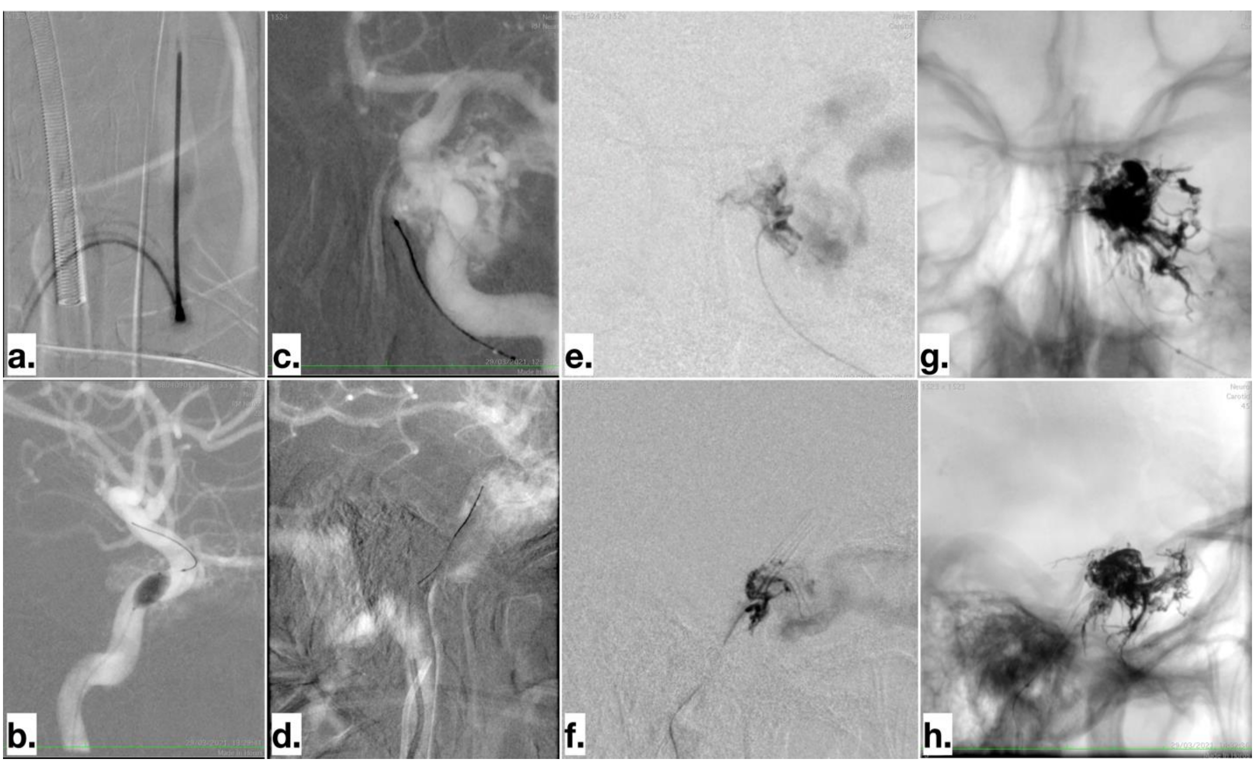

Figure 5. a. A 6F sheath was placed in the IJV under roadmap guidance; **b.** the compliant balloon inflated had the role in preventing reflux of the liquid embolic in the cavernous ICA; **c, d.** frontal and lateral projection roadmap images: a coronary wire was successfully navigated from the IJV through the IPS in the CS; **e, f.** frontal and lateral projection DSA images confirming the microcatheter position in the CS; **g, h.** frontal and lateral fluoroscopy images showing the final Onyx cast.
IJV=internal jugular vein; ICA=internal carotid artery; IPS=inferior petrosal sinus; CS=cavernous sinus; DSA=digital subtraction angiogram.

Discussion

Endovascular treatment of carotid-cavernous fistulae can be achieved by transarterial or transvenous approaches. Furthermore, multiple devices and occlusion strategies are available. The selection of a treatment strategy depends mainly on the angioarchitecture of the fistula. In the first case presented, transarterial occlusion with coils was, in our opinion, the safest plan due to a good visualization of the shunt and the easy access to the enlarged venous pouch. The treatment strategy in the second case was dictated by the absence of the left hemisphere, such that sacrifice of the ICA was deemed the safest and fastest cure possible.

Transarterial treatment of direct CCF's using detachable coils was first reported by Halbach et al. in 1991.[5] Since then, this technique has been assessed in multiple retrospective cohort studies. The reported immediate complete obliteration rate was around 70%, and up to 90% after later reinterventions, with no periprocedural morbidity or mortality.[5–7] The main reasons behind incomplete obliteration were a large arterial defect or a large aneurysmal dilation of the cavernous sinus.[6] To preserve the parent artery and prevent coil herniation in the ICA, non-detachable balloons or intracranial stents can be used as reconstructive treatment options.[6,8] In addition, incomplete closure of the fistula can lead to a change in hemodynamics, leading to a redirection of blood drainage, e.g., from the SOV to cortical cerebral veins, which can rupture and cause an intracerebral hemorrhage.

Detachable coils have potential disadvantages in the treatment of CCF's. Due to the dense packing needed to occlude the fistula, the coil mass can cause mechanical compression on the nerves that travel in the CS, with subsequent cranial nerve palsy (CNP). Worsening or developing new CNP after fistula closure seems to be associated with the total volume of coils used. Almost 80% of patients who needed a coil volume larger than 0.2 cm^3 developed new or worsening previous CNP. Only 70% of these patients recovered completely, requiring a more extended period than patients with lower coil volumes.[9] To achieve such a dense packing necessary to cure the fistula, a mean number of 14-22 coils are needed per patient.[6,7] This large number of coils increases the cost of the procedure and the irradiation to the patient.

If obliteration of the fistula with coils cannot be obtained transarterial, transvenous access is a viable alternative. It can be achieved by a posterior route to the CS through the internal jugular vein and inferior petrosal sinus or an anterior approach through the facial or superior ophthalmic vein.[4,9] Considering the internal trabecular structure of the CS, using only detachable coils might be partially responsible for the relatively low immediate occlusion rate. Consequently, coils can be combined with liquid embolic agents, either as a first-line approach or as a bail-out strategy to achieve complete obliteration.[4] This technique requires a combined transarterial and transvenous access. Usually, embolic agents are injected into the coil mass. At the same time, a non-compliant balloon is inflated in the ICA, covering the laceration to prevent emboli from occluding ICA branches. Immediate obliteration rates reported for this technique are higher than those for coiling alone, reaching 90%.[10] However, care should be taken when injecting liquid embolic in the CS, not to occlude normal draining veins and to cause a hemorrhagic infarct. In the third case reported, the structure of the fistula was so diffuse that we considered coils impossible to position properly. Therefore, only liquid embolic was slowly injected into the CS. Concomitantly, a compliant balloon was inflated in the cavernous segment of the ICA to prevent liquid from refluxing into the artery, causing an ischemic stroke.

Covered and flow-diverting stents can also occlude the tear in the cavernous segment of the ICA while preserving its patency.[4] However, covered stents are rigid, not designed for intracranial arteries, making them difficult to navigate and deploy. The first report of a direct CCF, caused by a ruptured aneurysm in a 72 years-old female patient, successfully obliterated with a Flow-Redirection Endoluminal Device (FRED) flow-diverter, was presented in 2012 at the ABC-WIN, Val d'Isere meeting by Stefanita Dima and Lucian Marginean. Since then, other reports have shown that transvenous or transarterial coiling of the cavernous sinus, with simultaneous flow-diverter implantation in the ICA, achieves a 100% obliteration rate.[11,12]

Conclusions

Carotid-cavernous fistulae represent a heterogeneous disease. This heterogeneity is caused by the different angioarchitecture of each fistula. These architectural particularities dictate the best endovascular treatment modalities. Transarterial balloon-assisted coiling is associated with relatively high immediate obliteration rates and a low number of complications. However, due to the large volumes of coils needed to occlude the fistula and their subsequent mass effect, cranial nerve palsies can persist for more extended periods. Transvenous occlusion with coils and liquid embolic has a higher cure rate than coiling alone; however, inadvertent reflux of liquid embolic into normal arteries can lead to devastating ischemic complications.

As technology evolves, endovascular choices will continue to expand, allowing higher immediate cure rates and shorter recovery intervals. Flow-diversion devices are promising adjuncts to the arsenal of devices aimed at carotid-cavernous fistulas. They can promote a faster thrombosis of the fistula, and more importantly, offer a scaffolding surface over the arterial laceration that can serve as a platform for endothelial growth and complete reconstruction of the internal carotid artery.

Ethical requirements

No personal data of the patient is available for identification.

Conflicts of Interest

The authors declare that there is no conflict of interest.

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