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Gilbert Gravino^{a,*}, Richard Pullicino^a, Mani Puthuran^a, Ynyr Edwards^a, Jawad Yousaf^{a,b}, Emmanuel Chavredakis^a, Arun Chandran^a

^a Department of Neuroradiology, The Walton Centre for Neurology and Neurosurgery, Liverpool, United Kingdom

^b Department of Neurosurgery, The Walton Centre for Neurology and Neurosurgery, Liverpool, United Kingdom

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ABSTRACT

Ethmoidal dural arteriovenous fistulas are a rare entity accounting for 10 % of all dAVFs.³⁻⁶ Haemorrhage occurs in up to 91 % of cases, which is a particularly high risk and warrants therapeutic intervention.⁸⁻⁹ Endovascular treatment for these fistulas using the conventional detachable microcatheter technique is associated with certain limitations and risks; 8.3 % rate of incomplete obliteration and an 8.3 % rate of complications. Complications include reflux of liquid embolic agent, posterior ischaemic optic neuropathy, acute visual loss, and small subdural haematoma secondary to a micro-perforation.^{8,10-12} We present our recent experience with the Scepter Mini Balloon Microcatheter for the endovascular treatment of ethmoidal dural arteriovenous fistulas in 3 patients, involving bilateral simultaneous inflation of the balloon. It demonstrates a novel application of this technology with good outcomes. It supports the use of this microcatheter in treating ethmoidal dural arteriovenous fistulas endovascularly, either as a first-line option or as an adjunct to surgery.

1. Introduction

High grade dural arteriovenous fistulas (dAVF) with cortical venous reflux (Borden types II and III) are generally associated with an annual risk of mortality in 10.4 %, haemorrhage in 8.1 %, and non-haemorrhagic neurological deficit in 6.9 %.¹ The rate of rebleed following an initial haemorrhage can be as high as 35 % in the first 2 weeks.² Ethmoidal dAVFs are a subtype which account for 10 % of all dAVFs.³⁻⁶ These may either be asymptomatic or present with varied symptoms including intracranial haemorrhage, headache, visual impairment or seizures.⁷ Haemorrhage occurs in up to 91 % of those cases with cortical venous reflux, which is a particularly high risk and warrants therapeutic intervention.^{8,9}

Embolisation of ethmoidal dAVF is particularly difficult because of the small calibre and tortuosity of the vessels involved, and the risk of non-target embolisation including the central retinal artery. While transvenous access may provide direct access to the fistulous point and allow direct occlusion, it can be difficult to navigate due to very tortuous cortical venous anatomy and thrombosed or stenotic sinuses, and poses high risk of rupture from enlarged and fragile cortical veins.^{8,10} In such cases, the transarterial approach to embolisation is a preferred option over transvenous embolisation.

In this case series we present our recent experience in treating ethmoidal dAVFs using a bilateral transarterial (ophthalmic arteries) approach with simultaneous inflation of the Scepter Mini Balloon Microcatheter (SMBM) (MicroVention). This received FDA approval on July 10, 2019. It is a dual-lumen balloon microcatheter with one lumen for balloon inflation and another lumen for injection of liquids such as embolic agents. It has a 2.2×9 mm hydrophilic-coated semi-compliant balloon and a distal outer diameter of 1.6Fr.

2. Case 1

A 52-year-old female patient experienced progressively worsening fatigue, headaches, dizziness, loss of balance and falls. She did not have any significant past medical history and was not on any regular medication. She was a non-smoker and consumed a moderate amount of alcohol.

Abbreviations: CT, Computed tomography; dAVF, Dural arteriovenous fistulas; DSA, Digital subtraction angiography; ICA, Internal carotid artery; LEA, Liquid embolic agent; MRA, Magnetic resonance angiography; SMBM, Scepter mini balloon microcatheter.

^c Corresponding author.

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E-mail address: gilbert.gravino@nhs.net (G. Gravino).

An investigation with an MRI scan of her head demonstrated multiple abnormal flow voids along the right side of the anterior falx, with no evidence of haemorrhage. A cerebral angiogram with digital subtraction angiography (DSA) confirmed a Boden Type III ethmoidal dAVF (Fig. 1A–H). This was supplied by bilateral ethmoidal arteries arising from the ophthalmic arteries and by bilateral frontal branches of the middle meningeal arteries. Its venous drainage was into a grossly dilated right frontopolar vein which drained into the superior sagittal sinus. Around 2 weeks after the angiogram the patient experienced fleeting paraesthesia in her right upper limb but was otherwise neurologically asymptomatic.

After discussing the risks and benefits of surgical and endovascular treatment, the patient elected to undergo endovascular embolisation. This treatment was carried out under general anaesthesia. Bilateral femoral arterial access was established with 6Fr sheaths and Envoy DA XB (Codman) guiding catheters were advanced into both Internal carotid arteries (ICAs). SMBMs were navigated into the ophthalmic arteries bilaterally and positioned at the origin of the arterial feeders of the



Fig. 1. A catheter angiogram with DSA in arterial phases (A–H) demonstrates arteriovenous shunting due to an ethmoidal dAVF - feeders from bilateral ethmoidal arteries and bilateral frontal branches of the middle meningeal arteries - venous drainage into a grossly dilated right frontopolar vein; (A) Right ECA injection – AP, (B) Right ECA injection – Lateral, (C) Left ECA injection – AP, (D) Left ECA injection – Lateral, (E) Right ICA injection – AP, (F) Right ICA injection – Lateral, (G) Left ICA injection – AP, (H) Left ICA injection – Lateral. Native fluoroscopic images demonstrate the position of the bilaterally inflated Scepter mini-balloons and the cast formed by the liquid embolic agent after embolisation; (I) Pre-embolisation – AP, (J) Pre-embolisation – Lateral, (K) Post-embolisation – AP, (L) Post-embolisation – Lateral. Post-embolisation AP and lateral imaging with CCA injections in arterial phase demonstrated resolution of the dAVF (M–P).

fistula (Fig. 1, I - J). With Scepter Mini Balloons inflated bilaterally, 25 % PHIL (Precipitating Hydrophobic Injectable Liquid) was slowly and sequentially injected on both sides resulting in good permeation of the dAVF and the venous tributaries (Fig. 1, K – L). There was also retrograde occlusion of a feeding middle meningeal branch. Angiographic evaluation post-injection did not show any residual filling of the ethmoidal dAVF and demonstrated preserved retinal blushes bilaterally.

The patient was well after the procedure and vision was not compromised. An MRA scan a week later showed no evidence of the fistula. Repeat imaging with a DSA 1 year after the intervention confirmed a sustained complete occlusion.

3. Case 2

A 48-year-old man presented to hospital following a tonic-clonic seizure with no post-ictal neurological deficits. He had a past medical history of depression, previous intravenous drug use, and surgical fixation of an ankle fracture. He smoked tobacco and drank approximately 10 units of alcohol every week.

A CT head showed intraparenchymal haemorrhage in the right frontal lobe (Fig. 2) with surrounding oedema and a dense parafalcine tortuous vessel (Fig. 3A). A subsequent contrast-enhanced CT head

confirmed a right frontal dAVF (Fig. 3B) and a cerebral angiogram with DSA further characterised the abnormality (Fig. 3, C–H). This demonstrated an ethmoidal dAVF supplied by bilateral ethmoidal arteries which arise from the ophthalmic arteries. Its drainage was into a dilated right orbitofrontal vein and an ectatic right superficial middle cerebral vein. It also demonstrated cortical venous reflux in keeping with a Borden type III dAVF.

After discussing the risks and benefits of surgical and endovascular treatment, the patient elected to undergo endovascular embolisation. This treatment was carried out under general anaesthesia and 5000iu of Heparin were administered intravenously at the start of the procedure. Bilateral femoral arterial access was established with 7Fr sheaths and 7Fr Asahi Fubuki (Asahi Intecc) guiding catheters which were advanced into both ICAs. SMBMs were navigated into the ophthalmic arteries bilaterally and positioned at the origin of the feeding ethmoidal arteries, distal to the origin of the central retinal artery. After bilateral inflation of the Scepter Mini Balloons, 25 % PHIL was slowly and sequentially injected on both sides resulting in good permeation of the dAVF and its venous tributaries. Angiographic evaluation post-injection did not show any residual filling of the ethmoidal dAVF and demonstrated preserved retinal blushes bilaterally.

The patient was well after the procedure and vision was not



Fig. 2. Unenhanced CT Head demonstrating hyperdense haemorrhage with surrounding vasogenic oedema in the right frontal pole.



Fig. 3. An unenhanced CT Head (A) shows oedema associated with an intraparenchymal haemorrhage in the right frontal lobe and a dense parafalcine tortuous structure that was then confirmed to be a prominent vein secondary to a dAVF on a CT angiogram (B). A catheter angiogram with DSA (C – H) demonstrates the ethmoidal dAVF - feeders from bilateral ethmoidal arteries - venous drainage into the right orbitofrontal vein and the right superficial middle cerebral vein; (C) Right ICA injection – AP (arterial phase), (D) Right ICA injection – Lateral (arterial phase), (E) Left ICA injection – AP (arterial phase), (F) Left ICA injection – Lateral (arterial phase), (G) Right ICA injection – AP (capillary phase), (H) Right ICA injection – Lateral (capillary phase). Native fluoroscopic images demonstrate the position of the bilaterally inflated Scepter mini-balloons pre-embolisation on AP (I) and Lateral (J) views, and the cast formed by the liquid embolic agent postembolisation on AP (K) and Lateral (L) views. Post-embolisation AP and lateral imaging with ICA injections in arterial phase demonstrated resolution of the dAVF (M–P).

compromised. A post operative MRA showed no evidence of the fistula and there was focal gliosis within the anterior inferior aspect of the right frontal lobe due to the previously described haemorrhagic insult. A follow-up MRA after 10 months also showed no evidence of any residual dAVF. A delayed DSA performed 25 months after endovascular embolisation confirmed a sustained complete obliteration of the dAVF.

4. Case 3

A 52-year-old man presented with a pulsatile swelling in the occiput which was associated with worsening headaches and a disturbed sleeping pattern over a two-year period. He also experienced pulsatile tinnitus and deteriorating vision in his left eye. His past medical history included well controlled asthma, previous deep vein thrombosis, appendicectomy and nasal sinus polyps. He smoked 5 cigarettes a day and did not use illicit drugs or consume alcohol.

The patient was initially investigated with a Gadolinium-enhanced MRI scan which showed two separate dAVFs, but no evidence of haemorrhage. A right occipital dAVF with a large extracranial component was tackled first, successfully treated without complications using coils, glue and liquid agent. A year later he was treated for an incidental ethmoidal dAVF. A cerebral angiogram with DSA characterised the latter to be a Boden Type III dAVF (Fig. 4A–D). It was supplied by bilateral ethmoidal arteries arising from dilated ophthalmic arteries. Its venous drainage was predominantly into bilateral dilated orbitofrontal veins, and a smaller component draining via an ectatic right frontopolar vein into the anterior aspect of the superior sagittal sinus.

After discussing the risks and benefits of surgical and endovascular treatment, the patient elected to undergo endovascular embolisation. This treatment was performed under general anaesthesia. Vascular access was achieved through the right radial artery using a 6Fr sheath and through the right common femoral artery using a 7Fr sheath. A 6Fr Benchmark (Penumbra) guiding catheter was advanced to the right ICA, and a 7Fr Asahi Fubuki (Asahi Intecc) guiding catheter was advanced to the left ICA. The SMBMs were navigated into the ophthalmic arteries

bilaterally and positioned at the origin of the arterial feeders of the fistula, distal to the origin of the central retinal artery (Fig. 4EandF). With Scepter Mini Balloons inflated bilaterally, 25 % PHIL was slowly and sequentially injected on both sides resulting in good permeation of the dAVF and its venous tributaries (Fig. 4GandH). Angiographic evaluation post-injection did not show any residual filling of the ethmoidal dAVF and demonstrated preserved retinal blushes bilaterally.

The patient was well after the procedure and vision was not compromised. A follow-up MRA after 4 months did not demonstrate residual filling of the ethmoidal dAVF. However, a cerebral DSA performed after 10 months identified a residual dAVF supplied by branches of the left ophthalmic artery. This drained into the anterior cerebral vein, through a cortical vein and into the superior sagittal sinus. The right occipital dAVF which was treated separately remained completely occluded. A superselective angiogram performed 17 months after initial embolisation demonstrated residual filling of the ethmoidal dAVF again. The patient did not experience any new onset symptoms in the interim. Treatment with a venous approach was considered but deemed too high risk. The case was discussed at the neurovascular multidisciplinary team meeting and the patient underwent surgery with complete disconnection of the fistula confirmed on a post-operative DSA.



Fig. 4. A catheter angiogram with DSA in arterial phases (A–D) demonstrates the ethmoidal dAVF - feeders from bilateral ethmoidal arteries - venous drainage into bilateral orbitofrontal veins and the right frontopolar vein; (A) Right ICA injection – AP, (B) Right ICA injection – Lateral, (C) Left ICA injection – AP, (D) Left ICA injection – AP, (D) Left ICA injection – Lateral. Native fluoroscopic images demonstrate the position of the bilaterally inflated Scepter mini-balloons and the cast formed by the liquid embolic agent after embolisation; (E) Pre-embolisation – AP, (F) Pre-embolisation – Lateral, (G) Post-embolisation – AP, (H) Post-embolisation – Lateral. Note: on the AP views (E, G) there is superimposition of the embolic cast from the previously treated occipital dAVF. Post-embolisation AP and lateral imaging with ICA injections in arterial phase demonstrated resolution of the dAVF (I–L).

5. Discussion

A meta-analysis by Giannopoulos et al (2019) which predates the availibility of SMBM focused specifically on surgical treatment of dAVFs in an ethmoidal location. This suggested that surgery is superior to endovascular treatment in achieving complete obliteration and has a similar complication rate.⁷ However, surgery is associated with longer hospital stays and longer recovery in addition to the morbidity associated with a craniotomy. Therefore, new technologies that have been developed in the realm of endovascular intervention must be regarded as an opportunity to overcome the current challenges in this approach. This way, more dAVF can become amenable to endovascular treatment.

Multiple cases of endovascular treatment for ethmoidal dAVFs using the conventional (non-balloon) and larger balloon (Scepter XC) microcatheters have been reported in the literature. However, this has been associated with certain limitations and risk of complications.^{8,11} Pre-dating the availability of SMBM, a meta-analysis published by Xu et al (2019) evaluated 48 patients treated for an anterior cranial fossa dAVF between 1997 and 2018 (the largest available series).⁸ This reported an 8.3 % incomplete obliteration and 8.3 % complications which included a retained microcatheter, LEA reflux, and parenchymal oedema - none of which resulted in permanent neurological deficit. Other complications reported in the literature include posterior ischaemic optic neuropathy (PION), small subdural haematoma secondary to a micro-perforation, and acute visual loss.^{10–12}

Initial in vivo and in vitro studies with the SMBM had suggested that this extra-small dual-lumen micro-balloon catheter is superior to standard microcatheters in embolisation procedures, with shorter procedural time, significantly lower risk of reflux, and more comprehensive embolisation.¹³ Subsequent clinical work with this device used for different applications was also promising, enabling treatment via small vascular feeders with the possibility of superselective flow arrest and navigation support.¹⁴ To our knowledge there has only been 3 cases previously documented in the literature which are specific to the use of the SMBM for transophthalmic embolisation of anterior cranial fossa dAVFs. These were part of a series of cases by Mayercik et al (2021), with two of these cases reported separately in more detail by Pulli et al (2020) (see Table 1).^{11,15}

All three patients in our series had ethmoidal dAVF with arterial feeders from both sides. These were treated with a simultaneous bilateral transarterial approach using the SMBM. Complete obliteration of the ethmoidal dAVF was achieved in a single embolisation session in two out of three cases. Although the other case required surgical resection of the dAVF, the partial occlusion achieved with this technique allowed for an easier disconnection of the fistula. No endovascular complications were encountered.

Our simultaneous bilateral transophthalmic (arterial) injection of 25 % PHIL differs from the approach applied by Pulli et al (2020) which involved unilateral transophthalmic (arterial) injection of Onyx-18.¹⁵ Similarly, we avoided the use of glue (N-butyl cyanoacrylate) to treat these dAVFs.. Although good outcomes have also been reported with glue, some literature suggests that transarterial embolisation with this agent may carry a higher risk of microcatheter occlusion and proximal small arterial feeders without reaching the fistulous point.^{9,16,17} This is also associated with greater inter-operator variability.¹⁷ Combined use of a LEA and coils has also been reported.¹¹

The SMBM was successfully navigated into the distal targeted arterial branches in all our cases. As reported by Gross et al (2016), distal access has been an issue in some cases that used conventional microcatheters.¹⁸ The SMBM also prevented reflux in all cases, which is another issue with conventional microcatheters as reported by Li et al (2013) and Limbucci et al (2018).^{12,19} Strict control of the embolic agent and avoidance of reflux is essential for several reasons. Importantly, it avoids the risk of retinal ischaemia with acute visual loss.¹² Other problems that arise from reflux include: a decreased chance of satisfactory distal penetration across the dAVF (with occlusion of the draining vein for a sustained cure), premature abortion of the procedure with incomplete treatment, and prohibits future access of the same arterial pedicle for subsequent embolisation. Another complication that has been prevented because of the low-profile property of SMBM is that of PION. This complication was reported by Mayercik et al (2021) and thought to be secondary to mechanical occlusion of the ophthalmic artery by the larger conventional microcatheter, rather than due to reflux of the embolic agent.¹¹

Comparisons and contrasts can be drawn between our case series and those of other authors in relation to a bilateral vs unilateral approach. Piergallini et al (2019) reported two cases with a bilateral approach to problem solve the issue of conventional microcatheters without a balloon that did not allow sufficient penetration of the embolic agent across the dAVF.¹⁰ In order to achieve definitive occlusion, each side was reportedly targeted separately, with one case targeting the contralateral side in a delayed intervention (1 month apart). Mayercik et al

Table 1

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Reported cases in the literature which are specific to the use of the SMBM for transophthalmic embolisation of anterior cranial fossa dAVFs.
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Author/ Year	Presentation	Feeding arteries	Venous drainage	Borden type	Approach	Agent	Obliteration	Complications
Pulli et al/ 2020 [17]	Asymptomatic	Bilateral ethmoidal and lacrimal, arteries, frontal branches of the bilateral middle meningeal arteries, and the left superficial temporal artery	Orbitofrontal and frontopolar veins into basal vein	Ш	Unilateral transarterial	Onyx- 18	Complete	No
Pulli et al/ 2020 [17]	Headache, left-sided pulsatile tinnitus	Unilateral ethmoidal arteries	Unilateral orbitofrontal vein into the superior sagittal sinus	III	Unilateral transarterial	Onyx- 18	Complete	No
Gravino et al/2022 [our series]	Headache, dizziness, poor balance, fatigue	Bilateral ethmoidal arteries and frontal branches of bilateral middle meningeal arteries	Right frontopolar vein	III	Bilateral transarterial	25 % PHIL	Complete	No
Gravino et al/2022 [our series]	Tonic-clonic seizures secondary to right frontal intracerebral haemorrhage	Bilateral ethmoidal arteries	Right orbitofrontal and superficial middle cerebral vein	III	Bilateral transarterial	25 % PHIL	Complete	No
Gravino et al/2022 [our series]	Headache, pulsatile tinnitus, vision impairment (left eye), disturbed sleep, occipital pulsatile swelling	Bilateral ethmoidal arteries	Bilateral orbitofrontal veins and right frontopolar vein	III	Bilateral transarterial	25 % PHIL	Partial (required surgical resection of residual fistulation)	No

(2021) also reported a case of an anterior cranial fossa dAVF with bilateral arterial feeders that was incompletely cured with a unliteral approach and required surgical resection.¹¹ Pulli et al (2020) reports an ethmoidal dAVF having bilateral arterial feeders that was successfully occluded with unilateral application of the SMBM. Although this is a single case, it is testament to the ability of the SMBM in providing good penetration while avoiding reflux.¹⁵

Endovascular embolisation with simultaneous bilateral application of the SMBM for balloon flow-arrest and bilateral injection of the embolic agent was the techqniue applied in the small series we report. This resulted in a cure in two cases and allowed for easier surgical resection in another case. The complication rate of our series was maintained at 0 %, equivalent to that by Pulli et al (2020) and less than the 8.3 % previously reported with conventional microcatheters.¹⁵

This approach appears to provide better penetration of the LEA towards the fistulous point, increasing the chances of a complete and sustained cure. Bilateral balloon inflation allows for flow arrest through the dominant feeders, such that even if the injected LEA is more voluminous from one side, the contralateral balloon inflation still serves to arrest flow through another dominant feeder. Some ethmoidal dAVFs may have other feeders aside from branches from the ophthalmic arteries, such as ECA feeders in Case 1. While it may not be possible to arrest flow from all feeders, the possibility of doing this from bilateral dominant intracranial feeders offers less resistance to the injected LEA. Although this approach theoretically poses a threat to both eyes, the low profile and greatly reduced risk of reflux with SMBM along with the improved flow dynamics of a bilateral approach, makes visual complications much less likely. In addition, the use of SMBM offers a significantly safer option than detachable microcatheters, since these require a pulling manoeuvre which can be traumatic in such small vasculature.

6. Conclusion

This report supports the application of the SMBM for endovascular embolisation of ethmoidal dAVFs and provides a new method to apply this technology. Simultaneous bilateral application of the SMBM for balloon flow-arrest and bilateral injection of the embolic agent is feasible and safe. Going forward, larger case series on the SMBM for this purpose will provide stronger evidence of its efficacy and perhaps also allow for comparison between unilateral and bilateral approaches in terms of the number of interventions required, rate of recurrence, and rate of complications.

CRediT authorship contribution statement

Gilbert Gravino: Writing - original draft, Conceptualization. Richard Pullicino: Writing - review & editing. Mani Puthuran: Methodology. Ynyr Edwards: Data curation. Jawad Yousaf: Methodology. Emmanuel Chavredakis: Methodology. Arun Chandran: Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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