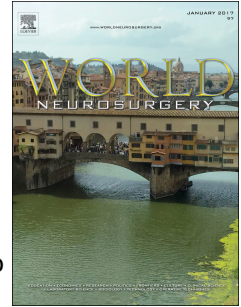


# Accepted Manuscript

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**Spontaneous resolution of a true intracanalicular ophthalmic artery aneurysm after endovascular treatment of an associated dural arteriovenous fistula**

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## **Spontaneous regression of a true intracanalicular fusiform ophthalmic artery aneurysm after endovascular treatment of an associated dural arteriovenous fistula**

### ABSTRACT

Ophthalmic trunk aneurysms associated to other vascular malformations are extremely rare, and little is known regarding its natural history and options of treatment. We present the interesting case of a 51-year-old man who was admitted with progressive visual loss secondary to a concurrent fusiform ophthalmic trunk aneurysm associated to a dural arteriovenous fistula (DAVF). He was treated with transarterial embolization of the fistulous point. A follow-up angiogram at 6 months showed the complete obliteration of the DAVF and regression of the ophthalmic trunk aneurysm. The literature is reviewed, and potential pathophysiological mechanisms leading to this association and regression in this subgroup of aneurysms are discussed.

### INTRODUCTION

Ophthalmic aneurysms arise from the superomedial wall of the internal carotid artery (ICA) just distal to the origin of the ophthalmic artery (OphA)(1). These aneurysms account for approximately 10-15 % percent of all intracranial aneurysms. However, aneurysms that arise from the trunk of ophthalmic artery are extremely rare, and only a few cases are reported in the literature(2).

The association between ophthalmic trunk aneurysms and vascular malformations has been previously described(3), although their relationship and natural history has not been clearly discussed.

We present a rare case of complete obliteration of an ophthalmic trunk aneurysm after endovascular treatment of an associated dural arteriovenous fistula (DAVF)

## CASE REPORT

A 51-year-old man was admitted with progressive visual loss of the right eye (visual acuity 20/50). He had no significant medical history, specifically no history of peripheral or cerebral vascular disease.

A Magnetic Resonance Imaging (MRI) showed a dilated vascular structure along the right ambiens cistern. Subsequent cerebral angiography revealed a tentorial DAVF, mainly fed by branches from the artery of Bernasconi-Cassinari and the right middle meningeal artery (MMA). It had also tributaries from ethmoidal branches of the OphA and the occipital artery (OccA). Selective ICA and external carotid artery (ECA) injections showed no choroidal blush. The DAVF was drained exclusively via the left basal vein of Rosenthal (●► Fig. 1). Furthermore, an unruptured 5 mm fusiform aneurysm arising from the intracanalicular segment of the right OphA was identified. (●► Fig. 2)

#### Treatment

After general anesthesia was induced, a 6-French guide catheter was placed in the right ECA. A Medtronic Marathon microcatheter was navigated over a Transend 0.010 microwire into the distal right MMA under roadmap guidance. The fistulous point was then occluded with Onyx-18. No embolism of the basal vein of Rosenthal or other veins was observed during the procedure, and a considerable reduction of the caliber of the ethmoidal arterial feeders (●► Fig. 3).

#### Outcome and follow-up

The angiographic follow-up 6 months later confirmed the complete obliteration of the DAVF. It also showed interval decrease of the caliber of the OphA and its ethmoidal branches, as well as regression of the ophthalmic trunk aneurysm. (●► Fig. 3).

The patient experienced partial improvement on his visual acuity at 6 months-follow-up (from 20/50 to 20/30).

## DISCUSSION

True ophthalmic trunk aneurysms usually present with visual symptoms, including visual loss and papilledema(2). Neuro-ophthalmological findings could be explained by direct compression or perfusion reduction of the optic nerve rather than aneurysmal rupture(4) . We present a case of DAVF and a concomitant ophthalmic trunk aneurysm found on a patient that presented with severe visual impairment on the right eye, which resolved after endovascular occlusion of the DAVF.

The most unusual angiographic finding in our case was the spontaneous regression of the untreated right ophthalmic trunk aneurysm. The association between ophthalmic trunk aneurysms and other vascular malformations has been previously reported, although spontaneous aneurysm regression after the treatment of the vascular malformation was not detected in any of them(2, 3). Kawaguchi et al.(5) reported a case with a symptomatic (frontal headaches) anterior fossa DAVF fed by an OphA harboring an unruptured aneurysm of the intraorbital segment. The DAVF was surgically obliterated, although angiographic images or information about the state of the aneurysm during follow-up were not reported. Kirsh et al(3) reported a slightly different case of a ruptured intraorbital ophthalmic artery aneurysm associated with a middle fossa DAVF with cortical venous drainage. The aneurysm was treated by endovascular coil occlusion and the DAVF was occluded by transvenous obliteration of the draining basal vein of Rosenthal.

Several factors, which may lead to aneurysm formation, have been described, including disruption of the internal elastic lamina, focal inflammatory changes and increased hemodynamic stress of the arterial wall(6). The contribution of hemodynamic changes after carotid artery occlusion to the development of flow-related aneurysms in humans was highlighted in a report by Senn et al(7). The hemodynamic theory was also proposed as a possible explanation of the *de novo*

formation of a giant posterior cerebral artery (PCA) aneurysm in a previous case report that was authored by this group(8). Kondziolska et al(9) described the phenomenon in which flow-related aneurysms regressed after AVM treatment, while those not directly associated to the AVM remained intact. Tsimpas et al(10) reported a case of a patient with an untreated, flow-related right frontal M2 aneurysm and a large, dysplastic, ruptured right supraclinoid ICA aneurysm that was treated with a flow diverting stent and coils. It was hypothesized that the flow diversion away from the M2 aneurysm reduced the tension against the aneurysmal wall and allowed its indirect repair. We hypothesize this was, most likely, the responsible mechanism in our case, where one ethmoidal branch was one of the main feeding to the tentorial DAVF. As increasing the hemodynamic stress may develop cerebral aneurysms in the feeding vessel, reduction of the blood flow and the hemodynamic stress may reverse the situation. As such, this hypothesis is enhanced, in our current case, after the endovascular treatment, in which, sequential angiograms demonstrated either progressive resolution of the true Ophthalmic trunk aneurysm as a narrower diameter in the ethmoidal branch (Figure 3).

It was also remarkable, in our case in regards to the radiological presentation is that the associated DAVF was located in a very uncommon location, such as the free margin of the tentorium, far away from the location of the OA aneurysm. Surgical(11) and/or endovascular obliteration(12, 13), particularly via the transvenous route, can be used to cure this type of DAVF. Surgical disconnection using a posterior interhemispheric route has been described, although this approach requires excellent surgical skills, and even so, potential damages to adjacent neurovascular structures cannot always be avoided(11). Endovascular treatment of DAVFs may be difficult due to limited transarterial or transvenous access to the fistulous point(13). The transvenous approach carries the risk of aggravating the situation by partially occluding the venous outlet without obliterating the DAVF(13). A surgeon needs to preserve all normal venous drainage, particularly in DAVFs with leptomeningeal venous drainage. Failure to do this may result in a catastrophic venous infarct(3).

Options of direct treatment of true ophthalmic trunk aneurysms include trapping, clipping and endovascular embolization(2). Although good clinical outcomes have been reported by some authors(4), treatment of true ophthalmic trunk aneurysms may lead to visual loss and oculomotor nerve impairment due to the inadvertent occlusion of the blood supply to the eye and orbit and direct manipulation to neural structures(14).

In our case, the DAVF was successfully obliterated after transarterial embolization of one of the feeders from the ECA, and the ophthalmic trunk aneurysm subsequently decreased its size. Occasionally, arterial embolization only may reduce the blood flow to the DAVF and provoke the closure of the abnormal arteriovenous communication and thus cure the DAVF without risking inadvertent embolization of the venous drainage(12). Our case suggests that treatment of the symptomatic vascular malformation only may lead to spontaneous regression of a distant, flow-related aneurysm that may be very difficult or risky to treat.

It is important to emphasize that our results are purely anecdotal and should not be generalized. Additional vascular and cross sectional studies are necessary to determine whether this is a safe and durable treatment method for treating this subgroup of aneurysms.

## CONCLUSIONS

True ophthalmic trunk aneurysms are very rare entities that may form as a consequence of increased hemodynamic stress on the OphA secondary to an associated vascular malformation. Endovascular obliteration of the malformation may lead to spontaneous regression of the aneurysm, likely due to reduction of the hemodynamic stress against the arterial wall.

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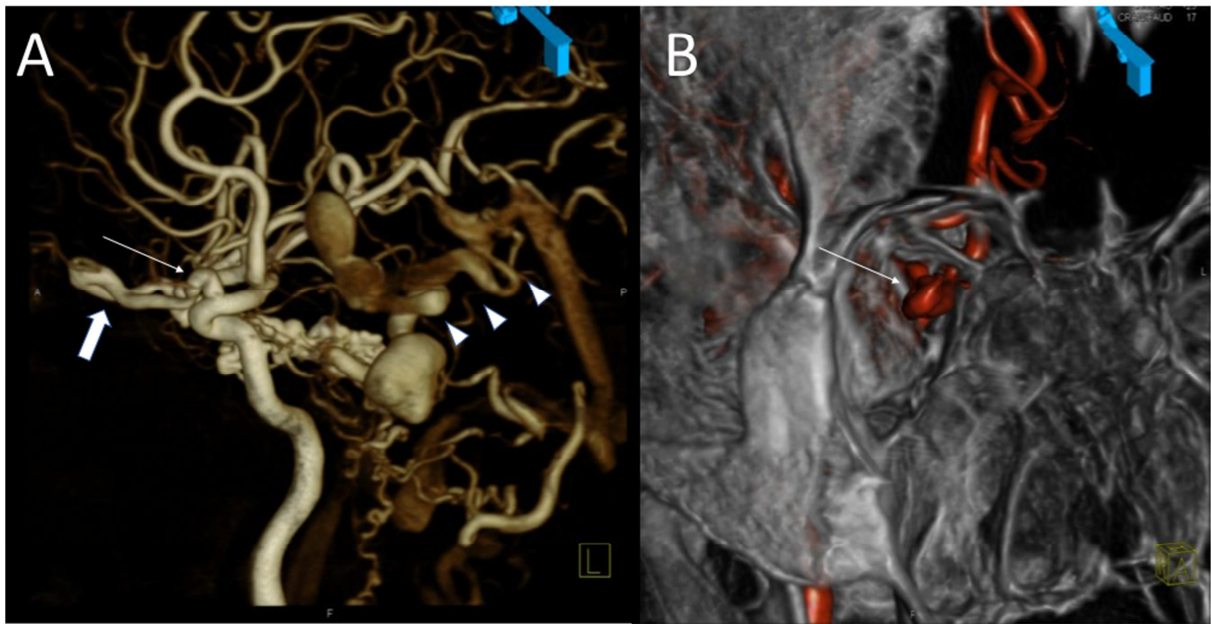
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## FIGURES

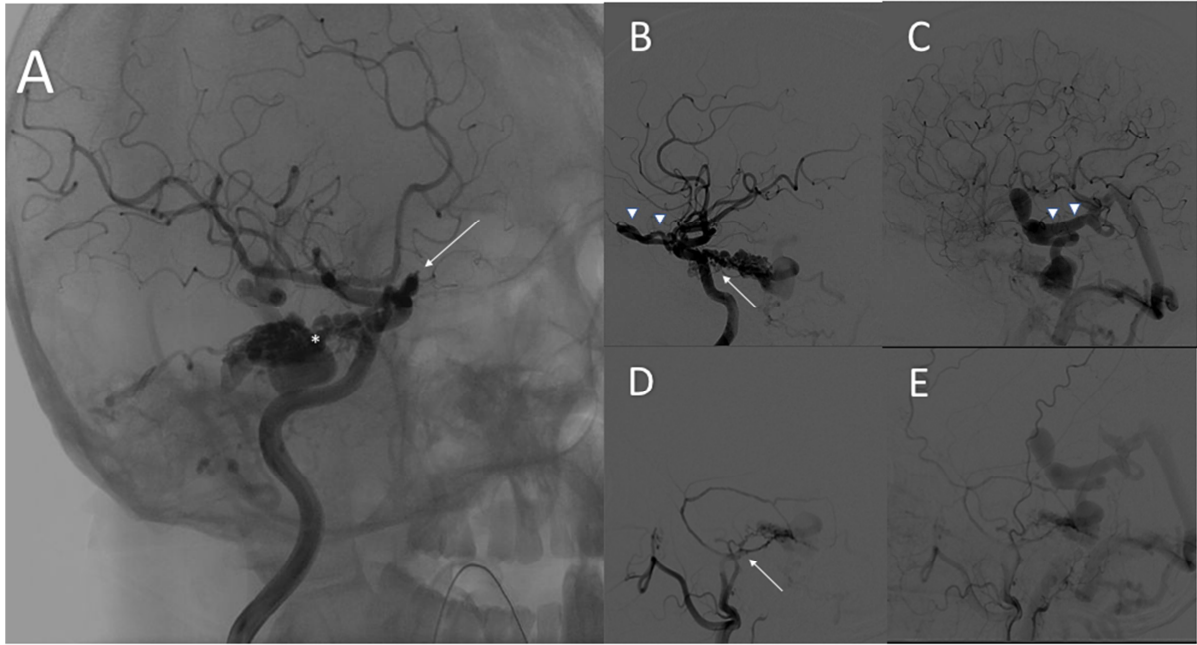
Figure 1. 3D angiogram reconstruction of the right ICA injection (A) shows dilated ophthalmic and ethmoidal branches (thick arrow) feeding incisural dAVF and draining into the sinus rectus via Basal Vein of Rosenthal (Arrow heads). There is a dilated fusiform aneurysm arising from the OA trunk (white arrow). Using the bone mask (B) it is observed that the OA aneurysm is located at the canalicular segment (white arrow).

Figure 2. Preoperative cerebral angiogram. Oblique projection of the right ICA injection (A) demonstrates a 4 mm-superiorly pointed OA trunk aneurysm (white arrow), associated with a tentorial incisura dAVF (asterisk). Lateral projection ICA injection (B and C) shows arterial dAVF feeders from ethmoidal branches (arrow heads) and tentorial branches coming from the cavernous ICA (Bernasconi-Casani artery) (white arrow) (B); and an early venous filling of the basal vein and Vein of Galen, draining into the sinus rectus. The lateral projection of an ECA injection also demonstrates some AVF arterial tributaries from the middle meningeal artery (white arrow) (D) and early venous filling (E).

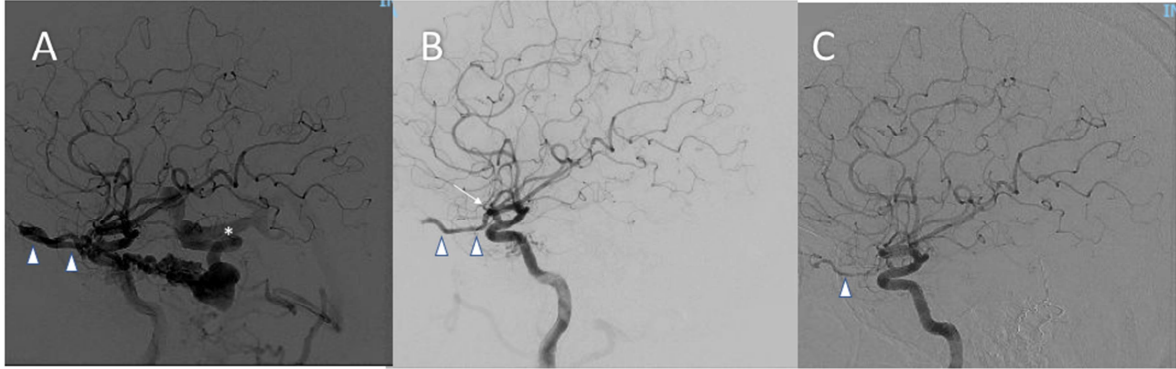
Figure 3. Cerebral angiogram ICA injection at admission (A), post-embolization (B) and 6 months follow-up (C). Preoperative angiogram (A) demonstrated dilated OA and ethmoidal branches feeding the dAVF (arrow heads) with early venous filling of basal vein of Rosenthal (asterisk) and vein of Galen. Immediate postoperative ICA injection (B) showed no signs of early venous filling nor filling of the ethmoidal branches. At 6 month-follow-up a diminution in caliber of the ophthalmic artery (arrow heads) and partial resolution of the OA trunk aneurysm (white arrow) is observed.



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## ABBREVIATIONS

dural arteriovenous fistula (DAVF)

ophthalmic artery (OphA)

internal carotid artery (ICA)

Magnetic Resonance Imaging (MRI)

middle meningeal artery (MMA)

occipital artery (OccA)

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## KEY POINTS

- True ophthalmic trunk aneurysms are very rare entities that normally present with visual symptoms. Neuro-ophthalmological findings are presumably due to direct compression or perfusion reduction of the optic nerve.
- True ophthalmic trunk aneurysms may form as a consequence of increased hemodynamic stress on the Ophthalmic artery, secondary to an associated vascular malformation.
- Endovascular obliteration of the malformation may lead to spontaneous regression of the aneurysm, likely due to reduction of the hemodynamic stress against the arterial wall.
- It is important to emphasize that our results are purely anecdotal and should not be generalized. Additional vascular and cross sectional studies are necessary to determine whether this is a safe and durable treatment method for treating this subgroup of aneurysms.