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Case Report

# Endovascular Approach to Concurrent Anterior Cranial Fossa Dural AVF and Concurrent Flow-Related Ophthalmic Artery Aneurysm: A Case Study

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## Abstract

Dural arteriovenous fistulas (DAVFs) with concurrent flow-related ophthalmic artery aneurysms, particularly intraorbital aneurysms, are rare. Retrograde cortical blood flow and sinus thrombosis heighten the risk of rupture and optic nerve compression, yet treatment strategies remain poorly defined. This study aims to explore effective management for these complex cases. A 66-year-old hypertensive man presented with an acute severe headache, loss of consciousness, and decreased vision in the left eye. Imaging revealed a left frontal intracranial hemorrhage and an anterior cranial fossa DAVF with a left ophthalmic artery aneurysm. Endovascular embolization via the left ophthalmic artery, using a microcatheter distal to the aneurysm and controlled reflux of glue injection (N-butyl cyanoacrylate and lipiodol), successfully treated the DAVF and ophthalmic artery aneurysm. Visual acuity improved to finger counting at 1 foot, with no residual DAVF at 6 months. This case demonstrates that controlled glue embolization is a viable alternative when microsurgery is not feasible due to complex anatomy.

**Keywords**

- concurrent ophthalmic aneurysm
- dural arteriovenous fistula
- glue embolization
- visual acuity

## Introduction

The natural history, origin, and future course of dural arteriovenous fistula (DAVF) have been subject to varied opinions. Due to flow-related changes in the arteries and draining veins, patients may initially present with milder symptoms such as bruit, vertigo, and headache. However, the subsequent progression to retrograde blood flow in the cortical vessels and sinus thrombosis complicates the condition by recruiting new arteries, developing aneurysms in its proximity to arteries, and causing congestion in draining cortical veins and sinuses, which increase the risk of bleeding of both the aneurysm and ectatic veins.[1–3]

The combined presence of concurrent aneurysms with DAVF significantly raises the likelihood of bleeding, estimated at nearly 20%. Specifically, concurrent flow-related aneurysms, such as intraorbital aneurysms associated with distal DAVF, are exceedingly rare, with only approximately 21 case reports documented.[4] The management of such cases becomes particularly challenging when bleeding is present, especially if there is an associated proximal feeding artery aneurysm. If these lesions are treated surgically, the intraorbital aneurysm may rupture due to hemodynamic changes, and the DAVF may not be completely obliterated due to persisting residual feeders. This aneurysm may compress

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adjacent critical structures, such as the optic nerve, leading to progressive worsening and a heightened risk of rupture and intraorbital bleeding if left untreated in such a scenario. Endovascular treatment using glue embolization carries an inherent risk of incomplete obliteration of dural arteriovenous fistulas (AVFs) and aneurysms, as well as the potential for unintentional blockage of vital arteries, including proximal retinal arteries. These risks underscore the critical importance of controlled microcatheter injection with meticulous precautions to prevent adverse outcomes.

Few case reports have suggested the possibility of spontaneous regression of aneurysms following the treatment of distal DAVFs on flow-unrelated aneurysms. Still, management strategies with presentations similar to our patients have not been reported.[5,6] While microsurgical options were unsuitable, we tried to present a treatment strategy that took care of both DAVF and concurrent aneurysms without affecting the flow in retinal vessels.

## Case Description

A 66-year-old hypertensive man presented with acute-onset severe headache, a fall, and loss of consciousness. He reported progressive left eye vision loss over 3 days, from counting fingers to light perception, while right eye vision remained normal. A seizure followed by unconsciousness was also noted. There was no history of drug use, bleeding disorders, or relevant family history. On examination, his Glasgow Coma Scale (GCS) score was E4 V5 M6, pulse was 86/min, BP was 152/92 mm Hg, and had a grade 3 papilledema in the left eye. A head noncontrast computed tomography revealed a left frontal intracranial hemorrhage with mild cerebral edema without mass effect or midline shift (►**Fig. 1A**).

Given the spontaneous intracerebral hemorrhage (ICH), the patient underwent computed tomography angiography (CTA) of the head (►**Fig. 1B–D**), which revealed an aneurysm on the ophthalmic artery and DAVF near the anterior cribriform plate with draining ecstatic dilated veins. Subsequently, the patient underwent a six-vessel selective digital subtraction angiography (DSA) in an emergency on a biplane DSA machine. The report revealed an anterior cranial fossa DAVF supplied by both ophthalmic arteries, anterior ethmoidal artery, and branches from the middle meningeal artery, with a retrograde reflux into cortical veins and a thrombosed anterosuperior sagittal sinus. Additionally, a left flow-related ophthalmic artery aneurysm measuring $5 \times 4$ mm with a neck of 3.3 mm was noted inside the orbit distal to the bend of the ophthalmic artery from an inferolateral position to a superomedial position with retinal vessel arising proximal to the bend (►**Fig. 2**).

The treatment plan was challenging due to the complex anatomy, which included an intraorbital aneurysm, multiple feeding arteries to the DAVF, a draining vein with reflux into the cortical draining vein, and a thrombosed anterosuperior sagittal sinus, precluding venous access for embolization and making an open surgical procedure cumbersome. Microsurgery was deferred as it could have led to incomplete closure of DAVF with residual feeding artery, which could have recruited more artery and veins and also could led to progression of

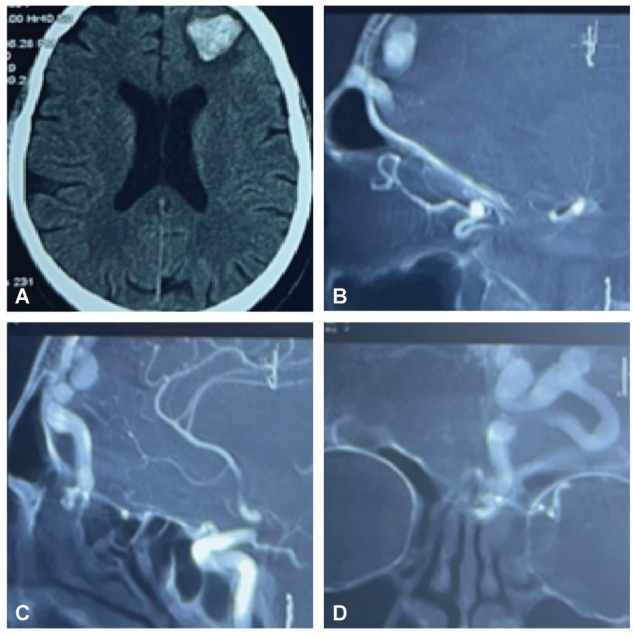


**Fig. 1** (**A**) A 66-year-old hypertensive, nondiabetic man presented with a 3-day history of visual blurring, ranging from the perception of finger movements at 1 foot to bare light perception. He experienced a sudden loss of consciousness, a severe headache, and two seizures within the past 3 days. On admission, his Glasgow Coma Scale (GCS) score was 15/15. Direct fundoscopic examination revealed grade 3 papilledema. A noncontrast computed tomography scan of the head identified a left frontal basal hematoma. (**B**) Computed tomography angiography (CTA) of the brain, sagittal view, showing an intracanalicular ophthalmic segment aneurysm. (**C**) Sagittal view CTA of the brain with a paramedian cut displaying a dural arteriovenous fistula at the planum near the anterior cribriform plate. Multiple feeders and a dilated draining vein are suspected. (**D**) Coronal view CTA of the brain revealing a dural arteriovenous fistula at the planum near the anterior cribriform plate in the basal frontal region, with multiple suspected feeders and a dilated draining vein.

untreated aneurysm progression of size and rupture with catastrophic bleeding. Consequently, an endovascular approach was chosen to address both the aneurysm and arteriovenous malformation (AVM) of the ophthalmic artery, with a plan for placing the microcatheter tip distal to the aneurysm near the fistulous opening and glue embolization of the AVF along with controlled backflow to completely embolize the aneurysm while preserving the retinal vessels arising very proximal to it, which had good collateral flow as demonstrated by a balloon occlusion test of the left internal carotid artery (ICA) near the ophthalmic artery origin (►**Figs. 2C, D** and **3**).

Although other treatment options, such as clip placement on the DAVF by open surgery, were considered, the patient and his family opted against open surgical procedures in favor of embolization. Hence, endovascular embolization with controlled reflux of the embolic agent was planned to also embolize the aneurysm while preserving the central retinal artery to prevent visual loss.

A 6-month follow-up revealed a normal left fundus, with vision improved to finger count at 1 foot. A follow-up DSA at 6 months suggested no residual DAVF, aneurysm, and patency of proximal ophthalmic and retinal artery (►**Fig. 4**).

 

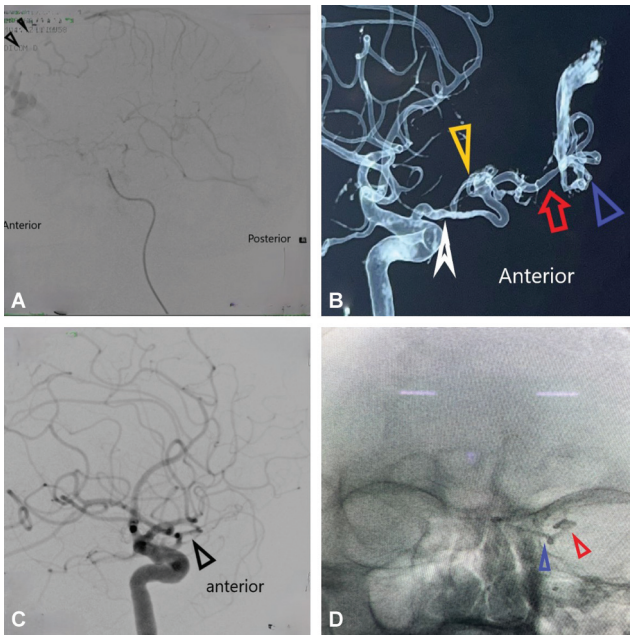


**Fig. 2** (**A**) Angiography of the left internal carotid artery in the venous phase demonstrating a thrombosed anterosuperior sagittal sinus with venous reflux into the dilated cortical venous system. (**B**) Lateral view and 3D reconstruction of left internal carotid artery angiography showing a left ophthalmic artery aneurysm and a distal dural arteriovenous fistula (AVF) draining through a dilated vein with venous reflux into the ectatic cortical vein. The *white arrowhead* indicates the origin of the retinal artery before the ophthalmic artery bends from an inferolateral to a superomedial position. The *yellow arrowhead* marks the aneurysm distal to the bend. The *blue arrowhead* denotes the fistula site, and the *red arrowhead* shows the microcatheter's position between the aneurysm and the fistula, where the glue was injected. (**C**) Lateral view of internal carotid artery injection showing successful obliteration of the left ophthalmic artery aneurysm and dural AVF, with preservation of the proximal ophthalmic artery. The *arrowhead* indicates the preserved segment of the ophthalmic artery and the retinal artery's origin. (**D**) Skull Towne's view showing the glue cast in the intraorbital segment of the ophthalmic artery, demonstrating complete obliteration of the aneurysm and fistula.

## Discussion

Unlike other AVMs, which may have a congenital origin, DAVFs are often considered acquired lesions, potentially resulting from head trauma, dural venous sinus thrombosis, or postcraniotomy changes.[1–3] In this study, we observed anterior sagittal sinus thrombosis, which may have led to cortical venous reflux, recruitment of vessels from the ipsilateral ethmoidal and middle meningeal arteries, and cortical venous ectasia. This phenomenon has been previously hypothesized in earlier studies.[1–3] No predisposing etiology for anterior sagittal sinus thrombosis was identified in our patient, who lacked a history of cardiac disease, coagulation defects, or thromboembolic sources, as revealed by echocardiography and duplex ultrasound of the limb vessels. Venous hypertension or an unnoticed trivial head injury may have contributed to the sagittal sinus thrombosis and subsequent DAVF development in our patient. The predominance of elderly male patients in this study and in previous case reports further supports an acquired rather than congenital etiology[7–10] (►**Table 1**).

Retrograde blood flow in the ophthalmic artery from ectatic cortical veins as anterior sagittal sinus was thrombosed, leading to further backward congestion of the retinal artery and, second, progressive enlargement of ophthalmic artery aneurysm causing compression over the retinal artery and optic nerve, which may have led to visual deterioration. This was also indicative of an impending aneurysmal rupture. While progressive visual loss due to enlargement of an intraorbital ophthalmic aneurysm in the context of a flow-related distal DAVF had not been previously reported, visual problems, exophthalmos, and proptosis due to ophthalmic vessel congestion have been documented in cases of ophthalmic aneurysmal rupture with or without concurrent DAVF.[7–10]

Our patient's preoperative DSA revealed multiple feeders from the left anterior ethmoidal artery, meningeal branches of the internal maxillary artery, and bilateral ophthalmic arteries, with venous reflux into the cerebral cortical veins and ectasia. Spontaneous regression of aneurysms with distal DAVF has been reported in only 6.5% of patients, necessitating definitive management through microsurgery or endovascular approaches.[1,2] While previous studies have favored venous approaches for fistula embolization, the absence of a prominent draining channel, such as the basal vein of Rosenthal, in our patient with dural sinus thrombosis rendered this approach unfeasible.[6,7] The anterior cranial fossa location of the DAVF is generally favorable for microsurgical approaches. Still, uncertainty regarding the exact localization of the DAVF and the potential for recruitment of feeding arterial vessels leading to recurrence of fistulas and progressive enlargement of untreated aneurysm causing its rupture in the present patient posed significant challenges, together with the unwillingness of patients to have surgery. In some cases, surgical excision of DAVF and coiling of the ophthalmic aneurysm have been employed.[8,11] However, this approach is less practical for aneurysms on flow-related proximal vessels prone to rupture due to hemodynamic changes following distal DAVF obliteration by surgery, specifically when the aneurysm is located intraorbitally. Radiosurgery was also deemed unsuitable due to the unstable nature of the DAVF with associated bleeding and the impending rupture of the flow-related aneurysm, as reported in anecdotal cases of concurrent aneurysms with anterior cranial fossa DAVF without bleeding.[10]

Based on existing studies and our experience of managing concurrent aneurysms with AVM following the aneurysm-first principle in patients with hemorrhagic manifestations,[12] we opted for glue embolization to treat the left ophthalmic artery aneurysm and DAVF. We positioned the flow-directed microcatheter distally near the fistulous communication ahead of the aneurysm, allowing backflow proximally up to the aneurysm while preserving the proximal segment of the retinal artery. The microcatheter used in this patient was a flow-guided Marathon microcatheter (Medtronic, Minneapolis, Minnesota, United States) with the Asahi Chikai 008 microwire (Asahi Intecc Co., Ltd., Aichi, Japan). More than 50% flow sluggishly in distal directions and

 

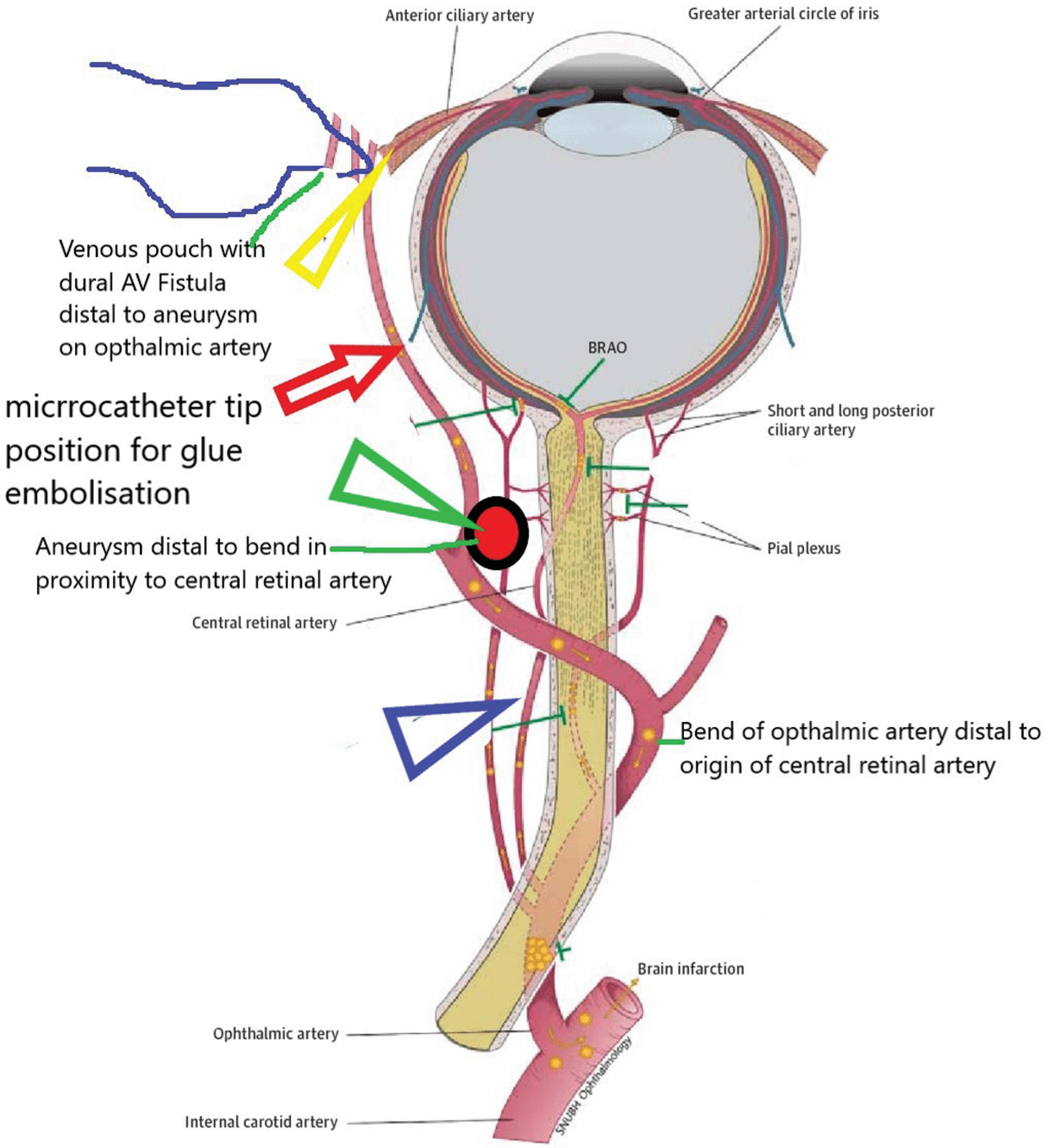


**Fig. 3** Schematic diagram illustrating the anatomical course of the ophthalmic artery originating from the internal carotid artery. Initially, the ophthalmic artery lies inferolateral to the optic nerve, bending over the optic nerve to a superomedial position. The retinal artery arises proximal to this bend (*blue arrowhead*). The aneurysm is located distal to the bend (*green arrowhead*), while the fistulous communication is situated distally (*yellow arrowhead*). The *red arrowhead* indicates the microcatheter's placement between the aneurysm and the fistula for glue injection, performed using the pressure cooker technique due to an inability to advance the microcatheter distally. AV, arteriovenous; BRAO, Branch Retinal Artery Occlusion.

it could have caused incomplete closure of fistulous communications and left residual feeding arteries. Similarly, more dilute 25 or 30% have more chances of reflux in a backward direction to close the retinal artery and whole ophthalmic artery, so a 40% concentration of N-butyl cyanoacrylate (NBCA) and lipiodol was used.

This approach led to improved visual acuity at the 6-month follow-up, as confirmed by direct fundoscopy and DSA, which showed no new or residual DAVF or aneurysm, an intact proximal ophthalmic artery, and good collateralization of the orbit with normalization of the fundus in the left eye with improved visual acuity.

## Conclusion

This study underscores the importance of recognizing DAVFs as acquired rather than congenital lesions, often developing in venous sinus thrombosis or other secondary insults. In this

 

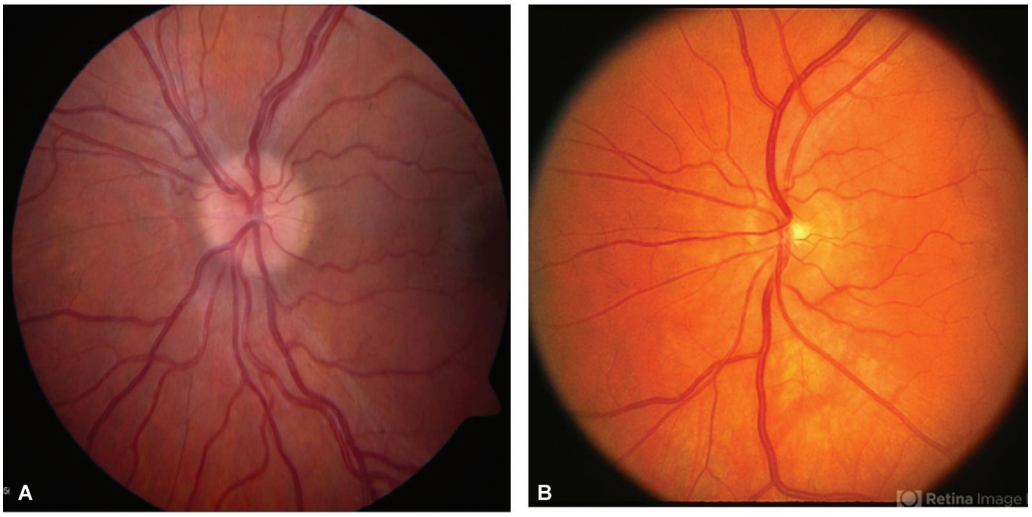


**Fig. 4** (A) Direct fundoscopic image of the left eye showing grade 3 papilledema. (B) Follow-up fundoscopic image of the left eye at 6 months, indicating complete resolution of the papilledema.

**Table 1** Earlier reported cases of flow-related ophthalmic aneurysms with distal dural arteriovenous fistulas and cases in the present study

| Study | Age (y)/ sex | Clinical presentation | DAVF, location, feeders, and drainage | Procedure performed | Follow-up |
|---|---|---|---|---|---|
| Martínez-Pérez et al[5] | 51/M | Progressive decreased visual acuity (20/50) | Right tentorial dural AVF supplied by a tentorial branch of the right ICA, right occipital, and middle meningeal artery with rt ophthalmic intracanalicular aneurysm draining through the basal vein of Rosenthal | Embolization through the middle meningeal artery only with obliteration of the fistulas | 6-mo resolution of right ophthalmic aneurysm with visual improvement to 20/30 |
| Andersson et al[6] | 77/F | Memory loss, seizures, and hypothyroidism | Left ophthalmic, anterior ethmoidal middle meningeal, and sphenopalatine branch | Glue embolization in the left ophthalmic artery and particle embolization in the meningeal artery | Spontaneous regression of aneurysm following embolization at 6 mo |
| Hellstern et al[7] | 77/M | Ventricular hemorrhage, H&H grade 3 | Tentorial DAVF. One aneurysm on intracranial ophthalmic artery, second on intraorbital, and third on the recurrent meningeal artery | Glue embolization through the ophthalmic artery to the deep recurrent meningeal artery. At the second stage, DAVF embolization through the basal vein of Rosenthal (PHIL, ONYX) intraorbital and intracranial aneurysm left untreated | Mild exophthalmos and fourth cranial nerve palsy at 4 wk |
| Kirsch and Henkes[8] | 62/M | Proptosis and visual deterioration, acute visual loss | DAVF at the anterior clinoidal process. Feeders from the left ophthalmic, anterior ethmoidal, and meningeal branch of the internal maxillary artery and draining into the basal vein of Rosenthal | Aneurysm coiling through the left ophthalmic artery and fistulas embolized through the basal vein of Rosenthal | NA |

(*Continued*)

 

**Table 1** (*Continued*)

| Study | Age (y)/ sex | Clinical presentation | DAVF, location, feeders, and drainage | Procedure performed | Follow-up |
|---|---|---|---|---|---|
| Kawaguchi et al[9] | — | Visual deterioration | Unruptured aneurysm on intraorbital ophthalmic artery aneurysm | Surgical treatment of the anterior cranial fossa DAVF | NA |
| Chen et al[10] | 63/M | Sudden loss of consciousness | Ophthalmic aneurysm with anterior cranial fossa DVAF with SAH | Coiling of the aneurysm with radiosurgery | NA |
| Lai et al[11] | 43/M | Bilateral bruit | Anterior cranial fossa DVAF with feeders from bilateral middle meningeal, superficial temporal artery, and right ophthalmic artery draining into sss with cortical venous reflux, flow-related aneurysm in A2 | Glue embolization and surgery | NA |
| Our patient | 66/M | Headache, seizures, loss of consciousness, loss of vision for 3 d with perception of light left frontal hemorrhage | Anterior cranial fossa DAVF with feeders from bilateral ophthalmic, anterior ethmoidal, middle meningeal with retrograde cortical venous reflux and proximal ophthalmic artery saccular aneurysm | Controlled glue embolization of DAVF and aneurysm with preservation of the retinal artery and collaterals with balloon assistance | 6-mo follow-up with finger count at one foot and normal fundus |

Abbreviations: DAVF, dural arteriovenous fistulas; F, female; H&H, Hunt and Hess; Lt, left; M, male; NA, not available; Rt, right; SAH, subarachnoid hemorrhage.

case, anterior sagittal sinus thrombosis and associated cortical venous ectasia likely triggered the DAVF, which, combined with an ophthalmic artery aneurysm, presented unique diagnostic and therapeutic challenges. The novel use of glue embolization, carefully planned to preserve essential arterial structures, resulted in the successful obliteration of the DAVF and aneurysm with marked improvement in visual outcomes. This case supports the efficacy of embolization in DAVF management, especially in anatomically complex scenarios, while highlighting the need for individualized treatment approaches to prevent complications such as aneurysmal rupture.

### Authors' Contributions

V.C.J., V.S.S., and N.K. were the operating surgeons. V.C.J., V.S.S., N.K., G.V., and R.J. conducted the literature search. V.S.S., N.K., G.V., and R.J. prepared the manuscript. V.C.J., V.S.S., N.K., and R.J. edited the final manuscript. V.S.S., N.K., G.V., and R.J. performed the patient's follow-up. All the authors have read and approved the manuscript.

### Patients' Consent

The patient gave written informed consent to participate and have their medical records used for research and academic purposes.

### Funding

None.

### Conflict of Interest

None declared.

## References

1 Abecassis IJ, Meyer RM, Levitt MR, et al; Consortium for Dural Arteriovenous Fistula Outcomes Research CONDOR Collaborators. Assessing the rate, natural history, and treatment trends of intracranial aneurysms in patients with intracranial dural arteriovenous fistulas: a Consortium for Dural Arteriovenous Fistula Outcomes Research (CONDOR) investigation. J Neurosurg 2021; 136(04):971–980

2 Cagnazzo F, Peluso A, Vannozzi R, Brinjikji W, Lanzino G, Perrini P. Arterial aneurysms associated with intracranial dural arteriovenous fistulas: epidemiology, natural history, and management. A systematic review. Neurosurg Rev 2019;42(02):277–285

3 Gross BA, Ropper AE, Du R. Cerebral dural arteriovenous fistulas and aneurysms. Neurosurg Focus 2012;32(05):E2

4 Garala P, Virdee J, Qureshi M, Gillow T. Intraorbital aneurysm of the ophthalmic artery. BMJ Case Rep 2019;12(04):e227044

5 Martínez-Pérez R, Tsimpas A, Ruiz Á, Montivero A, Mura J. Spontaneous regression of a true intracanalicular fusiform ophthalmic artery aneurysm after endovascular treatment of an associated dural arteriovenous fistula. World Neurosurg 2018; 119:362–365

6 Andersson T, Kihlström L, Söderman M. Regression of a flow-related ophthalmic artery aneurysm after treatment of a frontal DAVS. A CASE REPORT. Interv Neuroradiol 2004;10(03):265–268

7 Hellstern V, Aguilar Pérez M, AlMatter M, Bäzner H, Henkes H. Ophthalmic Artery aneurysm: flow-induced intracranial and intraorbital ("peripheral") ophthalmic artery aneurysms, associated with a tentorial dural arteriovenous fistula, supplied by the

 

ophthalmic artery; endovascular treatment of the dural arteriovenous fistula and one aneurysm; and conservative management of the remaining ophthalmic artery aneurysms. In: Henkes H, Lylyk P, Ganslandt O, eds. The Aneurysm Casebook. Cham: Springer; 2020

8 Kirsch M, Henkes H. A ruptured intraorbital ophthalmic artery aneurysm, associated with a dural arteriovenous fistula: combined transarterial and transvenous endovascular treatment. Minim Invasive Neurosurg 2011;54(03):128–131

9 Kawaguchi S, Sakaki T, Okuno S, Uchiyama Y, Nishioka T. Peripheral ophthalmic artery aneurysm. Report of two cases. J Neurosurg 2001;94(05):822–825

10 Chen Z, Zhu G, Feng H, Tang W, Wang X. Dural arteriovenous fistula of the anterior cranial fossa associated with a ruptured ophthalmic aneurysm: case report and review of the literature. Surg Neurol 2008;69(03):318–321

11 Lai CW, Agid R, van den Berg R, Ter Brugge K. Cerebral arteriovenous fistulas induced by dural arteriovenous shunts. AJNR Am J Neuroradiol 2005;26(05):1259–1262

12 Jha V, Behari S, Jaiswal AK, Bhaisora KS, Shende YP, Phadke RV. The "focus on aneurysm" principle: classification and surgical principles of management of concurrent arterial aneurysm with arteriovenous malformation causing intracranial hemorrhage. Asian J Neurosurg 2016;11(03):240–254

 