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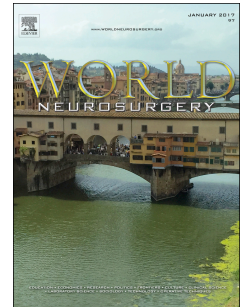
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Dural Arteriovenous Fistula of the Vein of Trolard Mimicking a Cavernous Sinus Fistula

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Conception and design: Dahl, Benndorf. Acquisition of data: Dahl, Benndorf. Analysis and interpretation of data: Dahl, Benndorf. Drafting the article: Dahl, Benndorf. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Benndorf. Administrative/technical/material support: Dahl, Benndorf. Study supervision: Benndorf.

Patient Consent

The patient has consented to the submission of the case report to the journal. Written informed consent from the patient has been obtained.

Abstract

Background: Arteriovenous fistulas (AVFs) involving the cavernous sinus usually become clinically apparent due to eye symptoms. Although rare, the same symptoms can be associated with AVFs located remote from the cavernous sinus when the shunt drains into its tributaries. We report the unusual case of a dural AVF where such communication was not immediately obvious from the diagnostic angiogram.

Case description: A 61-year-old male presented with increasing lid swelling, proptosis and redness of the right eye for 1 month. Digital subtraction angiography showed no evidence for a cavernous sinus fistula but revealed a dural AVF between the middle meningeal artery and the vein of Trolard. The fistula had a minor drainage through a small superficial middle cerebral vein towards the middle cranial fossa. Very late venous phase images eventually revealed faint opacification of the right cavernous sinus and superior ophthalmic vein. Endovascular treatment was performed by a transfemoral access and complete occlusion of the AVF was obtained after two sessions.

Conclusions: Dural AVFs involving cortical veins may cause atypical symptoms suggesting a cavernous sinus fistula due to rerouted venous drainage. Understanding venous anatomy helps to correlate vascular pathology and clinical symptoms, and thus allows efficient and safe treatment.

Introduction

Cavernous sinus fistulas (CSFs) usually present with ocular symptoms related to orbital venous congestion causing eye redness, proptosis, chemosis, and enlarged conjunctival vessels¹. The arteriovenous shunting increases the venous pressure in the cavernous sinus (CS) and superior ophthalmic vein² (SOV) and some patients may as well develop ophthalmoplegia, diplopia, reduced visual acuity, and bruit. Sometimes ocular symptoms are associated with dural arteriovenous fistulas (AVFs) located far from the CS. In such cases eye-related symptoms may rarely be caused by intracranial hypertension and direct compression of cranial nerves from a venous ectasia². However, a more common mechanism involves arterialization of the SOV due to rerouted venous drainage. Non-cavernous AVFs with eye symptoms often reside near the orbit^{3,4}, but some dural AVFs with extensive rerouting of venous drainage may be located remote from the CS⁵⁻⁸.

We report the unusual case of a dural AVF involving the vein of Trolard (VOT) presenting with eye symptoms where the communication to the CS was not immediately obvious from the diagnostic angiogram. Slight venous reflux into the CS and SOV was only noted after careful inspection of the digital subtraction angiography (DSA).

Case report

Clinical Presentation

A 61-year-old male presented with increasing lid swelling, proptosis, epiphora, and redness of the right eye for 1 month. On eye examination, the best-corrected visual acuity was 20/25 OD and 20/50 OS, and the intraocular pressure was 14 mmHg OU. Slit lamp and fundus examination of the right eye showed dilated and tortuous conjunctival, episcleral, and retinal vessels (**Fig. 1A-B**).

Diagnostic Workup

MRI revealed a right-sided proptosis, enhancement of the proximal SOV, and enlarged parietal cortical veins over the right hemisphere but failed to demonstrate an arteriovenous shunt involving the CS (**Fig. 2A-B**). Diagnostic DSA showed no evidence for a CSF but revealed a dural AVF between the right middle meningeal artery (MMA) and VOT with direct shunting and two venous ectasias (**Fig. 3A-B**). The main drainage was directed towards the right transverse-sigmoid sinus (TSS) via the vein of Labbé (VOL), and from here towards the jugular bulb. The proximal internal jugular vein appeared partially occluded and some of the venous drainage refluxed into the torcular region, superior sagittal sinus and left TSS. The

fistula also drained into several smaller cortical veins one of which coursed towards the middle cranial fossa and sphenoid wing as the superficial middle cerebral vein (SMCV). Only very late venous phase images eventually revealed faint opacification of the right CS and SOV (**Fig. 3C-D**).

Endovascular treatment

Endovascular treatment by transarterial embolization was performed under general anesthesia using a transfemoral access. A triaxial catheter system with a 6-Fr Neuron[™] MAX guiding catheter (Penumbra, Alameda, CA, USA), a 5-Fr Sofia[®] 55 distal access catheter (MicroVention, Tustin, CA, USA), and a 1.3-Fr Headway[®] Duo catheter (MicroVention) was navigated into the right external carotid artery and distal MMA (**Fig. 4A**). Here, a total of 2.55cc of PHIL[™] 25% (MicroVention) was injected (**Fig. 4B**) until cessation of the AV shunting was observed. Control runs showed complete occlusion of the arteriovenous shunt (**Fig. 4C**) and reversal of venous flow was noted in the right SMCV (**Fig. 4D**). The patient woke up with no neurological deficits. A daily dose of low-molecular heparin was administered for 1 week to prevent excessive thrombosis.

Postoperative course

The patient had an uneventful postoperative course with clinical improvement within one week. Eye examination one month later was normal. Follow-up DSA after 3 months showed a minor residual arteriovenous shunt between the right MMA and a short portion of the VOT draining through an additional small vein to the TSS, while the proximal VOT and VOL remained occluded. The fistula was successfully retreated by transarterial embolization with PHIL[™]. Follow-up DSA after 15 months showed complete occlusion and no eye symptoms were present (**Fig. 1C-D**).

Discussion

Arteriovenous shunts with direct drainage into dilated cortical veins are aggressive vascular lesions accounting for 13% of intracranial dural AVFs⁹. According to the Cognard classification¹⁰ these dural AVFs are considered type IV fistulas, which often present with progressive neurological symptoms, seizures, and intracranial hemorrhage, requiring early endovascular treatment¹¹.

Dural AVFs can induce profound hemodynamic changes to the venous system due to increased venous pressure and therefore focal symptomatology relates to the venous territory

of a dural AVF rather than the location of the nidus and arterial supply¹². Ocular symptoms are seen in 92% of CSFs⁹ but may as well occur in 4.3-5.7% of non-cavernous fistulas^{2,9}. Ocular symptoms related to non-cavernous dural AVFs are often caused by venous drainage into the CS and SOV increasing the orbital venous pressure². These non-cavernous fistulas mainly develop in the posterior fossa involving the jugular foramen^{2,9,13}, jugular bulb¹³, paracaval veins¹⁴, petrosal sinuses³, and hypoglossal canal¹⁵. The posterior fossa AVFs with ocular involvement drain via the dural sinuses en route to the CS. On the contrary dural AVFs involving the TSS^{2,6,8,9,16}, superior sagittal sinus^{7,9}, and ethmoid⁴ tend to drain via superficial cortical veins, while dural AVFs in the tentorium⁵ and torcular region⁹ may drain via the deep venous system. Cerebral venous sinus thrombosis, hypoplastic sinus segments, and high-flow fistulas can significantly change the venous drainage patterns sometimes directing the venous outflow toward the CS².

We only found a single report by Kobkitsuksakul et al. 2016⁸ describing a dural AVF between the MMA and a cortical vein mimicking a CSF. In this patient, an enlarged SOV was noted on MRI and DSA clearly showed that the fistula drained to the CS via an arterialized SMCV. The connection to the CS in our case was only noted after careful inspection of late venous phase DSA images. This anterior drainage was obscured because only a minor part of the AV shunting drained via the SMCV. Partial thrombosis in the internal jugular vein may have increased the venous pressure in the VOT rerouting the flow towards the CS via the small caliber SMCV. Furthermore, the distal SOV thrombosis blocking the connection to the facial venous system may have further increased the orbital venous pressure creating a low-flow AVF with symptoms of orbital congestion, that was not immediately obvious on the diagnostic DSA.

Conclusion

Symptoms suggesting a CSF may rarely be caused by dural AVFs involving parietal cortical veins and their rerouted venous drainage. The use of MRI for exclusion of a CSF in a patient with eye redness and proptosis is insufficient. Due to its superior resolution, DSA usually allows to identify even small, low flow communications between a remote arteriovenous shunt and the CS. Acquisition of very late venous images may be required to identify “angiographically obscured” AV shunts. Understanding venous anatomy and drainage pattern between cortical veins and the CS system are crucial to correlate vascular pathology and clinical symptoms.

Figure 1. Clinical pictures of the right eye. Eye redness caused by dilated conjunctival veins seen during admission of the patient (**A-B**). Complete resolution of eye symptoms 15 months after endovascular treatment (**C-D**).

Figure 2. Axial T1-weighted MRI shows a venous ectasia (arrow) in the right parietal region (**A**). Axial contrast-enhanced T1-weighted MRI demonstrates right-sided proptosis and enhancement of the right proximal SOV (arrow) (**B**).

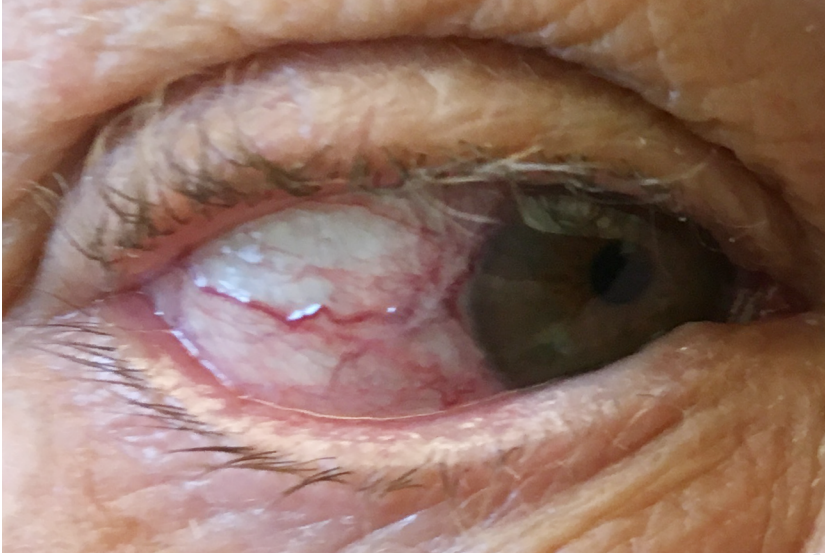
Figure 3. Digital subtraction angiograms with right proximal MMA injections in arterial (**A-B**) and late venous (**C-D**) phases. AP (**A**) and lateral (**B**) views showing a dural AVF (arrow) supplied by the MMA (double arrow) shunting directly into the VOT (arrowhead). The main venous drainage is directed into the TSS via the VOL (double arrowheads). A lesser venous outlet through a small SMCV (triple arrowheads) appears to drain anteriorly towards the CS. Two venous ectasias are present (asterisks). **C:** Magnified AP view with caudal angulation showing a faint but patent communication between the SMCV (arrow), CS (asterisk) and SOV (arrowhead). The anterior segment of the SOV appears thrombosed. **D:** Magnified lateral tilted view showing the SMCV (arrow) draining into the posteriorlateral part of the CS (asterisk) with filling of the SOV (arrowhead) and inferior ophthalmic vein (double arrowhead). The Sofia[®] distal access catheter placed in the right MMA is projected over part of the SMCV.

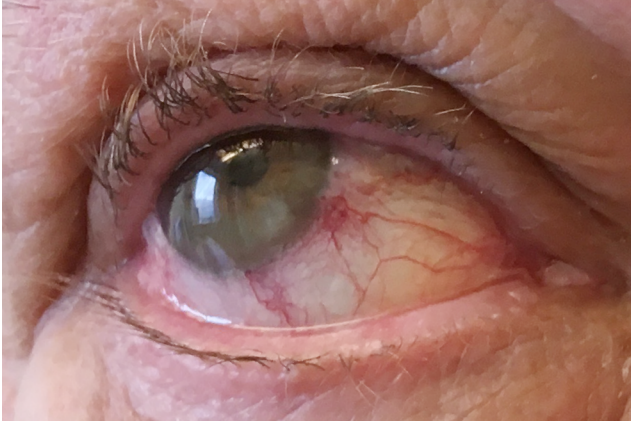
Figure 4. MMA injections in lateral views (**A-C**). The Headway[®] Duo microcatheter in the distal MMA reaching the fistula site (**A**) and the formation of an embolic cast after injection of PHIL 25% (**B**). Complete occlusion of the arteriovenous shunt (**C**). Late venous phase of a right internal carotid injection in lateral tilted view (**D**) showing faint opacification of the SMCV (arrows), CS (asterisk) and SOV (arrowhead) with flow reversal. The venous drainage was directed from the SOV into the CS and SMCV.

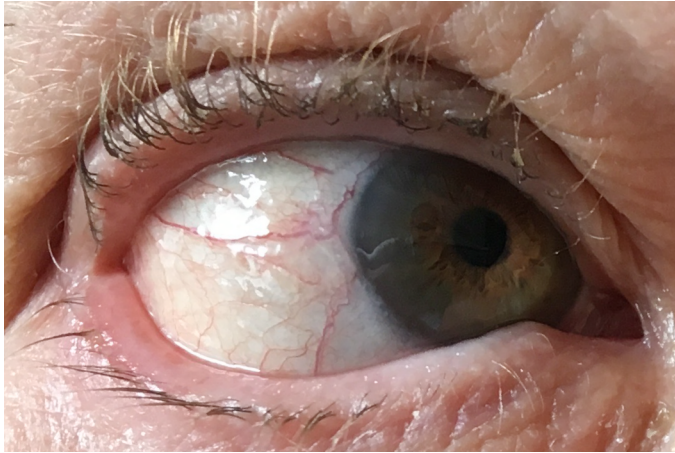
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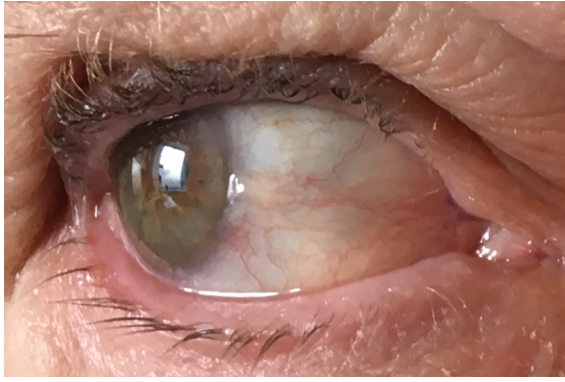
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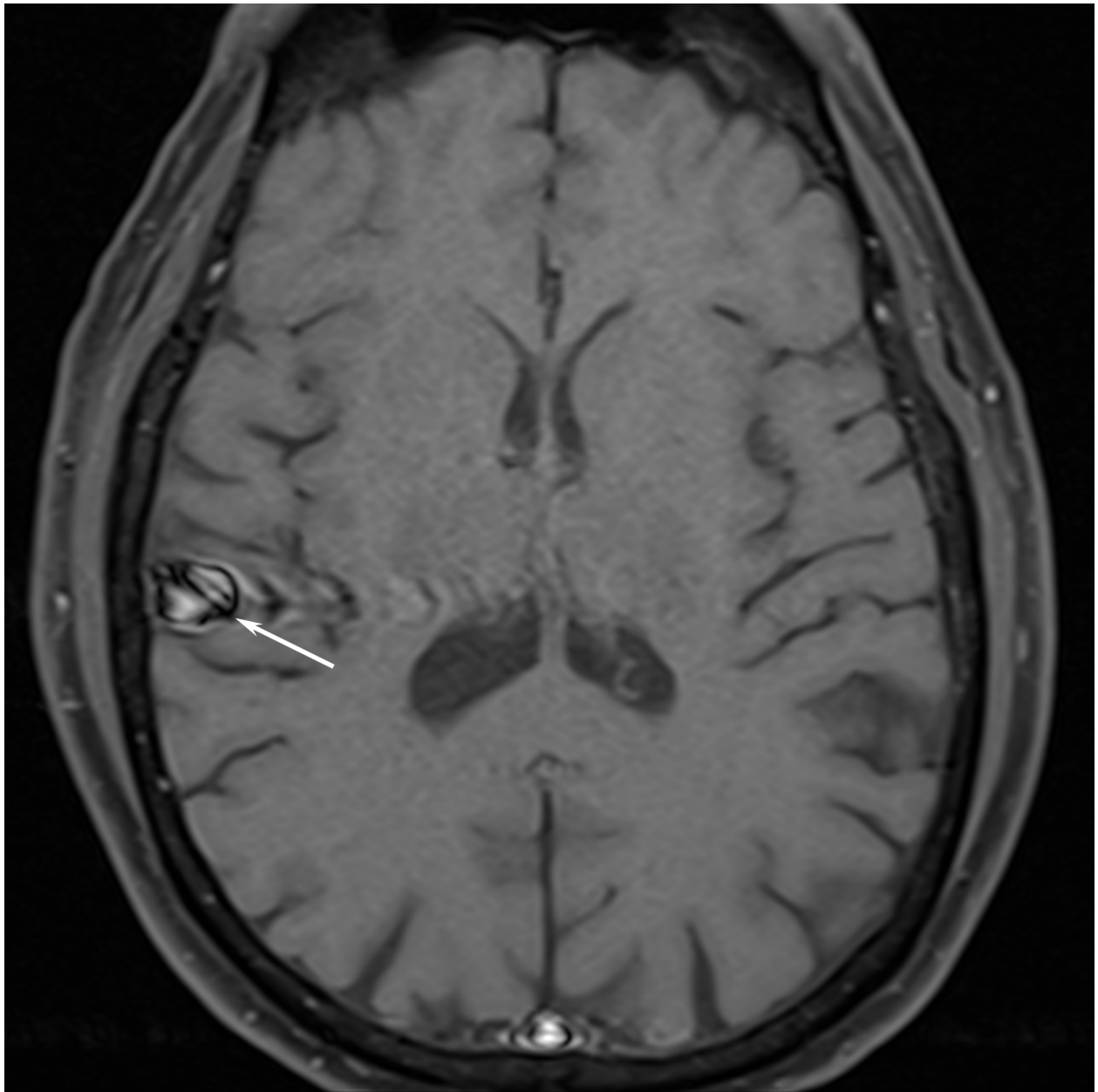


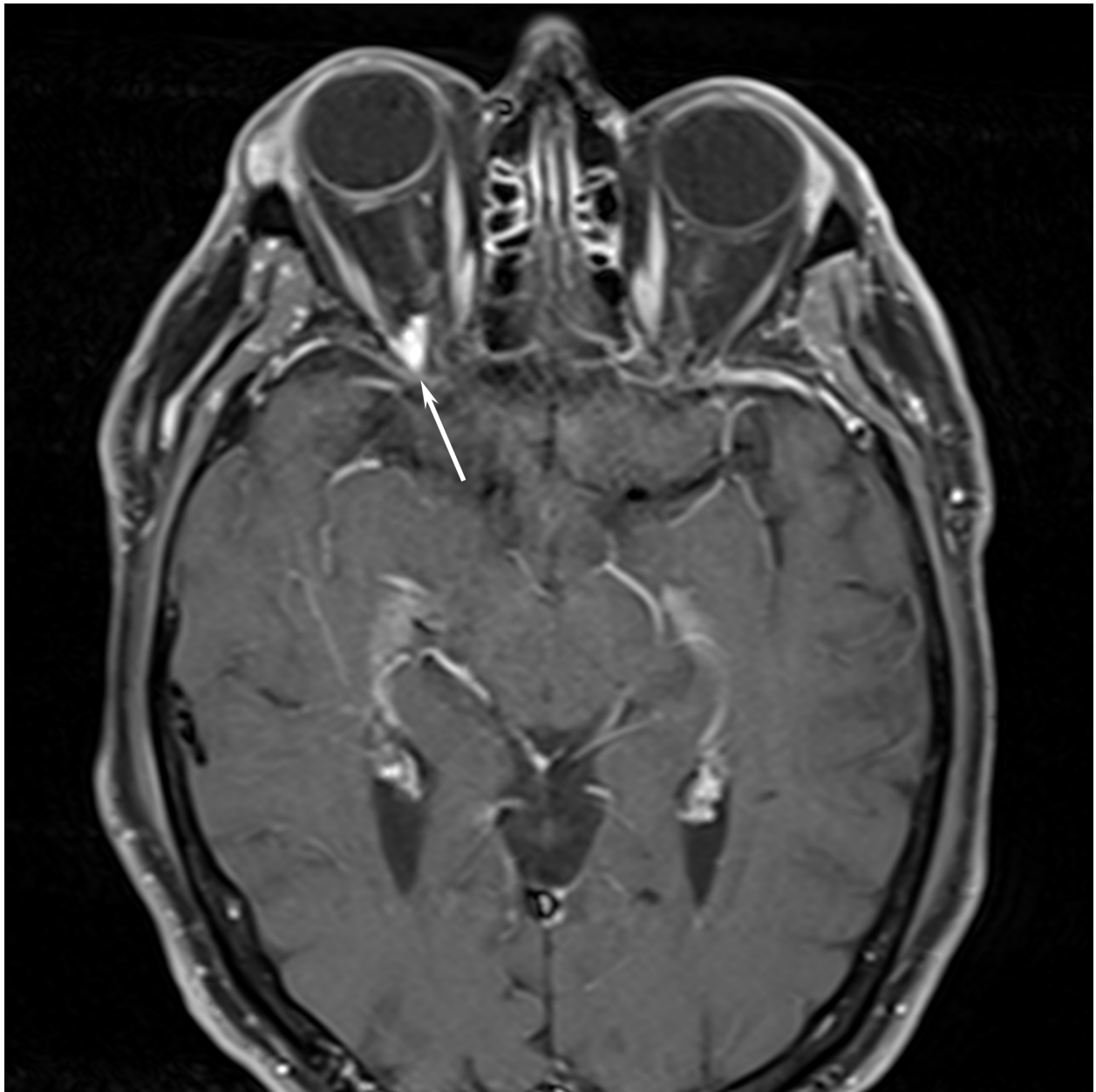


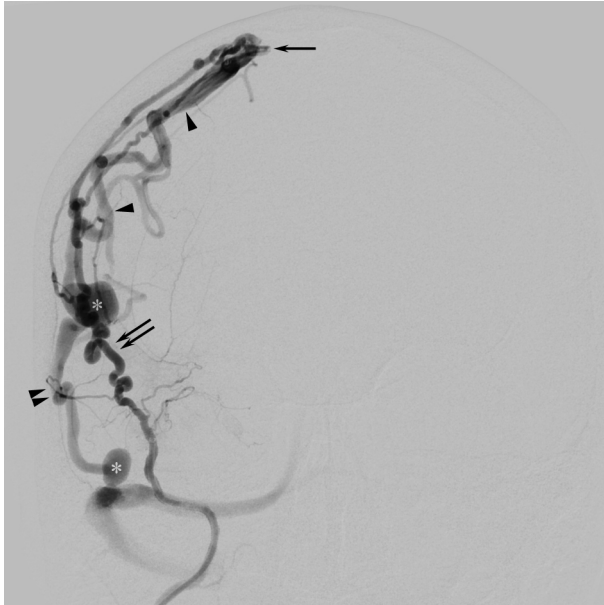


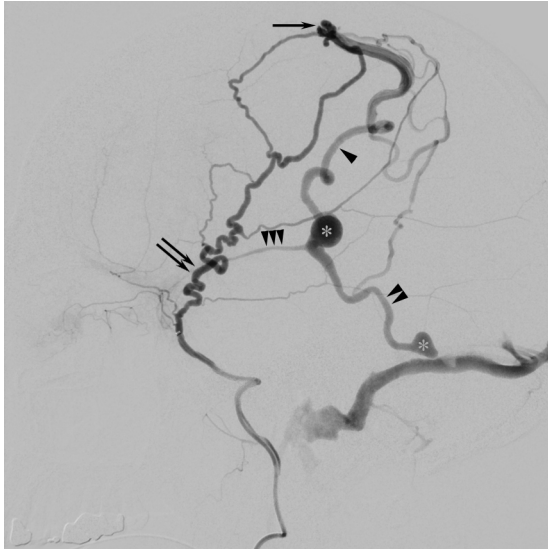


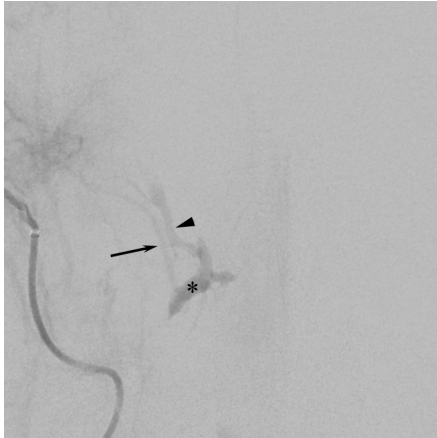
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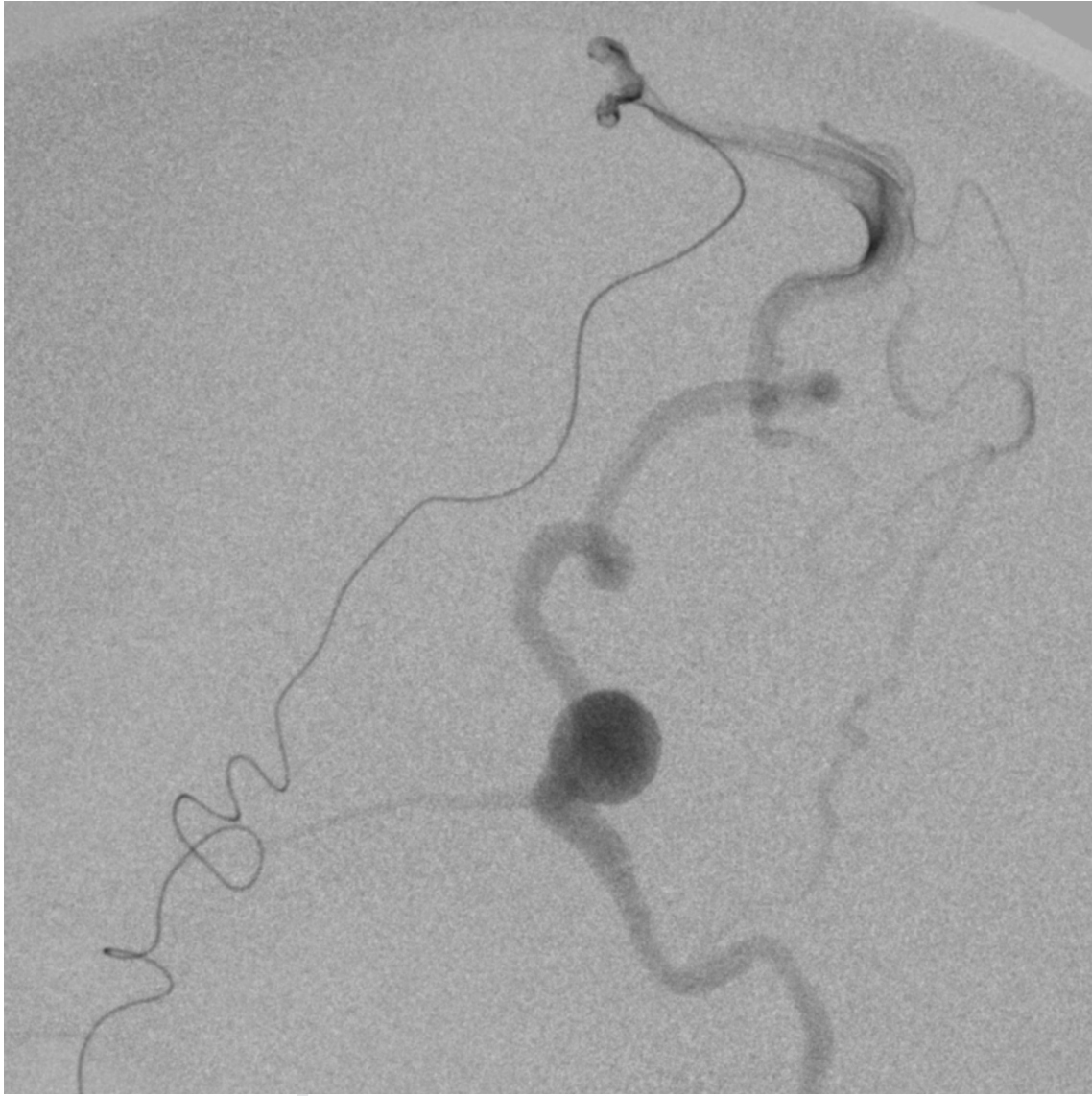


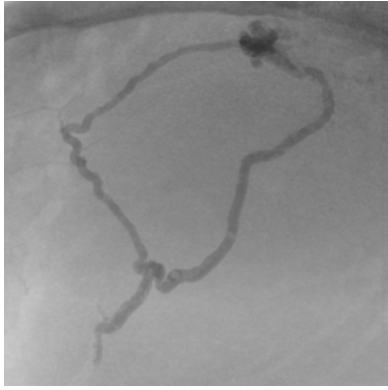




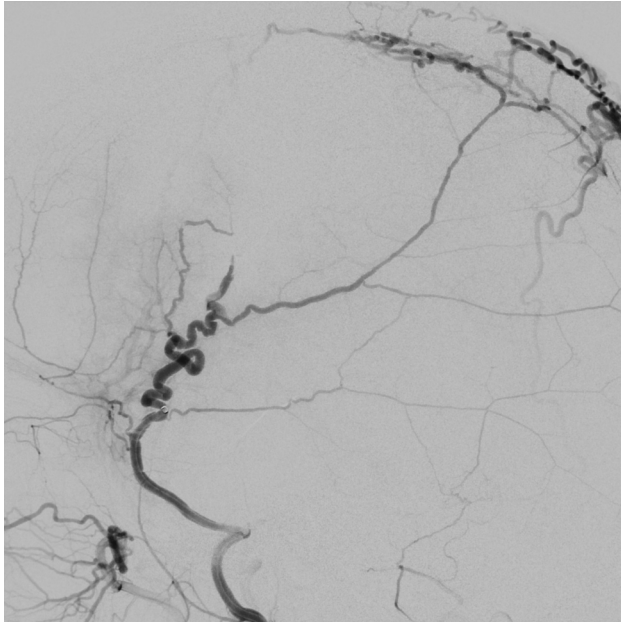








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