



Case Report

Oculomotor neuropathy from an unruptured arteriovenous malformation in the frontal operculum: A case report

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ABSTRACT

Background: Cerebral arteriovenous malformations (AVMs) are vascular lesions with a network of dysplastic vessels between an arterial and a venous tree with no intervening capillary bed. They most commonly present with an acute hemorrhage, seizures, or persistent headaches.

Case Description: The authors report the case of a 62-year-old male who presented with diplopia for 5 days. Magnetic resonance imaging and angiography demonstrated a Spetzler-Martin Grade 2 AVM located in the right frontal operculum with deep drainage into the basal vein of Rosenthal causing ipsilateral oculomotor neuropathy. The patient underwent staged embolizations of the feeding pedicles, which were derived from the internal as well as external carotid circulation. This was followed by a right pterional craniotomy for resection of the AVM. The patient reported complete resolution of the diplopia over 4 weeks with no recurrence at the 6-month follow-up appointment.

Conclusion: AVMs of the brain can present with atypical clinical symptoms that can be caused by the venous drainage pattern not the location. It is important to include vascular imaging studies in the work-up of patients who present with diplopia to rule out an AVM. Early diagnosis and treatment of the AVM can result in complete resolution of the diplopia.

Keywords: Arteriovenous malformation, Diplopia, Embolization, Endovascular, Microsurgery

INTRODUCTION

Cerebral arteriovenous malformations (AVMs) are vascular lesions with a network of dysplastic vessels or nidus between an arterial and a venous tree with no intervening capillary bed. This allows pathologic shunting of arterial blood into the venous vasculature causing venous hypertension and dilatation that can result in cerebral edema and compression of neural structures. Cerebral AVMs occur with an incidence of 1 in 10,000 and usually present in young adults with an acute hemorrhage, seizures, or chronic headache.^[1,2,5] The presentation often depends on the location, size, and configuration of the AVM. We report a case of a Spetzler-Martin Grade 2 AVM located in the right frontal operculum that exclusively presented with an ipsilateral persistent

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oculomotor nerve palsy as a result of nerve compression in the interpeduncular cistern.

CASE REPORT

A 62-year-old male presented to an outpatient ophthalmologist with a 5-day history of progressive binocular diplopia and was referred to our institution. On presentation, neurologic and ophthalmologic examination showed a right pupil-involving partial third nerve palsy with no other abnormal findings. Routine brain magnetic resonance imaging (MRI) and time-of-flight MR angiography (MRA) demonstrated an AVM located in the right frontal operculum extending to the anterior insula with venous drainage into the right basal vein of Rosenthal. There was no subarachnoid or intraparenchymal hemorrhage to suggest AVM rupture. A diagnostic cerebral angiogram was performed next and demonstrated a 1.9 cm × 2.2 cm AVM fed by the right middle and accessory meningeal and sphenopalatine arteries and opercular branches of the middle cerebral artery (MCA). The venous drainage followed a deep pattern into a dilated and tortuous right basal vein of Rosenthal to the vein of Galen and ultimately into the straight sinus [Figure 1]. The patient underwent staged embolizations of the feeding pedicles using Onyx-18 embolic material (Medtronic, Minneapolis, USA) to facilitate planned surgical resection. In the first stage, superselective catheterization of the right

middle meningeal, accessory meningeal, and sphenopalatine arteries was performed using a Marathon microcatheter (Medtronic, Minneapolis, USA) and a Synchro 10 microwire (Stryker, Kalamazoo, USA) [Figure 2]. In the second stage, the next day, the feeding branches of the opercular segment of the MCA were accessed with a Scepter balloon-guided microcatheter (MicroVention Inc., Tustin, USA) and a Synchro 2 microwire [Figure 3]. On the following day, the patient underwent a right pterional craniotomy for resection of the AVM. An intraoperative control angiogram confirmed complete obliteration of the AVM [Figure 3d and e]. The pathologic tissue examination was consistent with AVM.

At the 4-week follow-up, the patient reported complete resolution of the diplopia. Neurologic examination demonstrated no deficits and isocoric pupils with intact extraocular movements bilaterally. There was no recurrence of the diplopia 6 months after surgery.

DISCUSSION

Most AVMs are diagnosed by the fourth decade of life and usually present with an acute hemorrhage from rupture (40%–50%), seizures (30%), or headaches.^[1,2] The present case is unusual due to the patient's age and manifestation with a cranial mononeuropathy caused by venous compression. Recognizing that the patient's third nerve palsy involved that the pupil was important to raise suspicion for an underlying

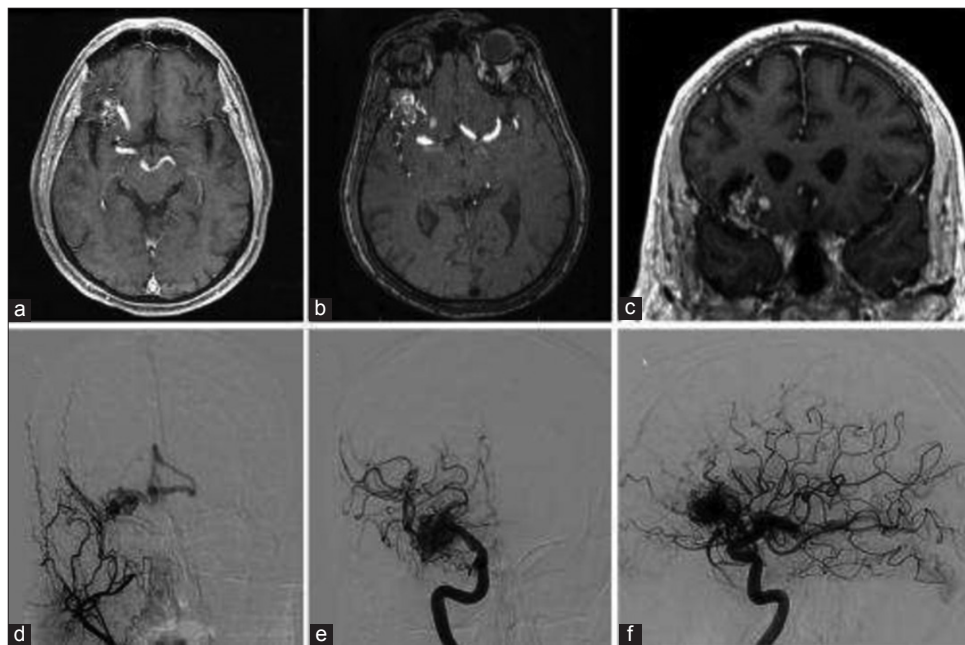


Figure 1: (a) Axial T1-weighted magnetic resonance imaging (MRI) with gadolinium and (b) MR angiography show the arteriovenous malformations (AVM) located in the right frontal operculum and anterior insula with prominent venous drainage in the interpeduncular cistern. (c) Coronal T1 MRI shows medial extension of the AVM. (d) Anteroposterior (AP) angiogram of the right common carotid shows the AVM and its venous drainage into a tortuous basal vein of Rosenthal and vein of Galen. (e) AP of the right internal carotid (ICA) shows the AVM. (f) Lateral projection of the right ICA angiogram shows filling from middle cerebral artery branches.

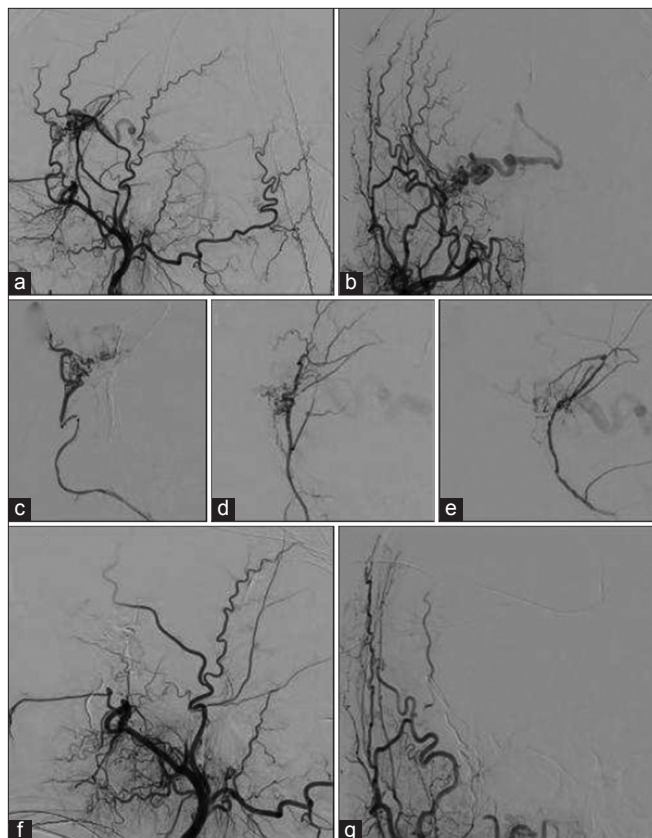


Figure 2: Lateral (a) and anteroposterior (AP) (b) right external carotid angiogram showing filling of the arteriovenous malformations (AVM) through the middle meningeal (MMA), accessory meningeal (AMA), and sphenopalatine (SPA) arteries. Superselective catheterization of the SPA (c), AMA (d), MMA (e), just before Onyx-18 embolization. Lateral (f) and AP (g) projections of the external carotid artery post-embolization with Onyx-18 not showing any filling of the AVM from the external circulation.

compressive lesion and, therefore, initiating the work-up with an MRI and MRA, which quickly led to the correct diagnosis.

Central to this patient's presentation is the high-flow shunting of blood across the nidus into tributaries of the basal vein of Rosenthal causing venous hypertension with increased tortuosity and dilatation of the vessel. The combination of direct mechanical compression and the transmission of pulsation onto the ipsilateral oculomotor nerve in the interpeduncular and crural cisterns was eventually responsible for the diplopia. A similar underlying mechanism is occasionally encountered in AVM-related trigeminal neuralgia. In such cases, AVMs located in the posterior fossa cause dilatation arteries and cause compression of the trigeminal nerve. Mori *et al.* reported a case of trigeminal neuralgia secondary to a vermian AVM that was successfully treated with endovascular embolization and radiosurgery.^[3]

Our treatment strategy for this patient's AVM consisted of embolization followed by surgical resection. We

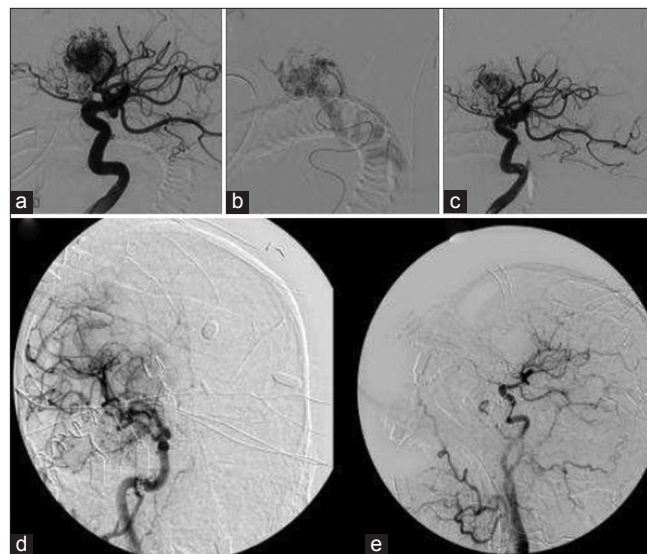


Figure 3: (a) Lateral right internal carotid (ICA) angiogram in early to midarterial phase showing filling of the nidus from opercular middle cerebral artery branches. (b) Superselective catheterization of opercular branches in preparation for injection of Onyx-18. (c) Lateral ICA projection after partial embolization of the arteriovenous malformations (AVM). Intraoperative AP (D) and lateral (E) common carotid angiogram showing complete resection of the AVM. The anterior cerebral circulation is not seen in this injection since it fills from the contralateral side.

performed embolization in multiple stages since our institutional experience has shown that this approach reduces periprocedural complications such as ischemia and microcatheter trapping. This practice is also preferred by Mounayer *et al.* in their series of AVM embolization using Onyx.^[4] Even though the AVM could not be cured embolization alone, it made microsurgical resection safer and more efficacious. In addition to reducing blood flow from surgically inaccessible feeders, the embolisate also helped define a dissection plane and reduced the duration of surgery.

CONCLUSION

AVMs of the brain can present with atypical clinical symptoms and patient demographics. Even though uncommon, the presented case demonstrates that AVMs may cause compression of the oculomotor nerve producing isolated diplopia. It is important to include comprehensive vascular imaging studies in the work-up of patients who present with diplopia to rule out an AVM. Our experience shows that early diagnosis and treatment of the AVM can result in complete resolution of the diplopia.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his

consent for his images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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