

## CASE REPORT

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# A Case Series of Ethmoidal Dural Arteriovenous Fistulas Treated by Endovascular Embolization with Onyx

Hoang Duc Ha<sup>1,2</sup>, Nguyen Minh Duc<sup>3</sup>, Januel Anne Christine<sup>4</sup>, Cognard Christophe<sup>4</sup>**ABSTRACT:****Introduction:** Ethmoidal dural arteriovenous fistulas (eDAVFs) are an unusual type of intracranial vascular lesion that commonly present with acute haemorrhage. **Aim:** We presented 3 cases of ethmoidal DAVFs treated effectively by endovascular embolization with Onyx.**Results:** 3 patients hospitalized by reasons of stroke (2 cases) and vision impairment of the right eye (1 case). The digital subtraction angiography (DSA) showed the fistulas supplied by ethmoidal branches of the ophthalmic arteries (3 cases) and an additional supply from the ethmoidal branches of the internal maxillary artery (1 case). The cortical venous drainage was dilated and flowed to the superior sagittal sinus. The superselective microcatheterization through the ophthalmic artery allowed injection of Onyx sufficient plug around the distal end of the catheter without any complications. **Conclusion:** eDAVFs with neurologic complications could be treated effectively by endovascular embolization with Onyx.**Keywords:** Ethmoidal dural arteriovenous fistulas, endovascular embolization, Onyx.**1. INTRODUCTION**

The dural arteriovenous fistulas (DAVFs) are abnormal arterio-venous shunts within the dura. This abnormality accounts for 10%–15% of all intracranial arteriovenous malformations (1). Based on the direction of dural sinus drainage, the presence or absence of cortical venous drainage, and venous outflow architecture, Cognard classified five types of DAVFs including: type I, drainage vein located in the main sinus with antegrade flow; type II, lesions drain with retrograde flow in dural sinus (IIa), directly into cortical veins (IIb), or both (IIa+b); type III, direct cortical venous drainage without venous ectasia; type IV, DAVFs with direct cortical venous drainage with venous ectasia; and type V, lesions in spinal stage (2). Ethmoidal dural arteriovenous fistulas (eDAVFs), rare intracranial lesions, account for approximately 2-3% of dural arteriovenous fistulas (1), are fed by ethmoid branches of the ophthalmic artery (3, 4, 5) and drained to dilated cortical vein. These lesions have a high risk of intracerebral hemorrhage and other aggressive neurologic complications (2, 6). Therefore, DAVFs need be completely and permanently embolized.

**2. AIM**

We performed a report of 3 cases in order to help interventional neuroradiologists know about an effective method to treat the eDAVFs with a new endovascular embolization material—Onyx (ev3, Irvine, CA, USA).

**3. CASE PRESENTATION**

This study consisted of 3 patients after clinical diagnosis and radiographic diagnosis (CT angiography and/or MRI angiography) and digital subtraction angiography (DSA) confirmed eDAVFs. All these patients presented type IV following the classification of Cognard (lesions with direct cortical venous drainage with venous ectasia) (2). These patients were treated by endovascular embolization at the Department of Interventional and Diagnostic Neuroradiology, Toulouse University Hospital, Toulouse, France.

**4. DISCUSSION**

In this case series, eDAVFs were all fed by ethmoidal branches of bilateral ophthalmic arteries with one side superiority and also by the middle meningeal artery (a branch of internal maxillary artery of external carotid artery) (3, 5). In all cases, the drainage vein flows into the frontal

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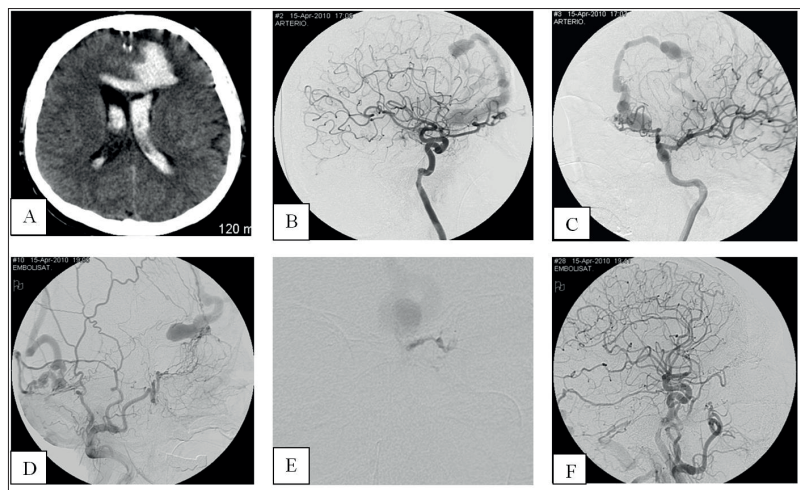
| Patient Sex/<br>Age | Clinical signs               | CT or/and MRI   | DSA   |   | Type | Embolization        |                    | Complication | Post-treatment recurrence                                |
|---------------------|------------------------------|---|---|---|------|---------------------|--------------------|--------------|--|
|                     |                              |   | Feeding artery  | Draining vein   |      | Embolized materials | Result             |              |  |
| #1: Female/73       | Stroke                       | Right frontal lobe hemorrhage   | Bilateral ophthalmic artery with right superiority  | Dilated cortical vein into superior sagittal sinus  | IV   | 0.9 ml Onyx         | Complete occlusion | No           | No   |
| #2: Male/60         | Stroke                       | Hemorrhage in the left frontal lobe, the genu of corpus callosum and ventricles | Bilateral artery with left superiority, and anterior left middle meningeal artery               | First dilated cortical vein into superior sagittal sinus; Second dilated cortical vein into frontal cortical vein and the same side vein of Labbé | IV   | 1.15 ml Onyx        | Complete occlusion | No           | No   |
| #3: Female/68       | Vision loss of the right eye | Some enlarged and tortuous vessels suspecting the DAVFs                         | Bilateral artery with right superiority and meningeal branches of right external carotid artery | Enlarged frontal cortical vein terminating by a narrow segment before draining to superior sagittal sinus   | IV   | 0.8 ml Onyx         | Complete occlusion | No           | No, but vision impairment of the right eye still existed |

**Table 1.** The detailed reports of 3 patients with eDAVFs

cortical vein and then into the superior sagittal sinus. The cerebral cortical veins are located in each side of falx cerebri, as a result, the drainage vein is usually in one side (3, 4). The enlarged venous drainage (type IV) with high hemodynamics had a high risk of cerebral hemorrhage. Therefore, these lesions must be treated as soon as possible. Many interventional methods have been applied, such as laser surgery, craniotomy surgery or endovascular embolization with artery or venous approach or a combination of multiple techniques (7).

Some reports presented the role of gamma knife radiosurgery (GKRS) in treatment of DAVFs. The study of Park KS et al., was implemented on 31 patients with DAVFs type IIb and III, presented the efficacy of GKRS with 90% cured patients and only one patient with a transient complication (8). However, these authors suggested a multicenter, prospective study was necessary to generalize their treatment algorithm. Yang HC et al. also reported that GKRS could be effective for DAVFs type II and III with only some sequelae such as intolerable headache, bruit, or ocular symptoms, but sometimes yielded severe complications (9). GKRS is not always effective for DAVFs because these lesions often do not have a nidus to target the ray as the arteriovenous malformation (AVM) (3). On the other hand, the time from beginning of therapy to the end of treatment process may be very long and so during this time the lesions often have a high risk of bleeding (3). As a result, this treatment method is considered only for mild cases and lesions with a favorable location.

The embolization technique with venous approach is also very difficult as the zigzag dilated drainage veins, which are very fragile, with several narrowed or obstruct-



**Figure 1.** A 60-year-old male patient with CT image presenting the hemorrhage in the left frontal lobe, genu corpus callosum and ventricle (A). DSA before treatment presented the eDAVFs fed by the left internal carotid artery (B), by the right internal carotid artery (C) and by the left external carotid artery (D). Superselective catheterization in the ethmoidal arteries of the left ophthalmic artery (E). Re-examination after 3 months (F).

ed segments before flowing into the superior sagittal sinus. The literature also presented several perforations of the veins during catheterization resulting intracerebral hemorrhages (10, 11). Another embolization technique has also been reported that a trans-arterial endovascular approach using diluted N-butyl cyanoacrylate glue was applied (12-14). The microcatheter was placed into at the distal segment of ophthalmic artery. The purpose of this technique was to embolize the proximal segment of drainage vein. However, the injection process of N-butyl cyanoacrylate glue, could be very unsafe and the use of glue agents, requires a good experience of interventional physicians. In the case, the glue agents were injected in the proximal segment, the drainage vein could not be blocked, then DAVFs would be revascularized. On the other hand, the glue agents were injected into the more distal segment. These agents could be difficultly controlled and caused cerebral infarction (7, 14, 15).

Onyx solution mixture (MTI-ev3) is a new non-adhesive material containing a mixture of copolymer, ethylene and dimethyl sulfoxide (DMSO). The advantage of this non-adhesive agent is to minimize the risk of adhesion around the distal microcatheter extremity. This help us to slowly inject and well control the volume of embolism agent. With safe reflux and proper penetration of Onyx, this method was not necessary to multiple embolizations. This also help avoiding the serious complications of material refluxing into central retinal artery (15, 16). In a recent study, Moenninghoff C et al. reported that the transarterial embolization with Onyx has established as the first-line treatment for cranial DAVFs. This technique had high cure rates and low rates of complications and recurrences (17). For many reasons, we had chosen the trans-arterial embolization technique by using the superselective microcatheter into the ophthalmic artery and injected a sufficient amount of Onyx into the proximal drainage vein without any complications during or after treatment in all 3 cases. The digital subtraction angiography (DSA), was performed after 3 months for these patients, confirmed that DAVFs was completely treated. In 2 cases (No #1 and No #2), there were no specific clinical symptoms, but the case No #3 still existed the vision impairment as before hospitalization. This showed the superior advantage of Onyx in embolization treatment.

## 5. CONCLUSION

DAVFs are an unusual type of intracranial vascular lesion with a very high risk of cerebral hemorrhage and confirmed by DSA technique. Up to now, many treatment methods of this disease have been reported but trans-arterial endovascular embolization with Onyx is the best method in DAVFs treatment, that has an efficient therapeutic effect and severe complications were not observed.

- **Ethical approval and Declaration of patient consent:** Owing to the case series design, the institutional review board approval was waived. Inform consent of patients was obtained.
- **Author's contribution:** Hoang Duc Ha and Nguyen Minh Duc contributed equally to this article. Hoang Duc Ha and Nguyen Minh Duc prepared, drafted, and revised manuscript critically for important intellectual content. Each author gave final approval of the version to be published and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.
- **Conflicts of interest:** There are no conflicts of interest to declare.
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