Case report

Computed tomographic and digital subtraction angiography evaluation of ophthalmic-ethmoidal artery dural arteriovenous fistula

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ABSTRACT

Ophthalmic-ethmoidal dural arteriovenous fistula (DAVFs) is a rare type of dural arteriovenous fistulas and usually presenting with spontaneous subarachnoid hemorrhage, subdural hemorrhage or ocular symptoms. We present a case of a 59-year-old gentleman presenting with acute headache, vomiting and generalized weakness. CT study of the brain revealed a large left frontal hematoma and abnormal aneurysmal sac with dilated cortical vein, communicating with the superior sagittal sinus. Conventional angiography confirmed diagnosis of ruptured ophthalmic-ethmoidal DAVF, resulting in a frontal intra-axial hemorrhage. Anterior fossa DAVFs are extremely rare, difficult to diagnose and treat. CT angiography is initial method of diagnosis, but digital subtraction angiography remains the gold standard of confirming dural fistulas.

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Introduction

Dural arteriovenous fistulas (DAVFs) are abnormal arteriovenous shunts located within the dura and accounts for up to 15% of all intracranial vascular malformations among all populations [1]. Across all types of DAVFs ethmoidal dural arteriovenous fistulas are considered very rare and seen in only 2%-3% of dural arteriovenous fistulas [1]. It usually receives its arterial supply by ethmoidal branches of ophthalmic artery and drains to cortical veins. These lesions are prone to rupture and result in intracerebral hemorrhage and cranial nerve palsies [2]. The general approach to treat symptomatic DAVFs is by endovascular permanent embolization or coiling, as well as through surgical clipping, which was performed in our case [1].

Case presentation

A 59-years-old man presented to emergency with history of vomiting, headache and generalized weakness for 4 days. It was associated with few episodes of imbalance and fall on day of presentation. He denied any history of loss of consciousness or seizures. He is a known case of hypertension and ischemic
Fig. 1 – CT of the brain. Non-enhanced images in axial (A) and sagittal (B) cuts showing a large intra-axial dense left frontal hematoma measuring 5.8 cm AP x 3.1 cm TS x 4.9 cm CC (volume of 44.0 mL). The hematoma was extending to the lateral (seen in A), third and fourth ventricles (not included). Contrast-enhanced axial (C) and sagittal images demonstrated a well-defined non-thrombosed aneurysmal sac measuring 1.2 cm in diameter corresponding to the large intra-axial hematoma (arrowhead in C) with a prominent cortical vein communicating the anterior part of the aneurysm to the enhancing superior sagittal sinus on arterial phase (white arrow in D). Another vessel was communicating the anterior surface of the aneurysm to the left olfactory fossa (open arrow in E).

heart disease and has history of cardiac stenting 2 years ago and currently on dual antiplatelets treatment.

On physical examination patient was conscious. Blood pressure was 147/77. Otherwise, he was vitally stable and afebrile. Neurological examination showed intact cranial nerves, intact sensory and muscle power, as well as regular gait.

Basic laboratory hematological findings were within normal limits. An emergency CT study of the brain was performed at time of presentation and revealed a left frontal hematoma
Fig. 2 – DSA of the left carotid artery done through right femoral artery percutaneous access in lateral projection. DSA demonstrating AV fistula in the left anterior cranial fossa with arterial supply from distal branches of bilateral ophthalmic arteries (A), with venous drainage to the superior sagittal sinus (B, C).

with intra-ventricular extension into the third and fourth ventricles and subdural hematoma over the left frontal, temporal, and parietal convexities. CT angiogram was done immediately, which demonstrated a contrast filled aneurysmal sac, likely a venous sac, at the left frontal inferior gyrus with prominent dilated cortical vein communicating to the anterior part of the superior sagittal sinus. A vessel was also seen communicating to the anterior surface of the aneurysm, more to the left olfactory fossa. No other definite feeders were seen (Fig. 1).

Bilateral internal and external carotid angiography confirmed the diagnosis of dural arteriovenous fistula and allowed a better visualization of abnormal vessels and surgical planning for arteriovenous (AV) fistula. The digital subtraction angiography (DSA) showed an AV fistula (Borden type III) in the left anterior cranial fossa with arterial supply from distal branches of bilateral ophthalmic arteries – ethmoidal branches and an ethmoidal branch from left external carotid artery. The venous drainage was found to be a tortuous cortical vein, which drains ultimately to the superior sagittal sinus (Fig. 2).

After multidisciplinary team discussion that included diagnostic, interventional radiology and neurosurgery, the patient was admitted under the care of the neurosurgery team and underwent surgical intervention to evacuate the intracerebral hemorrhage and ligate the fistula’s feeding vessels with permanent clips and the arterialized vein was cauterized, excised and sent for further histopathological evaluation.

Patient reported improvement in general status and relief of the initial symptoms. Patient remained under observation for few days and was later discharged in overall stable condition. He was scheduled for follow up CT in few weeks and neurosurgery outpatient follow up.

Discussion

Dural arteriovenous fistulas are fistulas that share arteriovenous shunts from dural vessels.

Reported incidence of all DAVFs is around 10%-15% among all populations. Usually they are asymptomatic, unless rup-
tured and present with intracranial or extracranial bleed [1]. Other symptoms may include seizures, tinnitus and orbital symptoms (in case of carotico-cavernous fistula, raised intracranial pressure and focal neurological deficits) [2]. The most known causes of DAVFs are previous episodes of dural sinus thrombosis, trauma, tumors or post-surgery conditions [3].

We should understand terms “aggressive” and “non-aggressive” dural fistulas. Factors predisposing to an aggressive outcome include: cortical venous drainage, galenic drainage (deep veins), variceal or aneurysmal venous dilatations. Non-aggressive fistulas are the ones with lack of cortical venous drainage [4].

There are 2 classification systems: Cognard and Borden. The Cognard classification of DAVFs correlates venous drainage patterns and direction of flow with aggressive neurological clinical course and estimates the risk of intracerebral hemorrhage. Another classification system is the Borden classification. It is a simpler (3 grades) and only takes into account the site of fistula and presence or absence of cortical venous drainage. It does not assess direction of flow or presence of venous ectasia. It has been suggested that it does not reflect the differences of hemorrhage rate, as included in Cognard classification [5,6].

Ethmoidal-ophthalmic fistulas are rare entity and receiving blood supply from ethmoidal arteries (branches of ophthalmic artery) with reported incidence of 5%. The commonest site of drainage is into superior sagittal sinus through intracranial cortical veins, as was shown in our case [4,7], (Fig. 2A).

Management of anterior fossa DAVFs is challenging due to adjacent anatomy, small ophthalmic vessels and high risk of embolization of central retinal artery, which may lead to visual loss. Another risk of treatment of dural AVF is a high chance of embolic agents refluxing into the ICA and central circulation, as ethmoidal arteries are originating from ICA circulation.

**Conclusion**

We have described an extremely rare case of a ruptured ethmoidal dural AVF, which was draining through inferior and superior ophthalmic arteries to the superior sagittal sinus and receive feeding from ethmoidal artery. CT brain with angiography is an initial method of choice in diagnosing DAVFs, however DSA remains the gold standard of visualization of dural fistulas.

**Patient consent**

A written consent was obtained from the patient for publication of this case and any accompanying images.

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