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Case Series

# Dural Arteriovenous Fistula with Unusual Drainage Route Beneath Vein of Foramen Caecum

**Pierluigi La Zazzera[1]*, Riccardo Russo[1], Umberto Gava[1] and Mauro Bergui[1]**

[1] *Department of Neuroradiology, University of Turin, Torino, Italy.*

***Corresponding Author:** Pierluigi La Zazzera, Department of Neuroradiology, University of Turin, Torino, Italy.*

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## Abstract

The existence of the vein of the foramen caecum (VFC) in humans is still controversial and its existence in humans is questioned by various authors. Vein of foramen caecum can be clinically important, since it is a potential pathway for intracranial spread of nasal infectious and tumoral processes. Due to small size, VFC may not be visible on normal angiograms. Arterio-venous shunts frequently lead to vein enlargement and finally better visibility. We report two cases of cranial Dural Arterio-Venous Fistula (DAVF) of the anterior cranial fossa, draining through an enlarged VFC. This demonstrates the role of VFC as a connection between vascular territories of ophthalmic and Sphenopalatine arteries; to our knowledge, a DAVF in this location has never been described: since occlusion of the primary draining vein is the target of the treatment, a correct identification is mandatory.

***Keywords:** Dural Arterio-Venous Fistula (DAVF); Vein; Creutzfeldt–Jakob disease; Foramen Caecum*

## Introduction

In classical anatomical books like Gray's anatomy, a vein coursing through foramen cecum, and draining nasal mucosa, is described [2]. This vein is supposed to explain spreading of infections from nasal cavities into the brain.

Foramen caecum is an embryologic dural projection of the anterior cranial fossa that regresses after birth and completely closes during early childhood. It is located between the crista galli and the crest of the frontal bone. Embryologic origin of foramen caecum occurs during anterior neuropore formation. Failure of the regression of the dural protrusion through the foramen cecum may lead to the development of various malformations, including nasal glioma and encephalocele [5].

Pathological studies showed contradictory results in neonates and during intrauterine life: Theile [3] believed that the VFC was present in neonates, although Padget (1957) found no traces of the vein of the foramen caecum in embryos up to 3 months, even if other emissary veins of the cranial cavity develop before 2.5 months. [9]

Post mortem studies in adults, demonstrated that foramen cecum was occluded by fibrous tissue in the vast majority of subjects [1][7].

VFC may be visible in various radiological investigations: Tutar et al. [12]demonstrated on a Computed Tomography (CT) and contrast-enhanced MRI the presence of the VFC passing through a patent foramen cecum. Using magnetic resonance imaging, Tsutsumi et al. have found tubular-shaped venous extensions lying in the Foramen Cecum (FC) arising from the rostral end of the falx cerebri [11]. Lewińska-Śmialek B et al, analysing 18 cranial computer tomography images, concluded that the foramen caecum was present in all investigated skulls, although with different sizes [8]. San Millan Ruiz D et al [10] performed angiography in two patient documenting intracranial drainage pathway for the septal nasal mucosa through the foramen cecum.

We describe two Patients with dural arterio-venous fistula of planum Ethmoidale, with primary venous drainage in the VFC.

The Institutional Review Board approved the study and waived informed consent based on the retrospective nature of the study.

## Case Presentation

**CASE 1**

An MRI on a 74 years old man with dizziness showed high flow, pathological vessels, in the anterior fossa. A DSA showed a Type 4 DAVF, fed by the bilateral anterior meningeal arteries and branches of the Sphenopalatine arteries. Ophthalmic artery involvement, frequently seen in such location, was not found.

Arteries converged on a venous structure located on the midline between the Crista Galli and the crest of the frontal bone, which was identified as the VFC. The vein drained subsequently in a tortuous pial venous channel finally joining the superior sagittal sinus (SSS). (Fig 1)

Due to inherent risk of hemorrhage of the DAVF, the patient was successfully operated.

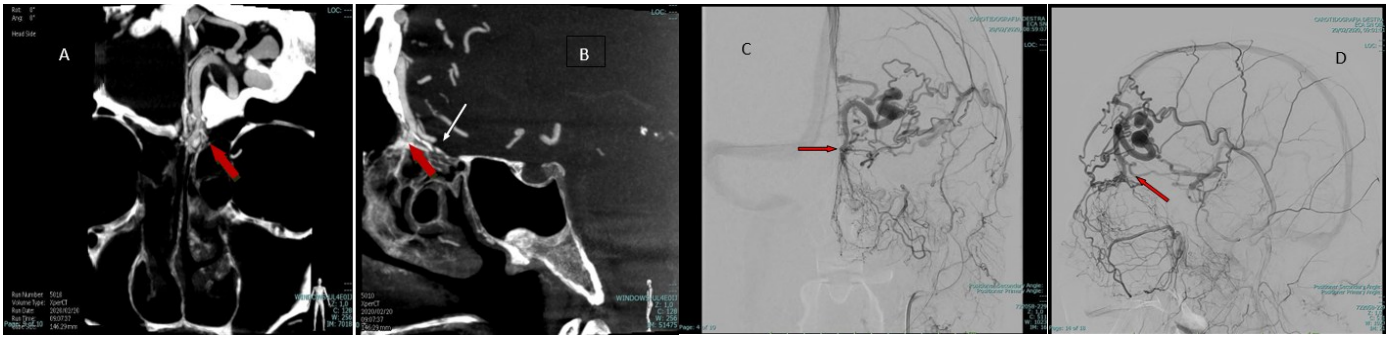


***Figure 1.*** *Patient 1: CT (A) sagittal slice; (B) coronal slice; DSA, arterial phase left ECA, (c) postero-anterior view Figure 1. (D) lateral view; Arterio-venous fistula of the anterior cranial fossa fed by the anterior meningeal arteries and branches of the Sphenopalatines and Infraorbitarian arteries drained towards the nasal cavity through the foramen caecum. Red arrows show enlarged VFC; white arrow shows Ethmoidal plane.*

**CASE 2**

A 58 years old woman was submitted to an MRI after an episode of transient amnesia. Because of the evidence of anomalous vascular structures in the anterior fossa, a DSA was performed. A type 4 DAVF of the anterior cranial fossa was found, with multiple arterial feeders, including Ophthalmic, Sphenopalatine and Meningeal branches of the external carotid arteries. (Fig 2)

Venous drainage involved primarily a venous structure located in front of the Cribriform plate of the Ethmoid bone and behind the frontal bone. This venous channel was identified as VFC. Subsequent drainage involved a pial vein with final destination to SSS. Endovascular treatment was performed, using coil and DMSO-based liquid embolic, to occlude the fistula point from the venous side.

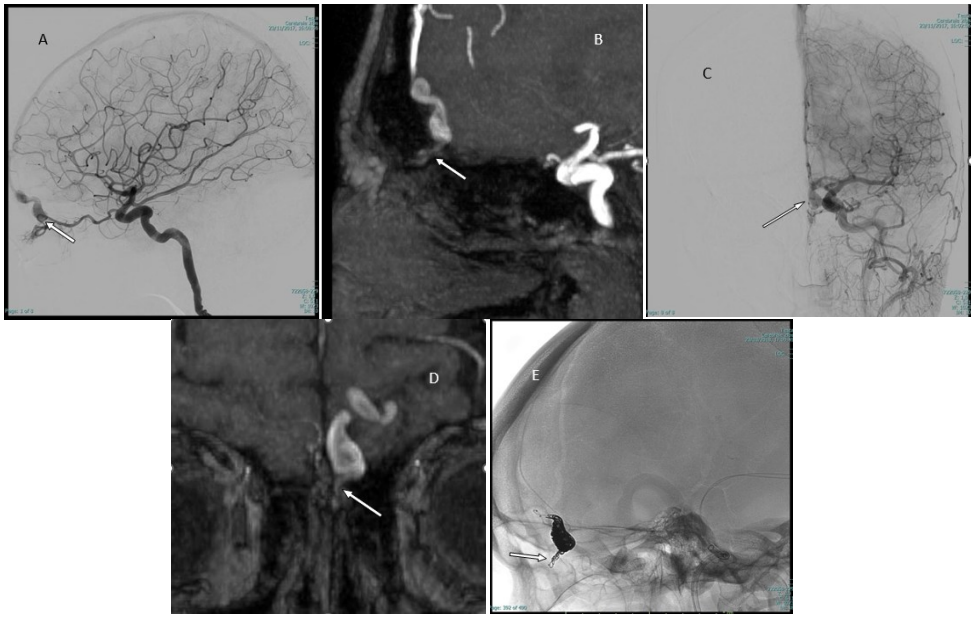


***Figure 2.*** *Patient 2: Type 4 fistula of the anterior cranial fossa drained by the VFC. DSA arterial phase left ICA (A) lateral view (C) antero-posterior view; IRM findings (B) sagittal view (D) coronal view; (E) lateral view post-treatment. Venous engorgement of pial veins in the anterior cranial fossa in connection with the vein of the foramen cecum (white arrows). The left Ophthalmic artery feeds the fistula with Ethmoidal branches. After treatment are visible coils in the lumen of the VFC (E).*

## Discussion

DAVF are a pathological, probably acquired, shunt between dural arteries and cerebral veins.

These two Patients showed hypertrophic VFC drained through the SSS; in the first Patient arterial feeders are arteries supplying the nasal mucosa, as found by San Millan Ruiz et al.[10]; in the second Patient arterial feeders of the DAVF are Ethmoidal branch of the Ophthalmic artery and Meningeal branch for the Cribriform plate.

Cribriform plate, Crista Galli are fed by Ethmoidal branches from Ophtalmic arteries, which represent as well the main feeders of DAVF of this region, with lesser contribution from middle Meningeal [4].

The roof of the nasal cavities and nasal septum are feed by Sphenopalatine arteries. Terminal anastomosis between these vessels are well known, and hemodinamic equilibrium is constituted on their respective border zones of vascularization. Foramen caecum and its vein, representing a link between extra and intracranial zones, are fed by both systems. In particular, an almost exclusive contribution from sphenopalatine arteries towards VFC, i.e. a VFC draining roof of nasal cavities and septum, as seen in patient 1, was described by San Millan Ruiz et al [10] in normal patients.

In both patients, VFC drained in hypertrophic, tortuous pial veins, finally ending in the SSS.

In our patients, the primary draining vein of the DAVF was the VFC, due to location and anatomical relationship with surrounding structures. The discrepancy between radiological studies, allowing to visualize VFC, [6][12] and pathological studies, finding no veins but fibrous tissue [7], in the majority of patients, remains unexplained. The small reticular vessels beneath the fibrous tissue occupying the foramen found by Kaplan et al [7] allowing functional flow communication without a macroscopic venous channel, may eventually explain those findings. In our cases, the existence of a DAVF could have caused enlargement of these small vascular channels, allowing us to identify a macroscopic vein.

Exact identification of the primary venous drainage is a critical point in management of DAVF. In effect, occlusion of this vein allows to achieve a definitive cure, while occlusion of the vein in a more distal position is usually not effective. VFC is located beneath the ethmoidal plane, and for this reason it may remain thin and difficult to identify and eventually to occlude.

## Conclusion

VFC represents a functional connection between dural, intracranial structures, and nasal cavities. In our patients, it also represented the primary draining vein of DAVF. Correct identification and obliteration are mandatory to successfully cure the patients.

## Conflicts of Interest

The authors have no conflicts of interest to declare.

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