

## Supernumerary fronto-orbital arteries arising from contralateral anterior cerebral artery associated with partially duplicated anterior communicating artery – case study and literature review

ALIN HORAȚIU NEDELCU<sup>1)</sup>, RĂZVAN TUDOR ȚEPORDEI<sup>1)</sup>, ANCA SAVA<sup>1)</sup>, CRISTINEL IONEL STAN<sup>1)</sup>, ANDA MARIA AIGNĂTOAEI<sup>2)</sup>, TRAIAN ȚĂRANU<sup>1)</sup>, MANUELA URSARU<sup>3)</sup>

<sup>1)</sup>Department of Morphofunctional Sciences, "Grigore T. Popa" University of Medicine and Pharmacy, Iassy, Romania

<sup>2)</sup>Department of Pathology, "Sf. Spiridon" Clinical Emergency Hospital, Iassy, Romania

<sup>3)</sup>Department of Radiology, "Grigore T. Popa" University of Medicine and Pharmacy, Iassy, Romania

### Abstract

The anatomical variations of the fronto-orbital artery are uncommon and rarely described in literature. During the dissection of a 64-year-old female formalized brain, we discovered a particular congenital abnormality regarding the arterial supply of the right fronto-orbital gyrus. We identified three arterial sources: a low caliber ipsilateral fronto-orbital artery that arises from the A2 segment of the right anterior cerebral artery and ends in the posterior part of the fronto-orbital gyrus, a large aberrant contralateral fronto-orbital artery that arises from the A2 segment of the left anterior cerebral artery, supplying most of the right fronto-orbital gyrus and a small accessory branch of the left anterior cerebral artery passing towards the contralateral fronto-orbital gyrus. These abnormalities are associated with a partially duplicated anterior communicating artery. This case shows a unique pattern of congenital brain vascular abnormalities that may have clinical and surgical implications.

**Keywords:** fronto-orbital artery, partially duplicated, anterior communicating artery, fronto-orbital gyrus.

### Introduction

Detailed knowledge of the cerebral arterial blood supply is crucial for neurosurgeons, neurologists and interventional radiologists. Due to particularities of the cerebral arterial vascularization, each major artery plays an important role in the brain's blood supply. Amongst them, the anterior cerebral artery particularly stands out due to its vast distribution territory. Moreover, anatomical variations of the anterior cerebral artery are quite common and are often described in the medical journals [1]. These variants refer to hypoplasia or aplasia, accessory anterior cerebral artery [2], anomalous origin of the callosomarginal artery [3] or uncommon origin of anterior communicating artery [4]. Despite that, only a small number of patterns regarding the fronto-orbital artery have been described, some authors mentioning its anomalous origins [5, 6].

The fronto-orbital artery (FOA) is the first cortical branch of the anterior cerebral artery (ACA), normally arising from the A2 segment [7, 8]. Some authors mention its origin from the A1 segment of ACA [1, 2], the pericallosal artery [2, 5], the callosal marginal artery or sometimes from the fronto-polar artery [9]. The trajectory of the fronto-orbital artery usually follows an anterior course, along the medial surface of the fronto-orbital gyrus. This artery typically supplies the *gyrus rectus*, the olfactory bulb and tract, the medial and inferior surface of the fronto-orbital gyrus and the anterior and medial surface of the superior frontal gyrus [8, 10].

In the current case report, we describe a unique vascular pattern represented by two anomalous right fronto-orbital arteries arising from the A2 segment of the contralateral anterior cerebral artery, associated with a partially duplicated anterior communicating artery (ACoA). These aberrant arteries are associated with a hypoplastic right fronto-orbital artery.

The aim of this study is to document the origin of the fronto-orbital artery in relation to its embryological development and to warn the clinicians about the anatomical variations of this artery. Furthermore, the duplication and fenestration of ACoA and their major complications, respectively arterial cerebral aneurism and subarachnoid hemorrhage are being discussed.

### Case presentation

The anatomical material consisted of a 64-year-old female brain fixed in a 10% formalin solution for 15 weeks. We dissected the arterial circle of Willis with the aid of an operator microscope (OPTRON – Zeiss OPMI 6), focusing on the anterior cerebral artery and the anterior communicating artery. In order to highlight the blood vessels and their distribution territory, we injected red ink into the left ACA, followed by injection of blue ink into the right ACA. The images were acquired with a Nikon D7000 digital camera equipped with a 60 mm AF Micro-Nikkor f/2.8D lens and were processed with Adobe Photoshop CS5 and Capture NX2 software.

During the dissection, we discovered a unique congenital abnormality regarding the arterial supply of the right fronto-orbital gyrus. Normally, this gyrus is vascularized by the fronto-orbital artery, which is the first cortical branch of the ipsilateral anterior cerebral artery. In our case, the arterial supply of the right fronto-orbital gyrus had three sources: one ipsilateral fronto-orbital artery, one contralateral fronto-orbital artery and another accessory branch originating from the contralateral ACA (Figure 1).

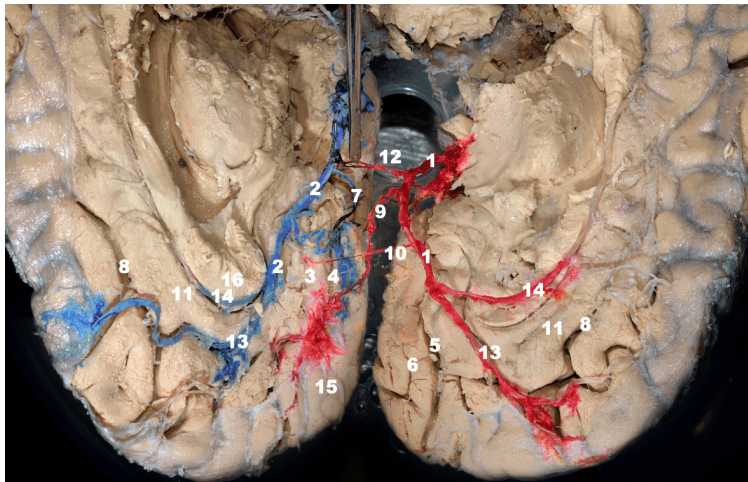
The first source is represented by the right fronto-orbital artery that originates from the A2 segment of the ipsilateral ACA. This artery has a short trajectory on the surface of the brain, situated into the *inferior rostral sulcus*, and then it splits in three branches: superior, inferior and anterior (Figure 2). The superior branch perforates the corresponding slope of the *sulcus* supplying the deep cerebral tissue. The inferior branch is directed towards the inferior border of the fronto-orbital gyrus. The anterior division is the terminal branch and continues the right fronto-orbital artery into the *inferior rostral sulcus*. At the distal extremity of the sulcus, this branch disappears into the cerebral tissue (Figure 2).

The second source, that we named aberrant right fronto-orbital artery (ARFOA), has the origin in the A2 segment of the left ACA, close to anterior communicating artery. This artery, that seems to be the main arterial source of the right fronto-orbital gyrus, is directed anteriorly and to the right, then passes under *falx cerebri* and before it

reaches the medial surface of the right fronto-orbital gyrus, it gives off thin collateral branches for the posterior part of the gyrus situated under *superior rostral sulcus* (Figure 2). The artery has a postero-anterior direction along with the medial surface of the fronto-orbital gyrus and splits in superficial branches, generating a “medusa head” aspect. The superior branches supply the inferior part of fronto-polar gyrus corresponding to area 10 Brodmann. The anterior branches exceed the superior border of the right hemisphere supplying the cortex of frontal pole, while the inferior branches direct to *gyrus rectus* crossing the hemispheric border. The main distribution territory is represented by the medial aspect of fronto-orbital gyrus (area 11 Brodmann) (Figure 2).

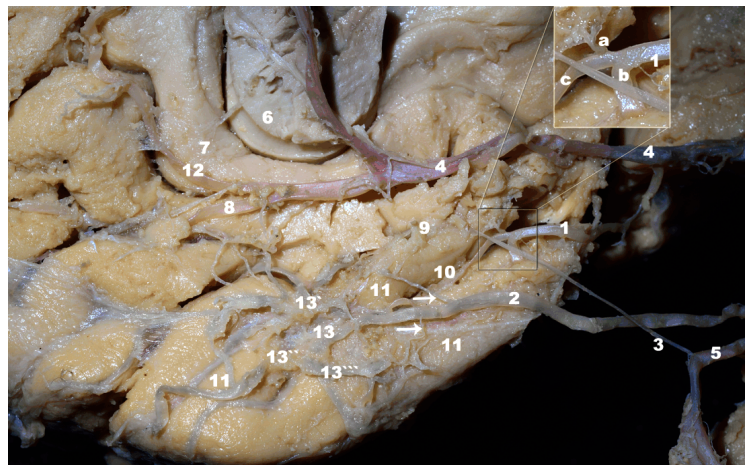
The third source is a small branch that we named accessory right fronto-orbital artery (AccRFOA). It originates in the A2 segment of the left anterior cerebral artery, distal to the previous branch. It is directed anteriorly and to the right, goes under the *falx cerebri*, crosses the previous branch on its dorsal aspect and reaches the medial surface of the right fronto-orbital gyrus. It supplies the segment situated above the *inferior rostral sulcus* (Figures 1 and 2).

In this case, the variations in the number and position of the FOA are also associated with a partially duplicated anterior communicating artery. The duplication occurs in the right half of ACoA. No aneurysm formation has been detected (Figure 3).



**Figure 1 – Ipsilateral and contralateral fronto-orbital arteries. Superior view of the medial aspect of both cerebral hemispheres. 1: Left anterior cerebral artery; 2: Right anterior cerebral artery; 3: Right superior rostral sulcus (Eberstaller); 4: Right (accessory) inferior rostral sulcus; 5: Left superior rostral sulcus; 6: Inferior rostral sulcus; 7: Right fronto-orbital artery (first source); 8: Cingulate sulcus; 9: Aberrant right fronto-orbital artery (ARFOA); 10: Accessory right fronto-orbital artery (AccRFOA); 11: Cingulate sulcus; 12: Partially duplicated anterior communicating artery; 13: Frontopolar artery; 14: Pericallosal artery; 15: Right fronto-orbital gyrus (*gyrus rectus*); 16: Genu of corpus callosum.**

**Figure 2 – Territory of distribution for the aberrant left (contralateral) fronto-orbital artery – second arterial source. Medial aspect of the right frontal lobe. 1: Right fronto-orbital artery; 2: Aberrant right fronto-orbital artery (ARFOA); 3: Accessory right fronto-orbital artery (AccRFOA); 4: Right anterior cerebral artery; 5: Left anterior cerebral artery; 6: Corpus callosum; 7: Cingulate gyrus; 8: Right fronto-polar artery; 9: Superior rostral sulcus (Eberstaller); 10: Inferior rostral sulcus; 11: Right fronto-orbital gyrus (*gyrus rectus*); 12: Right pericallosal artery; 13: “Medusa head” aspect of the aberrant contralateral fronto-orbital artery: 13’ – superior branches, 13’’ – anterior branches, 13''' – inferior branches. Inset: Branches of the right fronto-orbital artery: a – superior, b – inferior, and c – anterior branch. Arrows: collateral branches of the aberrant contralateral fronto-orbital artery.**







**Figure 3 – Fenestration of the anterior communicating artery. Basal view.**  
**1: Left frontal (orbital) lobe; 2: Right frontal (orbital) lobe; 3: Right internal carotid artery; 4: Left internal carotid artery; 5: Left anterior cerebral artery; 6: Partially duplicated anterior communicating artery; 7: Aberrant right fronto-orbital artery (ARFOA); 8: Accessory right fronto-orbital artery (AccRFOA); 9: Left frontopolar artery; 10: Left pericallosal artery.**

## Discussion

The vascular anatomy related to the anterior cerebral artery is quite complex due to its embryological development. During the formation process of the anterior communicating artery, a large plexiform anastomosis appears between the two anterior cerebral arteries. Normally, the vessels of this plexus will fuse together to form the anterior communicating artery. A fenestration process may appear instead, explaining the high incidence of anatomical variations [1, 11]. During this process, the mesenchymal tissue that normally forms the left fronto-orbital artery remains connected to the mesenchyme of the right hemisphere and supplies the contralateral fronto-orbital gyrus. Incomplete fusion of the plexiform anastomosis also explains the anatomical variants of ACoA, including duplication and fenestration [12].

We reported the unique case of an anomalous origin of the right fronto-orbital artery in the left ACA that is associated with an accessory branch emerging from the same source. To our knowledge, this is the first report of this pattern in the literature. Lee & Eastwood [5] and Maruyama *et al.* [6] reported two cases of anomalous origin of FOA observed during angiography. The current case report stands out by presenting a unique association of vascular malformations and by showing for the first time, an anomalous origin of FOA identified during anatomical dissection.

The abnormal position of the fronto-orbital artery can determine clinical symptoms such as tonic and clonic seizures related to an epilepsy focus in the right fronto-orbital gyrus [6]. These anatomical variants are also a predisposing factor for arterial aneurysms and intracranial hemorrhage [13], bringing the new techniques of angiography, especially angiographic projections, roadmapping [11] and endovascular access to ACA to a useful area of diagnosis and treatment [14].

The duplication of ACoA is a rare finding. In a large cadaveric study, Kapoor *et al.* [15] identified duplicated ACoA in 10% of cases, triplicated in 1.2% and plexiform in 0.4%. Gunnal *et al.* [16] mentioned in their anatomical study that the duplication was seen in 16 (10.66%) formalized brains. The imagistic studies realized by

Krzyżewski *et al.* [17] and Kovač *et al.* [18] report duplication of ACoA in 0.49% and respectively 0.4% of the investigated subjects. The discrepancy between the results of cadaveric and imagistic studies might be attributable to various factors: low diameters of the vessels, insufficient blood flow or imaging artifacts. To our knowledge, the partially duplicated ACoA has not been previously reported.

The difference between duplication and fenestration is a subject of debate for scientists. Makowicz *et al.* [1] considered that segmental duplications are, in fact, fenestrations as long they represent a division of a vessel. To sustain his findings, he proposes a morphological classification: type 1 – small, slit-like fenestration; type 2 – large, lenticular fenestration. Other authors such as Menshawi *et al.* [12] and Kovač *et al.* [18] concluded that the arterial fenestrations are completely different from arterial duplications. They claim that a fenestration is characterized by an artery that splits into two branches that rejoin. Furthermore, these authors affirm that the common origin is reserved to arterial fenestrations, while duplications should have distinct origins [12].

In contrast to duplications, cerebral arterial fenestrations are quite common. 2.1% of all the patients undergoing digital subtraction angiography were reported [19]. The fenestration is most often located in the posterior circulation (73.2%), especially in the basilar artery (52.6%). Fenestrations of the anterior circulation vessels are reported in 24.6% cases, mainly on ACoA. The rest of 2.2% are found on the internal carotid artery and the middle cerebral artery. The prevalence of aneurysms within the anterior and posterior fenestrations is almost similar: 60.7% vs. 61.1% [19].

de Gast *et al.* [11] concluded that there is a direct relation between fenestration of ACoA and aneurysms of the anterior brain circulation: 83% of the patients with fenestrated ACoA where diagnosed with ACoA aneurysm and 5.3% of the total number of patients with anterior circulation aneurysm associate ACoA fenestration. Moreover, in 4.4% of those cases, the aneurysms were located in ACoA and the rest of 0.9% in other arteries. The reported fenestration of ACoA in dissected bodies is between 7.5% and 40% [20].

Subarachnoid hemorrhage is often associated with arterial fenestration and especially with arterial aneurysms. The prevalence of hemorrhage in patients with aneurysms arising directly from fenestration is slightly higher (66.7%) compared to those patients with aneurysms situated at distance from the fenestration (58.6%) [19]. In a study which included 174 subjects, Hudák *et al.* [21] concluded that 95.2% of the patients with “unexplained subarachnoid hemorrhage” were diagnosed with arterial fenestration.

The treatment of the ACoA aneurysms hemorrhage is surgical or interventional. Most of the cases benefit of balloon assisted coil embolization, a minimally invasive technique with a low rate of procedure-related complications [20, 22]. The endovascular treatment ensures the complete or near-complete occlusion of the aneurysms for long-term and is a good alternative to traditional clipping procedure [22]. The classical surgical treatment is reserved for specific patients because of the potentially dangerous dissection of adherent vessels or vascular anatomical variations [23, 24]. A minimally surgical procedure was developed recently to expose the fronto-basal area of the brain. This approach, called supraorbital trans-eyebrow craniotomy, permits clipping or trapping aneurism techniques as well as the resection of different tumors such as gliomas and meningiomas, avoiding the classical pterional and fronto-temporo-orbito-zygomatic paths [25–28].

## ☒ Conclusions

Detailed knowledge of congenital anomalies related to cerebral vascular system is important for neurosurgeons and clinicians due to numerous arterial variations. The relevance of this case in medical practice lies in the fact that it brings to attention a rare anomaly of the anterior cerebral artery vascular system, where most of the cerebral aneurysms associated with congenital abnormalities occur. Nevertheless, recognizing and reporting the anatomic variants of the fronto-orbital arteries could be helpful in planning surgical procedures involving the ACA and ACoA, especially since these congenital abnormalities modify the regional landmarks.

## Conflict of interests

The authors declare that they have no conflict of interests.

## References

- [1] Makowicz G, Poniatowska R, Lusawa M. Variants of cerebral arteries – anterior circulation. *Pol J Radiol*, 2013, 78(3):42–47.
- [2] Maruyama N, Fukuma A, Ihara I, Ando S, Yamasaki T, Moritake K. Unusual variant of the anterior cerebral artery. *Neurol Med Chir (Tokyo)*, 2005, 45(5):246–248.
- [3] Krishnamoorthy T, Gupta AK, Bhattacharya RN, Rajesh BJ, Purkayastha S. Anomalous origin of the callosomarginal artery from the A1 segment with an associated saccular aneurysm. *AJNR Am J Neuroradiol*, 2006, 27(10):2075–2077.
- [4] Gurdal E, Cakmak O, Yalcinkaya M, Uzun I, Cavdar S. Two variations of the anterior communicating artery: a clinical reminder. *Neuroanatomy*, 2004, 3:32–34.
- [5] Lee ER, Eastwood JD. An unusual variant of the fronto-orbital artery. *AJNR Am J Neuroradiol*, 2000, 21(5):939–940.
- [6] Maruyama N, Fukuma A, Ihara I, Ando S, Moritake K. Epilepsy and variation in the frontal lobe artery. *Epilepsy Res*, 2005, 64(1–2):71–75.
- [7] Bhat DI, Shukla DP, Devi BI. Partially thrombosed distal orbito-frontal artery aneurysm mimicking an A1 segment aneurysm. *J Neurosci Rural Pract*, 2014, 5(4):436–437.
- [8] Avci E, Fossett D, Aslan M, Attar A, Egemen N. Branches of the anterior cerebral artery near the anterior communicating artery complex: an anatomic study and surgical perspective. *Neurol Med Chir (Tokyo)*, 2003, 43(7):329–333; discussion 333.
- [9] Aso K, Kashimura H, Takeda M, Chida K. An unusual variant of the common trunk of the fronto-orbital and frontopolar arteries associated with a ruptured aneurysm of the A1 segment of the anterior cerebral artery. *Surg Neurol Int*, 2015, 6(Suppl 16):S418–S420.
- [10] Rhoton AL Jr. The supratentorial arteries. *Neurosurgery*, 2002, 51(4 Suppl 1):S53–S120.
- [11] de Gast AN, van Rooij WJ, Sluzewski M. Fenestrations of the anterior communicating artery: incidence on 3D angiography and relationship to aneurysms. *AJNR Am J Neuroradiol*, 2008, 29(2):296–298.
- [12] Menshawi K, Mohr JP, Gutierrez J. A functional perspective on the embryology and anatomy of the cerebral blood supply. *J Stroke*, 2015, 17(2):144–158.
- [13] Hong SK. Ruptured proximal anterior cerebral artery (A1) aneurysm located at an anomalous branching of the fronto-orbital artery – a case report. *J Korean Med Sci*, 1997, 12(6): 576–580.
- [14] Graves VB. Advancing loop technique for endovascular access to the anterior cerebral artery. *AJNR Am J Neuroradiol*, 1998, 19(4):778–780.
- [15] Kapoor K, Singh B, Dewan LI. Variations in the configuration of the circle of Willis. *Anat Sci Int*, 2008, 83(2):96–106.
- [16] Gunnal SA, Farooqui MS, Wabale RN. Anatomical variations of the circulus arteriosus in cadaveric human brains. *Neurol Res Int*, 2014, 2014:687281.
- [17] Krzyżewski RM, Tomaszewski KA, Kochana M, Kopeć M, Klimek-Piotrowska W, Walocha JA. Anatomical variations of the anterior communicating artery complex: gender relationship. *Surg Radiol Anat*, 2015, 37(1):81–86.
- [18] Kovač JD, Stanković A, Stanković D, Kovač B, Šaranović D. Intracranial arterial variations: a comprehensive evaluation using CT angiography. *Med Sci Monit*, 2014, 20:420–427.
- [19] Cooke DL, Stout CE, Kim WT, Kansagra AP, Yu JP, Gu A, Jewell NP, Hetts SW, Higashida RT, Dowd CF, Halbach VV. Cerebral arterial fenestrations. *Interv Neuroradiol*, 2014, 20(3): 261–274.
- [20] Dimmick SJ, Faulder KC. Fenestrated anterior cerebral artery with associated arterial anomalies. Case reports and literature review. *Interv Neuroradiol*, 2008, 14(4):441–445.
- [21] Hudák I, Lenzsér G, Lunenkova V, Dóczi T. Cerebral arterial fenestrations: a common phenomenon in unexplained subarachnoid haemorrhage. *Acta Neurochir (Wien)*, 2013, 155(2): 217–222.
- [22] Fang S, Brinjikji W, Murad MH, Kallmes DF, Cloft HJ, Lanzino G. Endovascular treatment of anterior communicating artery aneurysms: a systematic review and meta-analysis. *AJNR Am J Neuroradiol*, 2014, 35(5):943–947.
- [23] Mugikura S, Kikuchi H, Fujii T, Murata T, Takase K, Mori E, Marinković S, Takahashi S. MR imaging of subcallosal artery infarct causing amnesia after surgery for anterior communicating artery aneurysm. *AJNR Am J Neuroradiol*, 2014, 35(12):2293–2301.
- [24] Zada G, Christian E, Liu CY, Giannotta SL. Fenestrated aneurysm clips in the surgical management of anterior communicating artery aneurysms: operative techniques and strategy. *Clinical article. Neurosurg Focus*, 2009, 26(5): E7.
- [25] Prat-Acín R, Galeano-Senabre I, Pancucci G, Evangelista R, Ayuso-Sacido A, Botella C. Supraorbital trans-eyebrow craniotomy and fluorescence-guided resection of fronto-basal high grade gliomas. *Clin Neurol Neurosurg*, 2013, 115(9):1586–1590.
- [26] Ladziński P, Majchrzak H, Kaspera W, Majchrzak K, Tymowski M, Adamczyk P. Medial sphenoid ridge meningiomas: early and

- long-term results of surgical removal using the fronto-temporo-orbito-zygomatic approach. *Neurol Neurochir Pol*, 2010, 44(5): 464–474.
- [27] Ormond DR, Hadjipanayis CG. The supraorbital keyhole craniotomy through an eyebrow incision: its origins and evolution. *Minim Invasive Surg*, 2013, 2013:296469.
- [28] Ormond DR, Hadjipanayis CG. The history of neurosurgery and its relation to the development and refinement of the frontotemporal craniotomy. *Neurosurg Focus*, 2014, 36(4): E12.

**Corresponding author**

Anca Sava, Professor, MD, PhD, Department of Morphofunctional Sciences, “Grigore T. Popa” University of Medicine and Pharmacy, 16 Universităţii Street, 700114 Iassy, Romania; Phone +40744–303 678, e-mail: dr.anca.sava.68@gmail.com

*Received: March 3, 2016*

*Accepted: December 2, 2016*