Bilateral Infraoptic Origin of Anterior Cerebral Arteries: a Rare Anomaly and its Embryological and Clinical Significance

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Summary

We describe a case of bilateral infraoptic origin of the anterior cerebral arteries associated with an anterior communicating artery (ACOM) aneurysm. Anatomical variations of the anterior cerebral artery (ACA) are common; however, bilateral infraoptic course of the anterior cerebral artery is extremely rare. Since an infraoptic course of the ACA is associated with ACOM aneurysm formation, an understanding of the cerebrovascular anatomy and embryology is important for appropriate management of the aneurysm.

Introduction

Variations of the anterior cerebral artery–anterior communicating artery complex are common findings in imaging studies and are usually of little clinical significance. An infraoptic course of the ACA is a rare subtype, with only approximately 35 cases reported to-date¹. About 44% of patients with an infraoptic course of the ACA are associated with ACOM aneurysm¹. Thus, it is important to recognize this cerebrovascular variation for treatment planning, especially for the endovascular approach. We describe the CT, MR and angiographic findings of a case of bilateral infraoptic ACA with an associated anterior communicating artery aneurysm.

Case Report

A 34-year-old male patient originally presented with anxiety related episodic hypertension. On physical examination, he was found to have a heart murmur and noted to have congenital absence of the ring and little fingers of his left-hand. There were no dysmorphic features, cognition and mental state were normal, and there was no significant family history. His medical history was significant for metastatic testicular cancer that was treated with orchidectomy and chemotherapy. He was also diagnosed as having a bicuspid aortic valve with post-ductal coarctation of the aorta, which was successfully treated with aortic stenting. Subsequent cerebral MR angiography showed a bilateral infraoptic course of both anterior cerebral arteries and a 7.5 mm anterior communicating artery aneurysm (figure 1). These appearances were further characterized by CT angiography and catheter angiogram (figures 2 and 3).

Discussion

In general the ACA arises from the internal carotid artery (ICA) terminus and traverses medially over the respective optic nerve (supraoptic course) and communicates with ACA of the other side through the anterior communicating artery. One of the most common variations of the ACA–anterior communicating ar-

Figure 1 A) Coronal maximal intensity projection image from a time of flight MRA shows bilateral low bifurcations of ICA (arrowhead) with an associated anterior communicating artery aneurysm. B) Axial T2-weighted image shows a pre-chiasmatic position of both ascending A1 segments (arrow points to left A1) that traverse medial and inferior to the optic nerves.

tery complex is a size asymmetry between two A1 segments, which is reported in approximately 80% of patients².

An infraoptic course of the A1 segment of the ACA is associated with a low ICA terminal bifurcation. The bifurcation is usually located at the proximal intradural ICA at or just above the level of the origin of the ophthalmic artery. In our case we document that both infraoptic origins of A1 are proximal to the origin of the ophthalmic arteries and arise below the optic strut (figure 2 and 3) possibly extradurally. The

optic strut is described as a reliable anatomic landmark for discrimination between intradural and extradural segments of ICA³. The A1 segments extend superomedially from their aberrant origin below the optic nerves and chiasm. Their path in relation to the optic chiasm is variable, with pre-, intra- and post-chiasmatic variants described. The artery then ascends medially to join normally positioned anterior communicating artery. Although a supraoptic A1 connection is not shown in our case, an associated normally positioned but extremely hypoplastic A1 has been reported^{4,5}. Infraoptic A1 is unilateral in most cases with only three cases of a bilateral infraoptic course reported in the anatomic and neurosurgical literature¹.

Certain anomalous arterial variations in the circle of Willis are known to have an associated increased incidence of cerebral aneurysms. Other associations include contralateral ICA agenesis⁴, common origin with an ophthalmic artery¹, azygos anterior cerebral artery⁶, arteriovenous malformation⁷ and aortic coarctation¹. Our patient had an associated anterior communicating artery aneurysm and coarctation of the aorta. However, the presence of a bicuspid aortic valve and postaxial phocomelia has never been described in cases of an infraoptic ACA. A review of human abortus, chromosomal, and teratogenic syndromes suggested a positive association of branchial defects

with cardiac and limb defects. It suggests the presence of a branchiocardiomeletic developmental field, which is involved in the coordinated development of the branchial arches, heart and limbs⁸. Disturbance of this putative developmental field may account for the various anomalies observed in this case. However, we cannot exclude the possibility of random development of various anomalies in the case.

The embryogenesis of the aberrant infraoptic A1 is controversial. The most likely explanation is related to an error in the development of the

Figure 2 A) Axial CT reconstructed angiogram shows possible extra-dural origin of these A1 segments bilaterally (arrows) proximal to the optic strut (asterix). B) CTA 2D coronal reformatted image shows bilateral bifurcations below the anterior clinoid process, absent normal supraoptic A1 segments and an anterior communicating artery aneurysm.

primitive ophthalmic artery⁹. At the 4-8 mm embryonic stage, two arteries supply the orbital region: the ventral ophthalmic artery supplying the cerebral derivative (the optic cup), and the dorsal ophthalmic artery supplying the mesodermal derivatives (rest of the orbital contents). The ventral ophthalmic artery originates from the future anterior cerebral artery and traversing downwards enters the orbit through the op-

tic canal. The dorsal ophthalmic artery originates from the intracavernous part of ICA and enters orbit through the superior orbital fissure. With further development anastomosis between the ICA and ventral ophthalmic artery develops near the optic canal. Normally this leads to caudal migration of the origin of ophthalmic artery to usual adult configuration. The proximal part between this new origin and the

Figure 3 A) Composite overlapped digital subtraction angiograms of both right and left internal carotid arteries demonstrate low terminal carotid artery bifurcations, absent supraoptic A1 segments and an associated anterior communicating artery aneurysm. B) Surface rendered 3D angiographic reconstructions of the internal carotid arteries show that the ophthalmic artery origin is distal to the origin of the anomalous A1 segment (arrows). The anterior communicating artery aneurysm fills during the left carotid angiogram.

ACA regresses. It is hypothesized that failure of this regression, coupled with regression of supraoptic A1 segment, gives rise to infraoptic aberrant course of A1.

Support for this theory comes from the observation that the infraoptic ACA can have a common origin with the ophthalmic artery. However, we note that infraoptic origin of ACA proximal to the origin of ophthalmic artery, as in our case, cannot be explained by this theory. Other hypotheses are persistent communication of the primitive ventral and dorsal ophthalmic arteries⁴ or that the infraoptic A1 is a persistent embryonic anastomosis between the primitive maxillary artery (which is thought to arise from the cavernous ICA) and the ACA¹⁰.

Associated aneurysm formation is most commonly seen in anterior communicating artery location. Recognition of an infraoptic ACA anomaly with an associated aneurysm allows for optimal planning prior to surgical or endovascular treatment. Based on the aneurysm size, location and the patient's age, this aneurysm has been treated endovascularly.

Conclusions

Bilateral infraoptic aberrant course of A1 is a rare anomaly that is associated with increased risk of aneurysm formation. An understanding of this anomaly is important to allow for optimal planning of surgical or endovascular aneurysm treatment.

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EDITORIAL COMMENT

Chakraborty et Al very elegantly report a bilateral infra optic course of the anterior cerebral artery in a patient presenting with an anterior communicating aneurysm. The arterial variations of the circle of Willis are known to be associated with arterial aneurysms thus pointing to the "location" of the disease on the arterial part of the vascular tree. Other arterial diseases (coarctation of the aorta, PHACE syndrome, ...) are associated with arterial anomalies. For other vascular lesions at the other extremity of the tree, like cavernomas, dural sinus malformation, lymphatic malformations of the face, the association with venous variations (DVA) are very common. AVMS and all AVShunts in general correspond to an impact on the venous side of the capillary. They are not associated with arterial anomalies or even DVA; the association of an arteriovenous malformation with an arterial anomaly or DVA is likely to be fortuitous.

Concerning the embryological discussion on the path and significance of the infraoptic course of the ACA, surgical or anatomic observations are enlightened by embryology and comparative anatomy. The significance of the ventral ophthalmic artery is that of the supply of the optic nerve (central nervous system), and it arises from a primary cerebral artery (the anterior cerebral artery). This artery originates in the subarachnoid space and as long as it courses into the optic canal it has to travel through the same space supplying similar CNS structures. The ventral ophthalmic artery cannot be extradural nor can the infraoptic ACA in this type of variant.

For the same reason arteries in the infero-lateral trunk would never be subarachnoid because its significance is the supply to non migrant structures in the epidural space. The hypothesis that the primitive maxillary could be involved in the discussion is doubtful as the remnant of this artery is the posterior hypophyseal artery. If so the ACA would run through the posterior part of the sella turcica¹. But this ACA variant is different from the infraoptic course which remains a clear entity.

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