World Journal of Neurology

Online Submissions: http://www.wjgnet.com/esps/
wjneurol@wjgnet.com
doi:10.5316/wjn.v3.i2.10

World J Neurol 2013 June 28; 3(2): 10-13
ISSN 2218-6212 (online)

CASE REPORT

Anterior communicating artery aneurysm associated with duplicated hypoplastic right A1 segment

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Received: February 11, 2013 Revised: April 15, 2013
Accepted: May 8, 2013
Published online: June 28, 2013

Abstract

Variations of the anterior cerebral artery (ACA)-anterior communicating artery (ACoA) complex are commonly observed when associated with a symptomatic intracranial aneurysm. We report an asymptomatic ACoA aneurysm associated with duplicated hypoplastic A1 segment of the right ACA, observed in a 70-year-old female cadaver. Furthermore, the aneurysm, practically substituting the ACoA, caused a remarkable depression on the internal surface of the right frontal lobe, anterior to the optic chiasm. Aneurysms and other anomalies of the ACA and ACoA are common and their microvascular surgical management requires sound knowledge of the normal and variant vascular anatomy. Persistence of some embryonic vessels that normally disappear, disappearance of vessels that would normally persist or sprouting of new vessels due to hemodynamic and genetic factors are the usual causes for such anomalies. The high incidence of coexisting vascular anomalies and aneurysm suggests that such abnormalities predispose to aneurysm formation due to changes in the regional blood flow. A1 segment duplication has been reported to occur in 4% of subjects in cadaveric studies and in up to 0.5%-9.7% of cases of ACoA aneurysm surgery. Angiographic hypoplasias and aplasias of the A1 segment have been also correlated with ACoA aneurysm patients.

Key words: Anterior cerebral artery; Anterior communicating artery; Aneurysm; Duplication; Hypoplasia

Core tip: Variations of the anterior cerebral artery (ACA)-anterior communicating artery (ACoA) complex are commonly observed when associated with a symptomatic intracranial aneurysm. We report an asymptomatic ACoA aneurysm associated with duplicated hypoplastic A1 segment of the right ACA, observed in a 70-year-old female. Furthermore, the aneurysm, practically substituting the ACoA, caused a remarkable depression on the internal surface of the right frontal lobe, antero-inferior to the anterior commissure and anterior to the optic chiasm. A literature review of similar cases and underlying mechanisms of such anomalies is presented.

Mavridis IN, Anagnostopoulou S. Anterior communicating artery aneurysm associated with duplicated hypoplastic right A1 segment. *World J Neurol* 2013; 3(2): 10-13 Available from: URL: http://www.wjgnet.com/2218-6212/full/v3/i2/10.htm DOI: http://dx.doi.org/10.5316/wjn.v3.i2.10

INTRODUCTION

Variations of the anterior cerebral artery (ACA)-anterior communicating artery (ACoA) complex are commonly observed when associated with a symptomatic intracranial aneurysm[1]. We report an asymptomatic ACoA aneurysm associated with duplicated hypoplastic A1 segment of the right ACA and causing a remarkable depression on the internal surface of the right frontal lobe. These anterior Willis circle anomalies were found during routine

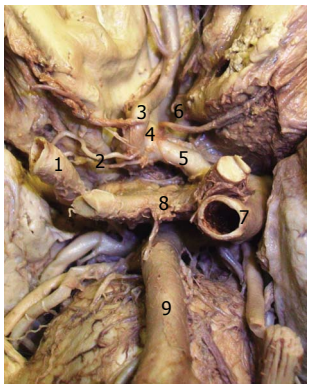

Figure 1 Duplicated hypoplastic A1 segment of the right anterior cerebral artery. 1: Right internal carotid artery (ICA); 2: Duplicated hypoplastic A1 segment of the right anterior cerebral artery (ACA); 3: A2 segment of the right ACA; 4: Anterior communicating artery (aneurysm); 5: A1 segment of the left ACA; 6: A2 segment of the left ACA; 7: Left ICA; 8: Optic chiasm; 9: Basilar artery.

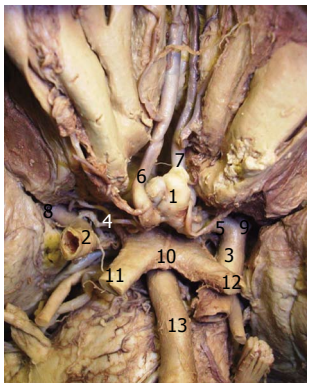

Figure 2 Duplicated hypoplastic A1 segment of the right anterior cerebral artery and anterior communicating artery aneurysm. 1: Anterior communicating artery aneurysm; 2: Right internal carotid artery (ICA); 3: Left ICA; 4: Duplicated hypoplastic A1 segment of the right anterior cerebral artery (ACA); 5: A1 segment of the left ACA; 6: A2 segment of the right ACA; 7: A2 segment of the left ACA; 8: Right middle cerebral artery (MCA); 9: Left MCA; 10: Optic chiasm; 11: Right optic nerve; 12: Left optic nerve; 13: Basilar artery.

dissection by chance. They were observed in a formalin-embalmed cadaver of a 70-year-old female who had died from heart disease. The details of this case as well as a relative literature-based discussion are presented below.

CASE REPORT

Dissecting the Willis circle arteries of this case's brain, it was observed no normal A1 (precommunicating) segment of the right ACA. Specifically, this segment was substituted by two hypoplastic branches (of about 1 mm in diameter) connecting the right internal carotid artery (ICA) with the A2 (postcommunicating) segment of the right ACA (Figures 1 and 2). Furthermore, an ACoA aneurysm (of about 10 mm in diameter), practically substituting the ACoA, was observed. Interestingly, it caused a remarkable depression on the internal surface of the right frontal lobe, antero-inferior to the anterior commissure and anterior (and slightly superior) to the optic chiasm (Figure 3).

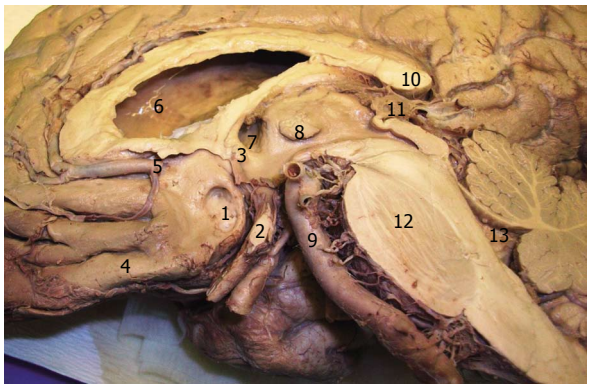

Figure 3 The depression on the internal surface of the right frontal lobe caused by anterior communicating artery aneurysm. 1: Cortical surface depression caused by aneurysm; 2: Optic chiasm; 3: Anterior commissure; 4: Gyrus rectus; 5: Right anterior cerebral artery; 6: Right lateral ventricle; 7: Right Monro foramen; 8: Massa intermedia; 9: Basilar artery; 10: Corpus callosum (splenium); 11: Pineal gland; 12: Pons (pontine nuclei); 13: Fourth ventricle.

DISCUSSION

Cases

More than half a century ago, Schuermann[2] discussed on unilateral ACA aplasia and surgical indication of ACoA aneurysm whereas Burmester *et al*[3] reported two cases of unilateral aplasia of the ICA in simultaneous aneurysm formation in the anterior area of the circle of Willis.

Matsumura *et al*[4] reported two patients with ruptured ACoA aneurysms associated with fenestration of the ACA. According to these authors, only few angiographic demonstrations of fenestration of the ACA have been reported in the literature. All fenestrations were limited to the distal half of the A1 portion, and approximately half of them were associated with aneurysms. The high incidence of coexisting fenestration and aneurysm suggests that congenital factors may play a role in the pathogenesis of cerebral aneurysm.

Kawakita *et al*[5] reported a case of a 25-year-old man with a ruptured saccular aneurysm in hypoplastic proximal ACA. He suffered a sudden onset of severe headache and vomiting due to subarachnoid hemorrhage. Cerebral angiography revealed this aneurysm in the hypoplastic A1 portion of the right ACA and no branch was present at the site of the aneurysmal neck.

Kim *et al*[6] reported a case of a 64-year-old female diagnosed with aneurysms of the proximal ACA and ACoA associated with middle cerebral artery (MCA) aplasia.

Weil *et al*[7] described a misleading case of a 70-year-old man with partially occluded A1 segment duplication that mimicked an ACoA aneurysm on computed tomography angiography and conventional angiography and led to surgical intervention. According to these authors, the location of such an anomaly at the ACoA on the side of least hemodynamic stress may provide a clue to recognizing this variant.

Anomalies of the ACA-ACoA-recurrent artery complex are frequently encountered, especially during ACoA

aneurysm surgery. Among these anatomic variations, duplication of the A1 segment of the ACA is infrequent. It has been reported to occur in 4% of subjects in cadaveric studies and in up to 0.5%-9.7% of cases of ACoA aneurysm surgery. Although A1 segment duplication can be identified on angiography, superimposition of vessels may render its identification difficult. Most clinically reported cases of A1 duplication are angiographically occult variations later identified during ACoA aneurysm surgery usually without consequences. Weil *et al*[7] concluded that A1 segment duplication with one arm occluded, can mimic an ACoA aneurysm on angiography and that the location of the anomaly at the ACoA on the side of least hemodynamic stress may provide a clue to suspect this variant.

Series

Kitami *et al*[8] analyzed angiographic features of the ACA and related vascular anomalies in a series of 704 aneurysm patients (mean age: 53 years). The total number of aneurysms was 866 with site distribution: ACoA 27%, MCA 31%, ICA 38%, ACA 6%, vertebrobasilar 5% and other arteries. More than one aneurysm was found out in 137 cases. Cases were divided into two groups, namely one who had the ACoA aneurysm and the other who had not. Angiographic calibers of A1 portion were compared statistically between these two groups. The ACoA aneurysm group showed significant asymmetry of A1 ($P < 0.005$) compared to the other cases. The left A1 portions were significantly dominant ($P < 0.05$) to the right in the ACoA aneurysm group. Angiographic hypoplasias and aplasias of A1 were found in the ACoA aneurysm group more frequently than in the other.

Karazincir *et al*[9] investigated the sites of intracranial aneurysms and incidence of associated congenital variations or anomalies. They retrospectively evaluated 190 cerebral angiography examinations that were positive for aneurysm. Fourteen cases with vasospasm were excluded and the remaining 176 patients were assessed for the location of the aneurysm and co-incidental vascular variations and/or anomalies. The most frequent locations of aneurysms were the supraclinoid ICA (32%), ACoA (30%) and MCA bifurcation (23%). Twenty-eight (17%) patients had multiple aneurysms. Ninety-one (52%) patients had a vascular anomaly or variation. Hypoplasia or agenesis of the A1 segment was found in 48 patients and an azygous ACA in one. The authors concluded that due to an increased hemodynamic stress, congenital anomalies of the intracranial arteries predispose to the formation of saccular aneurysms. Anomalies such as A1 hypoplasia or agenesis, azygous ACA, accessory MCA and persistent trigeminal artery are detected more frequently in patients with cerebral aneurysms compared to the normal population.

Aneurysms of the ACA and ACoA are common and their microvascular surgical management requires sound knowledge of the normal and variant vascular anatomy[10]. Saidi *et al*[10], from Kenya, evaluated ACA and ACoA variant anatomy by dissecting 36 adult brains. The ACA was observed to originate from the ipsilateral ICA in all cases. Unique variations observed include an accessory ACA from the ACoA, "bihemispheric pericallosal arteries", intertwining course of the A2 segments of the ACAs and crossing branches from one hemisphere to another. Variations of the ACoA were also observed including fenestration (26%) and duplication (13%). The majority of ACA bifurcations, in their study, were supracallosal suggesting the need for exploration of the interhemispheric fissure during surgical corrections of distal ACA aneurysms. ACoA fenestration was the most common variation raising concern as this has been shown to compromise collateral flow and predispose to aneurysm formation.

Kapoor *et al*[11] studied variations of the Willis circle using brains from 1000 medicolegal autopsy subjects of varying ages (Indian population). Out of 1000 specimens examined, 452 (45.2%) conformed to the typical pattern. In the rest of the specimens (54.8%) there were variations in the circulus arteriosus. The circle was deficient in 32 (3.2%). The ACA was absent in 0.4%, hypoplastic in 1.7%, duplicated in 2.6%, triple in 2.3% and single in 0.9%. The ACoA was absent in 1.8%, duplicate in 10%, triplicate in 1.2% and plexiform in 0.4%. Seventy-four brains (7.4%) had multiple variations. Intracranial saccular aneurysm was present in 10 (1%). Persistence of some embryonic vessel that normally disappear, disappearance of vessels that would normally persist or sprouting of new vessels due to hemodynamic and genetic factors are the usual causes for such anomalies.

Further, these authors reported that in four specimens (three male, one female) the proximal part of the ACA was absent on one side and larger than normal on the other side. Hypoplasticity of the proximal part of the ACA was present in 15 male specimens (10 right, 5 left) and two female specimens (one right, one left). But the distal part was normal in size. Loop formation in the proximal segment of the ACA was seen in 12 male specimens (seven right, five left) and 14 female specimens (six right, seven left, one on both sides). In each case the ACA divided into two components approximately 1 cm beyond its origin and rejoined to form a single artery. A loop was thus formed; no structure passed through this loop. The inside anteroposterior length of the loop varied from 2 to 12 mm. In one instance the loop extended 0.5 cm beyond ACoA, which joined the inner of the two constituent vessels of the loop. In 14 instances both the constituent vessels were apparently equal in caliber. In the remaining 12 instances one or the other artery of the loop was larger[11].

Finally, Kapoor *et al*[11] mentioned that hypoplasticity of the proximal part of the ACA is often described in the literature although a wide variation in the incidence, ranging from 4% to 44.3%, has been reported. Their study found hypoplasticity in only 17 specimens (1.70%). The wide variations in the incidence may be due to the fact that some workers have studied pathological or infarcted brains, while others based their studies on brains obtained from persons with mental disorder. Such structural

defects would resist the collateral blood flow. Duplication of a part of the ACA has been described by various authors under different names such as 'splitting' and 'island or loop formation'.

In conclusion, aneurysms and other anomalies of the ACA and ACoA are common and their microvascular surgical management requires sound knowledge of the normal and variant vascular anatomy. However, a case combining hypoplastic duplicated A1 segment, asymptomatic ACoA aneurysm and remarkable cortical surface depression is quite unusual. Persistence of some embryonic vessels that normally disappear, disappearance of vessels that would normally persist or sprouting of new vessels due to hemodynamic and genetic factors are the usual causes for such anomalies. The high incidence of coexisting vascular anomalies and aneurysms suggests that such abnormalities predispose to aneurysm formation due to changes in the regional blood flow.

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