CASE REPORTS

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Unruptured distal anterior cerebral artery mirror aneurysms associated with ruptured middle cerebral artery aneurysm: A case report

Nerupturisane distalne identične bilateralne aneurizme prednjih moždanih arterija udružene sa rupturisanom aneurizmom srednje moždane arterije

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Abstract

Introduction. Distal anterior cerebral artery (DACA) aneurysms, also known as pericallosal aneurysms are rare, while aneurysms in mirror position are extremely rare. These aneurysms have high tendency for rupture (PHASES score is always > 4). In more than a half of the patients with the DACA aneurysm rupture, imaging reveals intracerebral hematoma which is a predictor of poor outcome. **Case report.** A 49-year-old female patient was treated endovascularly in other institution, due to middle cerebral artery aneurysm (MCA) rupture, when the two small bilateral aneurysms at the distal segments of anterior cerebral artery (ACA) were revealed, left one measuring 4.5 mm and the right one measuring 6 mm in size, with the aneurysmal neck width of 3 mm and 4 mm, respectively. The decision was made by the interventional neuroradiologist only to treat the bleeding MCA aneurysm immediately. The patient was referred to our department six months later, and it was decided to perform microsurgical occlusion of the remaining DACA aneurysms. Unilateral interhemispheric approach was chosen to reach the distal ACAs and aneurysms at pericallosal-callosomarginal junction were clipped and completely excluded from the circulation. **Conclusion.** Management of DACA aneurysms is a surgical chellenge, even for experienced neurosurgeons. It is controversial whether these should be surgically clipped or coiled endovascularly, especially in cases like this one when a same-stage, endovascular coiling might look like a perfect approach. Surgical treatment should be prompt due to their tendency to early rupture. Careful evaluation for multiplicity is mandatory.

Key words:
aneurysm, ruptured; anterior cerebral artery; endovascular procedures; intracranial aneurysm; microsurgery; middle cerebral artery; neurosurgical procedures; treatment outcome.

Apstrakt

Uvod. Aneurizme distalnog segmenta prednje moždane arterije [*distal anterior cerebral artery* (DACA)], takođe poznate kao perikalozna arterija, retke su, dok su bilateralne aneurizme u identičnoj poziciji ekstremno retke. Te aneurizme imaju veliku tendenciju ka rupturi (PHASES skor je uvek > 4). U više od polovine bolesnika sa rupturom DACA aneurizme formira se intracerebralni hematom, koji je prediktor lošeg ishoda lečenja. **Prikaz bolesnika.** Bolesnica, stara 49 godina je, zbog rupture aneurizme na srednjemoždanoj arteriji [*middle cerebral artery* (MCA)], prethodno lečena endovaskularnom procedurom u drugoj ustanovi, kada su dijagnostikovane i dve male simetrične aneurizme na DACA obostrano. Dimenzija leve aneurizme bila je 4,5 mm, a desne 6 mm, dok su širine vrata bile 3 mm, odnosno 4 mm. Tada je interventni radiolog doneo odluku da leči samo krvareću aneurizmu na MCA. Bolesnica je upućena u našu ustanovu 6 meseci kasnije i doneta je odluka da se sprovede mikrohirurško lečenje aneurizmi na DACA. Uz pomoć unilateralnog interhemisferičnog pristupa i mikrohirurške tehnike obe simetrične aneurizme na kalozo-kalozomarginalnom spoju isključene su iz cirkulacije. **Zaključak.** Lečenje DACA aneurizmi je hirurški izazov, čak i za iskusne neurohirurge. I dalje postoji kontroverza u vezi izbora modaliteta lečenja – mikrohirurgija ili endovaskularna procedura, pogotovu u slučajevima kada se *coiling* u istom aktu sa udruženim aneurizmama čini kao odličan

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izbor. Zbog tendencije ka ranoj rupturi tih aneurizmi, mikrohirurško lečenje ne treba odlagati. Obavezna je provera postojanja udruženih aneurizmi.

Ključne reči:
aneurizma, ruptura; a. cerebri anterior; endovaskularne procedure; aneurizma, intrakranijalna; mikrohirurgija; a. cerebri media; neurohirurške procedure; lečenje ishod.

Introduction

Distal anterior cerebral artery (DACA) aneurysms, also known as pericallosal aneurysms are rare, and account for approximately 2%–9% of all ruptured intracranial aneurysms [1–4]. Studies have previously shown association of these aneurysms with multiple intracranial aneurysms disease, with multiple aneurysms presence in 55% of cases [4–6]. Several smaller series of DACA aneurysms indicated the frequency of bilateral aneurysms in 10%–20% of cases [1, 7], while mirror positioned DACA aneurysms are extremely rare [8–10]. DACA aneurysms are frequently associated with congenital anomalies and anatomic variations of DACA [8], although, there are reports of patients with DACA mirror aneurysms without any other vascular variation [9].

Typically, DACA aneurysms are small in size, with a wide neck, and with branches originating from the neck or fundus of the aneurysm [11]. The pericallosal-callosomarginal bifurcation is the most common location of DACA aneurysms [12, 13]. These aneurysms have high tendency for rupture (PHASES score is always > 4) [6, 14, 15]. In most of ruptures (67%–90%), DACA aneurysms were less than 7 mm in diameter [6, 16]. In more than a half of patients with the DACA aneurysm rupture, imaging reveals intracerebral hematoma (ICH), which is much more frequent then in other ruptured aneurysms (53%–73% vs. 26%) [16, 17]. Treatment options available include endovascular coiling, surgical clipping or by-pass surgery, which is the treatment of choice only in complex cases [4, 17–20].

We presented a case of surgically treated unruptured mirror aneurysms of DACA, accidentally seen during previous endovascular treatment after middle cerebral artery (MCA) aneurysm rupture.

Case report

Six months before admission to our department, a 49-year-old female patient was treated endovascularly in other institution due to MCA aneurysm rupture manifested with subarachnoid hemorrhage. Digital subtraction angiography (DSA), performed in the course, confirmed the existence of bilobular right MCA aneurysm, and also revealed two small bilateral aneurysms at the distal segments of anterior cerebral artery (ACA), left one measuring 4.5 mm and the right one measuring 6 mm in size, with the aneurysmal neck width of 3 mm and 4 mm, respectively, without other vascular malformations revealed (Figure 1).

The decision was made by the interventional neuroradiologist only to treat the bleeding MCA aneurysm immediately, while both ACA aneurysms were deemed unsuitable for endovascular treatment at the given moment. The post-procedural period passed without any complications. Follow-up multislice computed tomography (MSCT) angiography confirmed the existence of bilateral aneurysms on DACA segments one more time, as well as complete occlusion of the right MCA aneurysm (Figure 2).

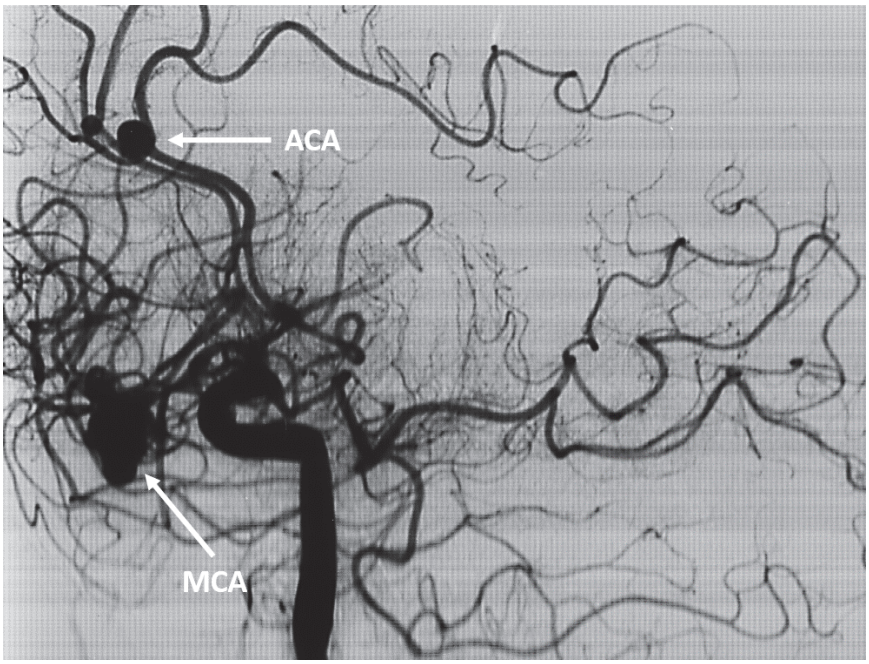

Fig. 1 – Digital subtraction angiography examination revealed middle cerebral artery bilobular aneurysm associated with two small bilateral aneurysms on the distal anterior cerebral artery segments.

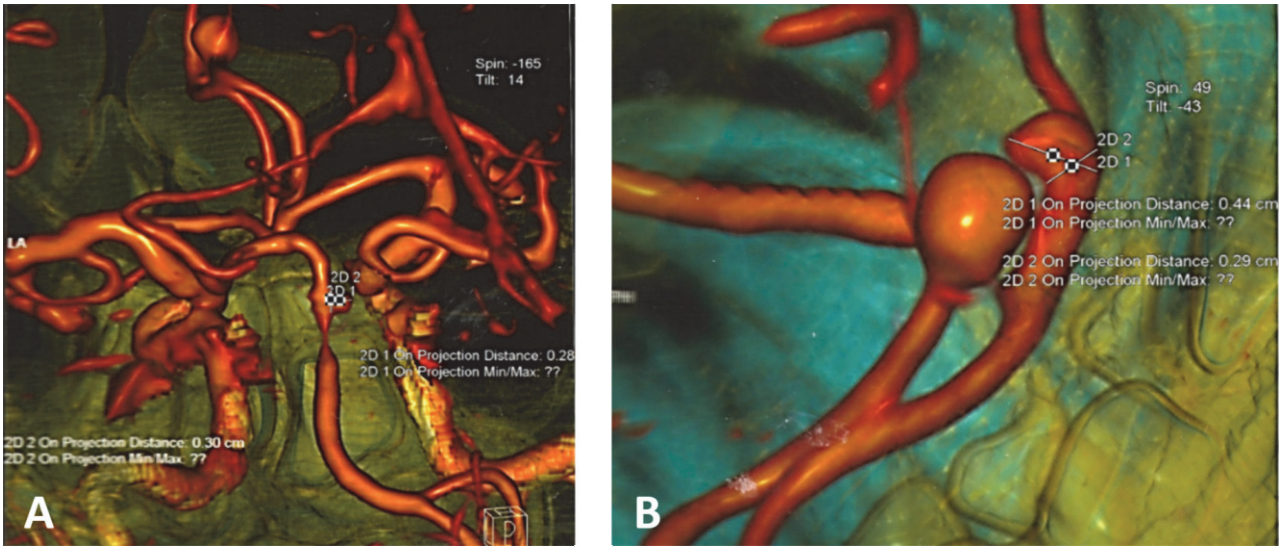

Fig. 2 – Multislice computed tomography angiography before the surgical procedure showed mirror anterior cerebral arteries aneurysms: A) anterior view; B) lateral magnified view with the smaller aneurysm measurement.

The patient was referred to our department six months later, and it was decided to perform microsurgical occlusion of the remaining ACA aneurysms. Unilateral interhemispheric approach was chosen to reach the distal ACAs. Retraction brain injury was prevented by evacuation of about 15 mL of cerebrospinal fluid by lumbar puncture preoperatively. Proximal pericallosal ACA segments were identified and then bilateral aneurysms at pericallosal-callosomarginal junction. Both aneurysms were clipped and excluded from the circulation completely, also major draining veins were preserved.

Postoperative course went well and the patient was discharged from our Department on the seventh postoperative day without any neurological deficit. Two months after the surgery, follow-up MSCT angiography revealed that all three aneurysms were completely excluded from circulation.

Discussion

Management of DACA aneurysms is a surgical challenge, therefore it is controversial whether these should be surgically clipped or coiled endovascularly, especially in cases when these are incidentally seen during the endovascular procedure for other aneurysm embolization, when a same-stage, endovascular coiling might look like a perfect approach to occlude mirror DACA aneurysms.

Although endovascular coiling is less invasive, and considered less harmful for the patient, it is associated with significantly higher periprocedural rupture [18] and procedure-related morbidity [21] than other circle of Willis aneurysms. Surgical clipping results, on the other hand, are same or slightly better than for aneurysms at other locations [16].

DACA aneurysms are still treated with microsurgical clipping more often than endovascular coiling due to their distal location and morphologic features, nevertheless surgical clipping remains demanding. Moreover, because of their rare occurrence, neurosurgeons often have the lack of experience in surgical treatment of these aneurysms [5, 7, 11, 16].

Non-experienced surgeons are avoiding to operate due to location of the DACA aneurysms in the narrow interhemispheric space [22], difficulties in establishing proximal control, and the high frequency of wide-necked and sclerotic aneurysms in this location, in particular those involving the origin of the branching arteries [4, 21].

Regarding the aneurysm size, only a few cases of a large and giant DACA aneurysms have been reported [5, 6, 14]. Average diameter at the moment of the rupture according to Gherasim et al. [19] was 5.5 mm vs. 9 mm compared with all other intracranial aneurysms which can be explained due to the lack of resistant arachnoid membranes at the level of the pericallosal cisterns. In our case, aneurysms at the distal segments of ACA, were measuring 4.5 mm on the left, and the right one measuring 6 mm in size, therefore demanding prompt surgical treatment.

According to meta-analysis of Petr et al. [23], aneurysm recurrence occurred in 3% after surgery and in 19.1% after endovascular treatment, although, in this series, there were no significant differences in procedure-related morbidity and mortality. The most important factor affecting the mortality and morbidity is the presence of associated aneurysms [5]. One stage surgery with unilateral craniotomy is suggested for bilateral DACA and mirror aneurysms, which is relatively straightforward due to their proximity [4], but also in cases when DACA aneurysms were associated with aneurysms at different location to reduce the morbidity and mortality [24, 25].

Initial haemorrhagic event related to DACA aneurysm rupture in more than a half of patients is ICH, which is considered to increase the risk of poor (lethal) outcome [26, 27]. The high incidence of ICHs, higher than for aneurysms elsewhere, is obviously related to the narrow pericallosal cistern and the dense attachments to the adjacent brain surface [26, 28]. Intraventricular hemorrhage is a little less frequent, appearing in 25%–30% of the patients [28]. This fact is also supporting our decision to proceed with surgical clipping, due to the possibility of immediate management of the intraoperative/intraprocedural rupture. Also, the

risk of ischemic event is better handled, due to better intraoperative overview and handling of the small branches originating from the aneurysm dome.

Bearing in mind the tendency for rupture regardless of small aneurysm size, high incidence of intracerebral hemorrhage, and a relatively high risk of aneurysm recurrence after endovascular treatment at this location [11, 16, 23, 26], we believe that both interventional radiologists and our decision for subsequent early microsurgical treatment was justified.

All patients with DACA aneurysms should be carefully evaluated with DSA or MSCT angiography for the presence of additional aneurysms due to the tendency for multiplicity [5, 26]. Even when DACA aneurysms were revealed during an endovascular procedure, surgical treatment should be undertaken as soon as possible [21, 26]. More than one aneurysm should not be treated in the same procedure. They should be aggressively treated even if they are very small because of their tendency to early rupture [15].

Conclusion

Successful surgical management of DACA aneurysms mostly depends on understanding of their unique microsurgical anatomy and the surgeon's experience, as well as careful preparation and examination of the patient.

Sufficient brain relaxation, accurate localization of the aneurysm, early identification of the proximal ACA segment, and preservation of the major draining veins remain necessary for a safe surgery.

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