Case Report

A rare anomaly of the anterior communicating artery complex hidden by a large broad-neck aneurysm and disclosed by three-dimensional rotational angiography

Jean G. de Oliveira¹, R. du Mesnil de Rochemont², J. Beck¹, E. Hermann¹,
R. Gerlach¹, A. Raabe¹, V. Seifert¹

¹ Neurocenter, Department of Neurosurgery, Johann Wolfgang Goethe-University, Frankfurt am Main, Germany

² Neurocenter, Institute of Neuroradiology, Johann Wolfgang Goethe-University, Frankfurt am Main, Germany

Received 1 May 2007; Accepted 11 September 2007; Published online 14 January 2008

© Springer-Verlag 2008

Summary

Double fenestration of the anterior communicating artery (ACoA) complex associated with an aneurysm is a very rare finding and is usually caused by ACoA duplication and the presence of a median artery of the corpus callosum (MACC). We present a patient in whom double fenestration was not associated with ACoA duplication or even with MACC, representing therefore, a previously unreported anatomic variation. A 43 year old woman experienced sudden headache and the CT scans showed subarachnoid haemorrhage (SAH). On admission, her clinical condition was consistent with Hunt and Hess grade II. Conventional digital subtraction angiography (DSA) was performed and revealed multiple intracranial aneurysms arising from both middle cerebral arteries (MCA) and from the ACoA. Three-dimensional rotational angiography (3D-RA) disclosed a double fenestration of the ACoA complex which was missed by DSA. The patient underwent a classic pterional approach in order to achieve occlusion of both left MCA and ACoA aneurysms by surgical clipping. The post-operative period was uneventful. A rare anatomical variation characterised by a double fenestration not associated with ACoA duplication or MACC is described. The DSA images missed the

double fenestration which was disclosed by 3D-RA, indicating the importance of 3D-RA in the diagnosis and surgical planning of intracranial aneurysms.

Keywords: Anatomical variation; aneurysm; anomaly; anterior communicating artery; double fenestration; three-dimensional angiography.

Introduction

Vascular anomalies of the Circle of Willis are commonly associated with aneurysm formation [1, 6, 8, 10]. The anterior communicating artery (ACoA) complex is a frequent site of arterial duplications and fenestrations, mainly when associated with aneurysms [10]. However, double fenestration of the ACoA complex is a rare finding, angiographically and surgically, which is normally caused by ACoA duplication and occurrence of an accessory anterior cerebral artery, also called the median artery of the corpus callosum (MACC) [8].

Although digital subtraction angiography (DSA) remains the gold standard method for the diagnosis of intracranial aneurysms, recent studies have shown excellent results with regard to detailed anatomical vessel information provided by three-dimensional rotational angiography (3D-RA) [2, 12].

This is, to the best of our knowledge, the first description of a double fenestration in the ACoA complex

Correspondence: Jean Gonçalves de Oliveira, M.D., Rua Sena Madureira, 1123 Apto. 61, 04021-051 São Paulo – SP, Brazil.
e-mail: jeangol@uol.com.br

associated with a broad-neck multilobulated ACoA aneurysm, not related to ACoA duplication or MACC, and DSA images failed to demonstrate the double fenestration.

Clinical details

History and examination

A 43 year old woman was referred to our department because she experienced a sudden headache. Computed tomography (CT) scans confirmed aneurysmal subarachnoid haemorrhage (SAH). On admission, her clinical condition was consistent with Hunt and Hess grade II. Computerised tomography (CT) showed SAH Fisher grade III (Fig. 1). Conventional DSA was performed and revealed multiple intracranial aneurysms arising from both middle cerebral arteries (MCA) and the ACoA

(Fig. 2). Three-dimensional rotational angiography (3D-RA) disclosed a double fenestration at the level of ACoA complex which was missed by DSA. In addition, anatomical details of the aneurysm like a broad-neck involving the ACoA and left A2 as well as multilobulation were also demonstrated (Fig. 3).

Treatment and postoperative course

Because of the complex anatomical characteristics of the aneurysms, surgical clipping was preferred over endovascular treatment. From the CT scans it was not clear as to which aneurysm had ruptured causing the subarachnoid haemorrhage. Therefore, the patient underwent a classic pterional approach in order to achieve occlusion of both left MCA and ACoA aneurysms by surgical clipping. The partially thrombosed MCA aneurysm was clipped first. After a complete dissection of the

Fig. 1. CT scan images showing subarachnoid haemorrhage Fisher grade III

Fig. 2. DSA images ((A) right ICA injection; (B) left ICA injection) demonstrating bilateral MCA aneurysms, ACoA aneurysm and hypoplasia of the right A1 segment

ACoA complex, the surgical findings were compatible with the 3D-RA results. However, in the ACoA aneurysm, the clip application was complicated by the wall

irregularities, broad-neck and association with the double fenestration, as well as the involvement of the ipsilateral A2 in the base of the aneurysm. Therefore, during

Fig. 3. (A and B) 3D-RA disclosing a double fenestration associated with a multilobulated ACoA aneurysm (views from below and above, respectively). (C and D) Anatomic details of the aneurysm with involvement of ACoA and left A2 by the broad-neck (lateral and back views, respectively)

Fig. 4. Post-operative DSA images ((A) AP view; (B) lateral view) demonstrating the residual ACoA aneurysm

the clipping application, we had to reconstruct the ACoA, to preserve the parent and branching arteries, as well as to occlude the aneurysm. Consequently, although intraoperative video angiography using indocyanine green confirmed absence of filling in both aneurysms with maintenance of regular perfusion in the parent and branching vessels, an expected small remnant at the broad-base ACoA aneurysm was observed on the post-operative DSA (Fig. 4). This remnant neck was wrapped. The post-operative period was uneventful.

Discussion

Double fenestration of the ACoA complex

Anomalies or variations of anatomy in intracranial arteries are frequently described [6, 10]. The most common variations of the ACoA complex are: unilateral anterior cerebral artery (ACA) hypoplasia, multiple vascular

channels, dimple, fenestration, duplication, fusion, MACC and azygous ACA [6, 11].

The incidence of fenestrations in the ACoA complex ranges between 5.7 and 13% of patients with aneurysms arising from the same segment [3, 6].

The genesis of the ACoA complex occurs around Day-35 in human embryos and at the early stages the arteries are relatively primitive and plexiform [9]. Incomplete fusion of the plexiform anastomosis may result in fenestrations or duplications [1, 10, 13].

Haemodynamic abnormalities resulting from these anomalies associated with a medial defect that is commonly present in the proximal end of a fenestration, can explain the commonly seen association of aneurysms in these sites [3–5, 7].

Whereas an ACoA aneurysm associated with fenestration in this segment is a not such a rare situation, the occurrence of double fenestration is extremely rare. Namiki and Doumoto [8] published a report of a double

Fig. 5. Schematic drawings of the ACoA complex. (A) ACoA aneurysm with the usual anatomy. (B) ACoA aneurysm associated with a double fenestration caused by ACoA duplication as well as the presence of a MACC. (C) The unusual anatomic variation presented, composed by hypoplasia of the right A1, both A2 vessels arising from the left A1 with fenestrations, and a broad-neck multilobulated aneurysm which assimilated the ACoA

fenestration associated with an ACoA aneurysm and stated that it was never described previously. In their patient, the fenestration resulted from the presence of a MACC and ACoA duplication. Nevertheless, this same finding was previously published by Kwak *et al.* [6] when they described twenty-six different patterns of ACoA aneurysms and anatomical variations in this region.

Our patient, however, is different from all others published up to now, since there was no MACC or ACoA duplication. Instead, it seemed to be an association of anatomical anomalies that included right A1 hypoplasia, fenestrations at the origin of both A2, and a broad-neck aneurysm that completely assimilated the ACoA (Fig. 5).

Diagnosis and surgical planning

Conventional DSA is the method widely used for diagnosis and treatment of intracranial aneurysms. However, the two dimensional images provided by this method sometimes makes the analysis of overlapping vessels difficult [12].

Consequently, anatomical anomalies or even details about size, shape and irregularities of the aneurysm are frequently disclosed only during the neurosurgical procedure. As such, a very detailed angiographic information is required for surgical planning of intracranial aneurysms, especially those located at the ACoA complex, where the recognition of, at least, five arteries is always necessary.

Based on three dimensional images, linked to multiplanar reconstruction, recent studies have shown that 3D-RA provides more detailed information for evaluation of the relevant aneurysm anatomy when compared with conventional 2D-DSA [2, 12].

We have used 3D-RA as a routine diagnostic procedure and it has brought a great improvement to our surgical planning for intracranial aneurysms. The clinical example presented was technically difficult not only because of the complicated anatomy, but more so by the aneurysm shape which assimilated the ACoA almost completely as well as the origin of left A2, making clip application extremely difficult. We believe that previous recognition of the anatomical variation and morphology of the aneurysm makes surgery safer and reduces the risk of an adverse outcome.

It is important to note that, because we perform 3D-RA routinely, only standard AP and lateral images are acquired by conventional 2D-DSA. Therefore, if oblique

series had been acquired, they could have disclosed the double fenestration.

Conclusions

Although vascular fenestration associated with aneurysms of the ACoA is not an uncommon finding, the occurrence of double fenestration, not associated with ACoA duplication or with MACC, is a rare anatomic anomaly that might be present.

We describe a patient with multiple intracranial aneurysms, where this anatomic variation was hidden by a broad-neck ACoA aneurysm which standard DSA images (AP and lateral) were not able to demonstrate. The information provided by 3D-RA not only disclosed this anomaly, but was also extremely helpful for surgical planning.

Acknowledgment

During this work, Jean G. de Oliveira, M.D., was a recipient of the International Fellowship Award in the Interchange Program between the Brazilian Society of Neurosurgery and the German Society of Neurosurgery for 2006.

References

1. Alpers BJ, Berry RG (1963) Circle of Willis in cerebral vascular disorders. The anatomical structure. *Arch Neurol* 8: 398–402
2. Beck J, Rohde S, Berkefeld J, Seifert V, Raabe A (2006) Size and location of ruptured and unruptured intracranial aneurysms measured by 3-dimensional rotational angiography. *Surg Neurol* 65: 18–25
3. Crompton MR (1966) The pathogenesis of cerebral aneurysms. *Brain* 89: 797–814
4. Ferguson GG (1972) Physical factors in the initiation, growth, and rupture of human intracranial saccular aneurysms. *J Neurosurg* 37: 666–677
5. Hashimoto N, Handa H, Nagata I, Hazama F (1980) Experimentally induced cerebral aneurysms in rats: Part V. Relation of haemodynamics in the circle of Willis to formation of aneurysms. *Surg Neurol* 13: 41–45
6. Kwak R, Niizuma H, Hatanaka M, Suzuki J (1980) Anterior communicating artery aneurysms with associated anomalies. *J Neurosurg* 52: 162–164
7. Nakasu Y, Nakasu S, Kidooka M, Handa J (1982) Aneurysm at the fenestration of basilar artery – case report. *Nippon Geka Hokan* 51: 344–348
8. Namiki J, Doumoto Y (2003) Microsurgically critical anomaly of the anterior communicating artery complex during the pterional approach to a ruptured aneurysm: double fenestration of the proximal A2 segments. *Neurol Med Chir (Tokyo)* 43: 304–307
9. Padgett DH (1948) The development of the cranial arteries in the human embryo. *Contrib Embryol* 32: 205–262
10. San-Galli F, Leman C, Kien P, Khazaal J, Phillips SD, Guerin J (1992) Cerebral arterial fenestrations associated with intracranial saccular aneurysms. *Neurosurgery* 30: 279–283
11. Serizawa T, Saeki N, Yamaura A (1997) Microsurgical anatomy and clinical significance of the anterior communicating artery and its perforating branches. *Neurosurgery* 40: 1211–1216

12. Sugahara T, Korogi Y, Nakashima K, Hamatake S, Honda S, Takahashi M (2002) Comparison of 2D and 3D digital subtraction angiography in evaluation of intracranial aneurysms. *Am J Neuroradiol* 23: 1545–1552
13. Teal JS, Rumbaugh CL, Bergeron RT, Segall HD (1973) Angiographic demonstration of fenestrations of the intradural intracranial arteries. *Radiology* 106: 123–126

Comment

The purpose of this paper is to present a rare case of an anomaly of the anterior communicating artery complex hidden by a large broad-neck

aneurysm. The anomaly was disclosed preoperatively by three-dimensional rotational angiography.

A lot of anomalies including various fenestrations of the AcoA complex have been described, some of them being surgical descriptions only, because these anomalies have not been detected preoperatively. The authors add a very rare case of a double fenestration of the AcoA with a broad neck aneurysm causing a difficult anatomical situation, which was preoperatively resolved by three-dimensional angiography.

The paper illustrates the advantage of 3D-angio in preoperative planning in cases of rare anomalies.

Wolfgang Deinsberger
Kassel