

PROGRESSION OF AN ARTERIAL INFUNDIBULUM TO ANEURYSM

Case report

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ABSTRACT - In this case an aneurysm of the right posterior communicating artery developed 11 months after an infundibular dilation of this artery had been angiographically and surgically demonstrated. In the best of the authors' knowledge, there are only eleven such cases reported in the literature. This report brings about diagnostic and therapeutic questions regarding arterial infundibula and the need of a better understanding of those lesions.

KEY WORDS: aneurysm, arterial infundibulum, bilateral supratentorial aneurysms, infundibular dilation.

Progressão de infundíbulo arterial a aneurisma: relato de caso

RESUMO - Relatamos a transformação de uma dilatação infundibular da origem da artéria comunicante posterior em aneurisma, após onze meses do diagnóstico angiográfico e cirúrgico. Apenas onze casos semelhantes foram descritos na literatura internacional. Relatos como esse trazem à tona os problemas diagnósticos e terapêuticos representados por essas lesões e suscitam a necessidade do melhor entendimento das dilatações infundibulares encontradas nas artérias cerebrais.

PALAVRAS-CHAVE: aneurismas, aneurismas bilaterais, dilatação infundibular, infundíbulo.

Infundibula (IFs) are funnel-shaped symmetrical enlargements of the origin of cerebral arteries. Most frequently they affect the origin of the posterior communicating artery (PCoMA) at its junction with the internal carotid artery (ICA), and are considered as normal anatomical variants devoid of pathogenic significance. Some authors do not agree with this statement and consider an IF a "pre-aneurysmal" lesion¹⁻⁴. This belief rests on the increasing incidence of infundibular widening with age and on the histological demonstration of changes in some IFs similar to that characteristic of saccular aneurysms¹.

We report the progression of an IF to aneurysm, with subsequent rupture, in a hypertensive patient previously operated of multiple and bilateral aneurysms, through a unilateral craniotomy.

CASE

A 53-year-old caucasian woman was admitted on March 30, 1998, complaining of a sudden bitemporal headache, started 12 hours before admission. She had no

loss of consciousness or vomiting. She scored 15 on GCE, but had a marked stiff neck. A CT performed on admission revealed widespread subarachnoid haemorrhage (SAH), mainly in the left Sylvian fissure. Five days later, the patient developed a complete III nerve palsy on the left side. Angiography revealed aneurysms on the origin of the left PCoMA, on the bifurcation of the middle cerebral artery and ICA bifurcation on the right side and an infundibular origin of the right PCoMA (Fig 1). She was submitted to a left pterional craniotomy and microsurgical clipping of the lesions in both sides (unilateral craniotomy to bilateral aneurysms) (Fig 2). The origin of the right PCoMA was explored and the presence of the IF confirmed. The postoperative period was unremarkable and the control angiogram performed on May 15, 1998 revealed no aneurysms. The follow up visits during the next nine months confirmed a good control of the mild hypertension (diagnosed during admission), no evidence of aortic coarctation, polycystic disease or inflammatory connective tissue disease, and a partial recovery of the left third nerve palsy - the residual deficit only seen during the upward gaze (deficit of the superior division of the oculomotor nerve).

On April 6, 1999, the patient was readmitted complain-

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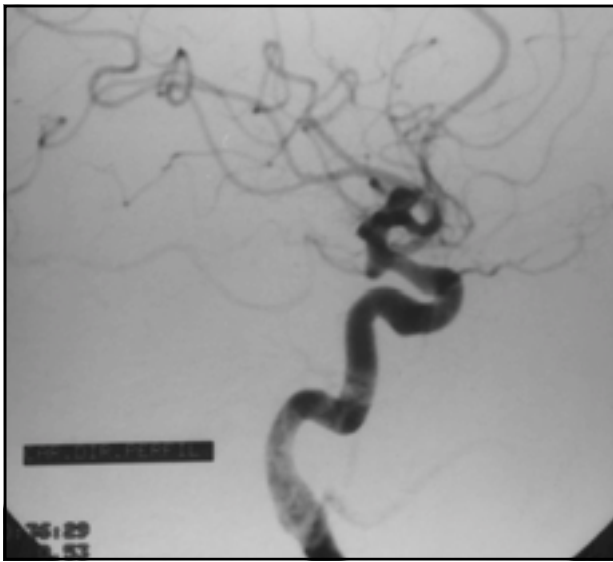


Fig 1. Diagnostic angiogram (profile) showing infundibular origin of right PComA.



Fig 2. Postoperative angiogram (AP) showing clips occluding aneurysms on the left PComA, right ICA and MCA, through an unilateral craniotomy.

ning of a "headache just like the other time". This event had taken place one week before and had been initially neglected by the patient until her eye became involved. On admission there was a heavy stiff neck and a 24-hour history of right ptosis. The CT performed at that time (nine days after the beginning of the complaint) failed to demonstrate HSA. The angiography disclosed an aneurysm at the origin of the right PComA (Fig 3). A right pterional craniotomy was performed and the aneurysm clipped. The microdissection on the right Sylvian fissure was accomplished uneventfully, but there were hard adherences on the lamina terminalis and right carotid cisterns. A control angiogram obtained on July 4, 1999 demonstrated complete occlusion of the lesion.

DISCUSSION

IFs are apparent on 7 to 25% of otherwise normal angiograms, and the incidence seems to be greater in cases of familial or multiple aneurysms³. Angiographically, IFs are triangular shaped lesions, which base does not measure more than 3 mm and the branch artery arises from its apex². An IF thus should appear as a symmetric bulge without a neck in marked contrast to an intracranial aneurysm, which bulges, asymmetrically from a well-defined neck³. Although this guidelines help in most cases, differentiation of aneurysms from infundibular dilations of the PComA remains a difficult radiological problem^{1,2,5}, especially when considering patients with subarachnoid haemorrhage and no other angiographic abnormality or patients with multiple aneurysms. According to Marshmann et al.², it may be unwise to report "negative" angiographic results in cases of SAH in which IFs are present.

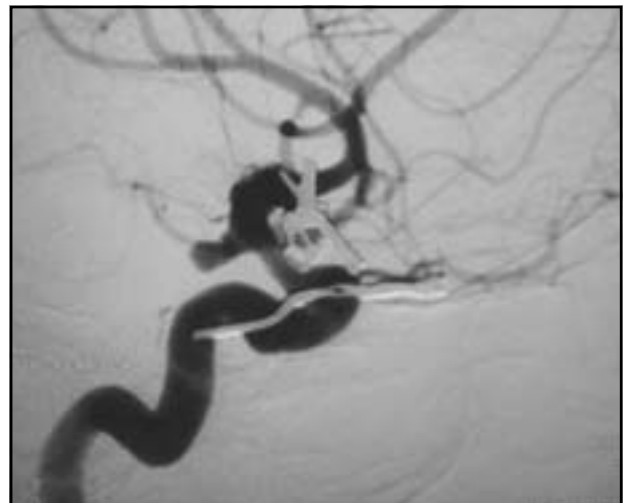


Fig 3. Diagnostic angiogram (profile) showing progression from infundibulum to aneurysm eleven month after the first angiogram (Fig 1).

Histologically, some IFs show medial defects, which are also present on normal artery bifurcation and aneurysms. Some authors believe that these defects occur first and allow elastica defects to develop from overstretching when hemodynamic factors such as ligation of one carotid artery or hypertension are added^{1,3}. The IF configuration itself could be hydraulically more disposed toward further dilation. The sudden decrease in velocity and kinetic energy at IFs results in increased wall tensions¹. This view can be supported by the radiological observation that a well-developed PComA could be a factor contributing to the progression of an IF to aneurysm⁵.

Our patient had one hemodynamic factor removed (treated hypertension) but remains to be understood if the effects of the presence of multiple aneurysms, or the results of their clipping could have contributed to the progression of the IF to aneurysm. This question is even more important when considering the time of progression in this patient: eleven months, in contrast with a mean period of seven years as described in literature¹⁻⁴.

Considering the pre-aneurysmal role that IF could play, some authors have advocated IF wrapping at craniotomy in precisely such cases³. Following this view imply that, added to the other benefits provided by the contralateral approach, this patient might have been spared of additional trouble. The question however, is not that simple: the co-operative study findings indicating that no signs of rupture were found in any bulging less than 3mm in size suggest that this procedure could be excessive^{3,6}. A systematic follow up, with angiography, especially in young patients who have IFs and multiple aneurysms is the recommendation of many authors^{1,3,7}. These reports, however, do not suggest any specific follow up times.

Based on the experience with this case, the literature data, the risks of the angiography itself and costs, the management of patients harbouring infundibular bulges at the Department of Neurosurgery at Hospital da Restauração include a careful watch of the cases and, in those patients in whom a high risk of progression would be foreseen, an annually based angiography is recommended. In centers where computer tomographic angiography is available, this relatively non-invasive method, which provides detailed information about vascular lesions' shape and direction as well as its relationship with adjacent structures⁸ may be used, with an even smaller interval, to follow these patients. It should be stated, though, that this method is still being perfected and few reports of its use to infundibular dilations have been made⁹.

Considering patients with multiple and bilateral aneurysms, which account for 32% of all patients

treated in our department annually¹⁰, a single craniotomy has the advantage of avoiding a second approach and anaesthesia, and reducing the risks of rebleeding¹¹, especially when an acute surgery is not possible. This technique, however, is not recommended for all the patients. The individual anatomy and clinical setting are the factors that have a major influence on the success¹². In our case the unilateral approach gave us an unexpected advantage, as say, not only the possibility of clipping all aneurysms through the same operative approach, but above all, the opportunity of inspecting the origin of the right PcomA and being able to confirm the progression of IF to aneurysm, co-validating, thus, this report.

REFERENCES

1. Stuntz JT, Ojemann GA, Alvord EC. Radiographic and histologic demonstration of an aneurysm developing of the infundibulum of the posterior communicating artery: case report. *J Neurosurg* 1970;33:591-595.
2. Archer CR, Silbert S. Infundibula may be clinically significant. *Neuroradiology*, 1978;15:247-251.
3. Marshman LAG, Ward PJ, Walter PH, Dossetor RS. The progression of an infundibulum to aneurysm formation and rupture: case report and literature review. *Neurosurgery* 1981;43:1445-1449.
4. Trassi S, Vincent LM, Zingesser LH. Development of aneurysm from infundibulum of posterior communicating artery with documentation of prior hemorrhage. *AJNR* 1981;2:368-370.
5. Endo S, Furuichi S, Takaba M, Hirashima Y, Nishijima M, Takaku A. Clinical study of enlarged infundibular dilation of the origin of the posterior communicating artery. *J Neurosurg* 1995;83:421-425.
6. Locksley HB: Report on the co-operative study of intracranial aneurysms and subarachnoid haemorrhage: section V-part II. Natural history of subarachnoid haemorrhage, intracranial aneurysms and arteriovenous malformations-based on 6368 cases in the co-operative study. *J Neurosurgery* 1966;25:321-368.
7. Itakura T, Fuminori O, Nakai E, Fujii T, Hayashi S, Komai N. Bilateral aneurysm formation developing from junctional dilation (infundibulum) of the posterior communicating arteries: case report. *J Neurosurg* 1983;58:117-119.
8. Sekhar LN, Kalia KK, Yonas H, et al. Cranial base approaches to intracranial aneurysms in the subarachnoid space. *Neurosurgery* 1994; 3:472-483.
9. Ng SH, Wong HF, Ko SF, et al. CT angiography of intracranial aneurysms: advantages and pitfalls. *Eur J Radiol* 1997;25:14-19.
10. Martins C, Lay J. Estudo das hemorragias aracnoideas aneurismáticas: revisão da literatura e análise dos casos atendidos no Hospital da Restauração no ano 2000. Monografia de Conclusão da Residência Médica em Neurocirurgia. Recife, 2001.
11. Oliveira E, Tedeschi H, Siqueira M, et al. Anatomical and technical aspects of the contralateral approach for multiple aneurysms. *Acta Neurochir (Wien)* 1996;138:1-11.
12. Oshiro EM, Rini DA, Tamargo RJ. Contralateral approaches to bilateral cerebral aneurysms: a microsurgical anatomical study. *J Neurosurg* 1997;87:163-169.