Treatment of endovascular aneurysms associated with fenestration of the basilar artery. A case report

Jorge Santos Franco, Leonardo Álvarez Vázquez, Rafael Saavedra Andrade and Miguel Antonio Sandoval Balanzario*

Neurosurgery Department, Specialties Hospital, Centro Médico Nacional "La Raza", México, D.F.

Abstract

Basilar artery fenestration is a rare anatomical variant that may be accompanied by saccular aneurysms; however, this association is uncommon. We report the case of a female patient who presented with a subarachnoid hemorrhage that was successfully treated with coil embolization and make a brief literature review. (Gac Med Mex. 2014;150:559-63)

Corresponding author: *Jorge Santos Franco, jorge_santos@unam.mx*

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Introduction

Basilar artery fenestration (BAF) is an anatomical variant observed in 0.6% of cerebral angiographies and in up to 5% of necropsies (Fig. 1)[1-3]. It may be accompanied by saccular aneurysms; however, this association is uncommon and is reported in less than 10% of cases[4]. We describe a case presenting with subarachnoid hemorrhage (SAH) that was succesfully treated by means of embolization with platinum coils.

Clinical case

Thirty-seven year-old woman presenting with sudden-onset severe headache and loss of alertness for 1 min with complete recovery. SAH diagnosis was made: Grade II in Hunt and Hess scale and grade IV in the Fisher scale (Figs. 2 A and B). Angiotomography (AT) and digital subtraction angiography (DSA) showed a large BAF at the proximal third associated with a saccular aneurysm (Figs. 1 C-D). Endovascular treatment was programmed three days afte the stroke.

Correspondence:
*Jorge Santos Franco
Servicio de Neurocirugía
Hospital de Especialidades
Centro Médico Nacional "La Raza"
Seris, s/n y Zaachila
Col. La Raza, Del. Azcapotzalco, C.P. 02990, México, D.F.
E-mail: jorge_santos@unam.mx

Under general anesthesia and using the Seldinger technique, right vertebral artery was catheterized with a 6F guiding catheter, and the aneurysm was immediately cathererized with microcatheter. The aneurysm was embolized with seven detachable platinum coils, leaving a small residual of the aneurysm (Figs. 3 A and B). The patient awakened without deficit and was discharged with no eventualities at the fifth day. The control DSA at six months showed complete occlusion of the aneurysm, with adequate arterial flow at the level of the arteries comprising the BAF (Figs. 3 C-E).

Discussion

A fenestration is defined as a single artery with two luminal canals that may or may not share their adventitious layer[5]. It is difficult quantifying its frequency in intracranial arteries; however, long time ago, Sanders et al.[6] demonstrated a frequency of 0.72% in a large series of cerebral angiographies. Basilar artery is formed by the fusion of bilateral longitudinal neural arteries at the fifth week of gestation[7]. At first stages of fusion, these arteries are connected by several bridging zones. If the fusion is not produced, it results in duplication; whereas if these bridges persist, BAF will occur. Its frequency is 1.3-5.3% in necropsies and 0.02-0.6% in DSA[3,8]. Islak et al.[9] reported a review of 2,000 DSAs where they found 20 BAF cases. Currently, TA and

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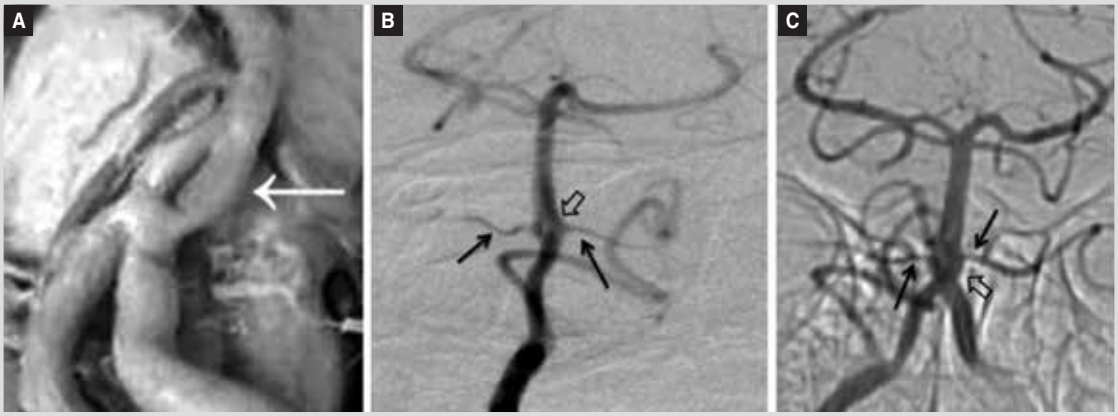

Figure 1. ***A:*** *post-mortem specimen showing a BAF at the proximal third (arrow).* (Reproduced with permission of Santos-Franco et al.[8]). **B** *and* ***C:*** *right vertebral artery selective angiography left oblique (B) and anteroposterior (C) projections showing a small BAF at the proximal third (hollow arrow). Both anteroinferior cerebelous arteries can be seen to originate from each branch of the BAF.*

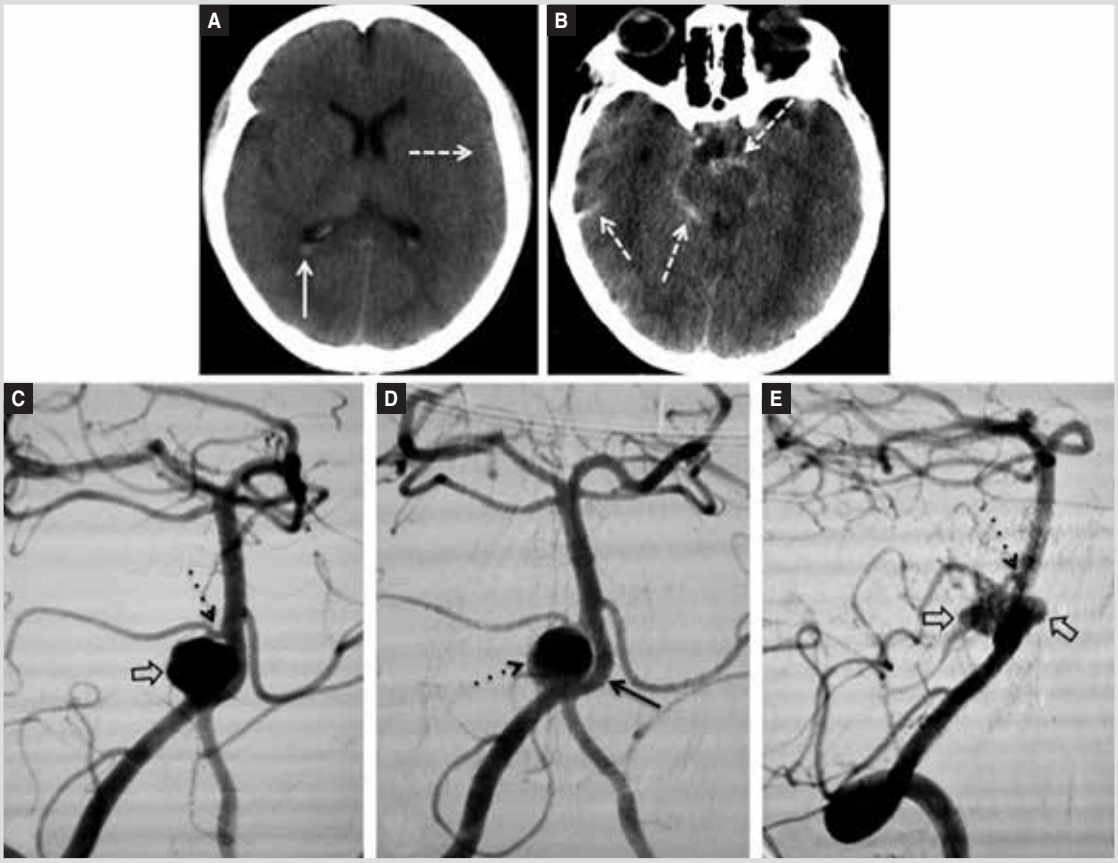

Figure 2. ***A*** *and* ***B:*** *simple tomography showing the SAH (discontinuous arrows) with higher density on the perimesencephalic cisterns. Additionally, ventricular system invasion is observed (arrow).* ***C-E:*** *right vertebral artery-selective SAH where an aneurysm feeding on a proximal third BAF (hollow arrow), which is larger at its anteroposterior diameter (hollow arrows in E), can be appreciated. The right branch of the fenestration (dotted arrow) is thinner than the left branch (arrow); however, it is evident that the right AICA originates in it.*

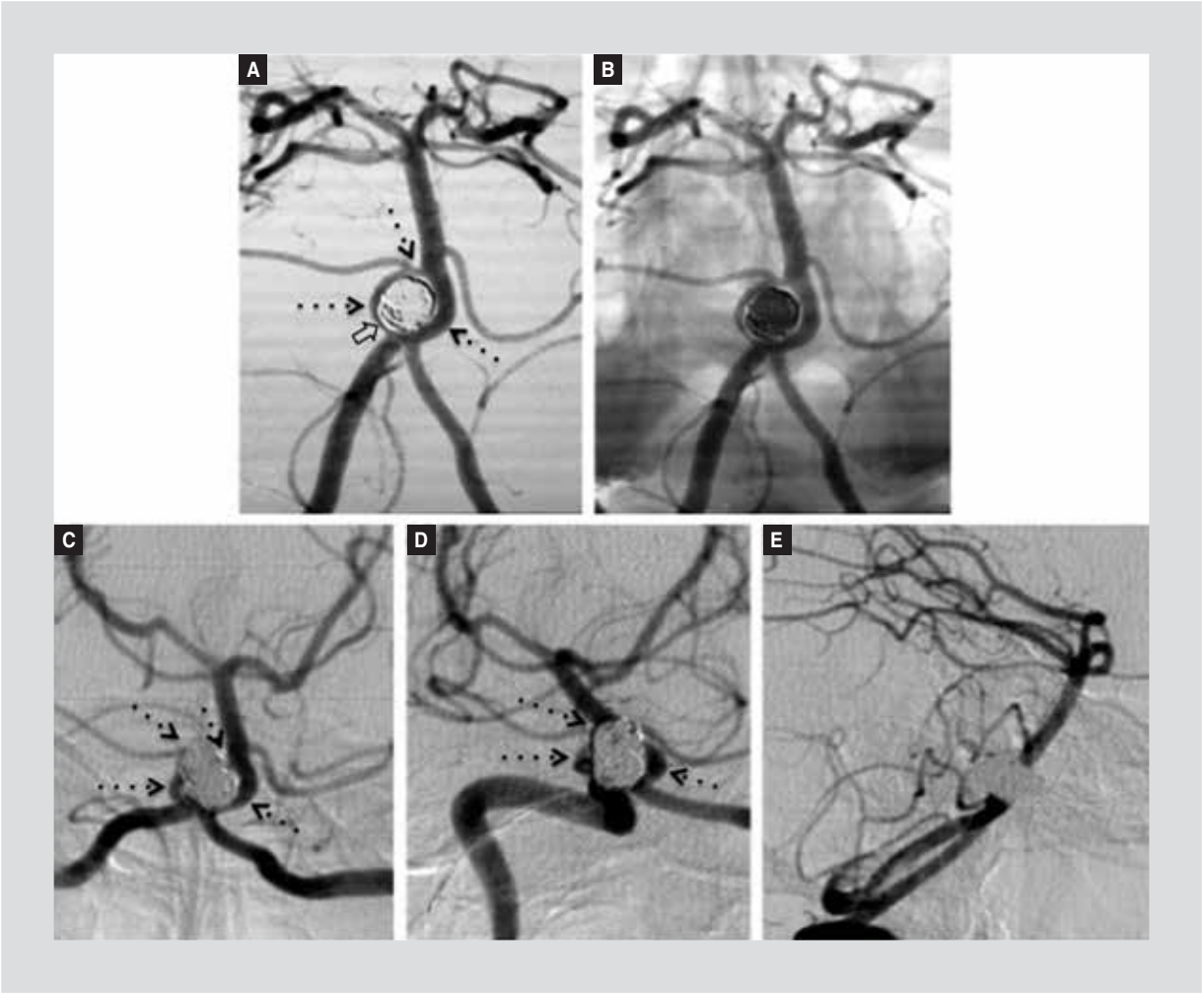

Figure 3. ***A*** *and* ***B:*** *DSA (A) and x-ray (B) controls immediately post-embolization with detachable coils, where it can be appreciated that the parental vessel was respected, including the components of the fenestration (dotted arrows). Residual filling of the aneurysm where the neck was found is clearly observed (hollow arrow).* ***C-E:*** *DSA at six months showing complete occlusion of the aneurysm and persistence of normal arterial flow in the parental vessel, including the components of the fenestration.*

angioresonance are excellent diagnostic methods[5,10,11]. In a recent study, BAFs were found in 2.33% of 5,657 TA images[10].

Basilar fenestration can occur at any segment of the artery; however, it is more common at its proximal third, generally in close proximity to the vertebrobasilar junction, and it is very uncommon on the medial and distal thirds[9,12-15]. BAF is classified according to its length as: a) small, with 0-3 mm; f) medium, from > 3.1 to 5 mm, and c) large, with more than 5 mm[12].

Lateral walls of the BAF have a normal intrinsic architecture; however, medial walls show structural defects at both ends of the fenestration with elastin discontinuity at the proximal end, in addition to subendothelial thinning[16]. These structural changes at the proximal end are consistent with those produced by hemodynamic stress, similar to those observed in cerebral arteries bifurcations and, therefore, consistent with the theory of intracranial aneurysms origin[1,5,12,17-19]. That is why the presence of aneurysms on this site is more frequent; however, in very few cases they can be found in the distal part.

Association between BAF and aneurysms has been reported to occur in 25-50% of cases[10,20]. Presence of two aneurysms has been reported in some patients[1,4,12,21,22]. Most aneurysms usually present with SAH, but a few can be incidental[1,12,20,22]. These aneurysms are complex due to their location and morphology and, therefore, surgical treatment is usually arduous due to the complex geometry of the fenestration and difficulties to obtain adequate surgical exposure, proximity to the brainstem and inferior cranial nerves

and presence of multiple perforating arteries, which can result in an inadequate clip clamping, associated with high morbidity and mortality[23].

Hoffman and Wilson[23] were the first to report a succesful case of clip clamping of an aneurysm of this type; however, the patient had craneal nerves VI, IX and X dysfunction for two weeks, with intermittent diplopia persisting for a prolonged period. In 1987, Campos et al.[22] reported on 20 cases treated using surgery, out of which only 17 could be clamped with clips and, of these, total occlusion was achieved only in 70%. During post-operative evolution, 65% of the patients showed cranial nerve paresis, one patient developed severe neurologic deficit and other died[22].

Endovascular therapy (ET) has been shown to be a safe and effective method in the management of intracranial aneurysms[19,24-28]. In a review by Itami et al.[29], 57 ET-treated BAF-associated aneurysm cases were collected. Based on this review and the one conducted by us, we can infere that ET has been useful in this particular type of aneurysms[1,4,9,12,20,29-43]. All reported cases were embolized with detachable platinum coils. Tasker and Byrne[4] reported six cases, out of which 61% of aneurysms were completely occluded. Graves et al.[30] described three cases where embolization with complete occlusion in two cases and incomplete in one was achieved. Islak et al.[9] describe 11 patients treated with embolization, out of which 91% were completely occluded, whereas a case of incomplete occlusion was subsequently successfully treated at a second moment. Recently, embolization with detachable coils using two microcatheters has been described[40].

Although complete embolization of the lesion is the primary objective, in the case we present here, a very small residual was left intentionally close to the aneurysm's neck. This decision was taken because during the infusion of the last detachable coil, protrusion into the parental vessel, as a result of the wide diameter of the aneurysm's neck, was observed. In a case described by Graves et al.[30] something similar happened. For such cases, Islak et al.[9] and Itami et al.[29] used the balloon-assisted embolization technique (remodelling) in 27 and 50% of cases, respectively. The display of one or two stents might ensure adequate compacting of the coils within the aneurysm; however, in our patient, we preferred avoiding this in order not to put her on a prolonged regimen with double oral antiaggregation agents, since she was at risk of developing hydrocephalus, which would have required surgical drainage.

It is important to preserve the branches that form the BAF, since pontine perforating bundles can emerge out of them or towards the vertebrobasilar junction, the occlusion of which might be accompanied by ischemia[8,44,45]. In our case, the anteroinferior cerebelous artery (AICA) originated in the right branch of the BAF, the occlusion of which might result in ischemia of the lateral portions of the brainstem, predominantly of the pons, the cerebral peduncle and right cerebelous hemisphere[46]. The degree of ischemic lesion and the symptoms are usually variable due to the diverse collaterality pattern of the AICA; however, the large diameter of the artery and its branching angiographic pattern led us to us expect an extensive ischemic lesion. Graves et al.[30] describe the complication with a thrombus on one of the branches of the BAF and on the apex of the basilar artery, which could be resolved with intra-arterial infusion of thrombolytic agents without any clinical consequence. Fujimoto et al.[20] reported one death; however, this was secondary to severe intracranial hypertension associated with the SAH.

In conclusion, aneurysms originating in BAF are infrequent and complex in their treatment. Surgery is usually risky, whereas ET offers good results with a direct, rapid and safe access. To our knowledge, this is the first formally reported case in Mexico and in Spanish-speaking Latin America.

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