Abstract

Background: Fusiform aneurysms are rare (<1%) and the underlying pathophysiology is not well known. Endovascular coiling is the standard of treatment; however, a surgical procedure with vascular reconstruction by excluding the pathological segment of the vessel and restoring the blood flow, seems to be the most effective and definitive treatment.

Case Description: We report a patient who presented a fusiform vertebral artery aneurysm previously coiled which developed a giant enlargement and a new contralateral fusiform aneurysm. Hemodynamic changes resulting in the formation of contralateral aneurysm might be the result of aneurysm occlusion without revascularization. In addition, continued blood flow to the aneurysmal wall through the vasa vasorum might result in aneurysm recanalization or regrowth. In order to account for these possible sources of complications, we performed a vascular reconstruction with high and low flow bypasses after trapping the aneurysm.

Conclusions: We hypothesize that, in this and similar cases, surgical vascular reconstruction should be the first and definitive treatment under experienced cerebrovascular surgeons.

Key Words: Endovascular coiling, fusiform aneurysm, revascularization, vasa vasorum, vertebral artery

INTRODUCTION

Fusiform aneurysms are rare (<1%)\(^{10,43,54}\) and the underlying pathophysiology is not well known. A preexisting weak vessel wall and the presence of hemodynamic changes may lead to the aneurysm formation.\(^{1,10,38,40,44,54}\) Currently, endovascular coilong is the standard of treatment, but the high rate of late complications\(^{14,15,31}\) should make us consider other treatment options. A surgical procedure with vascular...
reconstruction excluding the pathological segment of the vessel and restoring the blood flow seems to be the most effective and definitive treatment\cite{34,36,50,53} when performed by experienced cerebrovascular surgeons. The objective of this case report is to describe ideal vascular reconstruction treatment for a fusiform vertebral artery aneurysm.

**CASE REPORT**

An incidental left fusiform vertebral artery (VA) aneurysm was coiled in a 60-year-old man [Figure 1]. An early recanalization (after 4 months) was found and the attitude was conservative. After 7 years, the aneurysm enlarged and the patient underwent a second coil embolization. After 6 months, a new recanalization developed and a contralateral fusiform VA aneurysm appeared. Although the patient was asymptomatic, follow-up with magnetic resonance imaging (MRI) and digital subtraction angiography after 3 and 10 months, showed enlargement of both aneurysms [Figures 2 and 3]. The patient was referred to our hospital and underwent cerebrovascular reconstruction and aneurysm resection in June 2014. Antiplatelet treatment was initiated prior to surgery.\cite{9,41}

The surgery was performed under neurophysiological monitoring. The patient was placed in a right park bench position. A handheld Doppler was used to find the subcutaneous occipital artery (OA). An L-shape postauricular skin incision was made [Figure 4]. Skin flap was elevated and the OA was harvested. Simultaneously, the radial artery (RA) was harvested from the left arm. The suboccipital muscles were dissected layer by layer while the OA was safely exposed. Retrosigmoid and transcondylar approach was performed through a suboccipital craniotomy. The posterior half of the sigmoid sinus wall was skeletonized to maximize the lateral operative field. The V3 segment of VA was exposed in the suboccipital triangle. Hypoglossal canal was skeletonized by the drilling of condylar fossa. Posterior half of jugular tubercle could be removed extradurally under hip of sigmoid sinus securing wide operative field for intracranial proximal VA. The dura was opened. The giant coiled aneurysm was ventral to the caudal cranial nerves (CNs) [Figures 5a, b and 6a]. A small arterial branch arising from the proximal portion of the aneurysm was reconstructed by OA with end-to-side anastomosis [Figures 6b and 7a].

A fish mouth shape trimming was performed on the donor vessel to ensure endothelial layers of donor and recipient attached together.

After temporary trapping, the aneurysm coil mass was incised by monopolar coagulator [Figure 5c]. Microscissors and ultrasonic aspirator were used to cut and remove the tight packed coils. Careful and meticulous coil removal was performed to avoid damage of CNs and brainstem. At that point, aneurysm removal was made possible. The RA graft (RAG) was prepared in an albumin-containing heparinized saline solution after taking it from the forearm. Subsequently both ends of RAG were anastomosed with VA in a fish mouth trimming: RAG to intracranial V4 end-to-end and RAG to V3 end-to-side fashion [Figures 6b and 7b].

Intraoperative indocyanine green video angiography, Doppler sonography, and a transit-time blood flow meter were done to confirm bypass patency and perforator arteries integrity. Proximal aneurysm clipped reinforced was applied [Figure 8]. Operation time was 6 h.

The patient could walk without assistance after 7 days, although left thermal hypoalgesia, due to Wallenberg’s syndrome appeared because of small infarction of medulla oblongata [Figure 9]. Respiratory support with ventilator during sleep at night was transient for 2 months and a half. After 6 months, the patient had a modified Rankin scale of 2, with no changes 18 months after surgery.

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**Figure 1:** Initial digital subtraction angiography (DSA) (2004) shows a left fusiform vertebral artery (VA) aneurysm (a). DSA performed after the first embolization on the left fusiform VA aneurysm demonstrates aneurysm exclusion (b)

**Figure 2:** DSA performed 6 years after the first embolization on left fusiform VA aneurysm (2011) shows aneurysm recanalization and enlargement (a). DSA performed after the second embolization on left fusiform VA aneurysm shows a new contralateral fusiform VA aneurysm (2013) (b)
Radiological images confirmed no changes on the right fusiform VA aneurysm size [Figures 9 and 10].

**DISCUSSION**

Vertebral fusiform aneurysm may be caused by dissection or an atherosclerotic change. Many classifications have been proposed to differentiate between dolichoectatic, fusiform, transitional, giant serpentine, or dissecting aneurysms, but we consider them different stages of the same entity due to the underlying pathophysiological mechanism.

Endothelial damage on a preexisting weakness of the vessel wall (with/without an atherosclerotic plaque) and the presence of hemodynamic changes may lead to the fragmentation of the internal elastic lamina and to intimal hyperplasia with neovascularization of the wall. Intramural recurrent hemorrhage in newly formed capillaries between the aneurysm wall and the intraluminal thrombus facilitate continuous enlargement of the aneurysm dissection.
Intramural hemorrhage seems to be the most critical event because it forces the aneurysm to grow. The ideal moment for a surgical approach seems to be right before intima proliferation, which can be easily detected with MRI (high T1 signal).

Symptoms commonly result from compression of neuronal structures, ischemic stroke or aneurysm rupture. The annual risk of hemorrhage in a vertebrobasilar (VB) fusiform and transitional type aneurysm may reach 2.3%. The risk increases with progressive aneurysm enlargement, with the presence of symptoms at the time of diagnosis and according to Mizutani et al. after initial subarachnoid hemorrhage. Mangrum et al. reported a growth rate of 48% in 52 patients, and a mortality rate 5.7 times higher in patients suffering aneurysm enlargement compared to those without it. Mizutani et al., reported subsequent rupture after initial subarachnoid hemorrhage in VB dissecting aneurysms in 71.4% of cases, with a devastating outcome. These facts make surgical treatment a priority.

Endovascular procedures involve few periprocedural complications and it is the standard treatment if the contralateral VA is permeable, regardless of the presence of a contralateral fusiform aneurysm. However, complications do appear, as was the case with our patient. Compaction, recanalization, coils displacement, and/or delayed progression of the thrombosis appear in fusiform aneurysms. Flow diverters initially raised very high expectations given the facility of their use, the decreased mass effect they produce when compared to coils and the opportunity they provide to reach complex aneurysms. Nevertheless, their use produced serious complications. Anterior circulation aneurysms respond better to treatment than posterior circulation ones. The presence of perforator arteries and the nature and anatomy of the VB aneurysms make them behave differently. Ischemic strokes and hemorrhage may appear within days or months after the use of VB flow diverters. Endovascular manipulation during the procedure facilitate the thrombi formation and thus, the appearance of ischemic strokes (20–40%). As we increase the use of stents in a given patient (multiple overlapping stent), we also increase the likelihood of stroke appearance. Stent placement results in a decreased blood flow and thrombosis of the aneurysm sac. The resulting inflammatory response, along with the hemodynamic changes, sometimes results in the appearance of increased intraluminal pressure, increasing the risk of aneurysm sac rupture. Mortality rate remains high as shown by Siddiqui et al.

More data and understanding regarding the VB fusiform aneurysm nature and behavior are needed before we can consider it a safe treatment option.

It is logical to assume that an internal trapping by endovascular procedures cannot stop the blood feeding into the aneurysmal wall through the vasa vasorum. This promotes a potential source of recanalization on a coiled fusiform aneurysm through the transmural vascular connections.

Surgical procedures are the most curative and definitive treatments. A good outcome depends on the presence of adequate collateral circulation and the preoperative clinical grade. The location of the aneurysm and the expertise of the surgeon also influence the success rate. Drake reported a 67% success rate in a series of 120 surgically treated patients with giant fusiform aneurysm. Steinberg reported 87% excellent outcome in VA aneurysm. Recurrence after surgical treatment is not a common finding.

Series combining endovascular and surgical treatment have reported a mortality rate of 12.5–45%. Mortality does not seem to be related to bypass failure...
but to hemorrhagic and/or ischemic events secondary to antiplatelet/anticoagulant treatment and the nature of the aneurysm itself.\textsuperscript{21} Bypass series are limited. Isolated case reports demonstrate the effectiveness of the technique, with favorable short-term outcomes.\textsuperscript{24,42} Further series with longer term outcomes are needed to confirm the efficacy and safety of the technique.

We removed the aneurysm and performed a cerebrovascular reconstruction to avoid excessive hemodynamic load to the contralateral VA aneurysm. Unfortunately, because a perforator artery was arising from the VA close to the aneurysm, it may have been occluded after trapping the aneurysm. It could have been prevented if the ante grade flow through the intracranial VA had been maintained by an interposed graft reconstruction between intracranial proximal V4 and distal V4 segment after removing the aneurysm. Hemodynamic stress and preexisting arterial wall weakness might be responsible for contralateral aneurysm formation after sacrificing the left VA.\textsuperscript{18,22,24,42} Definitive treatment should ensure the ante grade flow to prevent hemodynamic changes.\textsuperscript{9,26}

**CONCLUSIONS**

Summarizing, the vascular wall pathophysiology on fusiform aneurysms makes endovascular treatment a noncurative option. A previously coiled fusiform aneurysm may grow and become giant. The surgical excision on a coiled aneurysm becomes more difficult and the prognosis worsens. A first microsurgical reconstruction approach in experienced hands can prevent recurrence of the aneurysm and improve the patients’ prognosis.

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**Conflicts of interest**

There are no conflicts of interest.

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