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CASE REPORT

Symptomatic enlargement of an occluded giant carotido-ophthalmic aneurysm after endovascular treatment: the vasa vasorum theory

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Abstract We describe a patient with a symptomatic left giant carotido-ophthalmic aneurysm who initially underwent coil embolization with subtotal obliteration. The patient's symptoms were initially stable, but 1 year later, she presented with a rapidly progressive contralateral visual deficit. Although angiogram showed a stable neck remnant, MR confirmed aneurysm growth and showed a new peripheral hematoma in the wall of the thrombosed aneurysm. Surgical exploration was undertaken, and even after trapping and intra-aneurysmal thrombectomy, constant bleeding was observed from the wall of the thrombosed aneurysm consistent with the vasa vasorum. Bleeding stopped after cauterization and partial resection of the aneurysm dome, and the aneurysm was clipped. The patient's recent visual deficit markedly improved, and the angiogram did not reveal any residue. Giant aneurysms may continue to grow due to a hypertrophic vasa vasorum and subadventitial hemorrhages. Surgery should be considered if complete thrombosis of the aneurysm does not alleviate patient's symptoms.

Keywords Aneurysmectomy · Coil embolization · Endovascular treatment · Giant aneurysm · Surgical clipping · Vasa vasorum

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Although there is a wide variety of endovascular therapeutic options for the treatment of giant intracranial aneurysms, none of the current techniques is completely successful and free of complications in the management of these complex lesions [2]. Giant aneurysms present also a formidable challenge to surgical treatment. The growth of giant aneurysms might not only be dependent upon continuity between the aneurysmal sac and the parent artery. We hereby describe a symptomatic growth of a coiled giant aneurysm because of the development of aneurysm wall disease by the vasa vasorum. To our knowledge, this is the first report of such an entity in the anterior circulation in which a surgical treatment with successful resection of the aneurysm and reconstruction of the parent artery were performed.

Clinical presentation

A 47-year-old woman presented initially to our clinic with a longstanding history of a left eye visual deficit, and the ophthalmological assessment confirmed an optic nerve atrophy with only perception to light on the left. There was no visual field cut. Brain MR and cerebral angiogram identified a left carotido-ophthalmic aneurysm with significant compression of the left optic nerve (Fig. 1a,b). The MRI also suggested the presence of a separate, completely thrombosed mirror aneurysm on the right ophthalmic segment (Fig. 2a), and the CTA did not reveal any patent component (Fig. 2b). Since the left visual loss was longstanding, the prospect of recovery was deemed unlikely, and we elected to treat the left aneurysm by endovascular means. The aneurysm was subtotally (neck remnant) occluded by GDC coils (Fig. 1c,d). A follow-up MR at 9 months had shown some contrast enhancement of the aneurysm wall without any change in the aneurysm size

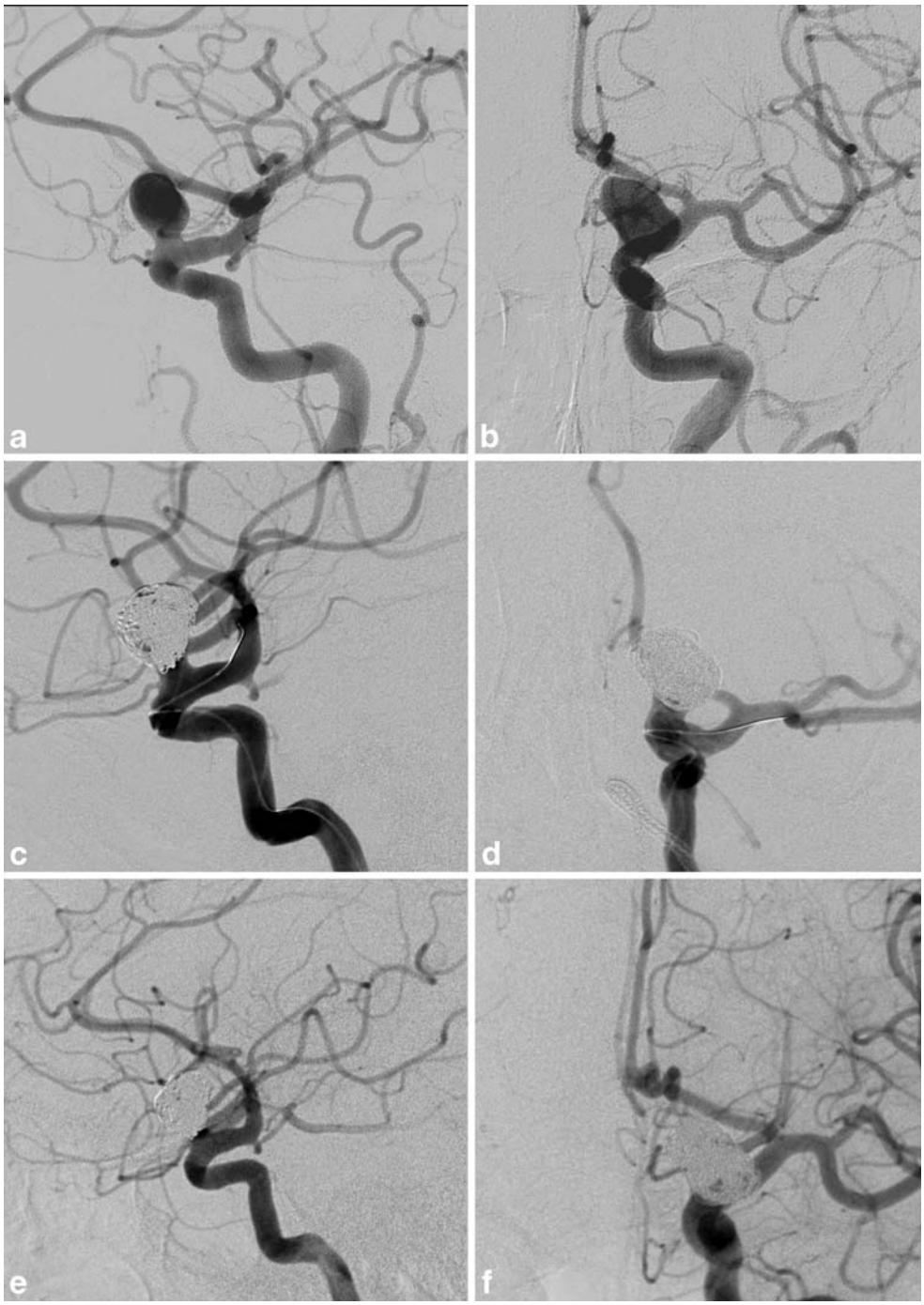

Fig. 1 (**a** and **b**) Lateral and A-P digital subtraction angiograms at the time of first presentation show the viable lumen of a left-sided giant partially thrombosed aneurysm. Lateral and A-P angiogram done right after the initial endovascular treatment (**c**,**d**) and at the second presentation (**e**,**f**) confirming a stable neck remnant without recanalization

(Fig. 2c). Afterwards, the patient was stable clinically for 1 year, when she presented again, this time with a rapidly progressive loss of right visual acuity and a right temporal hemianopia. A CT scan showed a hyperdensity in the wall of the aneurysm without any radiological signs of SAH. A conventional digital subtraction angiogram was unchanged from the post-treatment angiogram a year earlier, showing no aneurysm filling on the right ophthalmic segment and a stable neck remnant on the left carotid aneurysm (Fig. 1e,f). An MRI confirmed growth of the right-sided thrombosed aneurysm. When compared to the previous MRI, there was an onion-shaped hyperintense signal inside the aneurysm wall suggesting the presence of acute blood (Fig. 2d). At this point, and after re-evaluation of radiological images, we suspected that the aneurysm previously presumed to be right-sided might actually represent the thrombosed distal component of the coiled aneurysm on the left side, both components being connected by a narrow channel.

Due to the lack of complete certainty of this interpretation, it was felt that surgery should be planned, accounting

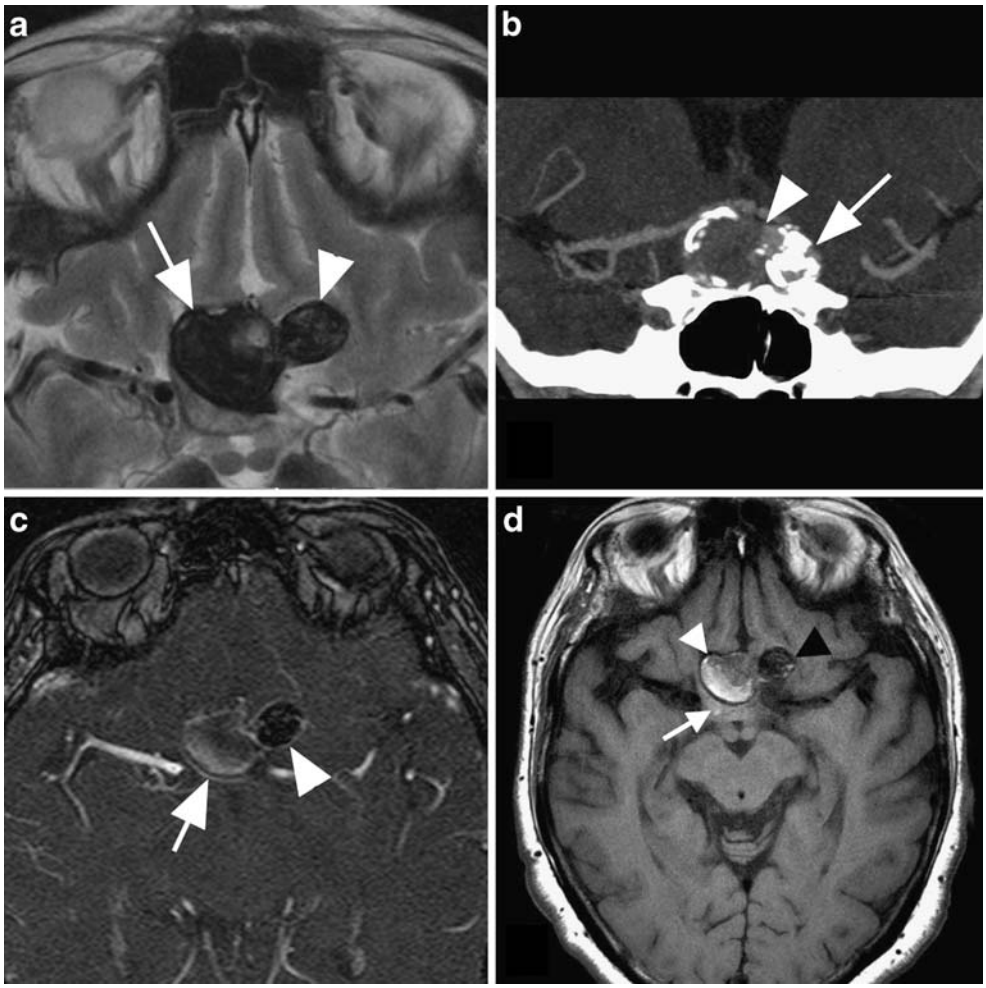

Fig. 2 (**a**) Axial MR at the time of first presentation showed a giant partially thrombosed left carotido-ophthalmic aneurysm, initially interpreted as bilateral aneurysms. The thrombotic part (white arrowhead) and the filling part (white arrow). (**b**) Coronal CTA showed the presence of calcification in the aneurysm wall and compression of neurovascular structures. The thrombotic part (white arrowhead) and the filling part (white arrow). (**c**) MR at 9-month interval shows the presence of a new wall enhancement (white arrow), suggesting an ongoing process, despite stability of aneurysm size and the neck remnant (arrow). (**d**) Axial non-enhanced T1-weighted MR showed the growth of the thrombotic part of the aneurysm (white arrowhead) with hyperintensity (arrow) along the aneurysmal wall, suggesting intramural hemorrhage. Black arrowhead showing the coiled part of the aneurysm

for the possibility of bilateral aneurysms. Although the patient had passed the balloon test occlusion of the left carotid artery, we decided to attempt the reconstruction of the carotid with resection of the aneurysm, while considering carotid occlusion as a rescue strategy.

Intervention After a left fronto-pterional craniotomy and exposure of both carotid arteries in the neck, the intracranial carotid artery was explored on the left side, and the origin of the posterior communicating artery and anterior choroidal artery were identified. The left optic nerve was totally deformed by the aneurysm, and there was a significant compression over the optic chiasm and the right optic nerve as well. Exploration through an inter-hemispheric exposure confirmed the presence of only one aneurysm arising from the ophthalmic segment of the left internal carotid artery. The aneurysm was dumbbell-shaped, with clear continuity with the thrombosed component, but no intra-luminal connection due to complete endothelialization of the previously patent (now coiled) proximal component (Fig. 3a). After temporary trapping of the left internal carotid artery, an intra-aneurysmal thrombectomy of the distal component was performed by the aid of an ultrasonic aspirator (Cavitron®; Valley Lab, Denver, CO). The presence of thrombosis had completely disconnected these two aneurysmal compartments, and there was no flow between the coiled part and the thrombosed part. Despite the absence of connection between the two lobes, there was a significant oozing from the aneurysm wall, which we attributed to the vasa vasorum (Fig. 3b). This bleeding stopped only when the aneurysm wall was subtotally resected and cauterized. Then the coils were removed from the proximal part of the aneurysm (Fig. 3c). The carotid artery was finally reconstructed and preserved by clipping the aneurysm neck (Fig. 3d). Certain portions of the aneurysm, with calcifications and adherence to neurovascular structures, were left after careful cauterization of the aneurysm wall.

A post-operative CTA and angiogram confirmed the total exclusion of the aneurysm (Fig. 4a,b), and the patient showed a marked improvement of her right eye's visual function, from 20/200 preoperatively to 20/40 postoperatively. The left eye did not show any recovery.

The histopathological report showed the presence of degenerative changes in the aneurysm wall with calcifica-

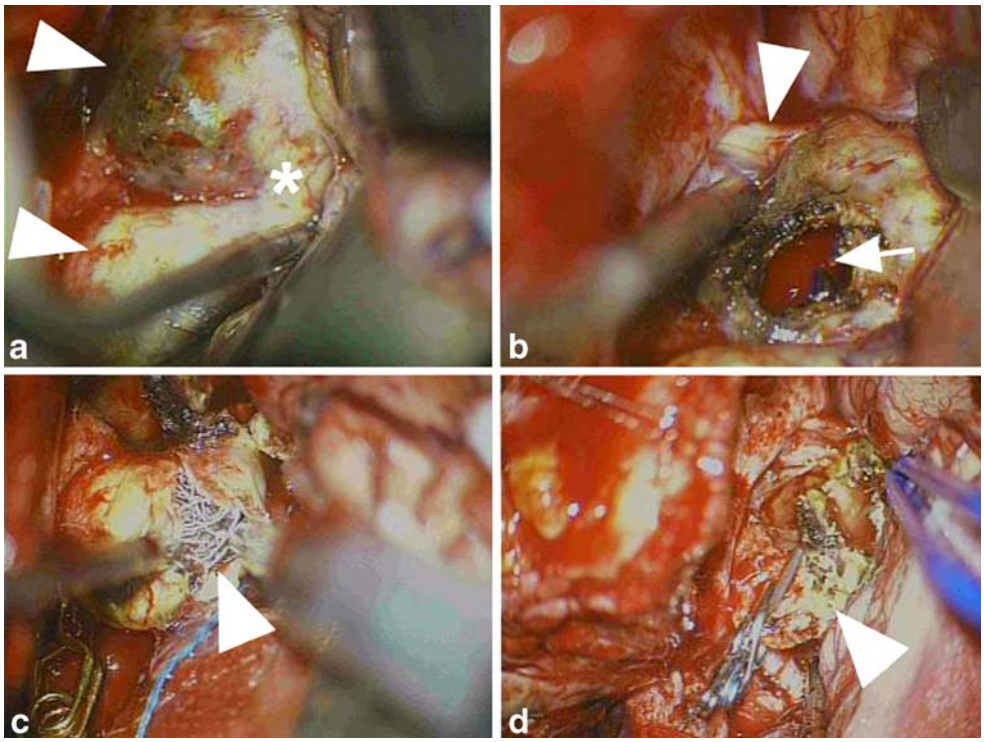

Fig. 3 (**a**) Intra-operative picture after a left pterional exposure showing the two lobules of the aneurysm (arrowheads) connected by a narrow channel (*). (**b**) Opening of the thrombotic part (right part) of the aneurysm causing blood oozing from the aneurysm wall. Note the compression of the right optic nerve (arrowhead). (**c**) Opening of the coiled part of the aneurysm with coil (arrowhead) removal. The temporary clip in this picture was placed distal to the aneurysm and proximal to the origin of the PcomA artery. (**d**) Final clip reconstruction of the parent artery after partial resection of the aneurysm

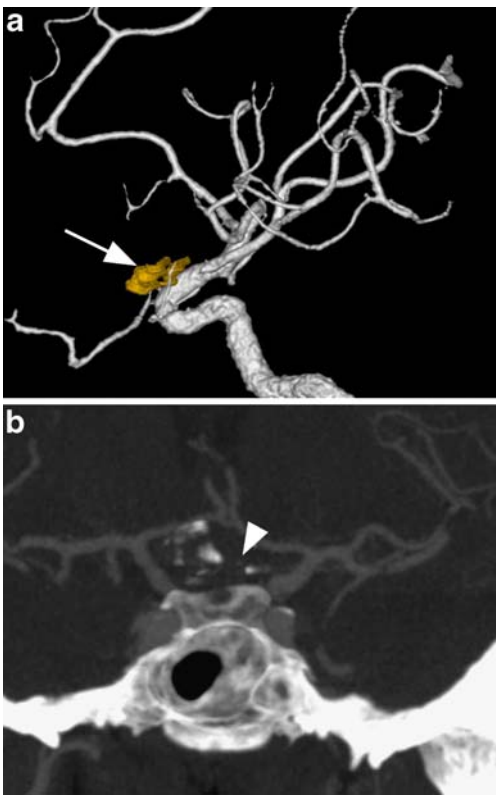

Fig. 4 (**a**) DSA confirmed total exclusion of the aneurysm by one fenestrated and one straight clip (arrow). (**b**) CTA showed subtotal resection of the aneurysm wall (arrowhead) with remaining calcifications adherent to the anterior cerebral artery

tions, hemosiderin deposition, and chronic inflammation marked by the presence of lymphocytes, hemosiderin-laden macrophages, and foreign body-type giant cells. Fresh fibrin mural thrombosis was found, and in the adventitia of the aneurysm wall, a plexus of small arterioles compatible with the vasa vasorum was identified (Fig. 5a,b).

The thrombus itself was void of any pathological evidence of neovascularization.

Discussion

Giant thrombosed intracranial aneurysms are some of the most challenging vascular pathological features in the central nervous system. Although trapping and aneurysmectomy by surgery is the established method for the treatment of these complex lesions [7, 14], endovascular techniques such as trapping, flow reversal, and covered stents offer promising alternatives because of their less invasive nature [11, 15]. In the present case, the choice of endovascular treatment at the initial presentation was based on the assumption of the presence of bilateral aneurysms, considering a left carotido-ophthalmic aneurysm suitable for coiling and a right thrombosed carotido-ophthalmic aneurysm with no viable lumen. The presence of partial thrombosis inside a single giant aneurysm would have been a relative contraindication for the initial endovascular coiling because of a high likelihood of coil migration and aneurysm recanalization. However, there was no recanalization or coil migration, but rather enlargement of the

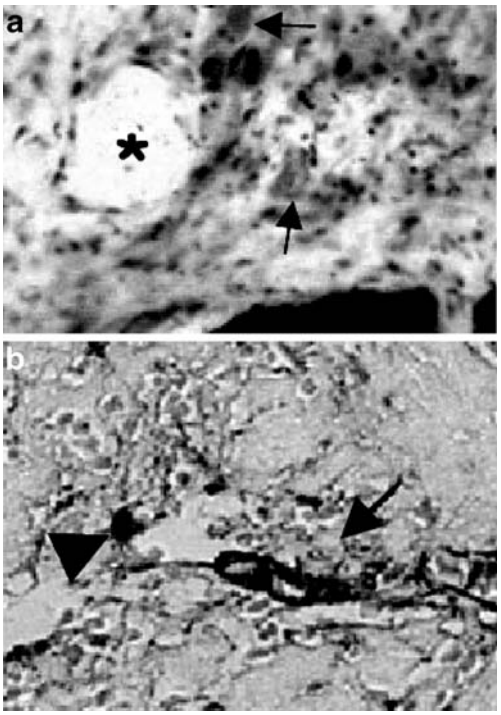

Fig. 5 Histopathological features of the resected aneurysm. (**a**) Neovascularization (*) and hemosiderin-laden macrophages (arrows) are visible in the aneurysm wall. (**b**) In the adventitia of the aneurysm wall, a plexus of small arterioles (arrow) compatible with the vasa vasorum and hemosiderin deposition (arrowhead) are seen. Original magnification ×400 (a); ×100 (b)

aneurysm in this case. Had the aneurysm been considered initially as a single giant partially thrombosed aneurysm, we would have favored a surgical treatment or an endovascular trapping procedure.

Halbach et al. have suggested that endovascular embolization can improve or alleviate presenting neurological signs resulting from the mass effect by stopping the progression of the aneurysm and preventing any further progress or injury to neuronal tissue[3]. Although the optimal treatment of intracranial aneurysms is to eliminate them from the intracranial circulation while preserving the parent vessel, this strategy might be difficult to achieve in some thrombosed giant aneurysms. Even after total occlusion by endosaccular coiling, continued growth and increasing symptoms have been reported [4, 5]. In our case, coiling left a stable neck remnant in this partially thrombosed giant aneuysm, but intra-aneurysmal wall bleeding and increased symptoms were observed at admission. The presence of bleeding from a markedly developed the vasa vasorum network identified at surgery was considered the underlying cause of the aneurysm growth. There are only two similar case reports with aneurysms in the posterior circulation. Hirasawa et al. reported on a giant basilar tip aneurysm with fatal regrowth despite the achievement of a complete thrombosis by endovascular treatment [4]. Iihara et al. reported the case of a patient with a partially thrombosed giant vertebral artery aneurysm who had undergone total endovascular occlusion and trapping [5]. Because of the symptomatic regrowth of the aneurysm, a surgical exploration and resection of the aneurysm were performed. The histological specimen confirmed the presence of an inflammatory reaction and the vasa vasorum in the aneurysm wall.

Although occluded intracranial arteries have been considered as a potent stimulus for the development of the vasa vasorum [1], in the present case they were derived without previous sacrifice of any major intracranial vessels, indicating the importance of the aneurysm wall per se in the development of neovascularization in giant thrombosed aneurysms. This might be due to other factors than a persistent continuity between the aneurysm and the parent vessel or intracranial vessel occlusion. Schubiger et al. suggested that formation of intracranial giant aneurysms could be due to a chronic dissection process associated with recurrent subadventitial hemorrhages from the vasa vasorum [12]. Naghiro et al. outlined that formation of intrathrombotic vascular channels and subsequent establishment of blood flow between the parent artery and those channels may be important factors in the growth of thrombosed aneurysms [10]. The role of the vasa vasorum in the formation of aneurysms is clearly emphasized by Zhao et al. They defined one potential mechanism responsible for the growth of arterial aneurysms, namely 5-lipoxygenase, which leads to leukotriene production, a potent mediator of inflammation. Neoangiogenesis of the vasa vasorum is encountered in close proximity to 5-lipoxygenase-activated macrophages [16]. Yasargil also reported that on a surgical exploration of giant intracranial aneurysms, a tremendous network of fine vessels covering the aneurysm could be seen [8]. Subsequently, it was noted that subarachnoid hemorrhage in partially thrombosed giant aneurysms did not always occur from the aneurysm itself, but instead from the vessel wall with formation of a fresh clot both inside and outside the aneurysm wall [13].

Neovascularization in the adventitia might be a potential source of recanalization of thrombosed aneurysm after successful coiling. The accumulation of inflammatory cells in the unorganized thrombus or an inflammatory reaction around the coils may induce a chronic inflammation and neovascularization in the aneurysm wall [9]. The new contrast enhancement in the aneurysm wall in the present case during follow-up was a representation of the neovascularization, which later caused the subadventitial hemorrhage. Those potential routes of blood supply from the vasa vasorum to the aneurysm cannot be interrupted by the endovascular coiling and are obvious limitations of endovascular packing in thrombosed giant aneurysms. Krings et al. have stated in a recent paper that endovascular

coiling might be insufficient to treat these aneurysms as the pathological process is likely to be maintained in the vessel wall employing a purely luminal therapeutic approach [6].

Conclusion

Symptomatic thrombosed giant aneurysms represent a formidable challenge for treatment. This special case illustrates the potential inefficacy of endosaccular coil embolization in the management of partially thrombosed giant ICA aneurysm even when near complete obliteration of the aneurysm lumen is achieved with no evidence of recanalization or coil compaction. The development of a rich vasa vasorum network, inaccessible by the endovascular routes, was clearly the cause of aneurysm growth by bleeding inside the aneurysm wall. Despite the rarity of regrowth of aneurysms following complete thrombosis or occlusion, the theory of the vasa vasorum should be kept in mind as a possible etiology in the progression of giant aneurysms. Surgical resection of giant aneurysms with or without parent artery sacrifice might be effective in refractory cases after complete coil embolization.

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