Rapid Revascularization After Therapeutic Parent Artery Occlusion for a Large Intracavernous Carotid Artery Aneurysm
—Case Report—

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Abstract

A 21-year-old woman presented with an unruptured large intracavernous aneurysm, which was spontaneously revascularized via unusual collateral pathways a short time after extracranial-intracranial bypass and surgical ligation of the proximal internal carotid artery. The patient had been treated for a large basilar trunk aneurysm with intraaneurysmal embolization using Guglielmi detachable coils, and an intracavernous carotid artery aneurysm treated conservatively. Two years later, the patient presented with right abducens nerve palsy, and was referred to our hospital. She had small nevi in the right forehead and eyelid. Cerebral angiography revealed enlargement of the intracavernous aneurysm. Superficial temporal artery-middle cerebral artery bypass followed by surgical carotid artery ligation were performed, and good patency of bypass and disappearance of the aneurysm were confirmed by intraoperative angiography. However, follow-up magnetic resonance angiography and cerebral angiography on the 20th postoperative day revealed revascularization of the internal carotid artery and the intracavernous carotid artery aneurysm via unusual collateral pathways. Subsequently, the recurrent aneurysm and the recanalized internal carotid artery were occluded by endovascular procedures. Histological examination of the nevus showed lack of properly organized vascular structures, and the diagnosis was angiodysplasia. The early development of unusual collateral pathway, and aneurysm formation at a young age might be related to the angiodysplasia. Revascularization is possible within a short time even in cases of intracavernous carotid artery aneurysm successfully treated with surgical ligation of the parent artery.

Key words: angiodysplasia, internal carotid artery occlusion, large aneurysm, revascularization

Introduction

Intracavernous carotid artery aneurysms account for 3% to 5% of all intracranial aneurysms, and often become enlarged and partially thrombosed, resulting in cranial nerve paresis caused by compression, and ischemic phenomena attributed to thromboembolic episodes. The patient often presents with symptoms such as ophthalmoplegia, diplopia, and orbital pain. However, 34% of patients with intracavernous carotid artery aneurysms are asymptomatic at diagnosis. Asymptomatic patients are often treated conservatively to avoid the risks of morbidity and mortality associated with attempts to treat the lesion. However, treatment should be considered for symptomatic intracavernous carotid artery aneurysms because of the high incidence of progressive ophthalmoplegia or visual disturbance, hemorrhage, or thromboembolic events. Nowadays, many hospitals manage these lesions using cervical carotid artery occlusion (using surgical ligation or endovascular procedures)
with or without extracranial-intracranial (EC-IC) bypass. The success rate has increased with recent advances in endovascular techniques and diagnostic tests such as the balloon occlusion test which can evaluate the risks of postoperative infarction before permanent internal carotid artery occlusion.\(^{3,6,9}\)

Here, we describe a case of unruptured large intracavernous aneurysm, which spontaneously revascularized within a short time after EC-IC bypass and surgical ligation of the proximal internal carotid artery.

**Case Report**

A 21-year-old woman suffered severe headache and vertigo, and visited a nearby hospital. Physical examination found pigmented skin lesions or nevi on the right forehead and upper eyelid, indicative of vascular abnormality (Fig. 1A). Magnetic resonance (MR) imaging and cerebral angiography demonstrated a large basilar trunk aneurysm and a large aneurysm of the intracavernous portion of the right internal carotid artery (Fig. 2A–D). Intraaneurysmal embolization using Guglielmi detachable coils (GDCs) was performed for the basilar trunk aneurysm, and the intracavernous carotid artery aneurysm was treated conservatively. The patient developed no new postoperative neurological deficit. Two years after treatment of the basilar trunk aneurysm, the patient presented with right abducens nerve palsy, and was referred to our hospital.

Right internal carotid angiography demonstrated complete obliteration of the basilar trunk aneurysm and the large intracavernous carotid artery aneurysm (20 × 15 mm), which had enlarged compared to 2 years previously (Fig. 2E–H). Right external carotid angiography revealed pooling of contrast medium in the venous phase supplied by the superficial temporal artery (STA), corresponding to the site of the cutaneous lesions (Fig. 1B, C). We decided to treat the symptomatic intracavernous carotid artery aneurysm.

The balloon occlusion test using technetium-99m ethyl cysteinate dimer-single photon emission computed tomography was performed to predict tolerance to therapeutic carotid artery occlusion. Occlusion of the internal carotid artery at the level of the C1–2 vertebral bodies caused decreased cerebral blood flow with a minimum of 71% hypoperfusion in the right hemisphere, compared to the contralateral homologous region, although there were no clinical symptoms. No collateral flows between

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**Fig. 1** A: Photograph showing pigmented skin lesions or nevi on the right forehead and upper eyelid. B, C: Right external carotid angiograms, lateral views in the early (B) and late (C) filling phases, showing pooling of the contrast medium in the frontal branch of the superficial temporal artery (arrow). D: Intraoperative photograph showing an abnormally tortuous and dilated vessel (asterisk) in a small portion of the superficial temporal artery.

**Fig. 3** A–C: Photomicrographs of specimens of the surgically resected abnormal vessels showing unusual structural abnormalities with an unevenly thickened wall (asterisk) surrounded by “cavernous angioma”-like anastomosing network of dilated cavities (arrow). Elastica-Masson stain, original magnifications A: \(\times 40\), B: \(\times 100\), C: \(\times 200\). D: Photomicrograph showing immunohistochemical staining for CD31 in the luminal surface of the central abnormal vessel (arrow) and surrounding anastomosing cavities (arrowhead). Original magnification \(\times 200\).
the external and internal carotid arteries were observed during occlusion.

STA-middle cerebral artery (MCA) bypass was performed followed by surgical internal carotid artery ligation at the cervical origin, and good patency of bypass and disappearance of the aneurysm were confirmed by intraoperative angiography. Reflection of the skin flap and dissection of the STA exposed abnormally tortuous and dilated venous pouch-like structures in the subcutaneous connective tissue layer (Fig. 1D), corresponding to the site of the pigmented skin lesions and angiographic findings. Histological examination of a specimen of these abnormal vessels demonstrated structurally abnormal vessels, consisting of unevenly thickened wall surrounded by “cavernous angioma”-like anastomosing dilated cavities (Fig. 3). The thickened vascular wall mainly consisted of collagen fibers, and displayed disrupted internal and external elastic lamina. The central thickened vascular walls contained many myofibroblasts, and only a few discontinuous vascular smooth muscle cells. The diagnosis was angiodysplasia based on the absence of proper organized vascular structures.

The postoperative course was uneventful. MR angiography obtained 1 day after the operation confirmed good patency of the STA-MCA bypass, and disappearance of the intracavernous carotid artery aneurysm (Fig. 4A). However, 11 days after the operation, follow-up MR angiography revealed revascularization of the aneurysm, although the STA-MCA bypass was well-preserved (Fig. 4B). Follow-up cerebral angiography was performed 20 days after the operation. Right common carotid angiography showed therapeutic occlusion of the right internal carotid artery, and antegrade revascularization 10 mm distal to the occluded portion through small vessels arising from the right external carotid artery (Fig. 5A). Right vertebral angiography demonstrated similar but more prominent small arterial channels from the vertebral artery, which provided antegrade flow to the internal carotid artery (Fig. 5B). These changes had resulted in reappearance of the intracavernous carotid artery aneurysm with partial thrombus formation.

The recurrent aneurysm and the recanalized inter-
Fig. 4  Magnetic resonance angiograms 1 day after operation (A) confirming complete obliteration of the intracavernous carotid artery aneurysm and good patency of superficial temporal artery-middle cerebral artery (STA-MCA) bypass, and 11 days after the operation (B) revealing revascularization of the intracavernous carotid artery aneurysm, and good patency of the STA-MCA bypass.

Fig. 5  A, B: Right common carotid angiogram 20 days after the operation (A) showing proximal occlusion of the right internal carotid artery, and revascularization of the right internal carotid artery (arrow) through small vessels arising from the right external carotid artery, and right vertebral angiogram (B) demonstrating multiple small arterial channels from the vertebral artery (arrowheads), providing antegrade flow to the internal carotid artery.  C, D: Right common carotid angiogram (C) and right vertebral angiogram (D) just after the second operation showing complete obliteration of the intracavernous carotid artery aneurysm and the internal carotid artery.  E: Lateral skull radiograph demonstrating Guglielmi detachable coils within the intracavernous carotid artery aneurysm and at the cervical portion of the internal carotid artery (arrowheads), as well as within the basilar trunk aneurysm (arrow).

Discussion

Rapid antegrade recanalization of the internal carotid artery is rare after permanent occlusion by surgical ligation for the treatment of intracavernous carotid artery aneurysm. In the present case, the angiographic and pathological findings suggested that this phenomenon occurred as a result of the development of unusual collateral pathways, probably facilitated by angiodysplasia. Angiodysplasia is a vascular malformation, and is a frequent source of recurrent gastrointestinal bleeding and anemia. Histologically, angiodysplasia resembles telangiectasia, and is characterized by dilated and tortuous vessels.16) Intracranial aneurysm associated with angiodysplasia is rarely reported.2,7) The present case has several unique points: two large intracranial aneurysms identified at a young age (21 years); association of nevi, which were histologically confirmed as angiodysplasia; and revascularization after surgical occlusion within a short time (11 days). Vascular changes including aneurysm formation may occur even in young patients with vasculitis or arteritis associated with Takayasu arteritis, Behçet's disease or human immunodeficiency virus infection,1,11,12) and connective tissue disorders such as Ehlers-Danlos syndrome and Marfan syndrome.13,14) However, the present patient did not exhibit any suggestive clinical and histological findings related to such diseases. Another explanation for this phenomenon is the involvement of intrinsic collateral routes between the intracranial and extracranial circulations. Formation of multiple large aneurysms at a young...
age and development of unusual collateral pathways during a short time are likely to be multifactorial. Local fragility of the vascular wall was attributed to angiodysplasia, that was most probably the reason for aneurysm formation in this young patient, and might have affected the early development of the unusual collateral pathways. However, we could show no direct relationship between the angiodysplasia and the cerebral aneurysms or unusual collateral pathways. More cases may clarify the causative processes.

Many therapeutic options including surgical, endovascular, and combined surgical and endovascular approaches are available for the treatment of intracavernous carotid artery aneurysm. The risk of morbidity in each procedure must be considered to determine the best treatment for individual patients. We could not exclude the possibility of dissecting aneurysm, because of the lack of histological examination of the intracavernous carotid artery aneurysm, so endovascular trapping of the parent artery might have been safer and easier. However, T1-weighted MR imaging showed no typical diagnostic signs of dissecting aneurysm such as double lumen, intimal flap, or high signal intensity around the arterial flow void due to intramural hematoma. Endovascular trapping of the parent artery including the origin of the ophthalmic artery (a possible collateral to the aneurysm) might also have been a treatment option in the first operation.

The present case indicates that rapid revascularization may occur even in cases of intracavernous carotid artery aneurysm successfully treated with cervical internal carotid artery ligation, with confirmation of disappearance of the aneurysm using intraoperative angiography. More cautious treatment and follow-up examinations are necessary in patients with cerebral aneurysms associated with any signs of vascular abnormalities such as nevus or angiodysplasia.

References

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