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Case Report

Emergent cervical surgical embolectomy to rescue total monocular blindness due to simultaneous cervical internal and external carotid artery occlusion by cardiogenic emboli

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Abstract

Background: Central retinal artery occlusion (CRAO) is a stroke of the retina and is associated with extremely poor prognosis. Although the pathophysiology of CRAO is diverse, including autoimmune or hematological disorders, neurosurgeons can perform carotid endarterectomy for the causal internal carotid artery stenosis or perform acute recanalization of the extra- or intracranial artery occlusion due to cardiogenic embolism.

Case Description: A 78-year-old male with a history of atrial fibrillation (Af) visited our hospital with a chief complaint of right monocular blindness. Magnetic resonance imaging revealed occlusion of the right internal and external carotid arteries. We performed emergent cervical surgical embolectomy for restoration of vision. Recanalization was accomplished within 8 h after onset, and the patient regained practical vision within 4 months.

Conclusion: In the diagnosis and treatment of CRAO, occlusion of the internal and/or external carotid artery due to large cardiac emboli should be taken in consideration, especially when the patient has a history of Af, since acute recanalization might restore vision.

Key Words: Acute ischemic stroke, atrial fibrillation, carotid artery occlusion, central retinal artery occlusion, retina, surgical embolectomy

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INTRODUCTION

Central retinal artery occlusion (CRAO) is a stroke of the retina that is associated with poor visual outcomes;[11] no effective treatments, including the use of tissue plasminogen activator (t-PA), have been established for management of this ocular crisis.[3,21] Large cardiogenic emboli due to valvular diseases or atrial fibrillation (Af) can cause cervical carotid artery occlusion and result in CRAO. Such acute cervical carotid artery occlusion can be treated with intravenous administration of t-PA (IV t-PA) and/or endovascular treatment or cervical surgical embolectomy.[5,17] We performed emergent cervical surgical embolectomy for right cervical simultaneous internal/external carotid artery occlusion due to Af-related emboli. Quick recanalization restored practical vision in

this patient with total monocular blindness. A detailed case illustration is presented.

CASE REPORT

History

A 78-year-old male presented to our emergency room with a chief complaint of total right monocular visual loss. Four hours and forty minutes prior, he had strained during a bowel movement and developed blurred vision in his right eye. He went to see an ophthalmologist. His visual acuity in the right eye was 20/25 (20/20, corrected) and the eye showed no abnormal conditions in his slit-lamp and funduscopic examination. The arteries of the retina were stenotic, however, the color of the retina was normal and an ischemic condition such as cherry red spot was not detected [Figure 1a (left)]. Optical coherence tomography demonstrated normal thickness of the retina [Figure 1b (right)]. Further, he recognized gradual improvement of his vision and returned home. Three hours later when he was taking a bath, he developed total blindness in the right eye. His wife noted his mydriasis of the right eye. His past medical history was remarkable for chronic Af, which had two episodes of cerebral infarction (5 and 3 years ago, respectively), for which he was taking warfarin. His prothrombin time-international normalized ratio (PT-INR) was 1.85 at the time of admission to our hospital. No obvious neurological focal deficits were noted except for findings in his right eye: His left pupil was 3 mm and reactive to light, but his right pupils was fixed at 6 mm on testing of both direct and consensual pupillary responses. He was alert and denied any other symptoms except his right total monocular visual loss. His National Institute of Health Stroke Scale (NIHSS) score was 0.

Examination

Initial diffusion-weighted images (DWIs) magnetic resonance imaging (MRI) of the brain did not demonstrate any high intensity lesions [Figure 2a]. Magnetic resonance angiography (MRA) showed occlusion of the right internal/external carotid artery (ICA/ECA) and the right ophthalmic artery was not visualized [Figure 2b]. Neck MRA did not show any signal intensity of the right ICA/ECA or of the right common carotid artery (CCA) [Figure 2c]. Magnetization prepared rapid gradient echo (MPRAGE) image showed loss of black signal in the right CCA with no obvious deposition of vulnerable plaque [Figure 2d].[7] Considering his past history of two embolic strokes attributable to chronic Af, the sudden-onset clinical course, and negative atherosclerotic arterial stenotic lesions in the MRA 10 months prior [Figure 2e and f], we inferred that cardiogenic emboli occluded both the right ECA and ICA, which caused his total monocular visual loss due to CRAO. IV t-PA was considered not appropriate in this condition, since large cervical emboli could migrate further into the intracranial ICA or middle cerebral artery (MCA), causing devastating cerebral infarction. Emergent endovascular treatment was not available at our institution. We decided to proceed with cervical surgical embolectomy. After thorough discussion with him and his family, the operation started 2 h and 28 min after the patient's arrival at our hospital.

Operation

With the patient in a neutral supine position under general anesthesia, an S-shaped curvilinear skin incision was made along the medial side of the sternocleidomastoid muscle. In order to prevent distal migration of emboli, the proximal portion of the CCA was clamped as soon as it was exposed. Linear longitudinal incision was made in the CCA, and bluish fresh large emboli came out of the incision spontaneously by back flow [Figure 3a]. Gross inspection of the emboli found simple bluish emboli rather than debris related to atherosclerotic plaque, suggesting cardiogenic embolism [Figure 3b]. After retrieval of the emboli, robust back flow of the blood from the distal portion of the CCA was seen. Considering good intracranial left to right cross flow signal in preoperative MRA, we

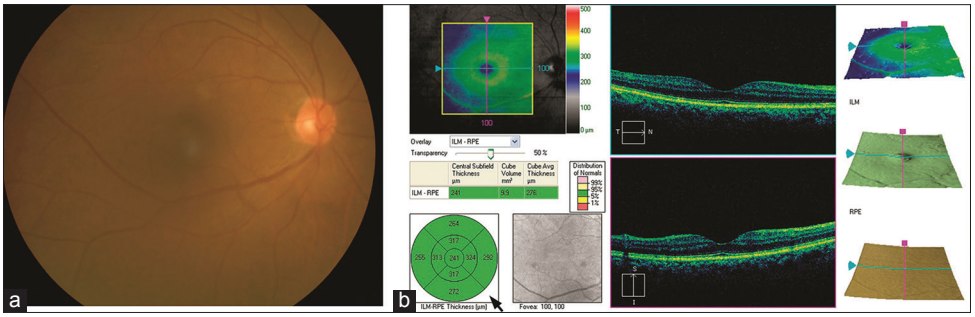

Figure 1: Ophthalmological studies at the initial onset of blurred vision in the right. In fundus photograph ((a), left), the arteries are stenotic, however, an ischemic change such as cherry red spot is not detected on the retina. In optical coherence tomography ((b), right), cross-section of the macular is in normal configuration. At the lower left of the (b) retinal thickness around the macula is shown. The macular area is divided into 9 segments and the thickness of each segment is within normal limits and is painted as green (arrow)

Figure 2: Preoperative imaging studies. Initial diffusion-weighted images (DWI) magnetic resonance imaging (MRI) demonstrates no high intensity lesions (a). Head magnetic resonance angiography (MRA) shows occlusion of the right internal carotid artery (ICA) and the right ophthalmic artery (b). Neck MRA shows occlusion of the right common carotid artery (CCA) (c). Magnetization prepared rapid gradient echo (MPRAGE) image shows loss of black signal in the right CCA (d). No stenosis is seen 10 months prior on head and neck MRA (e and f)

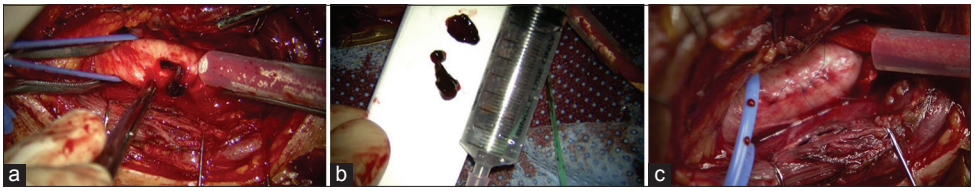

Figure 3: Intraoperative images of the cervical embolectomy. An embolus is coming out from the incision (a). The embolus consists of two components, each of which is 1 cm in size, and is composed of simple violet thrombus at gross inspection (b). The arteriotomy is sutured with 6-0 nylon (c)

judged that we had restored anterograde ICA flow. The distal portion of the CCA was also clamped, and the arteriotomy was sutured with 6-0 nylon [Figure 3c]. Doppler flowmeter confirmed good anterograde blood flow of the CCA.

The recanalization time was 19 min since the start of surgery, 2 h and 47 min since the arrival of the patient, and approximately 8 h and 12 min since symptom onset [Video 1].

Pathological findings

The pathological report described the embolus resected from cervical surgical embolectomy as erythrocyte rich mixed thrombus, consistent with cardiogenic embolism.

Postoperative course

Postoperatively, the patient had partial recovery of his vision of the right eye (he was able to recognize hand movement) from the status of total blindness. No obvious additional focal deficits were seen. Hyperbaric oxygen therapy and oral intake of kallidinogenase and limaprost alfadex were started for CRAO. Anticoagulation with warfarin was continued.

Postoperative DWI did not show any intracranial ischemic lesions, and MRA showed anterograde recanalization of the right cervical to intracranial ICA, including the right ophthalmic artery [Figure 4a]. Cervical MRA showed consistent occlusion of the ECA [Figure 4b]. However, follow-up MRA on postoperative day (POD) 7 demonstrated complete recanalization of the right ECA, suggesting that the occlusion was embolic in nature [Figure 4c].

On POD 2, the ophthalmologist reported improvement of blood flow to the right retina based on fundus fluorescein angiography [Figure 5a]. Although the retinal perfusion was recovered by the operation, the influence of retinal ischemia caused by occlusion of the carotid arteries still remained and therefore, cherry red spot was still seen [Figure 5b]. Optical coherence tomography on the same day showed thinning of the retina around the macular area [Figure 5c]. His visual acuity recovered to 20/1000 from total blindness.

He was able to recognize faces on POD 8. He returned

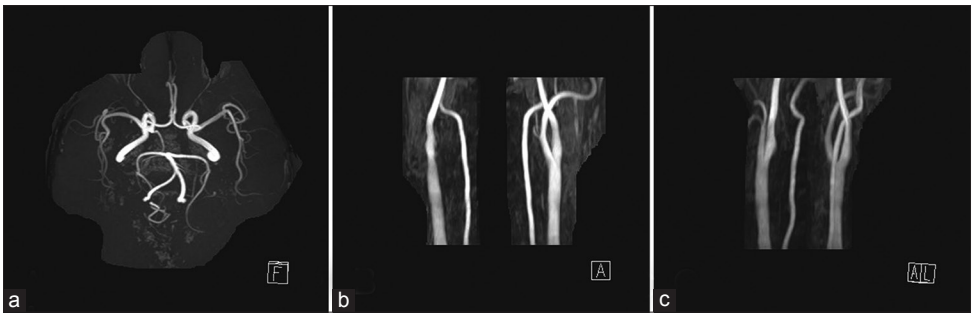

Figure 4: Magnetic resonance angiography (MRA) image after the cervical embolectomy. Head MRA shows total recanalization of the right internal carotid artery (ICA), including the right ophthalmic artery (a). Neck MRA shows good patency of the ICA and consistent occlusion of the external carotid artery (ECA) (b). Follow-up MRA on postoperative date (POD) 7 demonstrates recanalization of the right ECA (c)

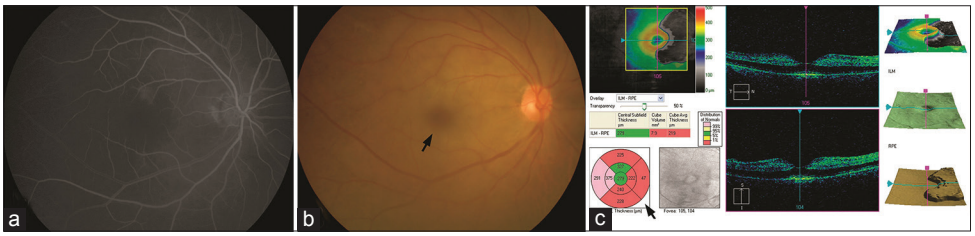

Figure 5: Postoperative ophthalmological studies. Fundus fluorescein angiography (a) demonstrates recovery of blood flow of the retinal arteries. As a result of retinal ischemia caused by occlusion of the carotid arteries, cherry red spot is seen in fundus photograph (b)arrow. Optical coherence tomography (c) also shows thinning of the retina around the macular area as painted red (arrow)

home 16 days after surgery. Four months after the surgery, his visual acuity increased to 20/40.

DISCUSSION

This is the first report to describe the efficacy of cervical surgical embolectomy for restoration of practical vision in a patient with simultaneous ECA and ICA occlusion due to cardiac embolism presenting with CRAO.

The typical presentation of CRAO is acute, painless loss of vision, and 80% of the affected eye has a final visual acuity of counting fingers or worse.[11,20] CRAO is classified into four groups according to its pathophysiology: Arteritic (such as giant cell arteritis), nonarteritic (classic clinical picture of retinal infarction, cherry-red spot, and absence of or poor residual retinal circulation on fluorescein angiography), nonarteritic with cilio-retinal artery sparing (a central island is spared thanks to the patent cilio-retinal artery), and transient nonarteritic.[11] The present case belongs to nonarteritic CRAO, in which visual improvement occurs only in 20–30% of patients.[11,26] From the standpoint of neurosurgeons, in the treatment of CRAO or transient monocular visual loss (TMLV), many studies emphasize the importance of carotid investigation and the potential indication for carotid endarterectomy.[1,2,18,23,24] Meanwhile, the contribution of cardiogenic embolism to CRAO has also been described, although the morbidity rate of Af in CRAO is still uncertain. One retrospective study of 77 patients with histories of transient or permanent monocular visual loss or evidence of asymptomatic retinal embolism reported that less than 10 patients (13%) had comorbid diseases, one of which was Af.[1] In a retrospective study of 37 eyes with CRAO, branch retinal artery occlusion, and cilioretinal artery occlusion, only three patients (8.7%) had a documented past history of Af.[14] It is not clear if Af in these two studies included paroxysmal Af, which is often difficult to detect.[19] Thus, we assume that large embolic simultaneous ICA/ECA occlusion as a cause of CRAO is quite rare.

Immediate reperfusion of the retina can result in restoration of vision. However, the use of t-PA is still controversial.[4] A prospective randomized multicenter clinical trial was unable to show that local intraarterial (IA) t-PA resulted in restoration of vision.[21] IV t-PA has not proven efficacy in restoring vision either.[3] Those studies excluded patients with carotid artery occlusion and thus we cannot assume whether IV t-PA would have been effective in our present case. However, the indication for IV t-PA was somewhat controversial in the present case; large emboli at cervical ICA, once partially resolved by IV t-PA, could migrate distally into intracranial ICA or MCA and result in devastating cerebral infarction. Endovascular treatment with proximal protection and aspiration might have been a good choice for treatment.[5,17] Thus, from the standpoint of the neurosurgeon, there are two gross revascularization strategies. If the occlusion site is distal to the ophthalmic artery, pharmacological recanalization by IV t-PA or IA t-PA could be a good option. In contrast, if the occlusion site is proximal to the cervical ICA,

surgical or endovascular clot retrieval with rigid proximal protection to prevent distal intracranial migration of emboli is ideal.

The golden time within which the human retina can be spared is not precisely known, but is no longer than 240 min.[12] Animal studies of rhesus monkeys report that the retina or the optic nerve suffer irreversible damage after a central retinal artery (CRA) occlusion of 105 min but may recover well within 97 min of the ictus.[9,10,26] In the clinical settings, recovery of vision has been reported in eyes treated up to 12 h after the onset of blindness, although loss of vision is often permanent after 2 h.[15,16,26] In the present case, retinal reperfusion was accomplished 3 h and 32 min after onset of total monocular blindness. In addition, the recanalization time was 19 min since the start of surgery, and 2 h and 47 min since the arrival of the patient to our hospital. Thus, from the standpoint of revascularization time, cervical surgical embolectomy could be an appropriate additional strategy to rescue acute visual loss by acute embolic cervical carotid occlusion.

There are numerous collaterals between the ICA and ECA, and the blood supply of the retina does not depend solely on the ICA/ophthalmic artery.[18] Typically, such collaterals open up as ICA stenosis progresses.[6,13] The ophthalmic artery forms important collateral pathway between the ECA and the ICA with ICA stenosis,[22] and such collateral pathways are summarized in great detail by Hayreh,[8] such as the distal maxillary artery, the ascending pharyngeal artery, the vidian artery, and so on. Thanks to these ECA to ICA collaterals, the proximal ophthalmic artery can sometimes be ligated without causing ischemia to the retina.[25] In our present case, the patient described stepwise aggravation of his vision in the right eye: Blurred vision that initially improved gradually but resulted in total blindness in 4.5 h. Based on this description of the history of illness, it is assumed that a cardiogenic embolus occluded the cervical ICA first when he strained during a bowel movement. Gradually collateral pathways opened and his visual symptom improved accordingly. However, another cardiogenic embolus or a secondary embolus originated from the dead end of the distal ICA occluded the proximal ECA. Due to suddenly decreased anterograde perfusion pressure secondary to proximal ECA occlusion, collateral flow to the retina from the ECA became suboptimal, which might have caused total blindness of his right eye. Our hypothesis is that minimal necessary blood flow to the retina to enable vision was maintained by collaterals from the ECA for as long as 4 h and 40 min and that the retina survived even more severe ischemia to cause blindness for 3 h and 32 min, during which only limited anterograde collateral perfusion into the right ophthalmic artery via the anterior communicating artery or the right posterior communicating artery maintained the right retina. Although very low flow may not be detectable with MRA despite an open vessel [Figure 2b], this might have left possibility of salvage from total blindness. Digital subtraction angiogram could have detected extremely limited flow of the right ophthalmic artery via collaterals, and might have been useful to support this hypothesis, although we did not perform it in order to start the treatment as soon as possible.

It is rare that patients with acute ICA and ECA occlusion manifest with only visual symptoms and are free from any motor or sensory deficits. We present a rare case in which acute reperfusiw of the retina restored practical vision after more than 8 h after onset of the symptoms or 3.5 h after assumed severe ischemia of the retina. There may be a chance to restore vision in patients with CRAO via investigation and recanalization of extra- or intracranial major arteries, especially when the patient is known to have stroke risk factors such as Af or major extra- or intracranial artery stenosis.

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