Central Retinal Artery Occlusion after the Endovascular Treatment of Unruptured Ophthalmic Artery Aneurysm: A Case Report and a Literature Review

Alaa Mohammed Elkordy,^{1,2,3,4} Kenichi Sato,³ Yasuhide Inoue,³ Yui Mano,³ Yasushi Matsumoto,³ Akira Takahashi,² and Teiji Tominaga¹

Endovascular coil embolization for ophthalmic artery (OphA) aneurysms has a risk of occlusion of the OphA, which can lead to loss of vision. The authors report a patient with unruptured OphA aneurysm which treated with endovascular coiling and were complicated by blindness due to OphA thromboembolic occlusion after the procedure. The OphA successfully recanalized using local intra-arterial fibrinolysis with complete regain of visual acuity. The risk of visual loss due to thromboembolic complications cannot be ignored during endovascular coiling of the OphA aneurysm despite of good retrograde flow during OphA occlusion test using a balloon catheter. Rapid intervention is required for recovering visual disturbance in such a situation.

Keywords: central retinal artery, complication, endovascular therapy, local intra-arterial fibrinolysis, ophthalmic artery aneurysm

Introduction

Endovascular treatment of the internal carotid artery (ICA) paraclinoid aneurysms has been proposed as an alternative to surgical clipping.¹⁻⁵⁾ Nevertheless, the risk of impaired vision due to unexpected thromboembolism or occlusion of the ophthalmic artery (OphA) after coiling is still a major concern.⁶⁻⁸⁾ Incidence of new visual deficits in paraophthalmic aneurysms treated surgically ranged from 4.8 to 7.5%. 9,10) The neurointerventional therapies have done little to reduce these risks as the incidence of visual complications following endovascular coiling of paraophthalmic aneurysms are estimated to be approximately 5%. 7,10,111) Rouchaud et al. found that the coverage of the OphA by flow diverters is not a harmless treatment because 17.9% of patients had permanent ophthalmic complications. These complications can be mild or severe, but some are only detected with a complete and extensive ophthalmic examination, which was not performed in most previous studies.¹²⁾ We present a case of an

Departments of ¹Neurosurgery and ²Neuroendovascular Therapy, Tohoku University Graduate School of Medicine, Sendai, Japan ³Department of Neuroendovascular Therapy, Kohnan Hospital,

Received: September 21, 2015; Accepted: February 11, 2016

OphA aneurysm that complicated by OphA occlusion with blindness after endovascular coiling and successfully recanalized by intra-arterial thrombolysis.

Case Report

Presentation

A 62-year-old female diagnosed by magnetic resonance angiography (MRA) in 2007 to have a right OphA aneurysm measuring about 5 mm in diameter. Follow-up MRA in 2015 showed enlargement of the aneurysm. Catheter angiography was performed and demonstrated asymptomatic increase in size of right OphA aneurysm to measure $6.9 \times 5.9 \times 4.6$ mm with wide neck measuring 6 mm and denoted that the right OphA was originating from the body of the aneurysm (Fig. 1).

Intervention

Therapeutic decision was reached through multidisciplinary deliberation of both neurosurgeons and nonsurgical neurointerventionalists, and informed consent was obtained from the patient and her family. Balloon-assisted coiling was elected to attempt dense packing of the wide necked aneurysm. A 75 mg of clopidogrel and a 100 mg of aspirin were administered 4 days before the procedure. After general anesthesia and femoral puncture, systemic heparinization was started with a loading dose of 5,000 units and maintained with a dose of 1,000 units hourly and the activated clotting time (ACT) kept above 250 s throughout the procedure. The right femoral artery was used to place a 6F guiding catheter (Fubuki; ASAHI-INTECC, Aichi, Japan) indwelled into the right cervical ICA. Excelsior SL-10 microcatheter (Stryker, Kalamazoo, MI, USA) and one balloon catheter (4 × 10 mm Scepter C balloon; Microvention, Tustin, CA, USA) were then positioned in the ICA clinoid segment. Balloon test occlusion (BTO) of the OphA was performed using Scepter C balloon inflated in the right ICA at the aneurysm neck, and then we performed cerebral angiography through the guiding catheter, which demonstrated right OphA reconstitution with a retinochoroidal blush through retrograde flow from the frontal branch of the right superficial temporal artery (Fig. 2).

The aneurysm was packed with coils and the last right ICA angiography showed patency of the right OphA and intraaneurysmal body filling. The heparin was reversed with protamine sulfate, and immediately after the procedure, the

⁴Neuroendovascular section, Department of Neurology, Faculty of Medicine, Tanta University, Tanta, Egypt



Fig. 1 Right internal carotid artery three-dimensional reconstructed angiogram demonstrates a wide necked ophthalmic artery aneurysm with the ophthalmic artery originating from the body of the aneurysm.

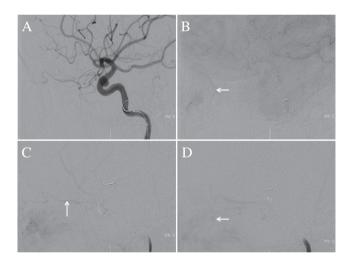


Fig. 2 (A, B) Right common carotid artery lateral angiograms reveal (A) anterograde filling of the ophthalmic artery in the mid arterial phase and (B) choroidoretinal blush in the late arterial phase (arrow). (C, D) Right common carotid artery lateral angiograms with balloon test occlusion reveal (C) complete blockage of the anterograde internal carotid arterial flow, retrograde filling of the ophthalmic artery via the external carotid artery branches in the mid arterial phase (arrow), and (D) choroidoretinal blush in the late arterial phase (arrow).

patient had no neurological deficit, and the patient was transferred to the recovery room.

Fifteen minutes later, the patient complained from blurred vision in her right eye. Although continuous heparin infusion was started, her visual disturbance progressed rapidly to complete blindness and total loss of light reflex in about 1 h after the endovascular procedure. The patient transferred emergently for cerebral angiography which demonstrated the complete occlusion of not only the treated aneurysm but also

the right OphA. The right external carotid artery (ECA) angiography revealed the disappearance of the retinochoroidal blush as seen in the OphA BTO. These findings suggested thromboembolic event occurring through the right OphA (Fig. 3).

Endovascular procedure for the right OphA occlusion was performed under local anesthesia and systemic heparinization which started with a loading dose of 5,000 units and was maintained with a dose of 1,000 units hourly keeping the ACT above 300 s. A 6F-guiding catheter placed into the right cervical ICA and a carnelian PIXIE (Tokai Medical Products, Aichi, Japan) with its distal tip placed close to the orifice of the right OphA confirmed by selective angiography. Mechanical disruption of the thrombus using a micro-guidewire and local intra-arterial fibrinolysis (LIF) using 1,20,000 units Urokinase was performed with evident recanalization of the OphA with a normal anterograde flow until the third segment (Fig. 4). At the same time the visual acuity of the right eye was improving until the patient restore normal vision through. Heparin reverse was not carried out the final ACT was 238 s. Postoperative systemic heparinization was continued for 48 h. Dual antiplatelet therapy was continued 6 months after the procedure.

Follow-up finding

The patient maintained normal visual acuity and the fundus photograph showed no ischemic lesion in her right retina.

Discussion

Periprocedural thromboembolic complications from endovascular treatment of intracranial aneurysms are estimated to occur in 2–15% of patients.¹³⁾ Aggressive treatment of acute intraprocedural thrombus formation with intra-arterial or intravenous administration of fibrinolytics

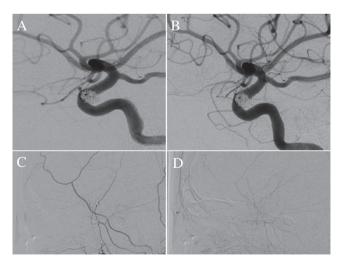


Fig. 3 (A) Right internal carotid artery lateral angiogram immediately after endovascular coiling reveals patent ophthalmic artery. (B) Right internal carotid artery lateral angiogram after the onset of the visual symptoms reveals ophthalmic artery occlusion. (C, D) Right external carotid angiograms after the onset of the visual symptoms reveal (C) no retrograde flow to the ophthalmic artery and (D) absent of choroidoretinal blush in the late arterial phase.

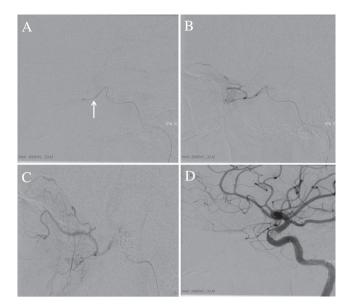


Fig. 4 (A) Microcatheter angiography before local fibrinolysis reveals occlusion of the right ophthalmic artery (arrow). (B) Microcatheter angiography during local fibrinolysis reveals incomplete recanalization of the right ophthalmic artery. (C) Microcatheter angiography postlocal fibrinolysis reveals complete recanalization of the right ophthalmic artery. (D) Right internal carotid lateral angiogram post-local fibrinolysis reveals normal anterograde flow to the ophthalmic artery.

or glycoprotein IIb/IIIa (GpIIb/IIIa) inhibitors has become a standard treatment for these complications. Recanalization of the treated artery is usually essential to avoid permanent neurological deficit.¹³⁾ Unfavorable aneurysmal configurations, including wide-neck lesions and those in which OphA entry is incorporated into the aneurysmal neck or the related ophthalmologic morbidities constitute a persistent challenge for endovascular coiling of OphA aneurysms.¹⁴⁾

BTO seems helpful in planning treatment strategies for coiling of OphA aneurysms that preserve vision. 30 Collateral channels between the branches of the ECA and OphA prevent permanent blindness after the occlusion of the OphA in 90% of patients. 15,16) Thrombotic occlusion at the OphA orifice is a major concern during the coil embolization procedure itself and in the follow-up period. 17) Although the probability of patent collaterals is high, however, the potential risk of ocular ischemia after coiling cannot be ignored.¹⁷⁾ Anterograde ophthalmic flow can be obliterated by thromboembolism during coil embolization without direct coil protrusion into the orifice of the OphA. Acknowledgment that tiny thrombi can occlude the central retinal artery regardless of the collateral flow, prompt handling of unexpected thromboembolic complications is vital to salvage visual function.18)

Visual prognosis in patients with central retinal artery occlusion (CRAO) has been reported to be very poor, because up to 92% have permanent loss of vision, with a final visual acuity of counting fingers of less. ^{19–26)} Visual acuity at the time of initial presentation ranges from counting fingers to light perception in 74–90% of eyes. ^{21,26)}

Conventional treatment modalities for CRAO include ocular massage, anterior chamber paracentesis, intraocular pressurelowering agents (e.g., mannitol, acetazolamide, and topical agents), hyperbaric oxygen, anticoagulants, and hemodilution, although efficacy regarding visual outcome of these treatments have not been clearly proved. 19) Several case series^{27,28,29)} and a meta-analysis³⁰⁾ indicated that LIF may improve visual outcome in patients with CRAO. Potential side effects of LIF include hemorrhagic or ischemic complication of the brain. For this reason, the benefit of LIF has also not generally been widely reported. Arnold et al. 19) proved the efficacy of LIF for the acute CRAO within 6 h after the onset compared with conventional therapy. They injected Urokinase, in a dose ranging from 1,00,000 units to 10,00,000 units (mean dose 6,77,000 units) manually through a microcatheter placed in the proximal segment of the OphA. Prospective randomized multicenter clinical trial in Europe to compare treatment outcome after conservative standard treatment with LIF using recombinant tissue plasminogen activator for patients with CRAO within 20 h after the onset was stopped after the first interim analysis because of apparently similar efficacy and the higher rate of adverse reactions in LIF group.31) These data suggest that LIF for CRAO should be performed by skilled neurointerventionalists as soon as possible after the onset.

In our case, the BTO revealed intact collateral circulation between the ECA and OphA and the immediate post-coiling angiography showed preserved anterograde flow of the OphA. However, rapid reverse of systemic heparinization by protamine sulfate might induce progressing thrombosis in coiled intraaneurysmal sac, which involved in the origin of the right OphA. The progressing thrombosis reached the third segment of the OphA, and that is the reason the collateral flow from the ECA could not work. The patient suffered from complete visual loss in the right eye. Urokinase infusion through the microcatheter placed in the origin of the right OphA successfully dissolved the thrombus and made the OphA recanalized. The patient's visual acuity completely recovered.

Conclusion

We need to highlight that during endovascular coiling of the OphA aneurysm the risk of visual loss due to thromboembolic complications cannot be ignored despite good retrograde flow during OphA occlusion test by a balloon catheter. Rapid endovascular intervention is required for recovering of visual disturbance in such a situation.

Conflicts of Interest Disclosure

The authors have no disclosure to report.

References

- Iihara K, Murao K, Sakai N, Shindo A, Sakai H, Higashi T, Kogure S, Takahashi JC, Hayashi K, Ishibashi T, Nagata I: Unruptured paraclinoid aneurysms: a management strategy. *J Neurosurg* 99: 241–247, 2003
- Kanagalingam S, Gailloud P, Tamargo RJ, Subramanian PS, Miller NR: Visual sequelae after consensus-based treatment of ophthalmic artery segment aneurysms: the Johns Hopkins experience. *J Neur-oophthalmol* 32: 27–32, 2012

- 3) Yadla S, Campbell PG, Grobelny B, Jallo J, Gonzalez LF, Rosenwasser RH, Jabbour PM: Open and endovascular treatment of unruptured carotid-ophthalmic aneurysms: clinical and radiographic outcomes. *Neurosurgery* 68: 1434–1443, 2011
- 4) Heran NS, Song JK, Kupersmith MJ, Niimi Y, Namba K, Langer DJ, Berenstein A: Large ophthalmic segment aneurysms with anterior optic pathway compression: assessment of anatomical and visual outcomes after endovascular coil therapy. *J Neurosurg* 106: 968–975, 2007
- Hoh BL, Carter BS, Budzik RF, Putman CM, Ogilvy CS: Results after surgical and endovascular treatment of paraclinoid aneurysms by a combined neurovascular team. *Neurosurgery* 48: 78–89, 2001
- Castillo B Jr, De Alba F, Thornton J, DeBrun G, Pulido J: Retinal artery occlusion following coil embolization of carotid-ophthalmic aneurysms. *Arch Ophthalmol* 118: 851–852, 2000
- 7) Ferrell AS, Lessne ML, Alexander MJ, Shah P, Golshani K, Zomorodi A, Smith TP, Britz GW: Visual complications after stentassisted endovascular embolization of paraophthalmic and suprasellar variant superior hypophyseal aneurysms: the Duke Cerebrovascular Center experience in 57 patients. World Neurosurg 78: 289– 294, 2012
- Sherif C, Gruber A, Dorfer C, Bavinzski G, Standhardt H, Knosp E: Ruptured carotid artery aneurysms of the ophthalmic (C6) segment: clinical and angiographic long-term follow-up of a multidisciplinary management strategy. J Neurol Neurosurg Psychiatry 80: 1261–1267, 2009
- Day AL: Aneurysms of the ophthalmic segment: a clinical and anatomical analysis. J Neurosurg 72: 677–691, 1990
- 10) Fulkerson DH, Horner TG, Payner TD, Leipzig TJ, Scott JA, DeNardo AJ, Redelman K, Goodman JM: Results, outcomes, and follow-up of remnants in the treatment of ophthalmic aneurysms: a 16-year experience of a combined neurosurgical and endovascular team. *Neurosurgery* 64: 218–229, 2009
- Turner RD, Byrne JV, Kelly ME, Mitsos AP, Gonugunta V, Lalloo S, Rasmussen PA, Fiorella D: Delayed visual deficits and monocular blindness after endovascular treatment of large and giant paraophthalmic aneurysms. *Neurosurgery* 63: 469–474, 2008
- 12) Rouchaud A, Leclerc O, Benayoun Y, Saleme S, Camilleri Y, D'Argento F, Boncoeur MP, Robert PY, Mounayer C: Visual outcomes with flow-diverter stents covering the ophthalmic artery for treatment of internal carotid artery aneurysms. Am J Neuroradiol 36: 330–336, 2015
- 13) Ries T, Siemonsen S, Grzyska U, Zeumer H, Fiehler J: Abciximab is a safe rescue therapy in thromboembolic events complicating cerebral aneurysm coil embolization: single center experience in 42 cases and review of the literature. Stroke 40: 1750–1757, 2009
- 14) Ahn JH, Cho YD, Kang HS, Kim JE, Cho WS, Jung SC, Kim CH, Han MH: Endovascular treatment of ophthalmic artery aneurysms: assessing balloon test occlusion and preservation of vision in coil embolization. Am J Neuroradiol 35: 2146–2152, 2014
- Schmidt GW, Oster SF, Golnik KC, Tumialán LM, Biousse V, Turbin R, Prestigiacomo CJ, Miller NR: Isolated progressive visual loss after coiling of paraclinoid aneurysms. Am J Neuroradiol 28: 1882–1889, 2007

- 16) Andaluz N, Beretta F, Keller JT, Zuccarello M: Aneurysms of the Ophthalmic (C6) segment of the internal carotid artery: clinical experience, treatment options, and strategies (Part 2). *Neurosurgery* 15: 91–102, 2005
- 17) Durst CR, Starke RM, Gaughen J, Nguyen Q, Patrie J, Jensen ME, Evans AJ: Vision outcomes and major complications after endovascular coil embolization of ophthalmic segment aneurysms. Am J Neuroradiol 35: 2140–2145, 2014
- 18) Kim B, Jeon P, Kim K, Yang N, Kim S, Kim H, Byun H, Jo KI: Endovascular treatment of unruptured ophthalmic artery aneurysms: clinical usefulness of the balloon occlusion test in predicting vision outcomes after coil embolization. J NeuroIntervent Surg 1–7, 2015
- 19) Arnold M, Koerner U, Remonda L, Nedeltchev K, Mattle HP, Schroth G, Sturzenegger M, Weber J, Koerner F: Comparison of intraarterial thrombolysis with conventional treatment in patients with acute central retinal artery occlusion. J Neurol Neurosurg Psychiatry 76: 196–199, 2005
- Augsburger JJ, Magargal LE: Visual prognosis following treatment of acute central retinal artery obstruction. Br J Ophthalmol 64: 913–917, 1980
- Brown GC, Magargal LE: Central retinal artery obstruction and visual acuity. Ophthalmology 89: 14–19, 1982
- 22) Arruga J, Sander MD: Ophthalmologic findings in 70 patients with evidence of retinal embolism. *Ophthalmology* 89: 1336–1347, 1982
- Atebara NH, Brown GC, Cater J: Efficacy of anterior chamber paracentesis and carbon in treating acute nonarteritic central retinal artery occlusion. Ophthalmology 102: 2029–2034, 1995
- 24) Mueller AJ, Neubauer AS, Schaller U, Kampik A; European Assessment Group for Lysis in the Eye: Evaluation of minimally invasive therapies and rationale for a prospective randomized trial to evaluate selective intra-arterial lysis for clinically complete central retinal artery occlusion. *Arch Ophthalmol* 121: 1377–1381, 2003
- Rumelt S, Brown GC: Update on treatment of retinal arterial occlusions. Curr Opin Ophthalmol 14: 139–141, 2003
- 26) Hayreh SS, Zimmerman MB: Central retinal artery occlusion: visual outcome. Am J Ophthalmol 140: 376–391, 2005
- 27) Ahn SJ, Kim JM, Hong JH, Woo SJ, Ahn J, Park KH, Han MK, Jung C: Efficacy and safety of intra-arterial thrombolysis in central retinal artery occlusion. *Invest Ophthalmol Vis Sci* 54: 7746–7755, 2013
- 28) Ahn SJ, Kim JM, Hong JH, Woo SJ, Ahn J, Park KH, Han MK, Jung C: Treatment of retinal arterial occlusion with local fibrinolysis using recombinant tissue plasminogen activator. *Ophthalmology* 106: 768–773, 1995
- Schmidt DP, Shulte-Monting J, Schumacher M: Prognosis of central retinal artery occlusion: local intraarterial fibrinolysis versus conservative treatment. Am J Neuroradiol 23: 1301–1307, 2002
- 30) Beatty S, Au Eong KG: Local intra-arterial fibrinolysis for acute occlusion of the central retinal artery: a meta-analysis of the published data. *Br J Ophthalmol* 84: 914–916, 2000
- 31) Schumacher M, Schmidt D, Jurklies B, Gall C, Wanke I, Schmoor C, Maier-Lenz H, Solymosi L, Brueckmann H, Neubauer AS, Wolf A, Feltgen N; EAGLE-Study Group.; EAGLE-Study Group: Central retinal artery occlusion: local intra-arterial fibrinolysis versus conservative treatment, a multicenter randomized trial. *Ophthalmology* 17: 1367–1375, 2010