pISSN 1738-6586 / eISSN 2005-5013 / J Clin Neurol 2023;19(3):324-326 / https://doi.org/10.3988/jcn.2022.0366

Blindsided: An Unusual Case of Ocular Ischemic Syndrome and Orthostatic Dizziness Caused by Brachiocephalic Trunk Stenosis

Yoon Seob Kim[a]
Je Hwan Won[b]
Seong-Joon Lee[a]

Departments of [a]Neurology and [b]Radiology, Ajou University School of Medicine, Suwon, Korea

Dear Editor,

We present a rare case of ocular ischemic syndrome (OIS) that arose from severe brachiocephalic trunk (BCT) stenosis, with preceding symptoms of orthostatic hypotension (OH), which was treated with BCT stenting.

A 60-year-old male truck-driver presented with orthostatic dizziness, followed by right monocular visual loss. He had histories of diabetes mellitus, hypertension, dyslipidemia, and coronary artery disease. Severe dizziness episodes occurred first, when getting down from the vehicle after driving, and lasted for 2−3 minutes. His right radial pulse was very weak. Initial carotid computed tomography (CT) angiography revealed severe right BCT stenosis (>90%), occlusion at the left proximal vertebral artery (VA) that was reconstituted by cervical collaterals at the V2 segment, and moderate vertebrobasilar junction stenosis (Fig. 1C). The anterior communicating artery was visible whereas the posterior communicating artery was not. Carotid duplex findings were unremarkable, with normal flow through the tested portions of the bilateral internal carotid artery and VA. Transcranial Doppler ultrasonography revealed increased mean flow velocities at the basilar artery stenotic portion (127 cm/sec), left anterior cerebral artery (117 cm/sec), and left VA (86 cm/sec), suggesting the presence of flow diversion from the neighboring vasculature to the right BCT territory. Flow reversal was not observed in the bilateral VA and ophthalmic artery. Based on the above results, the vertebrobasilar artery territory in the patient lacked primary collaterals and was highly reliant on the severely stenosed BCT, while partially being supported by secondary collaterals recruited from the cervical arteries to the occluded left VA. The tilt-table test revealed a diastolic blood pressure drop of 17 mm Hg after 65 sec of standing which then spontaneously recovered, indicating transient diastolic OH but did not meet the criteria for classic OH. The patient was managed using dual antiplatelets and high-dose statins.

After 6 months, the patient complained of seeing black lines that impeded vision in his right eye. The symptoms were more severe after rigorous exercise and hot baths. An ophthalmologic workup revealed retinal hemorrhage (Fig. 1A). His eyesight presented asymmetrically decreased visual acuity (OD/OS: 0.16/1.0). His visual acuity decreased on follow-up examinations, which revealed an increased intraocular pressure (IOP) of 28 mm Hg with iris neovascularization. Wide fundus angiography revealed delayed filling in the right eye, with retinal arterial staining and microaneurysm (Fig. 1B). The patient was diagnosed with neovascular glaucoma and OIS. The severe right BCT stenosis was considered to be responsible. Based on the cerebral hemodynamic condition of the patient, we presumed that the cause of his dizziness could also be attributed to his transient OH combined with a vertebrobasilar transient ischemic attack (VBTIA).

Angioplasty and stent insertion was performed at the right BCT. A 4-mm filter was de-

Received September 23, 2022
Revised December 14, 2022
Accepted January 16, 2023

Correspondence
Seong-Joon Lee, MD, PhD
Department of Neurology, Ajou University School of Medicine, 164 World cup-ro, Yeongtong-gu, Suwon 16499, Korea
Tel +82-31-219-5175
Fax +82-31-219-5178
E-mail editisan@gmail.com

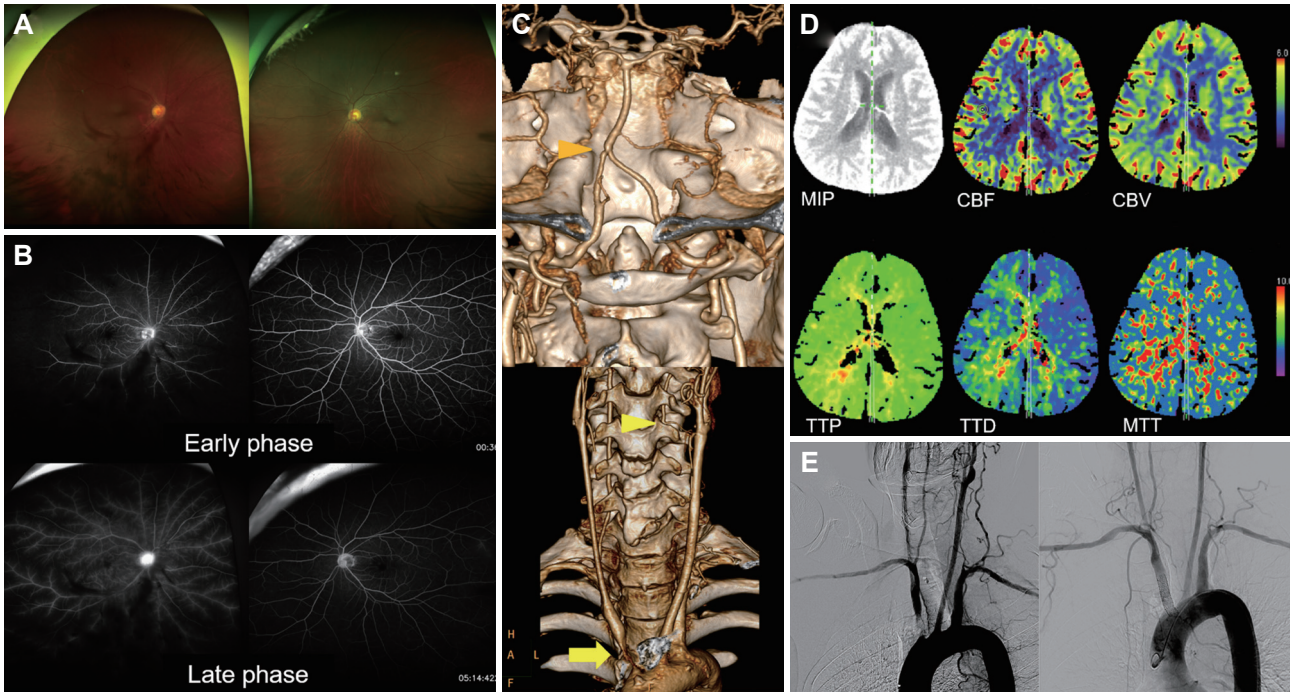

Fig. 1. Clinical and imaging findings of the patient. A: Bilateral fundus photography revealing retinal hemorrhage in the right eye. B: Wide fundus angiography revealing delayed filling in the retina of the right eye, with arterial staining and microaneurysms. C: CT angiography of the intracranial and extracranial vessels revealing severe right brachiocephalic trunk stenosis (yellow arrow) with occlusion of the contralateral vertebral artery that is reconstituted by cervical collaterals at the V2 level (yellow arrowhead), and moderate vertebrobasilar junction stenosis (orange arrowhead). D: CT perfusion images revealing mild perfusion delay in the right hemisphere border zone area. E: Stent insertion was performed at the right brachiocephalic trunk. CBF, cerebral blood flow; CBV, cerebral blood volume; CT, computed tomography; MIP, maximal intensity projection; MTT, mean transit time; TTD, time to drain; TTP, time to peak.

ployed at the right VA via the brachial artery. A 7-mm filter was deployed at the right internal carotid artery via the femoral artery, and an 8-mm-×-37-mm balloon-expandable stent was placed on the BCT (Fig. 1D).

The patient complained of a throbbing ocular pain and increased IOP after the procedure. He was treated using acetazolamide, and the pain subsided and the IOP decreased. His visual acuity did not recover after discharge, but ocular pain did not recur. The orthostatic dizziness also disappeared.

OIS manifests as visual loss, orbital pain, and various anterior and posterior segment signs such as iris neovascularization, secondary neovascular glaucoma, mid-peripheral retinal hemorrhages, and microaneurysms.[1] Clinicians must perform a thorough investigation of the carotid circulation when OIS is suspected. This case of OIS was caused by an artery that can easily blindside clinicians, namely the BCT. The current patient presented with three main characteristics: 1) fluctuating visual deficits, 2) diminished ipsilateral radial pulse, and 3) orthostatic dizziness, which led us to identify and confirm the presence of the culprit BCT stenosis.

It may be reasonable to consider that the orthostatic decrease in cerebral blood flow caused the VBTIA since the blood flow to the posterior circulation was largely dependent on the critically stenotic right BCT flow due to the contralateral VA occlusion. His dizziness also lasted unusually long and was unusually severe. While there are rare reports of orthostatic cerebral ischemia in the posterior circulation, there have been some[2] that were associated with severe vertebrobasilar steno-occlusions[3] and MRI evidence of acute ischemia. Symptom improvement after BCT stenting also supports the combined nature of OH and VB TIA in this patient.

The progression of neovascular glaucoma was irreversible even after revascularization. The abrupt ocular reperfusion instead caused increases in IOP and ocular pain, which has been described in previous reports of neovascular glaucoma aggravation after carotid reperfusion.[4] Such a phenomenon was hypothesized to arise from a sudden increase in aqueous humor production after reperfusion, combined with insufficient resorption caused by a long period of hypoperfusion.[5] However, if performed in a timely manner, revascularization procedures may stabilize or restore visual acuity,[6] especially if they are performed before iris neovascularization and secondary glaucoma development.[1]

This case demonstrates that endovascular treatment can be a safe and effective strategy for BCT stenosis management.[7] Recent reports indicate that perioperative stroke rates are

4.3%, which is comparable to carotid interventions, which supports stenting as the first-line treatment for BCT lesions over open transthoracic surgery, which achieves mortality and morbidity rates of 3%−16% and 15%−25%, respectively.[8] While femoral access is also feasible,[9] retrograde carotid access with general anesthesia may be preferred[8] due to the high stiffness of the aortic arch angle.[10] Restenosis is relatively common, but may be prevented by using covered stents.[8]

Ethics Statement

This study followed the tenets of the Declaration of Helsinki and was performed according to the guidelines of the Ajou University Hospital Institutional Review Board.

Availability of Data and Material

Data sharing not applicable to this article as no datasets were generated or analyzed during the study.

ORCID iDs

Yoon Seob Kim https://orcid.org/0000-0002-6363-1999
Je Hwan Won https://orcid.org/0000-0002-5901-295X
Seong-Joon Lee https://orcid.org/0000-0001-9735-6369

Author Contributions

Conceptualization: Seong-Joon Lee. Data curation: Yoon Seob Kim. Formal analysis: Seong-Joon Lee. Resources: Je Hwan Won, Seong-Joon Lee. Supervision: Seong-Joon Lee. Writing—original draft: Yoon Seob Kim, Seong-Joon Lee. Writing—review & editing: all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Funding Statement

This work was supported by the Basic Science Research Program through the National Research Foundation of Korea (NRF), funded by the Ministry of Education (grant number NRF-2021R1I1A1A01048331; S.J.L.).

REFERENCES

1. Terelak-Borys B, Skonieczna K, Grabska-Liberek I. Ocular ischemic syndrome - a systematic review. *Med Sci Monit* 2012;18:RA138-RA144.
2. Caplan LR, Sergay S. Positional cerebral ischaemia. *J Neurol Neurosurg Psychiatry* 1976;39:385-391.
3. Kim GS, Park HK, Song P, Lee JJ, Cho JY. Vertebrobasilar insufficiency presenting as orthostatic cerebellar ataxia. *Ann Indian Acad Neurol* 2021;24:459-461.
4. Lee KM, Kim EJ, Heo SH, Jin KH. Paradoxical development of neovascular glaucoma following carotid angioplasty and stenting. *Interv Neuroradiol* 2016;22:540-543.
5. Katsuta T, Fujimoto A, Oba K. Deterioration of glaucoma after carotid endarterectomy. *Neurol Med Chir (Tokyo)* 2013;53:418-421.
6. Kawaguchi S, Iida J, Uchiyama Y. Ocular circulation and chronic ocular ischemic syndrome before and after carotid artery revascularization surgery. *J Ophthalmol* 2012;2012:350475.
7. Onishi S, Sakamoto S, Sadatomo T, Shimizu K, Hara T, Kurisu K. Stenting for innominate artery stenosis under double-filter protection of the anterior and posterior circulation via the right brachial artery. *Ann Vasc Surg* 2020;63:454.e5-454.e9.
8. Ammi M, Henni S, Salomon Du Mont L, Settembre N, Loubiere H, Sobocinski J, et al. Lower rate of restenosis and reinterventions with covered vs bare metal stents following innominate artery stenting. *J Endovasc Ther* 2019;26:385-390.
9. Hüttl K, Nemes B, Simonffy A, Entz L, Bérczi V. Angioplasty of the innominate artery in 89 patients: experience over 19 years. *Cardiovasc Intervent Radiol* 2002;25:109-114.
10. de Borst GJ. Innominate artery stenting: the continuing saga of "who, when, and how"? *J Endovasc Ther* 2019;26:391-393.