Circumpapillary collateral vessel development in iatrogenic central retinal artery occlusion observed using OCT angiography

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ABSTRACT
Purpose: It has been reported that peripapillary loops develop after central retinal artery occlusion (CRAO). Although cilioretinal anastomoses have been shown by fluorescein angiography (FA) and indocyanine green angiography, these examinations cannot confirm the structural continuity between the retinal arteries and the posterior ciliary arteries. In the current report, we followed a patient with iatrogenic CRAO in which circumpapillary collaterals formed and assessed the connection between these two vascular systems using optical coherence tomography angiography (OCTA).

Observation: Iatrogenic CRAO developed in the left eye of a 30-year-old woman during preoperative embolization of a convexity meningioma. FA and OCTA showed complete impairment of the retinal circulation. Two weeks after this event, OCTA images showed flow in the retinal vessels and the beginning of collateral vessel development on the optic disc margin. Six months later, OCTA images showed that these circumpapillary collaterals connected branches of the retinal arteries with the vessels supplying the optic disc, which originate from the posterior ciliary arteries.

Conclusion and importance: OCTA aided our understanding of the three-dimensional configuration of the circumpapillary collaterals that developed after iatrogenic CRAO, which included anastomosis of the retinal and posterior ciliary artery systems. OCTA is useful for noninvasively monitoring the status of retinal circulatory dynamics after iatrogenic CRAO.

1. Introduction
Central retinal artery occlusion (CRAO), one of the common causes of permanent visual loss, can result from obstruction of blood flow from thrombotic, inflammatory, or traumatic causes or from iatrogenic causes during treatments such as embolization and cosmetic facial filler injections. It has been shown in both research and clinical settings that if vascular occlusion persists, tortuous collaterals may form which link blocked vessels with patent vessels nearby. While this is more common in branch retinal artery occlusion, and less common in CRAO, several reports have documented circumpapillary anastomotic vessels purportedly representing cilioretinal collateral circulation after CRAO. Although cilioretinal anastomoses have supposedly been seen during simultaneous infusion with choroidal flush on fluorescein angiography (FA), the technology cannot confirm the structural continuity between the retinal arteries and the posterior ciliary arteries. Optical coherence tomography angiography (OCTA) provides three-dimensional, chorioretinal microvascular images based on flow signals. In the current report, we followed a patient with iatrogenic CRAO in which circumpapillary collaterals formed and assessed the connection between retinal and posterior ciliary vascular systems three-dimensionally using OCTA (RTVue XR Avanti, Optovue, Fremont CA, USA; Triton DRI-OCT, Topcon, Tokyo, Japan, and Cirrus HD-OCT 5000, Carl Zeiss Meditec, Dublin, CA, USA).

2. Case report
A 30-year-old woman diagnosed with a convexity meningioma underwent preoperative embolization. She was generally healthy and had high myopia bilaterally (refractive error, –7 diopters). The best-cor-

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Rectified visual acuity in both eyes was 20/20. Under local anesthesia, transfemoral access was obtained with selective catheterization of the external carotid artery. A tumor was present in the parietal lobe, which was supplied by the left middle meningeal artery. The neurosurgeon confirmed that the catheter was advanced into a parietal branch of the middle meningeal artery and injected 300–500 μm embolic microspheres (Embosphere, Merit Medical Systems, South Jordan, UT, USA). During the procedure, the patient suddenly complained of acute visual loss of her left eye. The eye was found to have a relative afferent papillary defect and visual acuity had been reduced to light perception. The cherry-red spot with macular edema was seen in the left eye and the fellow eye was intact (Fig. 1a and b). She was treated immediately with ocular massage and hyperbaric oxygen therapy. FA and indocyanine green angiography (ICGA) were performed emergently (Fig. 1c–f). FA showed that, more than 20 seconds after dye injection, partial filling occurred only in the cilioretinal arteries; perfusion of the retinal arteries was impaired completely even in the late phase. The choroidal circulation also was delayed, but filling was observed on late-phase ICGA images. The OCTA image visualized only the proximal parts of the inferior retinal and cilioretinal arteries, while the OCTA image of the fellow eye visualized the retinal vessels and capillaries (Fig. 1g–j).

Two weeks later, although severe macular non-flow areas persisted, relatively large branches of the arterioles and venules were visualized by OCTA (Fig. 2b). We also found that a peripapillary vascular loop had developed (Fig. 2d). After 6 months, the retina was quite thin, but the major arterioles and venules were observed in the OCTA images (Fig. 2g). The peripapillary collaterals were developed and anastomosed with the superotemporal, superonasal, and inferonasal arterioles (Fig. 2i). En-face OCTA and corresponding flow-overlaid OCT B-scan images clearly showed collaterals connecting the retinal and posterior ciliary/choroidal circulation (Fig. 3-5a-d), and that the flow signals in these collaterals were in the deeper peripapillary margins (Figs. 3-5d). These collaterals supplied blood to the retina; however, the patient never recovered vision in this eye.

3. Discussion

In the current case, the central retinal and some of the short posterior ciliary arteries were occluded when embolization was performed through the middle meningeal artery, which is a branch of the external carotid artery system. This can be explained by the potential existence of anastomoses between the middle meningeal artery and the

Fig. 1. Images of both eyes obtained at the onset of iatrogenic central retinal artery occlusion. The right eye is intact (a), but the blood flow in the retinal arteries and veins of the left eye is severely impaired, and a cherry-red spot is present with macular edema (b). Fluorescein angiography (FA) (c) and indocyanine green angiography (ICGA) (d) obtained in the early phase (20 seconds after injection). The FA image shows only the proximal cilioretinal arteries and segmental choroidal flush (c). ICGA also shows a segmental infusion defect of the choroidal circulation (d). FA (e) and ICGA (f) images obtained in the middle phase (5 minutes after injection). Dye infusion in the retinal arteries stops around the peripapillary regions. ICGA shows choroidal filling even in the regions where dye infusion is impaired in the early phase. The 8 × 8-mm optical coherence tomography angiography (OCTA) image obtained using the RTVue XR Avanti shows that the retinal vasculature is intact in the right eye (g) and the horizontal B-scan image at the fovea visualizes the normal foveal structure of the right eye (h). The 6 × 6-mm OCTA image of the left eye obtained using the Cirrus HD-OCT 5000 visualizes only the proximal portions of the inferior retinal and cilioretinal arteries (i). The flow-overlaid B-scan image (at the horizontal blue line in (i)) shows rarefaction of the flow signals within the retina (j). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

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ophthalmic artery. Mames et al. reported central retinal and posterior ciliary artery occlusion after particle embolization through the internal maxillary artery.\(^1\) Unlike that case, however, not all of the short posterior ciliary arteries were occluded in our case. ICGA showed that more than one short posterior ciliary artery in the choroidal circulation was patent (Fig. 1c–f), and that peripapillary collaterals developed later (Fig. 2f).

Occasional development of arterial collaterals after occlusion of the retinal arteries has been recognized for many years.\(^5\)–\(^9\) Previous studies have reported that a suitable pressure gradient in the alternative blood supply plays a fundamental role in the initial formation of the collateral pathways around the obstructed vessels.\(^5\) Furthermore, at the optic disc, the superficial nerve fiber layer receives its primary blood supply from branches of the central retinal artery, and the deeper parts of the disc are nourished by branches from the arterial circle of Zinn-Haller (ZHAC) that originate from the short posterior ciliary arteries.\(^11\),\(^12\)

Therefore, potential communicating channels between these two vascular networks might exist in the optic disc; one could hypothesize in the current case that such channels (if they exist) developed into peripapillary collaterals between the two systems because of the complete and prolonged occlusion of the central retinal artery by artificial emboli.

It has been reported previously that circumpapillary anastomotic collaterals develop after CRAO\(^7\)–\(^9\) and are referred to as Nettleship collaterals after the man who first described them.\(^8\) Simultaneous dye infusion in these collaterals and retinal arteries during choroidal flushing seen in the FA images proves that the pathways of these collaterals extend from the posterior ciliary arteries to the branches of the central retinal artery.\(^7\),\(^9\) Although this coincident phase flow is important for clarifying the connection between the two vascular systems,
two-dimensional FA is limited in terms of visualizing structural continuity, especially between the retinal arteries and posterior ciliary artery system that is under the retinal pigment epithelium.

In contrast, three-dimensional OCTA imaging provides depth information about vessels visualized and defined by flow signals. Tsuboi et al. demonstrated that flow-overlaid OCT B-scan was useful for confirming the connection of the shunt vessel between the retina and the choroid in an eye with central retinal vein occlusion after radial optic neurotomy. Using OCTA, we visualized the presence of the collaterals not only at the distal anastomotic points between the collaterals and the
branches of the arterioles in the retina (Figs. 3–5a, c) but also that the flow signals of the proximal collaterals are definitely in the deeper peripapillary regions, which are considered to be tissues supplied by the posterior ciliary arterial circulation (Figs. 3–5b, d).

In highly myopic eyes, Ishida et al. used OCTA to visualize the ZHAC and ZHAC-derived cilioretinal artery. Although we could not visualize all of the original vessels from which the collaterals arose, en-face OCTA and flow-overlaid B-scan images facilitated identification and tracing of the proximal portions of the collaterals arising from the ZHAC, which as stated is derived from the posterior ciliary artery circulation (Figs. 3–5b, d). Using OCTA, the case presented thus demonstrates structurally, for the first time, that these circumpapillary collaterals developed after CRAO.

A study limitation was the use of multiple OCTA devices during the follow-up. Because the patient had poor fixation, we used more than one device to obtain OCTA images with acceptable quality. Another clinical limitation was that the formation of the collateral vessels unfortunately may not have contributed to the improvement of visual acuity in the current eye with iatrogenic CRAO. However, this case report showed that OCTA was useful for monitoring noninvasively the status of the retinal circulatory dynamics after iatrogenic CRAO as described previously in eyes with typical CRAO. Moreover, in the current case, OCTA aided our understanding of the three-dimensional configuration of the circumpapillary collaterals that developed after iatrogenic CRAO.
Patient consent

The patient’s legal guardian consented to publication of the case in writing.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

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