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Focal choroidal excavation disappearing after successful treatment of type 2 choroidal neovascularization with intravitreal aflibercept



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ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Aflibercept Choroidal neovascularization Focal choroidal excavation Optical coherence tomography Vascular endothelial growth factor	<i>Purpose:</i> To report a case with focal choroidal excavation (FCE) accompanied by type 2 choroidal neovascularization (CNV), in which the CNV was successfully treated with intravitreal aflibercept, after which the FCE was no longer detectable. <i>Observations:</i> A conforming type of FCE was detected in the left eye of a 34-year-old Japanese man who visited our hospital for a second opinion regarding treatment for his right eye. Three months later, type 2 CNV developed in the region corresponding to the FCE in the left eye. After treatment with intravitreal aflibercept, the CNV disappeared, and FCE was no longer detected. No recurrence has been observed in the 12 months following the treatment.

Conclusions and Importance: Although the etiology of FCE has not been fully elucidated, we discussed the mechanisms underlying its occurrence, its association with CNV, and the disappearance of both conditions after treatment.

1. Introduction

Focal choroidal excavation (FCE), first described by Jampol et al., in 2006, is a relatively rare clinical entity according to optical coherence tomography findings.¹ FCE is characterized by a localized area of excavation of the choroid without any evidence of either posterior staphyloma or scleral ectasia. In the conforming type of FCE, the outer segments of the photoreceptors are attached to a concave retinal pigment epithelium (RPE), while in the nonconforming type, there exists a separation between the photoreceptor tips and the RPE. In most cases, FCE is relatively stable; however, it can be associated with comorbidities, such as central serous chorioretinopathy and age-related macular degeneration (AMD), including polypoidal choroidal vasculopathy.^{2–8} Additionally, it has recently been proposed that FCE should be included in the pachychoroid spectrum.^{8,9}

Here, we report a case with FCE accompanied by choroidal neovascularization (CNV) in a 34-year-old Japanese man. In this case, CNV was successfully treated with intravitreal aflibercept (IVA; Eylea®, Leverkusen, Germany), after which the FCE was no longer detected.

2. Case report

The patient visited our hospital to obtain a second opinion about treatment for his right eye. He had noticed decreased vision in the right eye 3 months earlier. At the first visit, his best-corrected visual acuity (BCVA) was 20/70 in the right eye (spherical equivalent: 4.375 diopters) and 20/16 in the left eye (spherical equivalent: 3.875 diopters). Cicatrized subretinal neovascularization without subretinal fluid or intraretinal cystic changes was noted in the right eye, whereas the conforming type of FCE was detected 500 μ m temporal to the foveal in the left eye (Fig. 1). He had smoked cigarettes for 15 years. There was no remarkable family medical history. General examination did not reveal any systemic abnormality.

Three months later, he re-visited our hospital due to decreased vision in the left eye. The BCVA had decreased to 20/30 in the left eye. Ophthalmic examination confirmed type 2 CNV with subretinal fluid in the region corresponding to the FCE in the left eye (Fig. 2). After obtaining informed consent for the procedure, IVA was performed monthly for 3 months in the left eye. The study was conducted with the approval of the Institutional Review Board of Hayashi Eye Hospital and adhered to the tenets of the Declaration of Helsinki.

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Fig. 1. Fundus photographs showing subretinal cicatrized neovascularization in the right eye (A) and focal pigmentary changes in the left eye (B) of a 34-year-old male patient. Fundus autofluorescence images showing hypo- and hyper-autofluorescence corresponding to the cicatrized neovascularization in the right eye (C) and hypo-autofluorescence corresponding to the pigmentary changes in the left eye (D). Swept-source optical coherence tomography images showing subretinal cicatrized neovascularization in the right eye (F and G) and conforming type of focal choroidal excavation 500 µm distal to the fovea in the left eye (F and H).

0 µm 25 50 75 100 125 150 175 200 225 250 275 300 325 350 375 400 425 450 475 500

American Journal of Ophthalmology Case Reports 22 (2021) 101078

After the first IVA treatment, the CNV disappeared and the FCE was no longer detected. No recurrence has been observed at 12 months after the third IVA and BCVA has improved to 20/20 in the left eye (Fig. 3).

3. Discussion

We present a case with FCE accompanied by type 2 CNV. The clinical features reported are in good agreement with those of previous reports; that is, FCE accompanied by type 2 CNV was found in a young, myopic, Asian patient and was successfully treated with anti-vascular endothelial growth factor therapy.^{2,7–9}

The etiology of FCE in this case remains unclear. Some investigators

speculated that FCE may be a congenital posterior segment malformation; in contrast, others mentioned that FCE found in adults is more likely to involve an acquired type.^{1,2,10} In the posterior region of the eye, collagenous layers include the internal limiting membrane, Bruch's membrane, and the sclera. Scleral weakness may be associated with posterior staphyloma. Similarly, if Bruch's membrane was degraded for any reason, the overlying RPE and the outer retina may herniate outward due to intraocular pressure. This is supported by the following evidence: (1) the internal limiting membrane is not dislocated in eyes with FCE; and (2) nonconforming type of FCE with mild choroidal vascular leakage balanced with intraocular pressure often shows repositioning of the detached pigment epithelium or retina into the original



Fig. 2. Fundus photograph and autofluorescence image of the left eye showing an exudative lesion (A) and its corresponding hypoautofluorescence (B). Fluorescein (C) and indocyanine green (D) angiography images of the left eye showing fluorescein dye leakage in the center of the macula. Swept-source optical coherence tomography image of the left eye showing type 2 choroidal neovascularization arising from the location corresponding to the focal choroidal excavation, accompanied by subretinal fluid (E and F). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)



Fig. 3. Fundus photograph and autofluorescence image in the left eye showing pigmentary change in the area temporal to the fovea (A) and its corresponding hypoand hyper-autofluorescence (B). Swept-source optical coherence tomography image in the left eye showing neither focal choroidal excavation nor choroidal neovascularization after treatment (C and D).

curvature.⁹ Accordingly, we speculate that FCE development in this case may have been associated with the degradation of Bruch's membrane due to focal inflammation or other causes.

Most FCE lesions remain stable, although CSC and AMD may occur concurrently.^{2–8} In this case, FCE was accompanied by focal RPE alterations without remarkable pachychoroid in the left eye. Since cicatrized subretinal neovascularization had already formed in the right eye prior to the first visit, the preexistence of FCE was unknown. The patient had a smoking habit, which may have damaged the RPE.¹¹ RPE atrophy can then cause corresponding defects of both the choriocapillaris and Bruch's membrane.¹² Defects of Bruch's membrane can lead to FCE formation, while defects of the choriocapillaris may induce CNV development in the region corresponding to the FCE. Thus, in this case, formation of FCE, CNV, or both may have been related to smoking-related oxidative stress. We have now advised this patient to quit smoking and wear sunglasses according to the guidelines for AMD.

A novel finding in the case was that FCE was no longer detectable after the IVA treatment. There is no established theory that explains how FCE disappeared after the treatment of type 2 CNV with IVA. However, one possibility is that, after the successful treatment, collagenous material in the fibrovascular scar may have bridged the defect in Bruch's membrane, resulting in resolution of the FCE (Fig. 4).

Previous studies^{2,4,7,13–17} reported that 54 out of 76 eyes with FCE underwent anti-vascular endothelial growth factor therapy for CNV, including polypoidal choroidal vasculopathy, and showed good anatomical response to the treatment. While changes from non-conforming to conforming FCE were observed in some cases following

successful CNV treatment,^{15,17} an absence of change after successful treatment was seen in others.^{4,13} Nonetheless, this case report is the first to show the disappearance of FCE after successful CNV treatment.

4. Conclusion

We here reported a case with FCE accompanied by type 2 CNV, in which the CNV was successfully treated with IVA, and the FCE was no longer detected after this treatment. Although FCE is a relatively rare disease and its etiology has not been fully elucidated, we hope that this case report will assist other clinicians in the management of FCE.

Patient consent

Oral consent to publish the case report was obtained from the patient, although this report does not contain any personal information that could lead to the identification of the patient.

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Authorship

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



Fig. 4. Schematic diagram showing our speculations on how focal choroidal excavation (FCE) occurred, how choroidal neovascularization (CNV) developed, and how both conditions disappeared after CNV treatment. RPE: retinal pigment epithelium.

Declaration of competing interest

None.

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References

- Jampol LM, Shankle J, Schroeder R, Tornambe P, Spaide RF, Hee MR. Diagnostic and therapeutic challenges. *Retina*. 2006;26(9):1072–1076.
- Margolis R, Mukkamala SK, Jampol LM, et al. The expanded spectrum of focal choroidal excavation. Arch Ophthalmol. 2011;129(10):1320–1325.
- **3.** Ellabban AA, Tsujikawa A, Ooto S, et al. Focal choroidal excavation in eyes with central serous chorioretinopathy. *Am J Ophthalmol.* 2013;156(4):673–683.
- **4** Shinojima A, Kawamura A, Mori R, Yuzawa M. Morphologic features of focal choroidal excavation on spectral domain optical coherence tomography with simultaneous angiography. *Retina*. 2014;34(7):1407–1414.
- Wang Y, Chen ZQ, Wang W, Fang XY. Multimodal imaging evaluations of focal choroidal excavations in eyes with central serous chorioretinopathy. *J Ophthalmol.* 2016;2016:7073083.
- 6. Lim FP, Wong CW, Loh BK, et al. Prevalence and clinical correlates of focal choroidal excavation in eyes with age-related macular degeneration, polypoidal choroidal vasculopathy and central serous chorioretinopathy. *Br J Ophthalmol.* 2016;100(7): 918–923.

- **7.** Lee JH, Lee WK. Choroidal neovascularization associated with focal choroidal excavation. *Am J Ophthalmol.* 2014;157(3):710–718.e1.
- Chung HC, Byeon SH, Freund KB. Focal choroidal excavation and its association with pachychoroid spectrum disorders: a review of the literature and multimodal imaging findings. *Retina*. 2017;37(2):199–221.
- Cheung CMG, Lee WK, Koizumi H, Dansingani K, Lai TYY, Freund KB. Pachychoroid disease. *Eye.* 2019;33(1):14–33.
- Park KA, Oh SY. The absence of focal choroidal excavation in children and adolescents without retinal or choroidal disorders or ocular trauma. *Eye.* 2015;29 (6):841–842.
- Datta S, Cano M, Ebrahimi K, Wang L, Handa JT. The impact of oxidative stress and inflammation on RPE degeneration in non-neovascular AMD. *Prog Retin Eye Res.* 2017;60:201–218.
- Kurihara T, Westenskow PD, Bravo S, Aguilar E, Friedlander M. Targeted deletion of Vegfa in adult mice induces vision loss. J Clin Invest. 2012;122(11):4213–4217.
- Lim FP, Loh BK, Cheung CM, Lim LS, Chan CM, Wong DW. Evaluation of focal choroidal excavation in the macula using swept-source optical coherence tomography. *Eye*. 2014;28(9):1088–1094.
- Obata R, Takahashi H, Ueta T, Yuda K, Kure K, Yanagi Y. Tomographic and angiographic characteristics of eyes with macular focal choroidal excavation. *Retina*. 2013;33(6):1201–1210.
- Kuroda Y, Tsujikawa A, Ooto S, et al. Association of focal choroidal excavation with age-related macular degeneration. *Invest Ophthalmol Vis Sci.* 2014;55(9):6046–6054.
- **16.** Lee CS, Woo SJ, Kim YK, et al. Clinical and spectral-domain optical coherence tomography findings in patients with focal choroidal excavation. *Ophthalmology*. 2014;121(5):1029–1035.
- Xu H, Zeng F, Shi D, Sun X, Chen X, Bai Y. Focal choroidal excavation complicated by choroidal neovascularization. *Ophthalmology*. 2014;121(1):246–250.