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Spontaneous Resolution of Subretinal Detachment in Choroidal Osteoma Unresponsive to Antivascular Endothelial Growth Factor Agent and Multimodal Imaging: A Case Report

Dear Editor,

Choroidal osteoma (CO) is a rare benign ossifying choroidal tumor that typically affects young female patients [1]. While there is no standard treatment for CO, patients with complications, such as choroidal neovascularization (CNV) and serous retinal detachment (SRD), require treatment [2]. Several reports have discussed the role of antivascular endothelial growth factor (anti-VEGF) agents in the treatment of SRD with or without CNV, associated with CO [2,3]. We describe the spontaneous resolution of SRD in a patient with CO, that was unresponsive to intravitreal anti-VEGF treatment. The patient provided informed consent for publication of the research details and clinical images.

A healthy 9-year-old female patient presented with blurred vision in the left eye. The best-corrected visual acuity (BCVA) was 20 / 25 in the left eye. Funduscopic examination of the left eye revealed a juxtafoveal yellow-orange subretinal lesion with distinct borders (Fig. 1A). Fundus fluorescein angiography (FFA) revealed an early patchy hyperfluorescent choroidal filling pattern, dye staining in the area of retinal pigment epithelium (RPE) decompensation overlying the CO, pooling hyperfluorescence in the late phase, and no evidence of CNV (Fig. 1B, 1C). *En face* optical coherence tomography (OCT) showed the tumor border (Fig 1D, 1E). OCT angiography demonstrated the paucivascular tumor at the level of choroid, and absence of CNV (Fig. 1F). OCT showed SRD, a disrupted photoreceptor layer, and a hyperreflective mass posterior to the RPE (Fig. 1G). Therefore, the patient was diagnosed with CO with subretinal fluid in the absence of CNV.

To resolve the SRD, an intravitreal injection of bevacizumab (1.25 mg/0.05 mL, Avastin, Genentech) was administered to her left eye. One month after bevacizumab treatment, her BCVA decreased to 20 / 100, and OCT showed an increased SRD and intraretinal fluid (Fig. 1H). Because the lesion was unresponsive to bevacizumab, the patient was closely monitored without additional treatment. After 4 months, the SRD decreased spontaneously, leading to an improved BCVA of 20 / 50. After 1 year, complete resolution of SRD and intraretinal fluid was confirmed on OCT (Fig. 1I). However, disruption of the photoreceptor layer was detected, and the final BCVA remained unchanged at 20 / 50.

SRD, associated with CO, frequently occurs in the absence of CNV. It possibly resulted from multiple pinpoint RPE leakages over the osteoma noted on FFA [4]. Alternatively, the gradual atrophy of the RPE and Bruch's membrane likely decreased the capacity of the RPE to remove subretinal fluid from the disrupted outer blood-retinal barrier [3].

While it has been established to treat SRD with CNV, the significance of anti-VEGF in treating SRD without CNV remains controversial. Song et al. [3] reported that 83% of patients with CO and SRD did not present with signs of CNV. Following bevacizumab treatment, all patients showed a substantial decrease in SRD based on OCT, and 80% of the patients experienced improved visual acuity.

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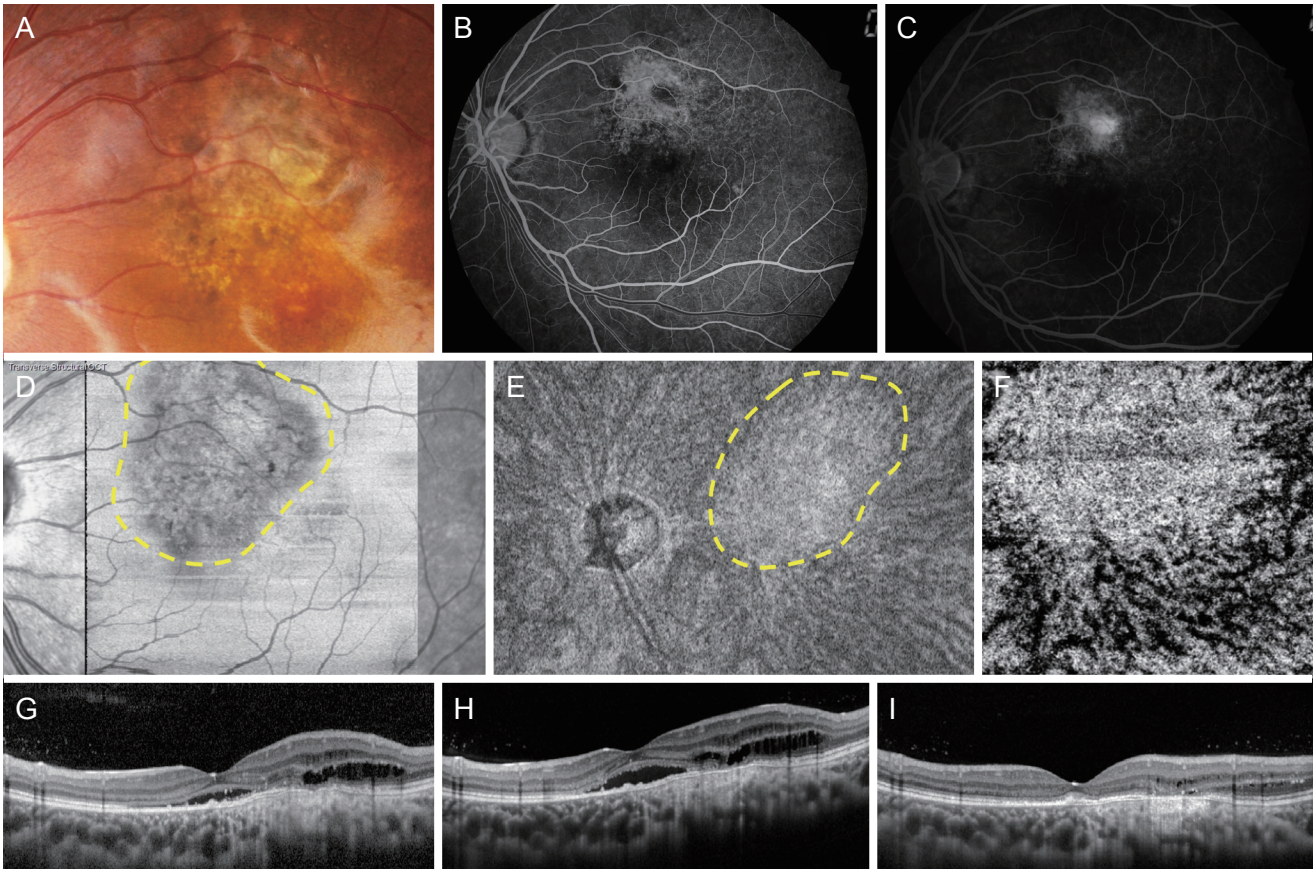

Fig. 1. Multimodal imaging of the choroidal osteoma. (A) Fundus photography showing well-defined yellow-orange subretinal lesion. (B) Fundus fluorescein angiography showing the area of early patchy hyperfluorescence. (C) Fundus fluorescein angiography at late phase showing dye staining in the area of retinal pigment epithelium decompensation overlying the choroidal osteoma and pooling hyperfluorescence. (D-F) *En face* optical coherence tomography (OCT) and OCT angiography showing the border (yellow dash lines) and the paucivascular content of the tumor. (G-I) OCT showing hyperreflective lesion casting a long shadow. (G) Initial OCT showing subretinal detachment, disrupted photoreceptor layer, and a hyperreflective band posterior to the retinal pigment epithelium. (H) OCT 1 month after the injection showing increased subretinal fluid and intraretinal fluid. (I) OCT 1 year after the injection showed complete resolution of subretinal detachment and intraretinal fluid.

To the best of our knowledge, this is the first report to investigate CO by applying *en face* OCT. *En face* OCT showed the border of CO with a great distinctness and aided noninvasive diagnosis. In addition, the present case was unique, compared to previously reported cases, because of the spontaneous resolution of SRD, which was unresponsive to intravitreal anti-VEGF treatment. Song et al. [3] reported that four out of five patients achieved complete resolution of SRD in response to anti-VEGF treatment, while one achieved only partial resolution of SRD after five months of treatment. Seong et al. [5] has shown that SRD without CNV responded worse to anti-VEGF treatment than did SRF with CNV. However, anti-VEGF was effective in about half of cases with SRF without CNV for the reason not clear at the moment [5]. In the present case, the SRD did not respond to anti-VEGF therapy. Interestingly, the SRD spontaneously resolved throughout the year while under observation without additional treatment. The underlying mechanism behind the spontaneous resolution of SRD in this case is unclear. Vascular leakage and focal RPE leakage on the surface of the tumor likely led to the unresponsiveness of SRD to anti-VEGF therapy. Spontaneous regression of the SRD may have begun with the recovery of the blood-retina barrier and RPE. In a young patient with CO-associated SRD without CNV, that was unresponsive to anti-VEGF agents, close observation might be a viable treatment option.

Conflicts of Interest: Se Joon Woo is a member of the Editorial Board of the *Korean Journal of Ophthalmology* since 2020. However, he was not involved in the peer reviewer selection, evaluation, or decision process of this article. No other potential conflicts of interest relevant to this article were reported.

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