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CASE REPORT

Longitudinal Follow-up after successful photodynamic therapy in two cases with unilateral choroidal hemangioma and serous macular detachment

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Abstract

Circumscribed choroidal hemangioma is a rare benign vascular choroidal tumor that may cause visual loss in regard to its location and/or be associated with either intraretinal or subretinal fluid leakage. We described the long-term good clinical outcome in two patients who underwent a single session of verteporfin photodynamic therapy (PDT) for the treatment of unilateral choroidal hemangioma and associated serous macular detachment with a follow-up duration of 13 and 14 years. PDT is a safe and effective therapy for the treatment of choroidal hemangioma and restores visual function in many cases without causing any apparent ocular or systemic side effects in the long run.

Keywords: Fluorescein angiography; choroidal hemangioma; optical coherence tomography; photodynamic therapy.

Choroidal hemangioma (CH) is a rare, benign vascular tumor that can either be circumscribed CH (CCH) or diffuse CH.^[1] CH may develop in early adulthood or be even present at birth.^[2] Circumscribed type is characterized as an orange-red, well-circumscribed choroidal mass, usually unilateral, located mostly at the posterior pole and without any associated systemic diseases.^[1-3] These vascular masses originate most likely from hemodynamic turbulences such as persistent arteriovenous shunts and are non-proliferative lesions with little to no tendency to enlarge, in contrast to other choroidal tumors.^[4] However, when an enlargement occurs, venous congestion is the primary cause.^[5]

The patients can be asymptomatic or experience mild-to-severe visual complaints including metamorphopsia,

photopsia, and/or visual loss.^[6] The tumor is usually diagnosed either by coincidence or by the help of visual symptoms.^[2] Visual disturbances may be related to tumor localization, presence of subretinal fluid (SRF), cystoid macular edema (CME), exudative retinal detachment, and very rarely choroidal neovascularization. The main treatment indications include SRF affecting the macula, the presence of CME, and choroidal neovascular membrane formation.^[2] Thermal laser photocoagulation, transpupillary thermotherapy (TTT), radiation therapy, photodynamic therapy (PDT), and anti-vascular endothelial growth factor injections are among the treatment modalities.^[7]

In this study, the long-term results of the two patients who received a single successful session of PDT were presented.



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Case 1

A 59-year-old healthy health-care worker was referred to us with the diagnosis of left acute central serous chorioretinopathy in 2009. On examination, his best-corrected visual acuity was 10/10 in OD and 5/10 in OS. Slit-lamp examination was unremarkable OU and intraocular pressure was within normal limits bilaterally. While the right fundus was normal, there was an orange-red choroidal mass with a size of 1.5 disk diameter in the left eye. On fundus autofluorescence imaging (Fig. 1a), the mass looked mostly hypoautofluorescent with ill-defined margins, and fluorescein angiogram (Fig. 1b) displayed the impaired retinal pigmented epithelium (RPE)-related patchy hyperfluorescence. Optical coherence tomography (OCT) depicted the presence of marked SRF (Fig. 1c). The patient was diagnosed with left CCH and treated with a single session of a standard dose of PDT. Intravenous verteporfin (6 mg/m²)

(Visudyne, Novartis Ophthalmicus, Hettlingen, Switzerland) was administered over a 10-min period. Five minutes after the completion of infusion, the laser was delivered at an intensity of 600 mw/cm² using a Reichel-Mainster 2 lens (Ocular Instruments, Bellevue, WA, USA) with a 3000-micron spot size. The treatment duration was 166 s. Fourteen years later, his visual acuity was 8/10 in the affected left eye. Although the previous SRF tract could be inferred from the pattern of RPE changes, there was no sign of SRF indicating the persistence of the residual or recurrent fluid (Fig. 2).

Case 2

A 46-year-old healthy man was presented with a left visual loss of 3-month duration in January 2007. On examination, his best-corrected visual acuity was 10/10 in the right eye and 4/10 in the left eye. Slit-lamp examination was unremarkable OU and intraocular pressure were within

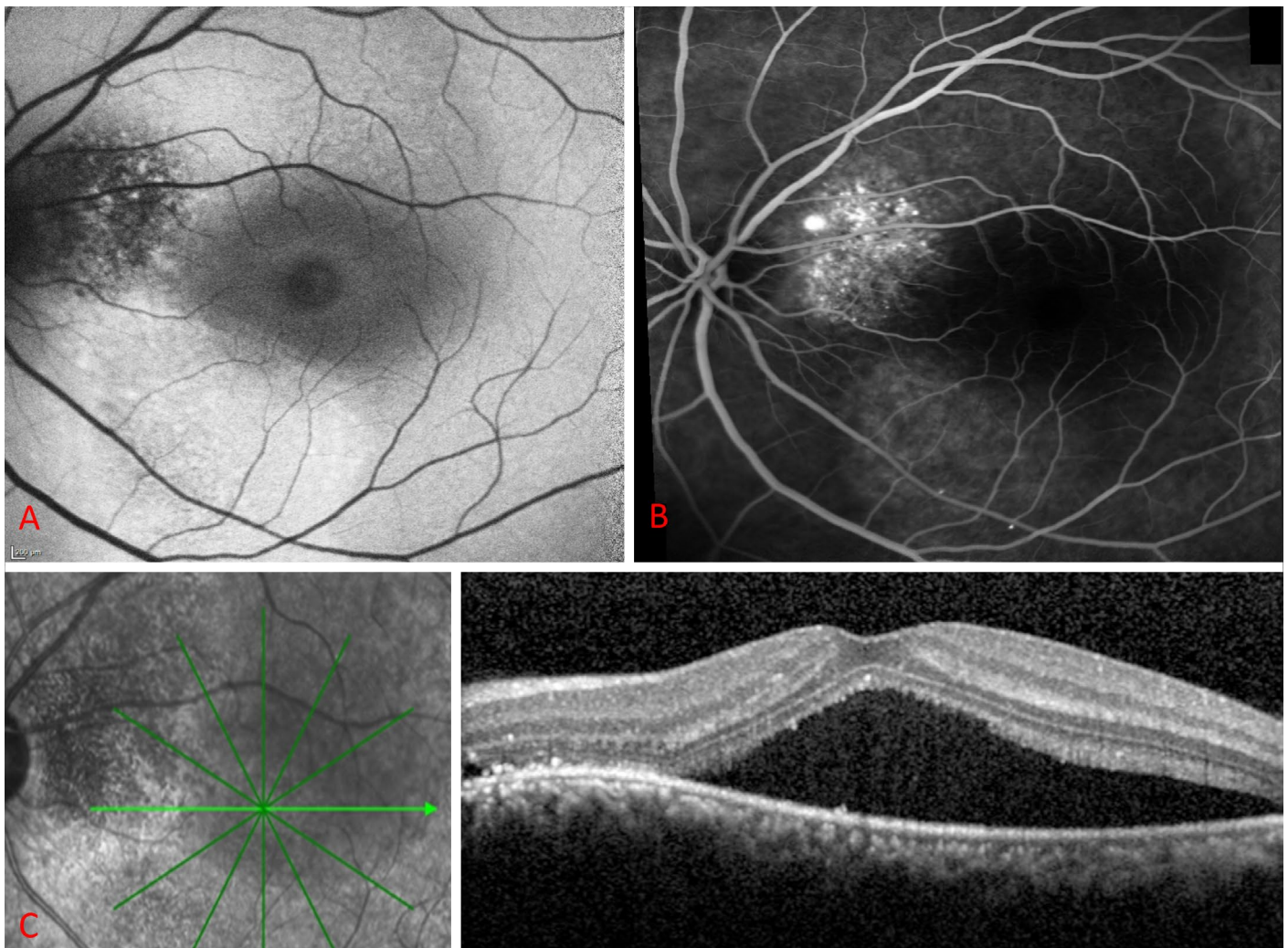


Fig. 1. Left eye (2009), ill-defined, peripapillary mostly hypoautofluorescent area showing the mass (a), Late phase of fluorescein angiogram depicting the hyperfluorescent lesion (b). Optical coherence tomographic section demonstrating the subfoveal extensive neurosensory detachment (c).

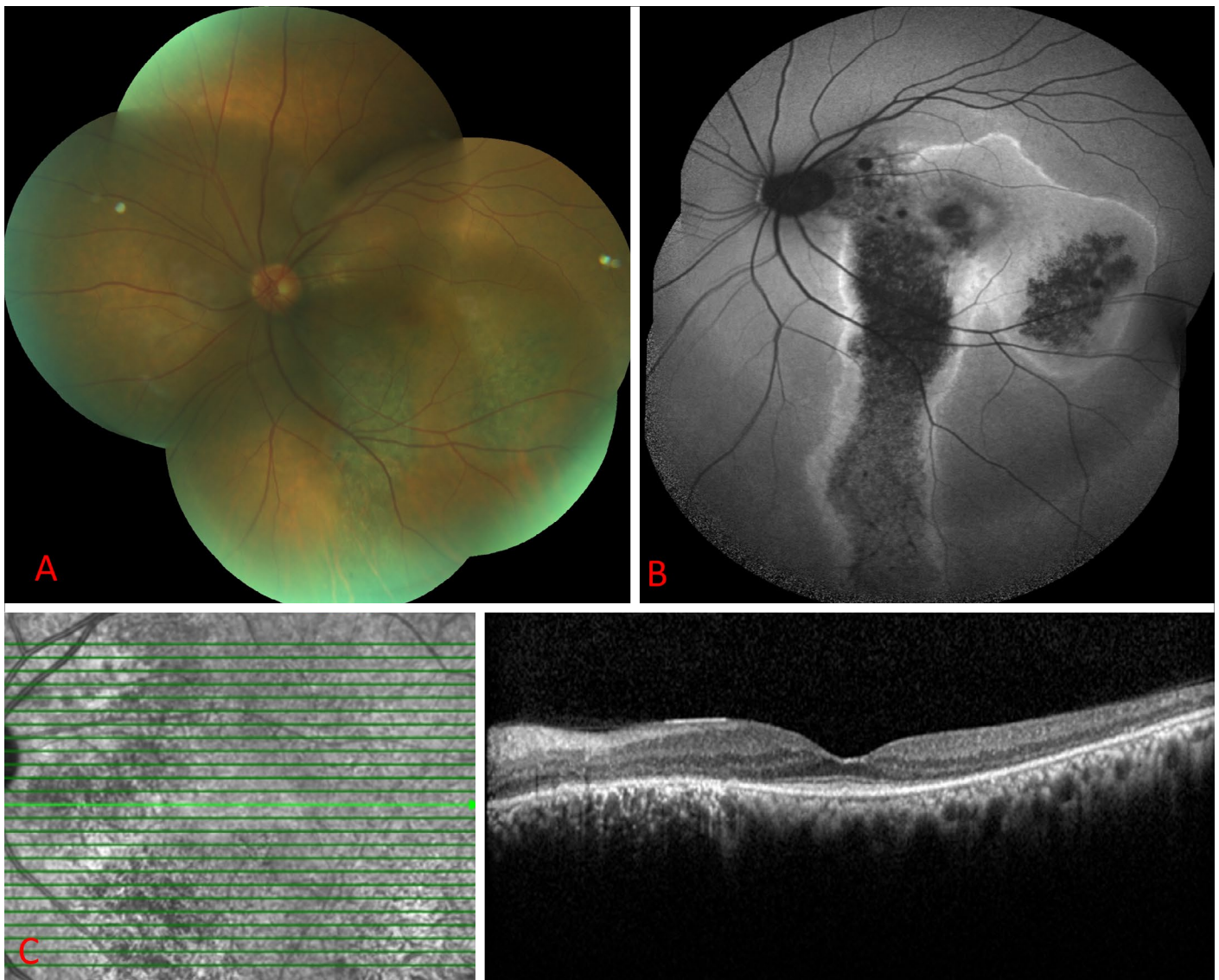


Fig. 2. Left eye (2023), color fundus image (a), and fundus autofluorescent images (b) depicting the mottled RPE changes. No sign of fluid was evident on optical coherence tomography (c).

normal limits bilaterally. On fundus examination, while the right fundus was normal, there was a prominent reddish choroidal mass of 4 disc diameter size and adjacent serous-looking retinal detachment in the left eye (Fig. 3a). Fluorescein angiographically, the mass looked hyperfluorescent with hypofluorescent margins (Fig. 3b). OCT revealed the presence of extensive serous detachment neighboring the mass (Fig. 3c). The patient was treated with a single session of PDT at an intensity of 100 J/cm² for 166 s 5 min after the 10-min verteporfin infusion at the dose of 6 mg/m² with the diagnosis of left CCH. Two overlapping spots were administered to cover the whole tumor by exceeding its edges by approximately 1000 μ . The treatment duration was 166 s. Thirteen years later, left vision was 10/10. On fundus examination, regressed CH

with residual RPE changes were noted (Fig. 4a). Besides the presence of choroidal mass, there was no evidence of active fluid tomographically (Figures 4b and c).

Discussion

Although CCH is a benign tumor, affected patients may experience visual deterioration. The aim of the treatment is to protect the integrity of the macula by eliminating the subfoveal fluid and CME if present and not to destroy the tumor such as the case in choroidal melanoma. TTT, laser photocoagulation, proton beam radiation therapy, external beam radiation therapy, cryotherapy, or plaque brachytherapy can be utilized to deal with CCHs.^[8] When PDT was first administered in 2000, it was successful in treating both acute and chronic exudative retinal

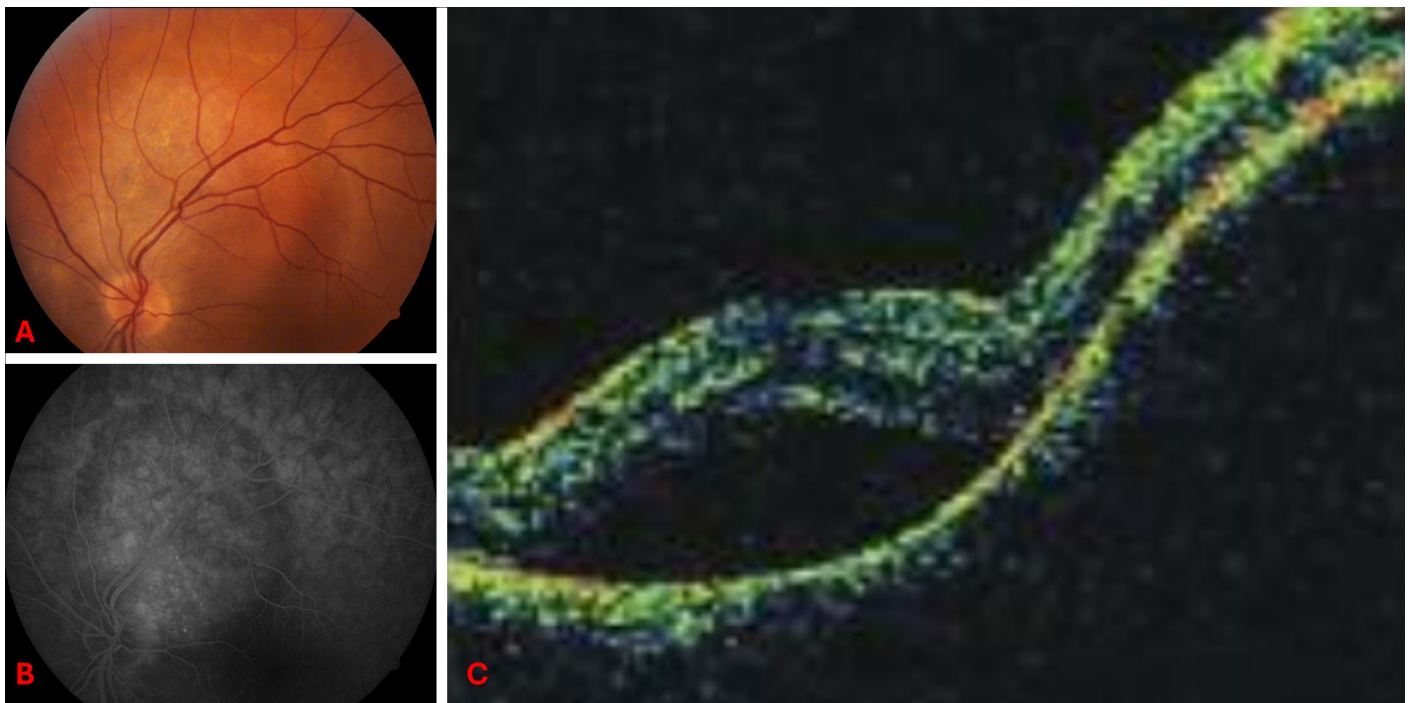


Fig. 3. Left eye (2007), A prominent reddish choroidal mass of 4 disc diameter size and adjacent serous-looking retinal detachment in the left eye (a). Late phase of fluorescein angiogram delineating the hyperfluorescent mass with relatively ill-defined margins (b). Time domain optical coherence tomographic section demonstrating the presence of extensive serous detachment (c).

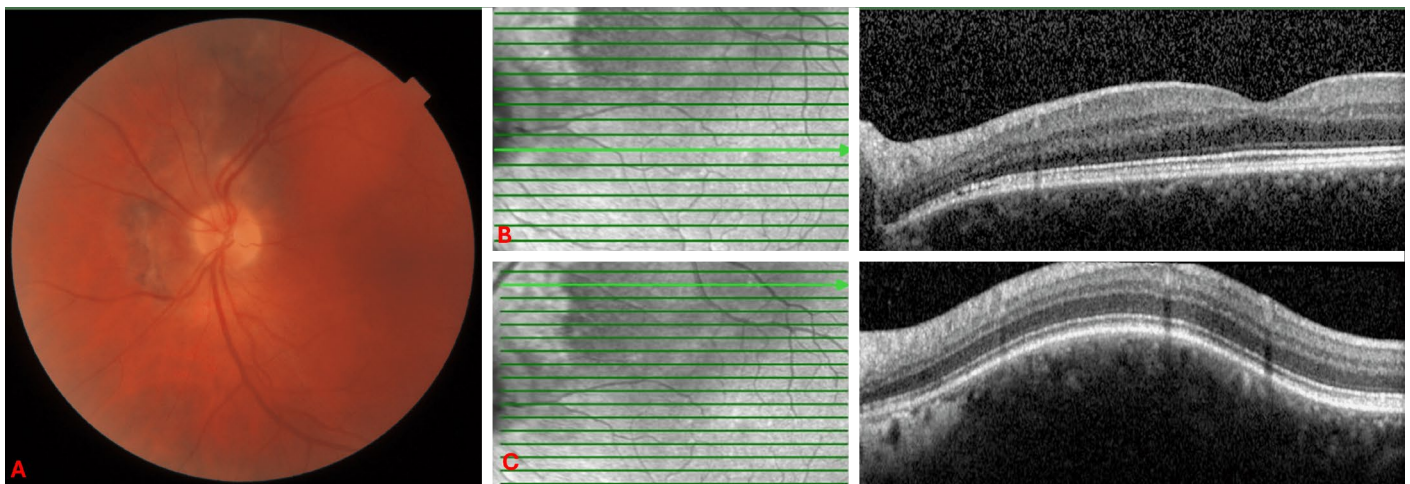


Fig. 4. Left eye (2020), color picture revealing the slightly regressed choroidal hemangioma with some residual RPE changes (a). OCT section depicting the normal foveal contour (b). OCT section over the mass without any evidence of fluid (c).

detachments in association with CCHs by preventing field loss, slightly flattening the tumor, reducing the SRF, and thereby improving the visual acuity.^[9-11] Studies consisting of 10 or more CCH patients treated with PDT are summarized in Table 1.^[12-21] PDT affects the neuroretinal structures selectively.^[22] Nowadays, PDT seems to be the most preferred option in eyes with CCH and associated serous detachment and facilitates the selective treatment of pathologic vessels and thereby results in the resorption of the serous fluid with relatively less collateral retinal

damage. Thus, visual improvement is usually achieved with a low recurrence and complication rate.^[22] PDT can be administered as a single session or multiple sessions,^[23] double dose,^[24,25] standard dose,^[12] half dose,^[26] or half-fluence dose.^[27] Although the exact mechanism of the PDT in CCH is still not understood, it is hypothesized that choriocapillaris sclerosis ensues following the treatment due to occlusion of the tumor's arteries, thereby reducing the leakage.^[6] PDT is more particularly effective in eyes where the hemangioma lies underneath the fovea when

Table 1. Manuscripts reporting 10 or more circumscribed choroidal hemangioma patients treated with photodynamic therapy

Author "Year"	Number of patients	Mean Age	Treatment Dose	Session (Min-Max)	Anatomic Success	Visual Success (Stable or improved)	Recurrence	Complication
Jurklic et al. ^[12] "2003"	19	51	Standard protocol	1–5	18 (94.8%)	18 (94.8%) 14 eyes-improved 4 eyes-stable	N/A	No
Zhang et al. ^[13] "2010"	25	17	Standard protocol	1–2	25 (100%)	21 (84%) 14 eyes-improved 7 stable	N/A	No
Michels et al. ^[14] "2005"	15	N/A	Standard protocol	1–4	15 (100%)	15 (100%) 13 eyes-improved 2 eyes-stable	No	Focal loss of choroid and RPE in all cases
Verbraak et al. ^[15] "2003"	13	50	Standard protocol	1–2	13 (100%)	13 (100%) 11 eyes-improved 2 eyes-stable	No	N/A
Lee et al. ^[16] "2019"	17 (7 standard dose verteporfin group, 10 double dose group)	52 for the double dose group and 51 for the standard dose group	Standard dose or double dose protocol	N/A	Complete SRF resorption 80% (double dose group) 57% (standard dose group)	Visual improvement was not statistically significant between the two groups	2 patients only in the standard dose group	No
Schmidt-Erfurth et al. ^[17] "2002"	15	53	Standard Protocol	1–4	15 (100%)	15 (100%) 13 eyes-improved 2 eyes-stable	No	Focal choroidal atrophy in 7 cases
Blasi et al. ^[18] "2010"	25	54	Standard Protocol	1–2	25 (100%)	22 (88%) 19 eyes-improved 3 eyes-stable	No	No
Porrini et al. ^[19] "2003"	10	N/A (Range: 38–64)	Standard Protocol	1–3	10 (100%)	10 (100%)	No	Intra-retinal edema in 2 cases
Jamison et al. ^[20] "2018"	17	56	Standard protocol	1–2	17 (100%)	17 (100%)	N/A	No
Singh et al. ^[21] "2004"	10	53	Standard protocol	1–2	10 (100%)	8 (80%) 4 eyes-improved 4 eyes-stable	N/A	Choroidal atrophy in 2 cases

N/A: Not applicable; RPE: Retinal Pigment Epithelium; SRF: Subretinal Fluid.

compared to other treatment techniques as PDT can be repeatable and has fewer side-effects than the other treatment modalities.^[28]

Conclusion

Verteporfin PDT is a safe and effective therapy for the treatment of CCH and restores visual function without causing any apparent ocular or systemic side effects in many eyes.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report.

Peer-review: Externally peer-reviewed.

Authorship Contributions: Concept: D.O.; Design: A.O.S.; Supervision: A.O.S.; Resource: D.O., A.O.S.; Materials: D.O.; Data Collection and/or Processing: D.O.; Analysis and/or Interpretation: D.O.; Literature Search: D.O.; Writing: D.O., A.O.S.; Critical Reviews: D.O., A.O.S.

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