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**Case Report**

# A Case of Bullous Central Serous Chorioretinopathy Treated with Surgical Removal of Submacular Fibrin and Subsequent Photodynamic Therapy under Silicone Oil

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**Keywords**

Central serous chorioretinopathy · Bullous retinal detachment · Photodynamic therapy · Subretinal fibrosis · Pars plana vitrectomy and Silicone oil tamponade

**Abstract**

Bullous retinal detachment is a rare complication in the chronic phase of central serous chorioretinopathy (CSC). Only a small subset of eyes with chronic CSC develops into the bullous variant of CSC (bCSC). In patients with bCSC, the elevated concentration of fibrin in the subretinal space leads to persistent retinal detachment and eventually, severe vision loss. We experienced a case of unilateral bCSC with a massive accumulation of subretinal fibrin. Multiple leakage points and dilated choroidal veins were also observed. The patient underwent surgical removal of subretinal fibrin and silicone oil injection followed by photodynamic therapy (PDT). After this treatment, the retina was successfully reattached, and the affected eye was free from recurrent exudative changes for more than 18 months. Massive subretinal fibrin could be surgically removed to prevent the formation of subretinal fibrosis and retinal fold, and PDT under silicone oil can control the underlying exudative changes in bCSC.



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## Introduction

Chronic central serous chorioretinopathy (CSC) is a subtype of CSC that is characterized by extensive retinal pigment epithelium (RPE) dysfunction and serous retinal detachments (RDs) persisting for more than 6 months. Bullous RD is a rare manifestation of chronic CSC [1, 2]. It remains unclear why only a small subset of eyes with chronic CSC develop into the bullous variant of CSC (bCSC), whereas most cases manifest RD of much lesser severity. Patients affected by bCSC usually have pigment epithelial detachment (PED) hidden beneath a cloudy subretinal fluid (SRF). This cloudy consistency is due to the elevated concentration of fibrin, which may eventually lead to subretinal fibrosis and a poor visual outcome [3]. Here, we report a case of bCSC that required surgical removal of massive subretinal fibrin and was successfully treated with photodynamic therapy (PDT).

## Case

A 51-year-old woman was diagnosed with CSC and received laser photocoagulation treatment in the right eye. After treatment, her best-corrected visual acuity (BCVA) in the right eye was better than 1.0 on the decimal scale (1.0 = 20/20). She noticed visual impairment in the right eye 5 years after the initial treatment and visited an ophthalmology clinic. BCVA in the right eye was 0.3 on the decimal scale, and she received sub-tenon injection of triamcinolone acetonide. However, the vision in her right eye did not fully recover, and she was referred to a hospital the next month. The decimal BCVA in the right eye was 0.6 at the hospital. A leakage under the submacular fibrin was observed on fluorescein angiography (FA). The anti-vascular endothelial growth factor agent aflibercept was intravitreally injected into the right eye once since age-related macular degeneration was suspected. Four months later, she was referred to our hospital. At the initial examination at our hospital, BCVA in the right eye was counting fingers. BCVA in the left eye was 1.2 on the decimal scale. Fundus examination revealed a bullous RD predominantly in the inferior two quadrants. There was dense white material occupying the subretinal space around the macula (Fig. 1a, 2a). FA and indocyanine green angiography revealed multiple leakage points and underlying dilated choroidal vessels around the macular area in the right eye (Fig. 1b, c). No apparent abnormality except a mild RPE abnormality in the temporal retina was detected in the left eye (Fig. 1d–f). Optical coherence tomography (OCT) detected a thick fibrin membrane, focal PED, and underlying dilated choroidal vessels (Fig. 2b–d). No RD or PED was observed in the left eye. The axial lengths in the right and left eye were 20.8 and 21.5 mm, respectively. She had no history of systemic disease or medication. Laboratory blood tests showed only a mild liver dysfunction, possibly due to a fatty liver. HLA blood tests confirmed negativity for DR4.

*Treatment*

To reattach the retina immediately, we performed pars plana vitrectomy using a 25-gauge vitrectomy system (CONSTELLATION Vision System; Alcon, Fort Worth, TX, USA) combined with phacoemulsification and intraocular lens implantation. In the intraoperative view, the retina was totally detached without any retinal break. Anterior and core vitrectomy was done after phacoemulsification and IOL implantation. Perfluoro-n-octane (Perfluoron, Alcon) was placed over the posterior pole during peripheral vitrectomy. After removing the perfluoro-n-octane, a retinotomy was created using microscissors (Disposable Microscissors, Vertical, 25 gauge; DORC, Zuidland, The Netherlands) near the inferior retinal arcade to gain access to the submacular fibrin (See online suppl. Video; for all online suppl. material, see www.karger.com/doi/10.1159/000524515). The whitish subretinal fibrin was smoothly extracted as a

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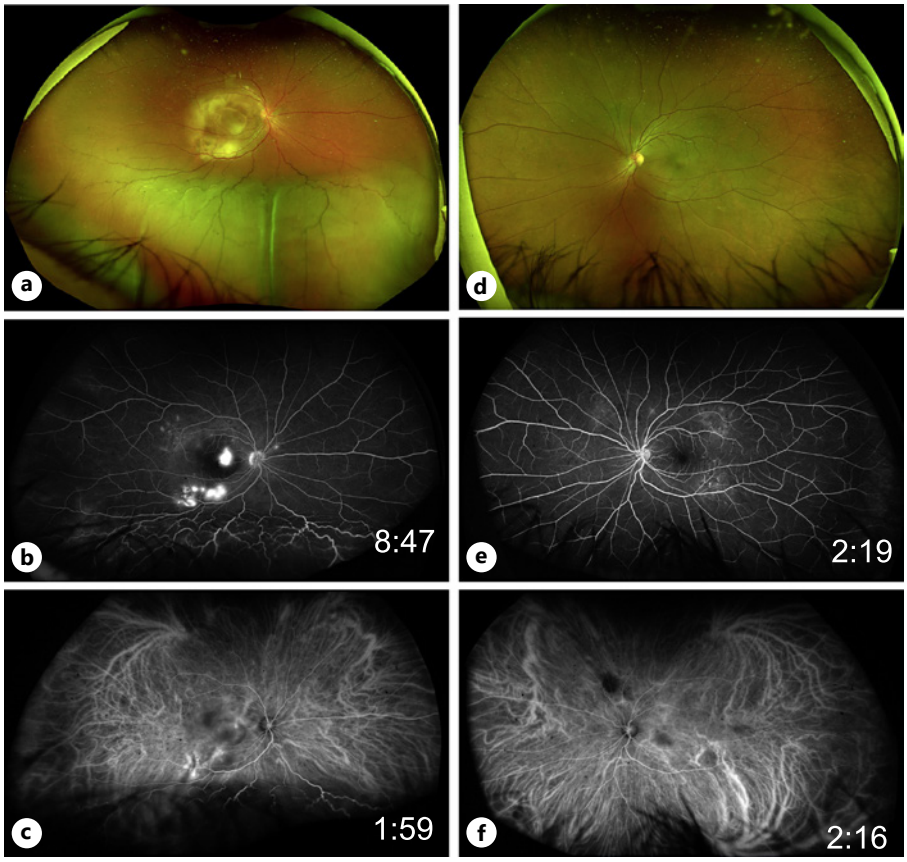


**Fig. 1.** Ultrawide-field fundus and FA/ICGA images from both eyes at the first visit. Bullous RD in the inferior quadrants and massive accumulation of subretinal fibrin in the posterior pole was observed in the right eye (**a**). FA detected vigorous leakage from several points, especially in the macular area and near the inferior retinal arcade (**b**). ICGA revealed dilated choroidal vessels crossing the posterior pole in the right eye (**c**). No abnormality in ultrawide-field fundus photography (**d**), mild window defects in the temporal retina with FA (**e**), and the dilatation of choroidal veins in the inferior temporal quadrant with ICGA (**f**) were observed in the left eye. ICGA, indocyanine green angiography.

single mass through the retinotomy using microforceps (V-ARTIST disposable micro forceps, asymmetrical tip; HOYA Corporation, Tokyo, Japan). Fluid-air exchange was performed using a flute needle to drain SRF through the intentional retinotomy, and silicone oil (SILIKON 1000; Alcon) was injected.

After the surgery, the submacular fibrin had disappeared, and the retina was almost attached under silicone oil tamponade (Fig. 3a). Laser photocoagulation (wavelength 647 nm, power 120 mW, duration 200 ms, spot size 100 μm, 78 shots) was postoperatively performed around the leakage points in the vicinity of the inferior arcade. However, there were remaining exudative changes including fluorescein leakage and shallow SRF as revealed by FA/indocyanine green angiography (Fig. 3b–d) and OCT (Fig. 4a, b). To suppress the exudative activity, we performed a full-dose PDT (greatest linear dimension; 6,562 μm; see Fig. 4c) at 1.5 months after the primary surgery. PDT successfully reduced those exudative changes (Fig. 5a, b). After the removal of silicone oil 2.5 months after PDT, the right eye was free from any SRF or PED. There was no recurrence of CSC through the final visit 14 months after PDT (Fig. 5c, d). Decimal BCVA in the right eye had improved up to 0.1 at 8 months after PDT and was 0.06 at the final visit. The time course of the treatment is summarized in the figure (Fig. 5e).

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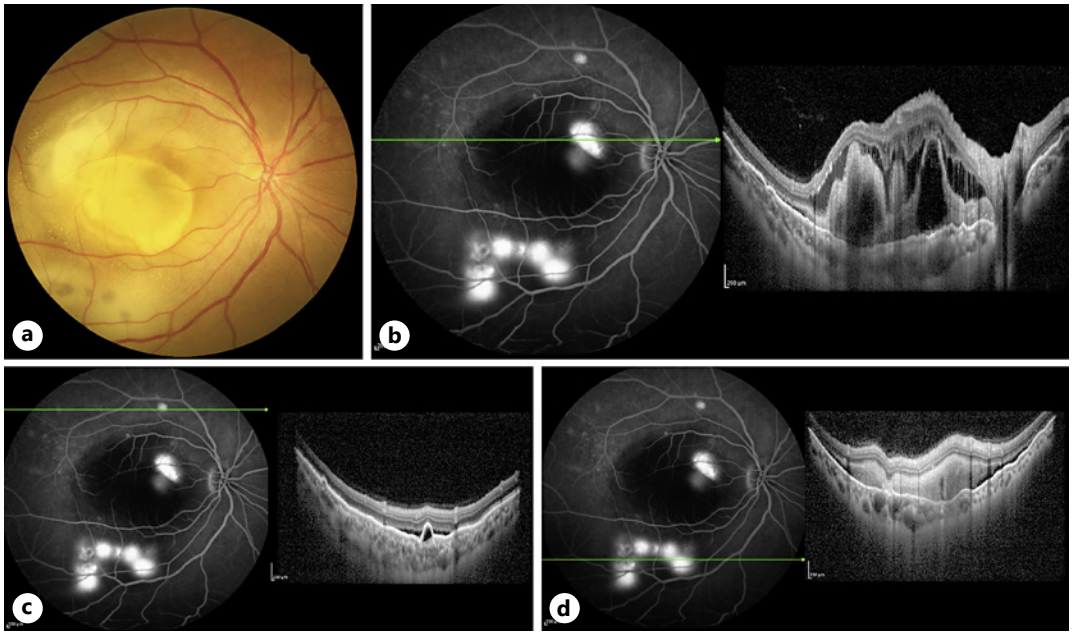


**Fig. 2.** A fundus photograph (**a**) and enhanced depth imaging OCT (**b–d**) of the massive submacular fibrin in the right eye at the first visit. A small PED was detected adjacent to the leakage point of fluorescein near the superior arcade (**c**). Thick subretinal fibrin accumulation was observed around the macular area (**b**, **d**).

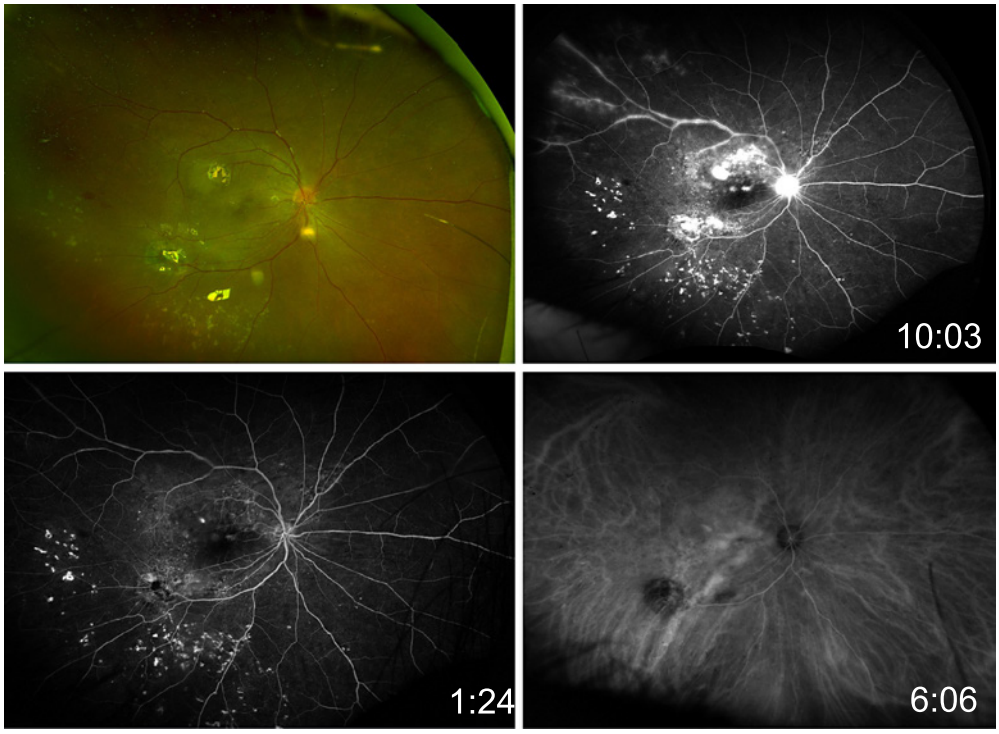


**Fig. 3.** An ultrawide-field fundus photograph from the right eye after silicone oil injection (**a**). FA showed a persistent fluorescein leakage from the macular area and near the inferior arcade (**b**, **c**), and ICGA indicated the dilated choroidal vessels with hyperpermeability (**d**). ICGA, indocyanine green angiography.

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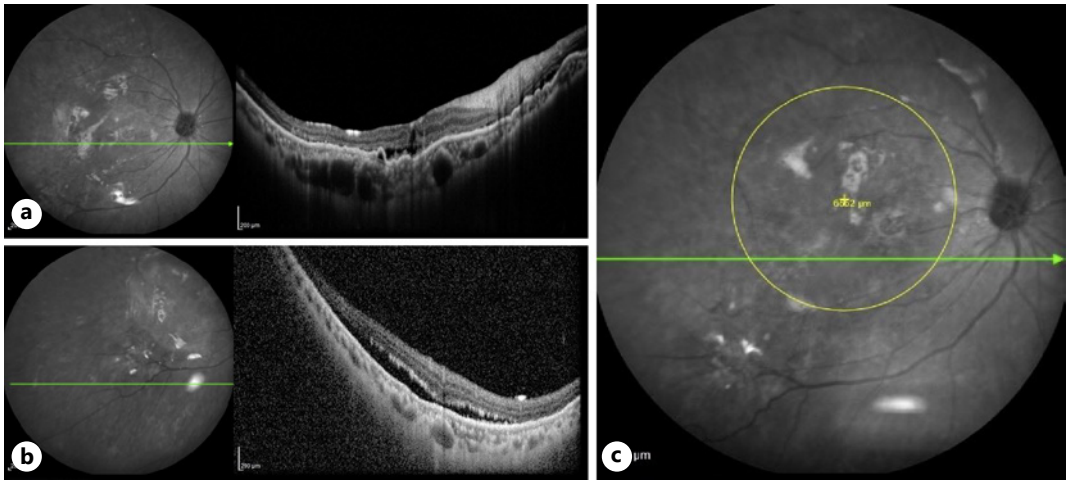

**Fig. 4.** Postoperative enhanced depth imaging OCT in the right eye. Dilated choroidal vessels were observed in the macular area (**a**), and shallow SRF was detected near the inferior arcade (**b**). The greatest linear dimension of performed PDT (6, 562 μm) is illustrated (c)

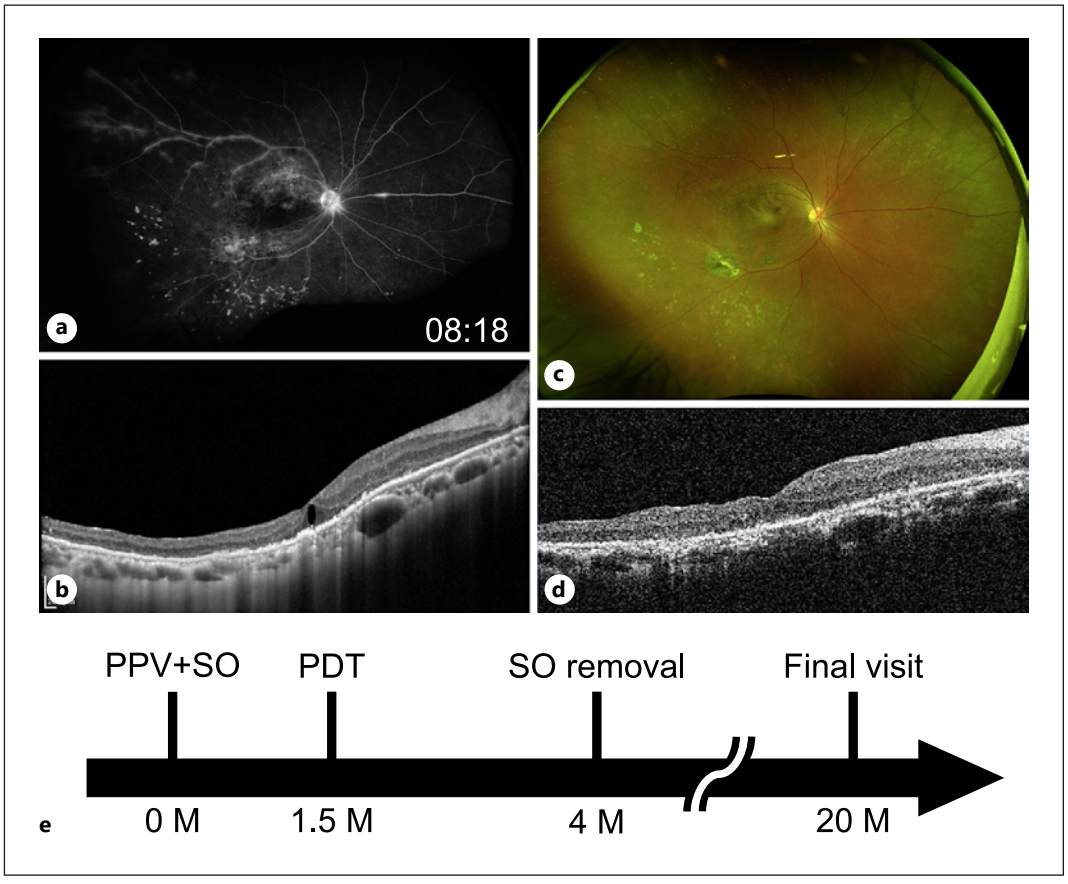


**Fig. 5.** FA and OCT images after PDT under silicone oil in the right eye. FA showed a significantly reduced fluorescein leakage around the macular area (**a**). Horizontal enhanced depth imaging OCT illustrated a diminished SRF/PED and a decreased dilatation of choroidal vessels after PDT (**b**). An ultrawide-field fundus image (**c**) and spectral-domain OCT of a horizontal section crossing the macula (**d**) at the final visit. Time course of the treatment is illustrated in (**e**). PPV, pars plana vitrectomy; SO, silicone oil.

## Discussion

The bCSC is occasionally complicated by a submacular accumulation of fibrin, which can eventually lead to subretinal fibrosis and a fixed retinal fold. Balaratnasingam et al. [4] reported that retinal folds and subretinal fibrin were identified in a greater proportion of eyes in the bullous CSC group than in the nonbullous CSC group. Indeed, subretinal bands were identified in 38.5% of eyes in the bullous CSC group at the final visit in their case series.

Several medical and surgical treatment options for bCSC have been reported, although the therapeutic algorithm is still a matter of debate due to the very low frequency of the disease. Otsuka et al. [5] enrolled 25 patients affected by bCSC with a mean follow-up time of 10.6 years, of whom 17 patients underwent laser photocoagulation for focal leaks. However, the final BCVAs were not significantly different between the treated and untreated group. PDT has been well established as a first-line treatment for chronic CSC [6–8]; however, few bCSC cases treated with PDT have been reported. Wykoff et al. [9] reported a case of bCSC presenting an RPE tear and the accumulation of a whitish subretinal material temporal to the fovea, which was treated by reduced-fluence PDT. Examination 7 months later revealed resolution of bullous RD and subretinal material with the development of a pigmented scar in the area of the RPE tear. Ng et al. [10] reported a case of bCSC that presented inferior RD with diffuse fluorescein leakage at the macula, which was treated with half-dose PDT. BCVA in that case improved from 20/70 to 20/25 within a month of treatment, and there was complete resolution of SRF and bullous RD 3 months after PDT.

Surgical approaches have also been attempted for bCSC treatment. Venkatesh et al. [11] reported a case of bCSC treated by two partial-thickness scleral resections in the inferior quadrants. BCVA improved from 20/630 to 20/200, and SRF resolved 4 months after the procedure. No recurrence was observed in the following 2 years [11]. John et al. [12] performed an encircling scleral buckle, phacoemulsification, and 20-gauge pars plana vitrectomy in a patient affected by bCSC with a BCVA of 20/80. The SRF was drained through a superior retinotomy, and silicone oil tamponade was applied. One year later, the silicone oil was removed, OCT showed no SRF, and BCVA was 20/200.

These cases treated with medical/surgical options were not accompanied by subretinal fibrin-like material except the one reported by Wykoff et al. [9] Even in the case Wykoff et al. [9] reported, the amount of subretinal fibrin was not as extensive as in our case. Note that the subretinal white material in their case was associated with an RPE tear, which might be involved in the resolution of subretinal fibrin, similar to cases of an RPE tear associated with bullous PED in which SRF can be resorbed without being treated [13]. In our bCSC case, the subretinal fibrin/fibrosis was so massive and extensive as to occupy the posterior pole; thus, we were afraid that a large subretinal fibrosis might remain beneath the macula as seen in the severe bCSC cases that Balaratnasingam et al. [4] reported. The existence of such thick subretinal fibrin/fibrosis may prolong RD by mechanically inhibiting photoreceptors attached to the RPE. Moreover, subretinal fibrin might cause direct damage to the retina by a similar mechanism as that observed in subretinal hemorrhage, in which a clot/fibrin can induce mechanical contraction to photoreceptors within 24 h [14]. Hence, we considered that surgical removal of submacular fibrin/fibrosis and silicone oil tamponade would be a reasonable option to allow immediate reattachment of the retina in this case.

In our bCSC case, PDT was performed in a silicone-oil injected eye, which enabled a safe follow-up until the disappearance of exudative changes was confirmed. There are limited numbers of case reports for the use of PDT under silicone oil. Soler et al. [15] reported a case of a myopia with macular neovascularization that was successfully treated with PDT in the presence of silicone oil. More recently, Kawakami et al. [16] reported a case of choroidal hemangioma with exudative RD treated by vitrectomy and silicone oil injection followed by

PDT. As a result, the circumscribed choroidal hemangioma disappeared. Thus, PDT under silicone oil seems to be a safe and effective treatment option in ocular diseases involving vascular exudation from the choroid.

## Conclusion

Our treatment for bCSC may have provided a chance for the retina to reattach, resulting in no recurrence of serous RD for over a year. Although the patient's retina might have already been damaged by preexisting submacular fibrin upon referral to our hospital, she did not require further treatment after the silicone oil removal. The effectiveness of PDT in chronic CSC has been well established, [6] although there are only two single-case reports of PDT for bCSC [9, 10]. Further investigations will be required to clarify better treatment options in bCSC. In this case report, we describe vitreoretinal surgery combined with PDT under silicone oil in severe bCSC with massive fibrin accumulation.

## Acknowledgment

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## Statement of Ethics

This study was reviewed and the need for approval was waived by the Institutional Review Board at Kyushu University Hospital. Written informed consent was obtained from the patient for the treatment (e.g., vitreous surgery and PDT) and for anonymized patient information and images to be published in this article.

## Conflict of Interest Statement

The authors have no conflicts of interest to declare.

## Funding Sources

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## Author Contributions

Shoji Notomi designed the study, drafted the manuscript, and uploaded the final version of manuscript. Satomi Shiose performed PDT and drafted the manuscript. Ri-ichiro and Kohno designed the treatment strategy. Sakurako Shimokawa performed imaging analysis. Keijiro Ishikawa critically reviewed and approved the manuscript. Kumiko Kano interpreted the data. Kenichiro Mori confirmed the patient's history. Iori Wada analyzed the imaging data. Shunji Nakatake collected the imaging data. Yosuke Fukuda reviewed and edited the manuscript. Muneo Yamaguchi reviewed the literature. Koh-Hei Sonoda approved the final version of the manuscript.

## Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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