CASE REPORT

Bilateral Bullous Central Serous Chorioretinopathy Successfully Treated with Vitrectomy and Spironolactone: A Case Report

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The authors report a case of a 46-year-old man with bilateral bullous central serous chorioretinopathy (bCSC). The patient experienced sequential vision loss in both eyes, occurring six months apart. He denied any comorbidities and steroid use. During active disease in the right eye (OD), the visual acuity (VA) was 20/100 in OD and 20/20 in the left eye (OS). The anterior segment appeared normal. Fundus examination and macular optical coherence tomography (OCT) revealed pigment epithelial detachment (PED) with subretinal fluid (SRF). The patient was initially diagnosed with macular neovascularization and received intravitreal bevacizumab injections. Two weeks after the third injection, VA declined to hand motion, and the fundus examination revealed the inferior bullous retinal detachment and subretinal fibrosis extending from PED. Ruling out rhegmatogenous retinal detachment, the patient underwent vitrectomy with gas tamponade. No retinal break was detected intraoperatively, and retinal attachment was successfully achieved. Six months later, the VA in OS declined from 20/20 to 20/60. Fundus examination showed inferior bullous detachment with ripped retinal pigment epithelium (RPE) and exudates. Fundus angiography revealed diffuse RPE leakages. The diagnosis was revised to bilateral bCSC. Due to a shortage of verteporfin and ongoing disease progression after observation, the patient was prescribed oral spironolactone, which led to SRF resolution. During the 6-month follow-up, VA was 20/25 (OD) and 20/20 (OS), with the disease remaining inactive. In conclusion, vitrectomy with internal drainage and spironolactone may serve as an effective alternative treatment for bCSC in the context of verteporfin shortage.

Keywords: Bullous central serous chorioretinopathy; Spironolactone; Vitrectomy

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Bullous central serous chorioretinopathy (bCSC) is an atypical variant of central serous chorioretinopathy (CSC). Its management remains controversial. Unlike typical CSC, bCSC often affects both eyes. Elimination of risk factors is the key management of all types of CSC. Interventions included laser photocoagulation, photodynamic therapy (PDT), mineralocorticoid receptor antagonists (MRAs), and surgery have been reported[1]. Verteporfin PDT is the promising treatment of choice for chronic CSC[2-4]. However, a shortage of verteporfin has adversely affected patient care. Previous meta-analyses assessing the efficacy of MRAs in chronic CSC have not demonstrated significant improvements in visual acuity or resolution of subretinal fluid (SRF)[3,5,6]. However, these studies did not specifically focus on bCSC. While vitrectomy is not typically recommended for CSC patients, few reports indicate its potential efficacy in managing SRF in bCSC[7]. This report discussed a case successfully treated by vitrectomy with internal drainage in the right eye and by oral spironolactone in the left eye.

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Case Report

A 46-year-old male patient was undergoing trabeculectomy for advanced primary angle-closure glaucoma in the right eye. The patient had no documented systemic underlying diseases. The patient also reported no significant medical history or substance used, with the exception of prescribed antiglaucoma eye drops. The baseline best corrected visual acuity (BCVA) was 20/40 in the right eye and 20/20 in the left eye. Two weeks post-surgery, the patient reported blurred vision in the right eye, with BCVA decreasing to 20/100. The intraocular pressure (IOP) decreased from 45 mmHg at baseline

to 12 mmHg without evidence of bleb leakage. The Fundus examination revealed a retinal fold, SRF, and pigment epithelial detachment (PED) at the macula. The optical coherence tomography (OCT) confirmed the presence of SRF, fibrovascular PED, and subfoveal choroidal thickening. Indocyanine green angiography (ICGA) exhibited early hypercyanescent lesions and retinal pigment epithelium (RPE) leakage (Figure 1). The patient was initially diagnosed with macular neovascularization and received intravitreal bevacizumab injections.

Two weeks following the third intravitreal injection, the BCVA further declined to hand motion. The IOP remained stable at 10 to 12 mmHg. Fundus examination revealed inferior bullous retinal detachment involving the macula, and subretinal fibrosis extending from PED. Rhegmatogenous retinal detachment (RRD), a potential complication following intravitreal injection, could not be excluded. Consequently, the patient underwent vitrectomy with internal SRF drainage and C3F8 tamponade. Intraoperatively, no retinal breaks were identified.

Six months after surgery, the BCVA in the right eye improved to 20/60. The IOP was 12 mmHg with functional blebs. The retina was successfully reattached. However, the patient reported the presence of central scotoma in the left eye, with BCVA decreasing from 20/20 to 20/60. Fundus examination revealed bullous retinal detachment involving the macula. Ripped RPE at the inferotemporal arcade was observed. Fundus fluorescein angiography (FFA) and ICGA demonstrated diffuse RPE leakage (Figure 2). Investigations for uveitis yielded negative results.

Following a comprehensive review, the diagnosis was revised to bilateral bCSC. The patient exhibited no systemic risk factors and was advised to pursue conservative management. However, the SRF and subretinal exudates continued to progress after two weeks of observation. Due to a shortage of verteporfin, the patient was prescribed spironolactone at a dosage of 50 mg/day for four weeks, followed by 25 mg/day for four weeks. Notably, two weeks after discontinuing spironolactone, the SRF had completely resolved, and simultaneous FFA and ICGA did not demonstrate any significant leakage (Figure 3).

Six months after the initiation of spironolactone, the BCVA in the left eye improved to 20/20. In the right eye, the BCVA achieved 20/25 after cataract surgery, which was thirteen months after vitrectomy with internal SRF drainage. The IOP remained stable, ranging from 10 to 12 mmHg. The anterior segment was normal, and no recurrence of disease activity was observed in either eye during the 2-year follow-up period.

Discussion

In the present case report, the authors presented the case of a middle-aged man who developed sequential bilateral bCSC. The clinical features observed in this case were consistent with the previously report of bCSC including inferior bullous retinal detachment, macular PED, subretinal exudates, and ripped RPE[1]. OCT demonstrated PED with pachychoroid features. FFA and ICGA demonstrated RPE leakage with choroidal vascular hyperpermeability. These findings were indicative of bCSC.

Diagnosing bCSC can be challenging as it shares clinical features with other conditions such as exudative retinal detachment (ERD), and RRD, potentially leading to misdiagnosis[1]. Furthermore, the absence of definitive diagnostic criteria for bCSC complicates its accurate identification and differentiation from mimicking conditions. It is crucial to rule out RRD through careful assessment for retinal breaks. In the present case, the right eye was initially diagnosed with RRD due to a history of blurred vision after intravitreal injection and the presence of subretinal fibrosis. However, no retinal breaks were identified during vitrectomy, prompting a revision of the diagnosis to bCSC. Moreover, the cause of ERD such as Vogt-Konayaki-Harada's disease, posterior scleritis, choroidal tumors must be excluded to avoid unnecessary corticosteroid therapy. Clinical signs of vitreous inflammation and ocular ultrasonography can aid in distinguishing between ERD and bCSC.

The management of bCSC remains controversial due to its rarity and the absence of well-established guidelines specific to this condition. Nevertheless, treatment options typically employed for chronic CSC have also been applied to bCSC patients. For chronic CSC, both PDT were the mainstay and showed significantly greater efficacy compared to control interventions. Studies also reported the effect of eplerenone, another MRAs that also successfully treats CSCR[8-10]. However, recent studies have demonstrated no significant therapeutic advantage of MRAs compared to placebo[3-6]. In this case, vitrectomy with internal drainage was performed in the right eye, leading to improvement, while spironolactone was administered to treat the left eye

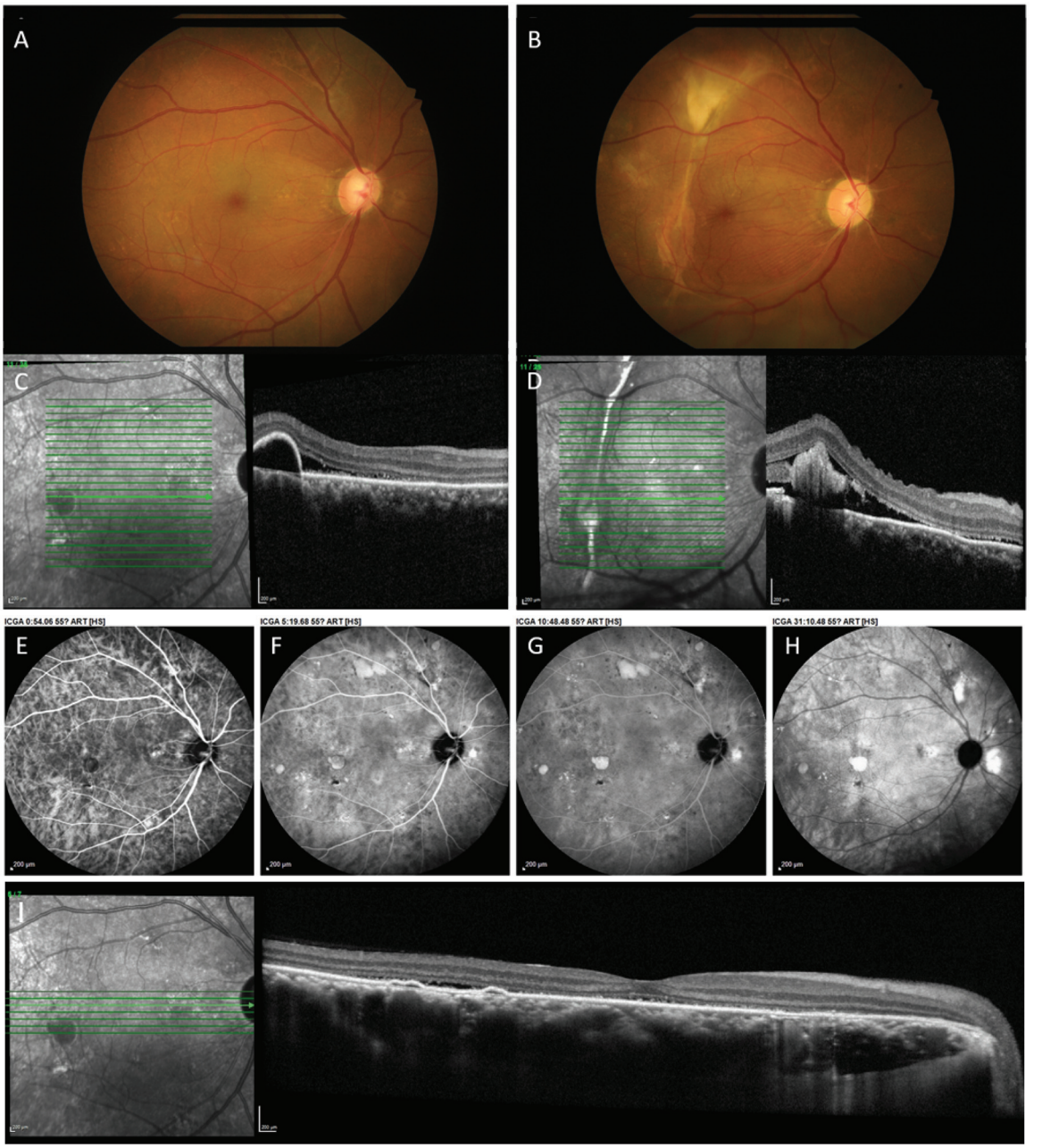

Figure 1. (A) Fundus photograph of the right eye at the first visit showed shallow subretinal fluid at the posterior pole. (B) Fundus photograph of the right eye after third intravitreal bevacizumab injection showed RPE detachment with crescent shaped subretinal fibrosis, and increased of subretinal fluid. (C) An optical coherence tomography of the macula demonstrated subretinal fluid, double layer sign, and dome-shaped serous PED in the right eye. (D) An optical coherence tomography of the right eye after third intravitreal bevacizumab injection showed RPE detachment with subretinal fibrosis, and increased of subretinal fluid. (E, F, G, H) The indocyanine green angiography at 1, 5, 10, 30 minutes respectively, showed early hypercyanescent lesions and RPE leakage in the right eye. (I) The enhanced depth imaging-optical coherence tomography of the right eye at the first visit showed double layer sign, pachyvessels and thickening of the subfoveal choroidal layer.

due to the shortage of PDT, resulting in a favorable response.

Given the limited evidence supporting the use of vitrectomy in bCSC along with the potential complications, the decision to perform vitrectomy in bCSC cases should be approached with caution. While vitrectomy is not considered a standard treatment approach for any type of CSC, there are

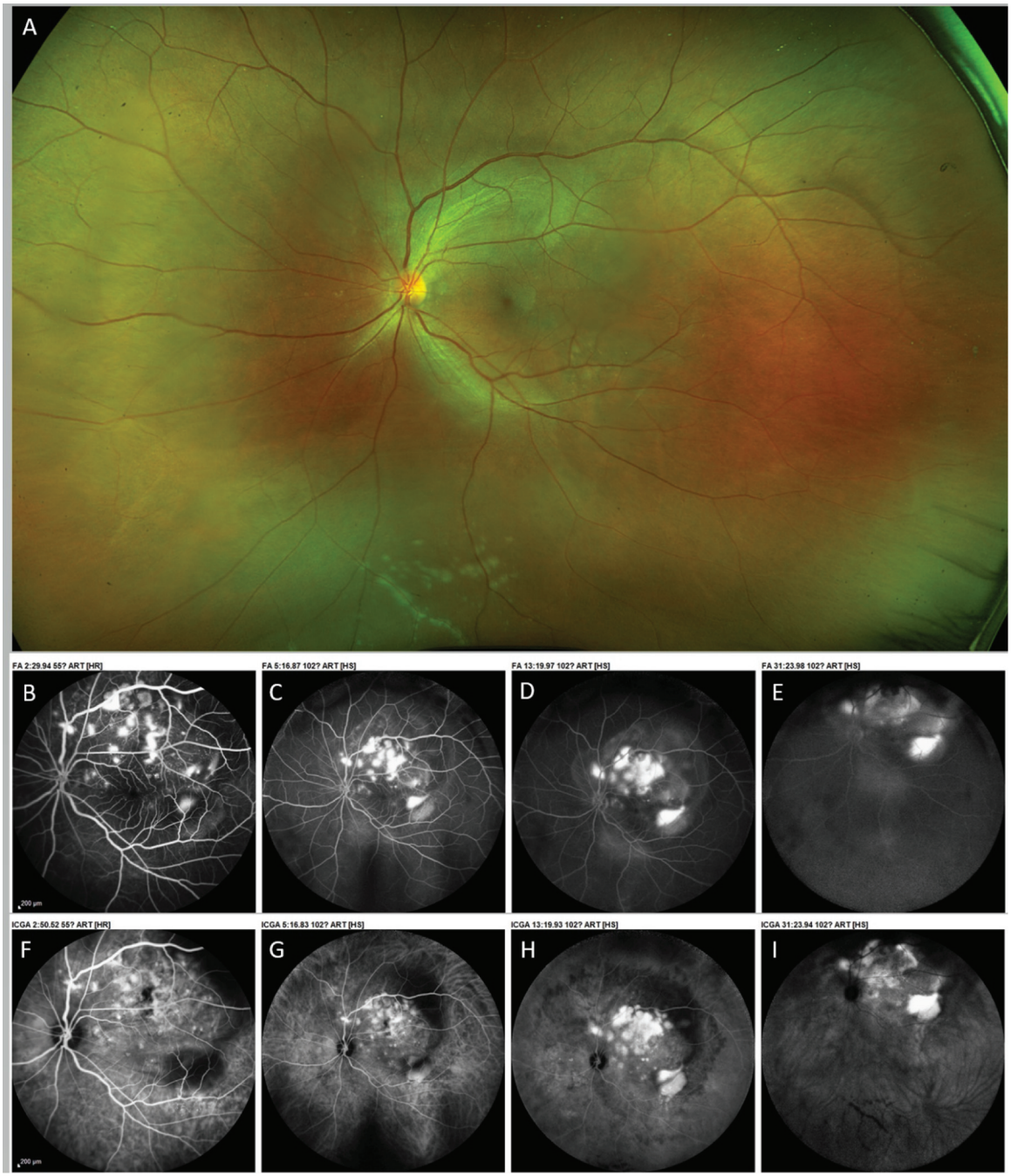

Figure 2. (A) Wide field fundus photo of the left eye. (B, C, D, E) The fluorescein angiography and (F, G, H, I) the indocyanine green angiography of the right eye at 1, 5, 10, 30 minutes respectively showed massive diffused RPE leakage and pooling to the subretinal fluid.

reports suggesting that vitrectomy with internal SRF drainage may facilitate the resolution of SRF in bCSC. However, the long-term outcomes and efficacy of this approach remain uncertain[7,11]. Mechanical drainage of accumulated fluid to the level below the threshold may aid RPE to reabsorb the remaining SRF. Additionally, the tamponade of intraocular gas or other vitreous substitutes may help reduce the hydrostatic pressure causing SRF leakage[12].

In addition, clinical findings in the left eye responded well eye after spironolactone administration. Spironolactone, as MRA, is

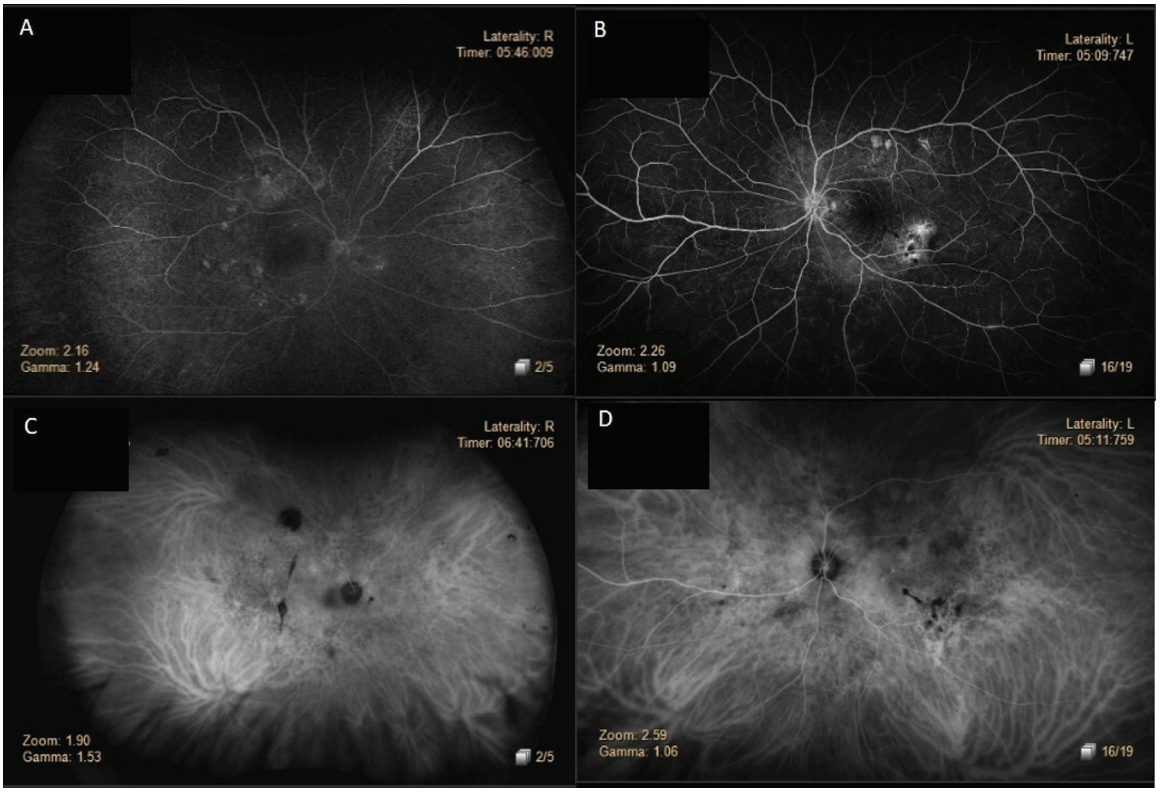

Figure 3. Two weeks after spironolactone was discontinued. (A) Fluorescein angiography of the right eye. (B) Fluorescein angiography of the left eye. (C) Indocyanine green angiography of the right eye. (D) Indocyanine green angiography of the left eye. There was no significant leakage in both eyes.

believed to reduce choroidal vasodilation and hyperpermeability[13], which are implicated in the pathogenesis of CSC. Existing data do not establish MRAs as superior to observation[3-6]. Although previous studies have recommended against the use of mineralocorticoid antagonists in chronic CSC, bCSC often presents with significantly larger volumes of SRF and extravascular substance accumulation. The exact pathophysiology of bCSC remains incompletely understood; however, it is hypothesized to involve increased choroidal vascular permeability and RPE dysfunction[9]. The accumulation of SRF and extracellular substances may further exacerbate fluid leakage. In this context, mineralocorticoid antagonists may offer enhanced efficacy in the resolution of SRF in bCSC compared to chronic CSC[14]. Limited studies have reported the effectiveness of spironolactone. Ramos-Yau et. al. successfully used spironolactone in the patient with bCSC who had previously received oral and peribulbar steroids[15]. While vitrectomy and spironolactone are not established as standard treatments, they may serve as alternative therapeutic options in scenarios where conventional therapies are unavailable or inaccessible due to supply limitations.

Limitations of the present case report stem primarily from its design as a representative case report of a male patient with bCSC, coupled with the lack of a control group receiving standard interventions such as PDT. Given these constraints, the findings lack sufficient generalizability to justify broad clinical application in all bCSC cases. Future research should focus on larger, well-controlled studies, including prospective cohort analyses or systematic reviews, to establish stronger evidence for therapeutic efficacy.

In conclusion, the optimal treatment approach of bCSC remains a subject of debate. The present case report highlights a case of bilateral involvement in which different management were employed for each eye. Vitrectomy with internal drainage and spironolactone proved to be effective in this particular case, indicating that they may serve as viable alternative treatment options for bCSC. Further research and larger studies are warranted to establish evidence-based guidelines for the management of bCSC.

What is already known about this topic?

The bCSC is recognized as an atypical variant of CSC, characterized by its bilateral presentation and the presence of features such as inferior bullous retinal detachment, macular PED, and subretinal exudates. Various treatment modalities have been explored, including PDT, laser photocoagulation, MRAs, and surgical interventions. However, the management of bCSC remains controversial due to its rarity and the absence of well-established treatment guidelines specific to this condition.

What does this study add?

This study enhances the current understanding of bCSC by presenting a case of sequential bilateral bCSC that effectively managed with a surgical intervention (vitrectomy with internal drainage) and pharmacological treatment (spironolactone). The favorable outcomes observed in this case underscore the potential effectiveness of these alternative treatment options in bCSC, particularly when verteporfin PDT is unavailable. Additionally, this case highlights the importance of accurately diagnosing bCSC to avoid mismanagement, emphasizing the need for careful evaluation of clinical features and the exclusion of differential diagnoses.

Ethical approval and consent to participate.

The protocol was exempted by Siriraj Institutional Review Board, Mahidol University (78.071/EC). Written informed consent was obtained from the participant for the publication of the case report with accompanying images.

Conflicts of interest

The authors declare no conflict of interest.

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