Presumed Sympathetic Ophthalmia After Scleral Buckling Surgery: Case Report

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Brief report

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Abstract

Background: We aimed to report a rare case of presumed sympathetic ophthalmia (SO) in a young female patient following scleral buckling (SB). As SB is usually considered an extraocular operation, it is presumed to have low risk of SO.

Case Presentation: A nineteen year-old female patient was referred for visual loss in her left eye because of macula off inferior longstanding rhegmatogenous retinal detachment (RD). Best corrected visual acuity (BCVA) was 20/400 in the left eye. SB with 360 degrees encircling band and inferior segmental tire, with one spot cryoretinopexy at the break site and subretinal fluid drainage was performed. One week after operation, visual acuity was improved to 20/80 and retina was totally attached. Six weeks later, patient came with severe visual loss in both eyes as counting finger 1 meter. Bilateral multifocal serous RD and vitreous cells was found. The patient was treated with intravenous corticosteroid pulse therapy and mycophenolate mofetile as a case of sympathetic ophthalmia. The inflammation was controlled and serous RD resolved after five days intravenous treatment and was not relapsed after six months. BCVA was 20/20 in right eye and 20/50 in the left eye. Systemic workup was negative for any extraocular disease or systemic involvement.

Conclusion: As SB usually considered as a procedure without manipulating intraocular tissues, it is considered to have low risk for SO. In this report, we have shown that SO may occur after successful SB. Inciting the choroid and retinal pigment epithelium with cryoretinopexy or perforating for drainage may induce SO.

Background

Sympathetic ophthalmia (SO) is a rare diffuse granulomatous bilateral panuveitis that may present usually after trauma or surgery in one eye, as penetrate the eye and expose the uvea. The incidence of SO is reported ranging from 0.2–0.5% and 0.01% after intraocular surgery. Surgeries with manipulation and irritation of the choroid and retina and also cyclodestructive surgeries are risk factors. The interval between the onset of SO and the ocular injury differs greatly, ranging from a few days to decades, with most of the cases usually take place within 3 months after injury to the exciting eye and 90% within the first year. The inciting surgeries were various ocular surgeries including trabeculectomy, cataract extraction or secondary intraocular lens placement, vitrectomy, cyclodestruction, iridectomy, and evisceration. Surprisingly, there is an increasing trend of SO after vitreo-retinal intraocular surgeries.

SB is usually considered an extraocular operation, a privilege over vitrectomy, SO is presumed to have a very low risk. Here, we aim to report a rare case successful scleral buckling surgery (SB) combined with subretinal fluid drainage (SRFD) and cryo-retinopexy proceeded to SO.

Case Presentation
A 19-year-old female was referred for visual loss and superior field defect in her left eye. Best corrected visual acuity (BCVA) was 20/20 in the right eye (RE) and 20/400 in the left eye (LE). Anterior segment examination was within normal limits except Shaffer sign in the LE. In fundus exam, macula off inferior longstanding rhegmatogenous retinal detachment (RRD) and three hours of proliferative vitreoretinopathy (PVR), retinal break and lattice degeneration were found. SB with 360 degrees encircling silicone band (silicone band type 240, FCI Inc., Paris, France) and inferior segmental silicone tire (asymmetrical silicone tire type 276, FCI Inc., Paris, France) with one spot cryo-retinopexy at the break site and subretinal fluid drainage due to chronicity of the RRD were performed. One week after operation, BCVA was improved to 20/80 and retina was totally attached.

Six weeks later, patient came back with severe visual loss in both eyes, since two days before. In examination, BCVA of both eyes were measured as counting finger at one meter and anterior segment was within normal limits, except 1+ anterior chamber. Anterior vitreous cells in both eyes and also bilateral multifocal serous retinal detachment were obvious (Fig. 1). In enhanced depth imaging optical coherent tomography (EDI-OCT) (Spectralis HRA+OCT, Heidelberg Engineering, Heidelberg, Germany) multi-lobular serous retinal detachment, septated subretinal spaces, hyperreflective dot reflexes in subretinal fluid, choroidal thickening and undulation of retinal pigment epithelium (RPE) (Fig. 1). Regarding to the examination and imaging, the diagnosis of SO following SB surgery was presumed. Therefore, the patient was admitted and treated for five days with high dose (1 g/day) intravenous infusion of methyl prednisolone followed by oral prednisolone 1 mg/kg. The inflammation was controlled and serous detachment resolved significantly after five days’ pulse therapy (Fig. 2). All of the systemic work-ups for common causes of choriditis including sarcoidosis, tuberculosis, syphilis and autoimmunity markers were inconclusive. Also familial, drug and past medical history were unremarkable. After pulse therapy, oral prednisolone and mycofenolate mofetil 2 gr/day was initiated. On the fourth day, fluorescein angiography (FA) was performed and showed no significant dye pooling except for mild leakage and mixed hypo and hyper-fluorescent dots scattered at the posterior poles presumably due to choroidal hypoperfusion and abnormal leakage (Fig. 2). Because of unavailability of indocyanine green dye at that time, indocyanine green angiography (ICGA) was not performed at the first presentation of SO.

After 3 months of immunsuppressive treatment, ICGA disclosed (Fig. 3) only a few hypofluorescent dark dots in the both eyes and mild background hyperflorescence at the posterior pole indicating response to treatment with mild subclinical inflammation of choroidal stroma. Because of increased blood glucose, prednisolone tapered rapidly and subtenon triamcinolone acetonide 40 mg/1 cc was injected bilaterally. Induced diabetes was controlled with oral medication which was ceased after one month. Prednisolone was reached to 5 mg/d at the end of second month with continuing of MMF 2 gr/d. Blood glucose was checked frequently and it was in normal limit during the follow-up, and was not relapsed during six months follow-ups. Following one year treatment with MMF 2 gr/d and prednisolone 5 mg/d inflammation was in complete remission with complete attached retina in both eyes. Final BCVA was 20/20 and 20/50 in the right and left eyes, respectively. The patient had not any symptom or sign of systemic disease with similar ocular manifestations, especially Vogt-Koyanagi-Harada (VKH).
Discussion

Herein, we report a rare case of SO following uncomplicated SB surgery with cryoretinopexy and SRFD. SO should be kept in mind in any case of bilateral panuveitis associated with multiple serous RD with history of penetrating ocular surgery. SO after vitreoretinal surgery has been reported in several studies. The mechanism of hypersensitivity in the sympathizing eye may be due to a cell-mediated immune response because of the exposure of the uveal tissue to conjunctiva. Use of trans-conjunctival sutureless vitrectomy may be the mechanism that explain the increase in incidence of SO following vitreoretinal surgery because of more uveal incarceration as it was happened in our case.

Kilmartin et al. reported RD surgeries to be the most common procedure associated with development of SO, with the risk of SO after vitrectomy being higher than scleral buckling. The onset of SO symptoms after operation usually occurs between 3 weeks and 6 months after surgery. In this case SO was developed 6 weeks later.

One of the most important differential diagnoses of our case is VKH. It is well known that SO and VKH are common uveitis etiologies with similar clinical and imaging findings in Asian. Although sympathizing eye in SO presents initially with nongranulomatous uveitis clinically and then progresses to granulomatous uveitis. VKH shows 3 successive stages: posterior uveitis, anterior segment involvement associated with posterior uveitis, and finally anterior granulomatous uveitis. SO has variable presentations. In 75% of cases presented with anterior chamber reaction. Anterior uveitis and posterior uveitis both are present in SO patients seen within 2 weeks of disease onset. In our case clinical features are more similar to SO than VKH, in the other hand our case had not any systemic sign or symptoms attributed to VKH or other systemic diseases causing choroiditis. The main differentiating clue between SO and VKH is the history of prior surgery or trauma in SO.

In terms of imaging findings in SO, on FAG multiple hyperfluorescent pinpoints leakages associated with late pooling resembling VKH and hypofluorescent foci during the early phase of angiography with late hyperfluorescence similar to acute posterior multifocal placoid pigment epitheliopathy (APMPPE) can be observed. In our case it seems that because of FAG timing after 4 days steroid therapy, hyperfluorescence was less and hypofluorescent that is compatible with location of granuloma and cellular infiltration was more prominant. On ICGA the most common features are multiple hypocyanescent spots. In acute phase of SO, EDI OCT discloses multiple serous RD associated with hyper-reflective septa, massive choroidal thickening and loss of normal choroidal vascular architectures, irregular outer segments of photoreceptors similar to what are observed in acute phase of VKH.

Visual outcome is worse in SO as compared with VKH disease. However, Vision was improved in our case because of early diagnosis and prompt aggressive and adequate treatment as it can be observed on
ICGA was taken 3 months after treatment.

**Conclusion**

In conclusion, it is noteworthy to keep in mind SO in any case of bilateral uveitis or bilateral serous RD following scleral buckling with uveal tract violation such as cryopexy or subretinal fluid drainage.

**Abbreviations**

BCVA: best corrected visual acuity

EDI-OCT: enhanced depth imaging optical coherent tomography

OCT: optical coherent tomography

FA: fluorescein angiography

RE: right eye

LE: left eye

RD retinal detachment

RRD: rhegmatogenous retinal detachment

RPE: retinal pigment epithelium

SB: scleral buckling

SRFD: subretinal fluid drainage

SO: sympathetic ophthalmia

**Declarations**

**Ethics Approval**: Not Applicable

**Consent for publication**: Written consent for images and data publication and identifying clinical details was obtained from the patient.

**Availability of data and material**: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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**Authors’ contributions:** All the authors contributed significantly to this research, and all authors agree to be accountable for all aspects of the work. SMH participated in patients’ participating in clinical analysis and interpretation of imaging, and in revising the draft. NS, MG, MAz helped to gathering patient’s evaluations and draft the manuscript. MAb participated in acquisition of clinical data, clinical analysis and interpretation, and revising and finalizing the manuscript. All authors read and approved the final manuscript.

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**Figures**
Figure 1

Fundus photography of the right (A) and the left (B) eye reveals bilateral disc swelling and multifocal serous RD at the posterior pole. EDI OCT of the right (C) and the left (D) eye at first presentation after development of SO discloses multi-lobular serous retinal detachment, septated subretinal spaces, hyper-reflective dots in subretinal fluid, choroidal thickening and undulation of RPE.
Figure 2

Five days after pulse corticosteroid therapy, fluorescein angiography of the right (A) and left (B) eye showed no significant dye pooling except for mild leakage and hypofluorescent dots scattered at the posterior poles, mostly in the left eye. EDI OCT of the right (C) and left (D) eyes showed significant response with resolution of serous retinal detachment and decreased choroidal thickening.
ICGA of the right (A,B) and the left eye (C,D) at third months disclosed only a few hypofluorescent dark dots (HDD) in the both eyes and mild background hyperfluorescence at the posterior pole indicating response to treatment with mild subclinical inflammation of choroidal stroma.
Figure 4

EDI OCT of the right (A) and left (B) eyes three months after corticosteroid therapy showed complete response with resolution of serous retinal detachment and decreased choroidal thickening and inflammation.

Supplementary Files

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- supplement8.pdf