Presumed Sympathetic Ophthalmia after Scleral Buckling Surgery: Case Report

Seyyedeh Maryam Hosseini  
Mashhad University of Medical Sciences

Nasser Shoeibi  
Mashhad University of Medical Sciences

Mahdieh Azimi Zadeh  
Mashhad University of Medical Sciences

Mahdi Ghasemi  
Mashhad University of Medical Sciences

Mojtaba Abrishami  
Tehran University of Medical Sciences  
https://orcid.org/0000-0003-2001-7929

Brief report

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Abstract

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

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, BCVA 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 diagnosed as sympathetic Ophthalmia, and treated with intravenous corticosteroid pulse therapy and mycophenolate mofetil. The inflammation was controlled and serous RD resolved after five days intravenous treatment and was not relapsed after six months. BCVA became 20/20 in right eye and 20/50 in the left eye after six months. 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 presented SO occurrence after successful SB. Inciting the choroid and retinal pigmen epithelium with cryoretinopexy or perforating for drainage may induce SO.

Introduction

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

Scleral buckling (SB) is usually considered as an extraocular operation, a privilege over vitrectomy, and SO is presumed to have a very low risk after SB. Here, we report an unusual rare case of SO following successful SB surgery which was combined with subretinal fluid drainage (SRFD) and cryo-retinopexy.

Case Presentation
A nineteen-year-old female was referred for visual loss and superior field defect in her left eye (LE). Best corrected visual acuity (BCVA) was 20/20 in the right eye (RE) and 20/400 in the 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), three hours of proliferative vitreoretinopathy (PVR), retinal break and lattice degeneration were found. Patient had no history of head or ocular trauma or any intraocular surgery. 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 SRFD due to chronicity of the RRD were performed. SRFD was not inadvertent. After passing the band and tire, scleral thinning was performed. After cautery of the choroid, the needle of polyester spatula suture (Mersilene, Ethicone LLC, Johnson and Johnson Inc., USA) was used to drain the fluid. One week after operation, BCVA was improved to 20/80 and retina was totally attached. The patient was followed by topical antibiotic (Chlobiotic, Sina Darou, Tehran, Iran) and topical corticosteroid (Bethasonate, Sina Darou, Tehran, Iran) for three weeks. 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 and anterior vitreous cells in both eyes. Bilateral multifocal serous retinal detachment was obvious in funduscopy(Figure 1 A,B). In enhanced depth imaging optical coherence 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) were found (Figure 1 C, D). Chorioscleral junction was not detected due to diffuse severe choroidal thickening. Regarding to the examination and imaging, absence of any other ocular trauma history except for SB surgery, the diagnosis of SO following SB surgery was presumed. Therefore, the patient was admitted and treated for five days with high dose (1g/day) intravenous infusion of methyl prednisolone. The inflammation was controlled and serous detachment resolved significantly after five days’ pulse therapy (Figure 2). All of the systemic work-ups for common causes of choroiditis including sarcoidosis, tuberculosis, syphilis and autoimmunity markers were inconclusive. Also familial, drug and past medical history were unremarkable. After pulse therapy, oral prednisolone (50 mg/day) and mycophenolate mofetil (MMF) (2 gr/day) were 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. Because of unavailability of indocyanine green dye at that time, indocyanine green angiography (ICGA) was not performed at the first presentation of SO. Because of increased blood glucose, prednisolone tapered rapidly and subtenon Triamcinolone acetonide (TriamHEXAL, Hexal AG, Holzkirchen, Germany) (40mg/1cc) , 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
2gr/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.

After 3 months of immunosuppressive treatment, ICGA disclosed (Figure 3) only a few hypofluorescent dark dots in the both eyes and mild background hyperfluorescence at the posterior pole indicating response to treatment with mild subclinical inflammation of choroidal stroma.

**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\(^2, 4-6\). It is postulated that trauma to the uveal tract in the context of inadvertent perforation, SRFD or cryotherapy during SB surgery might be deliberate uveal antigens, melanin or outer photoreceptor antigens resulting access to the lymphatic systems of the conjunctival tissue exciting delayed hypersensitivity reaction inside the eye.\(^4-6\) The mechanism of hypersensitivity in the sympathizing eye may be due to the exposure of the uveal tissue to the conjunctival lymphatic system resulting in a cell-mediated immune response.\(^6\) This mechanism can explain the association of increase in the trend of the use of trans-conjunctival sutureless vitrectomy with increase in incidence of SO following vitreoretinal surgery because of more uveal incarceration\(^6\). In our case, SRFD perhaps increased the risk of uveal exposure to conjunctival lymphatic tissue. In one of the largest study of SO following surgery, 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 twice of that external scleral buckling without any sex predilection.\(^7\)

The onset of SO symptoms after operation usually occurs between 3 weeks and 6 months after surgery because of delayed hypersensitivity.\(^8\) In this case SO was developed 6 weeks later.

In a case series, Ozbek et al.\(^9\) reported three cases of SO following SB, however 2 cases had combined vitrectomy with SB and only in one case SO occurred following encircling buckle combined with 360 indirect retinal photocoagulations and SRFD in contrast to our case that had only one cryoretinopexy spots and one drainage site. In another similar case report by Parvaresh and Falavarjani, SO was found in a case with history of SB revision after 4 years, which the second surgery was combined with SRFD and cryoretinopexy.\(^10\) However, in our case, SO was happened in the first episode, without any surgical past history.

In a recent study by Tyagi et al, incidence of SO following vitreoretinal surgery was 0.038 % of all vitrectomy cases, and 9% of all cases of SO had vitreoretinal surgery.\(^6\) Seventy-five percent of cases underwent multiple ocular surgery before development of SO.\(^6\) The most common anterior segment finding was non-granulomatous uveitis in 50 % in contrast to serous RD in 62.5 % of cases.\(^6\) In our case
we also found that anterior segment inflammation was less severe than posterior segment chorioretinal findings.

Most of the case presented in studies\textsuperscript{5-10} had simultaneous SB and vitrectomy or had previous trauma or multiple surgery, however our case had no extensive retinal or uveal tissue manipulation and had no combined surgery or previous surgery, so we assumed this would be of interesting. One of the plausible theory for this susceptibility might be Asian ethnicity and higher prevalence of VKH in our area.

One of the most important differential diagnoses of our case is VKH. It is well known that VKH is common uveitis etiology with similar clinical and imaging findings with SO\textsuperscript{1,11}. Although sympathizing eye in SO presents initially with nongranulomatous uveitis clinically, it progresses to granulomatous uveitis\textsuperscript{8}. VKH shows 3 successive stages: posterior uveitis, anterior segment involvement associated with posterior uveitis, and finally anterior granulomatous uveitis. Anterior uveitis and posterior uveitis both are present in SO patients seen within 2 weeks of disease onset\textsuperscript{8}. In our case clinical features are more similar to SO than VKH, as our patient had no 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, during the early phase of angiography multiple hyperfluorescent pinpoints leakages associated with late pooling resembling VKH and hypofluorescent foci were found. In the late phase of FAG, hyperfluorescence similar to acute posterior multifocal placoid pigment epitheliopathy (APMPPE) can be observed\textsuperscript{11}. In our case it seems that because of FAG was performed after 4 days of steroid therapy, hyperfluorescence was less and hypofluorescent that is compatible with location of granuloma and cellular infiltration was more prominent. On ICGA the most common features are multiple hypocyanescent spots. In acute phase of SO, EDI-OCT discloses multiple serous retinal detachments associated with hyperreflective 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\textsuperscript{8}. Moreover, BCVA was improved in our case because of early diagnosis and prompt aggressive and adequate treatment.

**Conclusion**

In conclusion, it is noteworthy to keep in mind diagnosis of sympathetic ophthalmia 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

ICGA: Indocyanine green 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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