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Case Report

Sympathetic ophthalmia caused by a severe ocular chemical burn: a case report and literature review

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Abstract: We herein presented a case of sympathetic ophthalmia induced by chemical burns. A 39-year-old male was referred to our retinal clinic complaining of sharply decreased vision in his right eye, with photophobia and headache. He had suffered severe chemical burns five months ago, and his left eye underwent amniotic membrane transplantation combined with tarsorrhaphy for persistent corneal ulceration. A comprehensive examination was performed. After excluding other infectious ocular diseases and systemic inflammatory disease, Vogt-Koyanagi-Harada was considered. A regimen of 1.5 mg/kg prednisone per day was initiated, and tapered to a weekly dose. The inflammation in the patient's right eye was improved, and the best-corrected visual acuity recovered to 20/50. He continued to be seen by his corneal specialist for follow-up on the left eye. A regular B-scan was performed, and atrophy of the left eyeball was detected 2 months later. Upon removal of the left eyelid sutures, corneal perforation with a prolapsed iris was found. Afterwards, the inflammation of the right eye reoccurred, and the diagnosis was revised to sympathetic ophthalmia. The patient underwent an enucleation procedure on the left eye, and a massive prednisolone regimen was initiated, and then tapered slowly over one year. Pathological results showed diffuse epithelioid cells, polymorphonuclear giant cells, and lymphocyte infiltration in the uveal tissue. Sympathetic ophthalmia was confirmed. The right eye remained unremarkable at 41 months of follow-up. An uncommon case is presented of sympathetic ophthalmia following severe chemical burns. It is notable that sympathetic ophthalmia developed after chemical burns to the ocular surface and corneal perforation, despite reconstructive treatment combined with tarsorrhaphy.

Keywords: Sympathetic ophthalmia, ocular burn, cornea perforation

Introduction

Sympathetic ophthalmia (SO) and Vogt-Koyanagi-Harada (VKH) disease are autoimmune disorders targeting melanin-bearing cells. Both diseases share similar ocular and systemic manifestations, such as bilateral granulomatous panuveitis, alopecia, poliosis, and vitiligo. According to the diagnostic criteria, SO patients should have a history of intraocular surgery or penetrating injury [1]. SO induced by ocular chemical burns is very uncommon. Recently, Zhang et al. reported that only 1 of 22 cases of SO was caused by alkali burns, from among 9,103 patients with globe injuries [2].

The management of SO remains controversial [1-4]. Some authors have recommended that early enucleation of the injured eye should be performed to prevent SO [5, 6], while others have suggested that this may not be absolutely preventative [2, 7]. Consequently, therapeutic strategies for treating SO induced by chemical burns have not yet been determined, and need further investigation.

Here, we present a case of SO in a patient who received severe chemical burns. The clinical manifestation, therapeutic regimen, and prognosis are reviewed.

Case report

A 39-year-old male was referred to our retinal clinic complaining of sharply decreased vision in his right eye, with photophobia and headache. He had suffered severe chemical burns five months previously, and his left eye under-

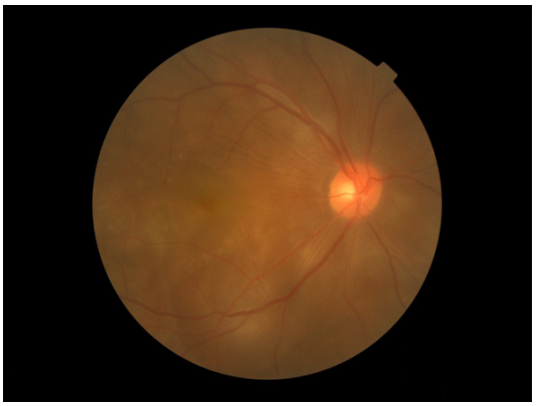

Figure 1. Examination of the right eye revealed a granulomatous panuveitis with diffuse mutton-fat keratic precipitates (KP), iris posterior synechiae, anterior chamber cells (1+), flares (1+), radial retinal folds surrounding the macula, and a subretinal yellow-white exudates.

went amniotic membrane transplantation (AMT) combined with tarsorrhaphy for persistent corneal ulceration. A comprehensive examination was performed. The best-corrected visual acuity (BCVA) of his right eye was counting fingers; the left eyelids had been sutured together. Examination of the right eye revealed a granulomatous panuveitis with diffuse mutton-fat keratic precipitates (KP), iris posterior synechiae, anterior chamber cells (1+), flares (1+), radial retinal folds surrounding the macula, and a subretinal yellow-white exudate (**Figure 1**). Fluorescence angiography (FA) revealed multiple pinpoint early-stage retinal pigment epithelium leaks, dye pooling with multiple lake-like areas, and late-stage optic disk hyperfluorescence (**Figure 2**). Optical coherence tomography (OCT) showed multifocal serous neural retinal detachment in the posterior pole. B-scan ultrasonography revealed a bilaterally thickened choroids and vitreous opacity (**Figure 3**). After excluding other infectious ocular diseases and systemic inflammatory disease, VKH was considered. A regimen of 1.5 mg/kg prednisone per day was initiated, and tapered to a weekly dose. The inflammation in the patient's right eye was improved, and the BCVA recovered to 20/50. He continued to be seen by his corneal specialist for follow-up on the left eye. A regular B-scan was performed, and atrophy of the left eyeball was detected 2 months later. Upon removal of the left eyelid sutures, corneal perforation with a prolapsed iris was found (**Figure 4**). Afterwards, the inflammation of the right eye reoccurred, and the diagnosis was revised to SO. The patient underwent an enucleation procedure on the left eye, and a massive prednisolone regimen was initiated, and then tapered slowly over one year. Pathological results showed diffuse epithelioid cells, polymorphonuclear giant cells, and lymphocyte infiltration in the uveal tissue (**Figure 5**). SO was confirmed.

The right eye remained unremarkable at 41 months of follow-up (**Figure 6**). The BCVA was 20/30, and the fundus showed a sunset glow appearance, with diffuse decreases in retinal thickness by OCT.

Discussion

The pathogenesis of SO is unclear. The possible mechanisms include an autoimmune hypersensitivity response directed against exposed uveal tissue in the traumatised (exciting) eye, which is responsive to several chorioretinal antigens, such as S antigen or Mart-1 melanoma antigen [1]. Factors that contribute to the development of clinically apparent sympathetic ophthalmia are: penetrating trauma, surgical repair 48 h or more after initial injury, the use of local and systemic steroids for more than 1 week after an initial injury, and injury to the ciliary body. However, there is also a risk of developing SO in the absence of a penetrating injury of the uveal tract [8, 9]. The diagnosis of sympathetic ophthalmia is based on the patient's history and clinical examination. In our case, due to the lack of obvious penetrating injury history and the sutured state of the left eyelids, Vogt-Koyanagi-Harada disease (VKH) was initially considered. VKH shares similar clinical features with SO, such as granulomatous panuveitis, and exudative retinal detachment. Furthermore, the immunologic mechanisms also seem to be similar [1]. A high-dosage systemic steroid therapy was administered, and the inflammation of the right eye was rapidly controlled. When the left eyelid sutures were removed, corneal perforation with iris prolapse was found, and the diagnosis was revised to SO.

The incidence of SO induced by chemical burns is low, and the optimal therapeutic methods remain unclear, due to the paucity of cases. Treatment of chemical and thermal injuries of

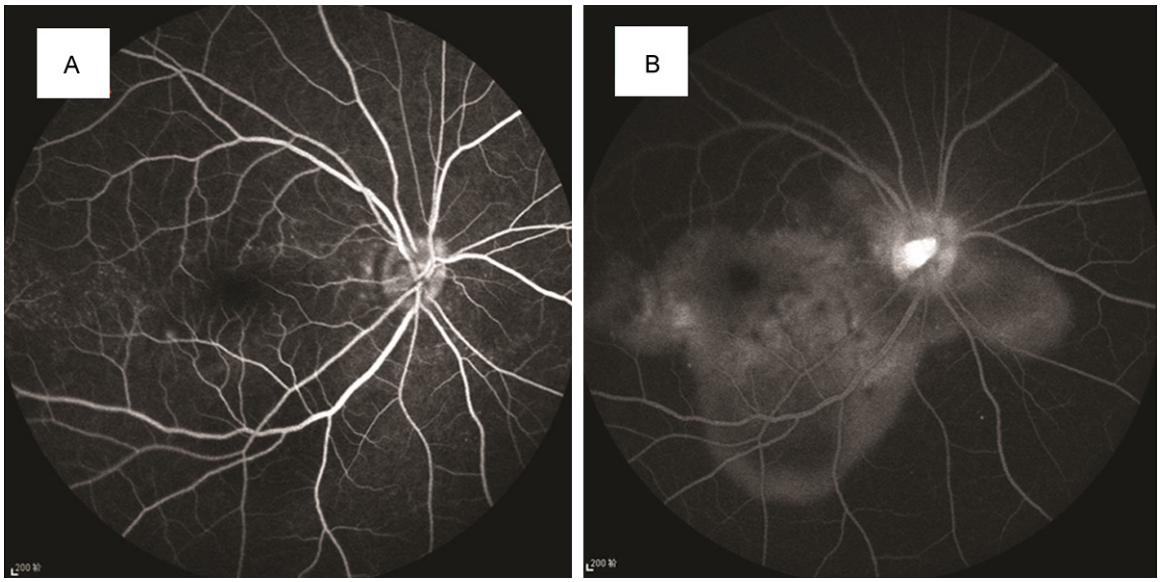

Figure 2. Fluorescent angiography revealed multiple pinpoint early-stage retinal pigment epithelium leaks (A), dye pooling with multiple lake-like areas, and late-stage optic disk hyperfluorescence (B).

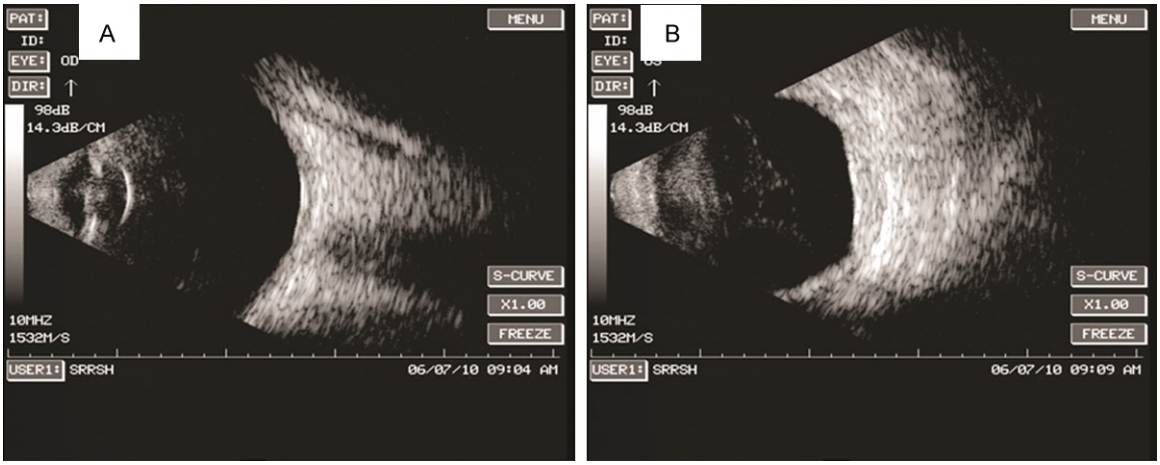

Figure 3. B-scan ultrasonography revealed a bilaterally thickened choroids and vitreous opacity (A: right eye; B: left eye).

the eye is a challenging problem. AMT with tarsorrhaphy is regarded as an efficacious treatment method for these patients. The purpose of the treatment is to restore the corneal and conjunctival epithelial surfaces, as well as to prevent ocular tissue melting. It is generally thought that enucleation of the exciting eye should be performed to prevent SO [5, 6]. However, some have suggested that care should be taken before making a final decision, even in situations where enucleation seems inevitable after ocular trauma [10]. In our case, corticosteroids were introduced initially in the right eye, and the final result was positive.

It is difficult to observe changes to the eyeball after tarsorrhaphy. We used B-scan ultrasonography to detect changes in the sutured eye. When the appearance of panuveitis was noted in the patient's right eye, the ultrasound images of both eyes revealed bilateral thickened choroids. When the constricted eyeball was noted, we removed the sutures in time. Therefore, after tarsorrhaphy, B-scan is useful for detecting changes in the eye during follow-up. Once SO is diagnosed, the enucleation of the damaged eyeball should be performed in time to prevent it from harming the contralateral eye.

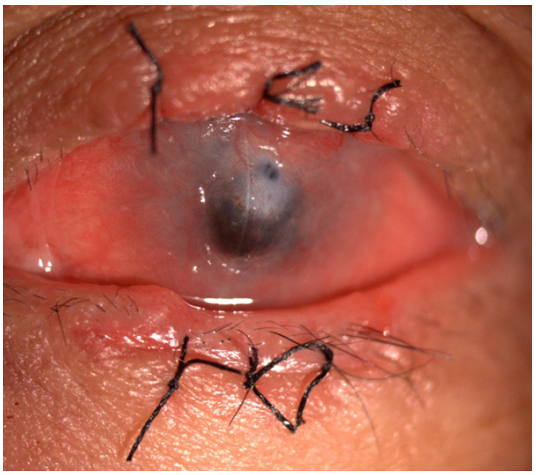

Figure 4. Upon removal of the left eyelid sutures, corneal perforation with a prolapsed iris was found.

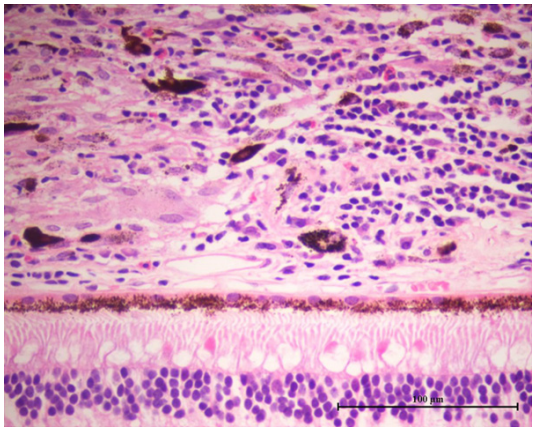

Figure 5. Postoperative pathological results showed diffuse epithelioid cells, polymorphonuclear giant cells, and lymphocyte infiltration in the uveal tissue.

In general, the time of SO development varies with the inducing event. SO after trauma occurs sooner than in surgically induced cases, at a median of 6.5 months after the inciting trauma, compared with 14.3 months after surgery [11]. The post-event interval in SO caused by chemical burns may be as long as 22 years [2]. In our case, SO developed 7.5 months after the patient received chemical burns.

Conclusion

An uncommon case is presented of sympathetic ophthalmia following severe chemical burns. It is notable that sympathetic ophthalmia developed after chemical burns to the ocular surface and corneal perforation, despite reconstructive treatment combined with tarsorrhaphy.

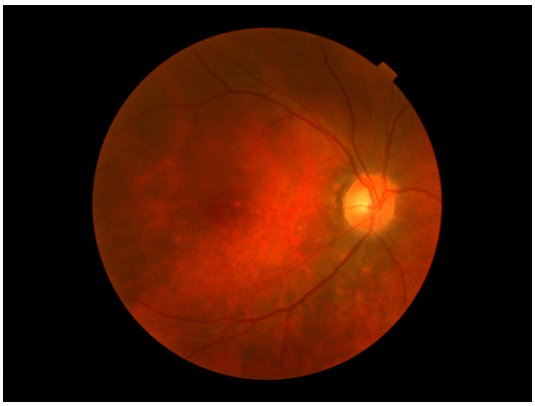

Figure 6. The colour photography showed a sunset glow appearance at 41 months of follow-up.

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Disclosure of conflict of interest

None.

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