

AN ATYPICAL CASE OF SYMPATHETIC OPHTHALMIA AFTER LIMBAL CORNEAL LACERATION

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Background/Purpose: To report a case of atypical sympathetic ophthalmia after limbal corneal laceration.

Methods and Results: An 11-year-old child had a successful left eye corneal laceration repair at the temporal limbus with excision of exposed nonnecrotic iris tissue, resulting in good visual acuity of 20/80 and 20/25 on postoperative Days 1 and 7, respectively. The patient was prescribed 1 mg/kg oral prednisolone in a tapering dose as prophylaxis. On postoperative Day 21, the patient presented with acute onset decreased vision in both eyes. Visual acuity was counting fingers 3 feet in both eyes. On examination, anterior segment examination was quiet without any inflammation, anterior vitreous face showed 1+ cells, and dilated funduscopy revealed bilateral symmetrical serous retinal detachments along the posterior pole. Optical coherence tomography demonstrated separation and elevation of inner neurosensory layers from the outer segment marking presence of hyperreflective material along with subretinal fluid between detached surfaces. There was stippled hyperfluorescence along the posterior pole as seen in fluorescein angiography. With a diagnosis of sympathetic ophthalmia confirmed, oral prednisolone (2 mg/kg body weight) was instituted after which, there was gradual decrease in macular elevation with corresponding improvement in visual acuity with no recurrence for the last 6 months.

Conclusion: To our knowledge, this is the first reported instance of an atypical presentation of sympathetic ophthalmia, and antecedent corticosteroid therapy would have mitigated robust anterior segment findings usually associated with the condition.

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Sympathetic ophthalmia is a rare phenomenon with an incidence of 0.03 per 100,000 per year.¹ Penetrating injuries involving uveal tissue and retinal surgeries are common causes.^{2–4} Plaque brachyther-

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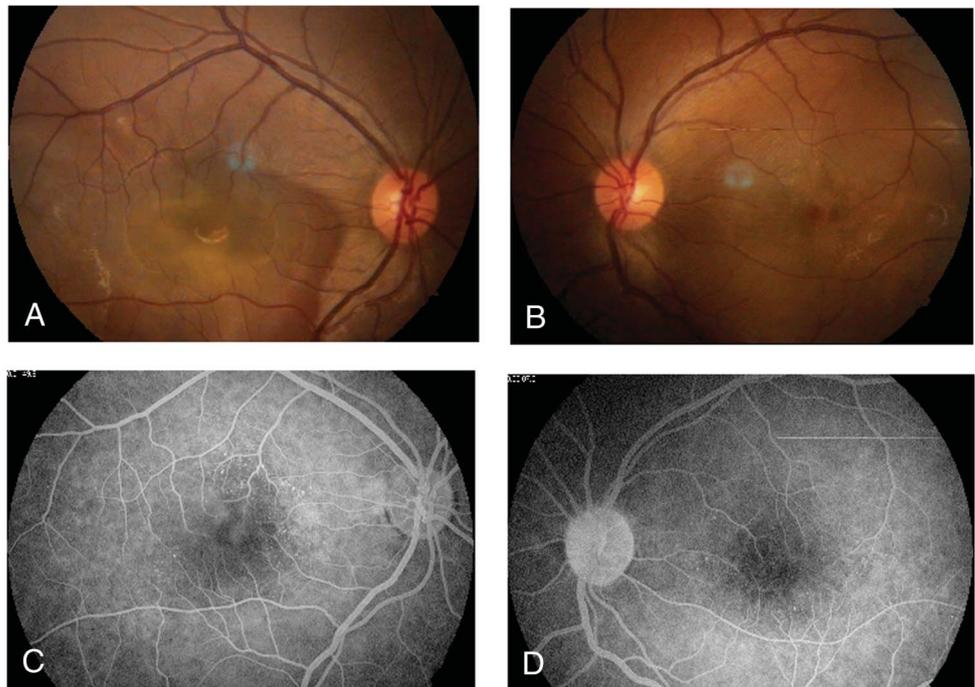
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apy,⁵ fungal keratitis,⁶ and cyclodestructive procedures⁷ have been reported to be rarely associated. There is a delayed hypersensitive reaction to sequestered uveal antigen leading to the damage of outer retinal pigment epithelium layer of the retina.^{8,9} Sympathetic ophthalmia has biphasic peaks in children and the elderly people because of greater incidence of accidental trauma and ocular surgery, respectively.¹⁰ In this study, we present a case of accidental corneal injury that developed sympathetic ophthalmia despite prophylactic systemic steroid therapy. The efficacy of optical coherence tomography (OCT) in following the course of the disease and correlating visual recovery with that of anatomical normalcy is also reported.¹¹

Case Report

An 11-year-old male child presented with complains of pain and decreased vision in the left eye for 3 days after penetrating

Fig. 1. A and B. Colour fundus photographs exhibiting serous macular elevation. C and D. Fundus fluorescein angiogram photographs in peak arteriovenous filling exhibiting stippled hyperfluorescence in the posterior pole.

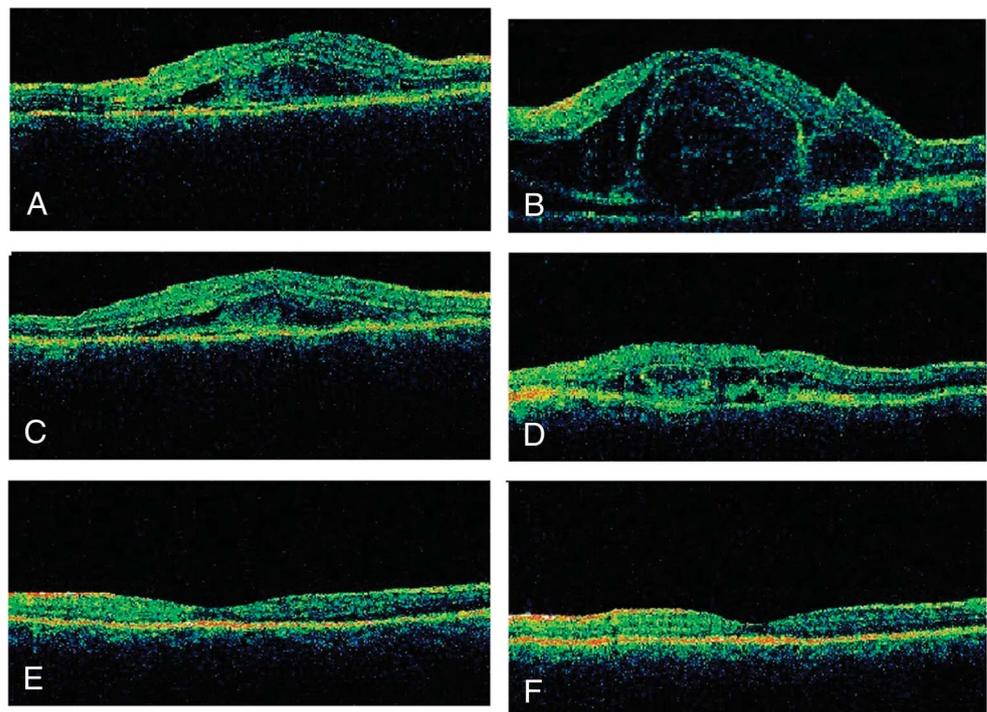


pencil injury. His visual acuity in the right eye was 20/20 and left eye 20/120. Examination of the left eye revealed full thickness corneal laceration at the temporal limbus with iris prolapse, clear lens, and normally appearing fundus. Corneal laceration repair was performed followed by excision of the exposed normal-appearing iris tissue, and apposition of corneal margins. The patient was treated with oral prednisolone 1 mg/kg

body weight, a plan to gradually taper the dose over 6 weeks. Visual acuity rapidly improved from 20/80 on postoperative Day 1 to 20/25 on postoperative Day 7.

On postoperative Day 21, the patient presented with sudden-onset, rapidly progressive visual loss in both eyes over the last 2 days. His visual acuity was counting fingers at 3 feet in both eyes. He was still on oral prednisolone therapy with a dose of 10 mg per

Fig. 2. A and B. Optical coherence tomography of both eyes showing exudative retinal detachment. C and D. Gradual reduction of macular elevation is seen. E and F. Complete resolution of serous macular elevation with attainment of normal contour with 15 days of high corticosteroid therapy.



day. Dilated fundus examination showed clear optical media with bilateral gross serous elevation of the macula (Figure 1, A and B) and occasional cells in the anterior vitreous face. Optical coherence tomography (Stratus OCT; Carl Zeiss Meditech, Dublin, CA) revealed separation of inner neurosensory layer from the outer hyperreflective area (retinal pigment epithelium layer) with accumulation of subretinal fluid along with exudation, but there was no evidence of cystoid spaces in the inner neurosensory layer (Figure 2, A and B). Fundus fluorescein angiogram (Figure 1, C and D) demonstrated stippled hyperfluorescence in the posterior pole. Analyzing the above features, a diagnosis of sympathetic ophthalmia was made; however, it was quite atypical owing to the absence of keratic precipitates and anterior chamber reaction and posterior synechiae. The patient was prescribed higher dose of oral prednisolone (2 mg/kg body weight), which was tapered by 10 mg every 10 days and terminated at 12 weeks. On Day 3 of increased steroid usage, OCT revealed reduction of macular elevation in both eyes with corresponding improvement in visual acuity (20/200 in both eyes) (Figure 2, C and D). At 15 days of increased steroid usage, his visual acuity was 20/20 in both eyes, and there was complete resolution of macular elevation with restoration of normal foveal contour (Figure 2, E and F). At subsequent follow-up for 6 months, the vision of the patient was well preserved and there was no evidence of recurrence of clinical signs of sympathetic ophthalmia.

Discussion

The diagnosis of sympathetic ophthalmia is based on clinical examination and evaluation of history.^{12,13} However, ocular investigations like fundus fluorescein angiogram and OCT are useful adjuncts in establishing the diagnosis.^{14,15} It classically manifest as bilateral granulomatous pan-uveitis with a definitive history of penetrating trauma and rarely by blunt trauma.¹⁶ Posterior segment shows moderate to dense vitritis, choroiditis, and papillitis with multiple exudative retinal detachments.^{17,18} The onset of disease is within 1 year in 90% of patients and 17% present within 1 month.^{17,19} Our patient presented on the 28th day of traumatic repair and 30th day of trauma. None of the anterior segment findings as seen in typical sympathetic ophthalmia could be elucidated in our patient possibly attributed to prior steroid therapy. Kumar et al²⁰ showed 30% of isolated posterior segment findings in their case series on sympathetic ophthalmia. Gupta et al²¹ demonstrated that 22 of their 40 patients presented with exudative retinal detachment with no evidence of anterior segment inflammation, leading to the conclusion that lone posterior segment findings may be indicative of early diagnosis where anterior segment has not yet involved or it is an atypical presentation. Our patient presented with lone posterior segment findings, which is very consistent with 2 of the previous case series.^{20,21} Isolated posterior segment findings could be explained by previous immunosuppression in the immediate postoperative period.

Optical coherence tomography is a useful noninvasive tool in the diagnosis and in determining the efficacy of treatment in sympathetic ophthalmia.^{22,23} Optical coherence tomography demonstrates exudative retinal detachments and its reduction marks the response to treatment. Our patient too had gradual reduction in exudative retinal separation in OCT after steroid therapy. Sympathetic ophthalmia is treated with immunosuppressive therapy. Because of the high risk of recurrence, patients needs timely follow-up. Recurrence calls for institution of other immunosuppressive therapy such as chlorambucil and azathioprine.²⁴ In our case, there was complete resolution of exudative retinal detachment with high-dose steroids, which was maintained for 6 months and showed no signs of recurrence undermining the need of immunosuppressants.

Conclusion

Sympathetic ophthalmia is a rare phenomenon and can still occur despite attempted prophylaxis with corticosteroid therapy and that OCT findings parallel clinical improvement. The present case is reported owing to its rarity and unusual presentation.

Key words: corneal laceration, OCT, open globe injury, exudative macular elevation, uveal prolapse.

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