Case Report

Ophthalmology and Allied Sciences
Volume 2 Number 1, January - June 2016
DOI: http://dx.doi.org/10.21088/oas.2454.7816.2116.6

An Atypical Case of Sympathetic Ophthalmia Following Zone 1 Corneal Injury

Praveen Subudhi*, B.N.R. Subudhi**

Authors Affiliation: *Consultant, Ruby Eye Hospital and Research Center, Govinda Vihar, Berhampur, Odisha. **Professor, M.K.C.G Medical College, Berhampur, Odisha, India.

Abstract

Purpose: To report a case of atypical sympathetic ophthalmia following limbal corneal laceration. *Methods and Results:* An eleven year oldchild had a successful left eye (OS) corneal laceration repair at the temporal limbus with excision of exposed non necrotic iris tissue, resulting in good visual acuity of 20/80 and 20/25 postoperative day 1 and 7 respectively. The patient was prescribed 1mg/kg oral prednisolonein a tapering dose as prophylaxis. Post operative day 21, 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 dilatedfundoscopy revealed bilateral symmetrical serous retinal detachmentsalongthe posterior pole. Optical coherence tomography (OCT)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 theposterior pole as seen in fluorescein angiography. With a diagnosis of sympathetic ophthalmia confirmed, oral prednisolne (2 mg/kg body weight) was instituted following which, there was gradual decrease in macular elevation with corresponding improvement in visual acuity with no recurrence for last 6 months. *Conclusion:* To our knowledge, this is the first reported instance of an atypical presentation of sympathetic ophthalmia and antecedant corticosteroid therapy would have mitigated robust anterior segment findings usually associated with the condition.

Keywords: Corneal Laceration; OCT; Open Globe Injury; Exudative Macular Elevation; Uveal Prolapse.

Introduction

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 brachytherapy [5], fungal keratitits [6]and cyclodestructive procedures [7] have been reported to be rarely associated. There is a delayed hypersensitivity reaction to sequestered uveal antigen leading to the damage of outer RPE layer of retina [8,9]. Sympathetic ophthalmia has biphasic peaks in children and the elderly because of greater incidence of accidental trauma and ocular surgery respectively.[10] Herebywe present a case of accidental corneal injury that developed sympathetic ophthalmia inspite of 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 anatomic normalcy is also reported [11].

Case Presentation

An 11 year old male child presented with complains of pain and decreased vision in left eye

Reprint Request: Praveen Subudhi,
Ruby Eye Hospital and Research center, Govinda Vihar, Berhampur-760001, Ganjam, Odisha India.
E-mail: subudhipraveen@gmail.com,

for 3 days following penetrating pencil injury.His visual acuity in right eye was 20/20 and in left eyewas 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 exposed normal appearing iris tissue and apposition of corneal margins (Figure 1). The patient was treated with oral prednisolone 1 mg/kgbody weight, a plan to gradually taper the doseover 6 weeks. Visual acuityrapidly improved from 20/80 on Post operative day 1to 20/25 on Post operative day 7.

On postoperative day 21, patient presentedwith sudden onset, rapidly progressive visual loss in both eyes(OU)over last 2 days.His visual acuitywas counting fingers at 3 feet in OU.He was still on oral prednisolone therapy with a dose of 10 mg/day. Dilated fundus examination showed clear optical media with bilateral gross serous elevation of macula (Figure 2a & b) and occasional cells in anterior vitreous face. Optical coherence tomography (StratusOCT, Carl ZEISS Meditech, Dublin, CA)revealed separation of inner neurosensory layer from outer hyper-reflective area (RPE layer) with accumulation of subretinal fluid along with exudation but there was no evidence of cystoid spaces in inner neurosensory layer (Figure 3a & b). Fundus fluorescein angiogram (figure 2c & d)demonstratedstippled hyperfluorescence in the posterior pole. Analyzing above features a diagnosis of sympathetic ophthalmia was made, however it was quite atypical owing to absence of keratic precipitates and anterior chamber reaction and posterior synechiae. The patient was prescribed higher dose of oral prednisolone (2mg/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 OU) (Figure 3c & d). On 15 days of increased steroid usage, his visual acuity was 20/20 in OU and there wascomplete resolution of macular elevation with restoration of normal foveal contour. (Figure 3e & f). Subsequent follow up for 6 months vision of the patient was well preserved and there was no evidence of recurrence of clinical signs of sympathetic ophthalmia.

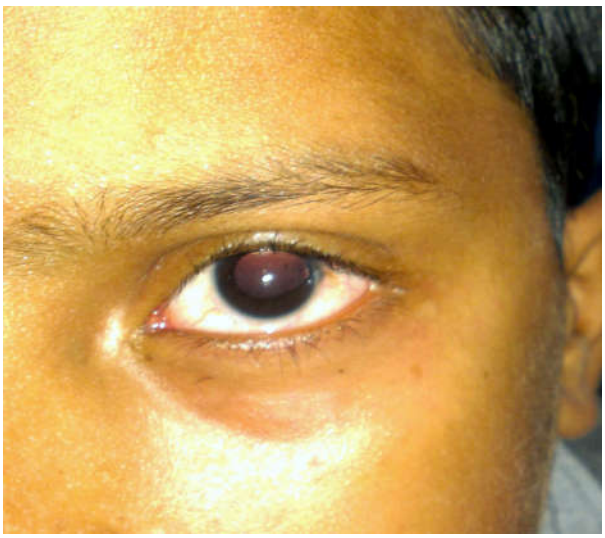

Fig. 1:

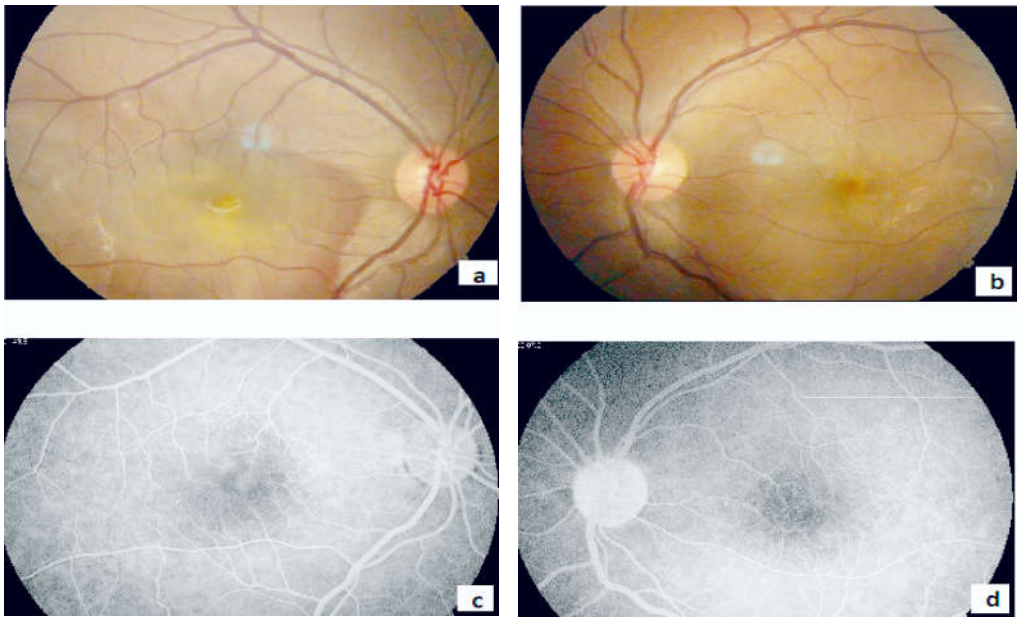

Fig. 1: A&B Colour fundus photographs exhibiting serous macular elevation. C&D Fundus fluorescein angiogram (FFA) photographs in peak arteriovenous filling exhibiting stippled hyperfluorescence in the posterior pole

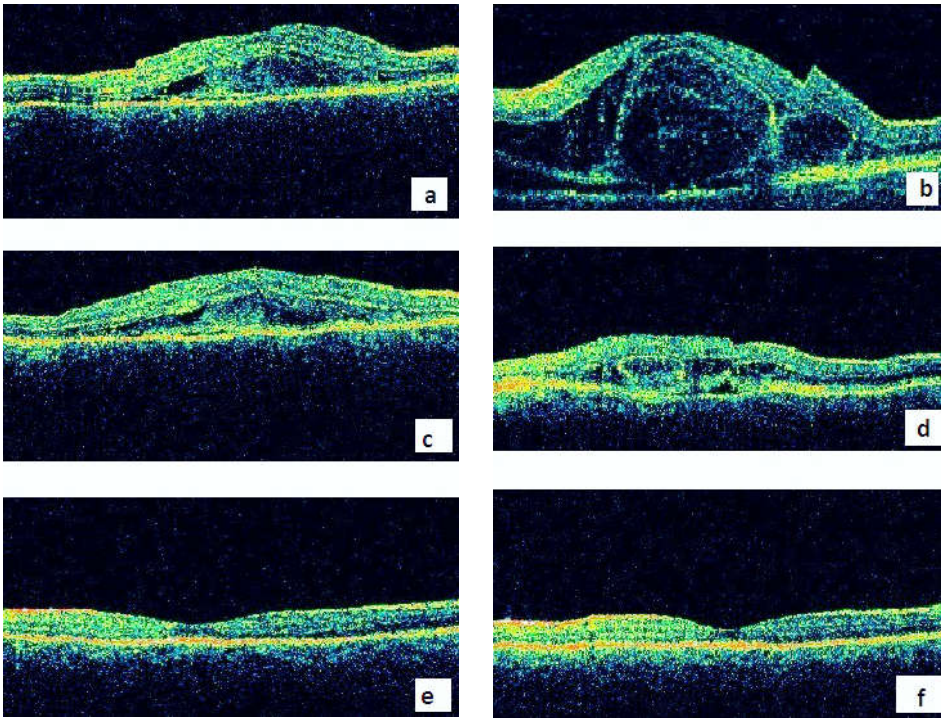

Fig. 3: *A&B:* Optical coherence tomography both eyes exhibiting exudative retinal detachment, *C&D:* showing gradual reduction of macular elevation, *E&F:* showing complete resolution of serous macular elevation with attainment of normal contour with 15 days of high corticosteroid therapy

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 optical coherence tomography 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 [18,19]. Onset of disease is within 1 year in 90% of cases and 17% present within 1 month [17,18]. Our case presented on 28th day of traumatic repair and 30th day of trauma. None of the anterior segment findings could be elucidated in our patient possibly attributed to prior steroid therapy.Kumar et al [20] showed 30% of isolated posterior segment findings in their case series on sympathetic ophthalmia. Gupta et al [21] demonstrated that 22 of their 40 cases presented with exudative retinal detachment with no evidence of anterior segment inflammation, leading to the conclusion that lone posterior segment findings may be an indicative of early diagnosis where anterior segment has not yet involved or it is an atypical presentation. Our case 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 prior immunosuppression in the immediate post operative period.

OCT is a useful noninvasive tool in the diagnosis and determining efficacy of treatment in sympathetic ophthalmia [22,23]. OCT demonstrates exudative retinal detachments and its reduction marks the response to treatment. Our patient too hadgradual reduction in exudative retinal separation in OCT following steroid therapy. Sympathetic ophthalmia is treated withimmunosuppressive therapy. Because of 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 phenomenoncan 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.

References

1. Kilmartin DJ, Dick AD, Forrester JV. Prospective surveillance of sympathetic ophthalmia in the United Kingdom and Republic of Ireland. Br J Ophthalmol. 2000; 84: 259–263.
2. Towler HMA, Lightman S. Sympathetic ophthalmia. Int Ophthalmol Clin. 1995; 35: 31–42.
3. Rao NA, Forster DJ, Spalton DJ. Sympathetic ophthalmia.In: Podos SM, Yanoff M (eds). The Uvea Uveitis and IntraocularNeoplasms, Vol. 2, Chapter 8. Mosby-Wolfe: USA. 1995; pp 8.10–8.13.
4. Nussenblatt RB. Sympathetic Ophthalmia. In: Nussenblatt RB, Whitcup SM (eds). Uveitis Fundamentals and ClinicalPractice, 3rd ed., Chapter 22. Elsevier: USA. 2004; pp 311–323.
5. Ahmad N, Soong TK, Salvi S, Rudle PA, Rennie IG. Sympathetic ophthalmia after ruthenium plaque brachytherapy. Br J Ophthalmol. 2007; 91: 399–401.
6. Buller AJ, Doris JP, Bonshek R, Brahma AK, Jones NP. Sympathetic ophthalmia following severe fungal keratitis. Eye. 2006; 20: 1306 –1307.
7. Jonas JB, Back W, Sauder G, Junemann U, Harder B, Spandau UH. Sympathetic ophthalmia in vater association combined with persisting hyperplastic primary vitreous after cyclodestructive procedure. Eur J Ophthalmol. 2006; 16: 171–172.
8. Chan CC, Benezra D, Rodrigues MM, Palestine AG, Hsu SM, Murphree AL, Nussenblatt RB. Immunohistochemistry and electron microscopy of choroidal infiltrates and Dalen-Fuchs nodules in sympathetic ophthalmia. *Ophthalmology*. 1985; 92: 580–90.
9. Jakobiec FA, Marboe CC, Knowles DM 2nd, Iwamoto T, HarrisonW, Chang S, Coleman DJ. Human sympathetic ophthalmia. An analysis of the inflammatory infiltrate by hybridoma-monoclonal antibodies, immunochemistry, and correlative electron microscopy. Ophthalmology. 1983; 90: 76–95.
10. Albert DM, Diaz-Rohena R. A historical review of sympathetic ophthalmia and its epidemiology. Surv Ophthalmol. Jul-Aug 1989; 34(1): 1–14.
11. Chan RV, Seiff BD, Lincoff HA, Coleman DJ. Rapid recovery of sympathetic ophthalmia with treatment augmented by intravitreal steroids. Retina. 2006 Feb; 26(2): 243-7.
12. Lubin JR, Albert DM, Weinstein M. Sixty-five years of sympathetic ophthalmia. A clinicopathologic review of 105 cases (1913–19788). *Ophthalmology*. Feb 1980; 87(2): 109–121.
13. Damico FM, Kiss S, Young LH. Sympathetic ophthalmia. Semin Ophthalmol. 2005; 20: 191–197.
14. David Fleischman, Emil Anthony T. Say, John D. Wright, and Maurice B. Landers Multimodality Diagnostic Imaging in a Case of Sympathetic Ophthalmia. August 2012; 20(4): 300-302.
15. Castiblanco, Claudia, Adelman, Ron A. Imaging for Sympathetic Ophthalmia: Impact on the Diagnosis and Management International Ophthalmology Clinics Issue: 2012 Fall; 52(4): 173–181.
16. Castiblanco CP, Adelman RA. Sympathetic ophthalmia. Graefes Arch Clin Exp Ophthalmol. 2009 Mar; 247(3): 289-302. doi: 10.1007/s00417-008-0939-8. Epub 2008 Sep 16.
17. Goto H, Rao NA. Sympathetic ophthalmia and Vogt-Koyanagi-Harada syndrome. Int Ophthalmol Clin. 1990 Fall; 30(4): 279-85.
18. Xi K Chu and Chi-Chao ChanSympathetic ophthalmia: to the twenty-first century and beyond J Ophthalmic Inflamm Infect. 2013; 3: 49.
19. Arevalo JF, Garcia RA, Al-Dhibi HA, Sanchez JG, Suarez-Tata L. Update on sympathetic ophthalmia. Middle East Afr J Ophthalmol. 2012 Jan; 19(1): 13-21.
20. Kumar K, Mathai A, Murthy SI, Jalali S, Sangwan V, Reddy Pappuru R, Pathangay A. Sympathetic ophthalmia in pediatric age group: clinical features and challenges in management in a tertiary center in southern India. Ocul ImmunolInflamm. 2014 Oct; 22(5): 367-72.
21. Gupta V, Gupta A, Dogra MR. Posterior sympathetic ophthalmia: a single centre long-term study of 40 patients from North India. Eye (Lond). 2008 Dec; 22(12): 1459-64.
22. Puliafito C. Acute sympathetic ophthalmia. In: Schuman, Puliafito, Fujimoto, eds. Optical Coherence Tomography of Ocular Diseases. 2nd ed. New York: Slack; 2003: 386–393
23. Gupta V, Gupta A, Dogra MR, Singh I. Reversible retinal changes in the acute stage of sympathetic ophthalmia seen on spectral domain optical coherence tomography. Int Ophthalmol. 2011 Apr; 31(2): 105-10.
24. Maruyama Y, Kishi S. Tomographic features of serous retinal detachment in Vogt- Koyanagi-Harada syndrome. Ophthalmic Surg Lasers Imaging 2004; 35: 239–242.