Case Report

# Central serous chorioretinopathy following an episode of angioedema: An interesting case report

**Anuradha Singh[1], Vikas Sharma[2]**

*From [1]Assistant Professor, Department of Ophthalmology, 167 Military Hospital, Pathankot, Punjab, India, [2]Assistant Professor, Department of Ophthalmology, AFCME, Subroto Park, New Delhi, India*

## ABSTRACT

Central serous chorioretinopathy (CSCR) is a common disease of the middle-aged population with largely unknown etiology. We report the case of CSCR in a 37-year-old patient following an episode of angioedema who presented with sudden diminution of vision in the left eye 2 days following an episode of angioedema, which had been managed with a short course of steroids and antihistamines. On evaluation, he was found to have subretinal fluid at the macula in the affected eye, suggestive of CSCR. The patient was conservatively managed. The patient had recovery of symptoms with conservative management over a long period. The association of angioedema and its treatment as an underlying cause of CSCR needs to be explored. To the best of our knowledge, this is the first such case to be reported worldwide.

**Key words:** Angioedema, Antihistamines, Central serous chorioretinopathy, Eplerenone, Steroids

Central serous chorioretinopathy (CSCR) is a common entity predominantly affecting the middle-aged population [1]. Clinically, it is an exudative collection between the retina and the retinal pigment epithelium (RPE)-Bruch's membrane complex, with a defect in the RPE leaking fluid into the subretinal space [2]. It typically presents as a sudden diminution of vision with a central scotoma, with distorted images. It is more common among males and those having a history of prolonged steroid intake, stress factors, or show traits of type A personality [3]. Most cases, however, are idiopathic [4]. The disease is self-resolving in a majority of cases, even after recurrence [5].

This case report discusses a patient who possibly developed unilateral CSCR after an attack of angioedema. This unusual etiological presentation of the condition generates interest in rare causes and pathophysiological mechanisms of CSCR. Informed consent has been obtained from the patient for publication of the case and relevant clinical photographs.

## CASE REPORT

A 37-year-old male patient presented to the Department of Internal Medicine of our zonal hospital with complaints of acute onset swelling of the face and neck region and difficulty in breathing, which started while the patient was tending to his garden (Fig. 1). He was managed with a single dose of intravenous corticosteroid and antihistamine followed by oral antihistamines for 2 days with which he had complete recovery of symptoms. Two days after the episode, he developed a sudden diminution of vision in the left eye and reported to the ophthalmology department. History revealed diminution of vision with a central scotoma, along with distortion of images, in the left eye.

General examination was within normal limits and vitals were stable. On ophthalmic evaluation, the patient was found to have a distant visual acuity of 6/6 in the right eye and 6/18(P) in the left eye by Snellen visual acuity chart, with no further improvement. Examination of the right eye revealed a normal anterior segment, while fundus examination revealed a cup-disc ratio (CDR) of 0.7:1 with a healthy neuroretinal rim. General fundus examination was otherwise normal in the right eye. Anterior segment evaluation of the left eye revealed a wedge-shaped cataract on the nasal aspect (Fig. 2a). On fundoscopy, the left eye showed a one disc diameter sized area of subretinal fluid (SRF) at the macula (Fig. 2b). The intraocular pressure was in the normal range in both eyes on repeated readings by Goldmann applanation tonometry.

Spectral-domain optical coherence tomography (OCT) macula showed a dome-shaped elevation of fovea along with hyporeflective shadow in the subretinal layers of the left eye, which suggested SRF (Fig. 3a). The central macular thickness

| Access this article online | |
|---|---|
| Received - 05 April 2021<br>Initial Review - 21 April 2021<br>Accepted - 07 July 2021 | Quick Response code |
| **DOI:** 10.32677/IJCR.2021.v07.i07.014 | |

**Correspondence to:** Dr. Anuradha Singh, Department of Ophthalmology, 167 Military Hospital, Dhangu Road, Pathankot - 145 001, Punjab, India. E-mail- anuradha9000@gmail.com

was 629 μ in the left eye. The findings of the right eye were within normal limits. Fundus fluorescence angiography showed the pooling of dye in an ink-blot pattern in the left eye (Fig. 4), while the right eye did not show any abnormality. Glaucoma workup done in view of raised CDR did not reveal any abnormality and was regarded as physiological disc cupping. The wedge-shaped cataract in the left eye was regarded as an incidental finding.

**Figure 1: Photograph of the patient showing angioedema**

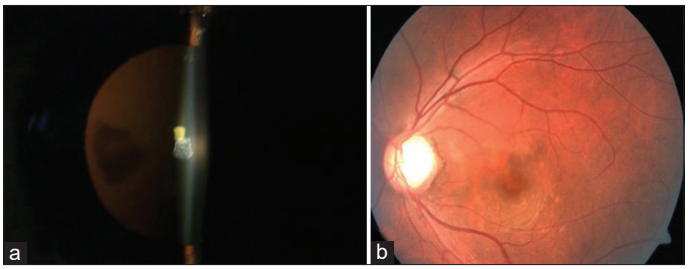


**Figure 2: (a) Slit-lamp photograph showing wedge-shaped cataract in the left eye; (b) fundus photograph of the left eye showing subretinal fluid at macula**

Based on the clinical and investigative findings, the patient was diagnosed as a case of CSCR of the left eye. Since this was the first episode of CSCR in this patient, the patient was observed for spontaneous resolution of SRF for 3 months, failing which, he was started on oral eplerenone 50 mg once daily. Over the next few months, there was a gradual reduction of SRF with an improvement of vision to 6/6 (P) with some residual metamorphopsia. OCT macula now showed signs of chronicity in the form of small discreet hyper-reflective shadows in the subretinal space followed by complete resorption of SRF and foveal thinning (Fig. 3b). The patient has been weaned off tablet eplerenone and has not reported a recurrence of symptoms.

## DISCUSSION

The RPE is the outermost layer of the retina which plays an important part in maintaining it in a dry state [6]. Therefore, any defect in the anatomical or physiological integrity of this layer can cause an inflow of fluid from the choroid into the subretinal space. Such an exudative retinal detachment leads to CSCR [7]. Many cases are idiopathic, while other risk factors are type A personality, chronically raised levels of steroids either endogenous or exogenous, systemic medications such as antibiotics, antihistamines and proton-pump inhibitors, gastrointestinal reflux disease, *Helicobacter pylori* infection, pregnancy, allergic respiratory condition, and uncontrolled hypertension [8]. The patients commonly present with sudden

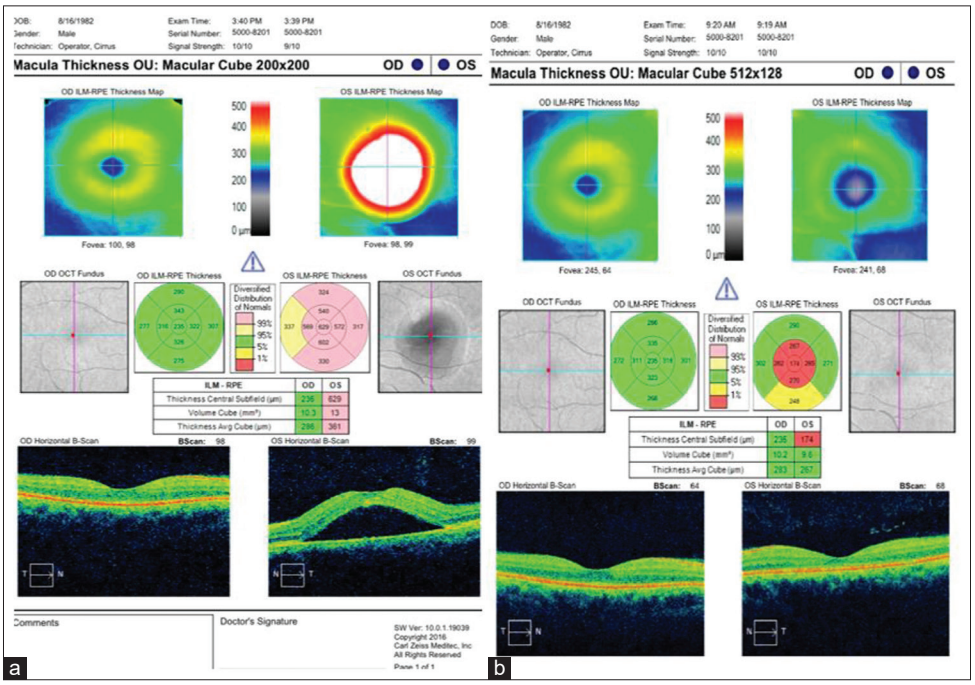


**Figure 3: Optical coherence tomography macula (a) at presentation showing dome shaped elevation of fovea with subretinal fluid; (b) resorption of fluid and foveal thinning in the left eye**

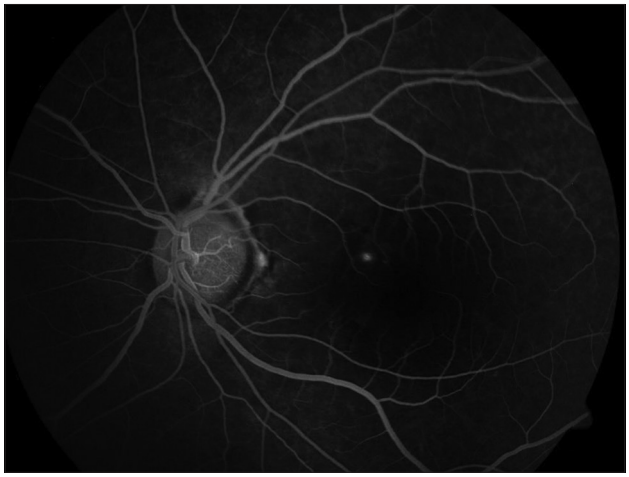

**Figure 4: Ink-blot pattern of fluorescein leak in the left eye**

painless diminution of vision in the form of a central scotoma, micropsia, and metamorphopsia, and a hyperopic change in their refraction [9].

The first line of treatment for CSCR remains conservative with observation and risk factor modification, as it is essentially a self-limiting disease [10]. Indications for intervention include persisting SRF after 3 months of observation; early treatment can be undertaken if the fellow eye has poor vision or there is a need for early rehabilitation due to occupational requirements [11]. Treatment modalities include conventional laser photocoagulation, micropulse diode laser photocoagulation, photodynamic therapy, transpupillary thermotherapy, and medical management with agents such as oral anti-steroid drugs (e.g., eplerenone), anti-*H. pylori* treatment, carbonic anhydrase inhibitors, and intravitreal anti-vascular endothelial growth factor injection [12].

This case was unique because of the mode of onset of CSCR, which was following an acute allergic reaction. A thorough literature search has not revealed any similar cases reported in the past. A case report published by Edalati *et al*. in 2009, described a case of CSCR in a patient with idiopathic non-histaminergic angioedema who was on tapering doses of oral steroids [13]. However, our patient had no history of chronic steroid intake and demonstrated a chronic course of CSCR rather than recurrence. Another case series of two patients who developed cataracts with antihistamine therapy was published by Alam *et al.* in 2019, though a causal relationship of wedge-shaped cataract seen in our patient could not be proved, as he did not have any history of chronic allergic condition or long-term antihistamine intake [14].

## CONCLUSION

CSCR is a common yet mysterious condition whose etiopathogenesis has been under research for decades. This case report indicates that the role of causative agents such as angioedema, short-term corticosteroid, and antihistamine therapy needs to be explored. To the best of our knowledge, this is the first case of CSCR following angioedema reported to date.

## REFERENCES


1. Tsai DC, Chen SJ, Huang CC, Chou P, Chung CM, Huang PH, *et al*. Epidemiology of idiopathic central serous chorioretinopathy in Taiwan, 2001-2006: A population-based study. PLoS One 2013;8:e66858.
2. Wang M, Munch IC, Hasler PW, Prünte C, Larsen M. Central serous chorioretinopathy. Acta Ophthalmol 2008;86:126-45.
3. Daruich A, Matet A, Dirani A, Bousquet E, Zhao M, Farman N, *et al*. Central serous chorioretinopathy: Recent findings and new physiopathology hypothesis. Prog Retin Eye Res 2015;48:82-118.
4. Vilela M, Mengue C. Central serous chorioretinopathy classification. Pharmaceuticals (Basel) 2021;14:26.
5. Mudvari SS, Goff MJ, Fu AD, McDonald HR, Johnson RN, Ai E, *et al*. The natural history of pigment epithelial detachment associated with central serous chorioretinopathy. Retina 2007;27:1168-73.
6. Gemenetzi M, De Salvo G, Lotery AJ. Central serous chorioretinopathy: An update on pathogenesis and treatment. Eye 2010;24:1743-56.
7. Semeraro F, Morescalchi F, Russo A, Gambicorti E, Pilotto A, Parmeggiani F, *et al*. Central serous chorioretinopathy: Pathogenesis and management. Clin Ophthalmol 2019;13:2341-52.
8. Haimovici R, Koh S, Gagnon DR, Lehrfeld T, Wellik S; Central Serous Chorioretinopathy Case-Control Study Group. Risk factors for central serous chorioretinopathy: A case-control study. Ophthalmology 2004;111:244-9.
9. Salmon JF. Kanski's Clinical Ophthalmology: A Systematic Approach. 9th ed. Amsterdam, Netherlands: Elsevier; 2019.
10. Liegl R, Ulbig MW. Central serous chorioretinopathy. Ophthalmologica 2014;232:65-76.
11. Nicholson B, Noble J, Forooghian F, Meyerle C. Central serous chorioretinopathy: Update on pathophysiology and treatment. Surv Ophthalmol 2013;58:103-26.
12. Manayath GJ, Ranjan R, Karandikar SS, Shah VS, Saravanan VR, Narendran V. Central serous chorioretinopathy: Current update on management. Oman J Ophthalmol 2018;11:200-6.
13. Edalati K, Roesch MT, Buchanan ML, Teeter M, Maberley DA. Central serous chorioretinopathy and idiopathic nonhistaminergic angioedema. Can J Ophthalmol 2009;44:606-76.
14. Alam MS, Somarajan AC. Presumed antihistamine-induced cataract in two young males. J Pharmacol Pharmacother 2019;10:75-6.


*Funding: None; Conflicts of Interest: None Stated.*

**How to cite this article:** Singh A, Sharma V. Central serous chorioretinopathy following an episode of angioedema: An interesting case report. Indian J Case Reports. 2021;7(7):310-312.