• Letter to the Editor •

Optic perineuritis as an initial presentation of ocular toxoplasmosis: a case report

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Received: 2021-08-24 Accepted: 2022-07-14

DOI:10.18240/ijo.2022.12.25

Citation: Chung SA, Kim C, Lim JY, Chung YR. Optic perineuritis as an initial presentation of ocular toxoplasmosis: a case report. *Int J Ophthalmol* 2022;15(12):2043-2046

Dear Editor,

Ocular toxoplasmosis, caused by *Toxoplasma gondii* infection, is one of the most common causes of posterior uveitis worldwide[1]. It typically manifests as white focal retinitis with overlying vitreous inflammation that results in a chorioretinal scar and associated scotoma[2]. However, the diagnosis of ocular toxoplasmosis can be challenging when it presents as an isolated papillitis without other typical signs of retinochoroiditis or vitritis[2]. Although optic nerve involvement in ocular toxoplasmosis was present in 5.3% of cases, isolated papillitis was extremely rare (3 eyes out of 926 patients) and was only considered a presumed diagnosis in such cases given the presence of old toxoplasmic retinochoroiditis lesions[3]. Few reports have described optic nerve involvement preceding toxoplasmic retinochoroiditis, while perineuritis, an uncommon form of orbital inflammatory disease involving the optic nerve sheath, has not been reported at all[4-5]. Herein, we describe a case of ocular toxoplasmosis that initially presented with optic perineuritis followed by typical retinochoroidal inflammation. To our knowledge, this is the first case report of ocular toxoplasmosis with perineuritis in an immunocompetent individual.

Ethical Approval The study was conducted in accordance with the Declaration of Helsinki. Informed consent was waived by the Institutional Review Board of Ajou University Hospital, Suwon, Korea (AJOUIRB-EX-2022-450).

Case Report A 56-year-old woman presented with a 1-day history of ocular pain and sudden diminution of the superior visual field in the left eye. Her corrected visual acuity was 20/20 in both eyes with a myopic correction of -2.50 diopters in the left eye. She had an unremarkable slit-lamp examination, pupils reactive to light with no relative afferent pupillary defect, and normal color vision in both eyes. Fundus examination revealed swelling and hemorrhage in the inferior sector of the left optic disc (Figure 1A) that correlated with a superior arcuate visual field defect (Figure 1B). Slit-lamp and fundus examination of the right eye were unremarkable. Magnetic resonance imaging (MRI) of the brain and orbits revealed wall thickening and enhanced signal intensity along the left optic nerve sheath that was suggestive of optic perineuritis (Figure 1C). The majority of the laboratory workups, which included negative serologies for viral markers, anti-aquaporin 4 antibody, and anti-myelin oligodendrocyte glycoprotein antibody, were noncontributory. The rapid plasma reagin test for syphilis and the polymerase chain reaction test for COVID-19 were also negative. The patient was started on a 3-day course of intravenous methylprednisolone (1 g/d) and experienced prompt resolution of the ocular pain; however, she did not show any improvement in the visual field defect, even after completing the steroid treatment.

A month after the initial presentation, she complained of worsening of vision and aggravated visual field defects in her left eye. The corrected visual acuity of the left eye deteriorated to 20/100. Slit-lamp examination revealed fine keratic precipitates and 2+ cells in the anterior chamber. A white focal retinal exudate with hemorrhage was noted in the inferior juxtapapillary area adjacent to the previous peripapillary lesion, and there was extension of the whitish retinal edema inferior to the fovea (Figure 2A). Fluorescein angiography showed blocked fluorescence at the retinitis lesion and delayed filling of the retinal arteries (Figure 2B). Optical coherence tomography (OCT) revealed an inner retinal edema with vitreal inflammation (Figure 2C). With the suspicion of ocular toxoplasmosis and combined branch retinal artery occlusion (BRAO), extensive laboratory workup was performed, which revealed positive serological results for anti-toxoplasma immunoglobulin (Ig)M and IgG and unremarkable findings in the other tests.

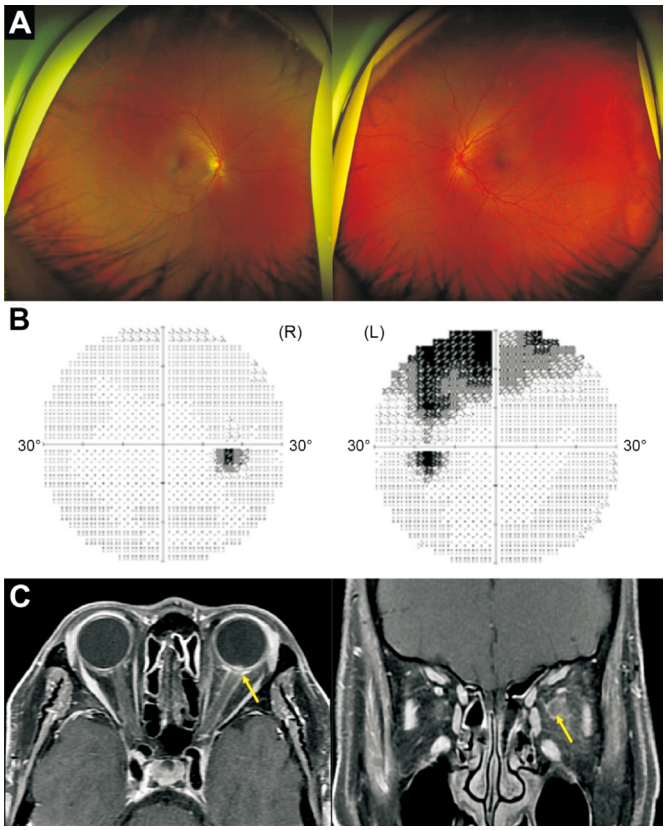

Figure 1 Ocular findings at initial presentation A: Wide-view fundus photographs showing a sectoral swelling in the inferior part of the left optic disc without any pre-existing retinochoroiditis scars; B: Humphrey perimetry showing superior arcuate field defect corresponding to the inferior lesion of the left optic disc; C: Axial and coronal views of contrast-enhanced, T1-weighted magnetic resonance imaging of the orbits demonstrating signal intensity along the left optic nerve sheath (yellow arrows), suggesting perineuritis.

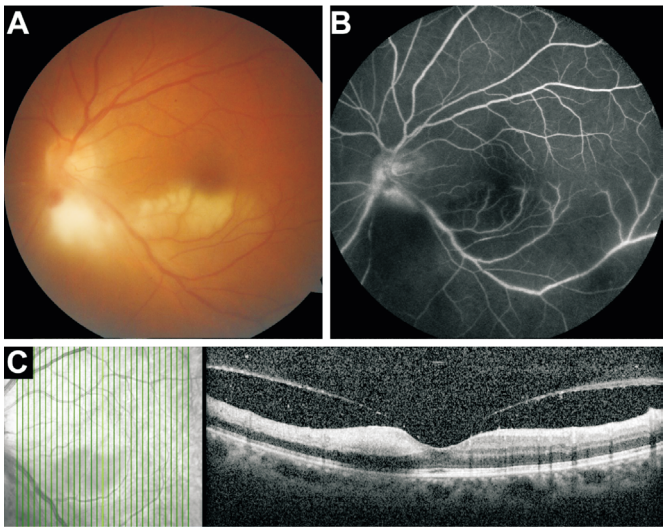

Figure 2 Fundus findings of the left eye 4wk after initial presentation A: Fundus photograph showing a whitish juxtapapillary lesion with hemorrhage inferior to the optic disc and whitish retinal edema inferior to the fovea; B: Fluorescein angiography showing blocked fluorescence inferior to the optic disc and leakage from vessels inferior to the fovea; C: Optical coherence tomography showing inner retinal edema inferior to the fovea and vitreal inflammation.

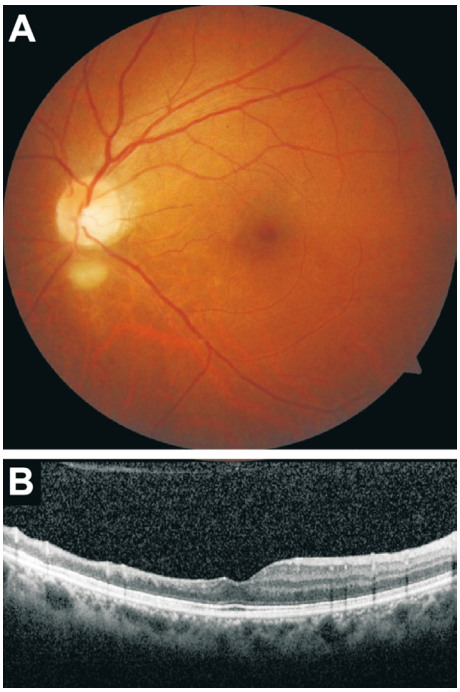

Figure 3 Fundus findings of the left eye at 5-month follow-up A: Fundus photograph with attenuation of inferior branch retinal vessels, inferior optic disc pallor, and a juxtapapillary whitish retinochoroidal scar; B: Optical coherence tomography showing thinning of the inner retinal layers with intact outer layers in the area with previous branch retinal artery occlusion.

The patient was started on anti-toxoplasmic antibiotics (sulfamethoxazole/trimethoprim and clindamycin) and oral prednisone. The retinal exudates and retinal edema started to improve a week after the initiation of anti-toxoplasmic medication, and the patient was able to complete a 6-week course of steroid tapering. At 5mo follow-up, the corrected visual acuity in the left eye was restored to 20/40. There was no recurrence but a white focal retinochoroidal scar on fundoscopy (Figure 3A) and inner retinal thinning at the previous BRAO lesion on OCT (Figure 3B) were noted.

Atypical presentations of ocular toxoplasmosis have been reported in the literature, such as optic neuropathy, neovascularization, retinal vascular occlusion, epiretinal membranes, and retinal detachment[2]. Moreover, BRAO has been reported to occur in 7% of ocular toxoplasmosis cases at the site of an active chorioretinitis lesion overlying or adjacent to the retinal vessels due to the direct compression of the artery by the inflammatory focus, vasoconstriction, and increased blood viscosity[6-7]. The low prevalence of optic nerve involvement in ocular toxoplasmosis, such as pure papillitis without concomitant chorioretinal inflammation, contributes to diagnostic challenges[2-4,8]. As summarized in Table 1[5,9-12], a few cases presenting with optic nerve lesions were diagnosed later as ocular toxoplasmosis based on subsequent typical retinochoroidal inflammation several weeks after optic nerve

Table 1 Summary of cases presenting optic nerve involvement in ocular toxoplasmosis

Authors	Sex/age	Medical history	Optic disc findings	Other findings
Mikhail and Varijjara[5]	M/14	None	Fluffy white lesion on inferotemporal optic disc margin	Neurosensory detachment with vitritis after 1wk. Serum IgG positive.
Song *et al*[9]	M/33	None	Optic disc edema, retinal hemorrhage, and vitreous opacity	Granuloma after 9d. Increased IgG titer.
Wong *et al*[10]	F/25	None	Optic disc swelling, non-granulomatous anterior uveitis, vitritis, and concomitant branch retinal vein occlusion	Neuroretinitis after 1wk, focal retinitis after 2wk. Serum IgG positive.
Lee *et al*[11]	M/46	Later AIDS	Optic disc swelling, cotton wool spots, flame-shaped retinal hemorrhages	Enhancement of optic nerve, extraocular muscles, and orbital flat on MRI. AIDS diagnosed during evaluation.
Lee *et al*[12]	F/53	None	Optic disc swelling with focal edema and hard exudates in nasal parapapillary retina	Serum IgM and IgG positive.

MRI: Magnetic resonance imaging; IgG: Immunoglobulin G; IgM: Immunoglobulin M.

involvement[5,9-10]. Consequently, some of these patients were started on an intravenous steroid regimen with a presumed diagnosis of optic neuritis and subsequently treated with anti-toxoplasmic medications after typical retinochoroidal lesions appeared[5,10].

Optic perineuritis, an uncommon inflammatory disorder, is generally considered an orbital inflammation[13-14]. MRI of the orbits may demonstrate circumferential enhancement along the optic nerve, occasionally accompanied by intraorbital inflammation[13]. Patients with perineuritis may complain of visual field defects with intact central vision[13]. A case of toxoplasmosis involving the optic nerve and orbital tissue has been reported as the initial manifestation of undiagnosed HIV infection[11], while ours was a case of perineuritis in an immunocompetent individual. A positive toxoplasmosis serology can provide additional clues for diagnosis in cases of isolated optic nerve involvement[12]. Serum anti-toxoplasma IgM levels can increase, and its titer can rise within 1-2wk of infection and become undetectable after 6-9mo[1]. We considered that optic perineuritis in our patient was the initial manifestation of ocular toxoplasmosis along with consecutive retinochoroidal inflammation and positive IgM serology with a 1-month interval. However, serology was not observed at the time of optic perineuritis. Our patient might have had a better prognosis if she were treated with anti-toxoplasmic antibiotics before chorioretinal lesions and BRAO developed. Steroid pulse therapy might have even aggravated the toxoplasmosis in this immunocompetent patient since the use of systemic steroids without anti-toxoplasmic antibiotics has been identified as a risk factor for recurrent ocular toxoplasmosis[15]. Cases of fulminant chorioretinitis in ocular toxoplasmosis have also been reported following the use of corticosteroids with or without antibiotics[16-17]. We need to suspect atypical ocular toxoplasmosis presenting with optic perineuritis in an immunocompetent patient and remind ourselves that the seropositivity for *Toxoplasma gondii* plays a role in supporting the diagnosis in this situation[1].

In summary, we report the first case of optic perineuritis that is thought to be attributed to ocular toxoplasmosis, given the subsequent typical retinochoroidal inflammation and complicated BRAO. Anti-toxoplasmic serology tests can be helpful, especially in cases with atypical presentations including optic perineuritis.

ACKNOWLEDGEMENTS

This case was presented as a free paper at the 21st Symposium of the Korean Uveitis Society in Seoul, Korea, May 15, 2021. We would like to thank Editage (www.editage.co.kr) for English language editing.

Conflicts of Interest: Chung SA, None; **Kim C,** None; **Lim JY,** None; **Chung YR,** None.

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