Ann Med Res

Current issue list available at AnnMedRes

Annals of Medical Research

journal page: www.annalsmedres.org

Papillitis in acute posterior multifocal placoid pigment epitheliopathy under oral steroid treatment

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ARTICLE INFO

Keywords:
Acute posterior multifocal placoid pigment epitheliopathy
Papillitis
Steroid

Received: Nov 04, 2024
Accepted: Nov 19, 2024
Available Online: 29.11.2024

DOI:
10.5455/annalsmedres.2024.11.235

Abstract

A 54-year-old woman presented with complaints of worsening vision in left eye. Funduscopic examination showed yellowish creamy placoid like lesions superotemporal to the optic nerve in the left eye with no evidence of disk edema and vitritis. She was diagnosed with APMPPE and after ruling out infectious causes started on a 1 mg/kg/day oral prednisone. 1 month later, she presented to with an acute worsening of vision in left eye. Her visual acuity was hand motion in the left eye. Funduscopy showed marked inflammatory lesions and papillitis in the left eye. OCT showed subretinal fluid and PED in the left eye. Oral prednisone was tapered gradually under close follow-up. 3 months later her visual acuity increased 20/200 and papillitis was relieved. This is the first reported case of APMPPE that evolving papillitis under treatment, and after cessation of treatment, improved.

Introduction

Acute posterior multifocal placoid pigment epitheliopathy (APMPPE) is related with a number of infectious (Lyme disease, Pulmonary tuberculosis, Mumps, antihepatitis B vaccination) and noninfectious systemic conditions (Sarcoidosis, Wegener's granulomatosis, ulcerative colitis, necrotizing vasculitis) [1]. Placoid lesions are the main feature of the retinal involvement. These yellowish white lesions can be varying size and multiple, arise from at the retinal pigment epithelium (RPE)/choriocapillaris level. The lesions vanish in weeks and after that RPE atrophy and hypo/hyperpigmentation occurs [2]. Fluorescein angiography (FFA) show early hypofluorescence followed by late hyperfluorescence in placoid lesions [3].

APMPPE has such atypical findings; exudative neurosensory retinal detachment, central retinal vein occlusion, periphlebitis, and papillitis [1]. About %35 percent of patients suffer from a viral flu-like prodromal phase and that usually precede visual symptoms [4].

Here we present a unilateral APMPPE case that developed transient papillitis under oral steroid treatment.

Case Report

A 54 year old woman patient, who is white descend, was suffering blurring vision in her left eye was seen in outpatient clinic. We have an informed consent form signed on 06/05/2020 by the patient for this case presentation. Visual acuity was 20/32 and biomicroscopic examination showed unremarkable findings. Funduscopic examination showed yellowish creamy placoid like lesions superotemporal to the optic nerve with no evidence of disk edema and vitritis in her left eye (Figure 1a). Spectral domain optical coherence tomography (SD-OCT) showed disorganization and disappearance of outer retina, partially disappeared atrophic RPE and partially thickened RPE hyperreflectivity (Figure 1b). FFA showed corresponding late staining of these placoid like lesions, and the focal atrophic chorioretinal lesions inferotemporal to macula. These atrophic lesions were surrounded by hyperfluorescence and the centers were like hypofluorescent scar (Figure 1c and 1d). Visual acuity was 20/20 and all biomicroscopic, fundus, FFA and OCT examinations were normal in her right eye.

She was diagnosed with APMPPE by initial appearance of the lesions, OCT and the FFA findings in the left eye and after ruling out infectious causes started on a 1 mg/kg/day oral prednisone. The patient was followed up

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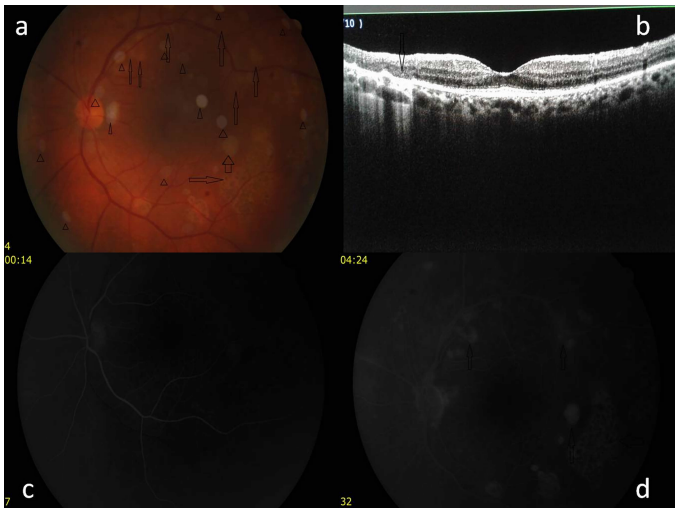

Figure 1. a) First presentation of the patients left eye. New placoid like lesions (vertical arrows), old atrophic chorioretinal lesions (Horizontal arrows), artefact (arrow heads). b) OCT shows disruption with partial disappearance of the elipsoid zone, and thickening with an irregularly thickened accentuated RPE hyperreflectivity.c) Early phase of FFA. d) Late phase of FFA. New placoid like lesions (vertical arrows).

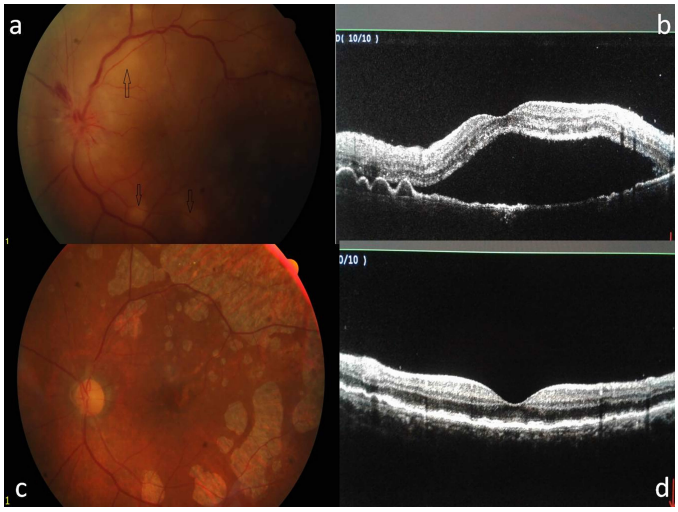

Figure 2. a) Marked inflammatory placoid lesions and papillitis development in the left eye under 1 month oral steroid treatment. b) OCT shows subretinal fluid and PED under 1 month oral steroid treatment. c) The color image of the patient 3 month later. No papillitis, and resolution of placoid lesions with gradual depigmentation. d) OCT images of the patient 3 month later.

weekly. There were not any changes in her eye examinations in that period.

1 month later, she was seen in emergency room with an acute decrease in left eye vision and had flu-like symptoms. At that time, her visual acuity was hand motion, biomicroscopic examination was normal. Funduscopic examination showed marked inflammatory lesions and papillitis in the left eye (Figure 2a). SD-OCT showed subretinal fluid and pigment epithelium detachment (PED) in the left eye (Figure 2b). Right eye vision and other examinations were normal.

Even tough erythrocyte sedimentation rate (ESR) increased to 56 mm/h and C-reactive protein (CRP) increased to 0.58 mg/dL, oral prednisone was tapered gradually under close follow-up. And she referred to pulmonary medicine because of cough and fever. She was diagnosed with flu and treated with conservative treatment.

3 months later her visual acuity increased 20/200 and papillitis was relieved. Her's imaging of retina demonstrated resolution of white placoid lesions with gradual depigmentation over a few months (Figure 2c). SD-OCT showed no subretinal fluid and accentuated RPE hyperreflectivity in the left eye (Figure 2d). At that time, ESR and CRP was within normal limits. On final examination, 1 year after presentation, best-corrected visual acuities were 20/200 on the left eye and other examinations and images were same as 3^{th} months.

Discussion

APMPPE typically affects the outer retina, choriocapillaris, and RPE [5]. Inflammatory lesions that correspond to acute RPE thickenning on SD-OCT are often associated with a disruption of IS/OS junction, disruption of the external limiting membrane (ELM) and photoreceptor layers [6].

APMPPE typically occurs in younger age groups and preceeding flu-like symptoms can associate approximately in one third of cases [1,4]. Our case was older age and flu symptoms started after 1 month oral steroid treatment. Altough APMPPE usuallay seen bilateral it can presented as unilateral rarely [4]. Our case was unilateral.

Papillitis and foveal subretinal fluid developed after 1 month oral steroid treatment. There are some report in the literature that APMPPE cases with unilateral involvement, retinal detachment and papillitis [4,7,8]. Our case presented these rare findings under oral steroid treatment. It can be explained by more severe recurrent attack under oral steroid as like optic neuritis [9]. In this respect, it differs from the literature about APMPPE.

In the APMPPE, the sensory retina overlying placoid lesions may detach, that like those seen in Harada disease [8]. The clinic, FFA and OCT characteristics of our patient did not match with those of VKH patients [10]. In addition the FFA properties of our case didn't match with central serous retinopathy features. An explanation for retinal and pigment epithelial detachment was that underliying choroidal inflamation might have broken the fluid exchange between outer retina and choroidal vascular system which was observed by OCT [11]. Moreover, in our patient, OCT of the left eye showed PED after anti-inflammatory therapy initiation. We couldn't perform indocyanine green angiography because of technical impossibilities.

There are no precise consensus about treatment of APMPPE but some authors have recommended the use of corticosteroids when serous detachment and papillitis are seen [8]. Oral corticosteroid treatment is the most common

option but sometimes before oral treatment iv methylprednisolone pulses are recommended. In our case, we started oral steroid treatment in the absence of these findings. However, these findings emerged while under treatment, and after cessation of treatment, improved.

We hypothesise that APMPPE associated papillitis seem to resembling arteritic ischemic optic neuropathy. Treatment of arteritic ischemic optic neuropathy is challenging about IV or oral steroid therapy.

Conclusion

To date, there has not been any report that steroid therapy worsening APMPPE patients. We assume that oral steroid therapy in our case induced papillitis because of upregülated inflammation due to low dose steroids. We didnt consider to give higher dose oral steroids or immunomodulatory prescriptions because of her systemic condition. It could be more effective, if we started high dose iv steroid pulses and then continued with oral steroid.

Financial supporting

No financial resources are used for this article.

Conflict of interest

There is no conflict of interest regarding this article.

References

1. Goldenberg D, Habot-Wilner Z, Loewenstein A, Goldstein M. Spectral domain optical coherence tomography classification of acute posterior multifocal placoid pigment epitheliopathy. Retina. 2012;32(7):1403-1410.
2. Kim DD, Ghorayeb G. Acute Posterior Multifocal Placoid Pigment Epitheliopathy Associated with Gastrointestinal Stromal Tumor and Hurthle Cell Tumor. Case Rep Ophthalmol Med. 2018 Sep 30;2018:1656131. doi: 10.1155/2018/1656131.
3. Pagliarini S, Piguet B, Ffytche TJ, Bird AC. Foveal involvement and lack of visual recovery in APMPPE associated with uncommon features. Eye (Lond). 1995;9: 42-47.
4. Khan HA, Shahzad MA. Atypical Acute Posterior Multifocal Placoid Pigment Epitheliopathy. Optom Vis Sci. 2016;93(1):103-106.
5. Hong IH, Park SP, Chen CL, et al. Cone photoreceptor abnormalities correlate with vision loss in a case of acute posterior multifocal placoid pigment epitheliopathy. Ophthalmic Surg Lasers Imaging Retina. 2014;45(1):74-78.
6. Kaplan AJ, Koushan K, Martin J. Novel optical coherence tomography description of acute posterior multifocal placoid pigment epitheliopathy. Can J Ophthalmol. 2013 Jun;48(3):e53-5. doi: 10.1016/j.jcjo.2012.12.012.
7. Vedantham V,Ramasamy K. Atypical manifestations of acute posterior multifocal placoid pigment epitheliopathy. Indian J Ophthalmol 2006;54: 49-52.
8. Tanigawa M, Tsukahara Y, Yamanaka H. A case of acute posterior multifocal placoid pigment epitheliopathy demonstrating Vogt-Koyanagi-Harada disease-like optical coherence tomography findings in the acute stage. Case Rep Ophthalmol 2013;4: 172-179.
9. Menon V, Saxena R, Misra R, Phuljhele S. Management of optic neuritis. Indian J Ophthalmol. 201; 59(2):117-122.
10. Ceylan OM, Uysal Y, Türk A, et al. Clinical Features of Cases of Vogt-Koyanagi-Harada Syndrome - Original Article. Turk J Ophthalmol 2010;40:250-254.
11. Lim LL, Watzke RC, Lauer AK, Smith JR. Ocular coherence tomography in acute posterior multifocal placoid pigment epitheliopathy. Clin Experiment Ophthalmol 2006;34: 810–812.