A rare ocular manifestation of Chikungunya – retinal vasculitis and cystoid macular oedema: a case report

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SUMMARY

Chikungunya, a mosquito-borne disease caused by the arbovirus chikungunya virus, is endemic in Malaysia. It is known for causing sudden fever and severe arthralgia. The most typical eye symptom associated with this disease is anterior uveitis. Additional ocular symptoms that may occur include conjunctival injection, episcleritis, scleritis, uveitis accompanied by glaucoma, isolated chorioretinitis, neuroretinitis and oculomotor nerve palsies. However, we describe a case in which a patient did not exhibit any signs of anterior chamber activity or those mentioned above. Instead, the patient had retinal vasculitis and cystoid macular oedema (CMO) after a quiescent interval following systemic infection. This highlights that viral uveitis can have a myriad of presentations. No dedicated antiviral treatment exists for chikungunya infection and randomised controlled trials have not been conducted to assess the specific treatment of ocular inflammatory conditions related to chikungunya.

INTRODUCTION

Chikungunya is a mosquito-borne viral disease that has been reported in Malaysia. It is caused by the Chikungunya virus – an arbovirus, primarily transmitted to humans through the bite of infected Aedes mosquitoes, particularly Aedes aegypti and Aedes albopictus.[1] During the initial phase of the illness (within 3 weeks after infection), patients experience a variety of non-specific symptoms including high fever (greater than 39°C), headache, fatigue, rash, muscle aches and joint pain. Among these, the most common and often incapacitating symptom is the intense swelling and discomfort in the joints.[1] The post-acute stage (extending from the third week to as long as three months after infection) is marked by the disappearance of the symptoms experienced during the acute phase, except for the persistent polyarthritis, which is frequently characterised by joint stiffness, pain and swelling.[1] Ocular manifestations do occur and have become more prevalent recently.[2] In Malaysia, chikungunya infection was first recorded in Port Klang in Year 1998-1999. Subsequently, outbreaks and re-emergence of Chikungunya infection occurred periodically in 2006-2009 throughout the states in Peninsular Malaysia.[3] Since year 2004, chikungunya has spread rapidly and has been identified in more than 60 countries throughout Asia, Africa, Europe and the Americas.[4] We report a rare case of retinal vasculitis and cystoid macular oedema (CMO) that happened at the post-acute stage i.e. after a quiescent interval following systemic infection.

CASE PRESENTATION

A 61-year-old Malay woman with no known medical illness presented with progressive bilateral painless blurring of vision for 3 weeks. It is associated with metamorphopsia. There was no associated eye redness or eye discharge during this episode. No visual field defect was reported. Systemic and ocular history are unremarkable other than a recent hospital admission for Chikungunya infection 6 weeks prior to her presentation, which was confirmed via real time PCR. During the hospital stay, her clinical course was stable with no haemodynamic compromise and no organ failure. She reported 3 days of bilateral eye mild redness and tearing which resolved spontaneously without any treatment during her hospital stay.

Her presenting vision was 6/18 OD and 6/24 OS. No relative afferent pupillary defect was detected. Slit lamp examination of the anterior segment was unremarkable. No anterior chamber activity and no anterior vitreous cells were detected. There was early nuclear sclerosis in both the eyes and normal intraocular pressure for both the eyes: 15 mmHg OD and 16 mmHg OS.

Posterior segment examination in both the eyes revealed perivascular sheathing, intraretinal haemorrhages, dull foveal light reflex and cystoid macula oedema (Figure 1). There was no optic disc swelling or vitritis, no retinitis or choroiditis seen. Macula oedema in both the eyes were further confirmed by SD-OCT findings (Figure 2).

A full blood panel including full blood count, renal profile, serum calcium and albumin levels, erythrocyte sedimentation rate, C-reactive protein were within normal range. Infectious work-up e.g., syphilis, HIV, hepatitis B were negative. Toxoplasma IgM was negative while IgG serology was positive. Mantoux test was negative, measured induration was 6 mm. Chest X-ray did not reveal any lung parenchymal or hilar abnormalities. Urinalysis was normal except for few white cells seen.

Fundus fluorescein angiography (FFA) was done. Arteriovenous transit time was normal at 12 seconds. No findings of hot disc. There was focal vascular leakage in the infero-temporal macula denoting vasculitis (Figure 2).

A provisional diagnosis of retinal vasculitis and cystoid macular oedema secondary to Chikungunya infection was made after ruling out other possible causes. Treatment option of anti-vascular endothelial growth factor or trial of topical

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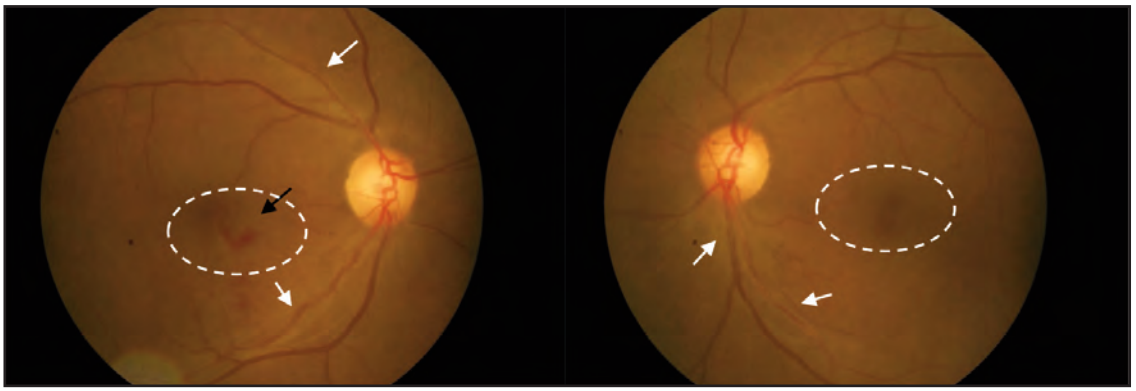

Fig. 1: Fundus photo showing perivascular sheathing (white arrowheads) predominantly in the inferotemporal vascular arcades, intraretinal haemorrhages (black arrowhead) in the macula and macula oedema (circled area).

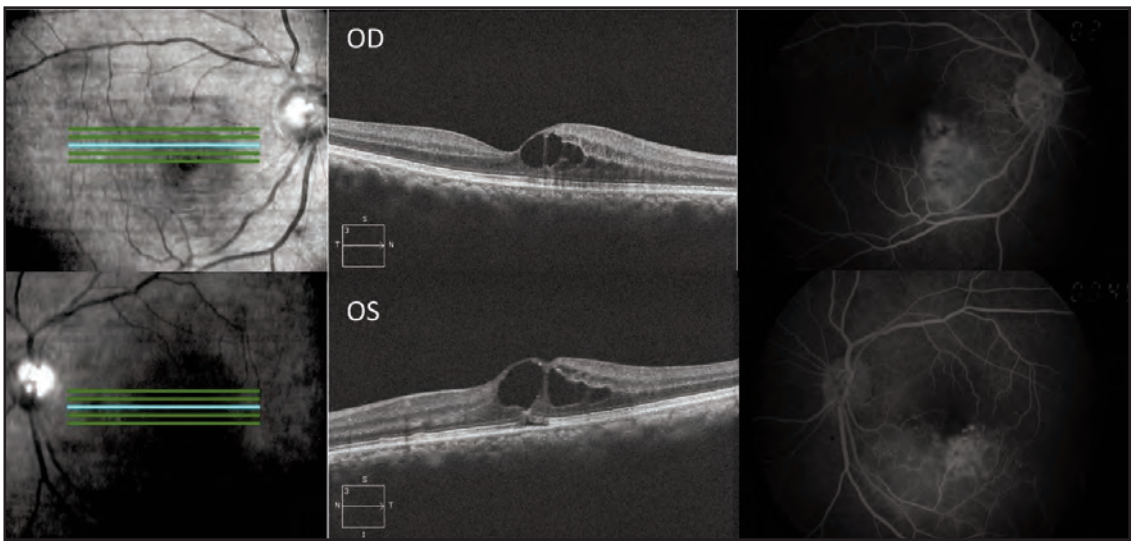

Fig. 2: HD 5-line Raster of the right and left eye showing cystoid macular oedema, worse in the left eye. Corresponding FFA image at late venous phase showing vascular leakage denoting vasculitis in the infero-temporal quadrant extending to the macula.

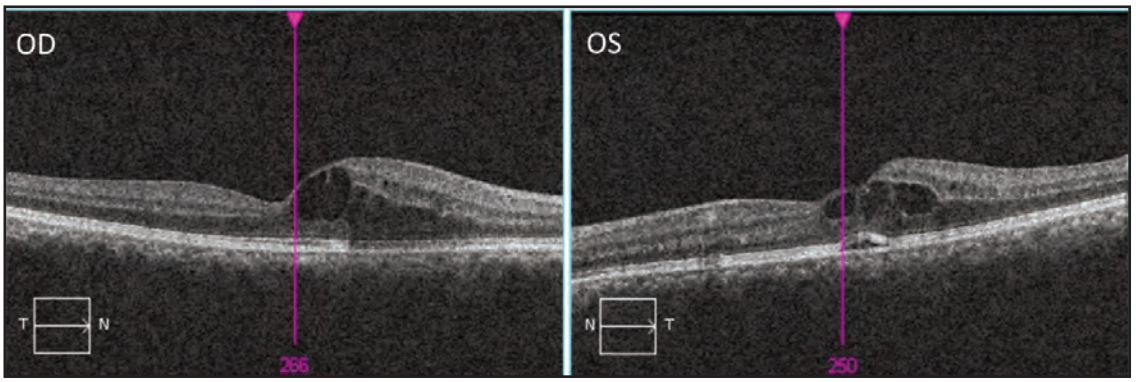

Fig. 3: OCT imaging of the macula 2 months after topical NSAIDs treatment was initiated. Her presenting vision of 6/18 OD and 6/24 OS improved to best corrected visual acuity of 6/12 OU.

NSAIDs were discussed with the patient and she opted for a more conservative approach with trial of topical NSAIDs. Intravitreal steroid therapy was not offered to her as she was still phakic with minimal cataract. After 2 months of treatment with topical NSAIDS, there was a modest reduction in central retinal thickness and improved vision following treatment. She achieved best corrected visual acuity of 6/12 OU was achieved. Subjectively she also reported less metamorphopsia.

Following that the patient was scheduled to be seen in another month but has since defaulted treatment and is lost to follow-up.

DISCUSSION

Chikungunya is a systemic infection that has many associated systemic manifestations.[5] In addition to inducing joint discomfort, the virus can also impact various other organs and systems, including the nervous system, cardiovascular system, skin and kidneys.[5] The exact mechanism of ocular involvement following Chikungunya infection has not yet been studied in detail. Ocular manifestations of Chikungunya can happen concurrent with systemic symptoms or can appear after a quiescent interval. A case series by Mittlal et.al found around two thirds of patients developed ocular symptoms concurrently with systemic illness, while the remaining presented within 6 weeks following resolution of initial illness.[5] The delayed onset of symptoms are postulated to be due to antigenic mimicry, delayed hypersensitivity reaction, or stimulation of a pathogenic lymphocyte reaction.[5] In clinical setting, it is a challenge to clarify the exact interval between the beginning of systemic symptoms and the establishment of eye symptoms. However, awareness about ocular manifestations of Chikungunya should be raised. In this case, we had to explore the patient's medical history in detail before the history of recent hospital admission was reported as it was thought by the patient to be irrelevant to the presenting symptoms.

In this particular case, Toxoplasma IgM was negative while IgG was positive. We do not think that it is toxoplasmosis as our patient lacks the vitritis that is usually present in toxoplasmosis and there are no chorioretinal scars seen. Other retinal vasculopathies like retinal vein occlusion is also less likely as the patient lacks any risk factors and the retinal haemorrhages occurring in this patient is at the macula instead of along the vascular arcades.

The ocular manifestations of Chikungunya are vast. The most common is anterior uveitis, followed by optic neuropathy.6 In a retrospective, observational case series conducted by Lalitha et al., anterior uveitis represented almost one third of cases in in the 37 cases of ocular complications related to Chikungunya infection.[6] Other reported ocular manifestations that can occur infrequently are conjunctival hyperaemia, episcleritis, scleritis, uveitis with glaucoma, monofocal chorioretinitis, neuroretinitis and oculomotor palsies. Conjunctival petechiae, intermediate uveitis, multifocal chorioretinitis, retinal vasculitis and maculopathy are thought to be rare complications.[6]

There is no specific antiviral therapy for Chikungunya infection and treatment is merely symptomatic. For treatment of its ocular complications, there is no difference from treatment of same manifestation due to other aetiologies. As of date, there are no randomised controlled trials for specific treatment of ocular inflammatory diseases associated with Chikungunya.

Our patient had modest improvement in visual acuity and decrease in central retinal thickness following initiation of treatment with topical NSAIDs drops. However, it is unclear whether the disease is by itself self-limiting or whether treatment is truly beneficial. Prognosis varies, ranging from full resolution to permanent vision loss despite intervention.[5]

CONCLUSIONS

Retinal vasculitis with cystoid macular oedema (CMO) is a rare ocular manifestation, but significantly affects vision resulting in morbidity. In recent years, due to the ease of international travel, Chikungunya infection is increasingly reported in both endemic and non-endemic regions, therefore awareness about vast ocular manifestations of Chikungunya should be raised. Other than ophthalmologists, it is also favourable if primary care doctors are made aware of the possible ocular complications that can occur so an early referral to a specialist can be instituted.

DISCLOSURE

No potential competing interest to declare. Informed consent was obtained from the patient in line with COPE standards.

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