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Case report

Successful treatment of tubercular multifocal serpiginous-like choroiditis without use of anti-inflammatory drugs: A case report with multimodal imaging

Mohammad Zarei, Navid Mohsenzadeh, Ramak Roohipoor, Hamid Riazi-Esfahani*

Eye Research Center, Farabi Eye Hospital, Tehran University of Medical Sciences, Tehran, Iran

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Abstract

Purpose: To report the successful management of a case of tuberculosis (TB) related multifocal serpiginous-like choroiditis with anti-TB medications alone.

Methods: A case report with multimodal imaging.

Results: A 28-year-old man presented with progressive loss of vision in his left eye. He mentioned a similar history for his right eye about 2 years earlier. Fundus examination showed multiple discrete greyish irregularly bordered lesions in both eyes. Based on a highly positive skin tuberculin test and exclusion of other possible causes, a diagnosis of presumed TB associated multifocal serpiginous-like choroiditis was made, and quadruple classic anti-TB therapy was initiated. One year after introduction of anti-TB treatment, active lesions turned into scars, with significant improvement of vision.

Conclusion: Anti-TB therapy may be enough as the only necessary treatment for TB-related serpiginous-like choroiditis.

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Keywords: Multifocal serpiginous-like choroiditis; Tuberculosis; Anti tuberculosis therapy

Introduction

Multifocal serpiginoid choroiditis is a progressive choroiditis, also referred to as serpiginous-like choroiditis. In contrast to classic serpiginous choroiditis (SC), multifocal serpiginous-like choroiditis entity is characterized by multifocal, irregular geographic lesions in the fundus with prominent vitritis; ocular involvement is frequently unilateral.

Tuberculosis (TB) has been proposed as the most common cause for multifocal serpiginous-like choroiditis. 1-3

TB-related multifocal serpiginous-like choroiditis requires specific anti-microbial regimen. The necessity of concomitant systemic corticosteroids or immunomodulatory agents is controversial.² Here, we report the successful management of a case of TB-related multifocal serpiginous-like choroiditis with anti-TB treatment alone, followed for one year by multimodal imaging.

E-mail address: Hamidriazi1364@gmail.com (H. Riazi-Esfahani). Peer review under responsibility of the Iranian Society of Ophthalmology.

Case report

A 28-year-old otherwise healthy man, who was born and raised in Afghanistan, presented with progressive loss of vision in his left eye over the past week. He mentioned a similar history for his right eye about 2 years earlier for which

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^{*} Corresponding author. Eye Research Center, Farabi Eye Hospital, Tehran University of Medical Sciences, Qazvin Square, South Kargar Street, 1336616351, Tehran, Iran.

he had not received any treatment that had led to severe vision loss in 2 weeks.

On ocular examination, his best corrected visual acuity was 20/200 in the right eye and 20/32 in the left eye. Anterior segment examination was unremarkable. In anterior vitreous, there was 1 + cells in the right eye and 3 + cells in the left eye [according to Multicenter Uveitis Steroid Treatment (MUST) Trial classification⁴]. Fundus examination of both eyes showed multiple discrete greyish irregularly bordered lesions, noncontiguous to the optic disc and involving the posterior pole and peripheral fundus with variable degree of pigmentation. In the left eye, there were two white-yellow patches with indistinct borders near the fovea (Fig. 1). Spectral domain optical coherence tomography (SD-OCT) scans of the lesions showed retinal thinning and loss of the outer retinal layers with central intraretinal edema in right eye (Fig. 2). Fundus autofluorescence (FAF) imaging depicted bilateral hypoautofluorescent areas corresponding to old lesions. The active lesions in the left eve were hyperautofluorescent and extended beyond the borders seen in fundoscopy (Fig. 1). Fluorescein angiography (FA) revealed vascular leakage, especially in left eye due to vasculitis. Disc leakage was evident in both eyes (Fig. 3).

Interestingly, the patient brought us images dated back to 2 years earlier. Based on this evidence, the left eye had been

lesion-free at that time, but in the right eye, there had been multiple lesions, some of which seemed to be active, especially in vicinity of the fovea (Fig. 1).

Mantoux tuberculin skin test (TST) showed 20 mm induration. Computed tomography scan of the chest, as well as sputum culture, were negative for pulmonary TB.

Considering the endemic status of TB in Afghanistan and based on clinical picture and a highly positive tuberculin test and exclusion of other possible causes, a presumed diagnosis of TB related multifocal serpiginous-like choroiditis was made. Quadruple anti-TB therapy with daily isoniazid 300 mg, ethambutol 1200 mg, pyrazinamide 1500 mg, rifampin 600 mg was initiated, with a plan to discontinue ethambutol and pyrazinamide after two months. Two weeks later, the patient noticed dramatic visual acuity improvement (OD: 20/100 and OS: 20/25). Since the patient showed good visual recovery without paradoxical worsening of inflammation (no Jarisch—Herxheimer-like reaction), it was deemed not to add corticosteroid or immunomodulatory agents to treatment.

One year later, the vision was improved to 20/32 in his right eye and 20/20 in his left eye. Examination revealed no aqueous or vitreous cells, and the active lesions were completely healed. FAF showed uniform hypoautofluorescence of the lesions that were hyperautofluorescent 1 year earlier (Fig. 1). There was also minimal disc leakage in

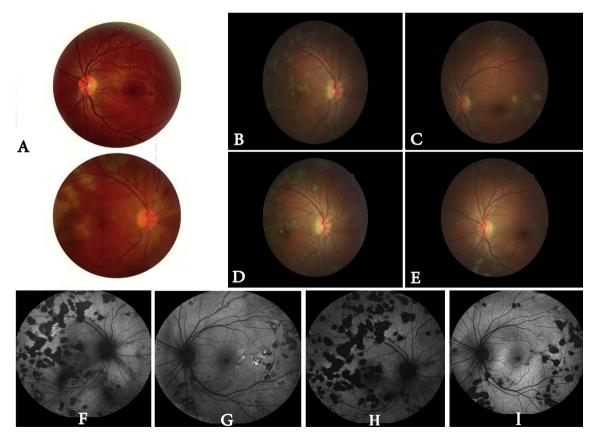


Fig. 1. A: Fundus photos of the patient 2 years before presentation. B and C: Fundus photo at presentation: multiple discrete greyish lesions with variable degree of pigmentation. D and E: Fundus photo after treatment. F and G: Autofluorescence at presentation: depicted bilateral hypoautofluorescent areas corresponding to old lesions. The active lesions in the left eye are hyperautofluorescent (temporal to fovea). H and I: Autofluorescence after treatment: uniform hypoautofluorescence of the lesions that were hyperautofluorescent 1 year earlier is evident.

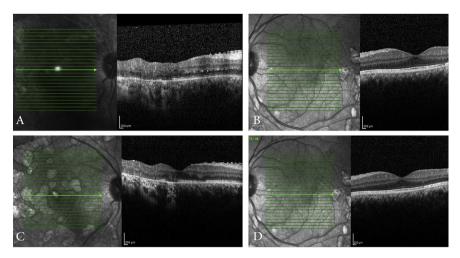


Fig. 2. A: Spectral domain optical coherence tomography (SD-OCT) scans of the lesions in right eye before treatment show central intraretinal edema, retinal thinning with loss of the ellipsoid zone, external limiting membrane and interdigitation zone. B: SD-OCT of left eye, before treatment, with no central lesion. C: SD-OCT of right eye after treatment: Although central edema improved, outer retinal attenuation with a correspondent increase in light reflectivity from the choroid are evident in healed lesions. Inner layers of retina are more distinct compared to pre-treatment appearance. D: SD-OCT of left eye after treatment.

both eyes, and the vascular leakages were improved dramatically based on FA. Although central edema was improved, outer retinal layers attenuation was evident in healed lesions on OCT of the right eye (Fig. 2).

Discussion

Here we reported a case of TB related multifocal serpiginous-like choroiditis, successfully treated with anti-TB medications, without any corticosteroid or immunomodulatory agents.

In a group of patients with multifocal serpiginous-like choroiditis, TB- Deoxyribonucleic acid (DNA) has been found in ocular fluids; therefore, anti-TB medication seems to be the main treatment option. ^{2,3,5,6} Unlike classic SC that is usually treated with immunomodulation, patients with TB-related multifocal serpiginous-like choroiditis might show progression of retinal lesions despite immunosuppressive treatments. ^{1,2}

There is no standard treatment for multifocal serpiginouslike choroiditis. However, there are reports of patients with multifocal choroiditis whose inflammation responded to anti-TB treatment alone. But some authors stated that systemic corticosteroids or immunomodulatory drugs are necessary to control active lesions. Recurrence rate seems to be significantly higher in cases who are treated with anti-inflammatory drugs alone.⁵ There was a wide heterogeneity in duration of TB treatment. A standard regimen of isoniazid, rifampin, ethambutol, and/or pyrazinamide for a minimum of 2 months (up to 3-4 months) was used, with subsequent administration of 2-drug therapy, namely isoniazid and rifampicin, for a minimum of 4 months up to 15 months based on different studies.⁸ Based on Ang et al. study, in patients with TBrelated uveitis, anti-TB treatment for more than 9 months can lead to an 11-fold reduction in the likelihood of recurrence.9

Guedes et al.⁶ and Zhang et al.¹⁰ have reported cases of tubercular serpiginous-like choroiditis, who had responded to anti-TB alone with no recurrence during 6-month follow-up. They showed treatment success based on visual acuity and fundus examination. Here, we have treated the patient with anti-TB alone, followed the patient after one year of treatment, and demonstrated the success of treatment in terms of visual acuity, fundus appearance, OCT, FAF, FA, and indocyanine green angiography (ICGA). We showed that all the lesions became hypoautofluorescent one year after treatment.

In contrast, Mackensen et al.¹¹ and Oray et al.¹² reported remission without relapses in patients with TB-related serpiginous-like choroiditis who had been treated with anti-TB and corticosteroid. They showed that these patients were relapse-free, based on clinical findings and visual acuity for about 1 year. They could not conclude whether systemic corticosteroid was necessary as an adjuvant therapy in all cases and suggested larger randomized clinical trials.

Based on a recent meta-analysis by Kee et al. 8 on treatment of different types of ocular TB, there was minimal difference in the outcome between patients treated with anti-TB therapy alone (85% successful outcome) and those with concomitant systemic corticosteroid (82% successful outcome). They concluded that use of anti-TB therapy may be sufficient for patients with intraocular TB; however, this conclusion is limited by the lack of control group analysis and standard treatment protocols.

The patient's right eye visual acuity was significantly improved with anti-TB treatment. Banasl et al. showed that multifocal lesions tend to spare the fovea even in eyes with macular involvement; therefore, final visual acuity can be good with appropriate treatment in TB-related multifocal serpiginous-like choroiditis. ¹³ Fluorescein leakage from disc and vessels and vitritis-which are not common features in pure "choroiditis" unless they occur in the setting of secondary choroiditis (TB, sarcoidosis, syphilis)- were significantly

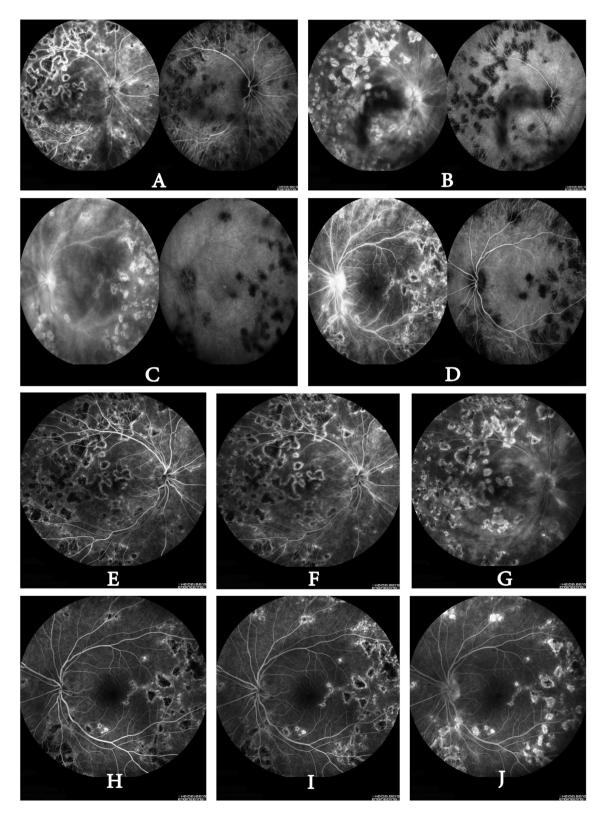


Fig. 3. A—D: Fluorescein angiography (FA) and indocyanine green angiography (ICGA) at presentation (early and late): on FA, the inactive lesions are hypofluorescent with late staining of borders. The active lesions have stronger hyperfluorescence of borders due to leakage. There is evidence of vascular leakage in left eye due to vacuities. Disc leakage is evident in both eyes. On ICGA, both the active and old lesions appear hypofluorescent in early and late phases. E—G: FA of right eye after treatment. H—J: FA of left eye after treatment. The disc and vascular leakages of both eyes improved dramatically.

improved after anti-TB treatment, which could be another explanation for visual acuity improvement. This is especially applicable to improved vision in the right eye, where the choroidal lesions were mostly inactive. It is important to note that after the completion of anti-TB treatment, significant improvement in all signs of disease activity (choroidal lesions, retinal vasculitis and optic disc fluorescein leakage and cellular reaction), which is not typical for untreated cases, were observed.

One concern while initiating anti-TB without any anti-inflammatory drug is paradoxical worsening or progression of the lesions due to a Jarisch—Herxheimer-like reaction. It is generally due to immune response to the antigens released from dying bacilli. It is important to be aware of the possibility of such reactions while using anti-TB, follow the patients in short intervals in first weeks of treatment and to start appropriate anti-inflammatory medication in this situation. 4,11,14 Our patient did not show such reaction and did well with anti-TB alone.

In this report, we documented changes in findings of multimodal imaging that can be used to monitor response to treatment in TB-related serpiginous-like choroiditis.

In conclusion, this case adds further evidence to support the idea that in some cases of TB-related serpiginous like choroiditis, anti-TB therapy maybe enough as the only necessary treatment. But to consolidate this notion, it is necessary to perform larger studies, preferably, controlled trials.

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