

Occlusive Vasculitis with Multifocal Choroiditis in Fungal Skin Infection

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Abstract

Multifocal choroiditis (MFC) with occlusive vasculitis of tubercular etiology is well known in a tuberculosis endemic country like India but presumed ocular histoplasmosis syndrome with multifocal choroiditis has been rarely reported. We describe 2 immunocompetent patients with presumed fungal multifocal choroiditis and associated occlusive vasculitis. In both the cases choroiditis resolved with antifungal monotherapy but vasculitis required oral steroids. Improvement of choroiditis with antifungal therapy as documented in our cases may instigate uveitis specialists to explore fungal etiology in MFC with occlusive vasculitis.

Keywords: Multifocal Choroiditis (MFC); Occlusive Vasculitis; Fungal Skin Infection

Introduction

Focal or Multifocal choroiditis (MFC) with or without retinal vasculitis of tubercular etiology in tuberculosis (TB) in an endemic country like India is widely reported and has also been well documented histopathologically [1-3]. But choroiditis and occlusive retinal vasculitis of other infectious etiologies especially fungal in immunocompetent individual is rarely been described unless it is presumed ocular histoplasmosis syndrome (POHS) or endogenous endophthalmitis [4,5].

Case Report

We hereby describe two unique immunocompetent cases of bilateral MFC and occlusive vasculitis associated with a fungal skin infection.

Case 1

A 23-year-old Indian male, previously diagnosed with idiopathic bilateral MFC and occlusive vasculitis (Figure 1a) with vitreous haemorrhage in the OS (left eye), presented with complaints of painless, distorted vision in the right eye (OD) of 5 days duration. Patient's previous investigations for vasculitis included Mantoux, TPHA (Treponema pallidum haemagglutination), Quantiferon Gold TB test, Serum Angiotensin Converting Enzyme (ACE) were all normal but ESR (erythrocyte sedimentation rate) and CRP (C-reactive protein) was slightly high (35 mm/Hr and 0.82 mg/L respectively). He had earlier been treated with oral methotrexate (for 6 months) and oral steroids (over 2 months) in tapering doses, for his retinal vasculitis. The patient had not been on any treatment for the past 15 months prior to this presentation in the right eye. Patient had undergone pars plana vitrectomy for persistent vitreous haemorrhage in the left eye 3 months ago. His last fundus fluorescein angiography (FFA) (done 3 months back) did not show vascular leakage or early hypo and late hyperfluorescence suggestive of resolved inflammation. Patient gave a history of having skin lesions over the abdomen and back on and off but had not undergone any treatment. Best corrected visual acuity (BCVA) in OD was 20/20 and in the OS was counting fingers at 2 meters. Examination of OD revealed quiet anterior segment, 1+ vitreous cells, resolved lasered vasculitis and a fresh lesion of choroiditis at the macula. Optical coherence tomography (OCT) showed small pocket of subretinal fluid, ellipsoid zone disruption and a small choroidal

elevation (Figure 1b). Left eye examination showed a complicated cataract and resolved lasered vasculitis. Patient was empirically started on indomethacin and topical nepafenac while the patient was being re-investigated. The patient defaulted follow up and was reviewed again after 2 months. The patient had taken the prescribed medications for two weeks and then discontinued. Patient had used unknown ointment for his skin lesions for a month and then he was started on oral antifungals (T. Itraconazole 100 mg BD) by his dermatologist, which he was on for about 20 days at his second eye review. On evaluation, he had dark pigmented, well circumscribed, granular round lesion with central clearing on his abdomen which was diagnosed as tinea cruris with tinea corporis (Figure 1e).

Right eye examination revealed multiple placoid circular inactive choroiditis lesions at the macula, OCT and fundus auto-fluorescence (FAF) suggested resolving multifocal choroiditis (Figure 1c and 1d). His fresh blood work up was negative for Mantoux, HIV, TPHA, but positive for quantiferon TB gold (10 IU/ml). After resolution of skin lesion the dermatologist discontinued antifungals and after a week patient came back with relapse of vasculitis and a new lesion of choroiditis (Figure 1f and 1h). Inspired from his previous response to antifungals, we re-started him on itraconazole 100mg twice daily and the patient discontinued again after two weeks. He did not review as advised and came for review after three weeks of stopping itraconazole. The new choroiditis patch had resolved but vasculitis recurred (Figure 1g and 1i). Oral steroids (40 mg in a tapering dose) along with itraconazole 100mg BD was prescribed over a period of 1 month to which vasculitis responded well.

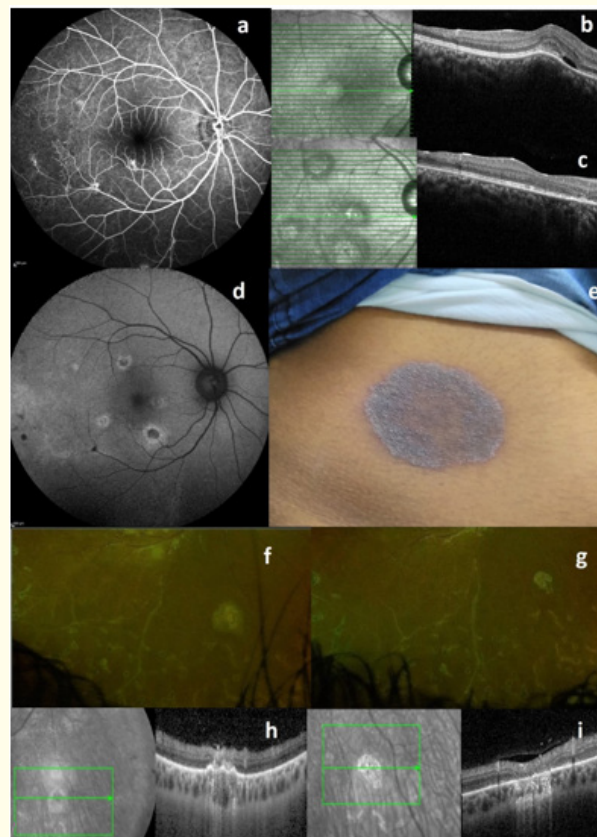


Figure 1: Case 1: Fundus Fluorescein angiography of OD in late phase showing areas of capillary non perfusion areas temporal to fovea and mild staining of small choroidal scars (a). OCT OD showing a choroidal elevation with small pocket of subretinal fluid and ellipsoid zone (EZ) disruption (b). Subsequent OCT done after treatment with itraconazole shows resolution of choroidal elevation and subretinal fluid along with mild EZ loss. (c) Fundus autofluorescence (FAF) showing central hypoautofluorescence with surrounding mild hyperautofluorescence suggestive of resolving choroiditis lesions (d).

A dark pigmented, well circumscribed, granular round lesion with central clearing on the right side of abdomen was diagnosed as Tinea cruris (e). Coloured fundus image of same eye shows appearance of a new chorio-retinal lesion in inferior mid-periphery (f) which again resolved after itraconazole monotherapy (g) OCT over same lesion showed mild choroidal thickening, retinal pigment epithelial undulations and indistinct retinal layers (h) which resolved after above treatment (i).

Case 2

30-year-old Indian male presented with a history of pain and floaters in the left eye for 3 days. Patient was on oral treatment for fungal skin infection (T. Fluconazole 150 mg BD for past 2 weeks). BCVA was 20/20 in OU and IOP was normal. Clinical examination revealed bilateral resolved MFC but had active occlusive vasculitis. FA and FAF confirmed clinical anatomical diagnosis (Figure 2a-2d). Patient's baseline investigations (CBC, TPHA, HIV, Mantoux, ESR and CRP) were normal. After dermatologist's clearance the patient was put on oral steroids (40 mg in tapering dose) and underwent pan-retinal photocoagulation to both the eyes. Oral steroids were gradually tapered off and stopped after 6 weeks. Patient was followed up for 5 years. His subsequent FA revealed no active inflammation (Figure 2e and 2f). Patient remained asymptomatic with BCVA 20/20 and no recurrences were noted at the end of five year follow up.

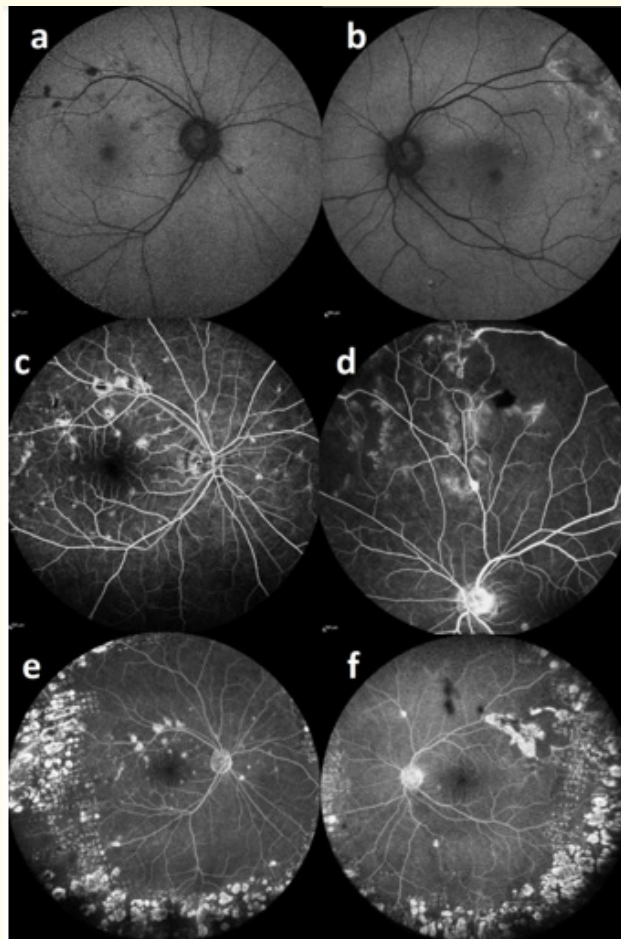


Figure 2: Case 2: Fundus autofluorescence shows multiple small hypoautofluorescence lesions at the macula in both the eyes (a, b) with a larger hypoautofluorescent lesion with hyperautofluorescent lesion in supero-temporal quadrant in the left eye(b). Fundus fluorescein angiography shows staining of choroidal scars in OD (c) and capillary non-perfusion area in OS (d). Repeated FA after 3 years shows staining of chorio-retinal scars and no vasculitic leakage from retinal vessels (e, f).

Discussion

Occlusive vasculitis with vitreous hemorrhage is a commonly encountered condition in clinical practice. Rarely vasculitis and choroiditis may precede each other or reactivate independently during the course of the disease as observed in our cases. History of occlusive vasculitis with vitreous haemorrhage, de-novo appearance of multifocal choroiditis with outer retinal involvement and positive

quantiferon TB gold test in case 1 may suggest the diagnosis of tuberculosis in TB endemic country. But their appearance along with fungal skin lesion prompted postulating a fungal etiology in our cases. Although PCR studies from ocular fluid were not done in our cases treatment response with anti-fungal (AFT) alone (in 2nd and 3rd visit) as seen in case 1 may support consideration of *Tinea cruris* as a causative agent for choroiditis directly or indirectly akin to POHS which is rarely reported from India [6]. Similar phenotypical presentation in case 2 with history of fungal skin infection and complete resolution of inflammation without recurrences in 5 years of follow up makes other possible causes of MFC with occlusive vasculitis less likely, nevertheless choroidal tuberculoma manifesting in a patient of Eales disease 6-years after initial presentation has recently been reported [2]. Given no history of skin infection case 1 would have also received anti-tubercular treatment (ATT) and steroids.

Case 1 who received only indomethacin therapy initially, developed few more choroiditis patches resembling his skin lesions (Figure 1c-1e) which surprisingly responded once the anti-fungal medications commenced by his dermatologist. After stopping anti-fungals without ophthalmologist's consultation, resulted in appearance of a new choroiditis lesion which again responded to itraconazole monotherapy (Figure 1f-1i), although his reactivated vasculitis required moderate dose of oral steroids along with AFT. We believe indomethacin had minimal role to play in this case as he developed more lesions while on indomethacin therapy. Spontaneous resolution of choroiditis of a different etiology can be well argued against presumed fungal etiology in our cases. Larger case series with similar phenotypical presentation with supporting molecular or pathological diagnostics are recommended.

Conclusion

Systemic antifungal treatment for skin lesions can accidentally lead to resolution of choroiditis lesions. It would be difficult to actually prove fungal aetiology in these cases. It is presumed that the lesions were of fungal origin. Improvement of choroiditis with AFT as documented in our cases may instigate uveitis specialists to explore fungal etiology in MFC with occlusive vasculitis.

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