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CASE REPORT

Non-neovascular subretinal fluid secondary to choroidal osteoma controlled with intravitreal aflibercept – A case report

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Abstract

The aim of the study was to describe a case of subretinal fluid without neovascularization due to choroidal osteoma that was treated successfully with intravitreal aflibercept. Choroidal osteoma is an ossifying tumor that can cause visual impairment through several pathological mechanisms, including fluid accumulation under the retina, with or without choroidal neovascularization (CNV). We described a young adult man who presented with unilateral visual impairment due to choroidal osteoma without CNV, resistant to bevacizumab intravitreal injections, which showed a marked response to aflibercept. Our experience provides support for the use of aflibercept in the treatment of non-neovascular choroidal osteoma; this is the first report describing this encouraging result. Nevertheless, the current observation merits further research.

Introduction

Choroidal osteoma is a benign ossifying tumor that is more prevalent among young individuals. Visual impairment is thought to be the result of several pathological mechanisms, including fluid accumulation under the retina, lesion decalcification, photoreceptors loss, retinal pigment epithelium (RPE) atrophy, and occasionally choroidal neovascularization (CNV).[1]

Case Report

A healthy 43-year-old man presented to the emergency room complaining of the right eye metamorphopsia and micropsia noted during his work as an architect. His visual acuity was 6/8.5 in the right eye and 6/7.5 in the left eye. Anterior segment examination was unremarkable. Dilated funduscopic examination of the right eye revealed a juxtapapillary white elevated lesion measuring four disc diameters extending over the superior vascular arcade to the superior macula. Figure 1a shows the fundus findings that were captured using a retinal camera (TRC-50XF, Topcon Medical Systems, Paramus, NJ, USA). Optical coherence tomography (Heidelberg Engineering, Heidelberg, Germany) demonstrated right eye macular subretinal fluid (SRF) with resultant retinal elevation, adjacent RPE changes, and hyperreflective dots. Central macular thickness (CMT) measured 443 μm. On fluorescein angiography (TRC-50XF, Topcon Medical Systems, Paramus, NJ, USA), nonspecific leakage was evident in the vicinity of the lesion; however, there was no evidence of CNV [Figure 1b]. Subsequent B-scan ultrasonography (Sonomed) revealed a hyperechoic mass casting an acoustic shadow over the posterior pole [Figure 1c]. The patient was diagnosed with choroidal osteoma and was treated with serial monthly intravitreal injections of 1.25 mg bevacizumab. This treatment strategy had yielded no significant visual or structural improvement, and following the fifth injection, it was abandoned. He received two bimonthly intravitreal 2 mg aflibercept injections that proved to be highly beneficial in terms of both anatomical and functional measures. On follow-up examination, 1 month after the last injection, visual acuity improved to 6/6.6, and CMT decreased to 197 μm [Figure 2]. Since then, for the past 2.5 years, he is treated with aflibercept injections every 6 months with preservation of vision.

Discussion

We described a patient with SRF secondary to non-neovascular choroidal osteoma who was recalcitrant to bevacizumab injections yet responded well to aflibercept injections. To date, no consensus exists regarding the optimal treatment strategy for choroidal osteoma. Given its rarity, there is a scarcity of publications regarding treatment options for

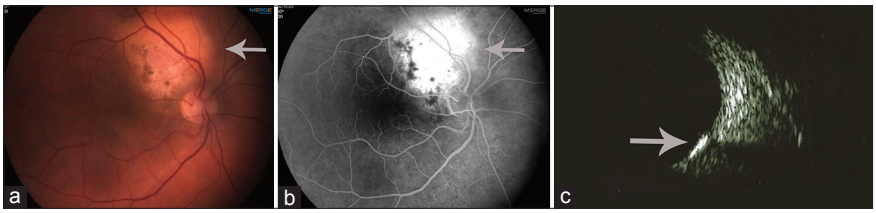

Figure 1: Choroidal osteoma demonstrated by color fundus photography (a), fluorescein angiography (b), and ultrasonography (c)

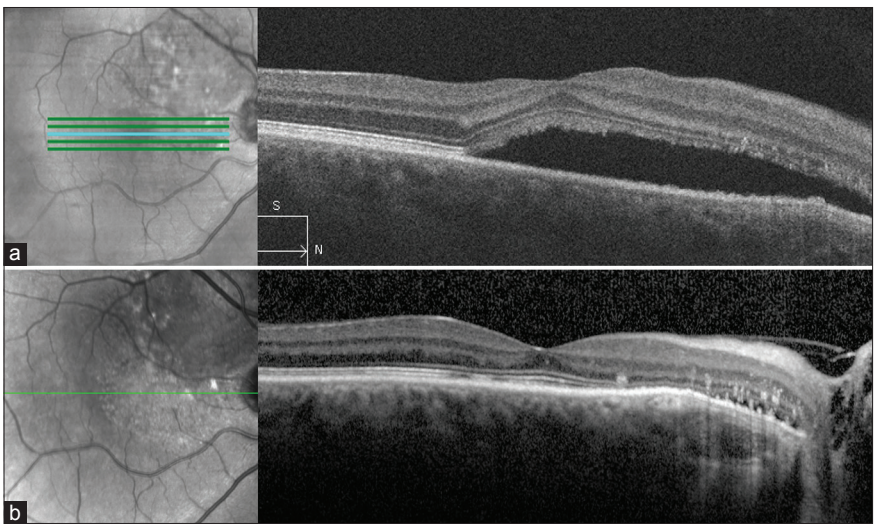

Figure 2: Subretinal fluid (a) and fluid reabsorption (b) as evident on optical coherence tomography

choroidal osteoma without CNV; focal laser, transpupillary thermotherapy, bevacizumab, and ranibizumab intravitreal injections have all been investigated.[2-6] With regard to anti-vascular endothelial growth factor (VEGF) therapy, in a retrospective case series of 6 eyes with choroidal osteoma-associated SRF, Song *et al.*[4] reported improved visual and anatomical outcomes following intravitreal injections of bevacizumab; to note, CNV was absent in all but one eye. Clinical response dictated a spectrum of 1, 2, or 3 injections spanning a mean follow up of approximately 11 months. Furthermore, Vayalambrone and Misra[6] described a successful treatment of intravitreal ranibizumab injections in an 11-year-old girl with non-neovascular choroidal osteoma. Previous research regarding the use of aflibercept has been restricted to choroidal osteoma associated with CNV. In an Italian case report, the use of VEGF-trap as a rescue treatment for CNV associated with choroidal osteoma was reviewed. Following the failure of both bevacizumab and ranibizumab administrated intravitreally, Saitta *et al.*[7] reported improved vision and retinal morphology after three intravitreal injections of aflibercept. To the best of our knowledge, the current report is the first to describe a case of choroidal osteoma without CNV, who failed to respond to bevacizumab, yet nevertheless was successfully treated with aflibercept. Apart from its crucial participation in the angiogenesis cascade, VEGF also plays an important role in fluid accumulation as it possesses permeability enhancing characteristics.[8] It is possible to speculate that the improved VEGF-affinity of aflibercept is responsible for the encouraging response in abolishing SRF in choroidal osteoma.

Conclusion and Clinical Significance

In summary, our experience provides support for the use of aflibercept for the treatment of non-neovascular choroidal osteoma; this is the first report describing this encouraging result. Nevertheless, the current observation merits further research.

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