Published in final edited form as:

Ophthalmic Surg Lasers Imaging Retina. 2013 ; 44(1): 77–80. doi:10.3928/23258160-20121221-17.

De-novo Appearance of a Choroidal Osteoma in an Eye with Previous Branch Retinal Vein Occlusion

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Abstract

This report describes the de-novo appearance of a choroidal osteoma in an eye, following years after laser photocoagulation for previous branch retinal vein occlusion (BRVO). A 62-year old male presented with an asymptomatic yellowish-orange lesion in the macula on fundus examination of his left eye, during a regular follow up visit for bilateral BRVO associated with macular edema, which had been treated with laser photocoagulation many years ago. The lesion was observed for 1.5 years until decrease in vision occurred. Fundus photography revealed a yellow-to-orange, well-defined lesion in the macular region. Fluorescein angiography was consistent with choroidal neovascularization (CNV). OCT and B-scan ultrasonography showed features consistent with choroidal osteoma. This is the first report of the de-novo appearance of a choroidal osteoma in an eye, following years after laser photocoagulation for BRVO. CNV developed secondary to the lesion, which was treated with intravitreal Bevacizumab, leading to subjective and anatomic improvement.

Keywords

BRVO; choroidal osteoma; CNV; intravitreal Bevacizumab; laser photocoagulation; OCT; sub-retinal fluid

INTRODUCTION

Choroidal osteoma is a rare, benign, ossifying tumor, typically presenting in the juxta-papillary region of young healthy females with no history of systemic or ocular disease, in their second or third decades of life.¹⁻³ The most striking feature of this rare entity is a slightly elevated well-defined mass, which is yellowish orange in appearance, as characteristically observed on ophthalmoscopic examination.^{2,4} There may be variation in color due to depigmentation, hyperplasia or thinning of the overlying retinal pigment epithelium (RPE).¹

Choroidal osteoma has been reported to rarely present in association with pregnancy and in eyes with previous history of ocular diseases such as optic atrophy, optic neuritis in Behcet's disease, retinitis pigmentosa, Stargardt's disease and bilateral optic nerve sheath calcification.^{3,5-9} To the best of our knowledge, it has never been shown to present in an eye

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Conflict of Interest

Jay S. Duker receives research support from Carl Zeiss Meditech, Inc., Optovue, Inc., and Topcon Medical Systems, Inc.

with previous BRVO. This report describes the de-novo appearance of choroidal osteoma occurring in an eye years after laser photocoagulation for BRVO.

CASE REPORT

A 62 year-old Asian male, with a known history of diabetes mellitus, hypertension and asthma, presented in April 2009, at the New England Eye Center, Tufts Medical Center, Boston, with an asymptomatic yellowish orange lesion on fundus examination of his left eye, during a regular follow up visit for bilateral BRVO. He had an ocular history of branch retinal vein occlusion (BRVO) with macular edema for 10 years in the right and 8 years in the left eye, which had been successfully treated with sectoral pan-retinal photocoagulation (PRP) and focal laser bilaterally [Figure 1]. Since the last 6 years, he maintained stable vision of 20/100 in the right and 20/30 in the left eye.

At the time of presentation, his best-corrected visual acuity (BCVA) was 20/200 in the right eye and 20/30 in the left eye. On dilated fundus examination, both eyes showed areas of sectoral PRP, sclerotic vessels in the periphery, and scarring with sub-retinal fibrosis, owing to the previous history of laser photocoagulation for BRVO. However, fundus photography of the left eye revealed a well-circumscribed yellowish orange lesion in the macular region [Figure 2], while OCT showed a slight disruption of the RPE [Figure 2]. The lesion was observed on follow up visits.

After 1.5 years of the initial appearance of the lesion, the patient presented with sudden distortion of vision in his left eye, and was evaluated with fundus photography, which again revealed the previously observed lesion [Figure 3]. Fluorescein angiography (FA) and OCT features were consistent with CNV and sub-retinal fluid collection [Figure 3]. A B-scan ultrasonogram revealed a hyper-reflective choroidal mass in the posterior pole, with posterior acoustic shadowing, consistent with the diagnosis of choroidal osteoma [Figure 3].

The patient was treated with 1.25mg/0.05ml intravitreal Bevacizumab, leading to subjective improvement in vision. He received a total of 8 injections during a period of 14 months, at intervals ranging from 1-3 months, for recurrent sub-retinal fluid collection due to the CNV. Following that, his visual acuity stabilized without therapy for 8 months. At the time of his last follow up, the BCVA in the left eye was 20/25, and fundus photography and OCT showed a substantial improvement [Figure 4].

DISCUSSION

This is the first report of a de-novo appearance of a choroidal osteoma in an eye with a previous history of BRVO treated with laser photocoagulation many years ago. The clinical appearance, B-scan ultrasonography and OCT features were characteristic for choroidal osteoma, as described above.

Gass et al in 1978 first described four healthy young women with characteristic ophthalmoscopy appearance of a slightly elevated, juxta-papillary, yellowish orange, choroidal lesion with well-defined margins.⁴ The pathogenesis of choroidal osteoma is obscure,⁵ although choristomatous, inflammatory and hereditary etiologies have been suggested.⁹⁻¹² These tumors are generally slow growing, leading to a slow but progressive decrease in vision.² In a follow-up study of 36 patients, the probability of loss of visual acuity (20/200 or worse) was more than 50% by 10 years.¹² CNV is the most frequent cause of visual impairment in choroidal osteoma.⁶ Despite the benign nature of the tumor, vision is usually compromised due to gradual atrophy of the overlying retina, atrophy of the RPE, sub-retinal detachment and accumulation of sub-retinal fluid/hemorrhage associated with CNV.^{2,3,13} Ultrasonography and computed tomography (CT) are valuable investigation

modalities, revealing high tissue density due to calcification.² With B-scan ultrasonography, the tumor is acoustically dense at high sensitivity, and shadowing is seen posterior to it, creating the appearance of a pseudo-optic nerve. CT scan reveals a plaque with the same density as bone at the level of choroid.¹ Fundus auto-fluorescence has also been described to assist in diagnosis.¹⁴ Recently, optical coherence tomography (OCT) imaging is being used to determine the features of choroidal osteoma, and demonstrates a plate-like hyper-intense appearance, involving RPE atrophy, specifically at an early stage of the tumor's evolution.¹

In our patient, choroidal osteoma appeared de-novo in an eye previously treated with laser photocoagulation for BRVO. This unique appearance may either be related to a dysfunction of the RPE owing to the previous history of frequent macular edema secondary to BRVO or a late complication of laser photocoagulation. However, a definitive comment cannot be made in this regard, as this occurrence may be purely co-incidental.

The definitive treatment for CNV associated with choroidal osteoma has not yet been elucidated. Although photocoagulation, surgical removal of CNV, photodynamic therapy, and transpupillary thermotherapy have been investigated, long-term vision is usually not well preserved.² Intravitreal Bevacizumab has been shown to be effective in preventing visual impairment in eyes with choroidal osteoma associated with CNV.^{2,13}

In conclusion, we describe the de-novo appearance of choroidal osteoma in an eye treated with laser photocoagulation many years ago for BRVO. CNV developed secondary to the lesion, which was treated with intravitreal Bevacizumab. The response to treatment was stable, leading to both subjective and anatomic improvement.

Acknowledgments

Financial Support

This work was supported in part by a Research to Prevent Blindness Unrestricted grant to the New England Eye Center/Department of Ophthalmology -Tufts University School of Medicine, NIH contracts RO1-EY11289-23, RO1-EY13178-07, RO1-EY013516-07, Air Force Office of Scientific Research FA9550-07-1-0101 and FA9550-07-1-0014, and the Massachusetts Lions Eye Research Fund.

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Figure 1.

(A) Red-free fundus image of the left eye depicting BRVO. Note the presence of intra-retinal hemorrhage typically involving the supero-temporal quadrant (black arrow), as well as scattered areas of hemorrhage temporal to the optic nerve head and involving the macular region (green box). (B) Red-free fundus image of the left eye following laser photocoagulation, showing the resolving intra-retinal hemorrhages. Note the previously observed scattered areas of hemorrhage temporal to the optic nerve head and in the macular region have now resolved, and small hard exudates have appeared typically in the macular region (black arrow). These exudates disappeared completely on follow-up dilated fundus examinations, following which the patient maintained stable vision of 20/30 in his left eye for 6 years. BRVO, branch retinal vein occlusion; PRP, pan-retinal photocoagulation.

Figure 2.

(A) Color fundus photograph of the left eye showing a yellowish orange, well-defined lesion in the macular region (black arrow). (B) OCT image of the left eye, showing a mild disruption of the RPE (red arrow). There is no evidence of CNV or sub-retinal fluid collection. OCT, optical coherence tomography; RPE, retinal pigment epithelium; CNV, choroidal neovascularization

Figure 3.

(A) Color fundus photograph of the left eye showing a yellowish orange, well-defined lesion with surface depressions and elevations in the macular region (black arrow). (B) OCT image of the left eye, showing an irregular plate-like hyper-reflectivity of the lesion (yellow asterisk) extending into the retina, with atrophy of the overlying RPE (green arrowheads). Sub-retinal fluid collection can also be observed (red arrow). (C) Fluorescein Angiography of the left eye showing leakage of the dye in the region of macula consistent with CNV secondary to the choroidal lesion (black arrow). (D) B-scan ultrasonography of the left eye showing a highly reflective choroidal lesion in the posterior pole (red arrow) and acoustic shadowing just posterior to the mass (green arrow). OCT, optical coherence tomography; RPE, retinal pigment epithelium; CNV, choroidal neovascularization

Figure 4.

(A) Color fundus photograph of the left eye following completion of treatment with intravitreal Bevacizumab, showing a reduction in size, as well as the elevations and depressions of the lesion in the region of macula (black arrow). (B) OCT image of the left eye showing a substantial reduction in the size of the lesion (yellow asterisk). Note that the sub-retinal fluid collection has resolved with treatment. OCT, optical coherence tomography