Ranibizumab for Choroidal Neovascularization Secondary to Choroidal Osteoma Associated and Central Serous Choroidopathy

Rosa Dolz-Marco¹, Sebastián Martínez-Castillo¹, Carol L. Shields², Roberto Gallego-Pinazo^{1,*} and Manuel Díaz-Llopis^{1,3}

¹Department of Ophthalmology, University and Polytechnic Hospital La Fe, Valencia, Spain

²Ocular Oncology Service, Wills Eye Institute, Thomas Jefferson University, Philadelphia, PA, USA

³Faculty of Medicine, University of Valencia, Valencia, Spain

Abstract: *Purpose*: To describe a case of atypical choroidal osteoma with secondary choroidal neovascularization associated with chronic central serous chorioretinopathy.

Case Report: A 46-year-old man presented with a 6-month history of vision loss in his left eye (OS). The visual acuity was 20/30 in his OS. Dilated fundus examination OS evidenced a whitish deep lesion in the superior temporal arcade suggestive of choroidal osteoma. Spectral-domain optical coherence tomography revealed a serous macular detachment with a heterogeneous hyperreflective intrachoroidal mass within the superior temporal major arcade with overlying cystoid macular edema, and a temporal macular pigment epithelial detachment. An intravitreal injection of ranibizumab was administered and one month later the visual acuity improved to 20/25 and the subretinal fluid partially regressed.

Conclusion: Although choroidal osteoma has no definite therapeutic approach, it may associate secondary complications. Patients should be monitored in order to detect changes suitable for potential therapies available and therefore minimize the visual burden of the disease.

Keywords: Central Serous Choroidopathy, Choroidal Neovascularization, Choroidal Osteoma, Ranibizumab, Subretinal Fluid.

INTRODUCTION

Choroidal osteomas are rare benign tumors firstly described by Gass in 1978 as yellowish slightly elevated juxtapapillary choroidal lesions with sharp geographic borders in healthy young women [1]. The typical features of choroidal osteomas include unilaterality, well-defined boundaries, slightly elevation, white-to-cream or orange color, and calcium density observed by ultrasonography and computed tomography.

These lesions may be asymptomatic, but may course with progressive vision decline due to tumor enlargement, partial or total tumor decalcification with consequent overlying retinal pigment epithelium alteration, and the development of secondary choroidal neovascularization (CNV). There is no established treatment for the CNV associated with choroidal osteoma. Different approaches have been reported including photocoagulation [2, 3], photodynamic therapy [4, 5] and transpupillary thermotherapy [6, 7]. Recently, the use of intravitreal anti-vascular endothelial growth factor (anti-VEGF) has shown positive outcomes [8-12]. Herein we report a case of choroidal osteoma with an atypical presentation successfully treated with intravitreal ranibizumab.

CASE REPORT

A 46-year-old man presented with visual loss in his left eye (OS) for 6 months. He did not refer any relevant systemic or ophthalmological history of diseases. His visual acuity (VA) was 20/20 in his right eye and 20/30 OS. Slit-lamp anterior segment examination was unremarkable. Dilated fundus examination revealed a yellowish choroidal mass within the superior temporal arcade with regular contour (Figure **1A**).

Fluorescein angiography (FA) images of the OS evidenced early patchy hyperfluorescence of the choroidal lesion with progressive staining with no obvious leakage (Figure **1B**). In addition, a temporal serous pigment epithelium detachment (PED) showed the typical progressive pooling with bright hyperfluorescence in the late frames (Figure **1B**). The FA was not definitive to rule out the presence of CNV over the choroidal lesion.

Ultrasound echography (B-mode) showed a highly hyperechogenic lesion with marked posterior acoustic shadowing consistent with a calcified choroidal lesion

^{*}Address correspondence to this author at the Department of Ophthalmology, University and Polytechnic Hospital La Fe, Valencia, Spain; E-mail: robertogallegopinazo@yahoo.es

Dolz-Marco et al.

(Figure **1C**). The magnetic resonance imaging examination did not revealed other significant findings.

The choroidal lesion was isoautofluorescent with central hyperautofluorescent dots consistent with lipofuscine deposition in the near infrared fundus autofluorescence (FAF) images (Figure **1D**); and an irregular hypo-hyperautofluorescent pattern was observed in the short-wave FAF (Figure **1E**). In both FAF images the serous retinal detachment was slightly hyperautofluorescent, being larger in near infrared FAF.

Spectral-domain optical coherence tomography (SD-OCT) scans showed the presence of a choroidal heterogeneous hyporeflective lesion in the superior temporal arcade associated with serous retinal detachment involving the fovea. Over the choroidal mass a focal vascular PED was observed consistent with a type 1 CNV and associated with cystoid macular edema without foveal involvement. Temporal to the choroidal osteoma a serous PED was also observed. Subfoveal choroidal thickness was measured on enhanced depth imaging scans resulting in 589 microns, which is significantly increased (Figure 1).

Due to the presence of intraretinal and subretinal fluid, and the suspicion of CNV, an intravitreal injection of ranibizumab was performed. One month later, VA improved to 20/25 with complete resolution of intraretinal fluid; and subtotal resolution of subretinal fluid was observed (Figure **2**).

DISCUSSION

Choroidal osteomas are benign intraocular tumors composed of mature cancellous bone that replace the full thickness of the choroid. They are frequently

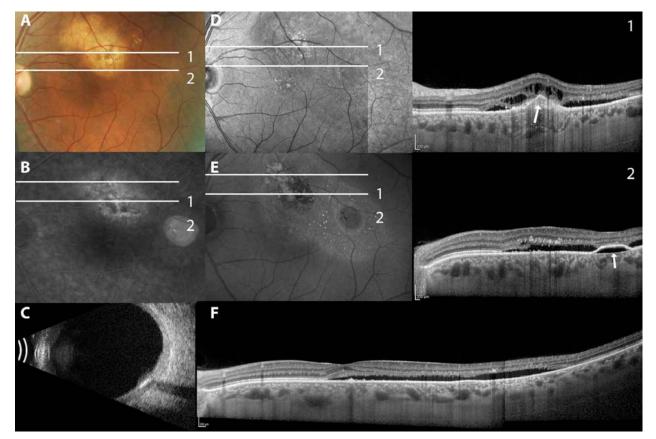


Figure 1: Color fundus photograph at baseline (**A**) showed a yellowish choroidal lesion with well-defined boundaries in the temporal superior arcade, and also a temporal pigment epithelium detachment was observed. The fluorescein angiography (FA) image (**B**) evidenced a choroidal hyperfluorescent lesion and a temporal serous PED. The B-mode ultrasound image (**C**) showed the typical hyperechogenic characteristics of choroidal osteoma with acoustic shadowing. The choroidal lesion appeared in the near-infrared fundus autofluorescence (FAF) (**D**) as isoautofluorescent with central mottling hyperautofluorescence, whereas the short-wavelength FAF (**E**) showed heterogeneous hypo-hyperautofluorescent characteristics. Serous retinal detachment was slightly hyperfautofluorescent in both near infrared and short wavelength FAF. The spectral-domain optical coherence tomography (SD-OCT) scan (**F**) showed the serous retinal detachment with foveal involvement. The SD-OCT scan over the choroidal lesion (1) showed an hyporeflective heterogeneous choroidal lesion with overlying vascular PED associated with cystoid macular edema. The SD-OCT scan over the temporal PED (2), showed a serous PED (arrow) with surrounding serous retinal detachment.

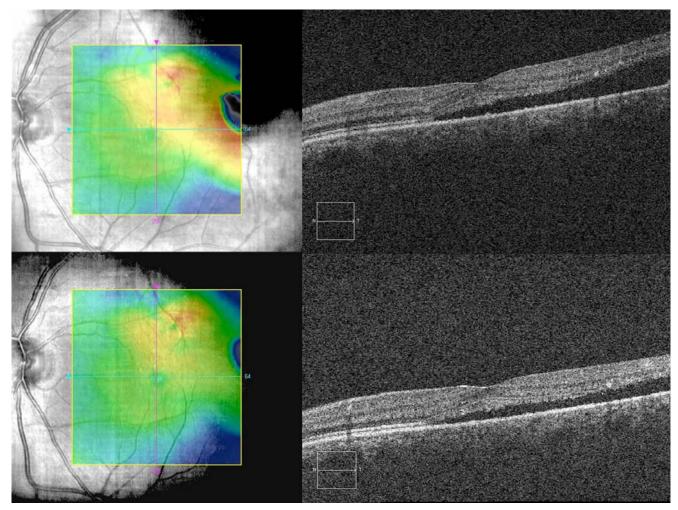


Figure 2: The color retinal thickness map and the spectral-domain optical coherence tomography at baseline (top images) showed important foveal involvement with serous retinal detachment that partially responded to intravitreal ranibizumab at 1-month follow-up (bottom images).

located in the juxtapapillary or macular regions. The etiology and pathogenesis of these lesions is unknown.

It has been reported a frequency of tumor growth and decalcification in nearly 50% of eyes by 10 years [13-14]. In addition one third of cases of choroidal osteoma may develop secondary CNV through a 10year follow-up [13]. Different therapeutic approaches have been reported for CNV secondary to choroidal osteoma with poor visual outcomes [2-7]. Recently, the use of intravitreal ranibizumab and bevacizumab has been reported achieving positive functional outcomes for CNV secondary to choroidal osteoma [8-12].

The multimodal imaging examination performed in our case showed an atypical case of choroidal osteoma complicated with overlying secondary CNV, and associated with chronic central serous choroidopathy (CSC). To the best of our knowledge there is no previous report about this association in the literature, although choroidal osteoma may masquerade CSC [15]; serous retinal detachment associated with choroidal osteoma can occur in the absence of CNV, being the most common cause of decreased vision in patients with this tumor [16-17].

We found typical characteristics of choroidal osteoma as a unilateral yellowish lesion with regular geographic boundaries. Echography showed marked shadowing and near infrared imaging showed an isoautofluorescent lesion [18]. The presence of cystoid macular edema overlying the choroidal tumor was suggestive of type 1 CNV. Finally, the presence of a large serous retinal detachment with the presence of a temporal serous PED and a subfoveal thickened choroid was consistent with CSC.

Although there is no established therapeutic approach for choroidal osteoma, our positive results with ranibizumab intravitreal treatment are consistent with previous reports. We present an atypical case of choroidal osteoma with CNV and CSC successfully treated with intravitreal ranibizumab. This antiangiogenic agent may be useful for these two entities, therefore improving the visual outcomes and restoring the normal retinal architecture.

REFERENCES

- Gass JD, Guerry RK, Jack RL, Harris G. Choroidal osteoma. Arch Ophthalmol 1978; 96: 428-35. <u>http://dx.doi.org/10.1001/archopht.1978.03910050204002</u>
- [2] Rose SJ, Burke JF, Brockhurst RJ. Argon laser photoablation of a choroidal osteoma. Retina 1991; 11: 224-8. http://dx.doi.org/10.1097/00006982-199111020-00007
- [3] Hoffman ME, Sorr EM. Photocoagulation of subretinal neovascularization associated with choroidal osteoma. Arch Ophthalmol 1987; 105: 99-9. <u>http://dx.doi.org/10.1001/archopht.1987.01060070142045</u>
- [4] Singh AD, Talbot JF, Rundle PA, Rennie IG. Choroidal neovascularization secondary to choroidal osteoma: successful treatment with photodynamic therapy. Eye 2005; 19: 482-4. http://dx.doi.org/10.1038/sj.eye.6701475
- [5] Blaise P, Duchateau E, Comhaire Y, Rakic JM. Improvement of visual acuity after photodynamic therapy for choroidal neovascularization in choroidal osteoma. Acta Ophthalmol Scand 2005; 83: 515-6. <u>http://dx.doi.org/10.1111/j.1600-0420.2005.00479.x</u>
- [6] Sharma S, Sribhargava N, Shanmugam MP. Choroidal neovascular membrane associated with choroidal osteoma (CO) treated with trans-pupillary thermo therapy. Indian J Ophthalmol 2004; 52: 329-30.
- [7] Shukla D, Tanawade RG, Ramasamy K. Transpupillary thermotherapy for subfoveal choroidal neovascular membrane in choroidal osteoma. Eye 2006; 20: 845-7. <u>http://dx.doi.org/10.1038/sj.eye.6702016</u>
- [8] Ahmadieh H, Vafi N. Dramatic response of choroidal neovascularization associated with choroidal osteoma to the intravitreal injection of bevacizumab (Avastin). Graefes Arch Clin Exp Ophthalmol 2007; 245: 1731-3. <u>http://dx.doi.org/10.1007/s00417-007-0636-z</u>

Received on 20-06-2013

Accepted on 21-06-2013

Published on 16-08-2013

DOI: http://dx.doi.org/10.12974/2309-6136.2013.01.01.9

© 2013 Dolz-Marco et al.; Licensee Savvy Science Publisher.

This is an open access article licensed under the terms of the Creative Commons Attribution Non-Commercial License (<u>http://creativecommons.org/licenses/by-nc/3.0/</u>) which permits unrestricted, non-commercial use, distribution and reproduction in any medium, provided the work is properly cited.

[9] Song WK, Koh HJ, Kwon OW, Byeon SH, Lee SC. Intravitreal bevacizumab for choroidal neovascularization secondary to choroidal osteoma. Acta Ophthalmol 2009; 87: 100-1.

http://dx.doi.org/10.1111/j.1600-0420.2007.01136.x

- [10] Pandey N, Guruprasad A. Choroidal osteoma with choroidal neovascular membrane: successful treatment with intravitreal bevacizumab. Clin Ophthalmol 2010; 4: 1081-4. http://dx.doi.org/10.2147/OPTH.S13730
- [11] Song MH, Roh YJ. Intravitreal ranibizumab in a patient with choroidal neovascularization secondary to choroidal osteoma. Eye (Lond) 2009; 23: 1745-6. <u>http://dx.doi.org/10.1038/eye.2008.313</u>
- [12] HY Wu Z, YY Wong M, YY Lai T. Long-Term Follow-Up of Intravitreal Ranibizumab for the Treatment of Choroidal Neovascularization due to Choroidal Osteoma. Case Rep Ophthalmol 2012; 3: 200-4. <u>http://dx.doi.org/10.1159/000339624</u>
- [13] Shields CL, Sun H, Demirci H, Shields J. Factors predictive of tumor growth, tumor decalcification, choroidal neovascularization, and visual outcome in 74 eyes with choroidal osteoma. Arch Ophthalmol 2005; 123: 1658-66. http://dx.doi.org/10.1001/archopht.123.12.1658
- [14] Aylward GW, Chang TS, Pautler SE, Gass JD. A long-term follow-up of choroidal osteoma. Arch Ophthalmol 1998; 116: 1337-41. http://dx.doi.org/10.1001/archopht.116.10.1337
- [15] Lee YC, Sheu SJ. Choroidal osteoma masquerading as central serous chorioretinopathy. Kaohsiung J Med Sci 2002; 18: 408-11.
- [16] Song JH, Bae JH, Rho MI, Lee SC. Intravitreal bevacizumab in the management of subretinal fluid associated with choroidal osteoma. Retina 2010; 30: 945-51. <u>http://dx.doi.org/10.1097/IAE.ob013e3181c720ca</u>
- [17] Aylward GW, Chang TS, Pautler SE, Gass JD. A long-term follow-up of choroidal osteoma. Arch Ophthalmol 1998; 116: 1337-41. http://dx.doi.org/10.1001/archopht.116.10.1337
- [18] Sisk RA, Riemann CD, Petersen MR, Foster RE, Miller DM, Murray TG, Augsburger JJ. Fundus autofluorescence findings of choroidal osteoma. Retina 2013; 33(1): 97-104. http://dx.doi.org/10.1097/IAE.0b013e31825c1cde