

## Sequential imaging of a case of choroidal osteoma using swept-source OCT and optical coherence tomography angiography: A 4-year follow-up study

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A 33-year-old gentleman was presented with metamorphopsia in the left eye due to choroidal osteoma (CO) complicated by choroidal neovascular membrane (CNVM). Optical coherence

tomography angiography (OCTA) proved to be a valuable, noninvasive tool in monitoring treatment response of CNVM. The tumor subsequently underwent decalcification over a period of 4 years. In addition, SS-OCT scans were instrumental in documenting the natural course of the tumor and focal choroidal excavations (FCE), which were found in correspondence with tumor decalcification. Close follow-up is warranted in FCE, secondary to decalcification of CO, as CNVM has been documented to occur on the slope or bottom of eyes with FCE.

**Key words:** Choroidal neovascular membrane, choroidal osteoma, decalcification, focal choroidal excavation, swept-source OCT, swept-source OCTA

Choroidal osteoma (CO) is a rare benign ossifying tumor, composed of mature bone replacing full thickness of the choroid.<sup>[1,2]</sup> It classically presents an orange-yellow subretinal mass in the juxtapapillary or macular region.<sup>[1,2]</sup> Visual impairment in CO is attributed to various factors such as choroidal neovascularization (CNVM), tumor decalcification, serous retinal detachment, and hemorrhages.<sup>[1,2]</sup> The tumor undergoes decalcification or involution, either spontaneously or secondary to treatment and is attributed to the development

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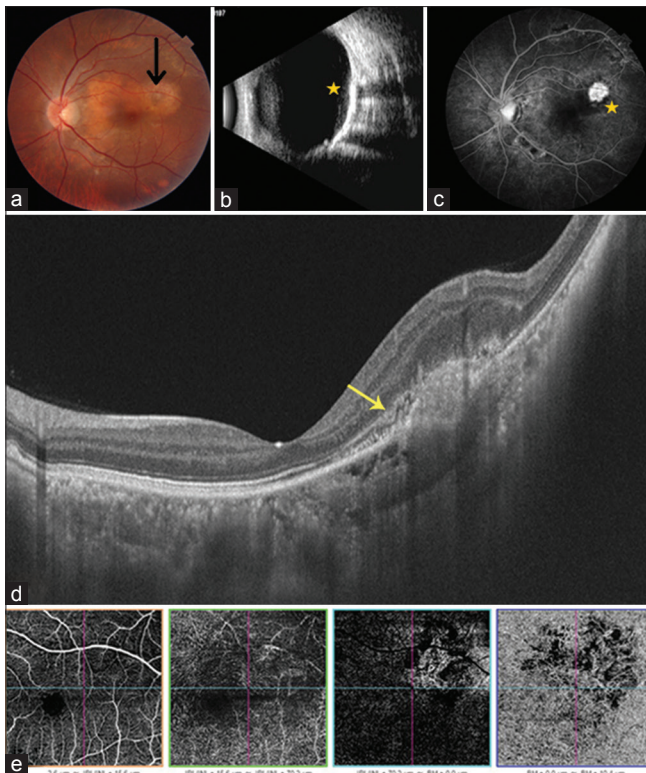
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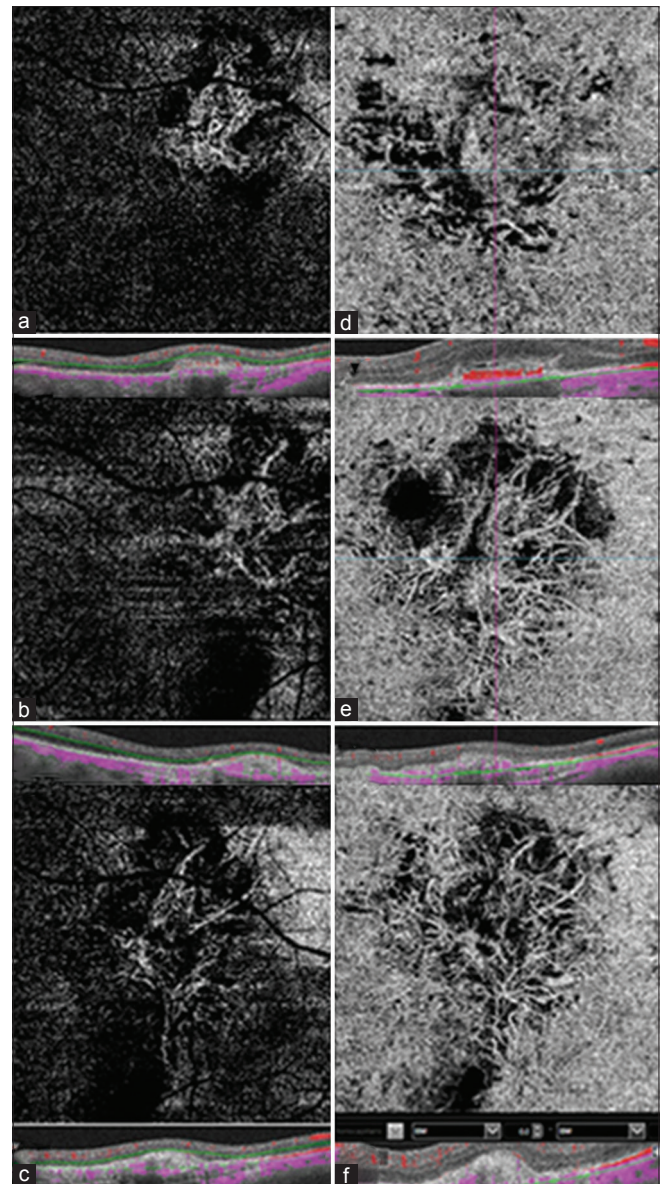


**Figure 1:** (a) Fundus photograph of the OS showing focal orange-yellow tumor with depigmentation (arrow). (b) Ultrasonography confirming CO (star) with high acoustic reflectivity and corresponding after-shadow. (c) FFA (late phase) showing hyperfluorescent type 2 CNVM (star) (d) SS-OCT (oblique scan) showing hyper-reflective lesion with underlying hyporeflexivity suggestive of CO. Disruption of RPE layer with SRF suggestive of CNVM (arrow). (e) SS-OCTA (automatic segmentation) showing fine, a thin network of vessels (CNVM) in deep retinal and choriocapillaris layer at the level of the tumor

of focal choroidal excavation (FCE) and CNVM in CO.<sup>[1-3]</sup> We hereby present a case report of a gentleman who was presented with unilateral CO and the natural course of the tumor had been illustrated in a unique manner through multimodal imaging such as fluorescein angiography (FA), swept-source OCT (SS-OCT), and OCT angiography (OCT-A, DRI OCT Triton, Topcon, Inc., Tokyo, Japan).

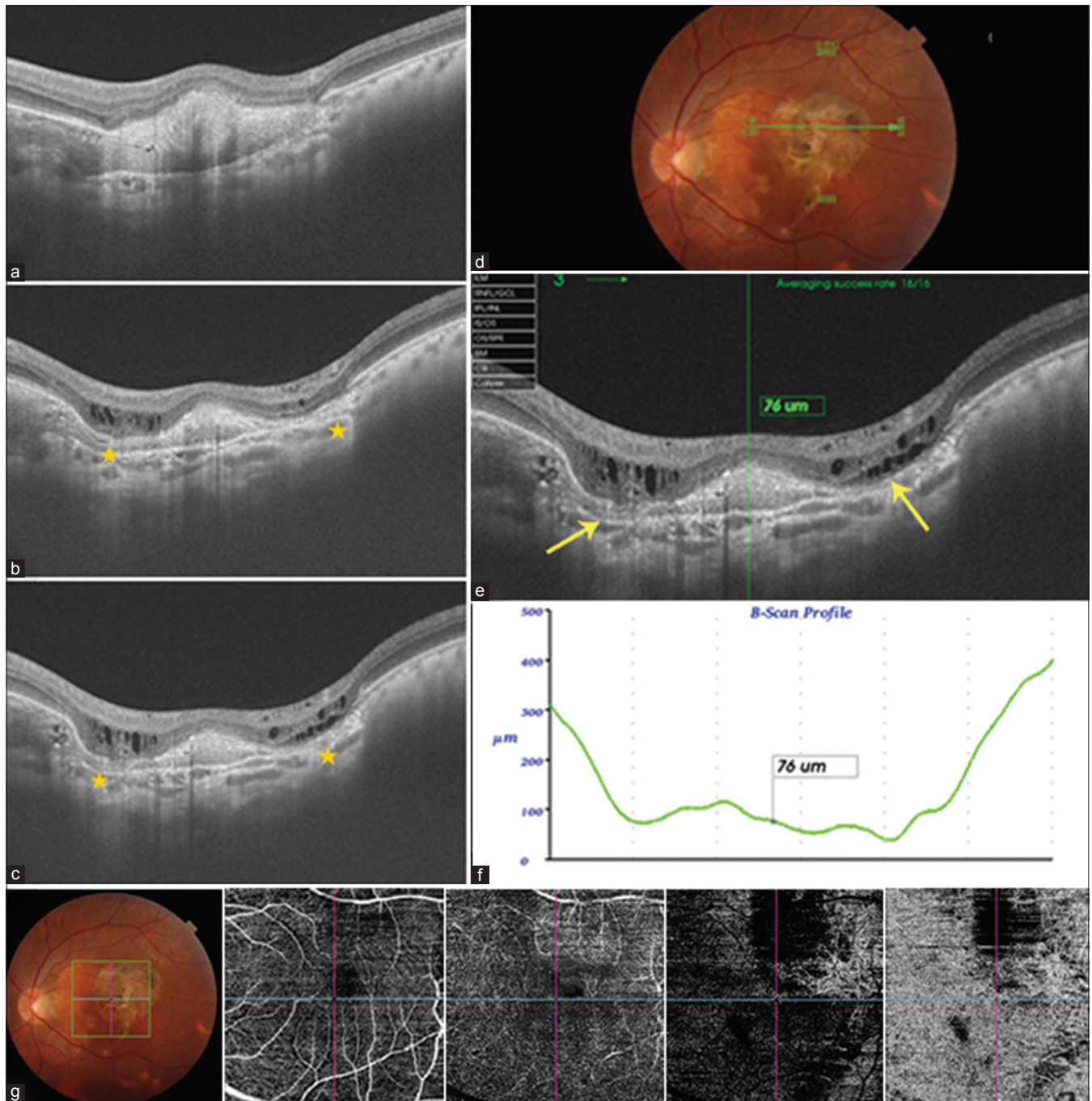
**Case Report**

A 33-year-old gentleman was presented with metamorphopsia in the left eye (LE) for 1 week. His best-corrected visual acuity (BCVA) was 20/20 in the right eye (RE) and 20/60 in LE. Anterior segment was unremarkable in both eyes. On LE posterior segment evaluation, a focal, orange-yellow, submacular lesion of 1 disc diameter (DD) size was noted 0.5 DD away from the fovea, temporally. Overlying area of depigmentation extended from the lesion to the temporal optic disc margin spanning an area of 3 DD and superior aspect of fovea involvement was eminent [Fig. 1a]. Hemorrhages and subretinal fluid surrounding the lesion indicated the presence of an underlying CNVM. Ultrasonography of LE found a hyper-reflective lesion with an acoustic shadow consistent with the lesion ratifying the diagnosis of CO [Fig. 1b]. SS-OCT in the LE revealed a subretinal hyper-reflective mass, temporal to



**Figure 2:** OCTA follow-up of CNVM showing treatment response with 3 doses of intravitreal ranibizumab given on a monthly basis. Each row represents a monthly follow-up post injection. CNVM noted in the deep retinal (a-c) and choriocapillaris layer (d-f) as a well-defined, cartwheel-patterned lesion at the level of the tumor, regressed and collapsed to lose its pattern with each injection

the fovea, with underlying hypo reflectivity in correspondence to CO. Disruption of IS-OS junction and retinal pigment layer (RPE) layer along with a collection of subretinal fluid (SRF) was observed overlying the tumor, corroborating the presence of an associated CNVM [Fig. 1d]. Fluorescein angiography (FA) revealed a type 2 CNVM characterized by early, lacy hyper fluorescence, and progressive leakage and was delineated well on OCTA [Fig. 1c]. A fine, thin network of vessels in outer retinal and choriocapillaris layer, noted on OCTA at the level of the CO was consistent with the CNVM [Fig. 1e]. Three doses of ranibizumab intravitreal injections were given on a monthly basis. CNVM regressed and collapsed with treatment and the same was monitored noninvasively by OCTA [Fig. 2a-f].



**Figure 3:** Locations of all scans correspond to the horizontal scan depicted in figure (d). (a-c) Serial SS-OCT scans of periodic follow-up post CNVM treatment showing progressive decalcification of tumor and development of FCE (stars). Each row represents a yearly visit. Final visit (fourth year) Fundus photograph (d) and choroidal thickness profile (e and f) with y-axis depicting the choroidal thickness in microns in the region traversed by the horizontal line scan (x-axis) in (d). Note FCE (e, arrow) and chorioretinal atrophy (f) beneath the decalcified tumor. (g) OCTA (final visit) CNVM remains regressed

One month after the final injection, BCVA improved to 20/20 in LE and was unaltered during the 4-year follow-up period [Fig. 3g]. No active intervention was warranted during the follow-up period. The CO underwent a natural progression of decalcification leading to flattening of the lesion with RPE thinning and scarring, underlying choroidal vessels [Fig. 3d]. Focal choroidal excavations (FCE) characterized by one or more focal areas of choroidal depression were found in

correspondence with the tumor decalcification on serial SS-OCT scans [Fig. 3a-c]. Chorioretinal atrophy was noted underneath the FCE and decalcified tumor [Fig. 3e and f]. Visual acuity remained stable at the final visit.

**Discussion**

Our patient presented unilateral CO complicated with CNVM which was treated with 3 doses of intravitreal ranibizumab

given on a monthly basis. Regression of CNVM was imaged by OCTA in a dye less, noninvasive manner and was ideal for monitoring treatment response of neovascularization associated with CO. The extent of neovascularization was well demarcated by OCTA unlike FA, where the leak from the new vessels often obscures the details. Following treatment for CNVM, our patient was merely observed for 4 years. Serial SS-OCT scans were instrumental in our follow up and provided detailed anatomical information on retinal and choroidal changes. The tumor underwent decalcification and focal choroidal excavation (FCE) of conforming type were noted at this point. FCE is a relatively recent OCT finding characterized by one or more focal areas of choroidal depression without any evidence of scleral ectasia or staphyloma.<sup>[4,5]</sup> Etiology of FCE remains unclear and has been classified by Margolis *et al.* into conforming lesions, in which the overlying retina is close to the retinal pigment epithelium (RPE), and nonconforming lesions, in which a hyporeflective space is visible between the retina and RPE on OCT scans.<sup>[5]</sup> CO and FCE in the proximity of CNV, as seen in our patient, has been described in two patients by Pierro *et al.* and decalcification of choroidal osteoma was cited as the common pathogenic pathway for the development of FCE and CNVM in choroidal osteoma.<sup>(3)</sup> Similarly, Olguin-Manriquez *et al.*, in their study of 16 eyes of 11 Hispanic patients with CO observed two eyes with CNV in the proximity of FCE and one eye with FCE in the boundaries of the osteoma and normal choroidal tissue. The authors conjectured that FCE might represent distinct stages of focal decalcification of the tumor and the depression may grow over time.<sup>[6]</sup> FCE observed in our patient correlates with their description as it was first noted at the onset of tumor involution and enlarged with progressive tumor decalcification. Though tumors with decalcification show no further growth, neovascularization occurs prior to or at the same time as decalcification and since CNVM has been documented to occur on the slope or the bottom of eyes with FCE in other pathologies, a closer follow-up is necessary in patients developing FCE secondary to CO decalcification.<sup>[1,7]</sup>

## Conclusion

In summary, to our knowledge, this is the first prospective case study of the natural course of CO through multimodal imaging.

SS-OCT and OCT-A are insightful in monitoring complicated CO.

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## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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