

Rapid enlargement of choroidal osteoma in an adult patient

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Dear Editor,

We present the first adult patient with rapid enlargement of choroidal osteoma. Choroidal osteoma is a rare benign tumor characterized by mature cancellous bone in the choroid^[1]. The usual progress of the tumor is not rapid. Herein, we report that choroidal osteoma enlarged rapidly in an adult patient with Rosai-Dorfman disease (RDD). This study adhered to the tenets of the Declaration of Helsinki, and the written informed consent was obtained from the patient (YKEC-KT-2022-024-P002).

A 34-year-old man presented with a 1-month history of metamorphopsia of the left eye. His best-corrected visual acuity was 20/20 in the right eye and 20/25 in the left eye. Fundus examination revealed bilateral orange-yellow choroidal lesions at the posterior pole (Figure 1A, 1B). On optical coherence tomography angiography, there was subretinal fluid involving the left fovea. B-scan ultrasonography demonstrated highly reflective choroidal lesions with posterior shadowing (Figure 1C, 1D). The maximal diameter of osteomas of the right eye and left eye were 3.8 mm and 8.6 mm, respectively. Serum C-reactive protein level was markedly elevated. Other systemic examination findings were normal. And cranial computed tomography scan was normal in December 2020.

A diagnosis of bilateral choroidal osteoma with (left-eye) subretinal fluid was made. The patient responded to three injections of ranibizumab with recovery of visual acuity and resolution of subretinal fluid 1mo after the procedure. At this time, his best-corrected visual acuity was 20/20 in both eyes. Unfortunately, the subretinal fluid reappeared 2mo later. Best-corrected visual acuity was 20/20 in the right eye and 20/25 in the left eye. Fundus examination (Figure 1E, 1F), and B-scan ultrasonography (Figure 1G, 1H) revealed rapid growth of the lesion during these 6mo. The maximal diameter of osteomas of the right eye and left eye were 6.7 mm and 11.9 mm, respectively. Horizontal enhanced depth imaging optical coherence tomography of choroidal osteoma revealed slightly elevated choroidal surface with intralesional horizontal lines and horizontal tubules (Figure 2A, 2B). At last follow-up, cranial magnetic resonance imaging (Figure 3A) showed a mass lesion and a preoperative diagnosis of meningioma was made in August 2021. The patient underwent resection of the intracranial mass. However, biopsy of the surgical specimen revealed characteristic histiocytes showing emperipolesis (Figure 3B). The diagnosis of intracranial RDD was made.

It is interesting that this case report highlights coexistence of two rare diseases in a single patient. Such an association is likely to be coincidental and has not been described before.

RDD is a rare systemic pseudolymphomatous disorder^[2]. Uveal involvement in RDD is rare. B-scan ultrasonography of uveal RDD revealed moderately low internal reflectivity consistent with uveal melanoma^[3]. However, B-scan ultrasonography of choroidal osteoma revealed highly reflective choroidal lesions with posterior shadowing. Besides, enhanced depth imaging optical coherence tomography of choroidal osteoma revealed intralesional horizontal lines. It helped to differentiate choroidal osteoma from other choroidal tumors^[4]. Therefore, this case reported coexistence of two rare diseases in a single patient.

Choroidal osteoma commonly manifests as a unilateral orange-yellow choroidal lesion in young adult females. It usually grows very slowly. Eyes with choroidal osteoma growth showed an increase in mean basal diameter of 0.37 mm per year^[5]. Mizota *et al*^[6] reported that choroidal osteoma enlarged rapidly in a 3-year-old girl. However, we report that choroidal

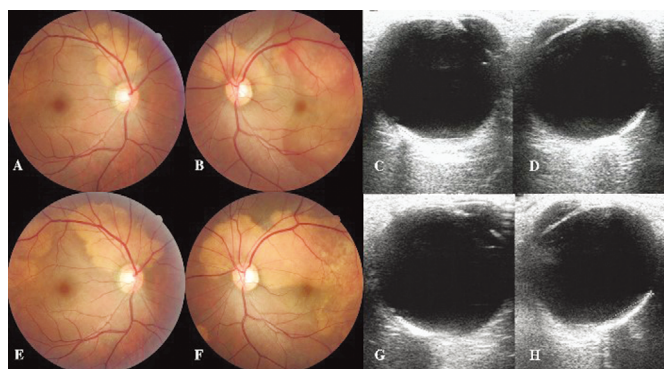


Figure 1 Fundus photographs and B-scan ultrasonography of right eye (A, C) and left eye (B, D) in December 2020, right eye (E, G) and left eye (F, H) in June 2021.

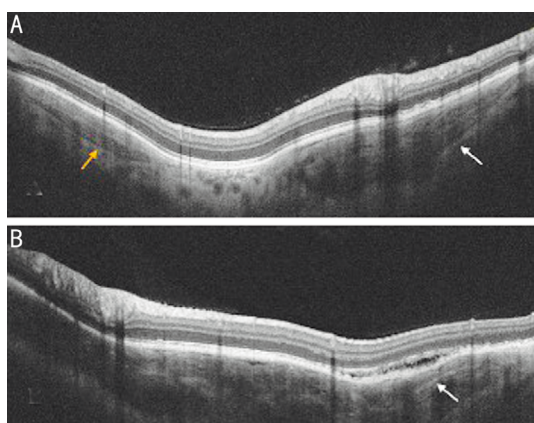


Figure 2 Horizontal enhanced depth imaging optical coherence tomography of choroidal osteoma through superior to disc revealed slightly elevated choroidal surface with intralesional horizontal lines (white arrow) and horizontal tubules (yellow arrow) in the right eye (A), and intralesional horizontal lines (white arrow) in the left eye (B) in June 2021.

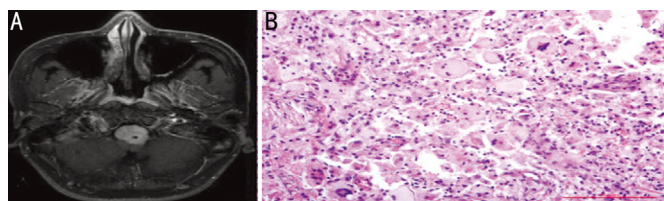


Figure 3 Cranial magnetic resonance imaging (A) showed a mass lesion preoperatively (August 2021) and biopsy of the surgical specimen (B) revealed characteristic histiocytes showing emperipolesis. Hematoxylin and eosin staining, original magnification $\times 200$.

osteoma enlarged rapidly in the adult patient. The average increase in the maximal diameter of choroidal osteoma was 6.2 mm per year. Besides, we found that serum C-reactive protein level was markedly elevated. Systemic studies of RDD demonstrated an elevated C-reactive protein^[3]. The severe inflammation may be correlated with the rapid enlargement of choroidal osteoma^[7]. However, the relationship between the growth of choroidal osteoma and the progression of RDD requires further investigation.

Subretinal fluid secondary to choroidal osteoma is a major cause of visual impairment. Antivascular endothelial growth factor treatments could decrease choroidal vascular permeability by inhibiting vascular endothelial growth factor, decrease the subretinal fluid, and subsequently improve the patients' visual acuity^[8-10]. In our case, the patient received an intravitreal injection of ranibizumab resulting in decrease of subretinal fluid, and improved visual acuity. However, the subretinal fluid reappeared. Antivascular endothelial growth factor treatments could not change the patient outcome^[10]. This case reported coexistence of two rare diseases in a single patient. The rapid growth of choroidal osteoma may be correlated with the progression of RDD.

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