Choroidal Osteoma in Oriental Patients

SUKHUMA WARRASAK, MD*,***,
ATAYA EUSWAS, MD*,***,
JIRAPORN LAOTHAMATAS, MD****
CHANET SUVARNAMANI, MD**,
VICHAI SUMETPIMOLCHAI, MD***,

Abstract

Background: Choroidal osteoma is a rare tumour of the choroid. This is the first report of cases of choroidal osteoma in Thai patients.

Objective: To report the clinical characteristics, imaging findings and long-term follow-up of choroidal osteoma in four Oriental patients.

Method: Four cases of choroidal osteoma were observed for 5 years or more.

Results: All patients were young female patients whose ages ranged from 24 to 37 years. Three were unilateral and one was bilateral. The tumors were located at the juxtapapillary and macular area with overlying serous retinal detachment. Two patients had previous thyroid diseases and one was pregnant when the tumors were diagnosed. Osteoma did not develop in the vicinity of posterior staphyloma of high myopic eyes. Echography showed acoustic features of a plano-convex sonically dense lesion with high reflectivity echoes which persisted despite lower system sensitivity. None had subretinal neovascularization. Subretinal fluid disappeared spontaneously within one to 14 months in three patients. Gradual growth of the tumor in a pseudopodium manner developed from two to six years after initial examination. Decalcification occurred spontaneously or after laser ablation.

Conclusions: The authors presented four Oriental patients with choroidal osteoma who were observed for at least 5 years. Echography is the best method for identifying this lesion and has unique acoustic features. Subretinal fluid can be seen in the absence of subretinal neovascularization and resorbs spontaneously. Decalcification occurred as a natural process or after laser ablative treatment. Hormonal changes may implicate the development of this tumor.

Key word : Choroidal Osteoma, Oriental Patients, Echography, MRI

WARRASAK S, SUVARNAMANI C, EUSWAS A, SUMETPIMOLCHAI V, LAOTHAMATAS J
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* Department of Ophthalmology, Faculty of Medicine, Ramathibodi Hospital,
** Pramongkutklao Hospital, Bangkok 10110,
*** Rutnin Eye Hospital, Bangkok 10110,
**** Department of Radiology, Faculty of Medicine, Ramathibodi Hospital, Mahidol University, Bangkok 10400, Thailand.

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Choroidal osteoma is a rare juxtapapillary and macular benign ossifying tumor of the choroid, typically unilateral, affecting young female patients. Gass et al first reported this tumor in a group of four patients in 1978 and since then others have subsequently reported some other clinical and investigative aspects. However, most of the cases mainly involved Caucasians as well as a few blacks while those from oriental descent were rarely reported in the literature(1-15).

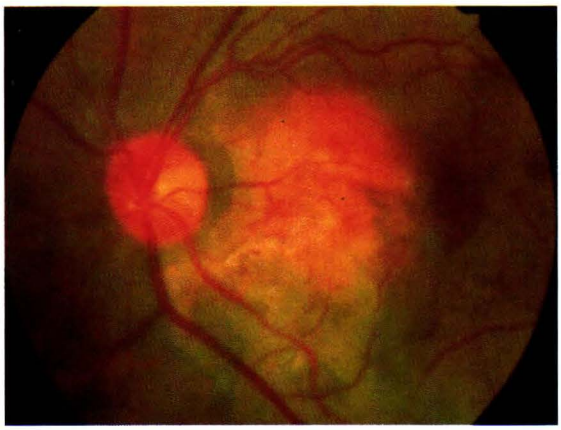


Fig. 1. Patient 1. Fundus photography of the left eye shows juxtapapillary yellow white placoid choroidal tumor involving papillomacular bundle and some part of fovea on the initial examination.

The authors present the outcome of clinical characteristics, echographic findings (Allergan Humphrey A/B scan, 835 Rev A9, San Leandro, USA), computerized scan (CT scan, 9800 quick, General Electrics, Milwaukee, USA) or magnetic resonance imaging (MRI,1.5 Tesla Signa, General Electrics, Milwaukee, USA) findings and, long-term follow-up of choroidal osteoma in four Oriental patients who attended the retina clinic on a tertiary eye care referral basis. This is the first report of cases of choroidal osteoma in Thai patients over the past 20 years. Echographic findings described in this report are quite distinct from other simulating choroidal lesions.

CASE REPORTS

Case 1:

In December 1990, a 34-year-old female Filipino singer reported blurred vision in the left eye for two weeks' duration. The patient had an allergic background but otherwise was healthy. Ocular examination revealed distant visual acuity of 20/20 in both eyes. The anterior segments were unremarkable. Dilated fundus examination disclosed juxtapapillary yellow white placoid choroidal tumor involving the papillomacular bundle and partly affecting the macula in the left eye (Fig. 1). Serous retinal detachment was seen overlying the tumor. Ultrasonography of the left eye demonstrated high reflectivity lesion with a flat top, plano-convex configuration which persisted despite reduced system sensitivity (Fig. 2A-

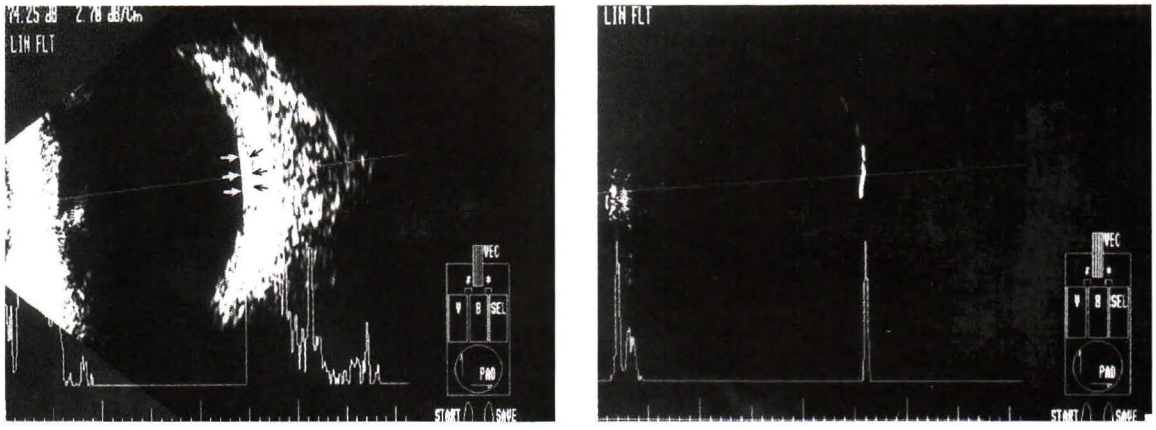


Fig. 2. Patient 1. Ultrasonography of the left eye A) high reflectivity echogenic mass (arrows). Note the flat-top bone density of the lesion and a plano-convex configuration. B) high reflectivity persisted even at reduced system sensitivity.

2B). Computerized scan (CT scan) of the orbit showed the presence of calcification (Fig. 3). MRI, performed with orbital surface coil and after gadolinium administration (Magnevist®, Schering AG, Germany) revealed a small nonenhancing hyposignal T_1 plaque with focal thinning of the involved choroid lateral to the optic nerve insertion but not seen in T2W image. One month later, subretinal fluid (SRF) subsided. Physical examination and laboratory investigations included T3, freeT4, total T4, TSH, serum calcium, serum phosphorus and alkaline phosphatase were all normal. In December 1995, subretinal fluid (SRF) leaked into the inferior retina and gradually disappeared over a period of two months. Intravenous

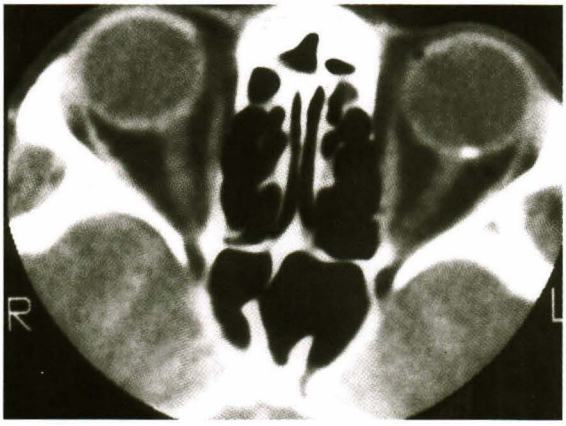


Fig. 3. **Patient 1. Enhanced axial CT scan of the left orbit shows a small calcified plaque just lateral to the optic nerve insertion.**

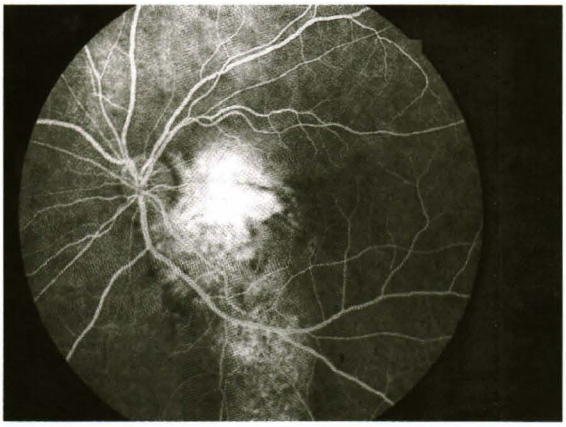


Fig. 4. **Patient 1. Fluorescein angiogram in the left eye shows irregular hyperfluorescence of the lesion. Note hypofluorescent streaks corresponding to furrows in the lesion probably represented intertrabecular spaces of the bone at the level of choroid. Other hyperfluorescence below inferior vascular arcades represented RPE alterations in serous retinal detachment of prolonged duration.**

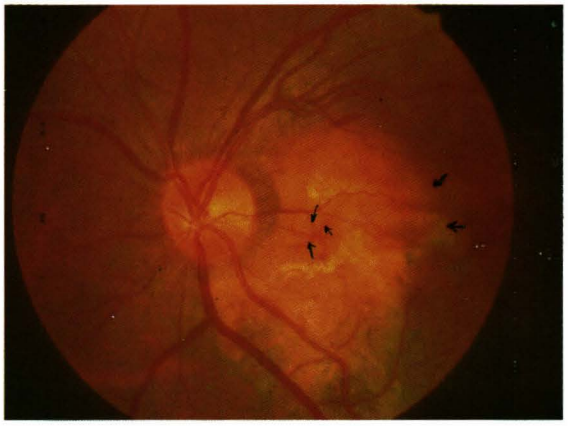


Fig. 5. **Patient 1. Fundus photography of the left eye shows slight enlargement of the lesion in a pseudopod manner (two arrows). Some large intratumoral vessels were further seen at 5 years of follow-up (three arrows).**

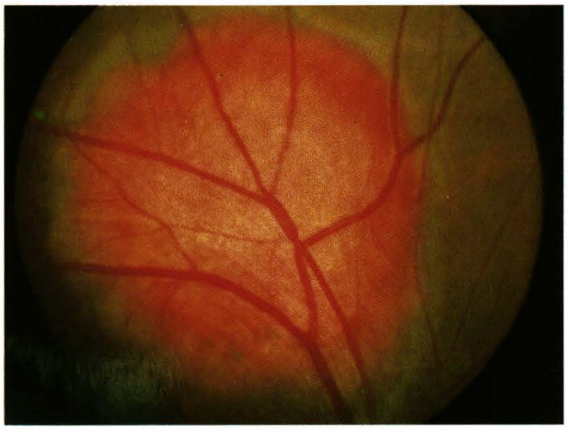


Fig. 6. **Patient 2. Fundus photography of the right eye shows a well circumscribed orange red choroidal mass with associated serous retinal detachment.**

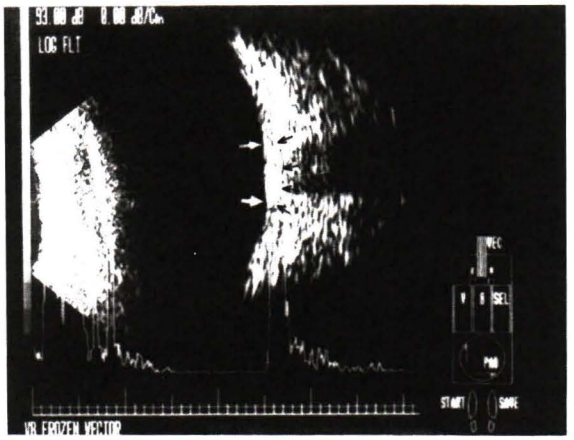


Fig. 7. Patient 2. Ultrasonography of the right eye shows the unique characteristic of a plano-convex configuration mass with high reflectivity echoes. Arrows outlined boundary of ossification within choroid and normal choroid which created a plano-convex configuration.

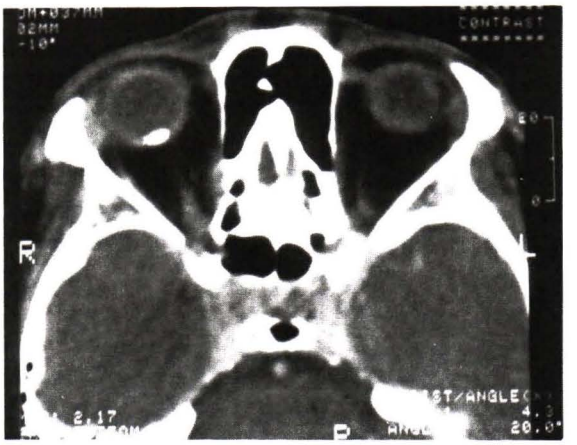


Fig. 8. Patient 2. Enhanced axial CT scan of the orbit demonstrates a moderate calcific choroidal lesion in the right eye just superolateral to the optic nerve insertion.

fluorescein angiography (IVFA) (Fluorescite™, Alcon Laboratories, Inc., Fortworth, USA) showed irregular hyperfluorescence of the lesion and late staining. There were hypofluorescent streaks which corresponded with furrows in the lesion (Fig. 4). In September 1996, her visual acuity in the left eye deteriorated to 20/100. On dilated fundus examination, the lesion showed slight enlargement in a pseudopod manner and some large intratumoral vessels were additionally seen (Fig. 5).

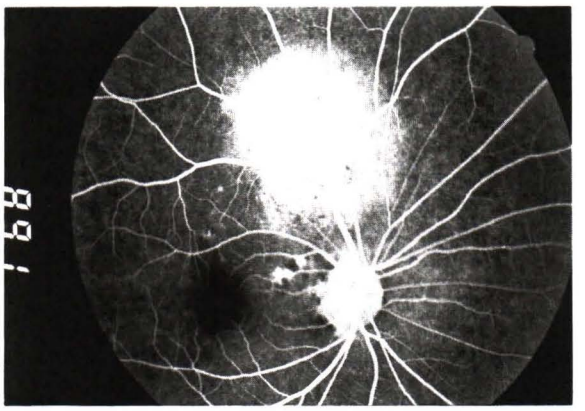


Fig. 9. Patient 2. Fluorescein angiography of the right eye shows irregular hyperfluorescence with late staining.

Case 2:

A 24-year-old Thai female employee presented in May 1991 with a one-week history of blurring central vision in the right eye. She had had a gastric ulcer for several years. The family history was noncontributory. Her visual acuity was corrected to 20/20 with +1.00 diopter sphere in the right eye and 20/20 without correction in the left. The right fundus showed a well-defined orange choroidal mass above the optic disc with associated serous detachment of the neurosensory retina (Fig. 6). SRF subsided in two months. Ultrasonography (Fig. 7) demonstrated a dense echogenic mass with a plano-convex configuration. A scan echography showed high reflectivity which still persisted at reduced sensitivity. Axial CT scan of the orbit (Fig. 8) demonstrated a moderate calcific plaque along the posterior choroid around the optic nerve insertion. MRI was not performed in this case due to the patient's refusal. IVFA (Fig. 9) showed irregular hyperfluorescence with late staining. Other work-up including complete blood count (CBC), urinalysis (UA), serum calcium, and thyroid function test was normal. In March 1997, the lesion showed no overlying serous detachment of sensory retina (SDSR). Her vision remained 20/20 without correction.

Case 3:

A 37 -year-old Chinese patient presented in June 1992 with a history of blurring central vision in the left eye during her third trimester of pregnancy. Her best corrected visual acuity was 20/20 in the right eye and 20/100 in the left. Her past medical history was noncontributory. Dilated fundus examination revealed a yellow white placoid juxtapapillary choroidal tumor located superotemporally and involved the papillomacular bundle and the macula in the left eye. Ultrasonography revealed a sonically dense lesion with a plano-convex configuration (Fig. 10). B-Scan echography showed high reflectivity echoes which persisted at lower system sensitivity. Physical examination and laboratory tests including CBC, serum

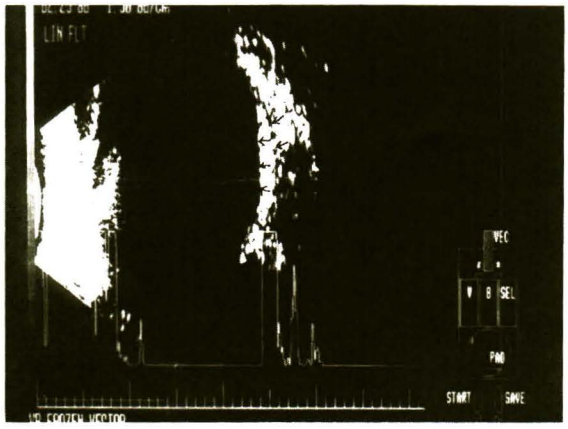


Fig. 10. Patient 3. Ultrasonography of the lesion in the left eye shows the characteristics of a high reflectivity echogenic mass with a plano-convex configuration (arrows).

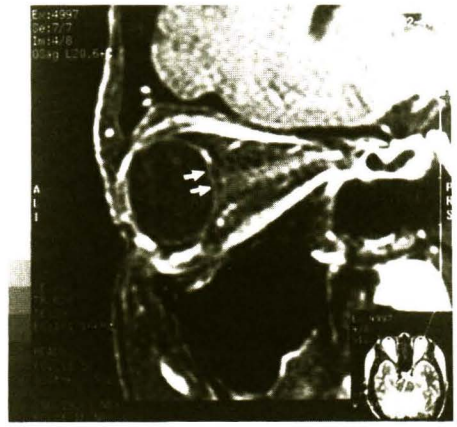


Fig. 12. Patient 3. Oblique sagittal T1W's postgadolinium MRI of the left orbit shows a small nonenhancing hyposignal T1 plaque (TR/TE/NEX=620/33/3) with thinning of the involved choroid adjacent to the optic nerve insertion (white arrows).

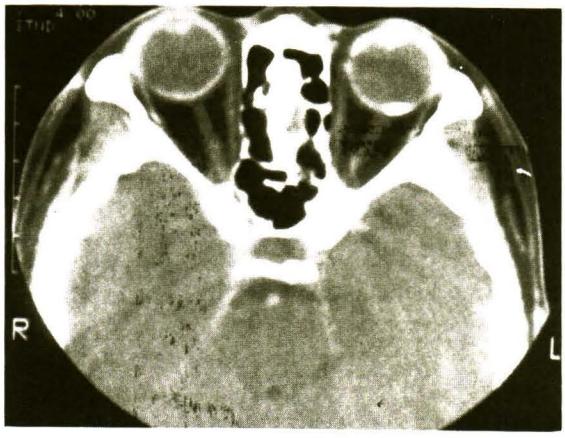


Fig. 11. Patient 3. Enhanced axial CT scan of the left orbit demonstrates a moderate calcific choroidal plaque posterolateral to the left optic nerve insertion.

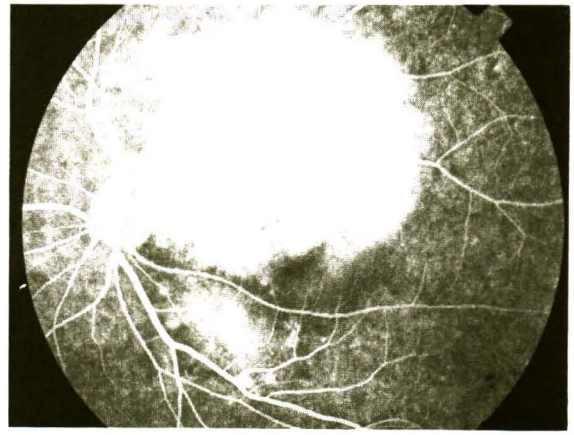


Fig. 13. Patient 3. Fluorescein angiography of the left eye demonstrates late intense staining of the lesion.

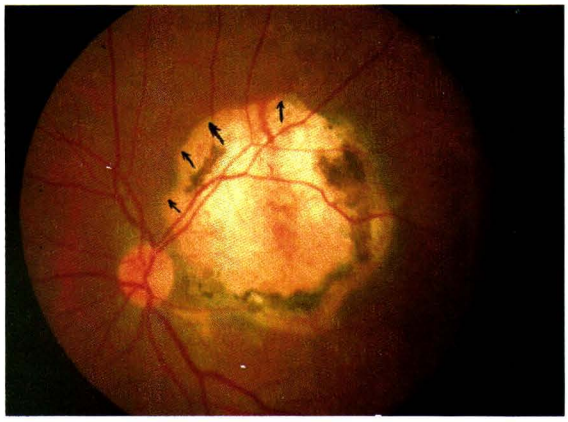


Fig. 14. **Patient 3. Fundus photography of the left eye shows a gradual growth of the lesion in a pseudopod manner (arrows) at one year post conception. Note thinning area along the lesion border and hyperpigmented RPE overlying the tumor following laser treatment.**

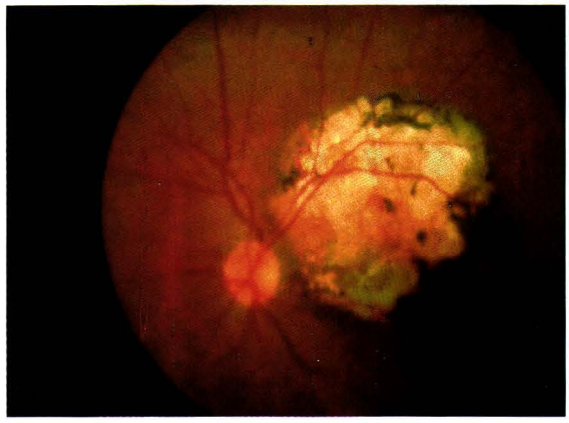
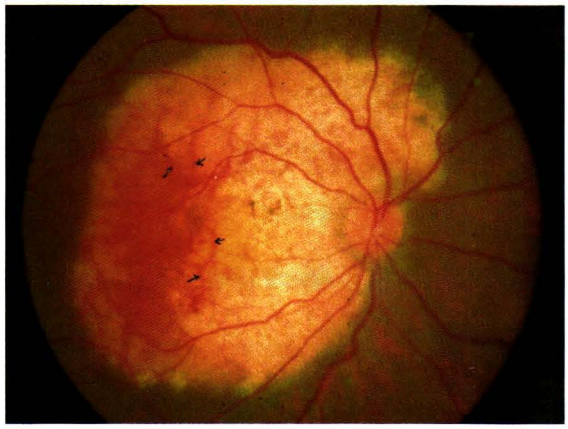


Fig. 15. **Patient 3. Fundus photography of the left eye shows decalcification of the lesion which occurred in the area of previous laser treatment and also in an area without laser treatment.**



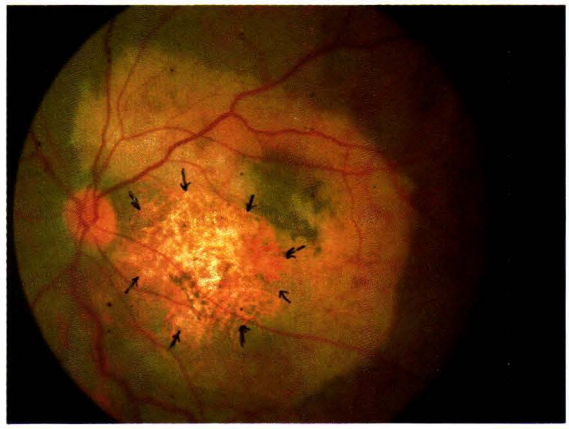


Fig. 16. **Patient 4. Fundus photography of both eyes shows a large placoid juxtapapillary and macular yellow-white choroidal tumor extending between the temporal arcades. A) Right eye. Note the prominent and numerous vascular tufts (arrows). B) Left eye. Arrows point to the area of posterior staphyloma.**

calcium, total T4, free T4, and T3 were within normal range. Other investigations were not performed before delivery. At 7 months following delivery, SRF still persisted and visual acuity had decreased to 20/400. Axial enhanced CT scan of the orbit demonstrated a moderate calcific plaque just superolateral to the optic nerve insertion (Fig. 11). Post gadolinium oblique sagittal T1W with fat suppression MRI of the left orbit revealed a small nonenhancing hyposignal T1 plaque with focal thinning of the involved choroid just lateral to the optic nerve head insertion of the left globe (Fig. 12). The IVFA (Fig. 13) demonstrated late staining of the lesion. Within two months SRF disappeared spontaneously and visual acuity declined to 5/200. Laser ablation utilizing green argon laser, a total of 688 spots, 200 μm in size, at 730 mw for 0.2 second duration was applied to the border of the lesion. The treated area became

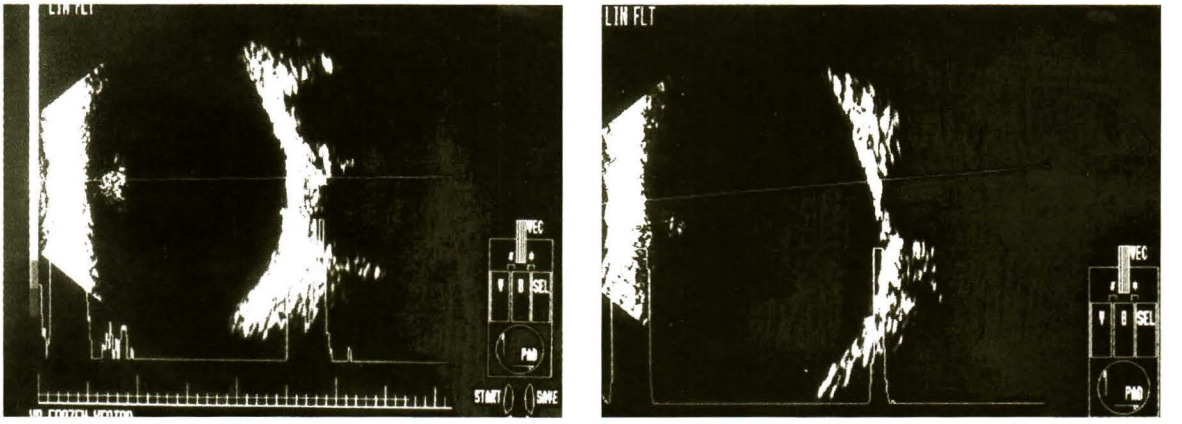


Fig. 17. Patient 4. Ultrasonography. A) Right eye. Acoustic shadowing of the orbit or a pseudo-optic nerve is seen posterior to the choroidal mass. B) Left eye. A plano-convex configuration mass with high reflectivity echoes.

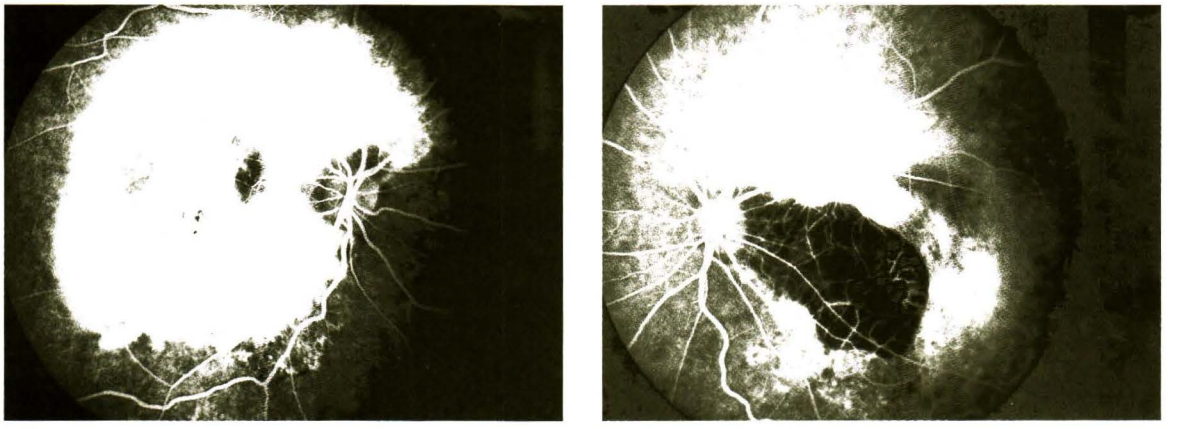


Fig. 18. Patient 4. Fluorescein angiography of both eyes. A) Diffuse staining throughout the lesion. B) A large hypofluorescent area contrasts to intense staining of the tumor in the late phase of the angiogram, corresponding to a posterior staphyloma where osteoma did not encroach.

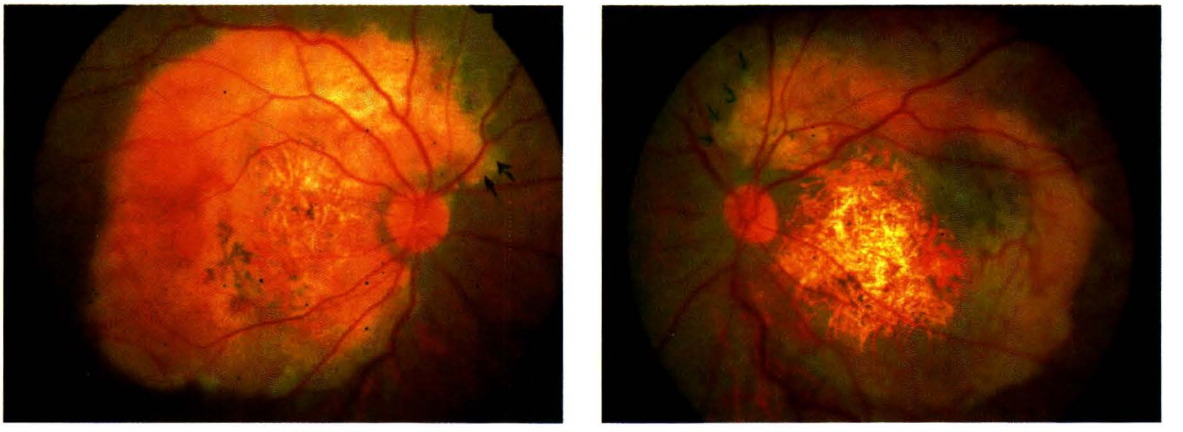


Fig. 19. Patient 4. Fundus photography of both eyes at two and a half years following initial examination. A) Right eye. B) Left eye. Note the gradual growth of the tumor in a pseudopodium manner (arrows).

thinner and, the retinal pigment epithelium overlying the tumor, was hyperpigmented. In May 1993, the lesion as seen in Fig. 14 was noted to have enlarged slightly towards superonasally and in July 1999, the lesion showed irregular thinning and had become decalcified in the area of previous laser treatment as well as in area without laser treatment (Fig. 15).

Case 4:

A 35 –year-old female school teacher was seen at the clinic in March 1994 with a history of blurred vision in both eyes for one month's duration. Her past medical history was positive for thyroid goiter and she had undergone treatment for the past three years. Her distant visual acuity was 20/100 with -3.00 Dsph ⊖ -1.00x180° in the right eye and 5/200 with –6.50 Dsph ⊖ -1.00x180° in the left. Dilated fundus examination disclosed bilateral large placoid juxtapapillary and macular yellow-white choroidal tumor extending between temporal arcades. The patient also had bilateral posterior staphyloma (Fig. 16A-16B). The vascular tufts in the inner surface of the tumor were numerous and more prominent in the right eye than in the left. The elevated choroidal lesion did not encroach upon the staphyloma. There was associated serous detachment of the neurosensory retina in each eye which gradually subsided over a period of 14 months. Ultrasonography of the right eye showed an acoustic shadowing of the orbit posterior to the choroidal mass giving the appearance of a pseudo-optic nerve (Fig. 17A) and in the left eye it demonstrated a plano-convex configuration mass with high reflectivity echoes which persisted at lower scanning sensitivity (Fig. 17B). MRI of the orbit, using surface coil and fat suppression technique showed a small nonenhanced hyposignal T1 area with focal thinning of the involved choroid. IVFA showed intense staining in late phase of the angiogram (Fig. 18A-18B). In September 1996, the tumor had slightly increased in size in both eyes (Fig. 19A-19B) but visual acuity remained unchanged. At the latest check up in June 1999, her visual acuity remained unchanged but the lesion had spontaneously decalcified in part.

DISCUSSION

The presented patients had orange red to yellow white choroidal lesions in the juxtapapillary and macular region which had to be differentiated from choroidal hemangioma, amelanotic choroidal malignant melanoma, optic nerve tumor involving the posterior pole of the eye, and from metastatic carcinoma to the choroid. However, the echographic findings in the present study demonstrated high reflectivity echo spikes which persisted at lower scanning sensitivity in A-scan mode and in B scan the unique characteristics of a plano-convex sonically dense lesion were demonstrated in all of the cases. This is because, when the sound beams stroke the large surface of ossification in the choroid, the ultrasonic pulses were reflected at boundaries between vitreous and ossified plaque in a regular manner. The residual sound energy then entered the homogeneous dense mass until they reached the posterior surface of the lesion which curved along the posterior surface of the globe. These characteristics are easy to recognize in addition to other known features of a pseudo-optic nerve, or an acoustic shadowing of the orbit posterior to the lesion, as previously described[16] and are very useful in differentiating choroidal osteoma from amelanotic malignant melanoma or other lesions such as choroidal hemangiomas, which may calcify but do not display such features. CT scan is useful in detecting the presence of hyperdense calcification or ossification in all patients while the high resolution MRI of the same three choroidal osteomas shows a hypointense signal on unenhanced T1W but not all T2W images. These findings were different from those reported by De Potter et al[17,18].

It was apparent that choroidal osteoma in the presented patients had varying colors and this may represent a different stage of bone development. The prominent branching vascular tufts in the first case were barely visible at the first examination but clearly developed later at follow-up while, in case 4, these vessels were prominently seen at of the first examination. The small tufts of branching blood vessels may simulate tufts of new vessels and cause serous or hemorrhagic detachment of the macula. By contrast, all patients had serous retinal detachment and no subretinal neovascular membranes (SRNVMs) were detected. In case 4, an excavated depression in the center of the tumor devoid of bony component, corresponded to the posterior staphyloma of sclera, where choroid was scarce or thinning.

The presented patients had sought ophthalmic advice because of blurring of central vision and metamorphopsia resulting from serous detachment of the neurosensory retina that spontaneously subsided within one to 14 months and, serous subretinal fluid leaked to the inferior retina in only one patient. However, all except one had good visual acuity. As for

etiology, previously suggested possible etiology included congenital, inflammatory, systemic diseases and endocrine influence[8,11,14,19-28]. The present study pointed out that hormonal changes may implicate the development of this choroidal tumor as all four patients were young or older female adults who had passed puberty. Only one of the 4 patients noticed blurred central vision during her first trimester of pregnancy while two others had a history of thyroid disease before they were found to have choroidal osteoma. However, none had decreased serum calcium or increased serum phosphorus or increased alkaline phosphatase.

Slight progressive enlargement of the choroidal osteoma was observed in three patients occurring in a pseudopod manner. In recent reports of larger numbers and long term follow-ups, a variable pattern of growth was documented in about 40 per cent, and rapid enlargement of the lesion was also known but rare[12,13]. Laser photoablation has been advocated in the treatment of bone resorption and also in SRNVM[29-31]. The proposed mechanism of laser bone ablation is that the laser may activate osteoclast to increase bone resorptive properties and the choriocapillaris overlying the tumor would also be destroyed. The lesion in one of the presented cases decalcified several years following laser ablation. The resorption of bone in this particular patient undergoing laser treatment was similar to that observed by Trimble et al in which complete decalcification occurred 18 months following argon laser photocoagulation for SRNVM[32]. However, spontaneous involution or decalcification can occur without laser ablative treatment[32,33]. This was also shown in another patient whose spontaneous decalcification began at the fifth year of follow-up. It is most likely that decalcification is part of the natural course of this disease.

SUMMARY

The authors reported four female Oriental patients who presented with central blurred vision from serous detachment of the macula secondary to choroidal osteoma. The importance is that choroidal osteomas are benign tumors which have a potential of growth in a gradual and pseudopod manner and may decalcify at late stages with or without laser ablative treatment. Clinical characteristics, ultrasonographic and CT scan findings are the keys to diagnosis of this benign tumor. Echography is the best method for identifying this lesion and has the unique acoustic features of a plano-convex sonically dense lesion with high reflectivity and persists at low system sensitivity.

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ออสตีโอมาของคอรอยด์ในผู้ป่วยชาวตะวันออก

สุชุมา วรศักดิ์, พบ,***, ชเนศ สุวรรณมณี, พบ**, อัทยา อยู่สวัสดิ์, พบ*,***, วิชัย สุเมธพิมลชัย, พบ***, จิรพร เหล่าธรรมทัศน์, พ.บ.*****

Choroidal osteoma เป็น benign ossifying tumour ของ choroids ที่พบน้อย รายงานฉบับนี้ เป็นรายงานฉบับแรกของ choroidal osteoma ในคนไทยในรอบ 20 ปีที่ผ่านมา

วัตถุประสงค์: เพื่อรายงานลักษณะทางคลินิกของโรค choroidal osteoma การตรวจด้วยคลื่นเสียงสะท้อน คอมพิวเตอร์สแกนของลูกตา หรือ magnetic resonance imaging (MRI) และติดตามการดำเนินโรคในระยะยาว

วิธีการศึกษา: ศึกษาย้อนหลังดูการเปลี่ยนแปลงของโรคและผลการรักษาผู้ป่วย choroidal osteoma 4 ราย ที่ตรวจพบระหว่างปี พ.ศ. 2533 – พ.ศ. 2543

ผลการศึกษา: พบผู้ป่วย 4 ราย อายุระหว่าง 24–37 ปี ทุกรายติดตามผลอย่างน้อย 5 ปี 3 ใน 4 ราย เป็นในตาข้างเดียว อีก 1 ราย เป็นทั้ง 2 ตา ตำแหน่งที่เป็น อยู่ใกล้ขั้วประสาทตาและจุดรับภาพ ทุกรายมี serous fluid ใต้จอรับภาพ แต่ไม่พบเส้นเลือดงอกใหม่ใต้จอรับภาพ 2 รายเคยเป็นไทรอยด์เป็นพิษมาก่อน อีก 1 ราย ตั้งครรภ์ขณะตรวจพบโรค ลักษณะเฉพาะที่เห็นได้จากคลื่นเสียงสะท้อน คือ เป็นก้อนทึบที่มีเงาสะท้อนสูง ด้านบนของก้อนจะแบน หรือก้อนมีลักษณะแบนส่วนบนและโค้งส่วนล่าง พบว่า choroidal osteoma จะเพิ่มขนาดอย่างช้า ๆ ในลักษณะ pseudopodium ในระยะ 2–6 ปี หลังตรวจพบครั้งแรก จอประสาทตากลับติดได้เอง ภายในเวลา 1–14 เดือน พบ decalcification ภายในเวลา 5 ปี โดยเกิดขึ้นเองหรือหลังได้รับการรักษาด้วยแสงเลเซอร์

สรุป: choroidal osteoma เป็นก้อนเนื้องอกที่พบน้อยในคนเอเชีย การเปลี่ยนแปลงของฮอร์โมนอาจมีผลต่อการเกิดโรคนี้ การตรวจด้วยคลื่นเสียงสะท้อนเป็นวิธีที่ดีที่สุดในการวินิจฉัย serous fluid ใต้จอรับภาพเกิดขึ้นได้โดยไม่ต้องมีเส้นเลือดงอกใหม่ และหายไปได้เอง decalcification เป็นส่วนหนึ่งของการดำเนินโรค

คำสำคัญ : ออสตีโอมาของคอรอยด์, Oriental Patients, Echography, MRI

สุชุมา วรศักดิ์, ชเนศ สุวรรณมณี, อัทยา อยู่สวัสดิ์,
วิชัย สุเมธพิมลชัย, จิรพร เหล่าธรรมทัศน์
จดหมายเหตุทางแพทย์ ฯ 2546; 86: 562–572

* ภาควิชาจักษุวิทยา, คณะแพทยศาสตร์ โรงพยาบาลรามาธิบดี,
** โรงพยาบาลพระมงกุฎเกล้า, กรุงเทพ ฯ 10110
*** โรงพยาบาลจักษุรัตนิน, 80/1 ซอยอโศก, เขตวัฒนา, กรุงเทพ ฯ 10110
**** ภาควิชารังสีวิทยา, คณะแพทยศาสตร์ โรงพยาบาลรามาธิบดี, กรุงเทพ ฯ 10400
† นำเสนอในที่ประชุมวิชาการครบรอบศตวรรษ ประชุมวิชาการประจำปีของ American Academy of Ophthalmology, ตุลาคม พ.ศ. 2539 มลรัฐชิคาโก, ประเทศสหรัฐอเมริกา
† ได้รับทุนบางส่วนจากคณะแพทยศาสตร์ โรงพยาบาลรามาธิบดีและมูลนิธิอุทัยรัตนิน