

Case Report/รายงานผู้ป่วย

# ผลการรักษา Papillary Capillary hemangioma ด้วยวิธี Photodynamic Therapy: รายงานผู้ป่วย

แมนสิงห์ รัตนสุคนธ์, พ.บ.\*

ณัฐวัฒน์ วงษ์ไชยคณากร, พ.บ.\*

## บทคัดย่อ

รายงานผู้ป่วยไทยที่มี papillary capillary hemangioma ในตาซ้าย ได้รับการรักษาด้วย photodynamic therapy (PDT) พบว่าระดับสายตาของผู้ป่วยดีขึ้นจาก 20/160 เป็น 20/63 และขนาดของก้อน, exudate และน้ำใต้จอตาลลดลง หลังการรักษาครบ 12 เดือน รวมทั้งไม่พบผลข้างเคียงใดๆ จากการรักษา **จักษุเวชสาร 2550 ; มกราคม-มิถุนายน 21(1) : 45-51.**

---

\* ภาควิชาจักษุวิทยา คณะแพทยศาสตร์ มหาวิทยาลัยสงขลานครินทร์ อ. หาดใหญ่ จ. สงขลา 90110

# Photodynamic Therapy for Papillary Capillary Hemangioma: A Case Report



Mansing Ratanasukon, M.D.\*

Nattawat Wongchaikanakorn, M.D.\*

## Abstract

The authors reported a Thai female patient with a papillary capillary hemangioma in the left eye treated with one session of standard photodynamic therapy (PDT) protocol. The patient's visual acuity improved from 20/160 to 20/63 with the reduction in lesion size, exudate and subretinal fluid within 12 months after treatment. There was no complication during the follow-up period. **Thai J Ophthalmol 2007 ; January-June 21(1) : 45-51.**

**Keywords:** *papillary capillary hemangioma, vertiporfin, photodynamic therapy*

---

\* Department of Ophthalmology, Faculty of Medicine, Prince of Songkla University, Hat Yai, Songkhla province, Thailand 90110

## Introduction

Capillary hemangiomas can occur as isolated tumors (von Hippel disease) or as part of the von Hippel-Lindau syndrome. Capillary hemangiomas of the optic disc (papillary capillary hemangioma) and juxtapapillary type can also occur and be endophytic or exophytic. The most clinically apparent of papillary capillary hemangioma is the endophytic type, which grows inward toward the vitreous cavity. The tumor is a smooth, reddish or orange, elevated, and well-defined nodule. It can have progressive leakage, resulting in visual loss caused by macular exudate or serous retinal detachment.

The clinical course of papillary capillary hemangioma is variable and difficult to predict but tends to have progressive visual deterioration.<sup>1-4</sup> In a large series of 72 eyes with juxtapapillary capillary hemangiomas, visual acuity (VA) was less than 20/40 in 39% of eyes at initial presentation and had further declined below this level in 65% of eyes after a mean follow-up of 5.4 years.<sup>3</sup> Schindler et al<sup>4</sup> reported visual deterioration in 28% of 14 eyes with papillary capillary hemangioma during 1 to 4 years of follow-up. Spontaneous regression of this tumor can occur but not common.<sup>5-6</sup>

Most peripheral capillary hemangiomas can be treated with laser photocoagulation or cryotherapy with a good result. However, the management of papillary capillary hemangioma is extremely difficult because of its anatomic location, near the optic disc and papillomacular bundle. Multiple technical approaches have been described, including laser photocoagulation, radiation therapy, transpupillary thermotherapy, and vitreoretinal surgery but their efficacy is very limited.<sup>7</sup>

Most recently, the efficacy of PDT for choroidal hemangioma is well reported and has been demonstrated to reduce exudation and subretinal fluids.<sup>8-9</sup> Because choroidal and retinal hemangioma

share a common histopathology in compromised angiomatous vessels, thus it should follow that PDT may be effective in treating papillary capillary hemangioma. Several authors have described the efficacy of PDT in capillary hemangiomas.<sup>10-14</sup> The authors reported the treatment of papillary capillary hemangioma by PDT in Thai patient with a good result.

## Case report

A 31-year-old Thai female patient presented with blurred vision in the left eye for 3 months at Department of Ophthalmology, Faculty of Medicine, Prince of Songkla University on February 2006. On presentation, visual acuity was 20/80 in left eye and funduscopy revealed a red, well-circumscribed lesion of one disc diameter above the left optic disc (Figure 1A). Also noted was exudate and serous detachment in the macula, confirmed by optical coherence tomography (OCT). There were no peripheral lesions identified, and no other abnormalities seen on examination. Fluorescein angiography demonstrated immediate hyperfluorescence with moderate progressive leakage late in the angiogram (Figure 2). A diagnosis of papillary capillary hemangioma was made with a decision to observe.

Over the following 3 months, her visual acuity deteriorated to 20/160 with increasing exudate and subretinal fluid. A decision was made to treat the lesion with PDT. The treatment was performed using verteporfin (Visudyne™ Novartis, Ophthalmics) according to standard protocol with the use of laser (Zeis Visulas 690s) and Visulink PDT adapter via a fiber optic and slit lamp using a contact lens. The dose of verteporfin was 6 mg/m<sup>2</sup> body surface area. This was dissolved and administered intravenously at a constant rate over a 10-minute period. Photoactivation was applied at 5 minutes following completion of infusion. A spot size was 3,800 μm (covering

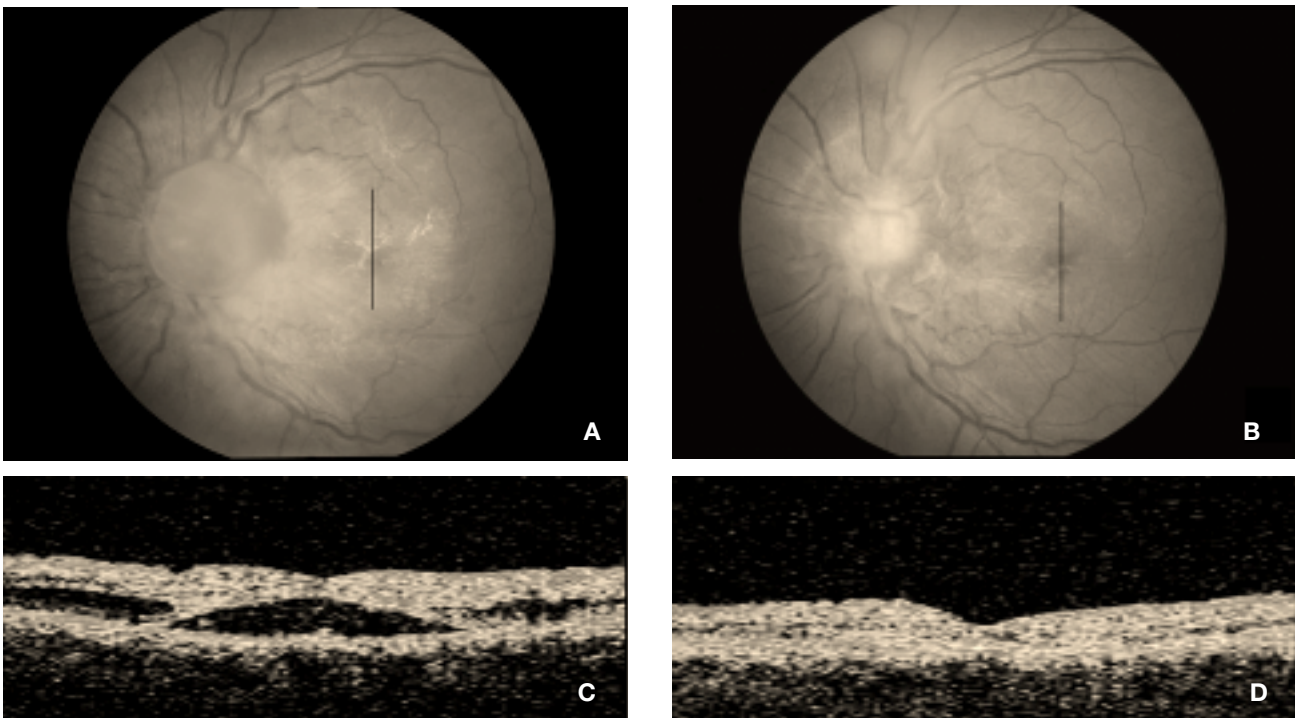
the largest dimension of the lesion). Irradiation of laser was 600 mW/cm for 83 seconds, achieving a total light dose of 50 J/cm<sup>2</sup>.

Following the treatment, no adverse effects from the treatment were noted. After 1 month, her visual acuity was 20/125 with reduction of lesion size, redness and subretinal fluid but angiography still revealed late leakage of lesion. The patient was then followed with serial acuity, angiography and OCT. At 6 months, the lesion was the same. The OCT demonstrated mild increasing of subretinal fluid. The late leakage of lesion still was observed with angiography but no visual deterioration. A decision was made to observe. At 12 months after the treatment, the patient noticed significant subjective improvement. Visual acuity was improve to 20/63 and a reduction of lesion size and

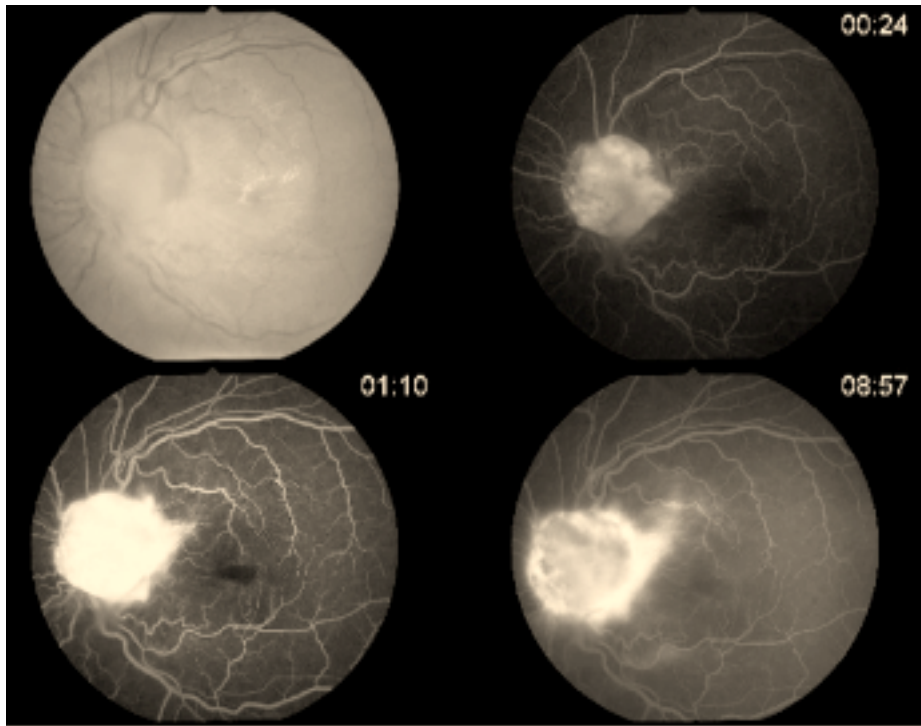
exudate (Figure 1B). No subretinal fluid was demonstrated by OCT (Figure 1C, 1D) and no leakage of lesion in angiography at the final follow-up (Figure 3).

## Discussion

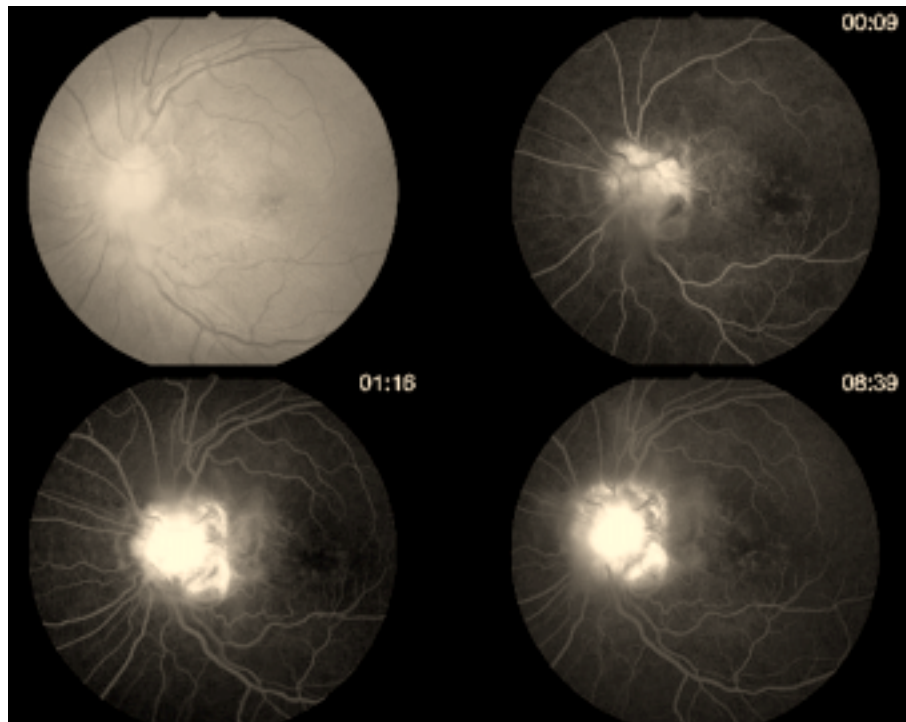
Verteporfin is a photosensitizer and has enhanced selectivity for tissues with up-regulated low density lipoprotein receptors. Choroidal neovascularization up-regulate low density lipoproteins, and consequently PDT with verteporfin is effective in this neovascularization. Moreover, retinal capillary hemangiomas are vascular hamatomas of the retina. The findings from histologic examination demonstrate normal endothelial cells, basement membranes and pericytes, but new vessels can develop on the ante-



**Figure 1.** A, Color fundus photograph of the left eye at initial presentation shows a red, well-circumscribed lesion above the left optic disc. B, Color fundus photograph of the left eye at 12 months after PDT shows a reduction of lesion size, subretinal fluid and exudate. No subretinal fluid was demonstrated by OCT (C, at initial presentation D, 12 months after PDT). (รูปสีที่ถ่ายเล่ม)



**Figure 2.** The fluorescein angiography at presentation shows immediate hyperfluorescence with moderate progressive leakage at late phase.



**Figure 3.** The fluorescein angiography at 12 months shows no leakage in angiogram with the improvement of visual acuity.

rior surface of the tumor.<sup>15</sup> The vascular endothelial cell lines have active uptake of acetylated low density lipoprotein.<sup>16</sup> It is reasonable to believe that PDT should also be effective in stimulating a thrombotic effect in these vascular tumors.

Several articles have shown the efficacy of PDT for capillary hemangiomas. Schmidt-Erfurth et al<sup>10</sup> treated five eyes with a standardize regimen of PDT. They found no improvement in visual acuity after one to three treatments in all patients. However, tumor regression with resolution of macular exudates and serous retinal detachment were obtained in all eyes after treatment. The discrepancy between functional and anatomical results was attributed to vascular complications including optic nerve ischemia and retinal vessel occlusions.

Atebara<sup>11</sup> reported a 55-year-old Asian male with large capillary hemangioma near the macula treated with 3 sessions of PDT. A first session of PDT was performed using standard protocol but second and third session of PDT were performed with a longer laser treatment duration of 166 seconds in three months interval. With 10 months of follow-up, involution of the hemangioma, reduction of subretinal fluid, and improvement of best-corrected visual acuity to 20/80 was seen.

The modified photodynamic protocols were also performed in 3 patients with large retinal capillary hemangiomas by Aaberg et al.<sup>12</sup> The modified protocols included shorter verteporfin perfusion times (infused over 5 minutes and laser was applied 6 minutes after infusion) and longer light exposure times (166 seconds). In 2005, Golshevsky et al<sup>13</sup> reported the successful treatment of juxtapapillary capillary hemangioma with PDT. The shorter verteporfin perfusion time (infused over 5 minutes) and longer light exposure times (166 seconds) were performed after failed treatment from previous standard protocol of

PDT.

In the presented report, the authors performed the one session of standard protocol of PDT in papillary capillary hemangioma. Visual acuity improved from 20/160 to 20/63 in 12-months follow-up and the reduction of lesion size, exudates and subretinal fluid was noted. Both anatomical and functional improvements were detected in our case.

The PDT protocols may influence in the results of treatment. The bolus infusion of verteporfin, rapid photoactivation and longer duration of laser applied were performed in a few articles as previous described.<sup>11-13</sup> Because of the relatively high-flow vascular tumor, a bolus of drug activated early would increase drug concentration and minimize time for drug clearance. Moreover, type of photosensitizers in PDT may also influence in the outcomes. Obana et al<sup>14</sup> treated a 36-year-old male with papillary capillary hemangioma with one session of PDT with hematoporphyrin derivatives, not verteporfin in a standard protocol with the different laser. Nevertheless, the vaso-occlusive effects at the level of the retina and optic nerve following PDT can compromise the functional outcomes. Optic nerve ischemia may occur in the treatment of papillary or juxtapapillary capillary hemangiomas with PDT that cause no improvement in visual function.<sup>10</sup> The parameters proven safe in choroidal neovascularization may be inappropriate in capillary hemangiomas.

In summary, while the optimal protocol for PDT in the treatment of capillary hemangiomas is unknown. From the presented case, the one session of standard protocol appeared to be effective in the treatment, with no any complications detected. The improvements of visual acuity, lesion size, exudates and subretinal fluid were observed in 12 months after the treatment.

## References

1. Gass JD, Braunstein R. Sessile and exophytic capillary angiomas of the juxtapapillary retina and optic nerve head. *Arch Ophthalmol* 1980;98:1790-7.
2. Atebara NH, Shields JA. Capillary hemangioma of the optic disc associated with a total retinal detachment. *Ophthalmic Surg* 1993;24:686-8.
3. McCabe CM, Flynn HW Jr, Shields CL, et al. Juxtapapillary capillary hemangiomas. Clinical features and visual acuity outcomes. *Ophthalmology* 2000;107:2240-8.
4. Schindler RF, Sarin LK, MacDonald PR. Hemangiomas of the optic disc. *Can J ophthalmol* 1975;10:305-18.
5. Whitson JT, Welch RB, Green RW. Von Hippel-Lindau disease: case report of a patient with spontaneous regression of retinal angioma. *Retina* 1986;6:253-9.
6. Milewski SA. Spontaneous regression of a capillary hemangioma of the optic disc. *Arch Ophthalmol* 2002;120:1100-1.
7. Garcia-Arumi J, Sararols LH, CAvero L, et al. Therapeutic options for capillary papillary hemangiomas. *Ophthalmology* 2000;107:48-54.
8. Barbazetto I, Schmidt-Erfurth U. Photodynamic therapy of choroidal hemangioma: two case reports. *Graefes Arch Clin Exp Ophthalmol* 2000;238:214-21.
9. Madreperla S. Choroidal haemangioma treated with photodynamic therapy using verteporfin. *Arch Ophthalmol* 2001; 87:84-9.
10. Schmidt-Erfurth UM, Kusserow C, Barbazetto IA, et al. Benefits and complications of photodynamic therapy of papillary capillary hemangiomas. *Ophthalmology* 2002;109:1256-66.
11. Atebara NH. Retinal capillary hemangioma treated with verteporfin photodynamic therapy. *Am J Ophthalmol* 2002; 134:788-90.
12. Aaberg TM Jr, Aaberg TM Sr, Martin DF, et al. Three cases of large retinal capillary hemangiomas treated with verteporfin and photodynamic therapy. *Arch Ophthalmol* 2005;123: 328-32.
13. Golshevsky JR, O'Day J. Photodynamic therapy in the management of juxtapapillary capillary haemangiomas. *Clin Experiment Ophthalmol* 2005;33:509-12.
14. Obana A, Goto Y, Ikoma M. A case of von Hippel-Lindau disease with papillary capillary hemangioma treated by photodynamic therapy. *Nippon Ganka Gakkai Zasshi* 2004;108: 226-32.
15. Gass JDM. Retinal vascular hamatomas. In: Gass JDM, ed. *Stereoscopic atlas of macular diseases: diagnosis and treatment*. St.Louis: Mosby,1997:844-58.
16. Obeso J, Weber J, Auerbach R. A hemangioendothelioma-derived cell line: its use as a model for the study of endothelial cell biology. *Lab Invest* 1990;63:259-69.