Tractional macular detachment caused by juxtapapillary retinal capillary hemangioma treated successfully by photodynamic therapy

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ABSTRACT

Introduction: To describe a case of tractional macular detachment caused by a juxtapapillary retinal capillary hemangioma (JRCH) treated successfully by photodynamic therapy (PDT).

Case Report: A 19-year-old man presented with reduced vision in his left eye of five months duration. Clinical examination revealed a tractional retinal detachment caused by a JRCH. He was treated with PDT, and the macular traction was released and the retinal detachment disappeared after seven weeks. Conclusion: Photodynamic therapy can be considered as a suitable treatment for the traction retinal detachment caused by a JRCH.

Keywords: Photodynamic therapy, Retinal capillary hemangioma, Retinal detachment, Vascular tumor

INTRODUCTION

A retinal capillary hemangioma (RCH) is a benign vascular tumor that can be solitary, without systemic disease or can be associated with the von Hippel-Lindau (VHL) syndrome [1–3]. Many RCHs are located in the juxtapapillary region [1]. It can cause visual impairments by leakage, edema, deposition of hard exudate, traction retinal detachment involving the macular, and ultimately neovascular glaucoma [4]. The management of a RCH depends on the size, location, complications, and whether the patient has the VHL syndrome [1]. Various treatments have been reported for the management of RCH, such as observation alone [2, 5], laser photocoagulation [1, 2], cryotherapy [1, 6], radiotherapy [7, 8], intravitreal anti-vascular endothelial growth factor injection [4], transpupillary thermotherapy [9], photodynamic therapy (PDT) [10, 11], surgery [1, 12], and combination of these treatments. The selection of the treatment is influenced by the location of the tumor, the size, and the presence of exudation or traction. In cases of juxtapapillary RCH (JRCH), these treatments have a risk of a significant reduction of visual acuity due to the injury of the optic nerve or major retinal vessels [2].

We present a case with a traction macular detachment caused by a JRCH that was successfully treated by only PDT.

CASE REPORT

A 19-year-old man presented with a history of reduced vision in his left eye of five months duration. Before the visit, he had undergone photocoagulation
around maculae area with gas tamponade for a retinal detachment of the macula twice at another clinic.

At the initial examination in our clinic, his best-corrected visual acuity (BCVA) was 0.8 in the left eye. Fundoscopic examination revealed an RCH located on the optic disk and a proliferative membrane on the optic disk (Figure 1A). Optical coherence tomography (OCT) showed a retinal detachment of the macular area. Subretinal fluid was present but was not connected to the JRCH (Figure 1B, C), and we believed that this retinal detachment was caused by the traction on the retina. Magnetic resonance imaging (MRI) revealed no hemangioma in his brain. We observed him for several months without any treatments.

At eight months after the initial visit, his decimal BCVA had decreased to 0.6, and we decided to perform vitreoretinal surgery after the activity of the RCH was reduced by PDT. Photodynamic therapy was performed with benzoporphyrin-MA (Visudyne, Novartis, Hettlingen, Switzerland) as a sensitizer, and the total light dose was 50 J/cm² (Figure 2). Three weeks after the PDT, the tumor had shrunk, the vitreous of temporal side had been detached and hemorrhage on the surface of the tumor was observed, but the tractional retinal detachment still remained (Figure 3). At seven weeks after the PDT, the tumor was further reduced, and the traction on the retina was released (Figure 4). We concluded that vitreous surgery was not needed. At 11 months after the PDT, no subretinal fluid was detected, and no enlargement of the tumor was observed. His decimal BCVA was 1.0.

**DISCUSSION**

Retinal capillary hemangioma is basically a benign tumor but it can cause visual impairment by leakage, edema, hard exudates, and traction on the macula [1, 2, 4]. A worse prognosis has been reported in eyes with a JRCH compared with peripheral RCHs [2]. The treatment of JRCH is difficult because of risks of damaging the optic nerve or major vessels [2], and various treatment modalities have been reported. The selection of the treatment is influenced by the location, i.e., juxtapapillary or extrapapillary, its size, and the presence of retinal damage [2]. Schmidt-Erfurth et al. reported five cases of JRCH treated by PDT and in all cases the tumor size and exudative activity were reduced [10]. Sachdeva et al. reported six cases of RCH treated by PDT including three cases of JRCH, and in their cases PDT resulted in tumor regression or stabilization as well as a reduction in the size of the subretinal fluid and liquid exudation [11].

In our case, we diagnosed that the retinal detachment was caused by retinal traction which caused the decrease in the visual acuity. Mariotti et al. reported on a 13-year-old JRCH case treated with vitreous surgery followed by PDT [12]. Following their treatment protocol, we had planned to perform vitreous surgery followed by PDT.

**Figure 1:** Fundus of the right eye (A) and OCT of macular (horizontal slice: B, vertical Slice: C). A round circumscribed orange lesion covered by a proliferative membrane was observed at optic disk (A). OCT showed a retinal detachment of the macular area (B, C).

**Figure 2:** The location of the treatment spot.
But we were concerned of the risk of severe hemorrhage from the tumor during the surgery, and we selected to perform PDT to reduce the risk of hemorrhage before the vitreous surgery. But the retinal traction was released by the PDT alone, and the subretinal fluid was not detected. Therefore, vitreous surgery became unnecessary.

Imasawa and Iijima reported a case of laser-induced posterior vitreal detachment (PVD) [13]. The exact mechanism causing the PVD in our case was not determined but we assume that the PDT caused a shrinkage of the tumor, and the tangential force from the optic disk to the macula was released.

About the side effects of PDT, Schmidt-Erfurth et al. concluded vaso-occlusive effects at the level of retina and optic nerve compromised the functional benefit [10]. Sachdeva et al. reported the treatment benefit may be limited by preexisting macular change and worsening of epiretinal membrane [11]. Fortunately, we have not observed these kind of side effects in our case until now. But we thought that the careful selection of patients must be necessary.

CONCLUSION

Photodynamic therapy can be one of the treatment options for the tractional retinal detachment caused by a JRCH. A larger prospective study is necessary about the selection of patients.

REFERENCES

9. Parmar DN, Mireskandari K, McHugh D. Transpupillary thermotherapy for retinal capillary

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All relevant data are within the paper and its Supporting Information files.

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