Case Report

Paracentral Acute Middle Maculopathy following Surgically Induced Branch Retinal Artery Occlusion During Vitrectomy

Panagiotis Stavrakas^a Efthymios Karmiris^b Paris Tranos^c
Konstantinos Ananikas^d Christos Tooulas^d Alexandros Rouvas^d
Angeliki Androu^d

^aDepartment of Ophthalmology, University of Patras Medical School, Patras, Greece;

^bDepartment of Ophthalmology, Hellenic Air Force General Hospital, Athens, Greece;

^c"Ophthalmica" Institute of Ophthalmology, Thessaloniki, Greece; ^d2nd Department of Ophthalmology, University of Athens Medical School, Attikon University General Hospital, Athens, Greece

Keywords

Paracentral acute middle maculopathy · Capillary ischaemia · Pars plana vitrectomy

Abstract

Paracentral acute middle maculopathy (PAMM) is a spectral-domain OCT finding of a thickened hyperreflective band at the level of the intermediate layers of the inner retina, attributed to the acute phase of intermediate capillary ischemia. The purpose of this paper is to report a case of PAMM deriving from a surgically induced branch retinal artery occlusion (BRAO) during vitrectomy for vitreous hemorrhage. A 70-year-old female referred to the posterior segment office with a 1-month history of visual loss in her right eye. Best-corrected visual acuity (BCVA) was "counting-fingers" (CF) due to a dense vitreous hemorrhage. She underwent a standard 23-gauge vitrectomy with a relatively close to the optic disc endodiathermy application to an avulsed vessel. Follow-up examination revealed a well-demarcated retinal whitening involving the inferior macula. SD OCT showed a surgically induced branch retinal artery occlusion exhibiting both a cotton wool spot and a PAMM lesion. Our case affords an insight into those

pathological processes involved with PAMM, providing angiographic evidence of the retinal ischemic changes responsible for its development, by angiographically proving that branch retinal artery occlusion leads to a prompt formation of both a cotton wool spot and a PAMM lesion. Our aim is to raise awareness of this potential complication of vitrectomy surgery, especially when involving multiple traumatic manipulations onto the retinal surface.

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Introduction

Paracentral acute middle maculopathy (PAMM) is a novel descriptive term introduced by Sarraf et al. in 2013 [1] referring to a spectral-domain optical coherence tomography (SD-OCT) finding of a thickened hyperreflective band at the level of the deeper layers of the inner retina [2]. PAMM has been attributed to the acute phase of deep capillary ischemia, referring to ischemia of intermediate and deep capillary plexuses [2]. Since then, PAMM has been associated with various retinal vascular diseases, including retinal artery and central retinal vein occlusion, diabetic retinopathy, sickle cell retinopathy, Purtscher's retinopathy, inflammatory occlusive retinal vasculitis and hypertensive retinopathy [3], all sharing a similar pathophysiological background.

Similarly, multiple reports in the published literature have focused on the incidence of PAMM lesions secondary to intraocular surgery [4–6]. Creese et al. [5] demonstrated PAMM incidence following uncomplicated phaco-emulsification cataract surgery under local anesthesia using retrobulbar, peribulbar or sub-tenon's techniques, causing severe irreversible vision loss in a case series report. They speculated that pre-existing compromised retinal perfusion, due to medical history of vasculopathy and/or blood dyscrasias, may render some eyes more susceptible to transient compression or spasm of the central retinal artery [5]. Additionally, Nakashima et al. [6] reported a percentage as high as 3.8% of PAMM following vitrectomy for proliferative diabetic retinopathy (PDR), also focusing on underlying retinal vascular dysfunction, leading to local perfusion insufficiencies in diabetic patients.

The purpose of this paper is to report a case of PAMM in combination with ischemic manifestations deriving from the superficial capillary plexus following a surgically induced branch retinal artery occlusion during vitrectomy for vitreous hemorrhage.

Case Presentation

A 70-year-old female with past medical history of arterial hypertension presented with a 1-month history of visual loss in her right eye. On evaluation, best-corrected visual acuity (BCVA) was "counting-fingers" (CF) and 20/20 in the right and left eye, respectively. Examination revealed a dense vitreous hemorrhage obscuring fundus view in her right eye, not suggestive of retinal detachment, based on b-scan prompt examination. Fundoscopic examination of the left eye was totally unremarkable. Intraocular pressure was 16 mm Hg in both eyes. On the basis of these findings, a both diagnostic and therapeutic right eye surgical intervention under local anesthesia was scheduled. She underwent a standard 23-gauge micro-incision sutureless vitrectomy with valved cannulas (Constellation Vision System, Alcon Laboratories, Inc, Forth Worth, TX, USA) under a sub-tenon's local anesthesia consisting of a mixture of xylocaine 2% and ropivacaine 0.8%. Honan's balloon was not used in this case. The balanced salt solution (BSS) for infusion had 0.5 mL of 1;1,000 adrenaline, diluting it to 1;1,000,000. After

confirmation of a posterior vitreous detachment, a core vitrectomy was carried out, without the use of adjuvant triamcinolone acetonide. The intraocular pressure (IOP) setting was switched on during surgery at a set pressure of 25 mm Hg. During core vitrectomy constant bleeding of an avulsed inferior temporal arteriole, relatively close to the optic disc, was recognized and was considered the source of the vitreous hemorrhage. As hemostasis was not achieved by IOP elevation to 80 mm Hg for 2 min, endodiathermy was used in order to ensure the clearance of the media and the continuation of the surgery. Peripheral vitrectomy and vitreous base shaving were then performed. Search of the periphery by means of scleral depression, without aspiration, failed to disclose any retinal breaks or holes or otherwise suspicious retinal areas. As a result, BSS was used to fill the vitreous cavity at the end of the surgery. Apart of the 2 min elevation of IOP in order to achieve hemostasis, in no other instance did the IOP surpass the set pressure of 25 mm Hg throughout surgery. At the end of the surgery, a subconjunctival injection of cefuroxime and dexamethasone was administered.

Postoperatively, on day 1, and while BCVA of the affected eye had already improved to 20/100, routine post-op follow-up examination revealed a subtle but sizable and well-demarcated retinal whitening involving the inferior macula. Upon questioning, the patient admitted the existence of a paracentral scotoma, corresponding to the fundoscopic finding. SD-OCT showed thickening and hyperreflectivity of both the inner and middle retinal layers, as seen in Figure 1. On the basis of this finding, a fluorescein angiography was carried out 1 week later in order to allow adequate media clearance. The fluorescein angiography identified a branch retinal artery occlusion, demonstrating delayed perfusion and patchy filling of an inferior retinal arteriole, starting at the site of the intraoperative endodiathermy application (Fig. 2). By that time, a diagnosis of a surgically induced branch retinal artery occlusion exhibiting both a cotton wool spot and a PAMM lesion and thus affecting both superficial and intermediate capillary plexuses, was made. Intraocular pressure at the time of diagnosis was 12 mm Hg.

Three months postoperatively and while BCVA has improved to 20/40, SD OCT demonstrates a remarkable thinning and atrophy of the inner and middle retinal layers and the patient continues to experience a stable paracentral scotoma (Fig. 2).

Discussion/Conclusion

In post-mortem studies in human donor eyes, Tan et al. identified four different capillary plexuses in the human retina: the first two located in superficial layers, i.e., the nerve fiber layer and the ganglion cell layer, thus referred to as the superficial capillary plexus, another located at the junction of the inner plexiform layer and superficial boundary of the inner nuclear layer (intermediate capillary plexus) and the last located at the junction of the deep inner nuclear and outer plexiform layer border (deep capillary plexus) [7]. Since its introduction, PAMM's underlying pathophysiological mechanism has been consistently hypothesized to be one of infarction of both the middle and inner retina representing the final stage of an ischemic cascade [8]. Our case affords an insight into those pathological processes involved providing angiographic evidence of the retinal ischemic changes responsible for PAMM development.

In our case, surgically induced branch retinal artery occlusion of a second order inferior temporal retinal artery resulted in the formation of a PAMM lesion in combination with ischemic manifestations deriving from the superficial capillary plexus in the form of a cotton wool spot, at as early as the first postoperative day. We speculate that the onset of PAMM and cotton wool spot formation was immediate but could not be detected during surgery since retinal whitening commences to appear 45–60 min after an ischemic event [9], by which time

surgery was already over. Fluorescein angiography revealed a delayed filling of the occluded artery as well as a relative hypofluorescence at the site of the lesion, probably representing a masking effect. So, an angiographically proven branch retinal artery occlusion leads to a prompt formation of both a cotton wool spot and a PAMM lesion, located focally to the parafoveal area and not to the whole perfusion area, representing ischemia separately involving both superficial and intermediate capillary systems. The parafoveal localization of these lesions may be attributed to the greater density of the infarct-related artery's capillary network in the area. A combination of both superficial and intermediate capillary ischemia is proved to be the most common finding in retinal artery occlusion as shown by a retrospective observational study of 40 eyes with some form of retinal artery occlusion, with a percentage as high as 78% among them exhibiting fundoscopic findings deriving from both plexuses [2].

Admittedly, in this case, more than one factors could have contributed to retinal ischemia leading to PAMM development postoperatively. Apart from the obvious endodiathermy application and subsequent surgically induced branch retinal artery occlusion, the marked elevation of intraocular pressure during vitrectomy could also induce retinal circulatory insufficiency. Additionally, the type of local anesthesia used has been proposed to contribute to transient retinal ischemia, as Swamy et al. [10] have suggested that the anesthetic fluid may become trapped in the periocular connective tissue causing focal mechanical compression of the central retinal artery. Even ropivacaine's and adrenaline's vasoconstrictive effects have been accused of causing a temporary reduction of the central retinal artery's blood flow [11]. Moreover, according to Sugiura et al. [12], rapid scleral depression without concomitant aspiration can provoke more severe elevation of intraocular pressure during search of the periphery. Finally, pre-existing vasculopathies, as is systemic hypertension in our case, are considered a risk factor for PAMM [13, 14].

In conclusion, our case provides angiographic evidence of ischemia involving both superficial and intermediate capillary plexuses leading to a prompt formation of both cotton wool spot and PAMM lesions. Our aim is to raise awareness of this potential complication of vitrectomy surgery, especially when involving multiple traumatic manipulations onto the retinal surface, in addition to the impact of the nature of the surgery on the central retinal artery's blood flow itself. Many of the patients' subjective complaints may be overlooked, especially when visual acuity is preserved or even improved and is therefore of ophthalmologists to suspect and identify ischemic lesions deriving from deeper capillary plexuses and counsel patients accordingly.

Statement of Ethics

The patient consented to publication of the case in writing.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

All authors attest that they meet the current ICMJE criteria for authorship. Surgeon: P.S., ancillary testing: A.R., manuscript draft: A.A., E.K., P.T., K.A., and C.T.

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Fig. 1. **a** SD-OCT image depicting thickening and hyperreflectivity of both the inner (yellow arrow) and middle (white thick arrow) retinal layers. **b** SD-OCT image 3 months postoperatively. Notice the remarkable thinning and atrophy of the inner and middle retinal layers (yellow arrows).

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Fig. 2. Fluorescein angiography image depicting delayed filling of the occluded artery and the site of intraoperative endodiathermy deliverance (yellow arrow). The cotton wool spot and PAMM lesion localization giving rise to a relative masking effect is also depicted (yellow line).