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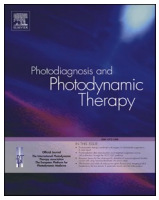

Case report

Paracentral acute middle maculopathy in a fasting patient after cataract surgery and its response to hyperbaric oxygen therapy

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A R T I C L E I N F O

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A B S T R A C T

Introduction: Paracentral acute middle maculopathy (PAMM) is a structural optical coherence tomography (OCT) sign secondary to ischemia in the intermediate and deep retinal vascular network, characterized by hyperreflectivity in the inner nuclear layer (INL).

Aim: Our objective is to demonstrate PAMM development following uncomplicated cataract surgery, possibly triggered by fasting and dehydration. We also aim to emphasize the potential role of hyperbaric oxygen therapy in treating PAMM.

Case presentation: A 66-year-old man with a past medical history of Neurofibromatosis type 1 and cardiovascular disease underwent uncomplicated cataract surgery in the left eye. The patient was also fasting due to Ramadan. The patient complained of very low vision during the routine postoperative examination on the third day. His-best-corrected visual acuity (BCVA) was counting fingers at 1 meter. His-anterior and posterior segment examination was unremarkable. In infrared imaging, a large hyporeflective area was observed in the parafoveal region, and structural OCT also showed increased hyperreflectivity in the middle retinal layers corresponding to the junction of INL and outer plexiform layer (OPL) involving the entire INL which suggested PAMM. Following 14 sessions of hyperbaric oxygen therapy, the patient's BCVA increased to 0.9 on the 14th day of diagnosing PAMM.

Conclusion: To the best of our knowledge, this is the first case representing a patient with PAMM triggered by fasting and cataract surgery who responded positively to hyperbaric oxygen therapy. However, triggering of PAMM by fasting is entirely unproven and that this observation occurred in a highly complex case with many other possible contributing factors. Also, the triggering of PAMM by some manipulation during surgery is equally unproven.

1. Introduction

Paracentral acute middle maculopathy (PAMM) is a structural optical coherence tomography (OCT) sign secondary to ischemia in the intermediate and deep retinal vascular network, characterized by hyperreflectivity in the inner nuclear layer (INL) [1]. Reports on PAMM cases secondary to cataract surgery are limited in the literature [2–4].

Hyperbaric oxygen therapy (HBOT) involves treating patients by inhaling 100% oxygen while within a pressurized treatment chamber [5]. The main purpose is to provide oxygen-rich plasma to tissue that lacks it, which helps to reduce reperfusion injury, enhance immune response, and promote the production of endothelial cells [5]. In a cost analysis study conducted in diseases where tissue hypoxia is considered to be an important factor, it was highlighted that HBOT is a cost-effective treatment [6].

Here, our objective is to demonstrate PAMM development in a case following uncomplicated cataract surgery, possibly triggered by fasting and dehydration in the presence of an underlying cardiovascular disorder. We also aim to emphasize the potential role of HBOT in the treatment of PAMM in the present particular case.

2. Case report

A 66-year-old man presented with gradual visual loss in his left eye over the last year. His-past medical history revealed Neurofibromatosis type 1 (NF-1) disease, hypertension and stent placement six years ago for coronary artery disease. He also underwent uncomplicated phacoemulsification surgery one year earlier by the same surgeon. His-body

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height and body weight were 160 cm and 75 kg, respectively, with a blood pressure of 120/70 mmHg. The patient was also fasting due to Ramadan. Snellen best-corrected visual acuity (BCVA) was 1.0 and 0.3 in the right and left eye, respectively. Slit lamp examination revealed pseudophakia in the right eye and grade 3 nuclear sclerotic cataract in the left eye. Intraocular pressures (IOP) were 10 mmHg (right eye) and 11 mmHg (left eye). Both fundi were unremarkable. OCT examination was normal in both eyes. In infrared imaging, hyperreflective patchy nodules were linked to hyperreflective signals in choroidal tissue in NF-1 (Fig. 1A) [7]. The patient was non-smoker and use no medication except for antihypertensive medication.

Following sub-Tenon anesthesia (sub-Tenon techniques, infero-medial quadrant, 2cc lidocaine, 1cc bupivacaine) without any compression to the globe, routine phacoemulsification with intraocular lens implantation was performed. Honan's balloon was not used. In brief, a 2.6 mm clear corneal incision was made, a viscoelastic substance

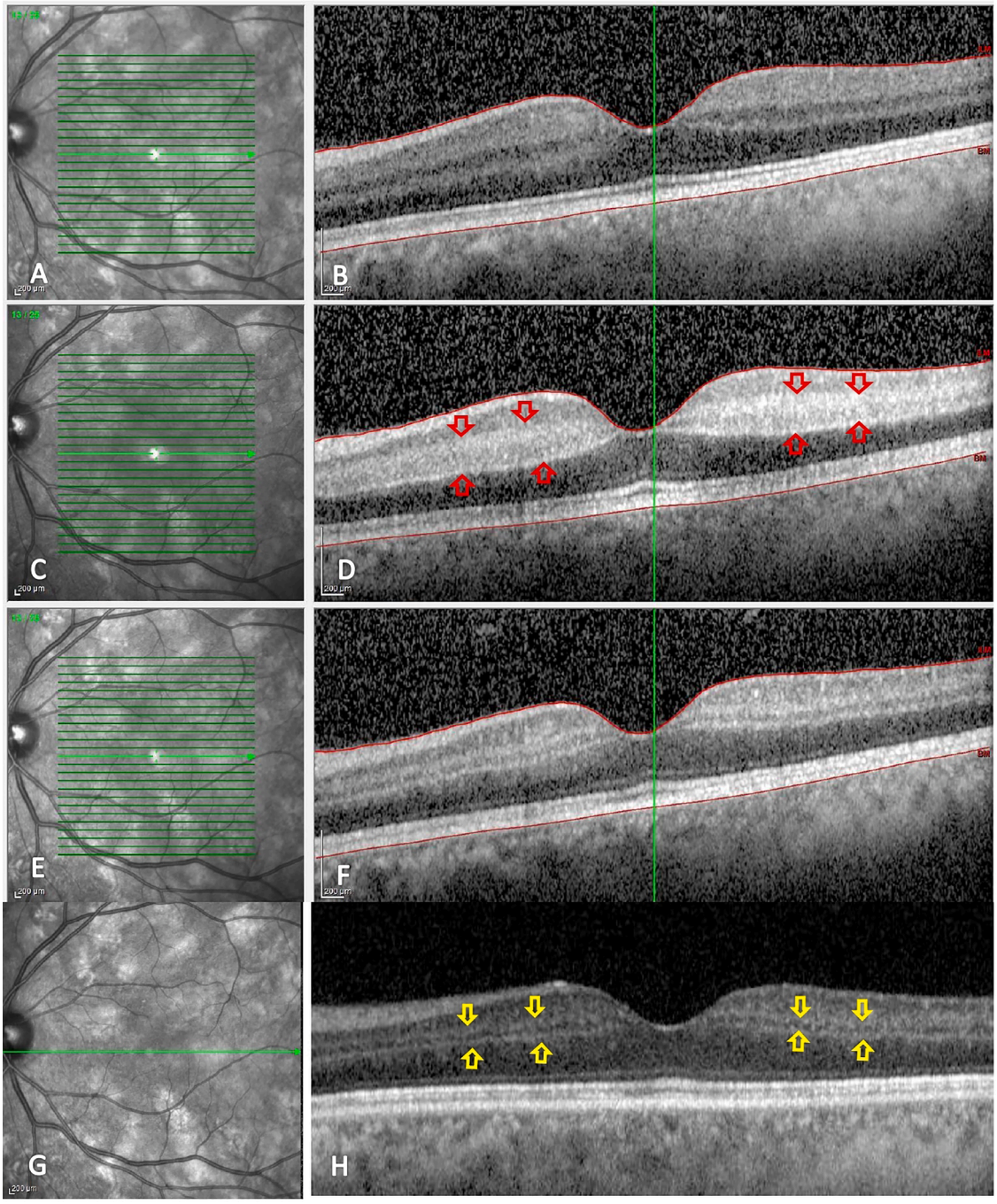

Fig. 1. Representative infrared and structural optical coherence tomography (OCT) images. (A) Preoperative infrared image, hyperreflective patchy nodules were linked to hyperreflective signals in choroidal tissue in NF-1. (B) Preoperative structural OCT image. (C) A postoperative third-day infrared image shows that a large hyporeflective area was prominent over the affected area. (D) Postoperative third-day structural OCT image shows increased hyperreflectivity in the junction of the inner nuclear layer (INL), and outer plexiform layer involving the entire INL (red arrows). (E) On the 5th day of the Paracentral acute middle maculopathy (PAMM) diagnosis hyporeflective area in the infrared image decreased. (F) On the 5th day of PAMM diagnosis, hyperreflectivity in the structural OCT scan decreased. (G) On the 14th day of PAMM diagnosis, hyperreflectivity disappeared in infrared imaging. (H) On the 14th day of PAMM diagnosis, atrophy in the inner retinal layers was seen in structural OCT (yellow arrows).

(Protectalon® 1.8% VSY Biotechnology) was injected into anterior chamber and a continuous curvilinear capsulorhexis was performed. Following gentle hydrodissection, stop and chop technique was used to remove the nucleus using the Constellation device (Alcon Laboratories, Inc). After aspiration of cortical material, a viscoelastic substance was again injected, and a monoblock monofocal intraocular lens was implanted in the capsular bag. The viscoelastic substance was thoroughly removed from the anterior chamber. Corneal wounds were hydrated, and intracamaral antibiotic (1 mg/0.1 mL cefuroxime) was administered. Surgery was completed with an IOP left at normotensive levels. The total surgical time was 15 minutes. The postoperative regimen included moxifloxacin and dexamethasone eye drops every two hours and per oral administration of one-a-day acetazolamide (250 mg).

On postoperative first-day slit lamp examination, there was 1+ cells in the anterior chamber in the left eye. On the third postoperative day, the patient complained about very low vision in his left eye. The BCVA was counting fingers at 1 meter. He had no evidence of a relative afferent pupillary defect. In anterior segment examination, the cornea was clear, there were 0.5+ cells in the anterior chamber with a well centered in the bag placed IOL in the posterior chamber. The IOP was 8 mmHg in the right eye and 9 mmHg in the left eye. No distinctive feature was observed in the posterior segment examination. In infrared imaging, a large hyporeflective area was observed in the parafoveal region (Fig. 1C), which was not seen during preoperative evaluation (Fig. 1A and 1B). Structural OCT also showed increased hyperreflectivity in the middle retinal layers corresponding to the junction of INL and outer plexiform layer (OPL) involving the entire INL, which suggested PAMM (Fig. 1D). After a detailed discussion with the patient regarding treatment options, the patient was immediately referred to HBOT without further examination. Because the hyperbaric oxygen unit in our city was temporarily out of service, the patient was referred to the nearest unit by ambulance helicopter. The patient was not fasting during HBOT. Hyperbaric oxygen therapy sessions were at 2.4 atmospheres, and each treatment session lasted 120 minutes. It was applied as two sessions on the first day of the treatment and once a day on the following days.

After four sessions of HBOT, the BCVA increased to 0.4 on the 5th day after the PAMM diagnosis. It was observed that both the hyporeflective area in infrared imaging and the hyperreflectivity in the structural OCT scan decreased, and the retinal layers could be distinguished (Fig. 1E and F). Fundus fluorescein angiography and color fundus imaging were unremarkable (Fig. 2A and B). At the end of 14 sessions of hyperbaric oxygen therapy with the recommendation of an underwater and hyperbaric medicine specialist, the BCVA was improved to 0.9 on the 14th day following PAMM diagnosis. Hyporeflectance on infrared imaging disappeared, and atrophy in the inner retinal layers was noted in structural OCT (Fig. 1G and H).

3. Discussion

Retinal capillary system is very important in supplying the metabolically hyperactive region of the inner and middle retina. The dynamics of blood flow through this region include both parallel and in series components where blood flows in a mainly serial direction between the superficial vascular complex and deep vascular complex [8]. The mechanism of PAMM is considered to be hypoperfusion-ischemia in the intermediate and deep capillary plexus of the retina [1]. The PAMM has been reported to be associated with several conditions [9]. Despite the normal fundus appearance on ophthalmoscopy, hypo-reflectance on infrared image and hyperreflectivity in the inner nuclear layer in structural OCT examination are often found [1,9]. The presence of PAMM following cataract surgery is rarely reported [2–4]. We believe that the cardiovascular condition in the present case, along with dehydration due to fasting and surgical trauma, might contribute to the development of PAMM. Furthermore, effective fasting during Ramadan with no water consumption might lead to blood thickening resulting in PAMM development. In contrast to other reports in the literature, our case had a better visual outcome at the final follow-up, which may be due to early diagnosis and prompt referral to HBOT [2–4].

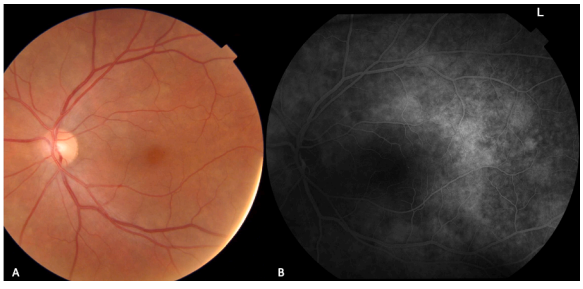

Fig. 2. Representative (A) color fundus imagining and (B) fundus fluorescein angiography on the 5th day after PAMM diagnosis (L: left eye).

Authors explained the development of PAMM after cataract surgery with the following hypotheses: arterial hypo-perfusion due to spasm or temporary occlusion of the retinal artery; the increase in orbital pressure related to anesthetic administration via subtenon, retrobulbar or peribulbar route, and/or the increase in intraocular pressure by phacoemulsification; abnormal IOP values at the end of surgery and an increase in IOP postoperatively (e.g., overhydration of corneal wounds, overly tight padding, viscoelastic retention) [2–4]. We utilized subtenon anesthesia in our case. The viscoelastic substance was thoroughly removed after intraocular lens implantation, and IOP was at normotensive limits at the end of the surgery. As a routine procedure, oral acetazolamide was given to prevent postoperative IOP spikes. Although IOP was normal limits on the first and third postoperative days, subclinical transient IOP elevations could not be totally excluded.

Our case underwent uncomplicated cataract surgery in his right eye by the same surgeon with the same procedure one year ago, and no postoperative complication developed. But the patient was fasting during recent cataract surgery. When we reviewed the literature, we could not find any article on the relationship between fasting and dehydration and PAMM. Still, previous studies have shown that hypovolemia could be a precipitating factor in the development of PAMM [10]. It has been known that fasting and dehydration increase blood hyperviscosity and related complications [11]. Dehydration might induce PAMM development via increased blood hyperviscosity in the presence of underlying cardiovascular disorder in our case. To the best of our knowledge, we report a possible association between fasting and PAMM development for the first time. However, there could be a number of other important factors as stated above that might have contributed to PAMM in the present case as well.

There is no current definitive treatment for PAMM. In a recent publication, low-dose aspirin was recommended to prevent vision loss due to PAMM [12]. Hyperbaric oxygen therapy has been applied to PAMM cases in the presence of accompanying arterial occlusion [13–15]. A recent case report demonstrated that HBOT results in favourable outcomes in acute macular neuroretinopathy (AMN) [16]. It has been known that AMN has clinical resemblance with PAMM sharing a similar mechanism regarding ischemia of the deep retinal vascular network. A recent study described the thinning effect of HBOT on the macula and suggested that HBOT can be used in vascular diseases affecting the macula [17]. Given that PAMM is an ischemia related disorder and no definitive treatment is present, we decided to employ HBOT empirically after a detailed discussion with the patient. And we observed improved visual outcome at last examination after HBOT. No literature data exists regarding HBOT in cases with isolated PAMM. But, as profound visual loss in the present case showed a dramatic improvement following HBOT, we may suggest a possible use of HBOT in PAMM treatment, at least based on the anecdotal evidence provided from the current case. However, hyperbaric oxygen chambers may not be widely available, hence patient referral should be considered in such situations in order to initiate a prompt treatment. Nevertheless, a case

report may not be enough to justify HBOT in PAMM patients.

Treatment with hyperbaric oxygen is associated with visual improvement in retinal artery occlusion, which should be a differential diagnosis in the present case. However, no evidence of retinal whitening or pallor, 'cherry red' macula, opacification of the nerve fiber layer and a thrombotic plaque was noted on fundus examination during the initial visit after cataract surgery. Although we could not totally exclude at least the presence of a partial retinal arterial occlusion, observation of hyporeflective lesion on infrared imaging limited to parafoveal area as well as prominent hyperreflectivity in the middle retinal layers on OCT suggested PAMM in our case. Another factor that may have caused all these could be the inadvertent administration of a higher dose of intracameral cefuroxime at the end of the surgery. However, we did not find any evidence to support this theory.

4. Conclusions

Dehydration due to fasting might play a role in PAMM development after cataract surgery with subsequent infarction of the inner retina along with comorbid cardiovascular compromise in the present case. Although the patient had a profound visual loss at the time of presentation, we observed a dramatic improvement in visual acuity after HBOT. However, triggering of PAMM by fasting is entirely unproven and that this observation occured in a highly complex case with many other possible contributing factors. Also, the triggering of PAMM by some manipulation during surgery is equally unproven. Furthermore, HBOT has successfully been applied in PAMM in the setting of retinal arterial occlusion so far. This might have occured in our case due to the operation, even though we did not find any associated finding supporting the presence of accompanying arterial occlusion. Although this anecdotal evidence may suggest a potential use of HBOT in the treatment of PAMM, studies with a larger patient population are needed in order to validate the results of the current work and to better ascertain the impact of HBOT on PAMM regression.

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Informed consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

CRediT authorship contribution statement

Betul Onal Gunay: Writing – review & editing, Writing – original draft, Visualization, Validation, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

Declaration of Competing Interest

The authors have no financial or proprietary interests in any material discussed in this article.

Data availability

All data will be made available upon request.

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