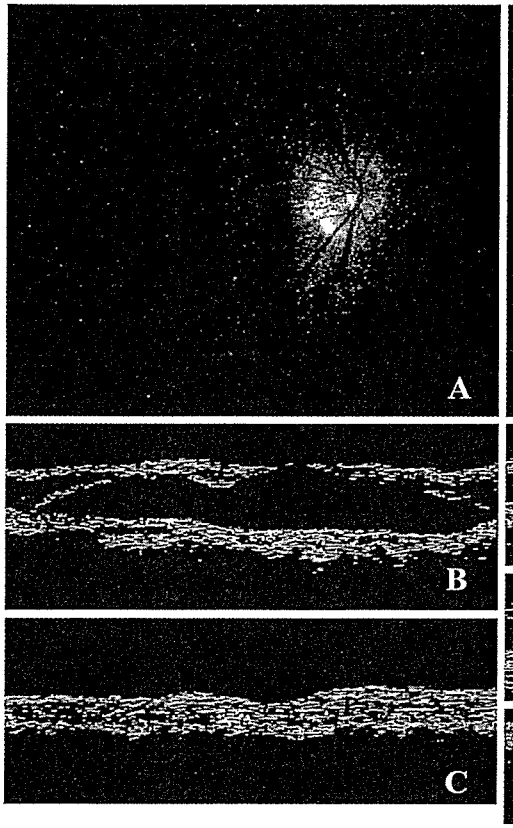

Figure 1

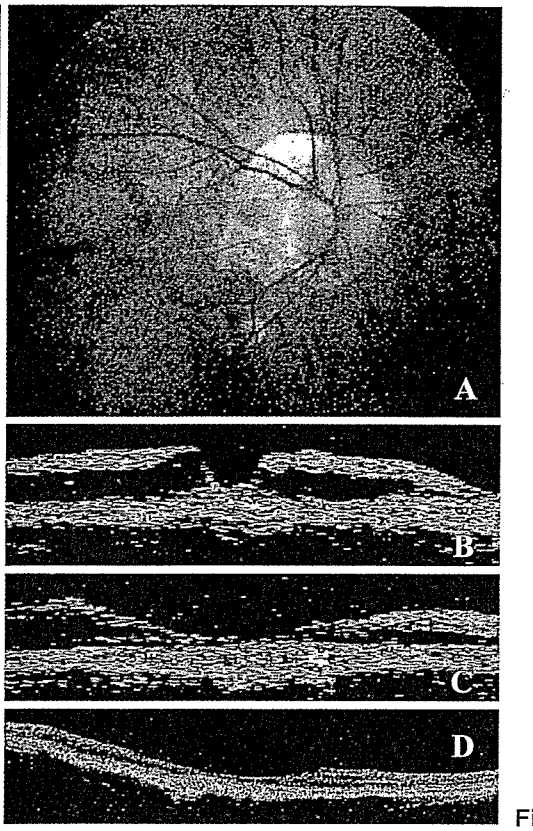

Figure 2

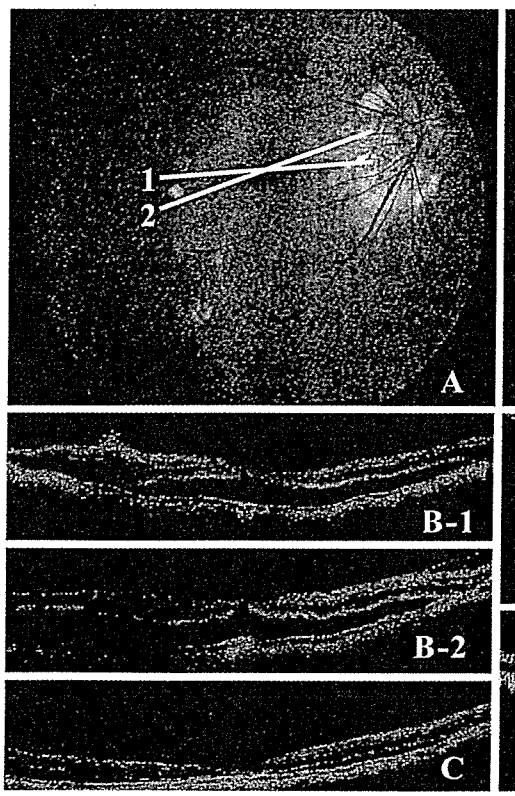

Figure 3

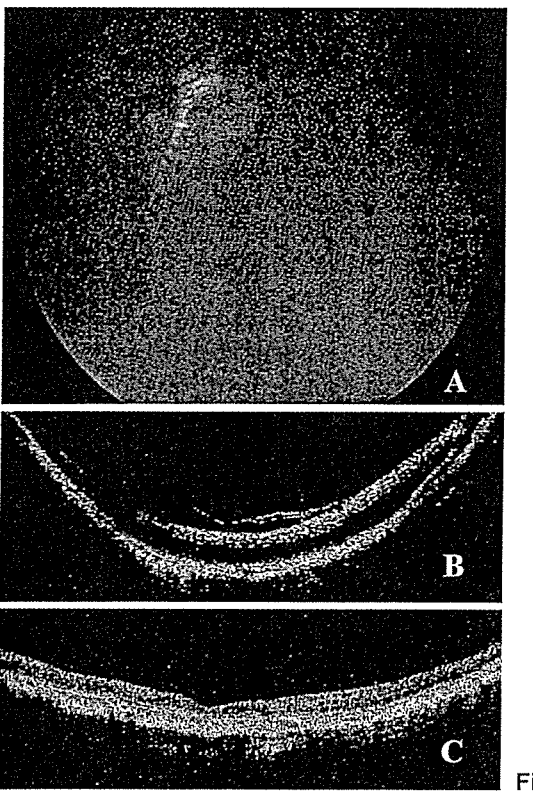

Figure 4

history of von Recklinghausen disease and refractive amblyopia. Examination of the right eye revealed a BCVA of 0.5 and a shallow serous macular detachment over the posterior staphyloma (Fig. 1A). OCT confirmed the presence of FD and RS (Fig. 1B).

After the patient had been observed for 3 months, he complained of increasing metamorphopsia, although an examination of his vision revealed no change. Vitrectomy was performed, with removal of the posterior hyaloid membrane and ILM peeling by viscodissection, followed by air–fluid exchange and 14% C_3F_8 gas tamponade.

Two months postoperatively, the BCVA was 0.6, and an OCT examination showed a marked resolution of the retinal detachment. The patient reported an improvement in the metamorphopsia. Six months postoperatively, the patient developed nuclear sclerosis of the lens, so phacoemulsification with intraocular lens implantation was performed. Consequently, the patient's BCVA further improved to 0.7. The posterior retina was found to be completely reattached, with no further changes during a 54-month follow-up examination. An OCT examination about 4 years later revealed a nearly normal configuration of the fovea (Fig. 1C).

Patient 2

A 53-year-old woman with a history of vitrectomy for a macular hole retinal detachment in her left eye 3 years earlier was noted to have a shallow elevation of the macula in her asymptomatic right eye during a routine follow-up examination. OCT revealed RS in the right eye.

One year later, she complained of a slight decrease in central vision and metamorphopsia in her right eye. Although her visual acuity had decreased from 0.8 to 0.4, a fundus examination showed no obvious changes in the severity of the RS (Figs. 2A, B). Vitrectomy and phacoemulsification with intraocular lens implantation were performed. Intraoperatively, marked syneresis of the vitreous was observed. Following core vitrectomy, we were able to easily induce posterior vitreous detachment (PVD) by suctioning with the vitreous cutter everywhere except over the posterior pole; ILM peeling was necessary to ensure the complete removal of the posterior hyaloid inside the staphyloma. Fluid–air exchange without subretinal drainage was performed, followed by gas tamponade with 14% C_3F_8. Two months after surgery, the patient reported that her metamorphopsia had diminished. An OCT examination showed a marked resolution of the RS (Fig. 2C). Six months after surgery, although the BCVA remained unchanged at 0.5, the patient reported an improvement in the metamorphopsia. Four years after the operation, an OCT examination confirmed the complete reattachment of the retina (Fig. 2D), and the patient's BCVA had been restored to 0.8.

Patient 11

A 70-year-old woman with a history of phacoemulsification with intraocular lens implantation complained of a 4-month history of metamorphopsia in her right eye. The patient's BCVA was 0.4, and a shallow retinal elevation was noted extending from the superotemporal to the inferotemporal arcades over posterior staphyloma (Fig. 3A). No retinal breaks were detected. OCT revealed an RS that appeared to be connected to the conus of the optic disc, as well as a large outer layer detachment at the macula and a partial posterior hyaloid separation surrounding the RS (Fig. 3B).

Vitrectomy and 20% SF_6 gas tamponade was performed with the adjunctive use of TA intraoperatively. The use of TA to observe the posterior hyaloid intraoperatively appears to be a useful technique when attempting to completely separate tight adhesions to the retina. One month after the surgery, the patient reported that her metamorphopsia had diminished. The patient's BCVA was 0.5, and an OCT examination showed a marked improvement in the inner layer separation and outer layer detachment. At 6 months after the operation, an OCT examination showed complete retinal reattachment (Fig. 3C). One year after the operation, the BCVA had been restored to 0.7.

Preoperative Clinical Characteristics

The clinical characteristics of all 14 patients are shown in Table 1. Eleven patients were women and three were men, and they ranged in age from 53 to 77 years (mean ± SD, 64.8 ± 7.7 years). All the patients were healthy and of Japanese ancestry, and all had complained of metamorphopsia or reduced vision in the affected eye for several months. The refractive errors ranged from –6.0 to –19.25 diopters (mean ± SD, –13.2 ± 3.8 D) in 12 phakic eyes. The axial lengths ranged from 24.9 to 30.4 mm (mean ± SD, 27.6 ± 1.6 mm). The eye of patient 7 was not highly myopic, but posterior staphyloma and glaucomatous cupping of the optic disc were present. The posterior staphyloma extended over a wide area in all 16 eyes. The decimal preoperative visual acuity values ranged from 0.01 to 0.5 (mean, 0.14).

OCT examinations disclosed various profiles of macular change. The presence of RS or FD was confirmed preoperatively in all eyes. Five eyes had RS alone. In four of these eyes, an extensive hyporefractive space had split the retina into a thick inner layer and a thin outer layer lying on the retinal pigment epithelium (RPE) [patients 1 (left eye, L), 3, 5 (R), and 6] (Fig. 4A, B). One eye (patient 2) had a similar appearance, but the fovea was not split and did not show a defect in the roof of the central cyst, giving the appearance of a lamellar hole (Fig. 2B). The BCVA in four of the five eyes with RS without FD was better than 0.2.

Eleven eyes had RS associated with FD (Figs. 1B, 3B, 5B). The BCVA of these eyes seemed to be worse than that

Table 1. Clinical characteristics

Patient	Age (years)	Sex	Eye	Ref (D)	Axial length (mm)	Symptom	Preoperative BCVA	OCT finding		Intraoperative finding				Final attachment	Final BCVA	Complications	Follow-up (months)	Fellow eye
								RS/FD	PVD	PVD Ind.	ILM peel	Tamp						
1	59	M	L	−8.75	26.7	Decreased VA	0.06	RS	—	+	−	C_3F_8		+	0.2	—	66	RS,FD
			R	−11.00	27.0	Decreased VA	0.5	RS,FD	—	+	+	C_3F_8		+	0.7	—	54	RS
2	53	F	R	−19.25	28.7	Metamorphopsia	0.4	RS	—	+	+	C_3F_8		+	0.8	—	52	MHRD
3	72	F	R	−9.75	27.9	Metamorphopsia	0.4	RS	—	+	+	—		+	0.8	—	45	—
4	75	F	R	IOL	26.2	VF defect	0.01	RS,FD	—	+	+	C_3F_8		+	0.07	MH	17	—
5	68	F	L	−15.00	27.5	Decreased VA	0.06	RS,FD	Part	+	−	SF_6		+	0.06	RRD	16	RS
			R	−17.75	30.0	Metamorphopsia	0.2	RS	Part	+	−	SO		+	0.6	Retinal break	10	RS,FD
6	63	F	L	−12.5	29.2	Metamorphopsia	0.4	RS	—	+	−	—		+	0.4	—	27	—
7	63	M	L	−6.0	24.9	Decreased VA	0.1	RS,FD	—	+	+	SF_6		+	0.5	—	27	Gla
8	54	F	R	−15.00	27.0	Metamorphopsia	0.01	RS,FD	Part	+	+	SF_6		+	0.1	MHRD	22	—
9	56	F	L	−14.00	29.2	Decreased VA	0.2	RS,FD	Part	+	−	SF_6		+	0.1	MHRD	17	RS,FD
10	77	F	L	IOL	27.4	Metamorphopsia	0.3	RS,FD	Part	+	−	SO		+	0.3	—	15	MHRD
11	70	F	R	IOL	30.2	Metamorphopsia	0.4	RS,FD	Part	+	−	SF_6		+	0.7	—	13	—
12	74	F	R	IOL	29.9	VF defect	0.06	RS,FD	Part	+	−	SF_6		+	0.2	—	6	RS
13	66	M	L	−15.00	29.6	Decreased VA	0.3→0.1	RS,FD→ MHRD	—	+	−	C_3F_8		+	0.2	MH	11	MHRD
14	59	F	R	−19.00	30.4	Decreased VA	0.2→0.06	RS,FD→ MHRD	—	+	−	C_3F_8		+	0.4	MH	12	—

BCVA, best-corrected visual acuity; C_3F_8, perfluoropropane; D, diopter; F, female; FD, foveal detachment; Gla, glaucoma; ILM, internal limiting membrane; Ind, induction; IOL, intraocular lens; L, left; M, male; MH, macular hole; MHRD, macular hole retinal detachment; OCT, optical computed tomography; Part, partial detachment; PVD, posterior vitreous detachment; R, right; Ref, refractive error; RRD, rhegmatogenous retinal detachment; RS, retinoschisis; SO, silicone oil; SF_6, sulfur hexafluoride; Tamp, tamponade; VA, visual acuity; VF, visual field.

of the eyes without FD, but 6 of the 11 eyes with FD had a BCVA that was better than 0.2. Two eyes developed macular hole retinal detachment during the routine follow-up period, and the BCVA decreased from 0.3 to 0.1 in patient 13 and from 0.2 to 0.06 in patient 14. Figures 6 and 7 show the changes in the macular configurations associated with the reduction in BCVA during the follow-up period in patients 13 and 14.

The presence of a partially detached posterior hyaloid was disclosed in seven eyes (Figs. 3, 4, 5). The separation of the posterior hyaloid beside the conus of the optic disc was observed in all of these seven eyes. Advanced vitreous syneresis was observed in all of these eyes, but PVD was not observed in the other nine eyes preoperatively.

Regarding the fellow eyes of the patients, three patients had a medical history of macular hole detachment, five patients had a history of RS and/or FD, one patient had a history of myopic posterior chorioretinal atrophy, and one patient had a history of primary open-angle glaucoma.

Anatomical Results

In all five eyes that had RS without FD, the retina reattached after the initial vitrectomy. Retinal reattachment was achieved in 8 out of 11 eyes with both RS and FD, including two cases that had progressed to macular hole retinal detachment (patients 13 and 14) before the initial vitrectomy. We removed the silicone oil about 5 and 2 months after the vitrectomies in patients 5 (R) and 10, respectively. The retina remained reattached after the removal.

Three eyes required reoperation because of recurrent retinal detachment. In two of these three eyes, a full-thickness macular hole associated with posterior retinal detachment occurred 1 month after vitrectomy with (patient 8) and without ILM peeling (patient 9; Fig. 8). We performed a second operation, consisting of extensive ILM peeling and silicone oil tamponade. At 5 months (patient 8) and 3 months (patient 9) after the second surgery, we removed the silicone oil, and retinal reattachment was confirmed by OCT in both eyes. One eye (patient 5, L) developed retinal detachment caused by a peripheral retinal break immediately after the initial vitrectomy. A second vitrectomy was thus performed to repair the retinal detachment.

Final retinal reattachment was achieved in all 16 eyes. No recurrences were observed during follow-up.

Visual Acuity Results

An improvement in the BCVA of 0.2 logMAR or greater was documented in 9 of the 16 eyes. The BCVA of seven eyes remained unchanged. The BCVA of the left eye of patient 5 improved from 0.06 to 0.1 at 12 months after the vitrectomy, but decreased again to 0.06 at 16 months, with no remarkable changes in the retinal findings. During this follow-up period, we performed a vitrectomy for the treat-

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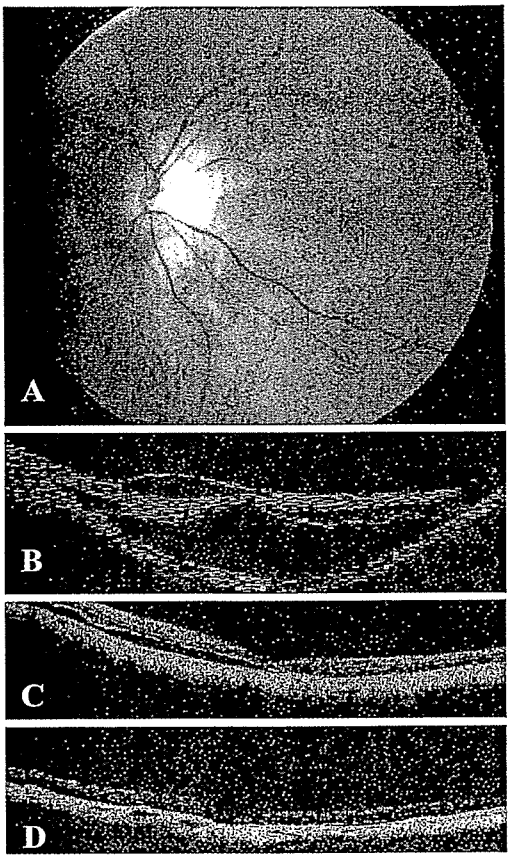

Figure 5

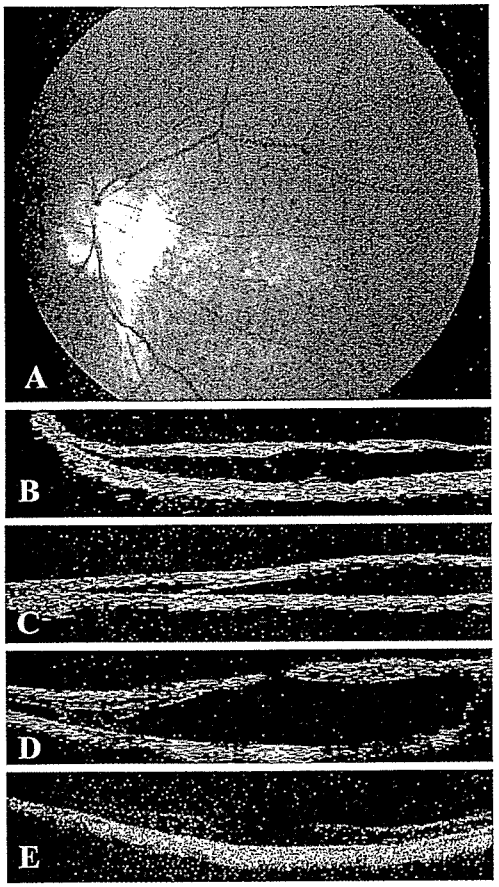

Figure 6

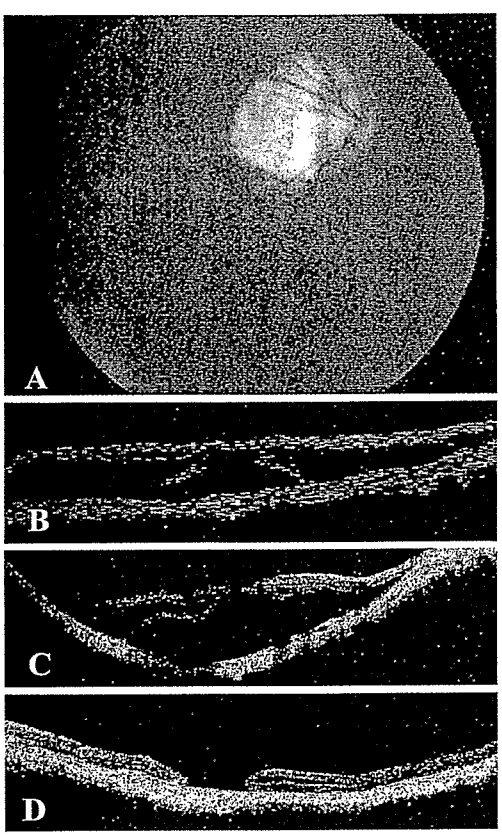

Figure 7

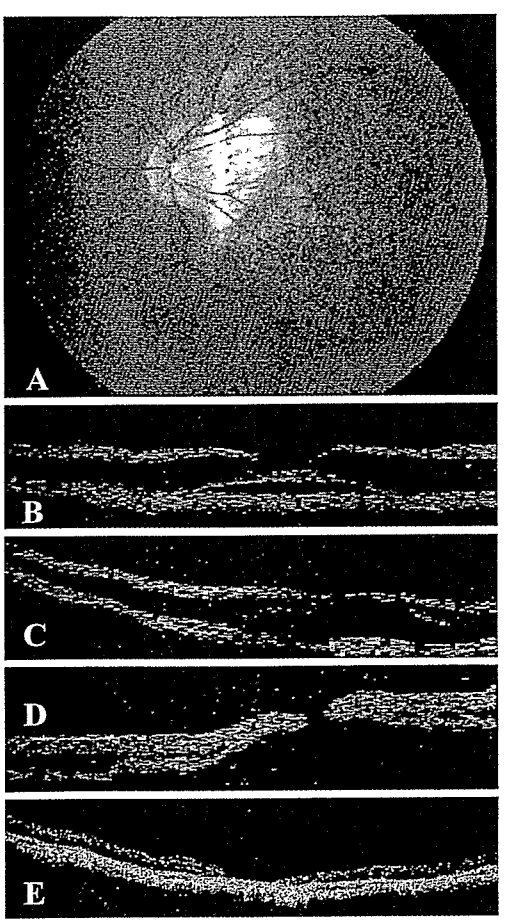

Figure 8

ment of RS in the right eye, which resulted in an improvement of VA from 0.2 to 0.6.

Fourteen of the 16 eyes had a final VA of 0.1 or better, and six eyes had a BCVA of better than 0.5.

Complications

Intraoperative or postoperative complications were observed in seven eyes. The retina redetached in three eyes [patients 5 (L), 8, and 9], as described above.

A full-thickness macular hole was observed after the initial vitrectomy in 5 of the 16 eyes. In four of these five eyes, the full-thickness macular hole associated with the retinal detachment appeared before (patients 13 and 14) or after the vitrectomy (patients 8 and 9), as described above. In the fifth eye (patient 4), a macular hole was found in the reattached retina after vitrectomy with ILM peeling and C_3F_8 gas tamponade. All five of these eyes had severe preoperative FD associated with RS and a poor VA.

Patient 5 developed a peripheral retinal break in the right eye intraoperatively during PVD induction that was treated successfully with laser photocoagulation.

Discussion

We confirmed that OCT is a very important tool for observing the pathological appearance of highly myopic eyes, as concluded in previous reports.[2–9] These previous reports also noted that RS and FD were not uncommon. Baba and associates[8] reported that none of seven patients with foveal retinal detachment complained of recent, progressive visual impairment. In addition, one case report described the spontaneous resolution of a myopic foveal retinoschisis.[13] Benhamou and associates[9] reported that this condition is fairly stable, in terms of visual acuity and retinal thickness, and changes slowly over time. In the present study, we studied eyes with RS and/or FD and progressive visual impairment. Fourteen of the 16 eyes exhibited a symptomatic visual impairment at the time of the initial patient visit, and the other two eyes were noted to have asymptomatic RS during routine follow-ups for the other eye, with subsequent visual impairment in the following few years. Two of the 11 eyes associated with FD developed macular hole retinal detachment during the preoperative follow-up period. The patients noticed an acute decrease in BCVA after the development of macular hole retinal detachment. In one eye, OCT revealed changes in the macular appearance during a follow-up examination, with no accompanying visual disturbance. This change detected by OCT and the symptomatic visual impairment associated with RS or FD may reflect the high risk of developing a macular hole.

A high incidence of macular hole retinal detachment in the opposite eye (in 3 out of 14 patients) and the preoperative progression leading to the development of macular hole retinal detachment in 2 out of the 16 eyes support the hypothesis of Takano et al.,[2] who suggested that RS and FD in highly myopic eyes may precede macular hole retinal detachment.

In this study, OCT examination revealed various macular profiles of myopic RS and FD, similar to the description by Benhamou et al.[9] RS involving the entire posterior pole connected to the conus of the optic disc was observed in all 16 cases; thus, we would like to propose that the term "posterior retinoschisis" is more appropriate than "macular retinoschisis" or "foveal retinoschisis." Although the BCVA in eyes with FD seemed to be poorer than that in eyes without FD, six eyes had a BCVA of better than 0.2, and the presence of FD could not be determined based only on visual acuity.

Figure 5A–D. Fundus photograph and OCT images of the left eye of patient 10. **A** A preoperative fundus photograph shows a shallow macular detachment in an eye with a BCVA of 0.3. **B** A preoperative OCT image shows a marked elevation of the posterior retina. A posterior retinoschisis is visible from the edge of the conus of the optic disc to the edge of the posterior staphyloma. The outer layer detachment is remarkable. A partial separation of the posterior hyaloid is visible between the fovea and the conus of the optic disc (scan length, 9.0 mm). **C** Two months after the vitrectomy, the posterior retina elevation was remarkably reduced (scan length, 10.0 mm). **D** Fifteen months postoperatively, the retina had completely reattached (scan length, 10.0 mm).

Figure 6A–E. Fundus photograph and OCT images of the left eye of patient 13. **A, B** A 66-year-old man with a history of macular hole retinal detachment surgery in the right eye was noted to have posterior retinoschisis of the asymptomatic left eye during a routine follow-up examination. The BCVA of the left eye was 0.5 (scan length, 9.0 mm). **C** After observation for 20 months, an OCT examination revealed the development of a foveal detachment associated with the posterior retinoschisis. The BCVA had decreased to 0.3, but the patient had not noticed the development of any visual disturbance (scan length, 9.0 mm). **D** After observation for 30 months, he complained of reduced vision and scored a BCVA of 0.1. An OCT examination revealed a posterior retinal detachment associated with a macular hole, and vitrectomy was performed (scan length, 10.0 mm). **E** Ten months postoperatively, the retina had completely reattached, but the macular hole persisted (scan length, 5.0 mm).

Figure 7A–D. Fundus photograph and OCT images of the right eye of patient 14. **A** A fundus photograph taken at presentation shows a shallow posterior detachment over a posterior staphyloma in an eye with a BCVA of 0.5. **B** An OCT image taken at presentation shows a foveal detachment with posterior retinoschisis (scan length, 9.0 mm). **C** Three months after presentation, the BCVA had decreased to 0.06; an OCT image shows macular hole retinal detachment. A vitrectomy was performed (scan length, 10.0 mm). **D** Three months after the operation, an OCT image shows complete retinal reattachment, but the macular hole remains visible (scan length, 5.0 mm).

Figure 8A–E. Fundus photograph and OCT images of the left eye of patient 9. **A** A preoperative fundus photograph shows a shallow posterior detachment over a posterior staphyloma in an eye with a BCVA of 0.2. **B** A preoperative OCT image shows posterior retinoschisis and foveal detachment (scan length, 2.8 mm). **C** Two weeks after the vitrectomy, the posterior retinoschisis elevation has decreased, but foveal detachment has increased (scan length, 5.0 mm). **D** One month after the vitrectomy, the patient noticed a reduced BCVA. An OCT image shows the development of macular hole retinal detachment (scan length, 5.0 mm). **E** About 1.5 years postoperatively, the retina had completely reattached, but the macular hole remained visible (scan length, 5.0 mm).

In 7 of the 16 eyes, OCT revealed the presence of a detached posterior hyaloid surrounding the macula, which may have widely stretched the posterior pole. The appearance of posterior vitreous adhesions over the macula was consistent with our experience of performing vitrectomies for retinal detachment associated with a macular hole[14] and a previous clinicopathological report,[15] which suggested that the posterior hyaloid might remain tightly attached to the macula, despite the presence of PVD in highly myopic eyes. Although PVD was not observed in the other nine eyes preoperatively, OCT examinations may be limited in detecting preretinal structures in highly myopic cases. In the present study, final reattachment was obtained in all 16 eyes, and no recurrences were observed in any eyes for over 6 months (the mean follow-up period was 23.3 months) after final surgery. This result suggests that the release of vitreous traction at the posterior pole may have an important role in the treatment of myopic RS and FS.

We performed vitrectomy, including vitreous cortex removal, in all eyes and internal limiting membrane (ILM) peeling in six eyes. All five eyes with RS and without FD achieved retinal reattachment after the initial vitrectomy. However, 3 of the 11 eyes with RS and FD required reoperation after the initial vitrectomy. In two of these three eyes, a full-thickness macular hole associated with posterior retinal detachment occurred about 1 month after vitrectomy with or without ILM peeling. The incidence of the development of macular hole retinal detachment seems to be higher than in previous reports,[3–7] in which most cases received ILM peeling. Kuhn[6] suggested that ILM may be responsible for macular detachment in highly myopic eyes. We also observed the proliferation of glial cells, which cause an abnormal ILM figure in highly myopic eyes.[14] ILM peeling may be highly beneficial for reducing the traction on the detached retina. However, the side effects of ILM peeling remain unknown. In addition, ILM peeling is technically difficult in highly myopic eyes, and ICG may be toxic to the neural retina as well as to the RPE.[16] The number of surgical reports remains insufficient, and we were able to obtain retinal reattachment in most of the eyes without ILM peeling. Thus, we were unable to conclude whether ILM peeling leads to a better anatomical prognosis. However, current techniques, such as TA-assisted vitrectomy or viscodissection, may be useful to avoid ILM peeling.

The role of gas tamponade in treatment is also uncertain. Gas tamponade can induce pneumatic displacement of outer-layer detachments and improve vision in retinoschisis associated with optic disc pits.[17] We expected intravitreal tamponade to have a similar effect in myopic eyes with RS and FD. However, intravitreal tamponade may push the subretinal fluid inside the limited area of the posterior staphyloma toward the weak point of the fovea, causing the formation of a macular hole. In two eyes with RS but without FD, retinal reattachment was obtained without using gas tamponade. Further study is needed to optimize surgical techniques.

Progression to a macular hole occurred preoperatively in 2 out of the 16 eyes and postoperatively in 3 out of 14 eyes. These eyes had relatively severe FD with posterior staphyloma prior to the development of a macular hole, and the BCVA of these eyes was worse than that of the others. These cases might have been at an advanced stage following the early development of a macular hole, similar to the two cases reported by Ikuno and Tano.[18] Patients and surgeons should regard this probable advanced stage with severe FD and poor vision as being equivalent to the early stage of macular hole retinal detachment when treating this condition.

Visual acuity improved or remained stable in all the eyes in the present study. Considering the poor prognosis of macular hole retinal detachment, vitrectomy using modern surgical techniques to induce PVD or to perform ILM peeling may be effective for the treatment of RS and/or FD in highly myopic eyes. However, further study is needed to select surgical indications and optimize surgical techniques to avoid complications, including macular hole.

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タイトル：拍動性眼球陥没凹を認めた2例
英文タイトル：Two cases of pulsating enophthalmos

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拍動性眼球陥凹を認めた2例

Two cases of pulsating enophthalmos

＜要約＞

眼窩脳髄膜瘤に片眼の拍動性眼球陥凹というまれな症状を合併した 2 症例を経験したので報告する。症例 1 は 59 歳女性。幼少期より神経線維腫を認め、von Recklinghausen 病と診断されていた。右眼の拍動性眼球陥凹を認め、眼窩 CT にて右蝶形骨の欠損と眼窩脳髄膜瘤を認めた。症例 2 は 75 歳男性。右眼の拍動性眼球陥凹を認め、眼窩 CT にて蝶形骨大翼の欠損と眼窩脳髄膜瘤を認めた。症例 2 については von Recklinghausen 病によるものが強く疑われたが確定診断はできなかった。拍動性眼球陥凹の予後の報告は少なく、球後麻酔等眼窩手術時の危険性もあるため患者への病態説明と慎重な経過観察が必要と思われた。

Abstract

We report two cases of unilateral pulsating enophthalmos complicated by orbital meningocele. A 59-year-old woman exhibited typical features of neurofibromatosis (von Recklinghausen disease), including neurofibroma and iris nodules in both eyes, and a 75-year-old man was suspected of having a previous history of neurofibromatosis. Computed tomography studies performed in both cases revealed the absence of sphenoid wings and encephalocele in the side of the pulsating enophthalmos. While the prognosis and pathogenesis of pulsating enophthalmos remains uncertain, the present clinical study suggests that continuous and careful monitoring of symptoms is necessary.

Key Words: pulsating enophthalmos, von Recklinghausen disease, encephalocele

緒言

眼球拍動を呈する疾患としては、頚動脈海綿静脈洞瘻、眼窩動静脈奇形、血管腫、眼窩脳髄膜瘤などが知られており、これらは一般的には眼球突出を呈する[1)]。今回我々は、眼窩脳髄膜瘤に、拍動性眼球陥凹というまれな症状を合併した2症例を経験したので報告する。

症例

症例1:59歳女性

主訴:複視

現病歴:平成14年より複視を自覚し近医を受診したところ、眼窩CTにて右眼窩壁の欠損を指摘された。その後放置していたものの視力低下が増悪し、平成16年5月14日当科受診した。

既往歴:幼少期より神経線維腫を認め、von Recklinghausen病と診断されていた。

眼科的所見:視力は右0.04(0.4×-9.5D=cyl-3.0DAx85°)、左0.01(0.2×-15.0D=cyl-3.5DAx90°)。眼圧は右眼12mmHg、左眼14mmHg。眼位は右内斜視。CUT(cover-uncover test)右 内斜視。角膜径は左右差なし。両眼に虹彩結節を認めた。両眼に中等度の白内障を認めたが、左眼の皮質白内障が右眼より強かった。両眼底には強度近視に伴う網脈絡膜萎縮がみられた。Hertel眼球突出計にて右眼13mm、左眼17mmと右眼が陥凹を示し(図1)、脈拍に一致して前後に約2mmの拍動がみられたが、bruitは聴取されなかった。眼球運動で右眼の外転障害がみられた。全方向で拍動に伴う複視があり、特に上下方向で訴えが強かった。

全身所見:顔面・頚部に神経線維腫が存在した。

放射線的検査:眼窩Computed Tomography(CT)にて右蝶形骨大翼の欠損と、眼窩脳髄膜瘤を認めた(図2)。腫瘍性病変は認めなかった。

経過:拍動性眼球陥凹に伴う上下方向の複視のみでは日常生活に大きな支障はないと考えられた。また視力低下の増悪は白内障によるものと考えられたが、手術の希望なく病態を説明して経過観察となった。3年間の経過で著明な変化はみられていない。

症例2:75歳男性

主訴:複視

現病歴:平成12年より複視を自覚した。改善しないため近医受診し右の拍動性眼球陥凹を指摘され、精査目的で平成14年8月9日当科受診した。

既往歴:昭和60年頃右前頭葉出血。平成4年交通外傷による腹部出血。外傷による頭蓋内病変はなかった。また、平成11年、ウイルス性小脳炎に罹患している。

家族歴:妹 喉頭がん

全身所見:背部に色素沈着あり

眼科的所見:視力は右0.9(n.c.)、左1.2(n.c.)。眼圧は右眼9mmHg、左眼8mmHgであった。眼位は正位。CUT正位。角膜径は左右差なし。虹彩結節は認めなかった。中間透光体では両眼に軽度の白内障を認めた。眼底には両眼とも異常はなかった。ヘス赤緑試験において軽度上転、

内転、外転障害を認めた。また両眼単一視野は 15° であった。拍動によって像がぶれることに加え、上方・右方視・左方視で複視を自覚していた。眼球突出度は Hertel 眼球突出計にて右眼 10mm、左眼 14mm と右眼が陥凹を示し(図3)、前後に脈拍に一致した拍動を認めた。bruit は聴取されなかった。

放射線的検査:眼窩 CT にて蝶形骨大翼欠損と眼窩脳髄膜瘤を認めた(図4)。また右前頭葉には出血の後遺症と思われる萎縮がみられた。

経過:拍動性眼球陥凹、軽度の複視だけでは日常生活には支障をきたさないため経過観察とした。3 年の経過で変化はみられていない。

考按

眼窩脳髄膜瘤はほとんどが von Recklinghausen 病に合併したものである[1,2,3]。脳髄膜瘤発症のメカニズムは、眼窩の後壁を構成する蝶形骨大翼の欠損部を通って脳実質が眼窩内に脱出したものであるが、骨欠損の原因には先天性骨形成不全説[4]と神経線維腫による二次的変化であるとする説[5]とがある。症例 1 は全身所見から von Recklinghausen 病と診断されているが、蝶形骨周囲には画像上明らかな腫瘍性病変がみられなかったので、前者と思われる。

本症例の注目すべき所見は眼球が陥凹を呈していながら前後に拍動するという印象的な臨床症状である。拍動の原因については、1961 年に Hunt ら[6]は、前頭骨と蝶形骨の欠損部位を通して、脳の脈動が眼窩内に伝達されるために拍動性眼球突出を呈すると述べ、この骨病変を von Recklinghausen 病に特有なものであるとしている。

今回の 2 症例においても、眼球の拍動は脈拍に一致し、頚動脈海綿静脈洞瘻などの血管病変を示唆する bruit も聴取されなかったことから、眼窩内に陥入した脳実質の脈動が眼球に伝達されたものと考えられた。ただ、このような脳髄膜瘤を原因とする眼球拍動は、1933 年に LeWald ら[7]が報告して以来、眼球突出を呈することが通例であり[8-10]、眼球陥凹を伴った報告は極めて少ない[1-3]。

眼球陥凹とは、眼窩内容が眼窩容積に比して相対的に減少する際に生じるもので、それは実際に内容が減少する場合と、眼窩の拡大による場合とがある。von Recklinghausen 病では眼窩の拡大がしばしば見られ、それによって眼球陥凹が生じたとする報告がある[1)]。今回の 2 症例では、CT にて明らかな眼窩の拡大がないので、長期間持続した脳髄膜瘤による圧迫のため、眼窩脂肪織が萎縮し、眼球陥凹に至ったものと考えられる。これまでにも、頚動脈海綿静脈洞瘻、眼窩静脈瘤などでそのような変化が起こることが知られている[11)]。

症例 2 の眼窩後壁欠損の原因は明らかではなかった。外傷の既往はあるが、傷害の主体は腹部で頭蓋骨骨折の記録はない。また、前頭葉出血の既往もあるが、開頭手術は受けておらず、血腫が長期間蝶形骨大翼を圧迫していたという記録もない。背部の色素沈着は皮膚科の診断では炎症後の色素沈着で von Recklinghausen 病を示唆する所見ではないとのことであったが、von Recklinghausen 病の表現形は多彩で、自分が病気であることに気付かない人もいることから、症例 2 においても von Recklinghausen 病が潜んでいる可能性は否定できない。また von Recklinghausen

病では80%程度みられるとされている虹彩結節[12]は認められなかったが年齢と共に目立たなくなった可能性もあると考えられた。von Recklinghausen病は17番染色体上の突然変異で起こるとされているが、原因遺伝子が巨大で遺伝子解析が難しいとされている。今回は本人が希望しなかったことより、遺伝子検査は行っていない。

治療については、今回の2症例では、拍動性眼球陥凹はあるものの、複視は軽度であり、日常生活には大きな支障をきたさなかったために経過観察とした。ただ、脳髄膜瘤が進行すれば、視神経、眼球運動神経、外眼筋が圧迫され、さらに高度の視機能、眼球運動障害をきたす可能性がある。しかし、眼窩後壁は複雑な立体構造を呈すために再建困難であるとする報告が多い[13]。

本症例のように脳髄膜瘤を認めている症例に対して球後麻酔を行った場合、球後針が脳髄膜瘤の範囲に到達してしまう可能性が高い。そのまま麻酔を施行すると、麻酔薬が脳脊髄液中に誤注入され、脳神経麻痺や脳幹麻痺を引き起こす可能性がある。[14]そのような危険性もふまえ、患者への病態説明と慎重な経過観察が必要と思われた。最後に、拍動性眼球陥凹という臨床所見は極めて稀なものであり、その原因のほとんどはvon Recklinghausen病によるものであるということを念頭に検査を進める必要があると思われた。

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LETTER TO THE EDITOR

Enlargement of optic nerve resembling orbital mass in case of optic neuritis

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Dear Editor,

Analyses of magnetic resonance images (MRI) and cerebrospinal fluid (CSF) can provide information to make a differential diagnosis of optic neuritis from other optic nerve diseases [1, 2]. In rare cases however, those findings resemble those of optic nerve tumors [3, 4]. We present a patient with acute visual loss and an unusual swelling of the optic nerve on MRI which resembled an optic nerve tumor but was found to have optic neuritis.

A 20-year-old woman complained of a sudden decrease of vision and periorbital pain associated with ocular movements in the right eye on April 7, 2005. The visual acuity was 0.05 OD and 1.2 OS, and a right relative afferent pupillary defect was present. The anterior chamber was quiet OU. Ophthalmoscopy revealed swelling of the right optic disc, and fluorescein angiography showed hyperfluorescence of the right optic disc (Fig. 1). Goldmann perimetry showed a peripheral island in the right eye (Fig. 1).

MRI of the brain and orbits demonstrated an unusually enlarged and twisted right optic nerve (Fig. 2). The visual evoked potentials stimulating OD were abnormal. Neither neurologic evaluation nor CSF revealed any abnormalities such as myelin basic protein and oligoclonal bands.

Although an optic nerve glioma was suspected from the MRI findings, the clinical symptoms were thought to be more consistent with optic neuritis. High dose intravenous corticosteroids were given for 3 days, and the right visual acuity improved to 1.2 after 8 days. Repeat MRIs showed no abnormalities and no recurrence has been found after more than one year. Although the possibility of lymphoma or metastatic cancer could not be completely excluded, the clinical course suggested optic neuritis.

The advancements in imaging technology, e.g., fast spin-echo and the short T1 inversion recovery method in MRI, have reduced the types of diseases from which a differential diagnosis of optic neuritis must be made [2, 5]. In some cases, however, the clinical presentations are so similar to that of an optic nerve tumor that a definitive diagnosis cannot be made [2–4]. A case with classic signs of optic neuritis, but was finally diagnosed as a pilocytic astrocytoma, was reported [4].

The clinical course in our case was compatible with optic neuritis in contrast to the MR images suggesting optic nerve glioma. Although an optic nerve neoplasm may have responded to corticosteroid therapy, we believe the most probable diagnosis was optic neuritis because the patient's symptoms and signs as well as the abnormal MRI findings disappeared quickly after the treatment and no recurrence has been observed.

Differentiating optic nerve neoplasms from inflammatory changes is not necessarily easy, and metabolic, infectious, and inflammatory work-ups as well as MRI are necessary. The response to corticosteroid can help in making a definitive diagnosis, and a biopsy of the optic nerve is done only after eliminating other possibilities.

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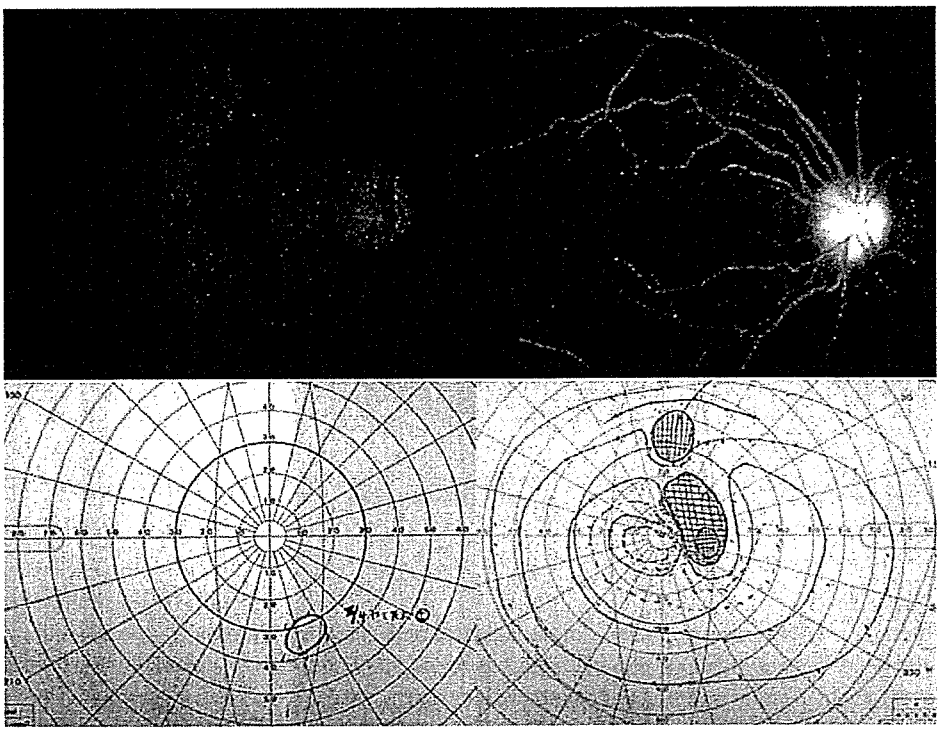

Fig. 1 Fundus photographs and visual fields of the patient's right eye. Top left. Fundus shows swelling of the right optic disc. Top right. Fluorescein fundus angiogram showing fluorescein staining of the right optic disc in the late phase. Bottom left. Goldmann visual field in the acute phase showing only a very small peripheral island in the right visual field. The corrected visual acuity was 0.01 OD. Bottom right: Goldmann visual field 19 days after high-dose intravenous corticosteroid showing a recovery in the right visual field. Visual acuity was 1.5 OD

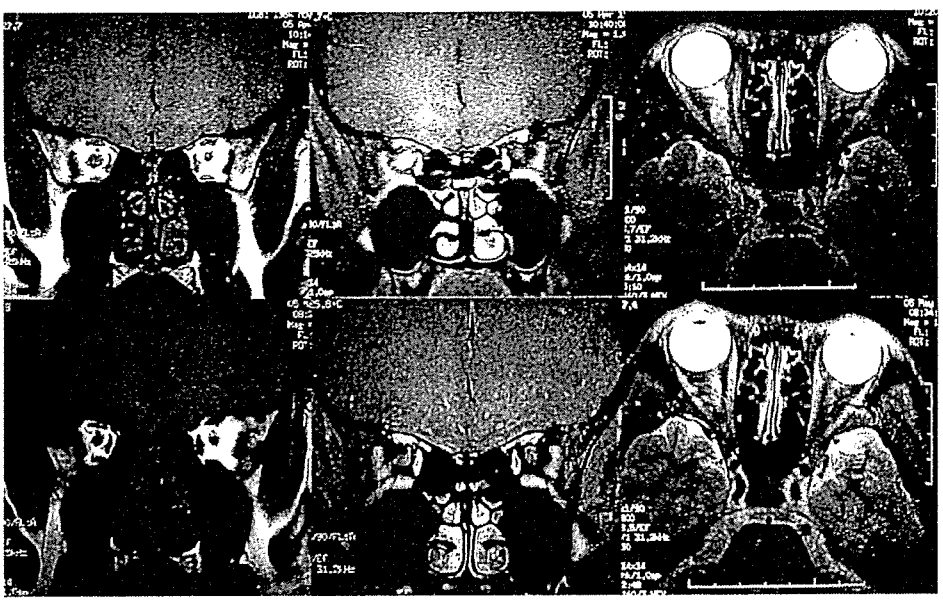

Fig. 2 Magnetic resonance imaging (MRI) of the brain and orbits in the patient. Upper row shows MRIs in the acute phase. Upper left. T1 weighted image (T1WI) of coronal section showing a very enlarged right optic nerve. No pathological lesion was observed in other white matter or the spinal cord. Upper middle: The coronal section of MRI with the short T1 inversion recovery method showed a uniform intense gadolinium enhancement in the optic nerve that suggested that the optic nerve and not the subarachnoidal space was swollen. Upper right: T2 weighted image (T2WI) of axial section showing an unusually homogeneously enlarged and twisting right optic nerve of 7 mm diameter. The signal intensity in the right optic nerve was abnormally increased. Lower row. MRIs obtained 20 days after high-dose intravenous corticosteroid therapy. Each picture corresponds with the upper one. No abnormal findings are present in the right optic nerve compared with unaffected left optic nerve

The current findings indicate that clinicians should still consider optic neuritis in cases with MR images that suggest an optic nerve neoplasm but the symptoms are more in keeping with optic neuritis.

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BRIEF COMMUNICATION

Relaxation of Encircling Buckle Improved Choroidal Blood Flow in a Patient with Visual Field Defect Following Encircling Procedure

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Abstract

Background: We report a patient with a visual field defect after retinal reattachment by the encircling procedure for rhegmatogenous retinal detachment. We confirmed improved ocular blood flow after relaxation of the buckle.

Case: A 24-year-old woman with a visual field defect appearing after an encircling procedure for rhegmatogenous retinal detachment.

Observations: Before and after relaxing the encircling buckle, we measured tissue blood flow in the fundus of each eye of the patient using a Heidelberg retina flow meter. Preoperative measurements showed a reduction of blood flow at the disc rim in the diseased fundus, while retinal blood flow was not reduced ($P = 0.026$, disc rim area versus retinal area, one-factor analysis of variance, ANOVA). Indocyanine green angiography showed extensive peripheral filling delay. Electroretinography showed low a-wave and b-wave amplitudes, but normal oscillatory potential. The base value of the electro-oculogram was severely reduced in the right eye. The blood flow values after surgery indicated a significant improvement of blood flow ($P = 0.01$, one-factor ANOVA). No further progression in the visual field defect was observed, and visual acuity of the right eye improved from 0.8 to more than 1.0.

Conclusions: These results suggest that the choroidal circulation disturbance, which was found after the encircling procedure, had a plausible role in the development of the visual field defect. **Jpn J Ophthalmol** 2006;50:554–556 © Japanese Ophthalmological Society 2006

Key Words: choroidal blood flow, encircling procedure, rhegmatogenous retinal detachment, scanning laser Doppler flowmetry, visual field defect

Introduction

There have been various reports concerning an adverse effect on ocular circulation of the scleral buckling procedure.[1–4] Choroidal and retinal blood flow was significantly decreased in eyes of rabbits buckled with encircling elements.[1] Further clinical evidence, for example, a 40% decrease[2] in the blood flow velocity of the choroid–retina on the buckled side and an average blood flow rate 50% lower[3] through the major temporal retinal arteries in the surgically treated eye compared with that in the fellow eye after scleral buckling for rhegmatogenous retinal detachment (RRD), has been reported. Compression mechanisms were cited as the cause of the reduced choroidal blood flow following scleral buckling procedures. However, although retinal blood flow in the macular area was reported to be disturbed in RRD patients without macular involvement, it recovered to an almost normal level 1 month after successful scleral buckling procedures.[4]

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We present a case of visual field defect (VFD) after retinal reattachment by an encircling procedure (EC) for peripheral RRD.

Case Report

In February 1999, in another hospital, a 24-year-old woman underwent EC for inferotemporal RRD in association with atopic dermatitis in her right eye. Six months later, she noticed an inferior VFD in her right eye, which had not existed before. The patient was referred to our clinic. Best-corrected visual acuities were 0.8 OD, 1.0 OS, and intraocular pressure (IOP) was 14 mmHg OU. Ophthalmoscopy showed no recurrence of RRD but a high indentation of the encircling buckle. Neither quadrant swelling of the optic disc nor retinal vessel branch obstruction in the area corresponding to the VFD was observed. Goldmann perimeter showed a wedge-shaped inferior peripheral VFD expanding to around 10° of the central visual field in the right eye (Fig. 1a), while the left eye showed a normal visual field. Single-flash electroretinogram (ERG) showed low a-wave and b-wave amplitudes, but normal oscillatory potential in the right eye, whereas the response of the left eye was normal (Fig. 1b). In an electro-oculogram (EOG), the base value was severely reduced in the patient's affected eye (Fig. 1c).

Fluorescein angiography (FA) showed no remarkable abnormal findings, but indocyanine green angiography (IA) indicated an extensive peripheral filling delay.

Tissue blood flow was measured at four retinal areas in both eyes with a Heidelberg retina flow meter (HRF, Heidelberg Engineering, Heidelberg, Germany), including the superior and inferior disc rim areas and paramacular areas. Mean blood flow (MBF) in each area was obtained using an automatic full-field perfusion image analyzer[5] (version 3.3, Heidelberg Engineering, Table 1). The ratio of the MBF of the affected eye to that of the fellow eye (a/f ratio) was calculated for each area to minimize the interexamination variation.[6] The a/f ratios at the paramacular area were 1.04 and 1.45, superior and inferior, respectively; whereas the

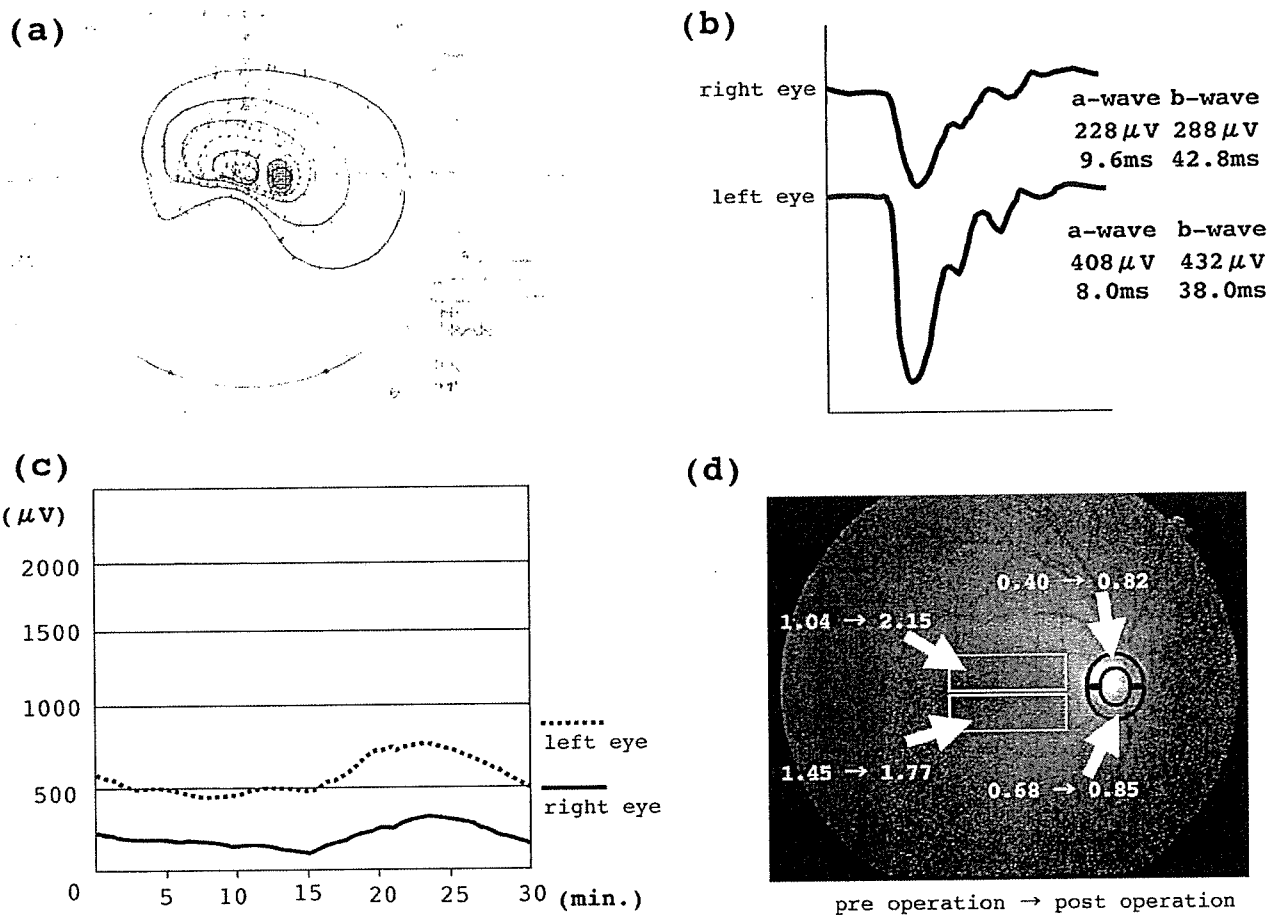

Figure 1. **a** Results of Goldmann perimetry 6 months after encircling procedure. **b** Single flash electroretinogram. Low amplitudes of a-wave in the right eye. **c** Electro-oculogram. Reduced base value in the right eye. Light peak/dark trough ratios were 2.142 in the right eye and 1.700 in the left eye. **d** Affected/fellow eye (a/f) ratio before buckle relaxation surgery and 6 weeks afterward. Preoperative measurements showed reduction of blood flow in disc rim of the diseased fundus ($P = 0.026$ for disc rim area versus retinal area). The mean ratio of each area indicated a significant improvement of blood flow after surgery ($P = 0.01$, one-factor analysis of variance).

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Table 1. Mean blood flow values and a/f ratios by HRF at each measurement area

	Neuroretinal rim area						Paramacular area					
	Superior			Inferior			Superior			Inferior		
	a	f	a/f	a	f	a/f	a	f	a/f	a	f	a/f
Pre-op	138.9	349.9	0.40	229.4	336.1	0.68	255.0	245.9	1.04	235.5	162.3	1.45
Post-op	215.8	262.1	0.82	253.9	297.2	0.85	572.9	266.4	2.15	323.3	245.0	1.77

Values are expressed in arbitrary units.
HRF, Heidelberg retina flow meter; a, affected eye; f, fellow eye, Pre-op, preoperation; Post-op, postoperation; a, affected eye; f, fellow eye; a/f, affected eye/fellow eye ratio.

ratios at the disc rim area were 0.40 and 0.68, superior and inferior, respectively, which is highly suggestive of decreased blood flow at the disc rim ($P = 0.026$, disc rim area versus retinal area, Fig. 1d). Therefore, the association of the VFD with the compromised choroidal circulation was highly suspected. Considering the possibility of retinal redetachment, removal of the encircling buckle was postponed.

In February 2001, with the intention of improving circulation but keeping indentation, the EC buckle was cut off at one part. The silicone band had been buckled about 15 mm from the corneal limbus. The degree of retinal elevation caused by the buckle did not change ophthalmoscopically before and after relaxing the EC. Six weeks later, the a/f ratios were 2.15 and 1.77, superior and inferior, respectively, at the paramacular area, and 0.82 and 0.85, superior and inferior, respectively, at the disc rim area, showing a significant improvement of blood flow ($P = 0.01$, one-factor analysis of variance, Fig. 1d). ERG, EOG, and Goldmann perimetry showed no remarkable change. Visual acuity of the right eye had ranged from 0.5 to 0.9 before relaxation of the EC buckle, but after buckle relaxation, visual acuity improved to 1.0 or higher, and has been steadily maintained at this level since. No further progression of the visual field defect has been observed after 3 years of follow-up.

Discussion

The current case demonstrated reduced tissue blood flow at the disc rim area that improved after relaxation of the EC buckle. However, the tissue blood flow at the paramacular area was normal. Blood flow of the inner retina and nerve fiber layer of the disc are supplied by the central retinal artery or its branches, while blood flow in the prelaminar region adjacent to nerve fiber layer is supplied by branches of juxtapapillary choroidal vessels. Flow values at the disc rim obtained by HRF are considered to be a mixture of retinal and choroidal blood flow contributions because the depth of laser penetration is about 400 μm. Therefore, the laser scanning depth includes the nerve fiber layer and prelaminar regions.

The findings in this case suggest that EC predominantly disturbs choroidal circulation while leaving retinal circulation intact over the long term following EC. The findings of FA and IA support these results. An alternative explanation is that the macular microcirculation is less susceptible to the compression force of the buckle, which could persistently affect the blood flow in main ocular vessels. Investigation of the retinal and choroidal blood flow changes shortly after the EC procedure would be helpful to clarify these hypotheses.

The electrophysiological parameters, such as low a-wave and b-wave amplitudes but normal oscillatory potential in ERG, and the severely reduced base value of EOG in the affected eye, suggest that VFD after EC is caused by dysfunction of the outer retinal layer, mainly due to the disturbance of choroidal circulation, supporting the MBF measurements. The patient noticed her visual field defect 6 months after the EC procedure. We speculate that her visual field disturbance progressed gradually during this period.

Electrophysiological testing together with ocular blood flow measurement might be useful in some cases to understand the mechanism of such complications as VFD.

In this case, visual acuities improved from under 1.0 to 1.0 or higher after relaxing the EC buckle. This change was inferred to result from choroidal and retinal circulation improvement caused by relaxing of the EC buckle.

Because preservation of the scleral indentation was preferred, the EC buckle was relaxed. This simple procedure can be considered for improving choroidal circulation, especially in patients who are suffering from VFD after EC.

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ORIGINAL PAPER

Phototoxic effects of commercial photographic flash lamp on rat eyes

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Abstract
Background To determine whether exposure of the cornea and retina of rats to flashes from a commercial photographic flash lamp is phototoxic.
Methods Sprague–Dawley rats were exposed to 10, 100, or 1,000 flashes of the OPTICAM 16M photographic flash lamp (Fujikoeki, Japan) placed 0.1, 1, or 3 m from the eyes. Corneal damage was assessed by a fluorescein staining score, and the retinal damage by eletroretinography (ERG) and histology before and 24 h after exposure.
Results Exposure of the eyes to 1,000 flashes at 0.1 m increased the fluorescein staining score significantly ($P = 0.009$, the Mann–Whitney test). Scanning electron microscopy (SEM) of the cornea showed a detachment of the epithelial cells from the surface after this exposure. The amplitude of the a-wave was decreased significantly by 23.0% ($P = 0.0026$) of the amplitude before the exposure, and the b-wave by 19.7% ($P = 0.0478$) following 1,000 flashes at 0.1 m but not by the other exposures. TUNEL-positive cells were present in the outer nuclear layer only after the extreme exposure, but no significant decrease in retinal thickness was seen under any condition. The fluorescein staining score and ERGs recovered to control levels within 1 week.
Conclusions Light exposure to a photographic flash lamp does not induce damage to the cornea and retina except when they are exposed to 1,000 flashes at 0.1 m.

Keywords Cornea · Electroretinograms · Photographic flash lamp · Phototoxicity · Retina

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Introduction

Human eyes are continuously exposed to sunlight and artificial lights, and excessive light exposure can induce tissue damage to different ocular structures. These changes have been studied both experimentally and clinically for photoreceptor damage by continuous low level