ELSEVIER

Ciliochoroidal Effusion Induced by Topical Latanoprost in a Patient with Sturge-Weber Syndrome

Hiroshi Sakai, Nariko Sakima, Yoshimi Nakamura,
Yuko Nakamura, Kazuhisa Hayakawa and Shoichi Sawaguchi

Department of Ophthalmology, University of the Ryukyus, Okinawa, Japan

Background: To report drug-induced ciliochoroidal effusion in a patient with Sturge-Weber syndrome.

Case: A 17-year-old man presented with unilateral glaucoma associated with Sturge-Weber syndrome.

Observations: His corrected visual acuity was RE 20/20 and LE 40/60. Intraocular pressure readings by Goldmann applanation tonometry were RE 32 mm Hg and LE 12 mm Hg. Fundus examination showed marked glaucomatous disc cupping in his right eye and normal finding in his left. The patient had a port-wine stain on his right upper eyelid ipsilateral to the glaucomatous eye. Antiglaucomatous medications were begun, including topical latanoprost, with a diagnosis of juvenile onset glaucoma associated with Sturge-Weber syndrome. Ultrasound biomicroscopy showed a 360° circumference ciliochoroidal effusion. Forty days after starting medication, latanoprost treatment was discontinued. Ten days later, ultrasound biomicroscopy showed a total disappearance of the ciliochoroidal effusion.

Conclusion: Interaction of the enhanced uveoscleral outflow with latanoprost in conjunction with elevated episcleral venous pressure may have caused the congestion of the aqueous humor in the supraciliary-choroidal space, resulting in the ciliochoroidal effusion. **Jpn J Ophthalmol 2002;46:553–555** © 2002 Japanese Ophthalmological Society

Key Words: Ciliochoroidal effusion, latanoprost, Sturge-Weber syndrome, ultrasound biomicroscopy, uveoscleral outflow.

Introduction

Patients with Sturge-Weber syndrome frequently develop ciliochoroidal effusion during or after trabeculectomy.[1,2] Surgical interventions such as filtration surgery, cataract extraction, or scleral buckling may cause ciliochoroidal effusion, and hypotony is usually associated. Ocular injury, uveitis, vascular diseases, malignancy, and scleral abnormality of nanophthalmos or uveal effusion syndrome[3] also may cause ciliochoroidal effusion.

Latanoprost, a prostaglandin analogue, has been reported to have the adverse side effect of choroidal detachment immediately or a long time after surgical treatment.[4–6] We report a first case of prominent ciliochoroidal effusion induced by topical latanoprost in a Sturge-Weber syndrome-associated glaucoma patient without surgery.

Case Report

A 17-year-old man was seen by a local ophthalmologist for myopia. He had no history of medical therapy in either eye. On initial examination his corrected visual acuity was RE 20/20 (−0.5 D) and LE 40/60 (−8.0 D). Intraocular pressure (IOP) by Goldmann applanation tonometry was RE 32 mm Hg and LE 12 mm Hg. Fundus examination showed marked glaucomatous disc cupping in his right eye and normal findings in his left. The patient had a port-wine stain on his right upper eyelid ipsilateral to the glaucomatous eye. Slit-lamp microscopy examination showed mildly engorged episcleral vessels in his right eye; the left eye was unremarkable. Application of

Received: September 25, 2001
Correspondence and reprint requests to: Hiroshi SAKAI, MD, Department of Ophthalmology, University of the Ryukyus, Faculty of Medicine, 207 Uehara, Nishihara-cho, Okinawa 903-0215, Japan

Published by Elsevier Science Inc.

0021-5155/02/$–see front matter
PII S0021-5155(02)00542-7

Jpn J Ophthalmol
Vol 46: 553–555, 2002

antiglaucoma drugs (0.5% timolol twice daily, 1% dorzolamide three times daily, and 0.005% latanoprost once daily) was started to his right eye to lower the IOP. The patient was then referred to our glaucoma clinic.

On day 11 after starting the antiglaucoma treatment, tonometry measured 24 mm Hg in the right eye and 17 mm Hg in the left. Gonioscopy revealed wide-open angle with minimum high insertion of iris roots in either eye. The cup-to-disc ratios were RE 0.9 and LE 0.4. Examination by Humphrey visual field analyzer showed superior arcuate defect in the right eye; the left was normal. Computed tomography ruled out a possible cerebral hemangioma. We concluded that the patient had juvenile onset glaucoma associated with Sturge-Weber syndrome. Antiglaucoma drugs (0.5% timolol, 1% dorzolamide, and 0.005% latanoprost) were continued.

On day 18, ultrasound biomicroscopy (UBM), performed as a routine procedure for glaucoma patients in our clinic, demonstrated marked ciliochoroidal effusion with a circumferential hypoechoic area, multiple ciliary cysts, and thick episclera in his right eye (Figure 1). The depth of the central anterior chamber of the right eye was measured by UBM to be 3.0 mm. The findings by UBM in his left eye were unremarkable. Fundus examination was repeated and revealed circumferential choroidal detachment in the right eye. Choroidal detachment lasted for about 3 weeks while the use of topical timolol, dorzolamide, and latanoprost was continued. Visual acuity of the RE was not changed.

On day 40 after starting the antiglaucoma treatment, latanoprost was discontinued. At this time point, choroidal detachment was observed by fundus examination. On day 47, the IOP was measured and found unchanged (RE 24 mm Hg). On day 50, ten days after stopping latanoprost, UBM examination was repeated and showed total disappearance of the ciliochoroidal effusion (Figure 2). The depth of the central anterior chamber of the right eye had increased to 3.5 mm. On day 53, the IOP of the RE was 23 mm Hg.

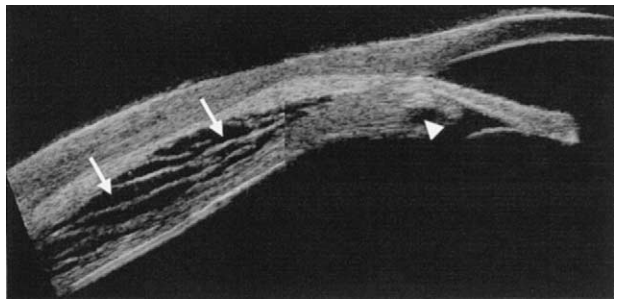

Figure 1. Ultrasound biomicroscopy images of the right eye of a 17-year-old man with Sturge-Weber syndrome. Marked ciliochoroidal effusion is shown as supraciliary-choroidal hypo-echographic space (arrows) together with ciliary body cyst (arrowhead) and thick episclera.

Discussion

Latanoprost reduces the IOP by enhancing the uveoscleral outflow. The ciliary muscle and its extracellular matrix play an important role in this pathway. Mishima et al had shown a decrease in the ciliary body thickness 4 hours after a single application of latanoprost in normal subjects.[7] Recent studies hypothesized that latanoprost relaxes the ciliary muscle and subsequently provokes the remodeling of the extracellular matrix.[8]

A few cases of choroidal detachment after topical latanoprost have been reported. It has been suggested that increased uveoscleral outflow may have an effect on the occurrence of hypotony and choroidal detachment.[4,5] These patients had undergone cataract extraction[5,6] or combined cataract extraction and trabeculectomy.[4] In one case,[6] hypotony had not been detected, but the patient had marked anterior uveitis that was a cause of ciliochoroidal effusion. It is believed that ciliochoroidal effusion, an abnormal accumulation of fluid in the suprachoroidal space, results from transudation from the choroidal capillary wall secondary to hypotony or abnormal vascular permeability. Otherwise, there may be a direct inflow of aqueous humor from the anterior

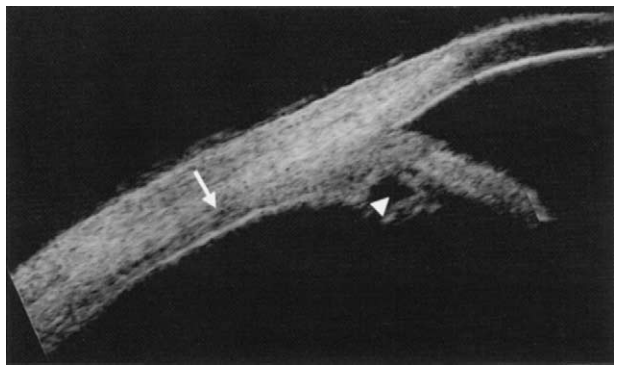

Figure 2. Ultrasound biomicroscopy images after discontinuation of latanoprost. Marked ciliochoroidal effusion has disappeared (arrow). Ciliary body cyst is unchanged (arrowhead).

chamber with or without a cyclodialysis. Ocular surgery is the major cause of ciliochoroidal effusion. Our patient had no history of surgery, and no evidence of hypotony, uveitis, or cyclodialysis was observed. On the other hand, the prominent episcleral vessels accompanying Sturge-Weber syndrome, indicative of increased episcleral venous pressure,[1] were observed. Increased episcleral venous pressure is thought to be a cause of ciliochoroidal effusion in Sturge-Weber syndrome patients.[1] In a Sturge-Weber syndrome-associated glaucoma patient,[9] a 360° ciliochoroidal effusion has been reported with the use of UBM (high-resolution echography). In our patient, the possibility of ciliochoroidal effusion before the use of topical latanoprost should be considered, because a small ciliochoroidal effusion cannot be observed by fundus examination. However, the mode of action of latanoprost may be an additional contributor to the formation of ciliochoroidal effusion, because the discontinuation of latanoprost resulted in the complete resolution of the effusion. We hypothesized that interaction of the effect of latanoprost and the mechanism of Sturge-Weber syndrome-associated glaucoma with increased episcleral venous pressure had caused prominent ciliochoroidal effusion in our case. Also, latanoprost has been found to have a limited intraocular pressure lowering effect on Sturge-Weber syndrome-associated glaucoma.[10] Further studies using UBM are needed to disclose the relation between latanoprost and ciliochoroidal effusion in Sturge-Weber syndrome.

References

1. Bellows AR, Chylack LT, Epstein DL, Hutchinson BT. Choroidal effusion during glaucoma surgery in patients with prominent episcleral vessels. Arch Ophthalmol 1979;97:493–497.
2. Iwach AG, Hoskins HD Jr, Hetherington J Jr, Shaffer RN. Analysis of surgical and medical management of glaucoma in Sturge-Weber syndrome. Ophthalmology 1990;97:904–909.
3. Brubaker RF, Pederson JE. Ciliochoroidal detachment. Surv Ophthalmol 1983;27:281–289.
4. Rowe JA, Hattenhauer MG, Herman DC. Adverse side effects associated with latanoprost. Am J Ophthalmol 1997; 124:683–685.
5. Wu G, Morrell A. Severe hypotony following cataract extraction in a patient on latanoprost. Eye 2000;14:915–916.
6. Gupta R, Vernon SA. Choroidal detachment following extracapsular cataract extraction in a patient treated with latanoprost. Br J Ophthalmol 2001;85:1268.
7. Mishima HK, Shoge K, Takamatsu M, et al. Ultrasound biomicroscopic study of ciliary body thickness after topical application of pharmacologic agents. Am J Ophthalmol 1996;121:319–321.
8. Sagara T, Gaton DD, Lindsey JD, Gabelt BT, Kaufman PL, Weinreb RN. Topical prostaglandin F2 treatment reduces collagen types I, III, and IV in the monkey uveoscleral outflow pathway. Arch Ophthalmol 1999;117:794–801.
9. Kranemann CF, Pavlin CJ, Trope GE. Ultrasound biomicroscopy in Sturge-Weber-associated glaucoma. Am J Ophthalmol 1998;125:119–121.
10. Altuna JC, Greenfield DS, Wand M, et al. Latanoprost in glaucoma associated with Sturge-Weber syndrome: Benefits and side-effects. J Glaucoma 1999;8:199–203.