Does Decreased Static Ocular Counter Rolling Account for Bielschowsky Head Tilt Test in Unilateral Superior Oblique Palsy?

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PURPOSE. To understand the relationship of static ocular counter rolling (s-OCR) and clinical manifestations in acquired unilateral superior oblique palsy subjects during the Bielschowsky head tilt test.

METHODS. Nineteen subjects that were diagnosed with acquired unilateral superior oblique palsy were included. Fundus photographs were obtained at different head tilt angles to evaluate static ocular counter rolling using a fundus camera with a cervical range of motion device. Using a graphics editing program, we calculated s-OCR from fundus photography.

RESULTS. The incycloductional s-OCR (OCR-I) in the paretic eye was significantly smaller than the OCR-I in the fellow eye ($P = 0.02$, <0.001, 0.002 for $10°$, $20°$, and $30°$, respectively, paired t-tests). In contrast, the excycloductional s-OCR (OCR-E) showed no significant difference between the paretic eye and the fellow eye for all angles. There was a significantly positive correlation between the amplitude of OCR-I in the paretic eye and the degree of hypertropia on ipsilesional head tilt ($\rho = 0.612$, 0.679, 0.474, $P = 0.02$, 0.002, 0.07 for $10°$, $20°$, and $30°$ respectively, Spearman's correlation). The amplitude of OCR-I in the paretic eye also showed a positive correlation with head tilt test difference, which is the degree of hyperdeviation difference between ipsilesional and contralesional head tilts ($\rho = 0.445$, 0.694, 0.579, $P = 0.09$, 0.002, 0.024 for $10°$, $20°$, and $30°$ respectively, Spearman's correlation).

CONCLUSIONS. In unilateral SOP, OCR-I in the paretic eye was smaller than that in the fellow eye, and this was positively associated with the degree of hypertropia during ipsilesional head tilting, as well as the head tilt test difference.

Keywords: superior oblique palsy, static ocular counter rolling, Bielschowsky head tilt test, cervical range of motion device

Ocular counter rolling (OCR) is a partially compensatory torsional eye movement that occurs when the head is tilted toward the shoulder.[1] Static OCR (s-OCR) is mediated by an otolith-ocular reflex in response to a gravitational direction change, while dynamic OCR is mediated by the semicircular canal-ocular reflex in response to torsional angular acceleration during active head movement.[2] The s-OCR is thought to be the biomechanic basis of the Bielschowsky head-tilt test (BHTT).[3–6] Clinically, superior oblique palsy (SOP) subjects exhibit positive BHTT, which means the paretic eye shows greater hypertropia during head tilt to the ipsilesional side compared to the contralesional shoulder. According to traditional s-OCR theory, when the head is tilted to the left shoulder, the intorters (superior oblique [SO] and superior rectus [SR]) of the left eye and the extorters (inferior oblique and inferior rectus) of the right eye are activated while extorters of the left eye and intorters of the right eye are inhibited. In left SOP, downward torque of the superior oblique is absent, so unopposed upward movement by the activated superior rectus is thought to contribute to hypertropia of the left eye during ipsilesional head tilt, which is observed in BHTT.[3] BHTT is still widely used as a means of diagnosing SOP, despite limitations that have become known over time.[7–9] To understand s-OCR and its role in BHTT, many investigators have used various methods.[2,10–12]

Recently, Lim et al.[13] published a novel method to measure s-OCR using fundus photography. In this study, we used the same fundus photographic method in clinically diagnosed acquired unilateral SOP subjects to elucidate the relationship between s-OCR and BHTT. We additionally used a cervical range of motion (CROM) device (Performance Attainment Associates, Roseville, MN, USA) for accurate recording of head tilt angle.

METHODS

Subjects

In this cross-sectional observational study, we enrolled 22 subjects who were diagnosed with acquired unilateral SOP from a single tertiary eye care center (Samsung Medical Center). Five subjects were excluded from data analysis due to poor cooperation. Thus, 17 subjects (11 men and 6 women) between the ages of 19 and 80 years (mean 47.12 ± 14.42) were included in our results. The study was performed between March 1, 2015, and August 31, 2015. Acquired unilateral SOP was diagnosed as follows: unilateral incomitant hypertropia that increased with adduction of the hypertropic eye; positive BHTT; no reversal of the hypertrophic eye on contralesional head tilt; excyclotropia of the paretic eye on

fundus photography in the primary position; and no evidence of congenital SOP, such as facial asymmetry, long standing symptoms of head tilt, or diplopia. Subjects were excluded if there was previous history of strabismus surgery, difficulty with cooperation during fundus photography examination for any reason, or suspicion of a skew deviation in part of the ocular tilt reaction. All enrolled subjects underwent a full ophthalmic examination including prism and alternative cover tests in all diagnostic directions, BHTT with 30° head tilting, extraocular motility test with photographs in nine cardinal positions of gaze, Double Maddox rod test, Lancaster or Hess screen test, and fundus photography with a CROM device during stepwise head tilting. Also, a complete history was taken including age of onset, presence of head tilt or facial asymmetry during childhood, presence of risk factors for ischemia, and history of head trauma. The study protocol complied with the tenets of the Declaration of Helsinki and was approved by the Institutional Review Board of Samsung Medical Center Hospital. Informed consent was obtained from all participants after study details were explained.

Measurement Procedure

After pupil dilation with 1% tropicamide and 2.5% phenylephrine HCl, all subjects were equipped with the CROM device to measure head tilt angle during the procedure. The CROM device is a head-mounted unit with a gravity metered scale that has been shown to measure head tilts with a reproducibility and accuracy of 1°.[14] High resolution (12.3 megapixels) 45° fundus photographs were taken with a fundus camera (Topcon Corp., Tokyo, Japan) in different head-tilt positions. The examiner tilted the subject's head about the naso-occipital axis in 10° steps over a range of 30° toward the right shoulder and then back to the upright position. This was followed by the same stepwise head tilt in the opposite direction. The subjects were asked to fixate on the camera's internal fixation marker. Fixation cooperation was monitored during examination using the camera, and five poorly cooperative subjects were excluded from data analysis. At least 10 seconds after the subject's head reached the correct head tilt angle, fundus photographs were taken to make sure there was no dynamic OCR effect. While obtaining fundus photographs, the examiner held each subject's head and watched the CROM angle to make sure subjects were maintaining the correct angle. Also, subject chin and forehead were firmly attached to the device during the entire examination to eliminate the possibility of chin up or chin down effects.

Image-Processing Procedure

To calculate s-OCR, all photographs were processed using a graphics editing program (Photoshop 6.0; Adobe Systems, Inc., San Jose, CA, USA). Image processing was performed in same manner as Lim's previous report.[13] Baseline primary head position was used as a baseline reference, and the images of each head tilt position were copied onto the reference image as semitransparent layers. We rotated the pasted semitransparent layer such that it overlapped the baseline image using the free rotate tool, so we could record the angle of ocular rotation expressed in the menu bar of the graphics editing program (Adobe Systems, Inc.; Fig. 1).

Measurement of Degree of Compensatory Torsional Movement

The OCR was calculated as the difference between head tilt angle and ocular torsion angle, which is measured by calculating the recorded angle of fundus rotation with the
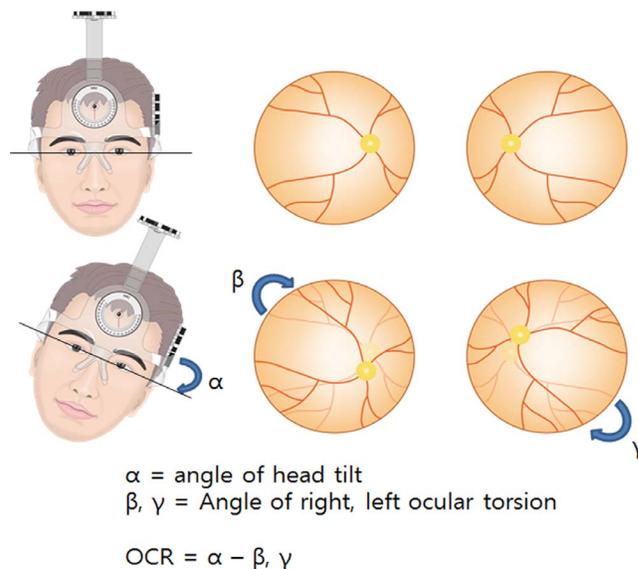

α = angle of head tilt
β, γ = Angle of right, left ocular torsion

OCR = $\alpha - \beta$, γ

Figure 1. Schematic of both ocular torsions according to head tilt. The degree of OCR is defined as the difference between the angle of head tilt (α) and the angle of ocular torsion (β = angle of right ocular torsion, γ = angle of left ocular torsion). This figure is modified from Lim HW, Kim JH, Park SH, Oh SY. Clinical measurement of compensatory torsional eye movement during head tilt. *Acta Ophthalmol.* 2017;95:e101–e106. © 2016 Acta Ophthalmologica Scandinavica Foundation. Published by John Wiley & Sons Ltd.

graphics editing program (Adobe Systems, Inc.; Fig. 1). To determine interobserver reliability, two independent observers (DDC, SML) measured the images using the above methods.

Definition of Terminology

The degree of OCR was defined as the difference between head tilt angle and fundus torsion angle. The word OCR-I was used to describe incycloductional s-OCR after ipsilateral head tilt for each eye. In the same manner, OCR-E was used to describe excycloductional s-OCR after contralateral head tilt for each eye. Due to our binocular fundus photography methodology, OCR-I in the paretic eye and OCR-E in the nonparetic eye were recorded simultaneously, as were OCR-E in the paretic eye and OCR-I in the nonparetic eye. The term head tilt test (HTT) difference is defined as the degree of hyperdeviation difference between an ipsilesional head tilt and a contralesional head tilt examination (ipsilesional hypertropia–contralesional hypertropia).

Statistical Analysis

Statistical analyses were performed using commercial software (SPSS for Windows version 21.0; SPSS, Inc., Chicago, IL, USA). A paired *t*-test was used to compare OCR of the paretic eye and OCR of the nonparetic eye for each angle. Additionally, the generalized estimating equation (GEE) was used to compare OCR values of paretic and nonparetic eyes for all angles together. Spearman's correlation was used to access the relationships between the degree of s-OCR, hyperdeviation of the paretic eye on ipsilateral head tilt, HTT difference, onset, objective torsion, subjective torsion, and grade of inferior oblique overaction (IOOA). Interobserver variability was determined using Cronbach's alpha. *P* values less than 0.05 were considered statistically significant.

TABLE 1. Baseline Characteristics of the Study Population

	Mean ± SD (Range)
Age, y	47.95 ± 13.87 (19–80)
Male:female, n	11:6
Paretic eye, right:left	9:8
Duration, mo	24.90 ± 23.31 (1–84)
Vertical deviation of paretic eye, prism diopter	
In primary position	6.71 ± 4.60
In ipsilateral HT	12.11 ± 5.77
In contralateral HT	3.79 ± 4.53
Difference between vertical deviation in ipsilateral and contralateral HT	8.32 ± 3.50
Objective torsion, deg	
In paretic eye	12.67 ± 3.70
In nonparetic eye	9.59 ± 1.56
Difference of objective torsion between paretic and nonparetic eye	3.08 ± 3.90
Subjective torsion by DMRT*, deg	6.54 ± 6.01
Inferior oblique overaction, grade*	0.32 ± 0.54
Cause of SOP, n	
Head trauma	6
Herpes infection	1
Trochlear nerve schwannoma	1
Diabetes mellitus	4
Unknown	5

DMRT, double Maddox rod test.
* Inferior oblique overaction was graded from 1+ to 4+.

RESULTS

Subject Baseline Characteristics

We recruited 17 subjects (11 male and 6 female) with a mean age of 47.95 ± 13.87 years. Mean symptom duration was 24.90 ± 23.31 months. All subjects showed compensatory OCR during head tilting. More detailed information is shown in Table 1.

Comparison of s-OCR in the Paretic and Nonparetic Eyes

As we hypothesized, OCR-I in the paretic eye was significantly smaller than OCR-I in the nonparetic eye at 10°, 20°, and 30° head tilts ($P = 0.02$, <0.001, and 0.002 for 10°, 20°, and 30°, respectively, paired t-test; Table 2). In contrast, OCR-E showed no significant difference between the paretic eye and the nonparetic eye at all angles ($P = 0.47$, 0.28, and 0.71 for 10, 20, and 30° respectively, paired t-test, Table 2). We also calculated the difference between the paretic eye and the fellow eye for all angles together using the GEE method. For OCR-I, the P value was 0.007, and the estimate was 1.391 (95% confidence interval [CI]: 0.374 to 2.408), representing a significant difference in OCR-I between paretic and fellow eyes, with a mean 1.391 larger size in the fellow eyes. On the contrary, the OCR-E difference between paretic and fellow eyes was not significant (P value = 0.539, 95% CI: −1.451 to 0.760).

Relationship Between OCR-I in the Paretic Eye and Clinical Hypertropia During BHTT

There was statistically significant positive correlation between the amplitude of OCR-I in the paretic eye and the degree of

TABLE 2. Comparison of OCR-I and OCR-E in Paretic and Fellow Eyes

	Paretic Eye, Mean ± SD	Fellow Eye, Mean ± SD	P Value*
OCR-I 10	1.28 ± 2.24	2.74 ± 1.24	0.02
OCR-I 20	3.93 ± 2.88	7.11 ± 2.39	<0.001
OCR-I 30	5.57 ± 3.28	8.31 ± 2.45	0.002
OCR-E 10	1.24 ± 2.09	1.02 ± 2.32	0.48
OCR-E 20	3.97 ± 2.58	4.35 ± 2.99	0.28
OCR-E 30	6.00 ± 3.42	6.20 ± 4.01	0.71

10, 20, 30, head tilt angles.
* Difference between paretic and fellow eyes for each angle, as assessed by paired t-test.

hypertropia on ipsilateral head tilt at 10° and 20° head tilts (Spearman's correlation, $\rho = 0.612$, 0.679, 0.474; $P = 0.02$, 0.002, 0.07 for 10°, 20°, and 30°, respectively; Fig. 2). A statistically significant positive correlation was also found between the OCR-I amplitude in the paretic eye and the HTT difference at 20° and 30° head tilts (Spearman's correlation, $\rho = 0.445$, 0.694, 0.579; $P = 0.09$, 0.002, 0.024 for 10, 20, and 30°, respectively; Fig. 2).

Relationships Between s-OCR and Symptom Onset, Objective Torsion, Subjective Torsion, and IOOA Function

We also evaluated the correlations between the degree of s-OCR and symptom onset, objective torsion, objective torsional difference between eyes, and IOOA function in the paretic eye, but there were no significant correlations.

Interobserver variability was determined using the results from two independent examiners (DDC, SML; Cronbach's $\alpha = 0.990$ [95% CI: 0.985~0.993] $P = < 0.001$).

DISCUSSION

The goal of this study was to understand the relationship of s-OCR and BHTT by measuring s-OCR in clinically diagnosed unilateral superior oblique palsy subjects. We used fundus photography to record s-OCR in unilateral SOP subjects. Unlike previous studies using photographic or video-based measurements to record the anterior part of the eye or invasive scleral coil methods, we noninvasively recorded the fundus itself to precisely measure torsional movement.[15-18] Moreover, we used a CROM device during the fundus photography procedure to maintain the head tilt angle and avoid chin-up or down effects. We calculated s-OCR from fundus photography using the graphics editing program (Adobe Systems, Inc.) as we rotated the fundus photograph of head tilt position until it matched the fundus photograph of the primary position. The s-OCR was calculated as "head tilt angle–fundus rotation angle." This methodology not only has the advantage of accurate calculation of s-OCR, but also allowed us to separate false torsion from vertical eye movements.[15] We confirmed that OCR-I in the paretic eye during ipsilesional head tilting was significantly smaller than OCR-I in the fellow eye during contralesional head tilting. Decreased OCR-I in the paretic eye was reasonable considering that the superior oblique, one of the intorters, was not functioning in the paretic eye. Furthermore, the degree of OCR-I in the paretic eye during ipsilesional head tilting was significantly correlated with the degree of hyperdeviation during ipsilesional head tilting, as
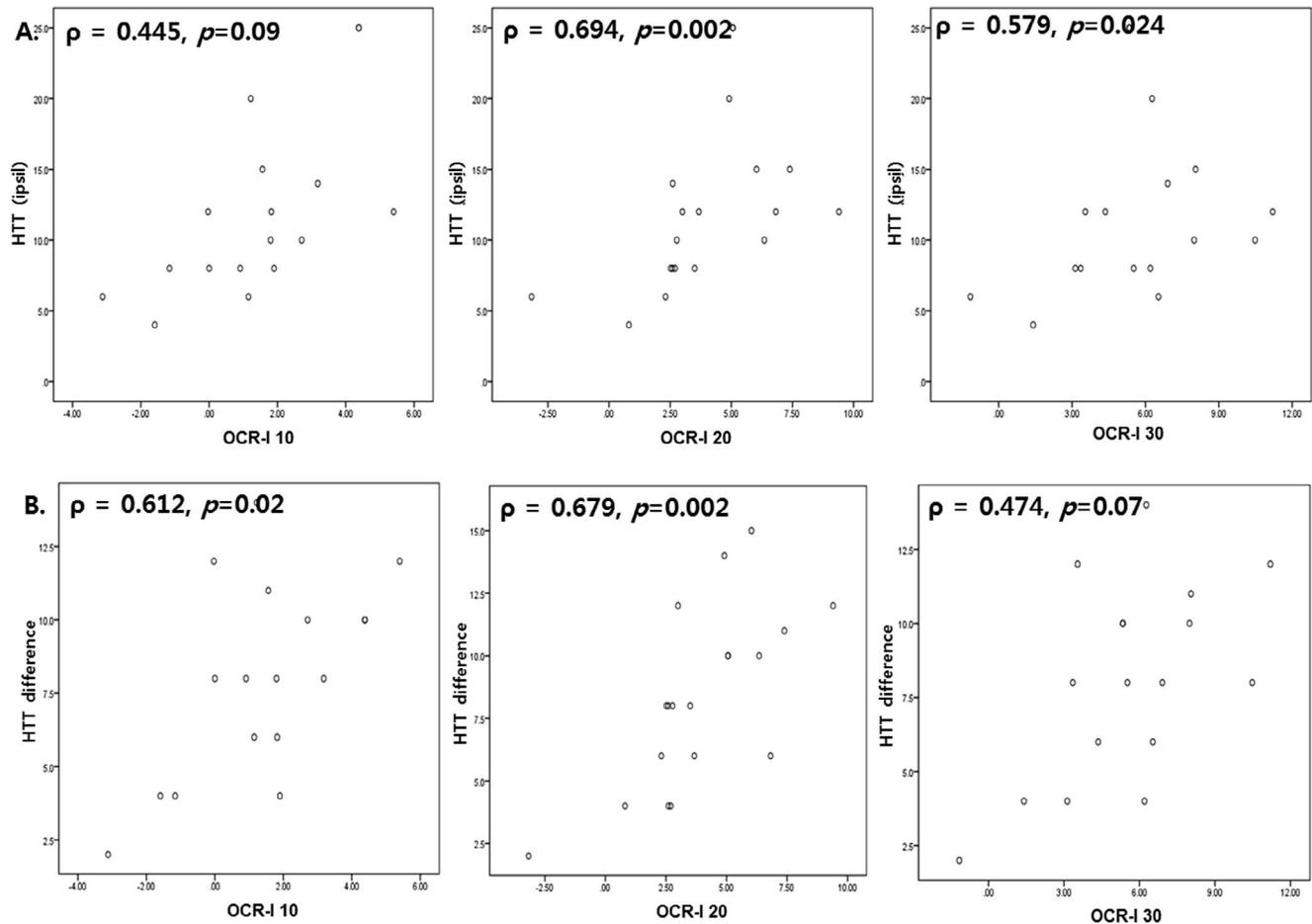
FIGURE 2. (**A**) Scatter plots showing the correlations between the degree of incyclotorsional static ocular counter rolling (OCR-I) in paretic eyes during 10°, 20°, and 30° head tilting and the amount of hyperdeviation during ipsilateral head tilt test. (**B**) Scatter plots showing the correlations between the degree of OCR-I in paretic eyes during 10°, 20°, 30° head tilting and the amount of HTT difference between ipsilesional head tilt and contralesional head tilt. HTT ipsil, hyperdeviation in the paretic eye during ipsilesional head tilt; 10°, 20°, 30°, angle; ρ, statistical value by Spearman's correlation test.

well as the difference between the hyperdeviation during BHTT.

Previously, Hamasaki et al.[19] used a video camera to record the iris and reported that the amplitude of OCR-I in a paretic eye during ipsilesional head tilting was smaller than s-OCR in control subjects during head tilting; however, they did not present their *P* value. They mainly compared s-OCR in ipsilesional and contralesional head tilting in each eye, but their analysis was based on the assumption that s-OCR in ipsilateral and contralateral eyes was equal in normal subjects. Because ocular torsional disconjugacy was observed in other studies, we compared OCR-I between paretic and fellow eyes.[13,20,21] Hamasaki et al.[19] also reported that there was no significant correlation between OCR-I in the paretic eye and the degree of hyperdeviation on ipsilesional head tilt. But, they found a significant negative relationship between OCR-I in the paretic eye and the difference between hyperdeviation with ipsilateral head tilt and in the head-upright position. In contrast, our results showed that OCR-I in the paretic eye and the degree of hyperdeviation on ipsilesional head tilt was positively correlated. Positive correlation was also found between the OCR-I in the paretic eye and the HTT difference. There are possible reasons for the difference between their and our results. First, they measured s-OCR using the iris pattern as a marker, which reflects the anterior part of the eye and can

easily cause measurement error. In our method, we were able to record the fundus itself using a CROM device in order to control its angle precisely. Secondly, the study of Hamasaki et al.[19] was limited to congenital SOP subjects, while our experiment was conducted exclusively with acquired SOP subjects.

To understand the positive correlation between OCR-I in paretic eyes and BHTT, we hypothesized that SO muscle function was negligible in paretic eyes. Then, OCR-I in paretic eyes would be mostly comprised of SR muscle function, another intorter of the eye. We also presumed that SR muscle function would be overactivated to compensate for the nonfunctioning SO muscle. Using an overstimulated SR muscle hypothesis, we could explain the positive correlation between OCR-I in paretic eyes and BHTT. As the SR effort to compensate for SO muscle—which is measured as the OCR-I of a paretic eye—becomes stronger, the degree of hyperdeviation of the paretic eye on ipsilesional head tilt increases. Although this is our speculation, we think it is a possible mechanism that is consistent with the observations made herein. Biomechanic simulation predicts that the vertical action of the SO muscle itself would be too weak to be responsible for the large hypertropia typical of SO palsy.[22] In comparison, the SR is a powerful elevator that is able to cause a markedly positive BHTT.[23,24]

Previously, Kono et al.[24] used magnetic resonance imaging (MRI) in clinically diagnosed SOP subjects and concluded that SO size did not account for the variation in BHTT, supporting the proposition that the BHTT is nonspecific for SO function. Our results are consistent with theirs, and we further suggest that BHTT does not account for SO function, but for the overstimulated SR that is measured as OCR-I in paretic eyes. In contrast, there is a claim that s-OCR in SOP subjects is associated with SO muscle function itself. Hamasaki et al.[25] reported that, from 12 unilateral SOP subjects, the mean ratio of s-OCR in nine subjects who showed SO muscle atrophy on MRI was significantly lower than s-OCR in three subjects with nonatrophy on ipsilesional head tilting. They concluded that s-OCR reflected the anatomic disorder of the superior oblique muscle in SOP. However, they did not consider compensatory mechanisms and ignored the function of another intorter, the SR muscle. Recently, Suh et al.[26] used high-resolution, surface-coil MRI in unilateral SOP subjects and observed significant changes in extended posterior partial volumes of extraocular muscles. They found hypertrophy in the contralesional SR, ipsilesional inferior rectus, and ipsilesional lateral rectus. Even though this finding is interesting, the contractility of extraocular muscles was not evaluated. Moreover, those findings cannot explain clinical manifestations of SOP like vertical hypertropia or BHTT. Their result could be due to the limitations of image analysis of extraocular muscles restricting direct evaluation of the amount of muscle function from image analysis. Also, MRI was not performed in the head tilting position, so the otolith-ocular reflex was not present.

The clinical diagnosis of unilateral SOP is made based on a three-step test including the BHTT.[5] But, as Demer et al.[27] pointed out earlier, there are "masquerading" SOPs that are clinically diagnosed as SOP but have normal cross-sectional area and normal contractility. On the contrary, we cannot rule out SOP even if there is a relatively normal SO configuration on MRI. Therefore, in clinical settings, the BHTT is still a significant physical examination for diagnosing SOP. Our results provide evidence that BHTT is positively associated with incycloductional s-OCR of the paretic eye, presumably the amount of overstimulated SR in acquired unilateral SOP subjects.

This study has a few limitations. Although prospectively collected, the sample size was limited to 17 subjects. Although this is relatively small number, a pilot study was conducted in advance and confirmed that 17 was a large enough sample size to obtain significant results. We included only acquired SOP subjects for uniformity within the sample, so further study including congenital SOP subjects and comparison between these two groups might be needed.

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