• Clinical Research •

Effect of unilateral inferior oblique weakening on fundus torsion in bilateral eyes of children with congenital superior oblique palsy

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Abstract
● **AIM:** To study the change of torsion in both eyes after unilateral inferior oblique (IO) weakening on children with congenital superior oblique palsy (SOP).
● **METHODS:** This retrospective study enrolled all patients diagnosed with unilateral congenital superior oblique palsy (UCSOP) accompanied by inferior oblique overaction (IOOA). A total of 120 eyes of 60 patients were divided into group 1 (more extorted paretic eye) and group 2 (more extorted nonparetic eye). The degree of fundus torsion was evaluated before and 1mo after the IO weakening procedure. The torsion of the fundus was recorded by measuring the disk-foveal angle (DFA) using fundus photography.
● **RESULTS:** Group 1 included 26 cases and group 2 included 34 cases, thus the rate of extorsion was insignificantly higher in the nonparetic eye (P=0.10). The preoperative DFA in the paretic and nonparetic eyes was 13.21±5.95, 7.97±4.25 in group 1, and 4.65±3.79, 13.16±5.35 in group 2 (both P<0.001). The postoperative DFA in the paretic and nonparetic eyes was 8.57±4.87, 7.32±4.27 in group 1 (P=0.24), and 3.85±6.00 and 9.94±5.45 in group 2 (P<0.001). The amount of postoperative reduction of the DFA in the paretic and nonparetic eyes was 4.64±3.90, 0.65±0.76 in group 1 (P=0.002), and 0.80±0.81, 3.21±5.50 in group 2 (P=0.01). The difference in the amount of reduction of DFA in the more extorted eye in group 1 (paretic eye) *vs* group 2 (nonparetic eye) was insignificant (P=0.30).

● **CONCLUSION:** Excyclotorsion in the nonparetic eye has a similar probability in the paretic eye in UCSOP children, and weakening of the ipsilateral IO has a more obvious effect on the decrement of extorsion in the more extorted eye regardless of which eye is paretic.
● **KEYWORDS:** superior oblique palsy; ocular torsion; disc-foveal angle; excyclotorsion

INTRODUCTION

Congenital superior oblique palsy (SOP) is the most common form of cyclovertical paralytic strabismus in children, usually characterized by an abnormal head posture, facial asymmetry, and vertical misalignment, which can also be accompanied by underaction of the superior oblique, overaction of the ipsilateral inferior oblique (IO) muscle, a positive head-tilt test result and fundus excyclotorsion[1-5]. A large proportion of studies have shown that there is significant ocular excyclotorsion in paretic eyes, but some others have found that excyclotorsion could also be present in the nonparetic contralateral eye[6-8]. IO weakening is the most common procedure used to reduce the vertical deviation and improve the abnormal fundus extorsion in the paretic eye, especially in cases with inferior oblique overaction (IOOA)[9-12], but little data are available regarding the impact of the IO weakening procedure on the change in torsion in the contralateral nonparetic eye.

Our study aimed to investigate the change of torsion in both eyes after unilateral IO weakening on children with unilateral congenital superior oblique palsy (USCOP).

SUBJECTS AND METHODS

Ethical Approval The study was approved by the Beijing Children's Hospital Ethics Committee and was conducted in accordance with the Declaration of Helsinki. Informed consent was obtained from the guardians of all the subjects.

We retrospectively reviewed the medical records of 60 consecutive patients who were diagnosed with UCSOP accompanied with IOOA and underwent IO weakening from February 2016 to July 2018 in the Department of Ophthalmology, Beijing Children's Hospital. The diagnosis of UCSOP was based on a history of an early-onset head tilt, vertical misalignment, apparent overelevation and underdepression in adduction of the affected eye and a positive Bielschowsky head-tilt test result. The exclusion criteria included previous ocular surgery, congenital cataracts, retinal diseases, history of head trauma, systemic diseases affecting extraocular movement, and poorly cooperative subjects; In addition, those with a suspicion of bilateral SOP were also excluded, based on the clinical signs including positive head tilt test to both sides, alternating hypertropia in lateral gaze, esotropia in downgaze, overreaction of bilateral IO muscle, or/and underaction of bilateral superior oblique muscle. The patients who underwent surgery on other extraocular muscles were also excluded from the analysis.

All patients underwent objective ocular torsion measurements of both eyes through fundus photography using a TRC-50DX retinal camera (Topcon Medical System, Tokyo, Japan) just before and 1mo after the surgery. The head of the patients was stabilized in the holder, and the eye position was controlled according to the marker in the camera. The patients were asked to look at the internal fixation target to align the eyes at the primary position while the contralateral eyes were occluded, and the photographer ensured that the patient's head was straight during the examination using the side markers as a guide. Each eye was examined separately. All measurements were performed by the same experienced photographer.

The degree of fundus torsion was measured using the disk-foveal angle (DFA). The DFA was defined as the angle formed by a horizontal line extending from the geometric center of the optic disc (the intersection of the horizontal and vertical lines that bisect the optic disc) and a line connecting the fovea and the optic disc center[13], measured using Photoshop (Microsoft Corp., Redmond, WA; Figure 1). All measurements were performed by two experienced examiners separately, and repeated twice for every photograph, thus, the average of four measurements was obtained for further analysis. According to the measurement of DFA, we divided the 60 patients into two subgroups according to the laterality of extorsion, group 1: more extorsion in the paretic eye; group 2: more extorsion in the nonparetic eye.

All patients underwent complete examinations just before the surgery, including visual acuity (Teller acuity was recorded and converted to logMAR visual acuity for those could not use logMAR visual chart), refractive error, ocular motility, eye dominance, the prism and alternate cover test and
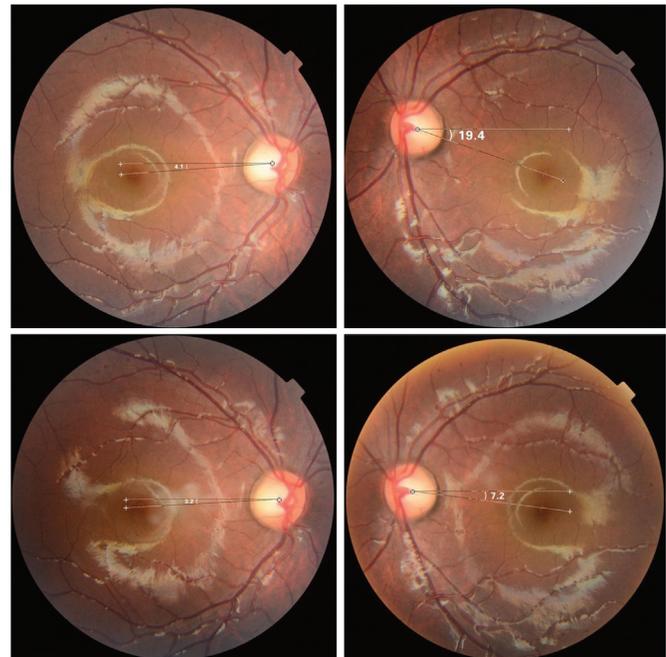

Figure 1 The DFA of the fundus pre- and post-operation In the nonparetic right eye, the DFA was 4.1 degrees and 3.2 degrees before and after the surgery, respectively. In the paretic left eye, the DFA was 19.4 degrees and 7.2 degrees before and after the surgery, respectively. DFA: Disk-foveal angle.

Bielschowsky's head-tilt test. Vertical deviation was measured by the prism cover test at the primary position and at distances of 33 cm and 5 m. The deviations of the other 8 diagnostic gaze positions were also measured. IOOA were graded according to the degree of over-elevation of the eye upon adduction scored on a scale ranging from 0 to +4 according to the distance between the lower limbus of cornea and the lower eyelid margin on superonasal fixation. If ≥4 mm, it means hyperactivity +4; if 3 mm, it means hyperactivity +3, 2 mm is hyperactivity +2, 1 mm is hyperactivity +1, <1 mm is defined as 0.

Different surgeries (myectomy or recession) were performed by one of three authors in consideration of the followings: deviation in primary gaze, the grading of IO overaction and SO underaction and forced duction test under general anesthesia during the surgery. Using a fornix incision, the IO muscle was isolated, for myectomy (IOOA +1 to +2 or vertical deviation <10), the IO muscle was disinserted, and clamped 10 mm from the original insertion. The part of the muscle between the clamp and the original insertion (approximately 6-8 mm) was transected and cauterized. For recession (IOOA +3 to +4 or vertical deviation ≥10), the procedure was performed as described by Park's with anchoring the IO to Park's point (3 mm posterior and 2 mm lateral to temporal edge of inferior rectus (IR) insertion[14].

Statistical Analysis Statistical analysis was conducted using SPSS version 23.0 (IBM Corporation, Armonk, NY,

Table 1 General characteristics

Parameters	All patients	Group 1	Group 2	P
Sex (M:F)	34:26	16:10	18:16	
Median age (range), y	3.9 (2.1-10.4)	3.8 (2.1-9.6)	4.0 (3.1-10.4)	
Visual acuity, logMAR	0.20±0.09 (0.1-0.6)	0.18±0.11 (0.1-0.5)	0.23±0.06 (0.1-0.6)	
Spherical equivalents, D	2.06±1.45 (-1.25 to +3.75)	2.78±1.95 (-0.5 to +3.75)	1.92±1.21 (-1.25 to +2.50)	
IOOA grade				
Preop.	+2.12±0.61 (+1 to +4)	+2.79±0.51 (+1 to +4)	+1.46±0.70 (+1 to +4)	0.89
Postop.	+0.31±0.08 (0 to +1)	+0.29±0.07 (0 to +1)	+0.34±0.10 (0 to +1)	1.00
P	0.021	0.014	0.035	
Vertical deviation, PD				
Preop.	13.55±5.73 (5-20)	14.16±4.79 (8-20)	12.95±6.68 (5-20)	0.65
Postop.	2.52±1.03 (0-5)	2.11±1.00 (0-4)	2.98±1.15 (0-5)	0.78
P	<0.001	<0.001	<0.001	

PD: Prism diopter; D: Degree; IOOA: Inferior oblique muscle overaction.

Table 2 Intra-group comparisons of DFA

Parameters	Group 1		P	Group 2		P
	Paretic eye	Nonparetic eye		Paretic eye	Nonparetic eye	
Preop.	13.21±5.95	7.97±4.25	<0.001	4.65±3.79[a]	13.16±5.35[a]	<0.001
Postop.	8.57±4.87	7.32±4.27	0.24	3.85±6.00[b]	9.94±5.45[c]	<0.001
t	4.87	0.85		0.99	3.41	
P	0.00	0.41		0.33	0.00	
Change in DFA	4.64±3.90	0.65±0.76	0.002	0.80±0.81	3.21±5.50	0.01

Group 1: More extorted paretic eye; Group 2: More extorted nonparetic eye; DFA: Disk-foveal angle. [a]$P<0.001$ vs Group 1; [b]$P=0.002$ vs Group 1; [c]$P=0.048$ vs Group 1.

USA). The Pearson Chi-square test was used to determine the difference in the percentage of excyclotorsion between the paretic and nonparetic eyes. The paired-samples t test was used to compare the DFA between the paretic and nonparetic eyes before and after surgery. The independent sample test was used to compare data between subgroups. A P value less than 0.05 was considered statistically significant.

RESULTS

A total of 60 patients who met the eligibility criteria were finally enrolled, including 34 males and 26 females. The median age of the participants was 3.9 (range: 2.1-10.4) y. The general clinical characteristics of the patients were listed in Table 1. Thirty-one patients presented with UCSOP in the right eye, and 29 patients presented with UCSOP in the left eye. Group 1 included 26 cases and group 2 included 34 cases, thus the rate of extorsion was higher in the nonparetic eye although insignificant ($P=0.10$).

The DFA in the paretic eye was compared before (8.36±6.43 D) and after (5.89±5.98 D) the surgery, and there was a significant reduction in the DFA ($P<0.001$). In the nonparetic eye, the difference in the DFA after (8.81±5.10 D) surgery compared to before (10.91±5.51 D) was also statistically significant ($P<0.001$). The change in the DFA was not significantly

different between the paretic (2.47±5.13 D) and nonparetic eyes (2.10±5.10 D; $P=0.73$).

We next analyzed the data by subgroups and between subgroups (Table 2). Interestingly, the change in the DFA of the more extorted eye in groups 1 (4.64±3.90) and 2 (3.21±5.50) was not significantly different ($P=0.30$).

DISCUSSION

The primary function of the superior oblique is incyclotorsion; thus, paralysis of the muscle can cause extorsion of the affected eye, which is one of the typical clinical manifestations of SOP[15-16]. However, this is not always the case; some researchers have found paradoxical results in that a large proportion of patients with unilateral SOP showed ocular excyclotorsion in the unaffected eyes[4-5]. Lee *et al*[6] also reported a rate of 38.7% for this discordance in acquired SOP. This study demonstrates extorsion at an even higher rate in the nonparetic eye (34:26) than in the paretic eye in children, although the difference was not statistically significant.

The reason why extorsion presents in the nonparetic eye may be due to the following two mechanisms. First, some patients with UCSOP have a large vertical fusional potential to overcome vertical diplopia, especially children, and when using fusional vertical vergence, there will be incycloduction

in the paretic eye and excycloduction in the contralateral eye; thus, extorsion may present in the unaffected eye[16]. Second, this presentation may also be related to the fixation during ocular development; when fixating with the paretic eye continuously, the amount of extorsion will decrease due to the increased strength of the superior oblique to reduce misalignment, which in turn will result in excycloduction in the nonparetic eye by conjugate movements, according to Hering's law[17]. However, if the torsion has been developed, the objective torsional measurements will not be altered by changing fixating eyes[18].

Although paretic and nonparetic eyes can both present with excyclotorsion, the surgery to correct the misalignment will often be performed on the affected eye, especially when accompanied by overelevation in adduction. All patients in our study presented with IOOA and underwent an IO weakening procedure. One month after the operation, the excyclotorsion of the paretic and nonparetic eyes was both reduced, the change of DFA in the paretic eyes and nonparetic eyes had no significant difference if all the patients were not divided into subgroups, which illustrates that surgery reduces excyclodeviation, and the torsional position of both eyes will change accordingly.

As a large proportion of patients presented with excyclotorsion in the nonparetic eye, which may have an impact on the statistical outcome, we analyzed the data of the two groups separately. Whether compared within or between groups, weakening of the IO could significantly reduce the excyclotorsion in the more extorted eye, regardless of which eye was paretic. There was no significant difference in the change in the DFA between the two groups. These outcomes illustrate that whether the paretic eye was the more or less extorted eye, the effect on the decrement of extorsion through the ipsilateral IO weakening procedure would reflect on the more extorted eye. The possible explanations of this phenomenon might be as the following: first, after IO weakening, the vertical misalignment will improve so that the need of fusional vertical vergence will reduce, thus the incycloduction in the paretic eye and excycloduction in the contralateral eye will reduce accordingly, so the more extorted nonparetic eye might be manifested as reduced extorsion. Second, if the paretic eye is the fixating eye, the fixation will prevent the paretic eye from dropping and incycloduction after IO weakening, accordingly, the excycloduction in the nonparetic eye will reduce by conjugate movements, according to Hering's law. It is considered that in patients with UCSOP accompanied by overelevation in adduction, fundus extorsion is a characteristic manifestation that can be alleviated or eliminated by IO weakening; thus, the reduction in extorsion is often used as a postoperative evaluation index. Through our research, we conclude that the pre- and postoperative evaluations of fundus torsion should always include both eyes, not only the paretic eye in case the unaffected eye is more extorted.

There were several limitations to this study. First, all of the participants in this study were children and some showed relatively poor cooperation, which may have had an impact on the fundus photography. We controlled for improper head positioning and fixation to minimize the effect of the error as mentioned in the methods. Second, Kim et al[19] found that ocular dominance can decrease the torsion in the dominant eye and hinder the abnormal ocular torsion from appearing in that eye. However, we did not determine the outcome of the dominant eye in some of the younger patients, which prevented us from analyzing the relationship between ocular dominance and extorsion. Third, the follow-up period was not long enough to evaluate the long-term effects of the surgery.

In conclusion, we demonstrate that excyclotorsion can present in the nonparetic eye in a large proportion of children with UCSOP and that weakening of the ipsilateral IO can have a more obvious effect on the decrement of extorsion in the more extorted eye regardless of which eye is paretic.

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