

Fusion can mask the relationships between fundus torsion, oblique muscle overaction/underaction, and A- and V-pattern strabismus

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PURPOSE	To evaluate relationships between fundus torsion, A- or V-pattern strabismus, and oblique muscle over- or underaction, and to explore the influence of stereopsis on these relationships.
METHODS	The medical records of patients with A or V patterns and/or abnormal ocular torsion seen at a single institution over nearly 30 years were retrospectively reviewed. Data collected were age, objective fundus torsion (estimated by indirect ophthalmoscopy), horizontal deviations in up- and downgaze, oblique muscle over- or underaction, and stereopsis.
RESULTS	A total of 396 patients were included. A patterns were observed in 121 patients (30.6%); V patterns in 90 (22.7%). Of the A-pattern patients, 73.6% had superior oblique muscle overaction, whereas 71.1% of the V-pattern patients had inferior oblique muscle overaction ($P < 0.0001$, $r = 0.71$), increasing to 78.6% and 86.3%, respectively, for patients without stereopsis ($r = 0.78$). Of the patients with fundus intorsion, 78.7% had superior oblique muscle overaction, whereas 74.4% of those with fundus extorsion had inferior oblique muscle overaction ($P < 0.0001$, $r = 0.79$), increasing to 83.5% and 82.8%, respectively, for patients without stereopsis ($r = 0.82$). Fundus intorsion occurred in 76% of the A-pattern patients, whereas fundus extorsion occurred in 71.1% of the V-pattern patients ($P < 0.0001$, $r = 0.73$), increasing to 78.6% and 86.3%, respectively, for patients without stereopsis ($r = 0.79$).
CONCLUSIONS	Strong correlations were found between fundus intorsion, superior oblique muscle overaction, and A patterns, and between fundus extorsion, inferior oblique muscle overaction, and V patterns. These correlations increased in patients without stereopsis, suggesting that the presence of binocular fusion can partially interfere with the close correlation of these parameters. (J AAPOS 2013;17:177-183)

Since 1897, when Duane first noted vertically intermittent horizontal strabismus,¹ A and V patterns have been attributed to various etiologies, including changes in the activity of the horizontal rectus muscles from up- to downgaze,^{2,3} paresis of the vertical rectus muscles,⁴ congenital nonparallelism of the planes of action of the oblique muscles,⁵ orbital pulley abnormalities,^{6,7} and oblique muscle dysfunction.⁸⁻¹¹ Numerous ophthalmologists have noted a relationship between the over- or underaction of the oblique muscles and the A

and V patterns of strabismus,⁸⁻¹⁶ and the oblique muscle dysfunction etiology is the predominant explanation for A- and V-pattern strabismus.¹² However, there have been few large-scale studies of the relationship between oblique muscle over- or underaction and A- and V-pattern strabismus. In one study of 118 patients with infantile esotropia,¹⁷ 81% of patients with inferior oblique muscle overaction had V patterns, and 83% with overacting superior oblique muscles had A patterns.

Fundus torsion in A and V patterns was first described by Piper^{18,19} and Weiss²⁰ in the 1960s.²¹ Weiss²² further proposed abnormal ocular torsion as the underlying factor causing A- and V-pattern strabismus and causing the clinical pattern of primary "overaction" or "underaction" of the oblique muscles simply by torsional rotation of the planes of action of the vertical and horizontal rectus muscles, contrary to the conventional interpretation.²³⁻²⁵ Guyton and Weingarten²⁶ resurrected Weiss's proposal and further suggested that the abnormal ocular torsion arises and persists as a consequence of reciprocal shortening or lengthening of the pairs of antagonist oblique

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muscles in the process that they dubbed “sensory torsion,” analogous to sensory exotropia in the horizontal plane.^{26,27} They believe that the shortening or lengthening of the pairs of oblique muscles occurs as a consequence of abnormal muscle length adaptation in the absence of, or deficiency in, the fusion-based neuromuscular control mechanism that normally maintains the proper torsional positions of the eyes.

With no or faulty feedback from the binocular fusion process, vergence is not properly guided, and if a small non-zero bias is present in the average vergence tonus, the eyes can slowly be driven inappropriately into abnormal strabismic deviations, which Guyton and Weingarten²⁶ call “sensory” deviations, via the vergence-guided processes of vergence adaptation and muscle length adaptation. When this process occurs horizontally, the result is usually sensory exotropia, with the deviation noted to occur bilaterally under general anesthesia.^{27,28} When this process occurs in the torsional control system, the result is “sensory torsion,” either bilateral extorsion with a V pattern and overacting inferior oblique muscles or bilateral intorsion with an A pattern and overacting superior oblique muscles.²⁶⁻²⁸

If Guyton and Weingarten’s hypothesis is correct, there should be close relationships and correlations between objective ocular torsion, A and V patterns, and oblique muscle overaction or underaction. Furthermore, if fusion is faulty but still present, ongoing vergence adaptation may hide the full correlation between these factors.^{26,28,29}

Brodsky and Donahue³⁰ proposed detailed neurologic mechanisms to explain why biases in cyclovergence tonus can exist, biases that are an integral part of Guyton and Weingarten’s proposed mechanism and assumed to exist by Guyton and Weingarten.²⁶ Brodsky and Donahue³⁰ appear to believe that the neurologic biases in cyclovergence tonus are the initial driving factor in oblique muscle over- or underaction, whereas in the view of Guyton and Weingarten,²⁶ any abnormality in the normal control mechanisms that maintain proper ocular torsion, most commonly a deficiency in binocular fusion, can allow even a small underlying bias in cyclovergence tonus to drive the eyes into abnormal torsional positions, with the end result being what they call “sensory torsion.” Brodsky and Donahue³⁰ appear to agree with this end result: “The ocular torsion produced by primary oblique muscle overaction also initiates a cascade of secondary mechanical events, including rotational displacement of the rectus muscle insertions, oblique muscle length adaptation, and mechanical tightening of the oblique muscles, as detailed elegantly by Guyton and Weingarten. These peripheral responses augment the overelevation or overdepression of the adducting eye and the corresponding A and V pattern observed clinically.” Whatever the initiating cause of this process, therefore, the result is that abnormal objective ocular torsion, A and V patterns, and oblique muscle overaction or underaction should be found to be closely interrelated.

Some authorities prefer the term *strabismus sursoadductorius* or simply *overelevation in adduction to inferior oblique*

Table 1. Exclusion criteria

Criteria	Number of patients excluded of 652
Previous horizontal rectus muscle surgery	113
Age <3 or inability to accurately assess pattern and torsion	47
Visual acuity <20/200 in one eye	29
Congenital syndromes, ^a cerebral palsy, thyroid disease, and dissociated vertical deviation	67
Total cases excluded	256

^aIncluding Brown, Duane, Ehlers-Danlos-like, congenital ablepharon, congenital fibrosis of extraocular muscles.

Table 2. Distance deviation and pattern of strabismus

	Exotropia	Esotropia	Hypertropia only	Totals
V pattern	45 (23)	44 (23)	1 (11.1)	90 (22.7)
No pattern	84 (42.9)	95 (49.7)	6 (66.7)	185 (46.7)
A pattern	67 (34.2)	52 (27.2)	2 (22.2)	121 (30.6)
Totals	196 (49.5)	191 (48.2)	9 (2.3)	396 (100)

(Values) are in %.

overaction, but in the combined Guyton/Weingarten and Brodsky/Donohue mechanisms, “overaction” can be considered appropriate if it simply means increased inferior oblique muscle effect, without implying etiology: the increased effect comes from oblique muscle length adaptation secondary to long-term, increased excyclovergence tonus to the oblique muscles, shortening the inferior oblique muscles and lengthening their antagonist superior oblique muscles.

The purpose of the present study was to analyze the relationships and correlations between the grade of objective ocular torsion, the amplitude of A and V patterns (the difference in the horizontal deviation in prism diopters between up- and downgaze), and the grade of oblique muscle overaction/underaction, and to further elucidate the role of residual fusion in confounding these correlations.

Subjects and Methods

The study was approved by the Johns Hopkins University School of Medicine Institutional Review Board in accordance with the guidelines of the Health Insurance Portability and Accountability Act. The medical records of all patients from the clinical practice of the senior author (DLG), coded with A- or V-pattern strabismus and/or abnormal fundus torsion, in the Krieger Children’s Eye Center at the Wilmer Institute between January 1980 and September 2009, were reviewed retrospectively. Exclusion criteria are listed in Table 1.

Data were collected on patient sex and age, objective amount of fundus torsion (grade estimated by indirect ophthalmoscopy from -4 to +4),³¹ horizontal deviations in up- and downgaze (in prism diopters), amount of oblique muscle overaction or underaction graded on a scale from +4 to -4, and stereoacuity.

An outward movement of the eyes of at least 10^Δ from up- to downgaze was considered to be a clinically significant A pattern,

Table 3. Correlation matrix for variables studied

	Patients with stereopsis			Patients without stereopsis			All patients		
	Size of pattern	Amount of oblique muscle over- or underaction	Amount of torsion	Size of pattern	Amount of oblique muscle over- or underaction	Amount of torsion	Size of pattern	Amount of oblique muscle over- or underaction	Amount of torsion
Size of pattern	1.00			1.00			1.00		
Amount of oblique muscle over- or underaction	0.57	1.00		0.78	1.00		0.71	1.00	
Amount of torsion	0.64	0.73	1.00	0.79	0.82	1.00	0.73	0.79	1.00

whereas an outward movement of at least 15^Δ from down- to up-gaze was considered to be a clinically significant V pattern.^{21,32}

The mean grade of oblique muscle overaction or underaction per patient was obtained by averaging the grade values for all 4 oblique muscles. The following sign convention was used in the averaging calculation: inferior oblique overaction and superior oblique underaction were both considered positive values; superior oblique overaction and inferior oblique underaction were both considered negative values. Trace muscle over/underaction was assigned a value of 0.5. For example, the maximum over/underaction mean grade of +4 occurs when there is bilateral +4 inferior oblique overaction and bilateral -4 superior oblique underaction, and the minimum over/underaction mean grade of -4 will occur when there is bilateral -4 inferior oblique underaction and bilateral +4 superior oblique overaction.

To analyze the relationship between horizontal deviation and each of the other variables, amounts of esotropia were assigned positive values, whereas amounts of exotropia were assigned negative values. Grades of fundus torsion were similarly assigned positive and negative values; intorsion was assigned a negative value, and extorsion was assigned a positive value. The net amount of fundus torsion between both eyes was then determined in each individual, with trace abnormal torsion being assigned a value of 0.5.

Statistical comparisons were performed using the χ^2 test and the *t* test. Correlations (for the data presented in the figures) were calculated with Pearson's correlation coefficient (*r*). A *P* value of <0.05 was considered to be statistically significant.

Results

Of 652 cases reviewed, 396 (186 males [47%]) were included. Of these, 196 (49.5%) had exotropia and 191 (48.2%) had esotropia. An A pattern was observed in 121 (30.6%) patients; a V pattern, in 90 (22.7%). No significant relationship with esotropia versus exotropia was found (Table 2). The mean age \pm SD of patients with an A pattern was 23.6 \pm 17.2 years, significantly older than the 17.6 \pm 19.2 years of patients with a V pattern (*P* = 0.02).

A correlation matrix (Table 3) illustrates that strong correlations exist between the size of the A or V pattern, the grade of oblique muscle overaction/underaction, and the grade of fundus torsion, decreasing in patients with stereopsis and increasing in patients without stereopsis.

Of the 121 patients with an A pattern, 89 (73.6%) had superior oblique muscle overaction. Of the 90 patients

with a V pattern, 64 (71.1%) had inferior oblique muscle overaction (Table 4). The relationships between superior oblique muscle overaction and A-pattern strabismus and between inferior oblique muscle overaction and V-pattern strabismus were statistically significant (*P* < 0.0001) based on χ^2 testing.

The percent of patients with superior oblique muscle overaction decreased to 62.2% (23/37) when only the patients with an A pattern who also had stereopsis were considered, and increased to 78.6% (66/84) when only considering the patients with an A pattern and no stereopsis. The percent of patients with inferior oblique muscle overaction was 86.3% (44/51) in V-pattern patients without stereopsis and 51.3% (20/39) in V-pattern patients with stereopsis. These differences between patients with and without stereopsis were statistically significant in both the V-pattern (*P* = 0.001) and A-pattern groups (*P* < 0.001).

Figure 1 illustrates the relationship between the amount of oblique muscle overaction or underaction and the size of either the A or V pattern in patients without (Figure 1A) and with stereopsis (Figure 1B). The strong correlation for this relationship (*r* = 0.71) increased slightly to *r* = 0.78 in patients without stereopsis and decreased to *r* = 0.57 in patients with stereopsis (Table 3).

To determine whether estimation of fundus torsion by indirect ophthalmoscopy was an accurate method, a Bland-Altman plot was constructed (Figure 2). Data for this plot were obtained from an unpublished study by the senior author (D.L.G.), in which estimates of ocular torsion by indirect ophthalmoscopy with measurements of torsion in the same eyes by fundus photography were compared. As one can see, estimation of fundus torsion by the use of indirect ophthalmoscopy is comparable with torsion assessment when fundus photography is used. The mean difference \pm 1.96 \times SD (95% CI) was -0.05 \pm 0.27 disk diameter, which corresponds to -0.4 \pm 2.16 grades of torsion (each grade equals 1/8 disk diameter of abnormal torsion outside the normal range).

Most patients with objective fundus intorsion (96/122 [78.7%]) were noted to have superior oblique muscle overaction. The opposite was true for patients with objective fundus extorsion, in whom most (116/156 [74.4%]) were noted to have inferior oblique muscle overaction (Table 5). Similarly, 96 of 121 of patients (79.3%) with

Table 4. Comparison of oblique muscle over- and underaction with A and V pattern in patients with and without fusion

	Without fusion	With fusion	Totals
V pattern (n = 90)			
IOM overaction	44 (86.3)	20 (51.3)	64 (71.1)
No overaction	6 (11.8)	17 (43.6)	23 (25.6)
SOM overaction	1 (1.9)	2 (5.1)	3 (3.3)
Totals	51 (56.7)	39 (43.4)	90 (100)
No pattern (n = 185)			
IOM overaction	42 (51.9)	32 (30.8)	74 (40)
No overaction	25 (30.9)	57 (54.8)	82 (44.3)
SOM overaction	14 (17.2)	15 (14.4)	29 (15.7)
Totals	81 (43.8)	104 (56.2)	185 (100)
A pattern (n = 121)			
IOM overaction	3 (3.6)	4 (10.8)	7 (5.8)
No overaction	15 (17.8)	10 (27)	25 (20.6)
SOM overaction	66 (78.6)	23 (62.2)	89 (73.6)
Totals	84 (69.4)	37 (30.6)	121 (100)

IOM, inferior oblique muscle; SOM, superior oblique muscle. (Values) are in %.

superior oblique overaction had fundus intorsion, whereas 116 of 145 (80%) with inferior oblique overaction had fundus extorsion. The χ^2 test revealed a statistically significant relationship between inferior oblique muscle overaction and fundus extorsion as well as a statistically significant relationship between superior oblique muscle overaction and fundus intorsion ($P < 0.0001$).

When considering only those patients with fundus intorsion but no stereopsis, the percent of patients with superior oblique muscle overaction increased to 83.5% (66/79) and decreased to 69.8% (30/43) when we considered only those who had stereopsis. This difference between with and without stereopsis was statistically significant ($P = 0.01$). For patients with fundus extorsion and no stereopsis, 82.8% (77/93) had inferior oblique muscle overaction, which decreased to 61.9% (39/63) in patients with fundus extorsion and stereopsis. This difference between with and without stereopsis was not statistically significant ($P = 0.17$).

The relationship between the grade of fundus torsion as estimated by indirect ophthalmoscopy, and the grade of oblique muscle over- or underaction is depicted in Figure 3. Oblique muscle over- or underaction and fundus torsion were strongly correlated ($r = 0.79$), with the correlation increasing slightly to $r = 0.82$ in patients without stereopsis (Figure 3A) and decreasing to $r = 0.73$ (Figure 3B) in those with stereopsis.

When A- or V-pattern strabismus and estimated fundus torsion were compared, we found strong correlations between V patterns and extorsion and between A patterns and intorsion ($r = 0.73$). This correlation coefficient also increased slightly to $r = 0.79$ when we considered only patients without stereopsis (Figure 4A) and decreased when we considered only patients with stereopsis (Figure 4B; $r = 0.64$).

Of 121 A-pattern patients, 92 (76%) had fundus intorsion, and 64 of 90 V-pattern patients (71.1%) had fundus extorsion. The percentages decreased in both groups

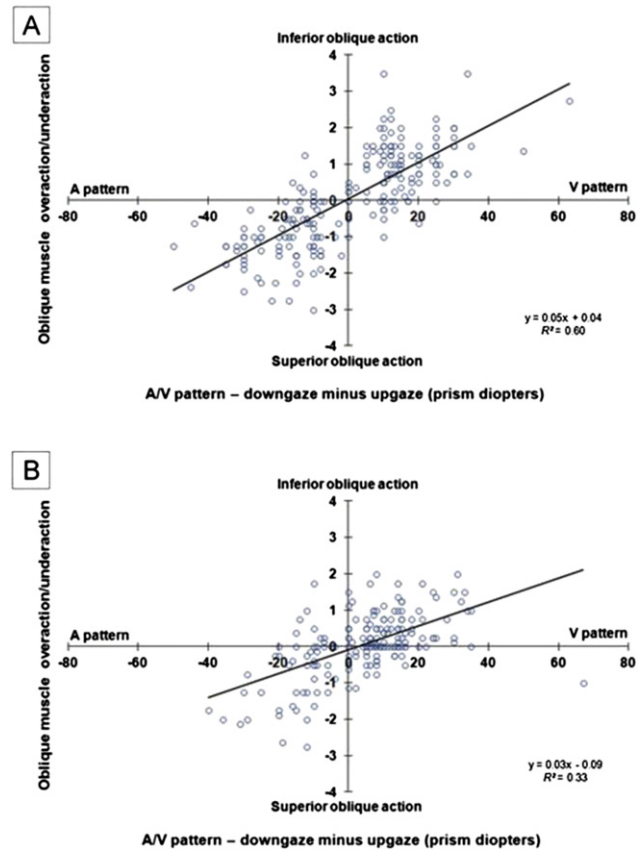


FIG 1. Relationship between oblique muscle overaction/underaction and the amount of A or V pattern in patients (A) without and (B) with stereopsis. Superior oblique muscle overaction is represented by negative values on the y-axis; inferior oblique muscle overaction, by positive values on the y-axis. The x-axis represents the degree of horizontal misalignment in prism diopters in downgaze minus the amount in upgaze (ie, positive values on the x-axis represent V patterns and negative values represent A patterns).

when we considered only patients with stereopsis and increased when considering only those without stereopsis. Specifically, the percent of patients with fundus intorsion was 78.6% (66/84) in A-pattern patients without stereopsis and 70.3% (26/37) in A-pattern patients with stereopsis, but the percent of patients with fundus extorsion was 86.3% (44/51) in V-pattern patients without stereopsis and 51.3% (20/39) in V-pattern patients with stereopsis. The χ^2 statistical test revealed a statistically significant relationship between fundus torsion and A- or V-pattern strabismus ($P < 0.0001$) as well as a statistically significant difference between patients with and without stereopsis in the V-pattern group ($P < 0.001$), but no significant difference existed between patients with and without stereopsis in the A-pattern group ($P = 0.27$; Table 6).

Discussion

This study revealed several findings that have not been previously reported in the literature and that should

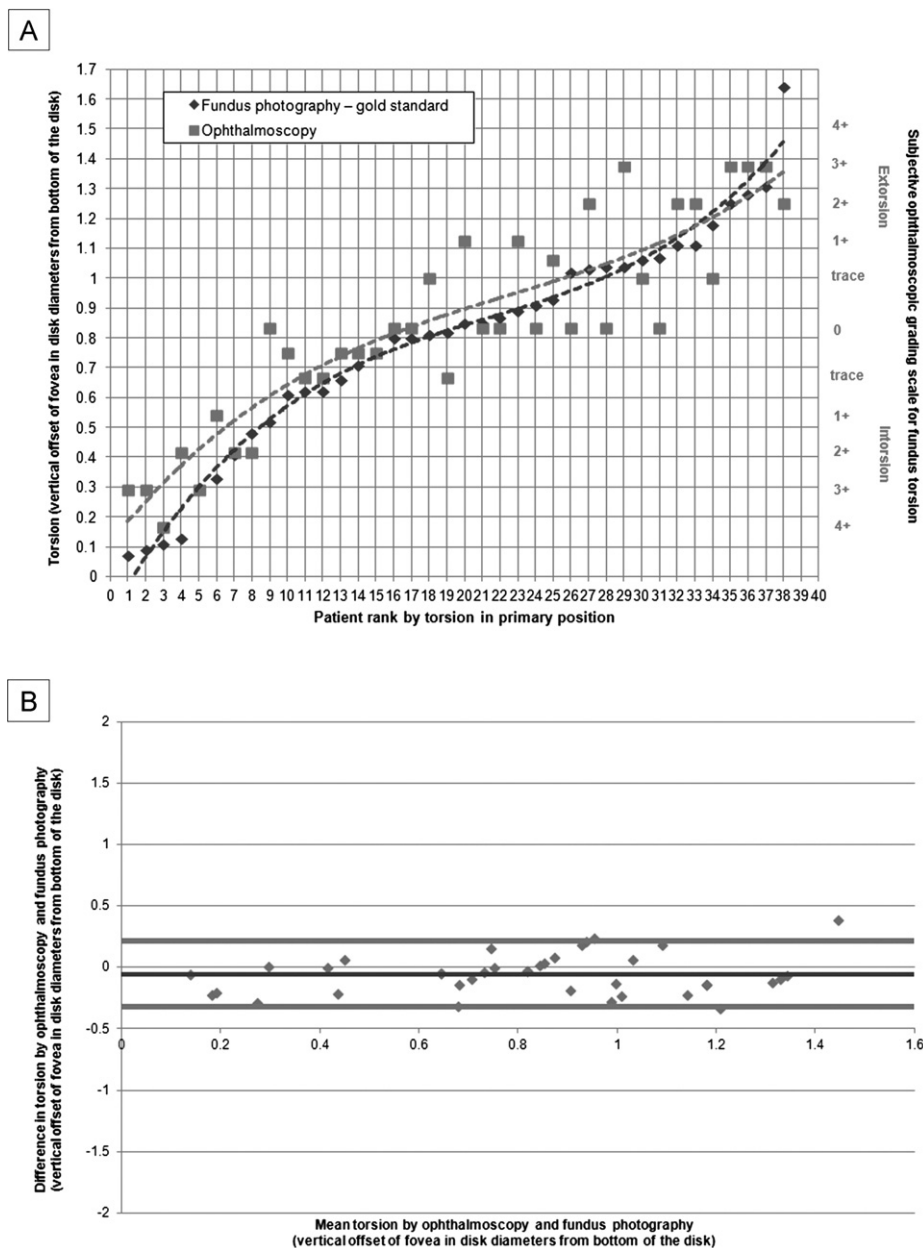


FIG 2. A, comparison of the measurement of fundus torsion with fundus photography and estimation with indirect ophthalmoscopy. The scale used for the former is on the left-hand vertical axis; for the latter, on the right hand. B, Bland-Altman plot in which the measurement of fundus torsion with fundus photography versus the estimation with indirect ophthalmoscopy is compared.

improve ophthalmologists' evaluation of patients and surgical planning. Various authors have noted that A and V patterns are related to overaction of the oblique muscles.⁸⁻¹⁶ This study confirms that V patterns are related to overaction of the inferior oblique muscles and that A patterns are related to overaction of the superior oblique muscles.

Weiss²² and later Guyton and Weingarten²⁶ proposed that abnormal ocular torsion, through rotating the planes of action of the rectus muscles, leads not only to A and V patterns in up- versus downgaze but also to apparent clinical over- or underaction of the oblique muscles on side gazes. The present study provides evidence supporting

such interrelationships. Our results also are consistent with the hypothesis that in the absence of significant binocular function (and thus when interference stemming from vergence adaptation is not possible), ocular torsion, A/V patterns, and oblique muscle over- or underaction are even better correlated, supporting the concept of the horizontal and vertical eye movements tending to follow the torted planes of action of the rectus muscles caused by "sensory torsion."^{26,28,29}

Liesch and Simonsz³³ reported that extorsion of eye movement patterns often developed when fusion was prevented in humans by monocular occlusion for 3 days. Miller and Guyton²⁹ reported the development of V

Table 5. Comparison of torsion with oblique muscle over- and underaction in patients with and without fusion

	Without fusion	With fusion	Totals
Extorsion (n = 156)			
IOM overaction	77 (82.8)	39 (61.9)	116 (74.4)
No overaction	13 (14)	21 (33.3)	34 (21.8)
SOM overaction	3 (3.2)	3 (4.8)	6 (3.8)
Totals	93 (59.6)	63 (40.4)	156 (100)
No torsion (n = 118)			
IOM overaction	10 (22.7)	16 (21.6)	26 (22)
No overaction	22 (50)	51 (68.9)	73 (61.9)
SOM overaction	12 (27.3)	7 (9.5)	19 (16.1)
Totals	44 (37.3)	74 (62.7)	118 (100)
Intorsion (n = 122)			
IOM overaction	2 (2.6)	1 (2.3)	3 (2.4)
No overaction	11 (13.9)	12 (27.9)	23 (18.9)
SOM overaction	66 (83.5)	30 (69.8)	96 (78.7)
Totals	79 (64.8)	43 (35.2)	122 (100)

IOM, inferior oblique muscle; SOM, superior oblique muscle. (Values) are in %.

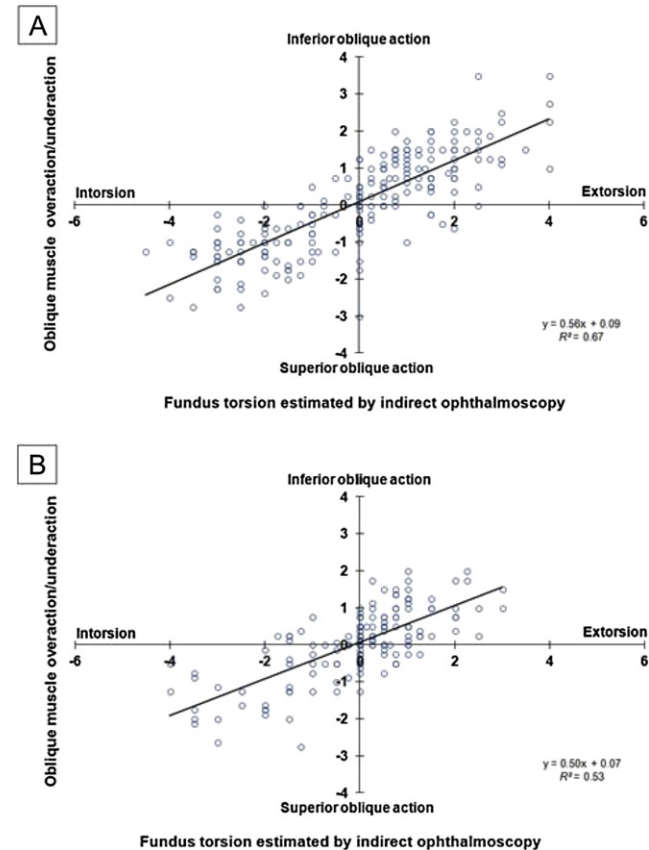


FIG 3. Relationship between the amount of fundus torsion as estimated by indirect ophthalmoscopy and the amount of oblique muscle overaction/underaction in patients without (A) and with (B) stereopsis.

patterns in 8 of 21 patients (38%) when losing fusion for at least 1 month after overcorrecting surgery for intermittent exotropia. Pott and colleagues³⁴ described elimination of vertical incomitancies and of V-pattern strabismus, with restoration of fusion after horizontal strabismus surgery

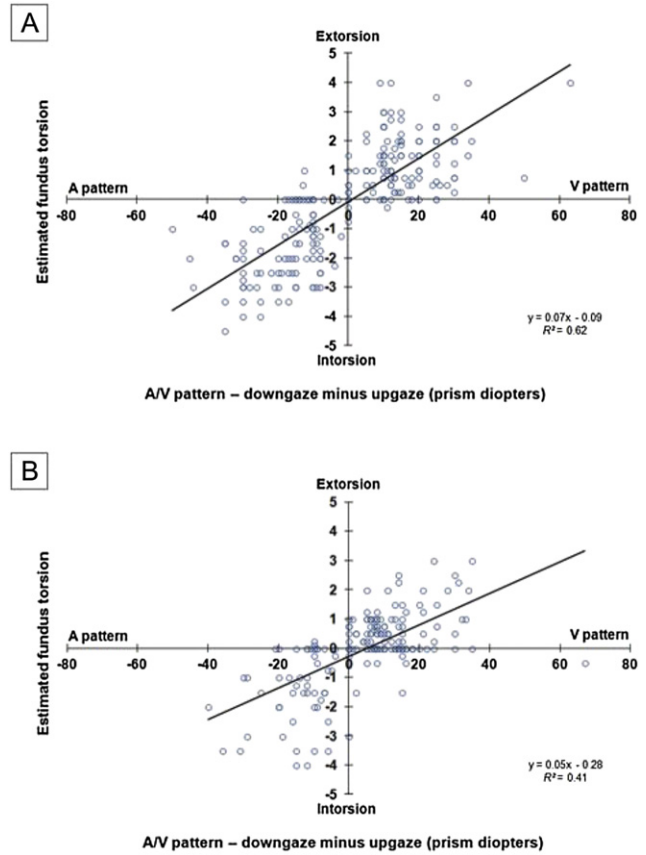


FIG 4. The relationship between the amount of A or V pattern and the amount of estimated fundus torsion in patients without (A) and with (B) stereopsis.

Table 6. Comparison of torsion with A or V pattern in patients with and without fusion

	Without fusion	With fusion	Totals
V pattern (n = 90)			
Extorsion	44 (86.3)	20 (51.3)	64 (71.1)
No torsion	7 (13.7)	16 (41)	23 (25.6)
Intorsion	0 (0)	3 (7.7)	3 (3.3)
Totals	51 (56.7)	39 (43.3)	90 (100)
No pattern (n = 185)			
Extorsion	47 (58.0)	43 (41.3)	90 (48.6)
No torsion	21 (25.9)	47 (45.2)	68 (36.8)
Intorsion	13 (16.1)	14 (13.5)	27 (14.6)
Totals	81 (43.8)	104 (56.2)	185 (100)
A pattern (n = 121)			
Extorsion	2 (2.4)	0 (0%)	2 (1.7)
No torsion	16 (19)	11 (29.7)	27 (22.3)
Intorsion	66 (78.6)	26 (70.3)	92 (76)
Totals	84 (69.4)	37 (30.6)	121 (100)

for cyclic esotropia and suggested loss of fusion as the likely cause for the development of V patterns and overelevation in adduction. These reports suggest that extorsion and V patterns are often held in check by binocular fusion, supporting our belief that residual fusion can interfere with

the correlations between ocular torsion, A and V patterns, and oblique muscle over- and underaction.

Our findings support current prevailing surgical treatments of patients with significant A- or V-pattern strabismus. For example, if the A or V pattern is accompanied by the typical fundus torsion and oblique muscle overaction, oblique muscle surgery is most likely the appropriate treatment. If, on the other hand, the typical fundus torsion and oblique muscle overaction do not accompany the A or V pattern, the oblique muscles are most likely not involved, and vertical offsets of the horizontal extraocular muscles will likely be more appropriate.

These clinical features and their interrelationships can also help guide diagnosis. For example, in the case of apparent overaction of the inferior oblique muscles without either fundus extorsion or a V pattern, the possibility of dissociated vertical deviation, which typically does not show abnormal fundus torsion, should be considered. Cases in which torsion, A or V pattern, and oblique muscle dysfunction are not related or correlated as expected may suggest an unusual mechanism at play that may not respond to customary surgical approaches. For instance, a V pattern associated with intorsion rather than extorsion is uncommon and could suggest an underlying Brown syndrome, in which this unusual pattern occurs secondary to an abnormally tight superior oblique muscle/tendon such that the involved eye flips outward from under the tendon when the patient attempts to look up.

In conclusion, this study revealed close relationships between fundus torsion, A and V patterns, and oblique muscle over- or underaction, namely, that fundus intorsion, A patterns, and superior oblique muscle overaction are strongly correlated, whereas fundus extorsion, V patterns, and inferior oblique overaction are strongly correlated. These correlations further increased in the absence of stereopsis, suggesting that binocular fusion can interfere with the close correlation between these parameters. These results can help ophthalmologists with clinical diagnosis and surgical planning by improving the understanding of the mechanisms underlying A- and V-pattern strabismus. In addition, they highlight the importance of accurately determining the type and degree of abnormal fundus torsion, which can assist in the differentiation between various clinical diagnoses.

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