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EXTENDED REPORT

Unexplained head tilt following surgical treatment of congenital esotropia: a postural manifestation of dissociated vertical divergence

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Background: Strabismus surgery for congenital esotropia can be complicated by the development of a postoperative head tilt.
Purpose: To determine the pathophysiology of acquired head tilting following horizontal realignment of the eyes in children with congenital esotropia.
Materials and methods: Retrospective analysis of nine children with congenital esotropia who developed unexplained head tilts following horizontal realignment of the eyes.
Results: Shortly after strabismus surgery, each child developed a head tilt in association with asymmetrical dissociated vertical divergence (DVD). Five children maintained a head tilt toward the side of the fixing eye (group 1), which did not serve to control the DVD. Four children maintained a head tilt toward the side of the hyperdeviating eye, which served to control the DVD (group 2). Children in group 2 had earlier horizontal muscle surgery and developed better stereopsis than those in group 1, suggesting that the higher degree of single binocular vision and stereopsis in these children may have led to a compensatory torticollis to control an asymmetrical DVD.
Conclusions: The onset of an unexpected head tilt after congenital esotropia surgery is usually a postural manifestation of asymmetrical DVD. In this setting, a head tilt toward the side of the fixing eye corresponds with a postural manifestation of the underlying central vestibular imbalance that produces DVD, while a head tilt toward the side of the hyperdeviating eye serves to counteract the hyperdeviation and stabilise binocular vision.

Congenital esotropia is characterised by a large angle crossing of the eyes that begins within the first six months of life.[1] Associated ocular motility disturbances such as inferior oblique overaction, latent nystagmus, and dissociated vertical divergence (DVD) may first appear following surgical realignment of the eyes.[1 2] Less commonly, a head tilt develops after successful horizontal realignment of the eyes.[3] This unexpected finding is often perplexing to the surgeon and frustrating to the parents, who complain that "the eyes are straight, but now he tilts his head all the time". In an attempt to elucidate the pathophysiology of this complication, we describe nine children with congenital esotropia who developed unexplained head tilts following successful horizontal realignment of the eyes.

MATERIALS AND METHODS

Medical records from children examined at Arkansas Children's Hospital and Texas Children's Hospital Eye Clinic, and San Paolo Hospital were retrospectively reviewed for unexplained head tilting following congenital esotropia surgery. After exclusion of patients with congenital nystagmus, cyclovertical muscle palsy, or primary oblique muscle overaction with baseline fundus torsion, nine children were found who met inclusion criteria. All nine children had been examined on multiple occasions. Historical findings included age at the time of strabismus surgery, surgical procedure performed, age at which the head tilt was first noted, age at which the DVD was first noted, presence or absence of a superimposed hypertropia, nature of strabismus surgery used to treat the head tilt, and surgical or non-surgical outcome.

A standard technique was used to measure the DVD and any associated hyperdeviation. The amplitude of DVD in each eye was measured by occluding the eye and adding base down prism behind the occluder until no hyperdeviation was observed when the occluder was shifted to the other eye. A superimposed hyperdeviation was detected by measuring a hypotropia of the other eye using alternate cover testing.

Sensory testing, occlusion testing, placement of prisms to match the vertical deviation, and Bielschowsky head tilt testing were used to determine whether the head tilt served to improve vertical alignment of the eyes and stabilise binocular vision. Sensory testing was performed with the head held in the compensatory position. A patient's head tilt was designated as *compensatory for binocular vision* when it improved vertical alignment of the eyes, and *non-compensatory for binocular vision* when it persisted despite of vertical misalignment of the eyes, when a contralateral head tilt restored improved vertical alignment, or when amblyopia was present in one eye.

RESULTS

Nine children developed enigmatic head tilts following horizontal strabismus surgery for congenital esotropia. In five children (group 1), the head tilt was directed towards the side of the fixing eye. In four children (group 2) the head tilt was directed towards the side of the hyperdeviating eye. In one child (case 3), a head tilt had been documented preoperatively and it became more conspicuous to parents and physicians after bimedial rectus muscle recessions were performed to treat congenital esotropia. In the other children, the onset of the postoperative head tilt usually preceded the detection of an asymmetrical DVD with a spontaneous hyperdeviation

Abbreviations: DVD, dissociated vertical divergence

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of one eye. In all children, the amplitude of the DVD was smaller in the fixing eye than in the spontaneously hyperdeviating eye. The associated head tilts were larger in group 2 (range 10–30 degrees) than group 1 (range 5–15 degrees) and were present during both near and distance fixation.

Four of five patients in group 1, and two of four patients in group 2 had latent nystagmus. Only one patient in each group had a coexistent head turn toward the side of the fixing eye. No child had an associated A or V pattern, although an apparent inferior oblique overaction of one eye, as manifested by a spontaneous elevation when the eye was adducted, was often an early manifestation of DVD. In both groups, the head tilt was present to a similar degree during distance and near fixation. When the spontaneous head tilt was directed towards the side of the fixing eye (group 1), a manifest DVD persisted despite the head tilt. Three of the five children in group 1 (cases 1, 3, and 4) maintained a spontaneous head tilt toward the side of the fixing eye that was found to resolve when the fixating eye was patched but not when the hyperdeviating eye was patched. One child (case 4) maintained a head tilt toward the side of the fixing eye despite the fact that Bielschowsky head tilt testing toward the opposite side eliminated the hyperdeviation. When the spontaneous head tilt was directed toward the side of the hyperdeviating eye (cases 6–9), Bielschowsky head tilt testing indicated that the head tilt served to control the DVD.

In general, children with head tilts toward the side of the hyperdeviating eye had earlier horizontal strabismus surgery and better stereopsis than those with a head tilt toward the side of the fixating eye (table 1). Patient age at the time of initial strabismus surgery ranged from 11 months to 4 years for group 1 and 7 months to 1 year for group 2. No patient in group 1 developed more than gross stereopsis (Titmus fly) whereas two patients in group 2 developed 100 seconds of arc. Since the head tilt in group 1 was not compensatory for binocular vision, subsequent vertical strabismus surgery to treat the head tilt was performed only in group 2. In one child in group 2 (case 6), the compensatory head tilt was reduced by vertical prisms and subsequently by vertical muscle surgery which reduced the asymmetry of the DVD in the two eyes. In one other patient in group 2 (case 8), vertical rectus muscle surgery reduced the size of the head tilt.

DISCUSSION

Previous studies have documented an association between the development of a head tilt in children with surgically corrected congenital esotropia and the presence of DVD.[3–9] The curious finding in these cases is that the head tilt may be directed towards or away from the side of the fixating eye. This observation raises the question of whether the head tilt arises from an underlying central vestibular imbalance that alters the patient's vertical orientation, or whether it serves to inhibit the hyperdeviation in one eye and thereby stabilise binocular vision. Making this determination requires the acquisition of sensory data, field measurements, and head tilt testing, which can be difficult to obtain in young children. For this reason, it is often necessary to follow affected children longitudinally to establish the underlying cause of the head tilt.

Crone found head tilting in 26 of 113 children with DVD.[4] In ''the great majority'', the head was tilted towards the shoulder on the side of the fixing eye. Crone proposed that this head tilt was compensatory for the incycloduction of the fixing eye. Since the hyperdeviation decreased rather than increased when the head was tilted toward the opposite shoulder, Crone concluded that ''the quest for binocular vision is quite excluded as a cause of torticollis in all these cases.'' Lang found an abnormal head tilt in 29 of 82 children with congenital esotropia and DVD.[3] In 18 children, the head was inclined toward the side of the fixing eye, while in 11 children the head was inclined toward the side of the hyperdeviating eye. Lang also noted that the head tilt was not adopted to avoid diplopia.[3]

In a more recent prospective study, Betchel *et al* found a manifest head tilt in 26/74 (35%) of patients with DVD, no prior vertical muscle surgery, and an ocular fixation preference.[5] Eighteen of 26 (69%) of these patients had a head tilt toward the side of the hyperdeviating eye. In 19% of patients with manifest head tilts, the head tilt produced no qualitative or quantitative change in the DVD. In another prospective study of anomalous head postures with DVD by Santiago and Rosenbaum, twelve of 14 patients tilted their heads towards the side of the fixing eye, whereas two patients tilted their heads towards the side of the hyperdeviating eye.[6] The authors noted that head tilting did not occur in their patients until after strabismus surgery. They postulated that surgical realignment permitted fusional mechanisms to function, and that head tilting was probably advantageous for fusion. In a series of consecutive cases with DVD, Prieto-Diaz *et al* found a head tilt in 26 patients (49%).[7 8] The head tilt was directed towards the side of the fixing eye in 19 patients and towards the side of the hyperdeviating eye in seven patients. De Decker and Dannheim-de Decker noted a head tilt in 29 of 73 patients who were operated on for bilateral DVD.[9] Nineteen patients had a head tilt toward the side of the fixing eye, and 10 had a head tilt toward the side of the hyperdeviating eye. These studies establish a dichotomy with respect to the direction of head tilt relative to the fixing eye, and the compensatory (that is, adapted to modulate the DVD and improve binocularity) versus non-compensatory (that is, centrally driven) nature of the tilt.

The evolutionary basis for DVD and its associated torticollis can be traced back to visuo-vestibular postural responses which are most pronounced in fish and insects.[10–12] In the upright fish, unequal light input to the two eyes produces a body tilt in the roll (frontal) plane, causing the dorsal aspect to rotate toward the side with greater light input (dorsal light reflex).[11 12] In a vertically restrained fish, unequal visual input to the two eyes produces a vertical divergence of the eyes, with depression of the eye that has greater visual input and elevation of the eye that has less visual input.[12] This primitive visuo-vestibular response orients the dorsal side of the animal toward the sky, and maintains equal visual input in the two eyes to maintain visual vertical orientation.[13] DVD conforms to a human dorsal light reflex, which realigns the eyes to a tilted visual orientation when congenital esotropia precludes normal binocular visual development.[10 13–15]

Invoking the human dorsal light reflex as the underlying cause for the head tilt in group 1 would explain why these patients maintain a head tilt despite a manifest hyperdeviation.[3 4] In the same way that a primitive dorsal light reflex encourages a body tilt towards the side of the eye with greater visual input, a human dorsal light reflex would be expected to encourage a head tilt toward the side of the fixing eye.[13 14] Although we have designated this head tilt as *non-compensatory for binocular vision*, it is a compensatory postural adaptation at the central vestibular level in the sense that it serves to realign the head to a tilted internal representation of vertical (fig 1, left).[13 14] This central vestibular imbalance may explain why one child in group 1 (case 4) maintained a left head tilt with a manifest hyperdeviation of the right eye despite the fact that the hyperdeviation resolved when the head was tilted to the right. A subjective visual tilt following occlusion of one eye has recently been shown in humans with DVD.[15]

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Table 1 Clinical Findings

Case	Age BMR performed	Direction of head tilt/age first noted	DVD characteristics	Head tilt test	Sensory	Response to patch test and to treatment
Group 1: Head tilt toward from side of fixing eye						
1	4 years	Left/postop	R>L	NP	Bagolini - ARC; Randot - no stereopsis	Head tilt resolved with patching OS but not OD
2	4 years	Left/postop	R>L	NP	Bagolini - ARC; Randot - no stereopsis	NP
3	21 months	Right/18 months	L>R	NP	Titmus - negative	Head tilt resolved with patching OD but not OS; symmetrical SR rec - NI
4	12 months	Left/25 months	R>L	↑ RHT with left head tilt; RHT resolves with right head tilt	Bagolini - ARC; W4D - supp OD	Head tilt resolved with patching OS but not OD
5	11 months	Right/18 months	L>R	LHT persists with head tilt to right	Bagolini - ARC; Titmus+fly W4D fusion	Head tilt resolved with patching; LHT resolved with LIOAP
Group 2: Head tilt toward side of hyperdeviating eye						
6	11 months	Left/18 months	L>R	↑ LHT with right head tilt resolves with left head tilt	DMR - no torsion; Bagolini - int supp OD; Titmus 100 sec/arc; W4D vertical diplopia	Vertical prisms eliminated head tilt; BSR+RIRres reduced head tilt
7	7 months	Right/18 months	R>L	↑ RHT with left head tilt; ↓ RHT with right head tilt	Bagolini - NRC; W4D - alt supp; Titmus+fly	NP
8	11 months	Right/postop	R>L	↑ RHT with left head tilt; ↓ RHT with right head tilt	Bagolini - ARC with int supp OS; Titmus 100 sec/arc; W4D -int supp OD	Head tilt decreased; RLRrec; LIRrec
9	1 year	Right/postop	R>L	↓ RHT with right head tilt; ↑ RHT with left head tilt	Bagolini ARC	Symmetrical BSR rec - NI

M, male; F, female; —, unknown; NP, not performed; L, left; R, right; ↑, increased; ↓, decreased; DMR, Double Maddox Rod; LHT, left hyperdeviation; RHT, right hyperdeviation; NI, no improvement; BMR, bimedial recession; W4D, Worth Four Dot; sec/arc, seconds of arc; NRC, normal retinal correspondence; ARC, anomalous retinal correspondence; int supp, intermittent suppression; alt supp, alternate suppression; IOAP, inferior oblique anteroplacement; SV, subjective vertical; postop, noted shortly after surgery; rec, recession; res, resection.

Some have questioned whether a head tilt toward the side of the fixing eye could serve to improve visual acuity by damping latent nystagmus.[16 17] This mechanism seems implausible since any static ocular counterroll would primarily damp the torsional component of latent nystagmus, which does not displace the image of regard from the fovea and therefore does not degrade visual acuity.[18] In this group, a head turn towards the side of the fixing eye would be the necessary strategy to lessen the horizontal component of latent nystagmus, stabilise foveation, and improve monocular visual acuity. Lang[3] reported that patients corresponding to group 1 commonly have a head turn toward the side of the fixing eye. Although four children in our group 1 had latent nystagmus, only one child had a coexisting head turn towards the side of the fixing eye. It is unlikely that the other three patients with latent nystagmus were maintaining a head tilt to selectively reduce the torsional component in the absence of a head turn to reduce the horizontal component.

The DVD associated head tilts in group 2 present a different set of diagnostic challenges. In these patients, it may be difficult to distinguish a manifest hyperdeviation from an occlusion hyperphoria since the compensatory head tilt serves to neutralise the manifest hyperdeviation. Furthermore, Bielschowsky head tilt testing is particularly difficult to quantify in young children with bilateral DVD. Therefore, a manifest hyperdeviation must be carefully sought after the patient's head has been placed in the upright position. Only when a manifest hyperdeviation of one eye can be identified, the amplitude of the DVD in both eyes measured, and Bielschowsky head tilt testing performed, can the compensatory nature of the postoperative head tilt be established.

Jampolsky has characterised the Bielschowsky head tilt response in DVD as one in which a head tilt to either side increases the hyperdeviation of the contralateral eye.[19 20] This response, which is opposite to that seen in patients with superior oblique palsy,[19] reflects the additive effects of utricular stimulation and oblique muscle innervation associated with DVD.[13 14] When unequal visual input evokes a hyperdeviation of the left eye, for example, visuo-vestibular innervation activates the right superior and left inferior oblique muscles to produce a cyclovertical divergence[13] (fig 1, right). This cyclovertical divergence then necessitates secondary fixational innervation to maintain monocular fixation with the lower (visually preferred) eye.[10 17] A dorsal light reflex in humans would cause the head to tilt away from the side of the hyperdeviating eye (fig 1, left) which would increase the left hyperdeviation as a right head tilt activates otolithic innervation predominantly to the right superior

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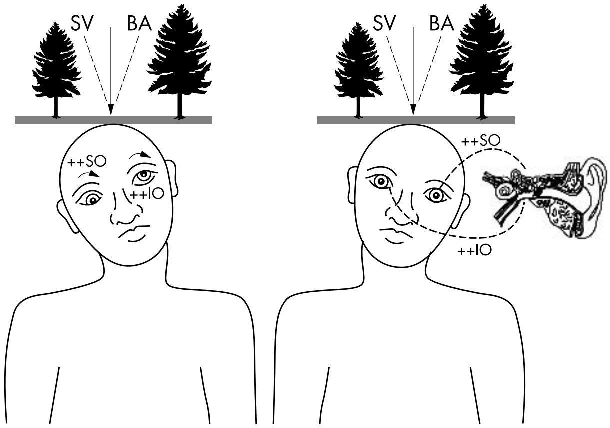

Figure 1 Components of the head tilt response in DVD. Left: head tilt toward the side of the fixing eye (group 1). A human dorsal light reflex induces a tilt of the subjective vertical (SV) towards the side of the fixing eye and a vertical divergence of the eyes. A head tilt to align the head with the tilted subjective vertical would be necessary to maintain vertical orientation. Right: compensatory head tilt in DVD. A patient with DVD and a hyperdeviation of the left eye (left figure) can use a compensatory head tilt to the left to recruit otolithic innervation to neutralise the DVD innervation and nullify the existing vertical divergence and restore binocular alignment (BA) of the eyes.

oblique and left inferior oblique muscles.[13 14] Conversely, a head tilt to the left would recruit otolithic innervation predominantly to the right inferior oblique and left superior oblique muscles, which serve to neutralise a left DVD associated hyperdeviation and improve vertical alignment (fig 1, right). Thus, by tilting the head towards the side of the more hyperdeviating eye, children in group 2 could calibrate their head position to modulate DVD in the two eyes and thereby stabilise binocular fusion.[13 14] Both patients who underwent strabismus surgery in an attempt to equalise the hyperdeviations showed reduction in the size of the head tilt and displayed some residual asymmetry in the hyperdeviations between the two eyes.

According to Jampolsky, DVD can eventually lead to secondary superior rectus contracture in the hyperdeviating eye.[20] Superior rectus contracture can reverse the head tilt response in DVD, causing a hyperdeviation in the affected eye to increase with ipsilateral head tilt and decrease with contralateral head tilt.[20] In this setting, a compensatory head tilt to the side of the fixating eye may be used to minimise otolithic innervation to the tight superior rectus muscle, while a head tilt towards the side of the higher eye will augment the hyperdeviation. When a superior rectus contracture develops, surgical recession of the tight superior rectus muscle can reduce or eliminate the compensatory head tilt.[6]

Only one of our patients in group 1 developed evidence of a superior rectus contracture (case 5), as evidenced by a head tilt away from the hyperdeviating eye and an induced hypotropia of the fixating eye when it was patched. Although three of our patients in group 2 had a superimposed hypertropia, they manifested a head tilt towards the side of the hyperdeviating eye, which is inconsistent with superior rectus muscle contracture. It therefore appears that a superimposed hypertropia can develop in some patients with asymmetrical DVD without other clinical signs of superior rectus contracture. It is also possible that these patients had a small coexistent hypertropia that was unrelated to the DVD (perhaps caused by unequal vertical positioning of the medial rectus muscles during surgery or by undetected unilateral inferior oblique muscle overaction). In a young child, a bilateral symmetrical DVD could mask a small hypertropia to produce a clinical appearance that simulates asymmetrical DVD. Figure 2 summarises the probable mechanisms by which DVD can manifest with head tilting in children with surgically treated congenital esotropia.

The dichotomy in the direction of head tilt relative to the fixing eye in this study suggests asymmetrical DVD induces a schizophrenic situation in which the need for vertical orientation and the need for vertical ocular alignment create conflicting postural drives. One the one hand, a head tilt towards the side of the fixing eye that is necessary to re-establish vertical orientation will increase the hyperdeviation of the contralateral eye (fig 1, left). On the other hand, a head tilt towards the side of the hyperdeviating eye that is necessary to minimise the DVD associated hyperdeviation will disrupt vertical orientation (fig 1, right). The neutral head position maintained by many patients with DVD may therefore represent a compromise position. To the extent that there is little binocular vision and an asymmetric DVD, however, one might expect the drive for vertical orientation to override, resulting in a head tilt towards the side of the fixing eye (that is, one that is driven by a human dorsal light reflex and non-compensatory for binocular vision).

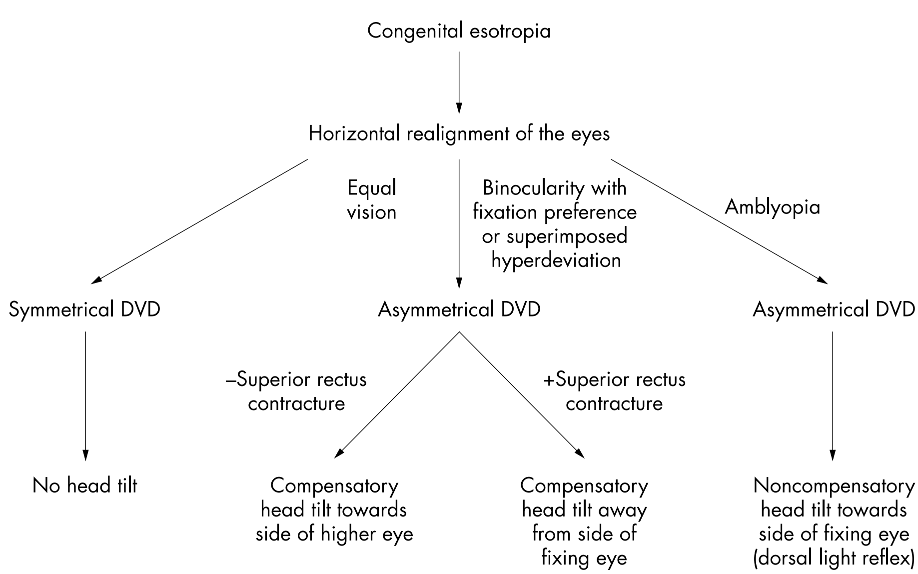

Figure 2 Clinical algorithm depicting postural mechanisms for head tilting in congenital esotropia.

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Alternatively, a strong potential for fusion and stereopsis would cause the drive for binocular vision to override, resulting in a head tilt towards the side of the hyperdeviating eye (which is compensatory for binocular vision). The earlier surgery and better development of stereopsis in group 2 compared with group 1 provides preliminary support for this mechanism.

From a therapeutic perspective, our findings suggest that strabismus surgery to neutralise an asymmetric hyperdeviation should eliminate the compensatory head tilt in group 2. As a head tilt attributable to a dorsal light reflex does not exist to realign the eyes, surgical treatment of the associated hyperdeviation should not eliminate it.[14] Prieto-Diaz and Souza-Dias[8] and von Noorden[21] have eliminated the head tilt in patients corresponding to group 1 by performing surgery to torsionally rotate the fixing eye in the direction of the head tilt. We believe that torsional rotation of the fixing eye may counterrotate a tilted subjective vertical back to true vertical, thereby eliminating the internal postural drive for the head tilt.

Our study needs to be viewed in terms of its inherent limitations. First, it is a retrospective study, so that all diagnostic tests were not performed on every patient. Ideally, it would be desirable to prospectively assess occlusion of each eye, prismatic neutralisation of the hyperdeviation, Bielschowsky head tilt test, and monocular subjective visual vertical in each patient. Second, our patient numbers for each group were small, so this study should be viewed as a pilot study that requires future confirmation by prospective studies. Third, our results cannot be applied to all patients with congenital esotropia who underwent horizontal strabismus surgery because we excluded patients with primary oblique muscle overaction to eliminate the potentially confounding effects of torsion in a monocularly viewing patient. Finally, while the findings in group 1 of a non-compensatory head tilt, a manifest hyperdeviation of the contralateral eye, and head tilt resolution with patching of the fixing eye but not the hyperdeviating eye (since the hyperdeviating eye is already suppressed) are all consistent with the visuo-vestibular disturbance that would occur with a human dorsal light reflex, definitive confirmation of this mechanism must await further study of the orientation of the subjective vertical and its correlation to the head position in these patients.

In conclusion, an acquired head tilt in the child with surgically corrected congenital esotropia seems to be a postural manifestation of asymmetrical DVD. The head tilt may be the initial sign of DVD, and its underlying mechanism may take years to elucidate. Our findings indicate that a head tilt towards the side of the fixing eye reflects a prenuclear imbalance in central vestibular tone corresponding to a dorsal light reflex, while a head tilt towards the side of the hyperdeviating eye is a compensatory posture to promote vertical binocular alignment. In both groups, the potential role of occlusion therapy in altering the head position needs to be elucidated. When a head tilt persists after occlusion therapy, strabismus surgery to equalise the hyperdeviation in the two eyes should eliminate a compensatory head tilt that is directed towards the side of the higher eye. When a head tilt is directed towards the side of the fixing eye, surgery to torsionally rotate the fixing eye in the direction of the head tilt may have application.

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