PEDIATRICS

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Objective ocular torsion outcomes after unilateral horizontal rectus surgery in infantile esotropia

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Abstract

Purpose To analyse objective ocular torsion among patients with infantile esotropia and to determine the effects of unilateral horizontal rectus surgery.

Methods Sixty-eight patients (136 eyes) (range 4 to 16 years) who underwent unilateral horizontal rectus surgery for infantile esotropia participated in this retrospective single-centre study. Objective ocular torsion using fundus photography was assessed before surgery and 1 year later. We defined three groups of patients based on preoperative qualitative objective ocular torsion: physiological extorsion and pathological extorsion and intorsion. For each group, the disc-foveal angle was measured and analysed both before and after surgery. We looked for possible correlations between amount of esodeviation and disc-foveal angle size.

Results Preoperatively, 28 (41%) patients had + 6.73 (± 2.66) degrees of physiological extorsion. Thirty-one (46%) patients had + 12.94 (± 3.67) degrees of pathological extorsion. Nine (13%) patients had − 1.99 (± 2.52) degrees of intorsion. After surgery, the number of subjects with physiological extorsion increased to 45 (66%). The number of patients with pathological extorsion decreased to 17 (25%) and the mean disc-foveal angle was significantly reduced by 1.80°. Six (9%) patients presented intorsion and the mean disc-foveal angle was significantly reduced by 2.28°. For the pathological extorsion group, the size of the disc-foveal angle before surgery was positively correlated to its reduction after surgery. Disc-foveal angle variation and distance esodeviation variation after surgery were positively correlated.

Conclusions These results highlight that pathological objective ocular torsion can be frequently found in infantile esotropia and is decreased after unilateral recession-plication surgery.

Keywords Infantile esotropia · Strabismus surgery · Torsion abnormality · Fundus photography

Introduction

Eye movements can be described around a theoretical centre according to Fick's theory [1] with three main axes. Horizontal rotation of the eye occurs around the vertical axis, vertical rotation around the horizontal axis, and torsional rotation around the visual axis. This three-dimensional oculomotor control minimizes image disparity, thus making fusion possible. Ocular torsion is an essential component of this system. It can be measured objectively or subjectively. Because of its simplicity and ease of application, fundus photography is currently employed as a standard for ocular torsion measurement. It has been reported that ocular torsion assessment using fundus photography is within a range in normal individuals [2]. On the other hand, some authors have rather focused on ocular torsion in cyclovertical strabismus, such as fourth nerve palsy, A-pattern or V-pattern strabismus, or dissociated vertical deviation [3–8]. In contrast, few studies have been published on ocular torsion among patients with infantile esotropia, characterized by primary esotropia with onset before 6 months of age at risk of impaired binocular vision. Most of these assessed ocular torsion as a predictive factor for the outcome of vertical disorders [9, 10]. Since infantile esotropia belongs to the category of horizontal strabismus, cyclotorsion

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is not regarded as a main factor influencing fusion capacities. However, infantile esotropia is observed in approximately 1% of children, making this an important issue for paediatric ophthalmologists [11]. Several other ocular motor signs can be observed, e.g., dissociated vertical deviation, latent nystagmus, inferior oblique overaction, and amblyopia. Those clinical findings have an influence on objective ocular torsion. Conversely, in their absence, ocular torsion should be expected to be within the range of normal individuals. In our clinical experiment, we observed fundus torsion in a large number of patients with infantile esotropia and also noticed variations after unilateral horizontal rectus surgery. We aimed to describe the objective ocular torsion among patients with infantile esotropia without significant vertical deviations and to determine the effect of unilateral horizontal rectus surgery.

Materials and methods

We conducted a retrospective observational single-centre study in our ophthalmology department, at the University Hospital of Tours from March 2009 to March 2015. Inclusion criteria for the patients were as follows: onset of esotropia before 6 months of age, distance or near esodeviation superior or equal to 15 prism dioptres as measured by the prism and alternate cover test, and first time horizontal strabismus surgery. Considering the potential influence of the following disorders on objective ocular torsion, patients with vertical deviation in primary position or in any gaze superior to 10 prism dioptres, A-V pattern strabismus, fourth nerve palsy, dissociated vertical deviation of with any fixation threatening condition such as amblyopia were excluded from this study. The study was performed in accordance with the ethical standards laid down in the Declaration of Helsinki and was approved by the Ethic Committee of the University Hospital of Tours, France. All data were collected and processed anonymously.

Surgical procedure

Patients underwent unilateral strabismus surgery on the most deviated eye under general anaesthesia. The lateral rectus was strengthened with plication, while the medial rectus was weakened either with recession or posterior fixation sutures (Cüppers' operation) or both combined.

Objective torsion assessment

Objective ocular torsion was measured on fundus photography using a Topcon fundus camera, both before surgery and 1 year later. Patients were asked to look at an internal fixation target in order to align their eyes in the primary position.

Photoshop CC 2014 software was used to evaluate objective quantitative and qualitative ocular torsion (Fig. 1).

Objective quantitative ocular torsion was assessed by measuring, in degrees, the angle between the line crossing the centre of the optic disc, the fovea, and the horizontal line passing through the centre of the optic disc, called the disc-foveal angle. The procedure used to determine the centre of the optic disc consisted in drawing the rectangle surrounding the edges of the optic disc and then the rectangle diagonals. The intersection of these two diagonals provided the centre of the optic disc.

Objective qualitative ocular torsion assessment was related to the position of the fovea. When the fovea was located between the horizontal line passing through the centre of the optic disc and the line at the bottom edge of the optic disc, the eye was considered to be in physiological extorsion. When the fovea was located bellow the horizontal line at the bottom edge of the optic disc, the eye was considered to be in pathological extorsion. When the fovea was located above the horizontal line passing through the centre of the optic disc, the eye was considered to be in pathological intorsion.

Accordingly to the position of the fovea, we defined three groups of patients based on preoperative qualitative objective ocular torsion: physiological extorsion and pathological extorsion and intorsion (Table 1). No patient presented one eye in pathological extorsion and the other in intorsion.

Extorsion angle was noted in positive values and intorsion angle in negative values. Statistical analyses were performed considering mean objective quantitative ocular torsion between both eyes.

Statistical analyses

Statistical analyses using R software version 3.2.3 were carried out to perform a paired Student's t test for normal distributed data. Otherwise, the paired Wilcoxon's test was used. Distribution normality was tested with a Shapiro test. Correlations were assessed by univariate regression. Results were considered statistically significant if the p value was less than 0.05. An α level of 0.05 was used for all statistical tests.

Results

A total of 68 subjects (136 eyes), including 36 (52%) males, were assessed in our study. The mean age was 6.9 years (± 2.6; age range 4 to 16 years). Surgery was performed on the left eye in 59 cases (87%) (Table 2). Surgery age distribution is described in Fig. 2.

Preoperatively, 28 (41%) patients had $+ 6.73°$ (± 2.66) of physiological extorsion. Thirty-one patients (46%) presented pathological extorsion with a mean disc-foveal angle of $+ 12.94°$ (± 3.67), though this was highly variable (range

Fig. 1 Assessment of objective ocular torsion on fundus photography. **a** Disc-foveal angle was measured from two straight lines: the first line passing through the geometric centre of the optic disc and the second one joining the centre of the optic disc to the fovea. **b** Physiological extorsion was defined by the fovea laying in the area between the centre and the lower edge of the optic disc. **c** Pathological extorsion was defined by the fovea being located below the lower edge of the optic disc. **d** Intorsion was defined by the fovea being located above the centre of the optic disc
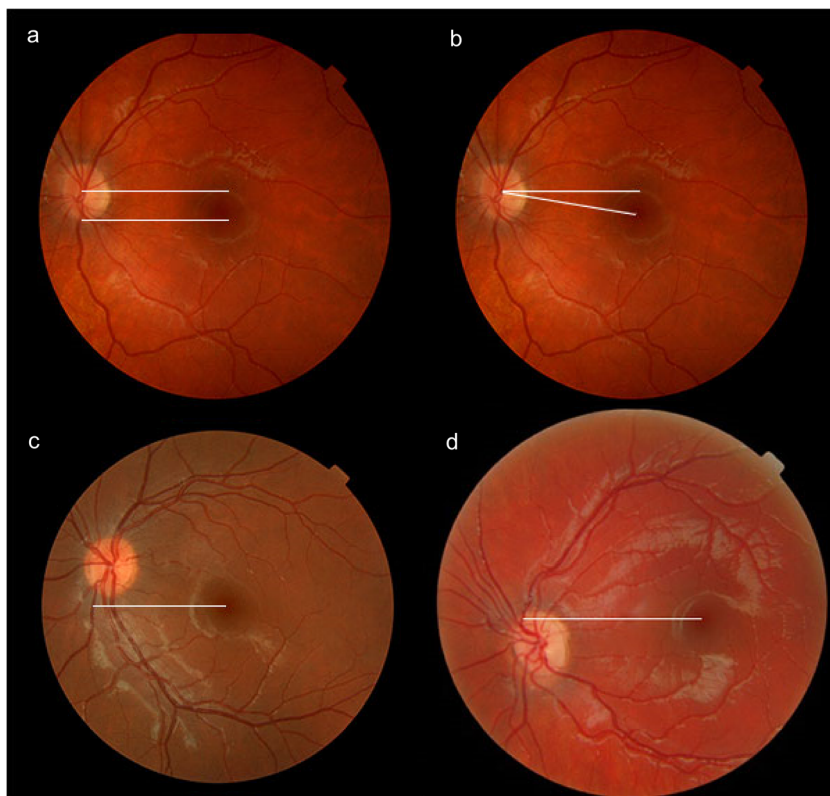
11.10 to 23.20°). Nine patients (13%) had intorsion with a disc-foveal angle of − 1.99° (± 2.52), though this was also highly variable (range − 2.50 to − 9.40°).

Preoperatively, mean distance esodeviation was 30.34 prism dioptres (± 13.22) and mean near esodeviation was 41.37 prism dioptres (± 12.45). After surgery, we found a significant decrease in distance esodeviation of 19.43 prism dioptres (95% CI [16.50–22.00], $p < 0.0001$). Near esodeviation was also significantly decreased by 21 prism dioptres (95% CI [18.50–24.00], $p < 0.0001$).

Among the 31 patients with pathological extorsion, mean disc-foveal angle was significantly reduced by 1.80° (95% CI [− 2.76, − 0.84], $p < 0.0001$). Among the nine patients with intorsion, mean disc-foveal angle was significantly increased by 2.28° (95% CI [0.16, 4.40], $p = 0.038$).

After surgery, the patient distribution according to qualitative torsion grading had changed, with more patients presenting physiological extorsion ($n = 45$, 66%) (Table 3).

Within the pathological extorsion group, regression analysis showed that the mean distance esodeviation reduction was positively correlated to the mean objective ocular torsion reduction after surgery (OR = 1.21, 95% CI [1.11, 1.31], $p < 0.0001$) (Fig. 3). Moreover, preoperative mean quantitative torsion was significantly and positively correlated to mean disc-foveal angle reduction after surgery (OR = 0.73, 95% CI [0.57, 0.93], $p = 0.010$) (Fig. 4).

Regression analyses failed to reveal any correlation between the severity of distance or near esodeviation and the amount of ocular torsion in any of the three groups studied (n.s.)

Table 1 Characterization of the three groups studied

Group	Right eye	Left eye
Normal	Physiological extorsion	Physiological extorsion
Extorsion	Pathological extorsion	Physiological extorsion
	Physiological extorsion	Pathological extorsion
	Pathological extorsion	Pathological extorsion
Intorsion	Intorsion	Physiological extorsion
	Physiological extorsion	Intorsion
	Intorsion	Intorsion

Table 2 Surgical procedures

Surgical procedure	n (%)	Mean in millimetres (sd)
Medial rectus muscle recession	53 (78)	4.24 (0.91)
Lateral rectus plication	67 (99)	5.45 (0.86)
Cüppers posterior fixation suture	25 (37)	
Plication + Cüppers	25 (37)	
Plication + Cüppers + recession	9 (13)	

Fig. 2 Surgery age distribution

Surgery age distribution
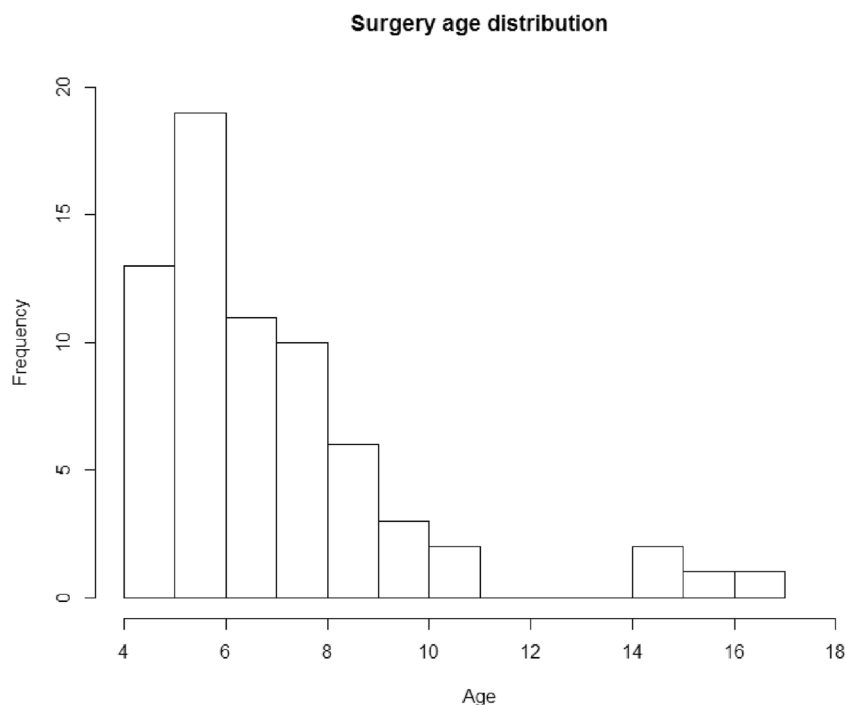
Discussion

In our study, patients with infantile esotropia, without significant vertical deviation, presented a high prevalence (59%) of pathological objective ocular torsion. Thirty-one patients (46%) presented pathological extorsion, whereas nine patients (13%) presented intorsion. This study suggests that pathological torsion might be frequently found in infantile esotropia even in the absence of significant vertical dissociation.

For the physiological torsion group ($n = 28$, 41%), the mean disc-foveal angle was + 6.7°. This result is similar to other previous studies in normal individuals without strabismus [2, 12].

Considering the pathological extorsion condition, distance esodeviation decrease after surgery was correlated to decrease in ocular torsion. We believe that medial rectus muscle overaction in infantile esotropia with significant horizontal deviation, i.e. mean distance and near esodeviations

were 30.34 and 41.37 prism dioptres respectively, could make the eye twist within the orbit leading to pathologic torsion. Ocular torsion could also be the consequence of abnormal pulleys or muscle displacement. This peripheral theory is supported by MRI studies [13, 14]. On the other side, the central hypothesis is supported by Graf's findings. Graf et al. reported that when binocular vision is artificially impaired, the ocular motor balance of the occluded eye tends to lead to an extorted position [15]. Conversely, ocular torsion might threaten fusion capacities, thus preventing development of binocular vision in childhood [16]. In a study, intorsion and extorsion have been reported being associated with oblique dysfunction in 78.7 and 74.4% of the participants with an increase in patients without stereopsis [17]. Interestingly, more than 20% of subjects present intorsion and extorsion without oblique dysfunction according to this study. This result suggests that torsion might not only be the expression of oblique muscles action as, in our study, weakening medial rectus with muscle recession tends to reduce ocular torsion. In children with infantile esotropia, extorsion could be part of a vicious circle, impairing binocular visual function, but also increasing with loss of fusion capacities. Unexpectedly, intorsion was found in 13% of the participants without significant vertical disorder in primary position or in lateral gaze and without significant oblique dysfunction at motility examination. Similarly to the peripheral hypothesis for extorsion, the presence of abnormal muscle pulleys or muscle displacement could partly explain intorsion and its reduction after horizontal rectus surgery [13, 14].

Table 3 Qualitative and quantitative torsion characteristics of the patients

Group before surgery	Before surgery, n (%)	After surgery, n (%)	Mean angle deviation before surgery, in degrees (sd)	Mean angle deviation after surgery, in degrees (sd)	p
Normal	28 (41)	45 (66)	6.73 (2.66)	6.73 (2.85)	0.501
Extorsion	31 (46)	17 (25)	12.94 (3.67)	11.14 (3.42)	< 0.001
Intorsion	9 (13)	6 (9)	− 1.99 (2.52)	0.29 (3.30)	0.038

Fig. 3 Correlation between mean distance esodeviation reduction after surgery and mean objective ocular torsion reduction after surgery in the pathological extorsion condition
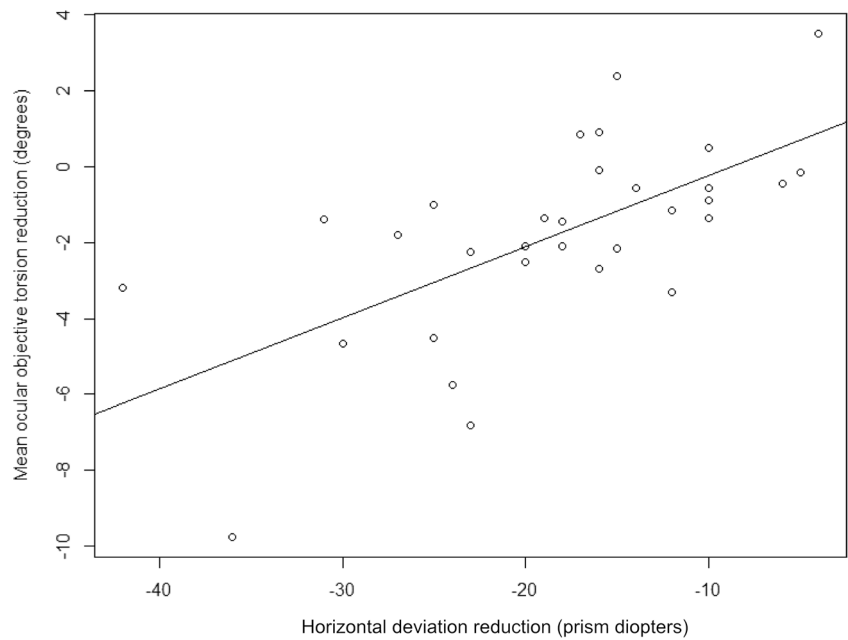
Ocular torsion is generally assessed in cyclovertical strabismus. Nevertheless, our study provides information about the status of ocular torsion, both quantitatively and qualitatively, in patients with infantile esotropia.

It is interesting to note that pathological ocular torsion can be found on the fixing eye. Over time, prolonged contraction of the oblique muscles could lead to their shortening. Thus, their arc of contact decreases, potentially producing ocular torsion in the primary position.

Several limitations to our study must be acknowledged. First, we could not compare the current findings to those in a population of patients with infantile esotropia who have not undergone horizontal surgery. The main limitation of our study is that the effect of ocular growth on objective ocular torsion is unknown. Investigating changes in ocular torsion after surgery in comparison to a control group matched on age would give more information about growth effect on children with infantile esotropia. However, Lefèvre et al.

Fig. 4 Correlation between mean objective ocular torsion reduction after surgery and mean disc-foveal angle before surgery in the pathological extorsion condition
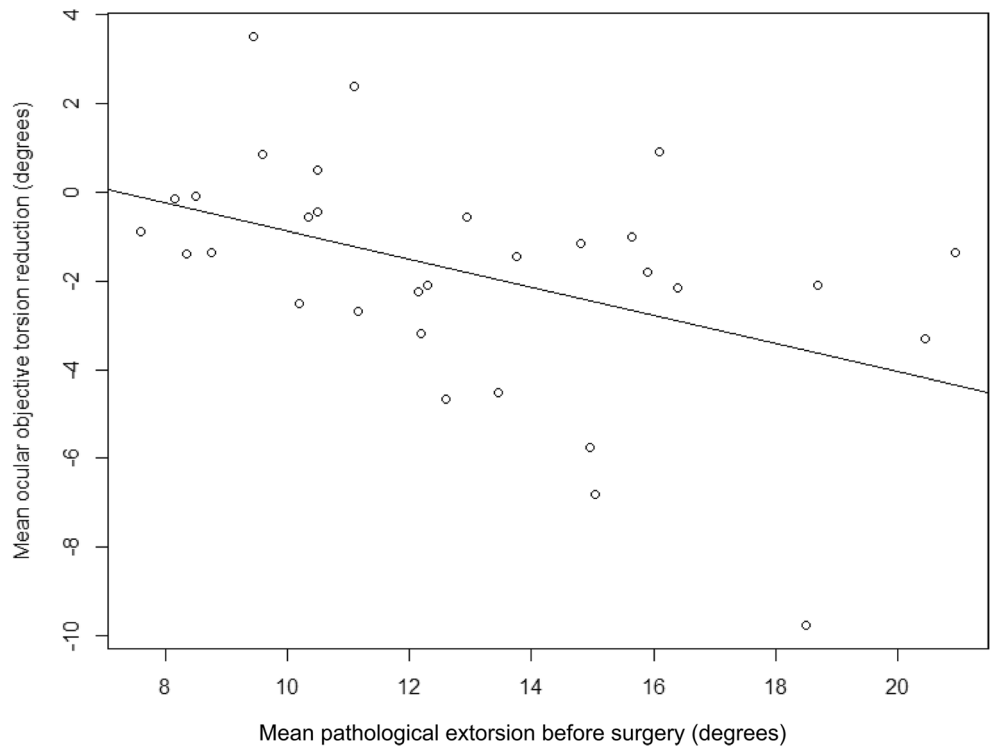
conducted a study in healthy patients (children and adults) and did not report significant change in objective ocular torsion over time (mean duration between two measures 131 days, range 16 to 201 days) [2]. Ocular torsion evaluation on fundus photography, even when performed in a rigorous manner, could be modified by head position. We limited this measurement bias by carefully checking head position and by avoiding any head tilt. In light of these limitations, fundus photography allows the evaluation of ocular torsion under monocular vision conditions. Ocular torsion could be different in the fixing or non-fixing eye, but could also be affected by monocular or binocular vision condition.

In terms of practical implications, pathologic torsion might not always be the expression of oblique dysfunction and should make clinicians consider with caution oblique muscle weakening or strengthening in children with large-angle infantile esotropia. For instance, we suggest that the frequently found pathologic torsion in infantile esotropia associated with alphabetic patterns or congenital fourth nerve palsy could be partly related to binocular vision impairment and medial rectus muscle overaction. Objective ocular torsion has also been assessed as a predictive factor of occurrence of subsequent vertical disorders such as vertical dissociated divergence or inferior oblique muscle overaction in patients with esotropia [10]. Furthermore, objective ocular torsion assessment could be a predictive factor of developing subsequent vertical disorders that should be assessed in future studies.

In conclusion, our study suggests a high prevalence of pathological objective torsion, mostly pathological extorsion, in patients with infantile esotropia without significant vertical deviations. Intorsion and extorsion tend to be normalized by unilateral horizontal rectus surgery. Furthermore, the severity of extorsion is correlated to its decrease after surgery. For patients with infantile esotropia associated with pathological torsion, unilateral horizontal rectus surgery seems to be an effective procedure. However, further investigations comparing unilateral plication-recession surgery to other procedures, e.g. unilateral recession-resection or bimedial recession, should be considered.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. For this type of study, formal consent is not required.

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