LETTER TO THE EDITOR

Cyclic esotropia and impaired vision after strabismus surgery

Ésotropie cyclique et dysfonction visuelle après une chirurgie du strabisme

Introduction

Cyclic esotropia is a rare condition in which 24–48 hours periods of squinting alternate with periods of orthotropia and normal binocular vision [1]. The condition affects around one in every 3000–5000 patients with strabismus. Cyclic esotropia usually presents in children and most reports describe an onset age of 2–6 years [2]. Adult-onset cyclic strabismus has been linked to numerous factors though most adult cases have been attributed to eye surgery or traumatic brain injury [3–5]. Although its pathophysiology remains unclear, some authors have suggested the dysregulation of central nervous system pathways involved in the biological clock affecting oculomotor control such as those of the hypothalamus or superior colliculus [6]. Esotropia is the most common ocular deviation, but it has also been reported esotropia, “V” patterns and isolated vertical alteration [7].

We present a peculiar case of cyclic esotropia detected after strabismus surgery, in which 36 hours cycles of mild esotropia coincided with impaired vision and with observed reactivated myopic choroid neovascularization.

Case report

A 61-year-old woman with degenerative myopia was referred to our department because of progressive diplopia and esotropia in the right eye (RE). Manifest refraction was −30.50, −0.75, 75° in the RE and −29.50, −0.50, 105° in the left eye (LE). Upon examination, best-corrected visual acuity (VA) was 20/40 RE and 20/32 LE. There was no afferent pupil defect. Slit-lamp biomicroscopy revealed a normal anterior segment and anterior vitreous. Intraocular pressure was normal. No choroid neovascularization was observed in a fundus examination (FE). Motility assessment revealed a right esotropia of 35 PD, limited abduction and elevation in both eyes, and a mild “V” pattern (Fig. 1). Forced duction test was positive in both eyes. Ice pack test and serum antibodies against the acetylcholine receptor were negative. A local lesion was ruled out in a CT scan and MRI. The patient was diagnosed with highly restrictive myopic esotropia and scheduled for strabismus surgery. Following surgery in the right eye involving a 10 mm medial rectus recession, 6 mm lateral rectus resection and superior replacement, she remained orthotrophic yet 5 days later, she complained of regular 36-hours cycles of convergent squint and diplopia accompanied by reduced and distorted vision in the RE.

During the “straight” interval (36 hours), she was free of diplopia and strabismus. In this period, tests indicated a small esophoria (4 PD), image fusion (4 dot test) and good stereoacuity (TNO test). Visual acuity (VA) was 20/63 in the RE and 20/50 in the LE.

During the “squinting” cycle (36 hours), esotropia was around 6 PD though until 10 PD was recorded in one visit, with 4 PD of right hypotropia. There was a right eye suppression and absent stereopsis in TNO test (480 seconds of arc). VA was usually between 20/200 and 20/100 in the RE and 20/50 in the LE (Fig. 2). Between visits, esotropia was fairly stable. Between visits, esotropia was fairly stable.

A FE and macular optic coherence tomography (OCT) revealed the presence of myopic choroid neovascularization, which was treated with ranibizumab (Fig. 3). In addition, 3 mg per day of melatonin was prescribed for two months. Cycles initially lengthened to 48 hours though after 4 months, they returned to 36 hours and melatonin was withdrawn.

Presently, right esotropia during strabismus episodes (every 36 hours) is fairly stable while the patient shows good fusion capacity, some stereoscopic vision and unmodified VA during straight periods. In 10 of 15 visits in periods of impaired vision and strabismus, reactivated choroid lesion was observed and OCT confirmed. The patient uses prism lenses during strabismus episodes.

Discussion

Late onset cyclic esotropia is extremely rare. The main mechanism proposed so far is innate biological clock disruption caused by a central nervous system trigger [3]. The trigger events described have been traumatic brain injury, strabismus surgery, extracapsular cataract surgery, retinal detachment with scleral buckling, orbital surgery and ocular trauma [4,8]. However, many cases have been associated with other eye diseases such as binocular optic atrophy or even with the menstrual cycle [2,9]. Only a few cases of adult cyclic esotropia and high myopia have been described [4,5,8]. Bagheri et al. reported the case of a myopic woman with bilateral retinal detachment who developed cyclic strabismus after retinal surgery and Troost et al., that of a woman who started with cyclic esotropia and hyperopia after unilateral scleral buckling surgery [3,8]. Our case is consistent with Troost’s hypothesis of abnormal sensory
Restrictive myopic esotropia with a mild "V pattern" observed in the patient before strabismus surgery.

"straight" day with no diplopia or strabismus. Right: "squinting" day with 6 PD esotropia and 4 PD right hypotropia.

input triggering an indefinite central adaptation in the ocular motor control system. The sensory input defect in our case would be myopic choroid neovascular lesion instead of retinal detachment.

Controversy exists over whether good prior binocular function precedes cyclic strabismus. Our patient showed good stereoscopic vision and fusion capacity on days when her vision was not impaired. Reactivation of myopic lesions leading to a worsened VA, especially in the right eye, may have limited her central fusion capacity. However, some authors disagree with this theory [2,5].

The 36 hours’ duration of esotropia and persistence of cycles for 6 years (so far) are striking. In our patient, upon melatonin treatment, cycles diminished to 48h yet recovered when treatment was suspended. Metz and Bigelow described a patient with cyclic esotropia in whom the cycle rhythm varied when traveling across six different time zones [1]. Both these findings are consistent with the theory of a disrupted biological clock. Some authors have noted that cycles tend to last a few months and then evolve into irregular ones until the deviation becomes constant after 4–12 months [3,6]. Accordingly, it has been suggested that these patients are basically strabismic with cycles of orthophoric vision rather than the other way round [7]. There have also been reports of cyclic esotropia lasting over one year or even longer than 7 years [8,10].

The surgical treatment of cyclic esotropia has proved successful. Surgery usually targets maximal deviation on the worse day [8]. In our patient, the deviation was small and rather than undergoing further strabismus surgery, she opted for the intermittent use of prism lenses.

In conclusion, we present the first case of late onset cyclic esotropia related to cyclic vision loss due to the reactivation of myopic choroidal neovascularization. We propose that cyclic esotropia may have been triggered by biological clock disruption due to strabismus surgery or to abnormal sensory input due to the recurrent activity of the patients’ myopic lesions.
Figure 3. Fundus exam and optical coherence tomography: myopic choroid neovascularization in both eyes.

Disclosure of interest

The authors declare that they have no competing interest. No financial support was received for this submission.

References


F. Pérez Bartolomé*, R. Gómez de Liaño, L. Morales Fernández, J. García Feijoo
Department of Ophthalmology, Hospital Clínico San Carlos, Complutense University of Madrid, Prof Martín Lagos Avenue, S/N 28040 Madrid, Espagne
* Corresponding author.
E-mail address: Franciscoperezbartolome@gmail.com
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