

Case Report

Cyclic Esotropia: a Case Report

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Article Notes:

Received: Aug. 6, 2017

Received in revised form:
Sep. 3, 2017

Accepted: Jan. 1, 2018

Available Online: Apr. 4, 2018

Keywords:

Cyclic

Esotropia

Treatment

Abstract

In this case report, two cases with diagnosis of cyclic esotropia are presented. Both patients complained of periodic alternative inside eye deviation with an interval of one day. Paraclinical tests including magnetic resonance imaging (MRI) of the brain and electroretinogram (ERG) were normal and no neurological abnormality or paralysis was detected. Patients were observed in two consecutive days to confirm the diagnosis of cyclic esotropia. Both patients underwent bilateral medial rectus recession surgery as routinely performed for non-cyclic esotropic patients, and the postoperative orthotropia with the best corrected visual acuity (BCVA) of 20/20 was obtained in both cases. In conclusion surgical and non-surgical treatments can be used for cyclic esotropia similar to what is routinely performed for non-cyclic strabismic patients. Successful therapeutic outcomes might be obtained in such patients achieving normal binocular fusion in several days with no ocular deviation.

How to cite this article: Rajavi Z, Sabbaghi H. Cyclic Esotropia: a Case Report . Journal of Ophthalmic and Optometric Sciences . 2018;2(2):25-9.



Introduction

Cyclic strabismus is a rare type of ocular deviation with an incidence rate of one patient in each 3000 to 5000 individuals with strabismus; and mostly appears as esotropia^{1,2}. It was first described by Costenbader and Mousel in 1964^{3,4}. These patients usually complain of periodic ocular deviation manifested with particular time intervals of one or several days (usually 48 hours)⁵. Commonly, cyclic esotropia manifests among children with average age of 3 to 4 years and with a large angle of deviation (30 to 40pd). Amblyopia and high accommodative convergence to accommodation ratio (AC/A ratio) may be present as complementary findings. Sometimes different severities of V-pattern may also be observed in these patients. No causative neurological or muscular disorders have been reported for cyclic esotropia. These patients would usually obtain a well-controlled binocular fusion following either surgical or non-surgical treatments. Medial rectus recession and unilateral recession-resection (R & R) methods are reported as the most common surgical techniques to treat patients with cyclic esotropia^{5,6}. In this manuscript, we report two cases with diagnosis of cyclic esotropia who referred to

our clinic complaining of periodic inside eye deviation.

Case Report

We present two patients with cyclic esotropia referred to the pediatric clinic of Torfeh Eye Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran. This case report was approved by the ethics committee of Shahid Beheshti University of Medical Sciences and patients' parents gave their consent before reporting the cases.

CASE I

A 3 year old girl with a history of strabismus since age of 1.5 years was referred to our clinic. Her parents reported a periodic manifestation of alternative esotropia at both eyes with an interval of one day. A primary diagnosis of lateral rectus paralysis was rolled out and her neurological examination using magnetic resonance imaging (MRI) of the brain and electroretinogram (ERG) were normal. She was not cooperative to perform visual acuity test. Cycloplegic refraction of +2.50D was found after 30 minutes installation of tropicamide 1 % and cyclopentolate 1 % eye drops. Patient was observed in two consecutive days to confirm the diagnosis of cyclic esotropia. We did not find any

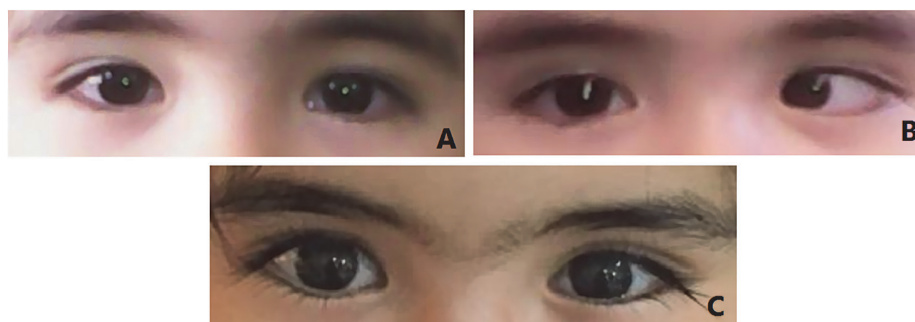


Figure 1: Patient appearance without (A) and with (B) strabismus manifestation at two consecutive days before the surgery, while no strabismus was observed postoperatively (C)

strabismus using alternative prism cover test at both far and near distances at the first day, while, esotropia of 40 pd was measured at both far and near distances in the second day (Figure 1). Based on the ocular motility an overaction of +2 was observed for the medial rectus and superior oblique muscles of both eyes. However, no lateral rectus underaction was observed. The patient underwent bilateral medial rectus recession surgery of 5 mm two months after confirmation of cyclic esotropia diagnosis. No strabismus was observed at the postoperative follow up of four years with BCVA of 10/10 in both eyes.

CASE II

A 6 year old girl with gestational age of 36 weeks, birth weight of 2400 grams and a one day history of hospitalization in neonatal intensive care unit (NICU) was referred to our clinic. Her chief complain was periodic manifestation of inside eye deviation with an interval of one day, which alternatively appeared in her eyes (Figure 2). Her cousin also suffered from strabismus.

Her presenting visual acuity (VA) was 10/10 and 8/10, which improved to 10/10 after prescription of the cycloplegic refraction of + 4.00 - 0.50 × 170 and + 5.75 - 0.50 × 10 for the right and the left eye, respectively. We

prescribed a pair of bifocal glasses with the near addition of + 2.50 D spheres. We confirmed the manifestation of cyclic esotropia in two consecutive days. Esotropia of 40 pd at far (6 m) and near (33 cm) using alternative prism cover test at the first day of examination was observed, while no deviation was observed at the second day of examination. We also found inferior oblique overaction of + 3 based on the ocular motility test with V-pattern. She underwent bilateral medial rectus recession surgery of 6 mm and bilateral inferior oblique myectomy of 5 mm. Postoperative BCVA was 10/10 with no strabismus manifestation at both far and near distances at all follow up examinations performed up to three years after the surgery.

Discussion

In the present case report, ophthalmic manifestations and therapeutic management of two patients with cyclic esotropia was presented. Although this type of deviation is very rare, clinical management is highly important.

The mechanism of cyclic esotropia is not well known. Some possible causes include the gradual esotropia manifestation due to non-compensatory component, incomplete cerebral dominancy, and strabismus following

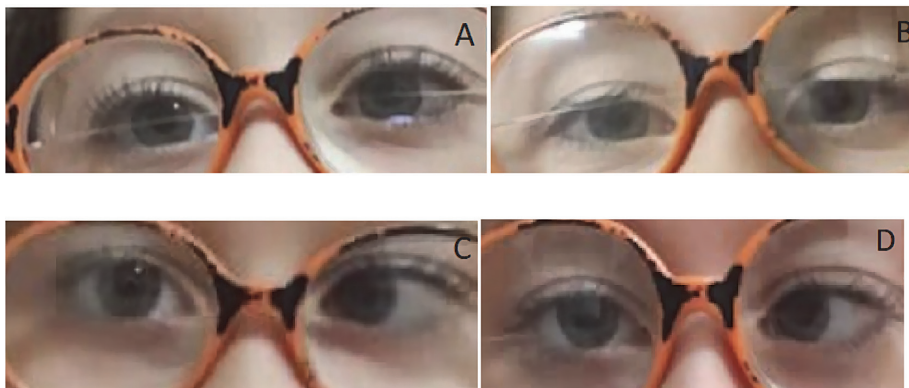


Figure 2: No strabismus was observed at far (D) and near (C), while esotropia manifested at far (B) and near (A) in the next day before the surgery

patch therapy⁶. Since central nervous system dysfunction has been reported by some studies, complete neurological imaging is recommended to rule out possible neurological dysfunctions^{7,8}. Consecutive patient observation based on strabismus manifestation interval reported by parents or care givers is also required to reach the definite diagnosis of cyclic deviation. Furthermore, childhood photos can be helpful in achieving the final confirmation.

Clinical management of cyclic esotropia is the same as simple esodeviation with no periodic manifestation. In this regard, providing BCVA is the preliminary step to obtain balancing between the accommodation and vergence systems. It is helpful in better controlling the binocular fusion and deviation reduction particularly in accommodative esotropia like our second reported case⁸.

There was a significant difference between the angles of deviation between far and near distances (high AC/A ratio) in one of our cases. This finding has been reported as an associated findings in cyclic esotropia and is corrected by prescription of bifocal glasses and surgery^{5,9}. Inferior oblique overaction and consequent V-pattern are some rare associated findings in cyclic esotropia, which were also observed in our second reported case and corrected significantly after inferior oblique myectomy

was performed^{5,10}.

As reported, we obtained high success after the surgery to achieve binocular fusion, which is line with a report by Souza-Dias et al.¹¹. However Riordan-Eva et al.,¹² reported that no binocular fusion was achieved in their patients due to lack of normal binocularity and monocular visual loss, and concluded that the level of binocularity in cyclic esotropic cases is correlated with the ocular alignment of the good eye. Interestingly Ma et al.,⁴ have suggested that sharp vision and good binocular fusion are not essential factors in achieving successful surgical outcomes in cyclic esotropic patients.

Conclusion

Surgical and non-surgical treatments can be used for cyclic esotropia similar to what is routinely performed for non-cyclic strabismic patients. Successful therapeutic outcomes might be obtained in such patients achieving normal binocular fusion in several days with no ocular deviation.

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Footnotes and Financial Disclosures

Conflict of Interest:

The authors have no conflict of interest with the subject matter of this manuscript.

