Original Article

Clinical Characteristics and Analysis of Spontaneous Consecutive Exotropia in Children with Refractive Accommodative Esotropia

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Abstract

Objectives: To assess the clinical characteristics and risk factors associated with spontaneous consecutive exotropia (ScXT) in children diagnosed with refractive accommodative esotropia (RAET).

Materials and Methods: A retrospective analysis of medical records was conducted on 19 patients who demonstrated a spontaneous transition from RAET to exotropia (XT). Patients who received strabismus surgery or botulinum toxin injection were excluded from the study. The control group consisted of 31 age-matched patients with RAET who demonstrated successful optical alignment at both near and distance. The ophthalmological examination findings of the study and control groups were compared. Independent two-sample t-test and Pearson's chi-square test were used to evaluate the data of the patients.

Results: The study examined patients diagnosed with RAET who developed consecutive XT. Among them, 15 (78.9%) were female and 4 (21.1%) were male. The mean age at esotropia (ET) onset was 22.68 months (standard deviation [SD]: 12.91). The control group consisted of 16 (51.6%) female and 15 (48.4%) male patients, with a mean age at ET onset of 25.09 months (SD: 15.47). Mean age at onset did not differ between the groups (p=0.55). The mean interval between ET onset and appearance of XT was 7.53 years (SD: 1.49). Cycloplegic refraction measurements taken during the initial examination indicated that the study group exhibited greater degrees of hypermetropia in their right (p=0.01) and left (p=0.04) eyes than did the control group. Furthermore, the incidence of inferior oblique muscle overaction was higher among the study group (p=0.03).

Conclusion: The findings indicate that patients with high hypermetropic refraction values should be monitored for an extended period due to the increased risk of developing subsequent XT. Concomitant inferior oblique overaction can increase the risk of ScXT.

Keywords: Spontaneous consecutive exotropia, hypermetropia, refractive accommodative esotropia, inferior oblique overaction

Introduction

Refractive accommodative esotropia (RAET) is a condition characterized by convergent strabismus that occurs due to accommodative convergence (AC) to hypermetropia. It is important to note that the AC/accommodation (A) ratio is normal. The mainstay of treatment in patients with RAET is the full correction of hyperopia with spectacles. Treatment should also be initiated if patients have amblyopia.[1,2,3]

The development of spontaneous consecutive exotropia (ScXT) differs from consecutive exotropia (XT) that occurs after strabismus surgery for esotropia (ET). The onset is gradual, and diplopia is not typically present. ScXT has been observed in 5% to 15% of patients with RAET following weeks to a mean of 5.5 years of full hyperopic correction.[1,4,5] High hyperopia of +5 diopter (D) or more, early onset of ET, initial amblyopia, weak or absent binocular single vision, a decrease in the AC/A ratio, and fusional vergence dysfunction have been suggested as triggers of ScXT.[6,7,8,9,10] A review of the literature reveals no reports on the association between inferior oblique muscle overaction (IOOA) and consecutive XT.

The purpose of this retrospective study was to analyze the clinical features of patients with RAET who developed ScXT.

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Materials and Methods

This study adhered to the tenets of the Declaration of Helsinki and was approved by the Institutional Review Board of Başkent University (project no: KA21/538, date: 04.01.2022). As it was a retrospective study, there was no need to obtain informed consent from the subjects. The records of patients with RAET corrected with spectacles were retrospectively reviewed, and those who developed ScXT were included in the study group. Patients with previous ocular surgery, botulinum toxin injection, and history of any systemic (neurological impairment or developmental delay) and ocular disease were excluded from the study. Age-matched patients with RAET who had maintained successful near and distance ocular alignment were included as the control group.

All patients underwent visual acuity testing, assessment of duction and versions, cycloplegic refraction, and anterior and posterior segment examination during follow-up.

Cycloplegic refraction was performed after two instillations of 1% cyclopentolate (Sikloplejin, Alcon Laboratories, Inc., Fort Worth, TX, USA) and 1% tropicamide (Tropamid, Bilim Pharmaceuticals, İstanbul, Türkiye) combination eye drops at 5-minute intervals. After maximum cycloplegia, manifest refraction was performed by retinoscopy and full hypermetropic correction was prescribed. The spherical equivalent (SE) refractive error was calculated as the sum of the SE plus half of the cylindrical power.

Anisometropia was defined as a difference of 1.5 D or more in SE refractive error between the eyes. Amblyopia was diagnosed when visual acuity was reduced by two lines or more in Snellen acuity or when central, steady, and maintained fixation was absent. Amblyopia was treated with occlusion therapy.

Ocular alignment was assessed by the cover and uncover test. The alternate prism cover test or Krimsky test was used to measure the angle of deviation in the primary position at near and distance fixation. Fusion was tested with the Worth four-dot test, and the Titmus fly stereo test (Stereo Optical Co., Inc., Chicago, IL, USA) was used to assess stereoacuity when cooperation was adequate.

Statistical Analysis

Patient data are expressed as number and percentage or as mean, standard deviation (SD), and range. Comparisons between the groups were made using independent two-sample t-test and Pearson's chi-square test. SPSS Statistics version 21 (IBM Corp., Armonk, NY, USA) was used. A p value less than 0.05 was considered statistically significant.

Results

Of the 19 patients with RAET who developed ScXT evaluated in this study, 15 (78.9%) were female and 4 (21.1%) were male. The control group consisted of 31 RAET patients without ScXT, 16 (51.6%) of whom were female and 15 (48.4%) were male. The demographic information and clinical characteristics of the groups are shown in Tables 1 and 2, respectively.

The mean age at onset of ET was 22.68 months (SD: 12.91, range 6-48 months) in the study group and 25.09 months (SD: 15.47, range 6-60 months) in the control group (p=0.55). The mean age at first eye examination in these groups was 31.89 months (SD: 14.42, range 12-60 months) and 34.25 months (SD: 14.79, range 12-72 months), respectively (p=0.45).

In the study group, the mean age at ScXT diagnosis was 9.42 years (SD: 3.41, range 4-13 years). The mean interval between ET onset and appearance of XT was 7.53 years (SD: 1.49 years).

The mean initial cycloplegic SE refractive error values in the study group were +5.72 D (SD: 1.85, range 3.25-8.00 D) for the right eye and +5.50 D (SD: 1.70, range 3.25-9.00 D) for the left eye. In the control group, these values were +4.41 D (SD: 1.78, range 2.75-8.75) for the right eye and +4.50 D (SD: 1.89, range 2.00-9.00) for the left eye. Initial cycloplegic refraction values showed that the study group had higher hyperopic values in the right (p=0.01) and left (p=0.04) eyes compared to the control group.

IOOA was present in 4 patients in the study group and 2 patients in the control group (p=0.03).

At initial presentation, 12 of 19 patients in the study group and 24 of 31 patients in the control group had amblyopia (p=0.36). All patients were treated with occlusion therapy. Anisometropia was detected at baseline in 6 patients in the ScXT group and 12 patients in the control group (p=0.61).

For near vision, the mean angle of esodeviation without spectacle correction was 32.69 prism diopters (PD) (SD: 9.50, range 16-40 PD) in the study group and 33.90 PD (SD: 13.65, range 16-80 PD) in the control group (p=0.34). With spectacle correction, the mean angle of esodeviation for near was 7.42 PD (SD: 0.97, range 4-8 PD) in the study group and 7.09 PD (SD: 1.77, range 0-8 PD) in the control group (p=0.24).

For distance, the mean angle of esodeviation without spectacle correction was 26.11 PD (SD: 7.80, range 14-40 PD) in the study group and 29.48 PD (SD: 13.92, range 10-75 PD) in the control group (p=0.96). With spectacle correction, the mean angle of esodeviation for distance was 4.57 PD (SD: 2.50, range 0-8 PD) in the study group and 4.83 PD (SD: 1.98, range 0-8 PD) in the control group (p=0.65).

We were able to perform the fusion and fly test on initial examination in 13 children in the study group and in 19 children in the control group. When comparing the initial fusion and the fly test, no difference was observed (p=0.44 and p=0.51, respectively).

ScXT was treated with reduced hypermetropia in 18 patients to stimulate AC. Despite undercorrection, one patient deteriorated to large angle XT and underwent surgery.

Mean follow-up was 74.10 months (SD: 46.74, range 23-204 months) in the ScXT group and 70.67 months (SD: 41.05, range 15-142 months) in the control group (p=0.93).

Table 1. Demographic data of the study and control groups			
	Study group (n=19)	**Control group (n=31)**	**p**
Sex			
Female	15 (78.9)	16 (51.6)	
Male	4 (21.1)	15 (48.4)	
Family history			
Strabismus	2 (10.5)	6 (19.4)	0.51
Amblyopia	2 (10.5)	6 (19.4)	0.51
Age at esotropia onset (months)	22.68±12.91 (6-48)	25.09±15.47 (6-60)	0.55
Age at initial visit (months)	31.89±14.42 (12-60)	34.25±14.79 (12-72)	0.45
Follow-up period (months)	74.10±46.74 (23-204)	70.67±41.05 (15-142)	0.93
Data are presented as number and percentage (Pearson's chi-square test) or mean ± standard deviation (range) (independent two-sample t-test)			

Table 2. Clinical features of the study and control groups			
	Study group (n=19)	**Control group (n=31)**	**p**
Cycloplegic refraction at initial visit (SE, D)			
Right eye	5.72±1.85 (3.25-8.00)	4.41±1.78 (2.75-8.75)	**0.01**
Left eye	5.50±1.70 (3.25-9.00)	4.50±1.89 (2.00-9.00)	**0.04**
Cycloplegic refraction at last visit (SE, D)			
Right eye	4.93±1.78 (1.50-7.50)	4.89±1.81 (2.25-10.00)	0.94
Left eye	4.88±1.81 (1.00-7.00)	4.92±2.10 (2.25-10.50)	0.93
Anisometropia	6 (31.6)	12 (38.7)	0.61
Amblyopia	12 (63.2)	24 (77.4)	0.36
Ocular patch treatment	12 (63.2)	24 (77.4)	0.36
Stereopsis*	2/13 (15.4)	6/19 (31.6)	0.51
Worth four-dot test fusion*	2/13 (15.4)	7/19 (36.8)	**0.44**
Inferior oblique overaction	4 (21.1)	2 (6.5)	**0.03**
Dissociated vertical deviation	0 (0)	0 (0)	
Angle of esodeviation at initial visit (without spectacles), (PD)			
Distance	26.11±7.80 (14-40)	29.48±13.92 (10-75)	0.96
Near	32.69±9.50 (16-40)	33.90±13.65 (16-80)	0.34
Angle of esodeviation at initial visit (with glasses), (PD)			
Distance	4.57±2.50 (0-8)	4.83±1.98 (0-8)	0.65
Near	7.42±0.97 (4-8)	7.09±1.77 (0-8)	0.24
Data are presented as number and percentage (Pearson's chi-square test) or mean ± standard deviation (range) (independent two-sample t-test). *Percentages given for the subsets of patients that could be tested, SE: Spherical equivalent, D: Diopter, PD: Prism diopter			

Discussion

ScXT is not uncommon in patients with RAET after correction of hyperopia without botulinum toxin injection or strabismus surgery.[9] Possible risk factors that may trigger the development of ScXT are high hypermetropic refraction, anisometropia, amblyopia, early onset of ET, vertical incomitance, lack of binocular single vision, decreased AC/A ratio, and fusional vergence abnormalities.[6,7,8,9]

Many previous studies have reported that high hyperopia of +5 D or more is considered to be the main cause of ScXT in RAET patients.[6,8-11] In the present study, highly hypermetropic refraction was found to be significantly more prevalent in the ScXT group than in the control group at the initial visit. Our patients in the study group had more than +5 D of hyperopia in each eye.

Senior et al.[9] determined that ET onset occurred before the age of 2 years in patients with ScXT. Contrary to their results, Watanabe-Numata et al.[4] reported that the age of correction was not a risk factor for ScXT. The age at onset of ET in the ScXT group in this study was similar to that in the study by Senior et al.[9] In our study, the age at onset in the study group was 22.7 months. There was no difference in onset age when compared to the control group. However, onset before the age of 2 years with more than +5 D of hyperopia might increase the chance of conversion to ScXT. In addition, early-onset ET without the protection of binocular single vision may contribute to conversion to ScXT.

Watanabe-Numata et al.[4] reported that the prevalence of amblyopia at the first visit was 89% in the ScXT group. Swan[5] reported that all RAET patients with monocular amblyopia progressed to ScXT. However, some studies found that amblyopia was not an important factor in the development of ScXT.[6,10,12] Similarly, we found that there was no statistical difference between the ScXT and control groups in terms of amblyopia.

Ciner and Herzberg[13] reported that ScXT can develop in childhood or much later in adulthood as the amplitude of A decreases. In the current study, the mean age at onset of ScXT was 9.42 years. This is consistent with the study by Moore[12], who reported a mean age at onset of 9 years.

It is difficult to estimate the exact time of onset of consecutive XT, as this information was not well recalled by patients or their relatives, so subjective descriptions of the timing of ScXT should be treated with some skepticism.[14] Beneish et al.[6] reported that the mean interval between the first ET and the appearance of ScXT was 20 months. Berk et al.[1] reported that ScXT developed an average of 5.5 years after hyperopic correction. Mohan and Sharma[15] found that patients with RAET treated with hyperopic correction alone developed ScXT at a mean follow-up of 6.70 years. In the current study, the mean time from initial diagnosis to development of ScXT was 7.53 years. The mean development time of consecutive XT was found to exceed the times reported in the literature. Therefore, long-term follow-up is necessary even if RAET is well corrected with hyperopic spectacles.

Some studies have evaluated vertical incomitance such as dissociated vertical deviation (DVD) and IOOA as risk factors for consecutive XT after ET surgery.[10,16] Patients with vertical incomitance are reported to have a higher chance of fusional vergence abnormalities leading to consecutive XT. The results of our study indicate that the prevalence of IOOA was significantly higher in patients with ScXT compared to the control group. This study is the first to document an association between IOOA and consecutive XT. Shin et al.[10] found that the presence of DVD was higher in their ScXT patients than in the control group. We did not detect DVD in either the study or control groups in our study, which we attribute to the relatively small number of cases.

Weir et al.[17] reported that the development of ScXT is not precluded by the presence of some level of binocular vision. Shin et al.[10] found no statistically significant results when comparing sensory fusion using the Worth four-dot test between the ScXT group and the control group. In the present study, we did not observe any significant between-group differences in sensory fusion using the Worth four-dot test or stereopsis using the stereo fly test.

The management of ScXT in RAET patients can be both conservative or surgical. Six out of 9 patients in the study by Watanabe-Numata et al.[4] underwent strabismus surgery. Berk et al.[1] observed that 62% of ScXT patients exhibited complete resolution upon hyperopia reduction while 12.5% had to resort to surgery. According to the findings of Beneish et al.[6], early recognition and hyperopia reduction by 50-60% could improve ScXT in patients with RAET. In the present study, only one patient (5.3%) underwent strabismus surgery, while in the other 18 patients (94.7%), ScXT improved after reducing hyperopia correction.

Study Limitations

The limitations of the study are that it was not carried out in a large population and that it was a retrospective study.

Conclusion

The present study indicates that high hyperopia and IOOA are risk factors for the development of ScXT. Further prospective studies with larger populations and long-term follow-up periods are necessary to determine if the degree of hyperopia and vertical incomitance are significantly associated with ScXT development.

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Ethics

Ethics Committee Approval: This study adhered to the tenets of the Declaration of Helsinki and was approved by the Institutional Review Board of Başkent University (project no: KA21/538, date: 04.01.2022).

Informed Consent: Retrospective study.

Declarations

Authorship Contributions

Surgical and Medical Practices: A.P., N.S.Y., Concept: A.P., N.S.Y., Design: A.P., N.S.Y., Data Collection or Processing: A.P., N.S.Y., Analysis or Interpretation: A.P., Literature Search: A.P., Writing: A.P., N.S.Y.

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