Research Article / Araştırma Makalesi

The Clinical Effect of Preoperative Deviation on Surgical Success and Sensory Function in Infantile Exotropia

İnfantil Ekzotropyada Preoperatif Deviasyonun Cerrahi Başarı ve Duyusal Fonksiyon Üzerindene Etkisi

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Abstract

Background: This study aims to assess the impact of preoperative deviation on surgical success and sensory function in infantile exotropia. Furthermore, to assess the effects of late surgical timing in these patients.
Materials and Methods: This retrospective study evaluated 16 subjects with infantile exotropia that underwent surgical management between 2012 and 2018. Patients with deviation ≤ 50 prism diopters (PD) were added to Group 1, and Patients with deviation > 50 PD were added to Group 2 to assess the impact of preoperative deviation on surgical success and sensory function. The surgical success is described as ≤5 PD esotropia and ≤10 PD exotropia for patients. The Titmus stereopsis and Worth 4-Dot tests were used for sensory function assessment.
Results: The average preoperative deviation was 50,63 ± 18,7 PD (20-90). The median age at the time of surgery was 73 (24-400) months. Surgical success was observed in 75% of the patients after the first surgery, and surgical success was observed in all patients after the second surgery. The sensory function was assessed in 11 testable patients (68.7%), of which 5 (45.4%) fusion was observed in the Worth 4-Point test, and 2 (18.1%) achieved measurable stereopsis. While the preoperative deviation had an effect on fusion, it had no effect on stereopsis and surgical success(p=0,015; p=0,45; p=0,77 respectively).
Conclusions: The smaller preoperative deviation may be associated with a higher rate of sensorial fusion development. Furthermore, relatively high surgical success can be achieved with late surgical timing in these patients.

Keywords: Exotropia, Preoperative deviation, Strabismus, Stereopsis

Öz

Amaç: Bu çalışma, infantil ekzotropyada preoperatif deviasyonun cerrahi başarı ve duyusal fonksiyon üzerine etkisini değerlendirmeyi amaçlamaktadır. Ayrıca, bu hastalarda geç cerrahi zamanlamanın etkilerini değerlendirmektedir.
Materyal ve Metod: Bu retrospektif çalışmada, 2012-2018 yılları arasında infantil ekzotropyası olan ve cerrahi tedavi uygulanan 16 hasta değerlendirdi. Preoperatif kayma açısının cerrahi başarı ve duyusal fonksiyon üzerine etkisini değerlendirmek için kayma açısı ≤ 50 prizm diyoptri (PD) olan hastalar Grup 1'e, > 50 PD olan hastalar ise Grup 2'ye dahil edildi. Cerrahi başarı ≤5 PD esotropya ve ≤10 PD ekzotropya olarak tanımlandı. Duyusal fonksiyon değerlendirmesi için Titmus stereopsis ve Worth 4-Nokta testleri kullanıldı.
Bulgular: Hastaların ortalama preoperatif kayma açısı 50,63 ± 18,7 PD (20-90) idi. Ameliyat anında ortanca yaş 73 (24-400) aydı. İlk ameliyattan sonra hastaların% 75'inde cerrahi başarı sağlanırken, ikinci ameliyattan sonra tüm hastalarda cerrahi başarı gözlendi. Duyusal fonksiyon 11 test edilebilir hastada (% 68.7) değerlendirildi. Bu hastaların 5'inde (% 45.4) Worth 4- nokta testinde füzyon görüldü ve 2'sinde (% 18.1) ölçülebilir stereopsis elde edildi. Preoperatif kayma açısının füzyon üzerine etkisi saptanırken stereopsis ve cerrahi başarı üzerine etkisi olmadığı gözlendi (sırasıyla p = 0,015; p = 0,45; p = 0,77).
Sonuç: Daha düşük preoperatif kayma açısı, daha yüksek oranda duyusal füzyon gelişimi ile ilişkili olabilir. Ayrıca bu hastalarda geç cerrahi zamanlama ile nispeten yüksek cerrahi başarı elde edilebilir.

Anahtar kelimeler: Ekzotropya, Preoperatif kayma açısı, Stereopsis, Şaşılık

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Introduction

Infantile exotropia begins in the first year of life. It is associated with constant wide-angle deviation and is seen in otherwise healthy patients with no systemic and ocular disease (1, 2). It is a rare motility defect characterized by divergent misalignment, occurring in 1 in 30,000 births in the general population (2). Hiles and Biglan first used the term infantile exotropia in the early-onset exotropia case report in 1983 (3). Choi and Kim described two types of infantile exotropia; early-onset intermittent exotropia and primary infantile exotropia (2).

More than 90% of patients with infantile exotropia eventually need surgery, resulting in successful alignment and limited binocularity in most patients (4). Although many studies have studied factors associated with infantile exotropia's surgical results, there is a shortage of knowledge about preoperative deviation effects on postoperative outcomes (5-8). There is no study to assess the impact of preoperative deviation on sensory function in infantile exotropia to the best of our knowledge.

This study aimed to assess the impact of preoperative deviation on surgical success and sensory function in infantile exotropia. Furthermore, to assess the effects of late surgical timing in these patients.

Materials and Methods

The medical documents of subjects diagnosed with infantile exotropia who underwent surgical treatment between 2012 and 2018 were retrospectively investigated. Informed consent was received before surgery. Local Ethics Committee approval was obtained for the study, and it was conducted following the Helsinki Declaration principles.

Infantile exotropia was described as exodeviation observed by participants' parents or ophthalmologists in the first year of life. Patients' exodeviations were stable over three consecutive visits. Patients with neurological diseases, systemic abnormalities, genetic defects or prior strabismus surgery were excluded. All participants were referred to the relevant specialist to rule out neurological diseases that may influence the ocular alignment. Sex, age at diagnosis, age at surgery, preoperative cycloplegic refractive error, preoperative visual acuity, preoperative and final angle of strabismus, primary and secondary surgical methods, associated ocular mobility defect, final sensory function, and follow-up time data were recorded for all participants.

The angle of strabismus was measured with appropriate spectacle correction at 6 m for distance deviation and 33 cm for near deviation. While the Krimsky and Hirschberg tests were used in uncooperative participants, the alternate prism cover test was used in cooperative participants doses were planned according to the average of distance and near deviations. All patients had medial rectus muscle resection and lateral rectus muscle recession in the nondominant eye. Some had an additional inferior oblique recession to correct for inferior oblique muscle overaction. for deviation measurement. The accompanying inferior oblique muscle overaction was graded from 1 to 4. Surgical

The Worth 4-Dot and Titmus stereopsis (Stereo Optical, Chicago, IL) tests were used for sensory function assessment. Postoperative motor and sensory outcomes were evaluated at the last follow-up visit. Patients with deviation ≤ 50 PD were added to Group 1, and Patients with deviation > 50 PD were added to Group 2 to assess the impact of preoperative deviation on surgical success and sensory function. The surgical success which was assigned according to the average of distance and near deviations was described as ≤5 PD esotropia and ≤10 PD exotropia for patients.

All data were analyzed with SPSS for Windows version 18.0 (SPSS Inc, Chicago, IL). Continuous data are reported as mean ± standard deviation (SD) or median (min-max). Categorical data are reported as count (n) and percent (%). The calculated power (1-beta) based on fusion analysis is 0.789, considering type I error (alfa) of 0.05, total sample size of 11(testable patients), and effect size of 0.83. In order to investigate the differences between the two groups, the Mann– Whitney U test and the Chi-square test were used. The confidence level in the analyzes was reported as 95% and a p-value of ≤0.05 was regarded statistically significant.

Results

A total of 16 cases of infantile exotropia with at least 3 months of follow-up time were included in the study: eleven boys and five girls. Demographic features, spherical equivalent, visual acuity, and patients' follow-up time are presented in Table 1. The median age at surgery was 73 (24-400) months. The best-corrected visual acuity was 0.81 ± 0.3 and 0.76 ± 0.3 in the right and left eyes, respectively. The three (18.8%) patients had inferior oblique muscle hyperfunction (AOH), 3 (18%) had abnormal head position and 2 (12.5%) had V pattern strabismus. The mean follow-up period was 23.0 ± 16.49 months.

The average preoperative deviation was 50,63 ± 18,7 PD (20-90). Surgical success was observed in 12 patients (75%) after the first surgery, and no surgical success was observed in 4 (25%) patients. A second surgery was performed on the other eye of these 4 patients, and all had surgical success. The Worth 4-Dot and Titmus stereopsis tests were performed in 11 testable patients (68.7%), 5 of them (45.4%) developed fusion in the Worth 4-Dot test, and 2 (18.1%) achieved measurable stereopsis. According to the Titmus stereopsis test, 40 arcsec of stereopsis was obtained in one patient and 100 arcsec in the other patient (Table 2).

There were eight patients in both groups. The average age of group 1 was 82.0±60.6 months, while the average age of group 2 was 171.2±142.2 months (p=0.105). After the first surgery, surgical success was observed in all patients of group 1, and 50 % of patients of group 2 (p=0,77). A second surgery was applied to the patients who had unsuccessful results with the first surgery in group 2, and surgical success

was observed in all patients. Sensory function was evaluated in six patients in group 1 and in five patients in group 2. Fusion was observed in five (83.3 %) of testable patients in group 1, while no fusion was observed in group 2. In the fusion evaluation performed with the Worth 4 Dot test, it was observed that this difference between the groups was statistically significant (p=0,015). In the stereopsis evaluation, no statistically significant difference was seen between the groups (p=0.45).

Table 1. Demographic features, visual acuity, spherical equivalent, and follow-up time of patients

Patients	Sex	Age at surgery(mo)	Visual Acuity at Surgery (Decimal),(R//L)	CR(SE)	Preoperative Follow-up Time(mo)	Postoperative Follow-up Time(mo)
1	M	38	1.0//0.8	Emmetrope	8	27
2	M	40	0.5//0.5	1.00//1.50	29	28
3	M	24	0.6//0.6	1.25//1.75	16	24
4	M	190	1.0//1.0	Emmetrope//-1.00	3	3
5	F	144	1.0//1.0	Emmetrope	2	26
6	F	118	1.0//1.0	Emmetrope	8	4
7	F	42	1.0//1.0	-1.00//-2.00	4	7
8	M	230	1.0//1.0	Emmetrope	3	48
9	F	186	1.0//1.0	Emmetrope	4	13
10	M	30	1.0//0.8	Emmetrope//-0,75	16	50
11	M	76	0.05//0.05	5,00//4.00	4	36
12	F	340	0.5//1.0	-4.00//-1.00	2	4
13	M	42	1.0//1.0	Emmetrope	34	12
14	M	56	1.0//0.7	0.75//1.00	18	50
15	M	70	0.3//0.3	1.50// Emmetrope	15	26
16	M	400	1.0//0.6	-5.00//-5.00	6	10

CR, cycloplegic refraction; F, female; M, male; SE, spherical equivalent

Discussion

The sensory function was assessed in 11 testable patients (68.7%), of which 5 (45.4%) fusion was observed in the Worth 4-Point test, and 2 (18.1%) achieved measurable stereopsis. While the preoperative deviation had an effect on fusion, it had no effect on stereopsis and surgical success. The postoperative successful surgical outcome has been reported in 60%-85% of cases (9). Many studies have assessed early and late surgical management of infantile exotropia. Park et al. notified that there is no significant correlation between age at the time of surgery and motor outcome (9). Several studies have been performed on early surgical outcomes (1, 10, 11). Rubin et al. obtained satisfactory outcomes in 11 of 13 (84 %) (11). In our study, the rate of surgical success was higher than in previous studies (75% after the 1st surgery, 100% after the 2nd surgery). The patients who required second surgery had higher preoperative deviation (62.5 ± 6.4 PD), and the patient informed that the second surgery might be necessary. The age at surgery of the patients in our study was higher. This is due to the low socio-economic status of our region and the fact that the patients applied to our clinic at an older age. The high rate of surgical success in our study may be because most of the patients had completed eye growth, which was more compatible with the standard tables used for surgery. Furthermore, another reason for the high surgical success rate may be the more reliable preoperative deviation measurement due to the older patients' age.

Many infantile exotropia cases do not achieve binocular vision despite the successful postoperative motor outcome. Biglan et al. reported 12 infantile exotropia patients followed for more than 4 years: all achieved satisfactory motor outcomes after surgery; but, only five (41 %) patients had a fusion, and two (%16) had stereopsis of 100 arcsec or better at long-term follow-up (12). Bagheri et al. observed successful postoperative ocular alignment of 66% after the first surgery and 92% after the second operation (5). They were able to evaluate sensory outcomes in 40% of cases (5). They found the binocular fixation rate (75%) was relatively high, and stereopsis (20%) was similar to previous studies (12-14). Similar to the study of Biglan et al., fusion was observed in 45.4% of the patients and stereopsis in 18.1% of the patients in our study.

Worth 4-Dot and Titmus stereopsis tests are used to evaluate second and third-order fusion, respectively (15). The lower orders of fusion are necessary levels that must be in place before a person can attain higher levels. Thus, the presence of sensory fusion does not confirm the pre-

sence of stereopsis. Some patients easily fuse similar images and have normal fusion amplitudes without good stereopsis. Third-order fusion (stereopsis) is a higher degree of fusion that requires more binocular cooperation and is more difficult to exhibit (13). This may explain the discrepancy between the relatively high rate of fusion and the low stereopsis rate in our study.

Table 2. Preoperative characteristics, surgical approaches, and results of the patients

Patients	Age at Surgery (mo)	Measurement Type	Preop XT (PD)	IOOA (R/L)	First Surgery	After First Surgery (PD)	Second Surgery	After Second Surgery (PD)	Fusion	Stereopsis, (arcsec)
Grup 1										
1	38	Krimsky	30	0/0	MR 4 mm resection LR 8 mm recession	5 ET	-	-	Good	No stereopsis
2	40	Krimsky	45	2/1	MR 5 mm resection LR 8 mm recession IO 10 mm recession	10 XT	-	-	No fusion	No stereopsis
3	24	Krimsky	45	0/0	MR 4 mm resection LR 8,5 mm recession	Ortho	-	-	-	-
5	144	Prism cover	35	0/0	MR 5 mm resection LR 8 mm recession	Ortho	-	-	Good	40
6	118	Prism cover	20	0/0	MR 4 mm resection LR 5 mm recession	6 XT	-	-	Good	100
9	186	Prism cover	40	0/0	MR 5,5 mm resection LR 8 mm recession	Ortho	-	-	Good	No stereopsis
10	30	Krimsky	45	0/0	MR 5 mm resection LR 8 mm recession	2 XT	-	-	Good	No stereopsis
11	76	Hirschberg	25	0/0	MR 4 mm resection LR 5,5 mm recession	Ortho	-	-	-	-
Grup 2										
4	190	Prism cover	52	1/0	MR 6 mm resection LR 8,5 mm recession	Ortho	-	-	-	-
7	42	Hirschberg	90	0/0	MR 5 mm resection LR 10 mm recession	Ortho	-	-	-	-
8	230	Prism cover	62	2/0	MR 6 mm resection LR 9 mm recession IO 10 mm recession	Ortho	-	-	No fusion	No stereopsis
12	340	Prism cover	65	0/0	MR 6 mm resection LR 12 mm recession	Ortho	-	-	No fusion	No stereopsis
13	42	Krimsky	65	0/0	MR 5,5 mm resection LR 10 mm recession	35 XT	MR 5 mm resection LR 8 mm recession	Ortho	-	-
14	56	Krimsky	55	0/0	MR 6 mm resection LR 8,5 mm recession	20 XT	MR 4 mm resection LR 6 mm recession	Ortho	No fusion	No stereopsis
15	70	Krimsky	60	0/0	MR 5,5 mm resection LR 8 mm recession	20 XT	LR 7 mm recession	Ortho	No fusion	No stereopsis
16	400	Krimsky	75	0/0	MR 6 mm resection LR 10 mm recession	40 XT	MR 6 mm resection LR 8 mm recession	10 XT	No fusion	No stereopsis

ET, esotropia; IO, inferior oblique; IOOA, inferior oblique overaction; LR, lateral rectus; MR, medial rectus; Ortho:orthotropic; XT, exotropia; PD, prism dioptri

Various studies have been conducted on the factors affecting exotropia surgery results in patients younger than 1-year-old. Though, the factors predicting the successful surgical outcome of early-onset exotropia have not been well determined (6, 9). Paik et al. suggested that early surgical approach resulted in better sensory function (1). However, Bagheri et al. reported that the early surgical approach did not significantly lead to more effective sensory functions,

and younger age had been associated with reoperation (13). Park and Kim have shown that no factors, including age at surgery, affect surgical results (9). In another research, postoperative results seem to be affected by misalignment duration rather than age at surgery (7). Shin et al. reported that preoperative part-time occlusion treatment improved the surgical result of ear-ly-onset exotropia and postsurgical stereopsis (8).

According to outcomes from research by Yam et al., the smaller preoperative exodevi-ation was related to higher surgical success at 6 weeks after surgery (6). In our study, we evaluated the impact of preoperative deviation on surgical success and sensorial function. The surgical success, fusion and stereopsis rates were higher in patients with smaller preoperative deviation (group 1). While the preoperative deviation had statistically significant an effect on fusion, it had no effect on stereopsis and surgical success. The limited cases of our study may have prevented finding a statistically significant difference between the groups in evaluating surgical success and stereopsis.

Unfortunately, our study has some limitations. Because of the low prevalence of the disease, a limited number of cases included in the study. In some cases, the beginning of exotropia was noted according to the parents' history, which may cause recall bias. The distribution of age at surgery was wider, which resulted in different cooperations of the patients and the measurement of preoperative deviation by different techniques. We recommend that more extensive prospective studies better understand the effect of preoperative deviation on surgical success and sensory function for these patients.

In conclusion, the smaller preoperative deviation may be associated with a higher rate of sensorial fusion development. Although a higher surgical success was observed with a smaller preoperative deviation, this was not statistically significant. Larger studies are required to assess the impact of preoperative deviation on surgical success. Furthermore, relatively high surgical success can be achieved in infantile exotropia patients at higher age of surgery due to a more accurate measurement of preoperative deviation and completed eye growth.

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