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Research Article

Surgical Outcomes of Late Surgery in Uncomplicated Infantile Exotropia

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Abstract

The purpose of our study is to determine the motor and sensory outcomes in children undergoing surgery for infantile exotropia after age of 2 years and to review the association of pre-operative variables with surgical outcomes. In this retrospective study, we included patients who were diagnosed as infantile exotropia with a deviation of > 25 Prism diopters (PD) before 12 months age and subsequently underwent strabismus surgery at a single centre from 2014 to 2019. Postoperative motor alignment was assessed at 6 weeks and binocular sensory status was assessed using Worth 4-Dot (WFDT) and Titmus stereoacuity tests at 6 months follow up. A total of 30 patients were analysed. At 6 weeks post-op, motor success was seen in 24 patients (80%), whereas 6 patients (20%) constituted failure group which included 5 patients (17%) with a recurrence and 1 (3%) with an overcorrection. At 6 months post-op, measurable stereopsis was present in 15 patients (50%) with a stereopsis better than 400 sec of arc in 12 patients (40%), binocular single vision was present in 6 patients (20%) on WFDT. Smaller pre-operative angle of deviation was significantly associated with motor success for distance (p = 0.009) and near (p = 0.020). Smaller pre-operative angle for distance was associated with better sensory outcomes (p = 0.041). Pre-operative vision, age at surgery and duration of misalignment did not show statistically significant association with motor and sensory outcomes. Thus we conclude, smaller pre-operative angle of deviation for distance and near are associated with good motor and sensory outcomes. Older age at surgery in this study had outcomes comparable to younger age group reported in previous studies. The association of surgical outcomes with pre-operative vision and duration of misalignment could not be proven by this study.

Keywords: Strabismus; Exotropia; Infantile; Surgery; Binocularity; Outcomes; Intermittent Exotropia

Introduction

Infantile or congenital exotropia (XT) is a rare motility disorder characterized by divergent strabismus that is apparent in infancy and persists beyond 6 months age [1]. It is reported to affect about one per 30,000 infants in the general population [2]. It has also been reported that there are two types of infantile exotropia before 1 year of age; early onset intermittent exotropia and primary infantile exotropia which do not have difference in their motor outcome [3]. Surgical management is necessary in most cases of infantile exotropia Based on previous reports, the eyes aligned successfully before 24 months of age have optimal motor and sensory outcomes [4-6]. Surgery at an older age might have an increased risk of recurrence and poor sensory outcomes. On the other hand ,proponents of surgery at an older age (beyond 2 years) note that early surgery might cause overcorrection and monofixation syndrome promoting amblyopia [7-9]. Older age at surgery has shown better sensory outcomes in a study by Rajavi., *et al* [10]. Authors have also mentioned poor surgical outcomes of infantile exotropia when operated early (both constant type and intermittent-type exotropia with onset before 1 year of age) due to postoperative exodrift [11,12].

Factors like constancy, angle of deviation, refractive error affecting surgical outcomes have been studied and it has been noted that, duration of misalignment has a significant contribution [13,14].

Through this study we intend to report the post-operative motor alignment and sensory outcomes and their association with age at surgery, pre-operative vision and angle of deviation, in patients of infantile exotropia who are operated beyond 2 years of age.

Subjects and Methods

The clinical records of all children with infantile exotropia who were operated between 2014 and 2019 at our tertiary referral centre, were retrospectively reviewed. Informed consent was obtained from all participants' parents prior to surgery. The study was approved by the Ethics Committee and was compliant with the principles of the Declaration of Helsinki (October 2008 revision). Infantile exotropia was defined as constant exodeviation of > 25 Prism diopters (PD) diagnosed before 12 months of age. The age of onset of exotropia was taken as age at which the misalignment was first noted by parents and was confirmed by reviewing past photographs. Children with any of the following were excluded: history of neurologic diseases, history of prematurity (defined as a gestational age < 35 weeks), trauma, significant refractive error, organic ocular or orbital pathology that could reduce vision, and exotropia caused by previous eye muscle surgery. Children with associations like amblyopia, latent nystagmus, oblique muscle over action, and dissociated vertical deviation were included. The following data were recorded for all subjects: gender; age at presentation, age at surgery; preoperative and final visual acuity; preoperative cycloplegic refractive error; primary, postoperative, and final angle of strabismus; primary and secondary surgical procedures; associated ocular motility disturbance; final binocular sensory status; follow-up period; and presence of amblyopia at final follow-up (defined as best corrected visual acuity worse than 20/40, difference of best-corrected visual acuity more than 2 lines, and fixation preference as noted by induced tropia test). In preverbal, nonverbal, or uncooperative children, visual acuity was evaluated as central-steady-maintained (CSM) using light reflex. The distance angle of deviation was measured at 6 m and near angle at 33cm in all cooperative children using prism alternate cover testing with spectacle correction. Sensory evaluation whenever possible was done using Worth 4-Dot and Titmus stereoacuity tests (Stereo Optical, Chicago, IL). Angle of deviation was measured over 2 or 3 consecutive visits. Angle of deviation was measured at least 2 times in consecutive visit. Surgical doses were based on surgeon's experience in guidance of the standard reference tables.

Surgery was performed by a single surgeon at one centre (SG). Motor outcomes were measured at 6 weeks post-op and at 6 months follow up. For motor outcome, patients were assigned to one of the following postsurgical groups at 6 weeks: (1) Success, defined as ocular alignment with esotropia of < 5PD or exotropia of < 10PD; (2) Failure group which constitutes; recurrence, defined as exotropia of > 10Δ; or overcorrection, defined as esotropia of > 5PD. Motor and sensory outcomes were evaluated at 6 month follow up.

Statistical analysis

Statistical analysis was performed using R version 4.0.2. The Mann Whitney was used for analysing motor outcomes and Kruskal Wallis test was used for sensory outcomes; P value of < 0.05 was considered statistically significant and all the p values were two sided.

Results

A total of 44 children were operated of which 14 were lost to follow up. Thirty patients (15 boys) were included in the study out of which, 10 were early onset intermittent variety and rest were constant. Table 1 summarizes baseline characteristics of patients enrolled. The average angle of deviation was 39.18 ± 21.35 PD.

Twenty-seven patients underwent bilateral lateral rectus recession, 3 underwent unilateral Lateral rectus (LR) recession with Medial resection (MR) resection. Mean age at surgery was 59.3 ± 27.05 months (Range 24 - 151 months). The surgical details are as mentioned in table 2.

Motor outcome was evaluated at 6 weeks post-operatively as shown in figure 1. Six weeks after the surgery, 24 patients (80%)

Parameter		Values
Sample (N)		30
Gender		Males 15
Age (months)	Mean ± SD	50.16 ± 32.80
	Range	8-144
Age of onset (months)	Mean ± SD	6.57 ± 5.08
	Range	0-12
Family history		1
Developmental delay		5
Birth history	Full term	22
	Pre-term	4
	Not known	4
RE vision Logmar		0-0.9
LE vision Logmar		0-0.5
Amblyopia		7
Distance angle (PD)	Mean ± SD	42.5 ± 19.20
	Range	25-120
Near angle (PD)	Mean ± SD	35.87 ± 23.59
	Range	5 - 120
Pattern	V	8
	A	2
	None	20
DVD		15
Nystagmus		7
IOOA		8
SOOA		2
Head posture		6
Preference	RE	11
	LE	9
	Alternate	10
Stereopsis	<400 sec	9
	400-3000 sec	2
	Absent	9
	Not co-operative	10
WFDT Distance	BSV	2
	One eye suppression	9
	Alternate suppression	5
	Diplopia	2
	Not co-operative	12
WFDT Near	BSV	1
	One eye suppression	9
	Alternate suppression	6
	Diplopia	2
	Not co-operative	12

Table 1: Baseline characteristics of patients.

SD: Standard deviation, RE: Right eye, LE: Left eye, PD: Prism diopters, DVD: Dissociated vertical deviation, IOOA: Inferior oblique over action, SOOA: Superior oblique over action, WFDT: Worth four dot test, BSV: Binocular single vision.

Parameter		Values
Age at surgery (months)	Mean ± SD	59.3 ± 27.05
	Range	24-151
Duration of misalignment (months)	Mean ± SD	52.73 ± 29.12
	Range	22-151
Type of surgery	U/L R-R	3
	B/L RECESS	27
Simultaneous surgery	IOAT	6
	Other surgeries	6
Second surgery		2

Table 2: Surgical details.

SD: Standard deviation, U/L R-R: Unilateral resection-recession, B/L RECESS: Bilateral Recession, IOAT: Inferior oblique anterior transposition.

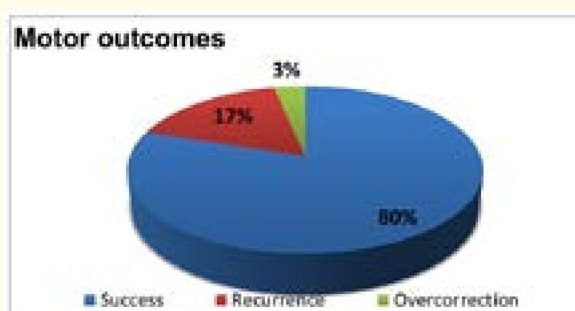

Figure 1: Showing pie chart describing the postoperative motor outcomes.

showed motor success, 6 patients (20%) showed failure out of which 5 patients (17%) showed recurrence and 1 (3%) had an overcorrection. Sensory status was recorded at 6 months follow up as shown in table 3. Measureable stereopsis was present in 15 patients (50%) with a stereopsis better than 400 sec of arc in 12 patients (40%). WFDT was recordable in 19 co-operative patients, out of which suppression was present in 9 patients (30%), binocular single vision was present in 6 patients (20%) and diplopia was present in 4 (13%). Successful motor alignment was maintained in 20 patients (67%) at the 6 month visit.

		Total	Distance		p-value	Near		p-value
			Failure	Success		Failure	Success	
	N	30	6	24		7	23	
Distance Angle	Mean	42.50	62.50	37.50	0.009	58.57	37.61	0.020
	SD	19.20	29.96	11.80		29.26	12.05	
Near Angle	Mean	35.87	62.50	29.21	0.003	57.14	29.39	0.010
	SD	23.59	31.58	15.92		31.87	16.39	
Age at Surgery (Months)	Mean	59.30	69.67	56.71	0.716	67.86	56.70	0.676
	SD	27.05	44.13	21.55		40.54	22.05	
Duration of Misalignment	Mean	52.73	65.50	49.54	0.640	62.43	49.78	0.659
	SD	29.12	46.78	23.24		43.24	23.85	

Table 3: Relationship between pre-operative variable and motor outcomes.

N: Number of patients, SD: Standard deviation.

Smaller pre-operative angle of deviation was significantly associated with better motor outcomes for distance (p = 0.019) and near (p = 0.008) (Table 4). Smaller pre-operative angle for distance was associated with better sensory outcomes (p = 0.041) (Table 3). Pre-operative vision, age at surgery and duration of misalignment did not show statistically significant association with motor and sensory outcomes (Table 5 and 6).

		Stereopsis			p-value
		>400 arc sec	400-3000 arc sec	No stereopsis	
	N	12	3	5	
Distance Angle	Mean	34.17	26.67	60.00	0.041
	SD	7.93	2.89	36.23	
Near Angle	Mean	24.50	25.00	60.00	0.064
	SD	13.60	5.00	37.42	
Age At Surgery (Months)	Mean	59.58	58.33	66.80	0.966
	SD	19.19	14.57	29.53	
Duration of Misalignment	Mean	50.83	54.33	66.80	0.612
	SD	19.00	21.08	29.53	

Table 4: Relationship between pre-operative variable and sensory outcomes.

N: Number of patients, SD: Standard deviation.

		Total	Distance		p-value	Near		p-value
			Failure	Success		Failure	Success	
	N	25	5	20		5	20	
RE vision	Mean	0.24	0.30	0.23	0.248	0.24	0.24	0.552
	SD	0.22	0.16	0.23		0.11	0.24	
LE vision	Mean	0.22	0.28	0.21	0.380	0.22	0.22	0.806
	SD	0.18	0.16	0.18		0.11	0.19	

Table 5: Relationship between pre-operative vision and motor outcomes.

N: Number of patients, RE: Right eye, LE: Left eye, SD: Standard deviation.

At the final follow up with a minimum period of 2 years, stereopsis was recordable in 9 patients (30%) as the remaining patients were lost to follow up.

Discussion

Infantile exotropia although considered a counterpart of infantile esotropia, there have been fewer studies discussing the clinical

		Stereopsis			p-value
		>400 arc sec	400-3000 arc sec	No stereopsis	
	N	12	2	4	
RE vision	Mean	0.18	0.20	0.43	0.075
	SD	0.17	0.00	0.17	
LE vision	Mean	0.17	0.20	0.40	0.094
	SD	0.18	0.00	0.14	

Table 6: Relationship between pre-operative vision and sensory outcomes.

N: Number of patients, RE: Right eye, LE: Left eye, SD: Standard deviation.

features, timing of surgical intervention and outcomes. According to Noorden, the deviation at distance fixation cannot be evaluated reliably before 2 years of age [15]. However, several studies show good sensory outcomes when operated before 2 years of age due to improvement in motor alignment and chances of improvement of binocular fusion [5,6]. Other factor which played a role was the constancy where surgical correction of early onset intermittent exotropia could be delayed since they had stronger sensorial adaptation [4]. Suh and Park., *et al*. have showed that age does not affect the surgical outcomes [14,16].

We evaluated surgical outcomes in infantile exotropia who underwent surgery after 2 years of age. Most of our children presented to the clinic at a later age, the mean age of presentation was 45.61 ± 27.67 months, therefore we were able to perform surgery only at a later age.

Successful motor alignment at 6 weeks was seen in 80% cases. This is higher than the motor success rate of 73% in a study by Paik., *et al*. who included patients undergoing surgery at age less than 2 years and a rate of 66% in a study by Bagheri., *et al*. who operated children less than 3 years age [6,15]. Biglan., *et al*. and Biedner., *et al*. noted that if surgical alignment is satisfactory at post-operative first and sixth week follow up, then chances of reoperation is lower [2,18]. In our study also the motor success was maintained in 67% of the children at 6 months follow up. Our results are comparable with the 60% long term success rate reported by Park and Kim where late surgery was performed [10].

In this study we found measurable near stereopsis in 15 patients (50%) and less than 400 seconds of arc in 12 patients (40%) at six month post-operative follow up. This is relatively lower as compared to a study by Rajavi., *et al*. on 54 patients of infantile exotropia, which reported measurable stereopsis in 77.7% patients operated after 5 years age [10]. They also reported binocular fusion in 75.9% patients as compared to 20% in our study. Similarly, Bagheri., *et al*. has reported worse sensory outcomes in children with younger age at surgery [17]. We also agree that older age at surgery can result in better stereopsis in infantile exotropia. However, it is also acceptable that sensory examination is reliable in older age group as compared to very young children due to better ability to respond. We also agree that we had included children with early onset intermittent exotropia which could be a possible explanation for the fair sensory outcome. In another study by Na and Kim., *et al*. age of onset ≤ 6 months was associated with worse sensory prognosis for children with infantile exotropia, irrespective of the constancy of its nature without a difference in motor outcomes [19]. In the present study stereopsis less than 400 seconds of arc was noted in 40% children.

In our study, we have included early onset intermittent exotropia and constant exotropia presenting before 1 year of age based on the history and photographic evidence. We assume the angle of deviation to be more definitive as children in our cohort presented at a later age, where cooperation is believed to be better. Both the groups had similar motor and sensory outcomes. A detailed subgroup analysis would be more desirable which was not done in our study due to smaller sample.

In the present study standard surgical dosage was used, without any planned augmentation or under-correction as few studies have reported change of surgical dosage does not affect the outcomes [20]. Although Cho and associates and Nam and associates recommend 1 - 2 mm under correction to achieve targeted 5PD overcorrection to maintain long term stable measurement [20,21]. Bilateral lateral rectus recession remained the procedure of choice in except 3 children with dense amblyopia. Moreover, in this study bilateral rectus recession did not affect the motor outcomes whether it was an early onset intermittent or constant exotropia as reported by Yam., *et al* [12].

According to our study, smaller pre-operative angle is significantly associated with higher chances of motor success and better sensory outcomes. This has been also noted by Rajavi., *et al*. where higher angle of deviation resulted in more suppression [10]. In another study by Yam., *et al*. older age at surgery, smaller pre-operative angle and longer interval between onset and surgery had better motor outcomes at 6 weeks post-op [11]. Yoo., *et al*. concluded shorter duration of misalignment to have significant association with risk of failure, which could not be proved in our study [13]. The association of surgical outcomes with pre-operative vision did not hold true from our results, unlike noted by Rajavi., *et al*. where better sensory outcomes were noted in children with Best Corrected Visual Acuity better than 0.3 Logmar. In a study by Park and Kim none of the factors including age at surgery, affected surgical outcomes [14].

Although older age of surgery in our cohort showed good surgical outcomes yet our study has its limitations. First, ours is a retrospective study. Second the study is limited by a smaller sample size. Considering its low lying prevalence, this issue of small sample remains unavoidable. Third, a short follow up duration of 6 months after surgery does not predict the long term results accurately. Fourth, the history of onset of exotropia was made based on recollections by the parents or relatives, or photographic documentation, which holds a potential source of recall bias.

The robustness of this study is favoured by the fact that there was consistency in surgical procedure and was operated by single surgeon (SG). The other strength is that since measurements were done at an older age and at least on two occasions, this was considerably reliable.

In our population awareness about correction of strabismus is still a setback and presence of strabismus in children is many a times seen as a social omen, surgical success can be attained despite presenting at a later age and operating beyond 2 years of age.

Conclusion

Thus, we conclude smaller pre-operative angle of deviation for distance and near are associated with good motor and sensory outcomes. Older age at surgery in this study had outcomes comparable to younger age group reported in previous studies. The association of surgical outcomes with pre-operative vision and duration of misalignment could not be proven by this study.

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