"Yokoyama Procedure" Efficacy on Non-Highly Myopic Patients with Acquired Esotropia and Hypotropia

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Running Title:
Yokoyama Procedure in Acquired Eso-Hypotropia

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Abstract

Objectives: To assess the efficacy of "Yokoyama Procedure", on non-highly myopic patients with acquired esotropia and hypotropia.

Methods: The study involved 10 eyes of 5 patients with eso-hypotropia. Inclusion criteria were: acquired esotropic-hypotropic strabismus with lateral rectus inferior displacement and superior rectus nasal displacement confirmed by MRI, refractive errors between ±6 Diopters and axial length<27 mm. Range of full duction movements and maximum angles of abduction-sursumduction was measured in each eye before and after surgery. All patients underwent T1- and T2-weighted MRI imaging. The surgery was aimed at creating a junction between the muscle bellies of the superior and lateral rectus muscles. This junction was made approximately 14 mm behind the insertions using a non-absorbable mersilene 5/0 suture (Yokoyama procedure).

Results: Mean patient age was 64,8±4,8 years. The mean globe axial length was 25,4±0,76 mm and a mean corresponding spherical equivalent refraction of -3,7±1,7 D. Eight eyes on 10 had mild limitation in abduction, while the remaining 2 had no limitation. Three out of ten eyes showed a moderate limited sursumduction, 5 eyes were categorized as mild, and the remaining 2 had no limitation. No evident post-operative limitation was present in any eye, in both abduction and sursumduction($p<0.01$). Pre-operative esotropia and hypotropia were respectively 32±11PD and 25±5PD, and they were significantly reduced after surgery as 9±1,7PD and 6±1PD($p=0,043$).

Conclusion: Yokoyama procedure is an effective, fast, reversible procedure to face eso-hypotropic acquired strabismus, even in patients with a clear MRI displacement

of superior and lateral rectus muscles, and absence of globe dislocation and of elevated myopia.

Introduction

Myopia and strabismus in adult patients are frequently related; in fact different acquired strabismus forms develop in myopic patients.

Certain of these patients develop progressive esotropia and hypotropia, with associated impaired abduction and sursumduction.

This condition was named "heavy eye syndrome" (HES)[1], and in the most advanced stage, as the eye can't be moved past the midline, it's called "myopic strabismus fixus"[2]. The pathophysiology was studied by Ohta et al. with orbital computed tomography[3] and by Krzizok and Schroeder by magnetic resonance imaging (MRI)[4], putting the blame on the downward displacement of the lateral rectus muscle. Later, Yokoyama et al. and Aoki et al. showed that the posterior portion of the myopic globe was elongated out from the muscle cone with a superotemporal dislocation[5,6]. Based on this premise, the same group showed a new surgical method (the "Yokoyama procedure") tying the muscle bellies of the superior rectus (SR) and the lateral rectus (LR) muscles with an non-absorbable suture[7]. They demonstrated that this procedure was effective both in restoring the dislocated globe back into the muscle cone and in improving ocular motility and deviation. In some patients with long lasting restricted abduction, a contractured medial rectus could recessed at the same time with excellent result[8].

Another peculiar condition characterized by esotropia and hypotropia for distance, associated with sursumduction and abduction deficit, has been named "sagging eye syndrome" (SES)[9], due to the aging related pulleys' inferior displacement (sag) at the level of the connective tissue suspending the lateral rectus(the LR-SR band)[9]. As consequence, the lateral rectus muscle slips

inferiorly similarly to myopic “heavy eye” syndrome. This condition was firstly associated to non-myopic patients because HES and SES were considered mutually exclusive, but later Tan and Demer showed that SES could occur in highly myopic patients too[10]. Authors described that these patients exhibited less globe back dislocation, with an associated superotemporal prolapse of soft tissue, which might directly limit the globe shift[10]. MRI studies are in fact essential in the differential diagnosis of these two syndromes, in view of the different surgical approach.

We present 5 adult patients with moderate myopia but the typical MRI features of HES, treated with the Yokoyama procedure. To our knowledge this is the first study that describes the results of this surgery in non-highly myopic eyes.

Methods

This research was designed as an interventional case series. Ten eyes of 5 patients with eso-hypotropia were examined at San Giuseppe Hospital in Milan between 2016 and 2018. Criteria for inclusion in this study were: acquired esotropic and hypotropic strabismus with MRI apparent lateral rectus inferior displacement and superior rectus nasal displacement, refractive errors between ± 5 Diopters and axial length < 27 mm, ocular axial length was measured by IOL Master 500 (Zeiss, Oberchen, Germany).

Patients with history of strabismus surgery and/or other possible causes of ocular movement limitation were excluded. Data were obtained interviewing the patient.

All patients underwent visual acuity assessment, motility examination, slit-lamp and dilated fundus examination and refraction. The range of full duction movements was measured in all patients. Duction limitation was graded from 0 (full movement) to -4 (no movement past the midline). Maximum angles of abduction and sursumduction were obtained in each eye, making the patient look at a fixation object with one eye at a time. The angle of ocular deviation was measured, at time 0 (pre-operative evaluation), with the alternate prism cover test in primary position at distance. All patients underwent axial and coronal T1 and T2-weighted MRI imaging. In order to investigate the anatomic relationships between the globe and the SR and LR muscles, a set of coronal images with 2 mm interval of each orbit, taken perpendicular to the orbital axis, was obtained. In all patients the diagnosis was confirmed and a displacement of both superior and lateral rectus muscle was present (Figure 1).

The surgery was aimed at creating a junction between the muscle bellies of the SR and LR muscles (Yokohama Procedure). This junction was made approximately 14 mm behind the insertions using a non-absorbable mersilene 5/0 suture. None of the patients required medial rectus recession because the movable range of the eyeball was arbitrarily considered sufficient by one of us (PN) after the union of the SR and the LR muscles.

To evaluate the surgical effects, we compared the maximum angles of abduction and sursumduction and the angle of ocular deviation before and after surgery. The post-surgical follow-up was usually between 30 days and 6 months from the day of the surgery

Statistical analysis was determined using non-parametric tests. Every specific test is specified when used. Data were presented as mean ± standard deviation.

Values of $P<0.05$ were considered significant. Statistical analysis was performed using SPSS Statistics 17.0 software (SPSS, Inc, Chicago, Illinois, USA).

Results

A summary of the series' characteristics is reported in table 1. Mean patient age was 64,8±4,8 years. Three of them were female. The mean globe axial length was 25,4±0,76 mm. Mean axial length of the 5 right eyes was 25,4±0,6 mm and a mean corresponding spherical equivalent refraction of -4±1,1 D. Left eyes axial length was 25,4±0,9 mm and their refraction were -3,4±2,3 D. We performed MRI in all patients and displacement of both superior and lateral rectus muscle was all of them.

No eye showed a limitation in the abduction movement larger than -2, as described in Table 1. Eight eyes on 10 had -1 limitation, while the remaining 2 had no limitation (patient 3 left eye and patient 4 right eye). Three out of ten eyes showed a limited sursumduction of -2 (patient 2 left eye and both eyes of patient 5), 5 eyes were categorized as -1, and the remaining 2 had no limitation (patient 3 left eye and patient 4 right eye) (Table 1). Post-operative limitation was absent in all the eyes, in both abduction and sursumduction. A significant difference was found regarding both abduction limitation ($p<0,01$, Wilcoxon Test) as sursumduction limitation ($p<0,01$, Wilcoxon Test).

Pre-operative esotropia was 32,0±11,5 prismatic diopters (PD), and it was significantly reduced after surgery as 8,6±1,7 PD ($p=0,043$, Wilcoxon Test). Hypotropia was as well significantly reduced after the surgical procedure from 24,6±4,6 PD to 5,8 ± 1,3 PD ($p=0,043$, Wilcoxon Test).

At 6 months follow up esotropia and hypotropia were stable.

Discussion

In our study we observed that the surgical procedure known as "Yokoyama procedure" is effective as treatment in non-highly myopic patients with acquired esotropia and hypotropia. The patients included in our study were all myopic, but no eye had severe myopia, in fact all eyes were shorter than 27 mm and the spherical equivalent refraction was lower than -6 diopters, neither they had the classic myopic elongation of the globe. After surgery, the parameters of eye movement and ocular deviation were both improved, without any significant complication.

Our patients showed an esotropia of 32,0±11,5 PD, and a hypotropia of 24,6±4,6 PD. These values are smaller than the one observed by Yamaguchi et al. in highly myopic patients with HES and globe dislocation (61±39 PD of esotropia, 26±21 of hypotropia)[7]. Esotropia deviation values are more similar to the ones described by Tan and Demer in their patients with SES (23±57 PD), while their patients' mean hypotropia is considerably smaller (2±2 PD)[10]. Lower values are shown in Chaudhuri and Demer's study, in which patients with non-myopic SES had a mean esotropia of 11.5±10.6 PD and hypotropia of 9.9±9.4 PD[11] (Table 2).

It seems we are face with a new form of eso-hypotropia associated to slippage of SR and LR that differs from HES because patients are not highly myopic and there is no globe dislocation, and from SES because patients show slippage of both SR and LR, without clear LR-SR band degeneration. Conversely, this form also seems caused by the partial combination of the underlying pathological mechanisms characteristics of HES and SES, as myopia and connective tissue

deterioration due to age, respectively. Even the angle of deviation values seems to fill the hole between the two pathologies, almost creating a continuum, in which connective degeneration, myopic axial elongation and globe posterior dislocation contribute with unequal weight.

Yokoyama et al. firstly described this surgery in order to treat highly myopic patients with superotemporal dislocation of the myopic globe and displacement of both the superior rectus and lateral rectus muscles[7]. They created a new suture anastomosis of the superior and lateral rectus muscles' posterior bellies in order to correct the position of the dislocated myopic globe. If needed, the medial rectus muscle recession was added to the surgery. This procedure was effective in restoring the dislocated globe and improving both ocular motility and deviation[7].

Rutar and Demer showed that in 3 strabismic, elderly and non-myopic patients the esotropia and hypotropia were caused by the LR-SR band degeneration and the subsequent inferior slippage of the lateral rectus muscle[9]. They treated their patients with two different techniques: suturing the lateral and superior rectus muscle margins together approximately 10 mm posterior to their insertions, with superior transposition of the lateral rectus muscle insertion; or suturing the margins of the lateral and superior rectus pulleys together using 5-0 non-absorbable suture[9].

Tan and Demer compared 5 patients with heavy eye syndrome and 6 with highly myopic patients with sagging eye syndrome and outlined that the difference in these two clinical entities lies in the anatomy, with a prolapsed myopic globe in HES and age-related slippage of the rectus muscle paths in SES[10]. In their study

authors didn't focused their attention on the surgical results, but they suggested that a conventional technique, as the medial rectus muscles recession, could be the best option in highly myopic patients with SES[10]. However, their group of patients were characterized by a large esotropia and a small hypotropia (Table 2). Considering this clinical appearance, a conventional approach was reasonable, instead a procedure that fixed both horizontal and vertical defects was necessary in our cohort of patients.

Acquired hypotropia and esotropia in moderate myopic eyes is a separate entity as it isn't included in the definition of both HES or SES, but at the same time it seems that it creates a clinical continuum between these two forms, probably due to common underlying pathological mechanisms.

So we hypothesized that the union of the muscles' bellies could have good effect even in non-highly myopic patients, because the vectors of muscle force of the superior rectus and of the lateral rectus should be adjusted thanks to Yokoyama surgery. It is also possible that we simply obtain a mechanical repositioning of both SR and LR. More importantly, as we have described, the absence of globe dislocation, and of a clear definition of HES, doesn't have to exclude this surgical option from the list.

Our study has some limitation: a limited number of patients, a short follow-up, and no comparison between Yokoyama surgery and other surgical techniques. However, this is a quite homogeneous group, with proved efficacy of SR-LR muscles' bellies union in a previously unexplored setting.

In conclusion we promote the Yokoyama surgery, as an effective, fast, potentially

reversible procedure even in non-highly myopic eyes with eso-hypotropic acquired strabismus due to a clear MRI displacement of superior and lateral rectus muscles.

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Titles and Legends to Figure

Figures

Figure 1. T2 Weighted Coronal MRI Scan of Patient **5**. It shows the bilateral slippage of both lateral and superior rectus muscles, responsible for eso-hypotropia. No globe enlargement is present, as no globe's back dislocation.

Tables

Table 1. Patients Characteristics

Patient	Age	Sex	Eye	Axial Length (mm)	Refraction (Diopters)	Abduction Limitation	Sursumdction Limitation	Pre-Surgery Esotropia	Post-Surgery Esotropia	Pre-Surgery Hypotropia	Post-Surgery Hypotropia
1	66	F	R	24.56	-2,5	-1	-1	35	10	25	5
			L	25.57	-4	-1	-1				
2	58	M	R	25.59	-4	-1	-1	40	8	28	6
			L	26.21	-4,5	-1	-2				
3	70	F	R	25.45	-4,75	-1	-1	20	10	20	8
			L	24.02	0,5	0	0				
4	62	F	R	24.96	-3,5	0	0	20	9	20	5
			L	25.01	-3,5	-1	-1				
5	68	M	R	26.23	-5,25	-1	-2	45	6	30	5
			L	26.31	-5,5	-1	-2				

Table 2. Esotropia and Hypotropia Prismatic Diopters Comparison between Different Studies

	Esotropia	Hypotropia
Our study	32,0 ± 11,5	24,6 ± 4,6
Yamaguchi et al.[7]	61 ± 39	26 ± 21
Tan and Demer[10]	23 ± 57	2 ± 2
Chaudhuri and Demer[11]	11.5 ± 10.6	9.9 ± 9.4