Spontaneous Consecutive Exotropia after Conservative Management of Accommodative Esotropia

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ABSTRACT

Spontaneous consecutive exotropia (SCXT) in conservatively managed accommodative esotropia is not widely discussed. The author presents a case series of 4 patients with SCXT describing patient characteristics and possible risk factors. Management modifications based on this series are proposed. Four cases of patients who initially presented with accommodative esotropia and later developed SCXT are presented. The age range at presentation was between 4 months to 3 years, with total follow-up ranging between 8–15 years. All four cases presented with esotropia on or before the age of 3 years, with initial hyperopia ranging between +1.50 to +4.25. The SCXT occurred between ages 2–7 years. Possible risk factors identified include amblyopia, dissociated vertical deviation, rapid control of esodeviation, inferior oblique overaction, and poor follow-up during the course of their strabismus. The author recommends earlier tapering of hyperopia correction following initial control with full cycloplegic refraction. Identified risk factors should alert the specialists to be wary of SCXT.

Keywords: spontaneous consecutive exotropia, accommodative esotropia

INTRODUCTION

Traditional teaching dictates that refractive accommodative esotropia needs to be managed with spectacle prescriptions obtained following full cycloplegic refraction.[1,2] The indications for tapering the hyperopic prescription are usually dictated by exotropia developing with the hyperopic prescription, or through the reading addition lenses, as well as poor vision with a full correction that is improved by reducing the hyperopic prescription, among others. Full cycloplegic refraction in spectacles is preferred over giving minimum hyperopic prescription and catching up with residual esotropia and hyperopia. Nonetheless, the goal of treatment remains clear – to control the esodeviation to within 8 PD of esotropia or better. This allows at least peripheral fusion to develop and expand a patient's fusional amplitudes.

While consecutive exotropia following esotropia surgery is widely recognized, spontaneous consecutive exotropia (SCXT) following accommodative esotropia managed conservatively is rarely discussed. This is an outward drift observed following management with hyperopic prescription of an esotropia that was previously controlled with full correction.

There are a few case series that discussed spontaneous consecutive exotropia written by Shin et al. (2020),[3] Senior et al. (2009),[4] Weir et al. (2001),[5] and Beneish et al. (1981).[6]

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The Japanese study by Shin et al. (2020)[3] had the largest series of 51 patients with SCXT following only full hyperopic prescription for refractive accommodative esotropia. Comparing patients who developed consecutive exotropia and those that remained controlled, more dissociated vertical deviations, more rapid reduction in hyperopia, and faster control of esotropia were found in patients with SCXT. Amblyopia and the size of the deviation were no different among the two groups.

Senior et al.[4] (2009) described a series of 14 patients who presented with SCXT. None of his patients was patching at the time of diagnosis of SCXT. The most commonly identified associations were the onset of esotropia before age 2 years, hyperopia of at least 5 diopters, and lack of binocular single vision.

Weir et al.[5] (2001), citing von Noorden, stated that SCXT usually starts as having partially accommodative esotropia with poor binocular function and weak motor fusion, thus preventing the attainment of stable eye alignment. In the 5 cases they reported, esotropia was first reported at age range 1.5 to 3.75 years, with esodeviation ranging from 15–50 PD, mostly measurements obtained at near fixation. The refractive errors range from +4.00 to +10.75 D, with 2 patients reported as having anisometropia as well. There was an absence of stereoacuity, but motor fusion was confirmed by fusing 20 PD of base-out prisms. Although monofixation esotropia was documented just before the preceding clinic visit, exotropia ranging from 2–16 PD was observed for near, and 4–20 PD for distance. In all patients, with unchanged cycloplegic refraction, hyperopic prescription was reduced by up to 1.5 diopters. This maneuver controlled the deviation to less than 10 PD of exotropia in 3, and less than 10 PD of esotropia in 2.

Beneish et al. (1981)[6] reported on 22 children who developed exotropia following hyperopic prescription given due to accommodative esotropia. Predisposing factors identified included the early onset of esodeviation, and the high hyperopia exceeding +5.0 diopters. In most cases, reduction of hyperopia by at least half resulted in good alignment control.

The author presents a case series of consecutive exotropia following previously controlled acquired accommodative esotropia. Possible risk factors and patient characteristics are identified and presented. Management modifications will be suggested based on lessons learned from these cases.

METHODS

A case series of four patients in the private practice of the author were retrospectively reviewed. Patients initially presented with accommodative esodeviation, either constant or intermittent, and later developed consecutive exotropia spontaneously. There were 3 males and a female, whose ages ranged from 4 months to 3 years at the time of initial presentation, with a follow-up period ranging from 8–15 years.

The study and data collection conformed to all local Philippine laws including the Data Privacy Act of 2012 and were compliant with the principles of the Declaration of Helsinki. A request for exemption for ethics review for this retrospective case series was requested and subsequently granted by the University of the Philippines Manila Research Ethics Board.

RESULTS

A summary of salient features of the cases and possible risk factors is shown in Table 1.

All patients had cycloplegic refractions either with short-acting cycloplegia or full cycloplegia that did not exceed +4.25 and presented esodeviations measuring between 25-40 PD without spectacle corrections. Three of four patients presented with a history of esotropia onset before age 1 year. Other possible risk factors identified include the presence of amblyopia in all cases, presence of dissociated vertical deviation in 75%, remote near the point of convergence in 75%, at least half had inadequate follow-up or missed scheduled follow-up for 2 years, and the presence of inferior oblique overaction in 50% of cases. Those who missed follow-up ended up wearing an outdated or wrong prescription. Other possible risk factors include the presence of pattern deviations found in 50% and prematurity found in one patient. Three of four patients had remote near point of convergence. Whether the latter is a consequence or the cause of consecutive exotropia is unclear.

Case 1

The patient was a 3-year-old who consulted for 15 degrees (~30 PD) of intermittent left esotropia (ET), suggestive of amblyopia in this eye. Atropine refraction was documented to be +2.50 OU. There was a small monofixation ET at distance, with near deviation measuring up to 30 PD. With +1.50 D correction (even while cycloplegic refraction was +2.50), a small angle of exotropia (XT) was already observed, so glasses were reduced to plano with +1.50 addition lenses. This controlled the deviation for both distance at 0–2 PD monofixation ET and orthotropic for near. Adequate control of esotropia was achieved by age 2 with a reduced plus of +1.00 despite a cycloplegic refraction of +2.00. Good alignment with subtle changes in refraction was noted till age 7. Unfortunately, the patient was lost to follow-up since then, and came back at age 9, wearing the last refraction on record from 2 years prior (+1.00 OU). He had developed poorly controlled exotropia measuring 30 PD intermittent exotropia [X(T)], with mild amblyopia of the left eye. His best correction was -0.50 in his right eye with which he saw 20/20, while the left eye required a -0.75 to see 20/40. Cycloplegic refraction was +0.50 OU. There was remote near point of convergence. Managed conservatively with his myopic prescription, patching, and convergence exercises, the patient's deviation was controlled, with measurements

Table 1. Profile of patients with spontaneous consecutive exotropia

Patient	1	2	3	4
Age at presentation	3 years	9 months	11 months	4 months
Sex	Male	Male	Male	Female
Acuity at presentation				
Right	Fixes, Follows, Maintains	Fixes, Follows, Maintains	Fixes, Follows, Maintains	Fixes, Follows, Maintains
Left	OU	OU	OU	OU
Cycloplegic refraction	Atropine +2.50 OU	Atropine +4.25* OU	Tropic-Phenyl +1.50 OU	Tropic-Phenyl +2.00 OU
Deviation at presentation	30 PD LE(T)'	40 PD ET'	35 PD ET'	25 PD E(T)'
Initial control of ET attained	2 months	6 months	3 months	3 months
Age XT documented	7 years	7 years	5 years	2 years
Cycloplegic refraction when exotropia documented	Atropine +2.50 OU	Tropic-Phenyl +1.00 OU	Tropic-Phenyl +1.00 OU	Tropic-Phenyl +1.00, OU
Exodeviation documented	4-30 PD X(T)	2-14 PD X(T)	8-35 PD X(T)	4-40 PD X(T)
Risk Factors				
Amblyopia	Yes	Yes	Yes	Yes
DVD	No	Yes	Yes	Yes
IOOA	Yes	No	Yes	No
Remote NPC	Yes	Yes	Yes	No
Wrong glasses	Yes	Yes	No	No
High AC/A	Yes	No	No	No
Period with lost follow-up	Yes (2 years)	Yes (2 years)	No	No
Preterm	No	No	Yes	No
E(T) recurred	No	No	No	Yes
Age, at last follow up	17	9	12	7
Subjective Refraction				
Right Eye	-1.00	-1.50	-1.50	-0.25
Left Eye	-1.00	-1.50	-1.50	-1.00 -0.50 x 180
Alignment				
without correction	14 PD X(T)	14 PD X(T)	12–16 PD X(T)	-not recorded
with correction	orthotropia	12 PD X(T)', DVD	10–16 PD X(T)', DVD	20PD XT w DVD
Disposition	Wear correction to control deviation	Surgery recommended; over-minus and orthoptics advised while hesitant to have surgery	Surgery recommended; over-minus and orthoptics advised while hesitant to have surgery	Surgery recommended; orthoptics while awaiting surgery

**Case 2, with a note of rapid drop to +2.50 OU in 3 months*
OU, both eyes; XT, Exotropia; X(T), Intermittent Exotropia; ET, Esotropia; L, left; E(T), intermittent esotropia; PD, prism diopters; DVD, Dissociated vertical deviation; IOOA, Inferior oblique overaction; NPC, Near point of convergence; AC/A, Accommodative convergence: Accommodation ratio; tropic-phenyl, tropicamide-phenylephrine combination

ranging from 0–10 PD X(T). At the time of the last follow-up at age 17, the patient's deviation was orthotropic with the best correction of -1.00 OU but measured 14 PD X(T) without correction.

Case 2

The patient first presented at age 9 months after being started less than a week on +3.00 OU by another eye care professional. The ET measured 40 PD without correction, improving to 30 PD with +3.00 OU. Atropine refraction revealed higher hyperopia of +4.25 OU which was promptly started. This partially controlled the deviation to 25 PD ET but repeat atropine refraction showed a rapid drop to +2.50 by age 1 year. By age 2, reduced plus prescription controlled the deviation. By age 5, the patient was noted to have mild amblyopia of the left eye with dissociated vertical deviation (DVD). He was wearing reduced plus of +0.75 OD, and +1.00 OS with which he saw 20/40 and 20/60, respectively. Short-acting cycloplegic refraction was +2.00. Controlled with reduced plus at age 5 years, the patient was lost to follow-up for 2 years, still wearing the last known prescription on record. At age 7, the patient showed monofixation exotropia even when glasses of +0,75 and +1.00 were removed. Patching was started for mild amblyopia, while the exodeviation was managed conservatively with orthoptics and tolerated over-minus lenses of -0.50 OU that gave a vision of at least 20/40. Cycloplegic refraction was +1.00 OU. At the time of the last follow-up at age 9, subjective refraction of -1.00 OU that improved vision to 20/30 at best, still gave a residual of 14 PD of X(T). This improved minimally to 12 PD with tolerated over-minus lenses of -1.50. Cycloplegic refraction was plano OU at this time. Surgery was recommended.

Case 3

Born preterm, this child's initial consultation was done at age 1, following alternating ET of 35 PD with right eye preferred for fixation persisting since age 11 months. Short-acting cyclorefraction was only +1.50 OU, surgery was advised, but the patient opted to try membrane prisms and patching. This controlled the deviation except for the XT in upgaze with inferior oblique overaction (IOOA) and DVD. By age 5 however, there was frank XT of 18–25 PD in the primary gaze which was controlled with over-minus lenses of -1.50 (short-acting cycloplegic refraction was +1.00, OU) to within monofixation range. There was also remote near point of convergence (NPC). At the time of the last follow-up at age 12, over-minus lenses sometimes controlled the deviation to 10 PD XT but deteriorates at times to 16 PD XT. Without correction, the exodeviation measured 12–16 PD. Despite advice to have a surgical intervention, parents opted to stay with conservative management that included over-minus lenses and orthoptics.

Case 4

The only female patient first consulted at age 4 months with intermittent esotropia measuring 25 PD that was mostly tropic. The mother also suffered from uncorrected esotropia from childhood. Short-acting cyclorefraction of +2.00 OU was started, which controlled the deviation for 2 years until XT was observed albeit in the monofixation range with full correction. This XT increased to 14 PD without glasses. Control of the deviation remained fair, with deterioration to 20 PD of XT, as well as DVD observed, with eye preference that shifted between the 2 eyes. At some point at age 5 years, the patient's alignment shifted to monofixational esotropia, only to revert to monofixation exotropia with dissociated strabismus. At the time of the last follow-up, at age 8 years, the patient's deviation measured 20 PD XT, with DVD, with a slightly better vision on the left eye of 20/20 with -1.00 -0.50 x 180, while the right had -0.25 refraction but saw only 20/25. Surgery was advised.

DISCUSSION

Spontaneous consecutive exotropia (SCXT) occurs in up to 18.4% of patients following correction of hyperopia in children with refractive accommodative esotropia.[7] SCXT was associated with esotropia that presented initially from early weeks to less than 4 years of age.[4,6] Our patients all presented at less than 3 years of age, consistent with their cohort, with most patients (75% in our series) presenting at less than 1 year of age.

Amblyopia as a risk factor in the development of consecutive exotropia following esotropia surgery was identified by a prior study,[8] and reported to be as high as 89% of spontaneous consecutive exotropia in refractive accommodative esotropia treated with hyperopic glasses.[7] On the contrary, however, there are studies where amblyopia was not a prominent finding in patients who develop SCXT.[4,6] Anisometropia was also identified as a risk factor but only one of our patients developed anisometropia and was already late in the course of the disease (Case 4).

Shin et al found that the mean interval to correct esotropia in patients with accommodative esotropia was shorter in those that eventually developed SCXT,[3] showing also more dissociated vertical deviation (DVD). DVD was found in 75% of the patients in our series. The faster reduction in hyperopia was also identified as a risk factor. This phenomenon was observed in Case 2, which had shown hyperopia of +4.25 that rapidly dropped to +2.50 in less than 6 months.

Prior studies documented SCXT patients had presented commonly with high hypermetropia of +5.00 D,[4,6] contrary to our series where none exceeded +4.25 OU, and most were below +2.50.

In the Shin et al study,[3] SCXT was documented after a little over three years from hyperopia correction. Our series showed an average of about 4 years before this occurrence.

The inherent limitation of this series is its retrospective nature and small sample size. Nonetheless, some lessons that could impact a change in management to avoid SCXT are suggested by this review.

The gold standard in managing accommodative esotropia remains to be prescribing the full cycloplegic refraction in patients usually younger than 5 years, and the maximum tolerated plus prescription in patients who cannot tolerate the full prescription, usually the older child. To avert the development of SCXT in accommodative esotropia patients, following initial control with full cycloplegic refraction for at least two cycles, a paradigm shift of prescribing only the minimum hyperopia required to control an esotropia deviation should be considered. This allows the medial rectus muscle to retain its muscle tone, and not fully "relax" as this is most likely contributory to the development of remote near points of convergence. This practice will also provide a "buffer" of under-correction for those who will miss follow-ups and the need to reduce hyperopia when exotropia, no matter how small, is observed. Half of our series missed appointments as long as two years wearing an outdated prescription.

A more drastic reduction of hyperopia by 50–60% suggested by an earlier report, allowed patients to have satisfactory realignment with the restoration of binocularity albeit rudimentary.[6] Ha et al.[9] recommended tapering of hyperopia at around age 6 when esotropia without correction and visual acuity can reliably be obtained in patients. Their protocol, however, recommended tapering only up to 1 diopter of hyperopia, unlike Beneish et al.[6] who recommended up to 60% reduction. This leads to the recommendation espoused by the author of this series following initial control of esodeviation with full cycloplegic refraction, subsequent visits should consider early tapering of hyperopia that will still control the esodeviation at least to within monofixation range.

The presence of features that may predispose to the development of spontaneous consecutive exotropia, such as but not limited to dissociated vertical deviation, inferior oblique overaction, and amblyopia, calls for closer monitoring of these patients. Instead of quarterly visits where opportunities to taper refraction were missed, return visits as often as monthly for refraction and alignment check may be more prudent.

Authors have also identified the lack of binocular vision as a risk factor for developing spontaneous consecutive exotropia,[4,5] despite having motor fusion confirmed by the ability to overcome up to 20 PD base-out prisms.[5] This parameter, however, was not routinely obtained in our cohort, but should perhaps be considered as a potential risk factor for spontaneous consecutive exotropia and should likewise be monitored.

Statement of Authorship

The author contributed in the conceptualization of work, acquisition and analysis of data, drafting and revising and approved the final version submitted.

Author Disclosure

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