CONGENITAL ESOTROPIA:

CASE REPORT

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ABSTRACT

Documented is the successful treatment of an esotropic child from 4 months of age to 5 years, 4 months of age. The strategy used to change the visual status from esotropia to good binocularity and acuity are outlined. Techniques included prudent patching, use of gingham patterns, development of equal visual circuits, two-sided gross body activity, eye movements, motoric and visual activities along the Z axis, and three-dimensional activities along the mid-line.

KEY WORDS

congenital esotropia, infantile esotropia, occlusion, gingham pattern, amblyopia, critical period, gross motor, fine motor

The term congenital esotropia has been assigned a number of definitions. It is frequently used synonymously with infantile esotropia; however, this has recently gained disfavor. Helverston and Ellis[1] define congenital esotropia as an esodeviation occurring in an infant less than 1 year of age. Griffin[2] categorizes strabismus according to time of onset; congenital esotropia is described as esodeviation whose onset is 6 months of age or earlier. Von Noorden[3] uses a similar time set for his characterization of congenital esotropia. He believes the terms congenital and infantile are interchangeable; however, the deviation is usually in the magnitude of 50^{Δ} . He further states that during the first week of life binocular posture varies from straight to deviated, but by 3 months of age the eyes should gain binocular coordination and alignment. Christenson et al.[4] note that frequently a distinction is made between congenital and infantile esotropia and each are ascribed various characteristics. The concept advanced by Archer[5] is that congenital esotropia develops between 2 and 4 months of age. This would more readily conform to Von Noorden's thought.[3] In general all these authors require congenital esotropia to occur no later than the first year of life.

Another means of characterizing esotropia is by the patient's refractive status. Some authors chose to make this category separate or non-inclusive with congenital esotropia. However, in most instances, they acknowledge that refractive (with a normal accommodative convergence to accommodation ratio [AC/A]) and congenital esotropia can coexist.[3] Pollard[6] makes reference to the successful treatment of accommodative esotropia in patients under 6 months of age with the appropriate hyperopic prescription. Rethy and Gel[7] postulate that a great number of congenital esotropes are accommodative in nature. Von Noorden[3] believes that most congenital esotropes are not accommodative and display a similar angle of deviation at distance and near. He also believes that the majority of accommodative esotropes manifest after 2 years of age. This age category would correspond to that found by Forrest and FitzGerald.[8]

There also exists the esotropic category some authors refer to as nonrefractive. Here the patient has a high AC/A and displays a greater turn at near than at distance.[3] Parks[9] describes the age of onset from 8 months to 7 years; however, this category of esotropia tends to cluster at 2 1/2 years of age. With all things considered it would appear that a patient could be both an accommodative and congenital esotrope. However, it is also evident that most tend to adhere to a more classic definition, i.e., time of onset.

MANAGEMENT CONSIDERATIONS OF CONGENITAL ESOTROPIA

An infant with an esodeviation presents a number of interlacing considerations for the optometrist. He needs to resolve the following questions:

1. Is the deviation a result of pathology?
2. Is the deviation caused by uncorrected hyperopia, and if so, is the turn completely eliminated with the prescription?
3. Are there mechanical or neurological extraocular muscle (EOM) involvements?
4. Is the visual acuity (VA) equal in each eye and appropriate for the baby's age?
5. As per the critical period, is the VA at risk?
6. As per the critical period, will binocularity be compromised at the cortical level?
7. Are the baby's gross motor skills appropriate for his age?
8. Are the baby's fine motor skills appropriate for his age?

The answers to these questions and the priority of need will be the beacon of treatment strategy. An elaboration of each consideration follows.

The first step of the diagnostic process is to determine whether a pathological process or insult is the cause of the deviation. The involvement could be ocular, systemic or neurologically based.[10,11,12] Thus, an appropriate evaluation is in order.

In considering hyperopia and accommodative esotropia there are three basic categories:

1. True accommodative esotropia secondary to hyperopia
2. Accommodative esotropia created by a high AC/A ratio
3. Accommodative esotropia with a residual deviation

The first category will have alignment with the proper prescription. The second category may require an add for the deviation which is likely to be greater at near than at a distance. Patients in this category may be aligned at a distance and deviate only at near. The third category will show an esotropia even with the proper prescription. For all these categories, Von Noorden recommends the correction of the refractive error and if this creates alignment "surgery is definitely contraindicated."[3] If a plus prescription creates alignment, the cosmetic or motor element of the turn is resolved. Evaluation of functional binocularity and VA would follow.

If a residual deviation occurs or no plus is accepted, muscle limitations and overactions should be evaluated. Surgical intervention definitely should be considered with residual turns. A vertical component is frequently present; however, this may be secondary to the original deviation.[3] A vertical muscle imbalance has been reported in as many as 90% of the cases with congenital esotropia.[13] Other authors have reported an average of 77%[14] to 63%.[15] Early surgical resolution of residual turns is essential if a good functional prognosis is desired.[16] Usually surgery by 12 months, but not much later than 18 months, enhances good binocular results.[17,18]

Another mode of angle resolution in accommodative esotropia with residual deviation is the use of Fresnel prisms, but these devices often create reduction in VA and could be counterproductive in some infants.[19] Nevertheless, this is a viable approach in some cases. For the sake of completeness, miotic therapy should be considered. Anticholinesterases, i.e., acetylcholine or a cholinergic drug, are used to increase the accommodative response without increasing convergence. This method is sometimes employed when the patient is resistant to wearing the spectacles or as a substitute for a near add. The drugs routinely used for this therapy are isoflurophate or echothiophate. It seems to me that this therapy mode is cyclic in its popularity because it presents undesirable side effects,[20] namely anterior subcapsular opacities, iris cysts and, consequently, prolonged therapy is not desirable.

The possibility of amblyopia and the development of binocularity become companion concerns for an infant with esotropia. The practitioner must consider the concept of critical periods of visual development. Hickey and Peduzzi[21] found that stereopsis emerges between the 2nd and 6th month of life and that the visual cortex is not adult-like until the 6th month. Blakemore[22] makes the following points about the neurological development in the visual cortex. The babe comes into the world with most of the neutral circuitry and there is little subsequent increase in the number of neurons. However, the neural elements continue to grow and differentiate. The process is basically gene driven, but the environment plays a part. Positive stimuli foster development of the system, while negative occurrences retard the process. Blakemore's work with monkeys showed equal development and differentiation of neurological cells in each eye. However, when he created a depriving condition, the result was a reduction in the amount of space occupied by the affected terminals in the cortex layer. He further goes on to state: "Early deprivation (2 to 5 1/2 weeks) caused a very clear imbalance in the fraction of cells in layer IVc dominated by the two eyes, reflecting the change in the relative size of ocular dominance bands. Late deprivation, starting at 11 months, had no obvious effect at all on the physiologically determined right eye and left eye region in layer IVc."[22]

Hubel and Wiesel[23] also gave evidence supporting the concept of a critical period. They created a visually depriving condition in kittens and found a marked reduction in binocular function when monodeprivation took place.[23]

Hoyt and Caltrider[24] used electrophysiological measures and found no intrinsic basis for the lack of development of fusion and stereopsis in congenital esotropes. Consequently there exists the potential for the development of fusion. This concept leads one to consider neutralization of the angle of deviation by prism and, in addition, the potential to develop binocular cells even in the absence of alignment.[24] Blakemore and Van Sluyters demonstrated that with the use of gingham or other repetitious patterns binocular cells could be developed.[25] Forrest successfully used the concept in the treatment of a congenital esotrope.[26]

Early diagnosis and treatment is essential for a better prognosis, not only for the alignment but for the prevention of amblyopia. Von Noorden makes reference to the first part of the statement when he says that every child whose eyes are not aligned by 3 months of age should have a complete visual evaluation. He refers to the frequently associated amblyopia when he states that if it is not treated early it will be a "severe obstacle to the return to normal binocular function."[3] Consequently, the treatment of the amblyopia should be timely yet conservative. The optometrist must seek to establish equal monocular systems but not at the expense of the better eye. Here the work of Blakemore[22] and Hubel and Wiesel[23] should be kept in mind, i.e., critical periods.

General motor abilities are acquired in a cephalo-caudal and a proximal-distal manner.[27] The critical period of neural development and growth is from the second trimester of pregnancy through most of the first year of life. During this period neurons and macroglia are being replicated and much of the brain growth takes place.[28] The skull growth during this first year is from 33 cm to 46 cm.[29] During this period the generalized movement controlled by the extrapyramidal tract is replaced by voluntary movement mediated by cortical centers and the pyramidal tract. The palledum receives an inhibitory effect as a result of the development of the corpus striatum. This occurs in conjunction with the cortical and pyramidal tract changes. Thus purposeful and goal-oriented movement gradually replaces gross symmetrical and asymmetrical activity.[30] Large muscle behavior leads to smaller muscle performance.[31] Interhemispheric activity and nerve development occur with the development of activity in the corpus callosum, which serves as the great communicator between the two cerebral hemispheres.[32] One of the developmental milestones which occurs between the 4th and 5th month of life is the grasping for objects within the infant's field of view.[28] This is initially a more reflex style response and is performed one-handed. Mirroring the continuing overall neurological development, this activity will also become more goal-oriented and motor perfected.[34] At this point visual control and the harmonious synchronization of both hands emerges.[33] Bower[34] notes a phasic approach to this motor control and thus an eventual development of reach/grasp/release.

This development is certainly gene-driven but, again, the environment can play a major part. Bower[34] states it this way: "The emergence of new behaviors is very much a matter of gene expression, with the environment operating as accelerator or brake, rather than a switch." Studies of twins have shown that a slight variation of the timing of motor skills can be achieved, but most developmental psychologists believe that motor milestones occur on a programmed schedule. However, they do agree that early experiences are important.[33] When we consider Blakemore's[22] notion of the development of the visual system, we recognize the similarities of process between these two systems.

The next consideration is determining the criteria for cure. Griffin suggests that there are two types of resolutions, i.e., functional and cosmetic.[2] Flom[35] uses the following as a criteria of a functional cure, namely: "... clear, comfortable, single, binocular vision being present at all distances, from the farpoint to a normal NPC (nearpoint of convergence)." Stereopsis and normal ranges of motor fusion are also required and a deviation that is present 1% of the time is acceptable provided that there is awareness of diplopia. Corrective lenses and prism correction no greater than 5^{Δ} are also acceptable.[35] Manley requires a stereo threshold of 67 seconds of arc to be achieved in order to qualify a patient as having bifoveal fusion.[36] Thus on the Titmus Wirt Circles[a] the patient must pass plate number 7.

With this research as a backdrop, the following case report and management scheme is presented.

CASE STUDY

In order to integrate the examination data and the treatment plan an impression statement will frequently follow visits.

Evaluation I

Date 09/30/83, Patient's age (aet) 4 months. (DOB 05/23/83)

The Caucasian female (K.Y.) was referred for an evaluation by another optometrist. Her history included the mother's observation of a left eye that turned in since birth. The mother had a Duane's Syndrome which was surgically treated. Other significant familial ocular history included a great aunt who was esotropic. Obstetrical and pediatric data was unremarkable. The babe was the first child of the family and her developmental milestones were as expected. One other significant aspect of the history was that the infant was breastfed.

An Optokinetic Drum was used in order to assess the infant's visual awareness; however, no response was elicited. Eyegrounds and externals were within normal limits. There were no extra ocular muscle limitations but the patient avoided extreme left gaze. Cover testing revealed a constant 25^{Δ} to 30^{Δ} left esotropia at distance and near. Keratoscopic examination showed the cornea of each eye to be spherical and distortion free. Retinoscopy was +1.50 sph OD and OS with intermittent dulling of the left eye reflex. The deviation was frequently reduced when the child performed gross motor activities, especially playing the game "So Big" with her arms extended.

The diagnosis was a constant left congenital esotropia. The home therapy program was:

1. Eye Movement Development
   Even though there appeared to be no extraocular muscle (EOM) limitations, the familial history of Duane's remained an issue. If the infant continued to avoid left lateral gaze, she would reduce the frequency of action of the left lateral rectus as well as her awareness of the left peripheral field.
2. High Contrast Gingham Pattern Viewing
   The gingham pattern was used to enhance the development of binocularity. The mother was to drape the material arrangement in the crib so that the child's full field of vision would be exposed to the pattern.
3. Crib Placement
   The crib was to be in the center of the room and the child's placement in the crib routinely varied.
4. Nursing
   If nursing were to be discontinued, the child's feeding position with the bottle was to be alternated from side to side. Activities 3 and 4 were prescribed in order to enhance the development of equal monocular visual "circuits" and acuity via equal visual stimuli.
5. Gross Motor Activities
   Activities such as "So Big," "Patty Cake," or raising and lowering both legs at the same time were to be performed. Gross motor activity yields a systemic input and communication to each hemisphere. At this stage the baby should be in a bilateral phase of symmetry, i.e., right-left (RL) as noted by the previous grasp discussion.[37]

The refractive error fell within the expected range for a child of her age. Some authors would not prescribe for this expected level of hyperopia.[38] At this time, New York state had not passed DPA legislation, but were a cycloplegic refraction done, +3.00 D would have been the anticipated finding.[39] Other studies would put this mean finding at +2.00 D.[40] At this time I chose to defer prescribing until the next visit. Because of K.Y.'s excellent subsequent progress the prescription was not needed.

Evaluation II

Date 10/27/83, aet 5 months

The mother now reported that at times the babe's eyes were aligned. Cover testing revealed an intermittent 25^{Δ} left esotropia at distance and near. As before, there were no extraocular muscle limitations. This remained true for the remainder of this case report and no further reference will be made to it. The child displayed only slight head resistance in looking to the extreme left. Retinoscopy was OD +1.75 sph and OS +2.25 sph. The left reflex continued to be duller. The home program remained intact with the addition of direct patching (OD patched) for 15-minute periods four times a day.

Because the deviation persisted, I instituted direct occlusion. In order to preserve the VA of the fixating eye, the occlusion periods were short. I believe that during occlusion therapy the patient should be required to perform active tasks within his or her capabilities. In this case the mother presented stimuli to encourage motilities and did playing which required some eye-hand performance, e.g., "get the toy rabbit's ears." As the patient increases in age, activities such as placing pennies in a cup or pegs in a pegboard, threading, lacing, spearing raisins, etc. can be employed. Sometimes cycloplegics are instilled to created deprivation of the "good eye."[41] Atropine is frequently used in this more passive method; however, I felt that the prolonged action of the drug contraindicated its use for this infant.

Fresnel prisms could be used to neutralize the deviation. However, as stated previously, they will usually reduce VA. Thus, because of the infant's age and VA consideration this was not done. A second factor was the improvement in alignment, as will be documented, by the overall therapy plan employed.

Evaluation III

Date 11/10/83 aet 5 months 2 weeks

Throughout this case the mother routinely returned for progress visits as designated on the previous visit. The patient had a cold this day and was somewhat debilitated. This visit was to determine whether the patching regimen should be altered. The mother's report and the Hirschberg evaluation indicated alignment. Because of this improvement, direct patching was reduced to two 15-minute periods a day.

Evaluation IV

Date 12/08/83 aet 6 months 1 week

The patient's cold had reoccurred. The Optokinetic Drum testing yielded nystagmoid responses to 12 feet in each eye. The Hirschberg indicated orthophoria, and the NPC was to 2 inches. On probing Z axis reach/grasp/release, the child overconverged and lapsed into a 25^{Δ} left esotropia. The home program was to remain intact with the addition of the following:

1. "Give and Take" of an object. This task was designed to help the baby tactually experience the reach/grasp/release (R/G/R) response. At this stage the child learns more from motoric input. Visually, overconvergence represents a malfunctioning of the R/G/R response.
2. Child holds object as it is moved along the Z axis. This task was prescribed to enhance her tactual awareness of moving in and out within her spatial world.
3. Object movement in the Z axis. An example is rolling a ball toward and away from the patient. This task was designed to enhance her projection into her world. The ball served as an extension of herself beyond her grasp.

Each one of these tasks was to be done along the patient's mid-line. The OKN response indicated that the two visual systems were somewhat equal. It was noteworthy that the debilitating aspects of the cold did not seem to impair her binocular progress.

Evaluation V

Date 01/05/84 aet 7 months

The mother reported that the deviation occurred only when the patient was tired and was now less frequent. She noted that K.Y. initially had difficulty with the "Give and Take" game. The mother now added that K.Y.'s second cousin had an intermittent esotropia. Cover tests were ortho at distance and near. The NPC was at two inches. This finding remained essentially unchanged for the remainder of the visits and will not be mentioned again. The patient didn't overconverge. Versions and rotations were essentially accomplished visually, especially in the horizontal meridian. Retinoscopy was +1.50 D, OD and OS. The reflexes were essentially equal. Stereo tests yielded questionable results. The home program remained unchanged with the exception that the gingham pattern viewing was altered to match the patient's age. Here the parent hid behind the pattern and played Peek-a-Boo.

It was understandable that the "Give and Take" game was difficult for K.Y.; her maturational level of grasping was in its elementary stage. Because of her age, the gingham pattern viewing sequence needed to be altered. The overall impression of this visit was that K.Y. continued to progress regarding alignment and binocular responses.

Evaluation VI

Date 02/02/84 aet 8 months

OKN responses were equal with each eye. The patient broke into a left esotropia only on prolonged monocular cover, but regained alignment immediately upon cover removal. Binocularity was probed by using the Keystone Basic Binocular Series plates 6, 9 and 11.[b] Plate number 6 is an elephant presenting a base-out trunk and a base-in tail when the right eye wears the red lens of the red-green glasses. Plate 9 is a duck with base-out demand. Plate 11 has two spheres; one base-out and one base-in. At times youngsters who depend on tactual information more than visual will grasp a base-out stereo target in space and feel nothing. Thus they proceed to move their hand toward the plane of the target until they feel something. Consequently the mismatch of systems occurs. K.Y.'s response was that of the mismatch to a questionable pass. The home program was continued.

Evaluation VII

Date 03/01/84 aet 9 months

The history presented was that the patient was aligned almost all the time and the deviation occurred only very infrequently. The Keystone Basic Binocular Series plates[b] were used for testing. The patient passed the plates, using both a base-in and base-out stimuli.

First she localized the base-out target

by grasping the apparent target in space. When presented a base-in target, she reached into the page. Cover tests were orthophoria at distance and near. Doing a repeated Z axis jump duction, the patient broke once into a 25^{Δ} right esotropia. Patching was discontinued. Gross motor activities, such as crawling and creeping, were to be encouraged, as were three-dimensional tasks done along the patient's mid-line. The balance of the home program remained.

The major finding of this visit was the occurrence of alternation. Previously K.Y. deviated with only the left eye. This turning of the OD on the Z axis jump duction implied relatively equal systems and VA. Consequently, I discontinued the patching regimen. The utilization of gross and fine motor skills and their subsequent hemispheric interplay was continued. These motor tasks are commensurate with expected developmental skills.[37]

Evaluation VIII

Date 05/23/84 aet 1 year

OKN responses were to 20 feet in each eye. Cover tests were ortho at distance, near and in all cardinal positions of gaze. Z axis jump ductions were smooth, equal and aligned. The same stereo tests as on visit VII were passed.

The VA was essentially equal and K.Y. was aligned in all positions of gaze. Alignment was also present when gaze was shifted from one object to another along her Z axis, i.e., in- out.

Evaluation IX

Date 08/29/84 aet 1 year 3 months

The patient was walking. OKN and cover tests remained unchanged. Retinoscopy was +1.00 sph for each eye with equal quality of the reflexes. Stereo testing resulted in passing performances on the Stereofly[a] and the Wirt Animals,[a] A through C. Thus the baby was now able to pass greater demand stereo tests to 100 sec of arc.[a]

Evaluation X

Date 04/25/85 aet 1 year 11 months

The findings were unchanged from visits VIII and IX. In addition K.Y. passed Wirt Circles[a] 1 through 4, indicating 140 sec of arc. She was able to visually complete a Three Piece Form Board[c] and stack a tower of seven one-inch cubes.[d] Her bimanual performance displayed some lead-support, with her right hand being preferred.

The performance level of binocularity had increased over the last few visits. Her visual perceptual skills and bimanual performance were equal to, if not advanced, for her age.[37]

Evaluation XI

Date 5/15/86 aet 2 years 11 months

VA at near using the AO Picture Chart[e] was at least 20/30 in each eye and binocularly. Eyegrounds were unremarkable. Cover tests were ortho at distance and near. Versions and rotations were full and performed without head support. Reach/grasp/release functions along the Z axis were good. The patient now passed all the Wirt Circles, indicating stereo acuity of 40 sec of arc.[a] Refraction was +0.50 sph OU. Near retinoscopy yielded a fast "with" motion in each eye. In this procedure, the patient views a hand-held mask while the examiner does retinoscopy through the eye opening in it.

At this point the patient met Flom's and Manley's criteria for success. The acuity in each eye was equal and she attained a 40 sec of arc stereo demand.[a]

Evaluation XII

Date 06/19/87 aet 4 years

Broken Wheel[e] VA was OD 20/20 and OS 20/25. All other findings were essentially unchanged. Bell retinoscopy was $16''/16\frac{1}{2}''$ in each eye. This test measures the patient's lag of accommodation. Retinoscopy is done at 20" while the patient views a bell which is slowly moved toward her. The examiner notes the first "against" motion and then proceeds to move the bell away. The first "with" motion is then noted, thus the fraction. The expected lag is usually 0.50 D, which it was in K.Y. The patient passed the Pseudoisochromate Plates (PIP).[e]

The PIP testing was done by having the child pretend her finger was a race car which finds the roads on which to drive.

Evaluation XIII

Date 10/07/88 aet 5 years 4 months

Snellen VA was OD 20/25 and OS 20/20. All other findings were basically as in the previous visit. Distance habitual phoria was 3^{Δ} eso and the near habitual was 3^{Δ} exo. Vertical phorias were orthophoria.

At this point the patient had good VA, alignment, and binocularity that met Flom's criteria.

DISCUSSION

The first consideration in managing the infant with esotropia as to classification (infantile, congenital, or infantile-congenital) is in this instance academic. Obviously the mother was not a professional. However, from her own experience she was more expert in evaluating a turn than the average observer. Secondly, in order to avoid redundance, there were some findings which varied slightly from visit to visit and were not repeatedly enumerated. These findings remained relatively the same throughout the case.

The treatment plan considered the general and visual development of the baby. Initially, glasses were not prescribed because the hyperopia measured was within normal limits for the patient's age. However, had she not made the type of progress she did, I would have prescribed by the second visit.

The treatment plan stressed gross movement and the development of the two sides of the body, as well as eye movements and peripheral visual stimulation. The mother was very cooperative and aggressively pursued the home program. Eye movements were also employed to eliminate any question of mechanical muscle limitation and/or congenital muscle involvement. The gingham pattern was used to enhance development of binocular cells during the critical period. Patching was instituted to eliminate the possible development of amblyopia, but not to the detriment of the fixating eye. The patching was discontinued once the patient displayed alternation and achieved alignment most of the time. The concept of R/G/R was at first developed tactually with the expectation that it would then develop on a visual basis in regard to convergence and accommodation. The Z axis games were used in conjunction with the above concept, i.e., in a tactical-motoric aspect in the patient's 3-D spatial world and then on a more visual basis.

CONCLUSION

The history highlights include a congenital esotropic baby whose familial background included various esotropias.

The mechanism was in place for the development of amblyopia as evidenced by the deviation and a difference in retinal retinoscopic reflexes. It should be noted that the mother and child were very cooperative and responsive to the treatment strategies. Maturation is certainly an essential element for success. Thus K.Y.'s treatment plan predisposed her to the full benefit of maturation.

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