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Journal Title: Developmental Medicine and Child Neurology
Volume: Volume 58, Number 2
Publisher: Wiley: 12 months | 2016-02-01, Pages 154-159
Type of Work: Article | Post-print: After Peer Review
Publisher DOI: 10.1111/dmcn.12832
Permanent URL: https://pid.emory.edu/ark:/25593/rn95p

Final published version: http://dx.doi.org/10.1111/dmcn.12832

HHS Public Access
Author manuscript
Dev Med Child Neurol. Author manuscript; available in PMC 2016 May 17.

Published in final edited form as:
Dev Med Child Neurol. 2016 February ; 58(2): 154–159. doi:10.1111/dmcn.12832.

Motor Skills of Children with Unilateral Visual Impairment in the Infant Aphakia Treatment Study

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Abstract

Aim—To assess motor functioning in 4.5 year olds enrolled in the Infant Aphakia Treatment Study, and to determine contributions of visual acuity and stereopsis to measured motor skills.

Method—Children with unilateral aphakia randomized to intraocular lens (IOL) or contact lens (CL) treatment were evaluated at 4.5 years for monocular recognition visual acuity, motor skills, and stereopsis by a traveling examiner masked to treatment condition. Motor skills were assessed with the Movement ABC-2. Visual acuity was operationalized as logMAR value for treated eye, best logMAR value for either eye, and intraocular logMAR difference.

Results—T-tests showed no significant differences in MABC-2 scores between the IOL and CL groups. The mean total score was low (6.43; 18th percentile) compared to the normative reference group. Motor functioning was not related to visual acuity in the treated eye or to intraocular logMAR difference, but was predicted in a regression model by the better visual acuity of either eye (usually the fellow eye), even after accounting for the influence of age at surgery, examiner, orthotropic ocular alignment, and stereopsis.

Interpretation—Children with unilateral congenital cataract may have delayed motor functioning at 4.5 years, which may adversely affect their social and academic functioning.

Although unilateral visual impairment (UVI) is a common pediatric vision disorder, its functional significance is not fully understood[1]. Hyperopic refractive errors identified at infant screening are associated with poor visuomotor test performance in early childhood[2], but it is unclear to what extent UVI affects children's motor development. UVI and associated abnormalities of binocular depth perception result from a number of conditions, including eye misalignment (strabismus), refractive imbalance (anisometropia), or image deprivation (e.g., from cataract). Treatment of these conditions typically includes occlusion of the eye with better vision during a sensitive period for visual development, which coincides with the stage during which children develop and integrate key motor skills,

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including reaching/grasping and postural stability and control. Therefore, children with UVI may be at risk for delayed development of motor skills due to poor vision, lack of binocular function, and/or to lack of practice in ambulating or manipulating objects.

Children with UVI resulting in amblyopia generally perform worse on tests of motor skills than age-matched children with normal vision, particularly on fine motor tasks that require speed and dexterity[1, 3–5]. However, only some of these studies used standardized, norm-referenced tests to assess motor skills, and none examined multiple domains of motor functioning. Most of this research assesses only fine motor skills or eye-hand coordination. Relatively few studies measured postural control/stability or gross motor skills of children with UVI. Arguably, UVI and associated deficits in binocular vision might interfere in performance of tasks assessing balance, posture, and gait, as these activities require integration of vestibular signals relating to head or body position/motion with proprioceptive information and visual perception of the environment. There are no published reports of motor skills in children with UVI secondary to unilateral congenital cataract.

It is unclear to what extent the relatively poor motor test performance of children with UVI is due to poor visual acuity in the affected eye, to reduced binocular vision, or both. Multiple regression analyses suggest that strabismus[5] or reduced stereovision[4] has a greater adverse impact on fine motor performance than the degree to which the two eyes differ in acuity among children with amblyopia. However, the relative importance of stereopsis versus degree of amblyopia may vary according to the child's age[3], given developmental changes in reach-to-grasp strategies[6].

The Movement Assessment Battery for Children – 2nd edition (MABC-2[7]), a standardized, norm-referenced assessment of children's everyday motor competence, includes eight tasks assessing three domains: manual dexterity, ball skills, and static and dynamic balance. The primary aim of this study was to assess motor functioning using the MABC-2 in 4.5 year old children enrolled in the Infant Aphakia Treatment Study (IATS), a randomized, multicenter (n = 12) trial comparing treatment with a primary intraocular lens (IOL) or contact lens (CL) in 114 infants with surgery for unilateral cataract extraction between 1 and 7 months of age. Key research questions include the following: (a) Are there significant differences in motor functioning between the treatment groups? (b) How does the motor functioning of the IATS sample compare to that of the MABC-2 standardization sample? (c) To what extent are motor skills predicted by visual acuity and/or stereopsis? Multivariate analyses control for earlier age of surgery and the absence of strabismus, as these variables have been associated with better stereoacuity in previous studies (8, 9).

METHOD

Information about the overall design and clinical measures of the IATS has previously been published[10]. Briefly, the IATS is a randomized trial comparing two treatments for 114 children with unilateral congenital cataract: early removal of the cataractous lens followed by contact lens correction of aphakia versus IOL implantation at the time of lens extraction. Randomization was stratified for clinical center (three groups based on the surgeon's experience in implanting IOLs in infants) and patient age (28 to 48 days versus 49 to 209 days) after caregivers gave informed consent. Children with an IOL were prescribed spectacles for residual refractive errors of > 1 diopter (D) hyperopia, >3 D myopia or > 1.5 D astigmatism. Aphakic children were prescribed a contact lens to be worn when awake. Aphakic children's refractive status was assessed at regular intervals, and they were prescribed new contact lenses when their refractive errors changed. Starting at age two years, all children were corrected for both near and far vision; they were provided with bifocal correction of 2.5 diopters. Patching of the phakic eye was prescribed for all patients: 1 hour/day per month of age until the child was 8 months old, and thereafter for 50% of the child's waking hours. Children were not restricted from any physical or motor activities. The study was approved by the institutional review boards of all participating institutions and was in accordance with the tenets of the Declaration of Helsinki.

During the 4.5 year visit (mean, 4.5 years; range, 4.5–4.9 years), monocular recognition visual acuity and monocular fixation stability were evaluated, followed by testing of motor skills, and then assessment of stereopsis[11]. Motor skills were assessed while the child had optimal visual correction and was not patched. All assessments were conducted by one of two trained traveling testers masked to treatment condition; the primary tester examined 79% of the patients.

Assessment Procedures

Monocular recognition acuity was assessed using the Amblyopia Treatment Study HOTV test[12]. Patients were tested wearing their best correction (updated 3 months earlier). Visual acuity testing was standardized using a video monitor for stimulus display and a computer algorithm for the sequence of presentation. Single optotype stimuli with surround bars were presented on the video monitor, initially set at a distance of 3 meters. The child was not required to name the letters, but could identify them using a matching card. If the child was unable to see the HOTV letters, this distance was decreased to 1 meter. If the child still could not identify the letters, the Low Vision Card (Teller Acuity Card, 0.32 cy/cm) was used to differentiate among some pattern vision, light perception, or no light perception following standard protocols. This procedure yielded logMAR values for the treated eye and the fellow eye. Binocular alignment was assessed to measure the full magnitude of strabismus, which includes the manifest tropia and any latent deviation. Children were categorized as orthotropic or strabismic.

Motor skills were assessed with age band 1 of the MABC-2[7], a norm-referenced performance test designed to evaluate gross and fine motor functioning in children aged 3:0–16:11 years. The test comprises eight tasks assessing three domains and takes about 30 minutes to complete. Standard scores (mean=10, standard deviation=3) are provided for individual tasks and for composite scores based on respondent age. The instrument has adequate test-retest reliability for composite scores[7] and for most tasks for age band 1[13]. The validity of the MABC-2 is supported by its relationship with other broad-based movement tests[7], and by confirmatory factor analyses[14].

Traveling testers completed training in MABC-2 administration prior to data collection, and the IATS Vision Scientist (secondary tester) accompanied the traveling tester to the first 4.5 year visit at each site to ensure that the testing environment was adequate and consistent with test developer recommendations. To determine validity and reliability of MABC-2 procedures and scoring, 34 test administrations from both testers and 11 of 12 sites were videotaped and reviewed (*n*=32) or observed live (*n*=2) by the IATS psychologist. Videotaped MABC-2 administrations were rated for fidelity to procedure and scoring according to a 45-item form developed by the authors. Items were scored as present or absent, yielding a total score ranging from 0 to 100%.

Stereopsis was evaluated using the Frisby Stereotest (Richmond Products, New Mexico), which consists of a series of plates with four square areas composed of randomly placed triangular shapes. On one of the squares the triangular shapes create a circle that can be detected only if the patient has some degree of stereopsis. The plates vary in thickness and are viewed without special glasses. Children were initially tested with a “practice” plate (6 mm), with the viewing distance from 20 to 40 cm. If the child correctly identified the circular pattern on 3 out of 4 presentations with the 6 mm plate, the same procedure was repeated with the 3 mm plate and then the 1.5 mm plate. If the child was unable to detect the circular pattern on the 6 mm plate at 40 cm, the child was tested at 30 cm. If the child was unable to detect the circular pattern on the 6 mm plate at 30 cm, the test was stopped. This procedure yielded a level of stereopsis that was dichotomized as present or absent.

The data were tested for normality by computing z-scores for skewness and kurtosis. T-tests were conducted (two sided) to test for differences in demographic variables and motor skills between the two treatment groups, and to test for differences in MABC-2 scores between examiners. The visual acuities were compared between the treatment groups using the nonparametric Mann-Whitney U Test because of the skewed distribution of the data and because of the assignment of visual acuity values for patients with low vision (11). Nonparametric statistical tests (Spearman’s rho) were also used to assess the relationships between MABC-2 summary scores and vision variables. Regression analyses were conducted to evaluate the potentially unique and combined contribution of the five independent variables (age at surgery, examiner, orthotropic ocular alignment, minimum logMAR visual acuity, stereopsis) to each of the four MABC-2 summary scores. Statistical significance was defined as alpha < 0.05.

RESULTS

The baseline clinical characteristics of the patients in the IATS have been previously published[15]. Of the 114 patients enrolled (Figure 1), 112 had vision testing and 113 received MABC-2 testing at the 4.5 year visit. For three cases, MABC-2 testing was not complete, and six additional cases were removed from the analyses because these patients had non-ocular conditions that might affect motor functioning (1 with cochlear implants, 3 with developmental delay, 1 with foot braces, and 1 with Stickler’s syndrome). Of the remaining sample of 104 children, 51 had been randomized to the CL group and 53 to the IOL group.

The sample was 53% female, 87% Caucasian, with about a third reporting that they received Medicaid. The majority of caregivers (74% mothers, 70% fathers) reported that they had vocational training, some college, or a college or graduate school degree. Children underwent cataract surgery at 28 days to 6.83 months of age (mean = 2.51 months, SD=1.64). Visual acuity in the treated eye at 4.5 years ranged from −0.22 (20/12) to 2.93 (worse than 20/2400) logMAR (mean=1.03, median=0.85, SD=.81); 48% had vision 20/200 or worse. Visual acuity in the fellow eye ranged from −0.22 (20/12) to 0.70 (20/100) logMAR (mean=0.10, median = 0.10, SD=.15); 88% had acuity better than 20/40. For six children, there was no difference in visual acuity between treated and fellow eyes; two children had better visual acuity in the treated eye than in the fellow eye (logMAR differences of −0.19 and −0.20). The mean logMAR intraocular difference (treated eye minus fellow eye) was 0.92 (median difference = 0.70, range = −0.20 to 2.93).

There were no significant differences between the IOL and CL groups in patient age at surgery, gender, race, mother's education, father's education, Medicaid status, or visual acuity of either the affected or non-affected eye. There were also no significant differences between the two groups in MABC-2 Total or Domain standard scores (Table 1).

Given the absence of group differences, the two groups were collapsed for subsequent analyses. MABC-2 scores were low compared to the normative reference group (Table 2). The mean Total Standard score of 6.43 and median of 6.0 correspond to the 18th percentile; 40% obtained scores at or below the 5th percentile. Mean scores on the MABC-2 domains were also low, corresponding to the 22nd percentile (Manual Dexterity), 31st percentile (Aiming and Catching), and 22nd percentile (Balance). The distributions of domain standard scores indicate that these relatively low mean values are not due solely to clustering of scores at the low end.

Patients assessed by the primary tester had significantly higher scores than those assessed by the secondary tester on the Manual Dexterity domain (t=2.00, p=.048) and the Aiming and Catching domain (t=2.43, p=.017), but not on the MABC-2 Total score (t=1.76, p=.081). This difference is unlikely to be contaminated by time, as the secondary tester assessed the first 12 patients in 2009, one in 2010, and 9 in 2012 to 2013, while the primary tester assessed patients in 2010, 2011, and 2012. Fidelity scores for the MABC-2 ranged from 81% to 100% for test administration (38 items; mean=96.7%, SD=3.89), 71% to 100% for scoring (7 items; mean=89.7%, SD=10.6), and 79% to 100% (45 items; mean=95.7%, SD=4.3) for both combined. While there was no significant difference between the two examiners in fidelity for scoring, there was a significant difference for test administration (t=2.061, p=.049).

Performance on the MABC-2 was not related to visual acuity in the treated eye or to intraocular difference in logMAR values (Table 3). Better MABC-2 Total scores were associated only with better visual acuity (minimum logMAR value for either eye); Aiming & Catching domain scores were significantly associated with better visual acuity, stereopsis, and examiner.

The regression model for MABC-2 total score was significant (F=3.097, p =.013), accounting for 14% of the variance; however, only visual acuity uniquely accounted for measured motor skills (beta = −.288, p=.004). Similar findings were obtained for the Manual Dexterity domain (12% variance), with visual acuity again contributing unique variance (beta=0.231, p=.021). For the Aiming and Catching domain, the overall model was significant, accounting for 17% of the variance; however, both examiner and visual acuity contributed significantly to the prediction of MABC-2 scores. For the Balance domain, the overall model was not significant, accounting for only 7% of the variance in MABC-2 scores. However, visual acuity was the sole measured factor responsible for higher levels of motor skills.

DISCUSSION

Motor skills, as assessed by the MABC-2, were generally low (18th percentile) for the IATS sample at 4.5 years of age, with no significant differences between those treated with an IOL versus those treated with CL. Forty percent obtained MABC-2 total scores at or below the 5th percentile, denoting a significant movement difficulty[7]. While children performed somewhat better on aiming and catching tasks than they did on manual dexterity tasks or balance tasks, analyses of 3 to 6 year olds' MABC-2 scores do not provide sufficient evidence for three domain-specific factors, suggesting that only the total score should be interpreted[14]. To our knowledge this is the first report on the motor functioning of preschool children treated for unilateral congenital cataract. As motor impairments among 7 to 9 year olds may be associated with increased difficulties with reading and spelling[16], our results raise questions about the school readiness of preschool children treated for unilateral congenital cataract.

It is unclear why the motor functioning of our sample was comparatively low. Performance on the MABC-2 was not related to visual acuity in the treated eye, but was associated with the better visual acuity of either eye (usually the fellow eye), even after accounting for the influence of age at surgery, examiner, ocular alignment, and stereopsis. In contrast to findings from other studies[5,17] stereopsis did not contribute uniquely to the prediction of measured motor skills. However, this discrepancy may be due to sampling differences in child age, as binocularity may confer increasing benefits for eye-hand coordination as children mature (3). In addition, only 11% of our sample had stereopsis, in comparison to the more even distributions of stereopsis reported for children with amblyopia[5,17]. Further, stereopsis may not be critical for successful performance on some MABC-2 tasks given the presence of monocular clues.

As the majority of the sample (88%) had vision better than 20/40 in the fellow eye, poor visual acuity alone cannot account for the overall low MABC-2 scores. Unmeasured factors such as intellectual ability, motor experience, and confidence in motor skills may have contributed to MABC-2 performance. Participants' motor functioning may have been delayed due to avoidance of physical risk-taking or lack of motor skill practice during times when they were patched, which was prescribed 50% of waking hours from 8 months and a significant portion of time in infancy. While it is not clear that our standard patching regimens[8,18] contributed to delayed motor functioning in the IATS sample, Yang and Lambert[19] found iatrogenic effects, including delayed fine and gross motor skills, of prolonged occlusion therapy for five children with UVI secondary to structural abnormalities. As all of the IATS children had at least 2 general anesthesias before 14 months, it is also possible that their lower than expected scores on the MABC-2 are due to the neurotoxic effects of general anesthesia[20].

The strengths of this study include the use of a norm-referenced, standardized instrument to measure three domains of motor functioning for a well characterized sample, a traveling tester masked to treatment group, procedures to assess fidelity of MABC-2 administration and scoring, and analyses that controlled for the effect of co-varying and confounding variables. The primary limitation is that only one aspect of binocular functioning was assessed, with only 11% assessed to have stereopsis. However, only a minority of children treated for unilateral congenital cataract achieve stereopsis[21]. Second, the tester was not masked to visual acuity, which was assessed before motor skills. However, the possible effect of known visual acuity on motor test outcomes is mitigated in part by the fact that the IATS psychologist, masked to visual acuity, scored the Drawing Trail task and computed standard scores. Third, findings from the regression analyses should be interpreted with caution as logMAR acuities were not normally distributed. Finally, assessment of visual acuity with both eyes open may be a more valid assessment of visual acuity related to motor functioning.

Our data suggest that children with UVI secondary to unilateral congenital cataract may have delayed motor functioning across the areas of fine motor, ball skills, and balance during the year before they enter kindergarten. Delayed motor functioning may have implications for children’s academic performance as they progress through elementary school[16]. In addition, the importance of motor competence to children’s social interaction with peers has been well documented[16, 22]. Children with delayed motor skills and UVI should be referred for evaluation by physical and occupational therapists to determine if therapeutic intervention is indicated, consistent with recommended guidelines for evaluating motor delays[23]. Future research should assess IATS patients’ motor functioning at a later age to determine whether children with unilateral congenital cataract “catch up” with their peers or continue to experience delayed motor skills.

Acknowledgments

The Infant Aphakia Treatment Study Group

Funding Source: All phases of this study were supported by a grant from the National Eye Institute (# U10 EY013287 and U10 EY13272).

Clinical Trial Registration: Infant Aphakia Treatment Study (IATS) (NCT00212134) in ClinicalTrials.gov

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What this Paper Adds

- Children treated for unilateral congenital cataract had delayed motor functioning at 4.5 years.
- There were no differences in motor functioning between children randomized to intraocular lens versus contact lens treatment.
- Motor test performance was associated with best monocular visual acuity.
- Visual acuity uniquely accounted for measured motor skills after controlling for age at surgery, examiner, orthotropic ocular alignment, and stereopsis.

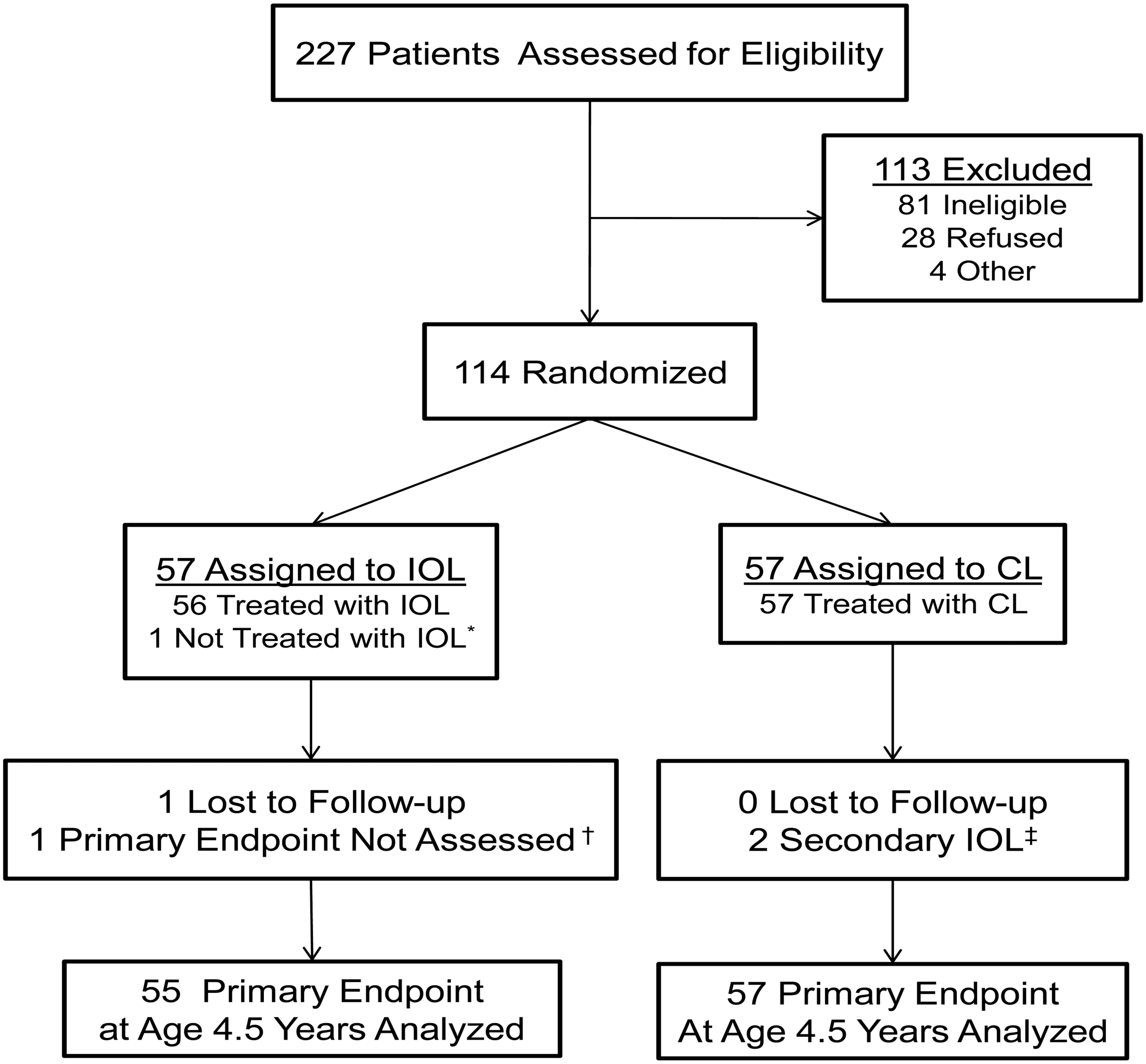

Figure 1.
Consort Diagram for IATS

Table 1

MABC-2 Domain and Total Standard Scores by Treatment Group

Test	CL (*n*=51) Mean (SD)	IOL (*n*=53) Mean (SD)
Manual Dexterity	6.88 (2.81)	6.75 (2.81)
Aiming & Catching	8.16 (2.36)	8.17 (2.82)
Balance	6.96 (2.48)	7.42 (2.55)
Total MABC-2 Score	6.35 (2.58)	6.51 (2.49)

CL, contact lens; IOL, intraocular lens; MABC-2, Movement Assessment Battery for Children (2nd ed.). $P > .05$ for t-test between treatment groups for all MABC-2 scores.

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Table 2

MABC-2 Standard Scores, Visual Acuity, Stereopsis, and Strabismus

Variable	Mean (SD) or %, Range	
MABC-2 Manual Dexterity domain	6.82 (2.80), 2 to 13	
Posting Coins	7.56 (2.47), 1 to 14	
Threading Beads	8.04 (2.82), 1 to 13	
Drawing Trails	5.63 (4.40), 1 to 13	
MABC-2 Aiming & Catching domain	8.16 (2.59), 3 to 15	
Catching Beanbag	7.49 (2.72), 3 to 16	
Throwing Beanbag onto Mat	8.65 (2.48), 6 to 14	
MABC-2 Balance domain	7.19 (2.51), 2 to 16	
One Leg Balance	6.74 (2.20), 3 to 14	
Walking Heels Raised	6.79 (3.63), 3 to 13	
Jumping on Mats	9.82 (3.54), 1 to 12	
MABC-2 Total Score	6.43 (2.52), 1 to 14	
Minimum logMAR of either eye	0.10 (0.14), −0.22 to 0.70	
VA category treated eye	Better than 20/40	18%
	20/40 – <20/80	22%
	20/80 – <20/200	12%
	20/200 or worse	48%
VA category fellow eye[c]	Better than 20/40	88%
	20/40 – <20/80d	10%
	20/80 – <20/200	2%
Frisby Stereotest[a]	11% with stereopsis	
Ocular alignment[b]	27% orthotropic	

MABC-2, Movement Assessment Battery for Children (2nd ed.); VA, visual acuity; logMAR, log base 10 of the Minimum Angle of Resolution (used to calculate population means)

[a] Assessed for 100 participants.

[b] Assessed for 102 participants.

[c] Two children had better logMAR values for the treated eye than the fellow eye (difference = 0.20 logMAR).

[d] 7 had 20/40 vision.

Table 3

Spearman's rho correlations between MABC-2 Scores and Independent Variables

	Manual Dexterity	Aiming & Catching	Balance	MABC-2 Total Score
logMAR of treated eye	−.08	.05	−.04	−.03
Intraocular logMAR difference[a]	−.06	.09	−.00	−.00
Minimum logMAR of either eye	−.17	−.29[**]	−.19	−.24[*]
Stereopsis	.18	.20[*]	.05	.19
Orthotropic alignment	−.05	.17	.04	.06
Age at surgery	−.10	−.17	−.13	−.17
Examiner	−.20[*]	−.26[**]	−.02	−.18

MABC-2, Movement Assessment Battery for Children (2nd ed.); logMAR, log base 10 of the Minimum Angle of Resolution.

[a]logMAR of treated eye minus logMAR of fellow eye.

[*] $p<.05$.

[**] $p<.01$.