Original paper



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Infant vision screening predicts failures on motor and cognitive tests up to school age

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Abstract In a population-based infant vision screening programme, 5295 infants were screened and those with significant refractive errors were followed up. To assess the relationship between the development of vision and other domains, we report a longitudinal study comparing infants with significant hyperopia, identified at age 9 months ('hyperopes') with infants with normal refractions ('controls'). Children are included who completed at each age a broad set of visual, cognitive, motor and language measures taken over a series of follow-up visits up to age 5.5 years. Hyperopes performed significantly worse than controls on the Atkinson Battery of Child Development for Examining Functional Vision at 14 months and 3.5 years and the Henderson Movement Assessment Battery for Children at 3.5 and 5.5 years. The Griffiths Child Development Scales, MacArthur Communicative Development Inventory and British Picture Vocabulary Scales showed no significant differences. Exclusion of those infants who became amblyopic and strabismic did not substantially alter these results, suggesting that the differences between groups were not a consequence of these disorders. These results indicate that early hyperopia is associated with a range of developmental deficits that persist at least to age 5.5 years. These effects are concentrated in visuocognitive and visuomotor domains rather than the linguistic domain.

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Key words Infant vision; visual development; vision screening; hyperopia; developmental delay

Introduction The First Cambridge Infant Vision Screening Programme,¹⁻³ using cycloplegic photorefraction confirmed by retinoscopy, found that infants with a significant hyperopic (long-sighted) refractive error identified at age 9 months had poorer visual outcome at 4 years than children who were refractively normal as infants. Observations in the course of this programme suggested that this outcome might be associated with a broader range of developmental delays or deficits. Therefore, in a second screening programme^{3,4} with a new total population of infants from the same Health District, we assessed visual outcome and the relation between early vision and possible developmental delay. A large number of visual, cognitive and motor measures were taken over a series of follow-up visits up to age 5.5 years. In the present paper we present a comparison of infants with significant hyperopia and refractively normal controls using such measures, taken from standard developmental tests - the Atkinson Battery of Child Development for Examining Functional Vision (ABCDEFV),⁵⁻⁸ The Movement Assessment Battery for Children (Movement ABC)9 and the Griffiths Child Development Scales.¹⁰ The ABCDEFV was administered at 14 months, 2 years and 3.5 years; the Movement ABC and Griffiths at 3.5 and 5.5 years. Vocabulary tests at 2 years and 5.5 years were also included. These tests were chosen to be appropriate at each age.

Methods

SUBJECTS

Screening All infants in the Cambridge Health District were invited for vision screening at age 8 months in well-baby clinics. Videorefraction or photorefraction¹ is a method of rapidly obtaining a measurement of the subject's plane of focus, from a defocused video image of the eyes, making it suitable for use with passive or uncooperative subjects such as infants and very young children. Of all those invited, 5142 infants (76%) were screened (at average age 8.07 months, s.d. 0.79) using non-cycloplegic videorefraction between 1993 and 1995.34 From this population, the potentially hyperopic group was identified as any infant showing a hyperopic error of focus greater than or equal to +1.5D in any axis on non-cycloplegic videorefraction. Refractions were confirmed by cycloplegic retinoscopy at the first follow-up visit, at average age 9.3 months (s.d. 0.85 months). The 260 infants with at least one axis greater than or equal to +3.5D on cycloplegic retinoscopy were considered hyperopic and invited to attend further follow-up visits. A control group was recruited from the same clinics; these were children who, at non-cycloplegic screening, showed no error of focus equal or greater than +1.5D or -3D in any axis, and were confirmed by cycloplegic retinoscopy as having all axes <3.5D and >-2D; 267 such children were included in the study. The parents of these children gave informed consent for participation in the study. Those with strabismus,

1.5D or more of anisometropia or any axis of +6D or more were referred to a hospital ophthalmological outpatient clinic for appropriate treatment.

Subjects in the present analysis We report here two forms of the analysis. First, we consider only those infants who attended and completed testing at all follow-ups, i.e. at ages 14 months, 2 years, 3.5 years and 5.5 years, a total of over 75 measures. These were 71 (31 male + 40 female) hyperopes and 66 (29 male + 37 female) controls. The reduced numbers are almost entirely due to children who failed to attend at a particular age, not to failure to complete testing among those who attended at a particular visit. In many cases, children who did not attend at a particular age nonetheless attended a later session.

Within this group, the mean value of the greatest axis of the hyperopic group at age 9 months was +5.2D (s.d. 1.4D), while for the control group the mean was +1.9D (s.d. 0.7D). Figure 1 is a histogram showing the distribution of children's refractions (value of most hyperopic axis) in the longitudinal hyperopic and control groups. Note that this should not be treated as a single continuous distribution since the two groups had specific selection criteria, giving the distribution for each group a sharp cut-off. The distribution within each group was closely representative of the distributions of refraction in the groups initially designated by cycloplegic retinoscopy at the first follow-up. Because screening was non-cycloplegic, we do not know the full distribution of refractions in the screened population; however, the proportion in the hyperopic groups is consistent with that found in our earlier study which screened an entire infant population from the same Health District using photorefraction under cycloplegia.^{1,3} The average cycloplegic refraction at follow-up from the hyperopic group and the control group is also consistent between the two studies.

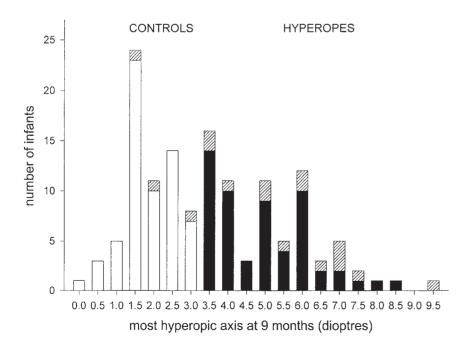


Fig. 1. Distribution of refractions (most hyperopic axis) in the control group (white bars) and hyperopic group (black bars). Children who became strabismic or amblyopic in the course of the study are indicated by cross-hatched parts of the bars. Note that the two groups do not represent a continuous population because (a) the controls are a sample from a much larger group of screened children; (b) the cut-off at 3.5D was used to select these groups from the screened population.

We refer to these groups of children who attended all follow-ups as the 'longitudinal groups'. Analysis of these groups has the benefit that all results relate to the same group of children with uniform selection criteria. It could, however, be argued that the requirement to participate in all follow-ups might introduce some selection bias, and a larger number of children can be included if this requirement is relaxed. We have therefore carried out a second, cross-sectional, analysis in which each test at a given age is compared for all hyperopes and controls who participated in the relevant follow-up. For this analysis, the number of children differs at each age, and is indicated in the two bottom rows of Table I.

Strabismus and amblyopia Fourteen of the 71 hyperopes developed strabismus over the course of the study; 7 of these were also amblyopic

| Test | Function tested | 14mo | 2 yr | 3.5 yr | 5.5 yr |
|---|------------------------------------|------------|------------|------------|------------|
| Tests from ABCDEFV | | | | | |
| retrieve totally & partially covered object; pick up black/white cotton; pincer grasp with cotton | visuo-cognitive & spatial skills | * | | | |
| stack 2+ blocks | visuo-spatial & motor skills | * | | | |
| <pre>shape matching; recognise embedded animal figures[13]; find hidden cats[13]; insert card in envelope</pre> | visuo-cognitive & spatial skills | | | * | |
| copy block constructions ^a | spatial cognition, motor skills | | | | |
| MOVEMENT ABC | | | | | |
| <i>manual dexterity:</i> post coins in slot (each hand); thread 6 beads; follow trail with pen. <i>static & dynamic balance:</i> balance on either leg; jump over cord; walk line with heels raised; <i>ball skills:</i> catch beanbags; roll balls into goal | visuo-motor skills | | | * | * |
| GRIFFITHS CHILD DEVELOPMENT SCALE eye/hand co-ordination performance ^{a,b} practical reasoning | S | | | | * |
| personal-social hearing & speech | | | | | |
| VOCABULARY TESTS | | | _ | | |
| MacArthur Communicative Development Invento British Picture Vocabulary Scales | ry (British adaptation) | | | | |
| number of children from hyperopic group participating number of children from control group participating | | 199 178 | 150 142 | 110 131 | 105 115 |

 \Box = test performed, no significant difference between groups; * = hyperopic group performed significantly worse than controls. See text for statistical procedures and results of individual subtests.

^adifferent, age-appropriate versions used at each age tested.

TABLE I. Tests used at the four

testing ages, with results of the analysis of the longitudinal group.

not carried out at this age.

Blank cell: test not appropriate or

^bbead threading omitted from Griffiths due to similarity with Movement ABC item.

190 J. Atkinson et al.

(defined as acuity differing by a factor of 2 or more between the eyes, e.g. 6/9 LE, 6/18 RE, on Cambridge Crowding Cards⁹) at 5.5 years. A further 3 were amblyopic but not strabismic. Of the 66 in the control group, 3 developed strabismus; none were amblyopic at 5.5 years. The refractions of the strabismic children are indicated in the histogram of Figure 1. A supplementary analysis was carried out to determine whether any of the differences found between hyperopes and controls were due to the higher incidence of strabismus and amblyopia in the hyperopic group. Amblyopic and strabismic children were taken out of the groups and the remaining controls and hyperopes were compared as before.

DEVELOPMENTAL TESTS Table I summarises the tests analysed and the ages at which they were administered. The ABCDEFV^{5,6} consists of two parts: a set of core vision tests (results reported elsewhere)²⁸ and a set of additional tests to assess various aspects of perceptual, motor and cognitive vision, including the copying of age-appropriate block constructions. Table 2 briefly describes the latter tests analysed in this paper for each age and the 'pass' criterion used in the analysis of the data. The Movement Assessment Battery for Children¹⁰ provides an assessment of the everyday motor competence of children between the ages of 4 and 12 years. Most of the tasks were appropriate for the 3.5 year-olds, although the balance tests were found to be rather difficult for children of this age. Due to the length of the battery only one attempt was given on each test. The Griffiths Child Development Scales¹¹ are widely used in paediatrics as a set of tasks to measure various aspects of general development between the ages of o and 8 years. Each scale comprises 6 tasks. The MacArthur Communicative Development Inventory¹² (adapted for British English in our unit) is a checklist of vocabulary by parental report, while the British Picture Vocabulary Scale¹³ tests vocabulary by asking children to choose the picture, from a selection of four, that matches a word. These vocabulary tests were selected as appropriate for specific age groups: the BPVS cannot be used below 3.5 years, and so the MCDI was used at the younger ages.

By 3.5 years, children who still had a significant refractive error were referred for prescription of a suitable spectacle correction, which was therefore worn as necessary for testing at 3.5 and 5.5 years.

STATISTICAL METHODS Each of the tests analysed here from the ABCDEFV battery at ages 14 months, 2 years and 3.5 years were scored as pass (1) or fail (0) by the criteria listed in Table 2. An individual subject's scores at a given age were summed to give two totals: one for block copying tasks and another for the rest, since the blocks constituted a different proportion of the battery at different ages. The hyperopic and control groups were compared on both blocks and non-blocks test totals at each age using the Mann-Whitney U test. Non-parametric statistics were used because these tests are designed for the majority of children to gain the passing score, leading to highly skewed distributions. At 5.5 years performance on block designs was measured by the time taken to complete them (the ability to copy the designs was

| Age used | Test | Pass criterion |
|---------------|---|--|
| 14 months | retrieve partially covered object | child sees cloth placed to partially cover small toy: retrieves toy on first trial or on 2/3 trials |
| | retrieve totally covered object | child sees cloth placed to completely cover small toy: retrieves toy on first trial or on 2/3 trials |
| | pick up black/white cotton | child attempts to pick up (any form of grasp) 10cm lengths of both black and white cotton from light coloured table. |
| | pincer grasp for cotton | child uses pincer grasp opposing finger to thumb, in test above |
| | stack blocks | given blocks on table top, child spontaneously stacks at least two. |
| 2–3.5 years | shape matching: shape sorter board with templates for square, circle, triangle, rectangle, six-pointed star | fit 4/5 shapes, presented individually, into correct template |
| | embedded animal figures (card with five overlapping outline drawings of animals) | child asked to point out animals, finds 3/5 |
| | find hidden cats (4 'cat' figures defined by areas of vertical shading in shaded tree drawing) | child, shown sample cat figure and asked to point out those hidden, finds 2/4 |
| | insert card in rectangular envelope | child aligns long axis of card with long opening of envelope, without trial and error. |
| 2 years | copy block constructions | child achieves partially successful copies of: eight-block stack, eight-block row, joined stack and row (4 blocks each) |
| 3.5–5.5 years | copy block constructions | child achieves partially successful copies of 2 year old test plus:, pair of stacks, four sided enclosure, bridge (two horizontals on top of three verticals), cross of eight blocks |

TABLE 2. Component tests of the ABCDEFV battery used at each age in the present study.

no longer an issue at this age). Controls' and hyperopes' mean total times to complete all three constructions were compared using the Mann-Whitney U test.

Movement ABC tests were analysed both individually and as combined scores. The raw scores of the two groups were compared on each individual test using the Mann-Whitney U statistic. To obtain combined scores, the raw data have to be transformed to a common scale, since the tests involved a variety of different measures (e.g., number of catches, time in seconds). The common scale was a z-score based on the mean and s.d. of the control group scores, which were used to transform both the control group and the hyperopic group scores. The summary measure for each subject was then the mean of their z-scores, with the two separate coin and one-leg balance scores weighted as half a test each. This method was used in preference to calculating normalised Movement ABC impairment scores¹⁰ since no normalisation has been previously published for children as young as 3.5 years. This mean score was compared between the control and the hyperopic groups using two-tailed U tests.

The published Movement ABC normalisation¹⁰ was used in a secondary analysis to provide a measure of the number of children with clinically significant levels of impairment in each group. It must be remembered that this normalisation applies to a testing procedure in which more than one attempt is given on several of the tests, and is not intended for children under 4 years; therefore a higher than average number of children with deficits on this measure may be expected in both groups.

Each of the Griffiths tests was scored as pass (I) or fail (0) according to the test's published age norms; these were summed to give totals for the five individual scales administered at 3.5 years, a total for all scales together at 3.5 years, and a total for the single scale administered at 5.5 years. Hyperopes and controls were compared using the Mann-Whitney U test.

Mean numbers of reported words on the MCDI vocabulary checklist and standardised BPVS scores were compared for the control and hyperopic groups using Mann-Whitney U-tests.

INDIVIDUAL IMPAIRMENT The differences between groups were examined in terms of the number of subjects in each group that gained very low scores, indicating a clinically significant impairment or deficit. This issue is addressed in the Discussion.

Results

COMPARISON OF HYPEROPES AND CONTROLS ON THE ABCDEFV (LONGITUDINAL GROUPS) On the subtests of the ABCDEFV, excluding copying block constructions, controls were significantly better than hyperopes at 14 months (Mann-Whitney U = 1915, p < 0.02) and 3.5 years (Mann-Whitney U = 2080, p < 0.02): 85% of controls scored full marks (3/3) compared with 66% of hyperopes – in other words, hyperopes were more than twice as likely as controls to fail one or more of the tests (33% compared with 15%). At the older ages, this difference was in the same direction but did not reach significance. Full marks were not expected on the tests administered at 2 years, since some of the tests included are passed, on average, at a later age -83%of controls scored 2/4 or higher, compared with 76% of hyperopes. At age 3.5, 98% of controls scored 4/4 compared with 87% of hyperopes - only I of the 66 controls achieved less than full marks, compared with 9 of the 71 hyperopes. From the control data, and from experience in the use of these test items at 2.5-3 years,⁶ a score under full marks is expected in less than 5% of the general population at 3.5 years. Such scores can therefore be taken as an indicator that would be of clinical concern.

By contrast, the groups performed similarly on the block constructions. The only significant difference was on the very first construction at age 14 months, placing one block on top of another (chi square = 3.88, p < 0.05): 71% of controls succeeded in this compared with 55% of hyperopes. At none of the later ages did the blocks scores show a significant difference between groups.

MOVEMENT ABC (LONGITUDINAL GROUPS) In terms of overall mean z-score, controls performed significantly better than hyperopes at both ages tested. At 3.5 years the hyperopes had a mean combined

z-score of -0.23, s.d. 0.57, compared to the control group, with mean combined z-score of 0.00 by definition and s.d. 0.46 (Mann-Whitney U = 1781, p < 0.02). At 5.5 years, the hyperopes' mean was -0.34, s.d. 0.58, compared to the controls' 0.00, s.d. 0.50 (Mann-Whitney U = 1478, p < 0.001).

Analysing the tests individually, controls were significantly better than hyperopes on two tests at 3.5 years – jumping and walking along a line – and on five tests at 5.5 years: bean bag catching, balance (preferred and non-preferred leg), coin posting and bead threading.

Given the overall difference between groups, we wished to determine the proportion of children with clinically significant impairment by calculating normalised total impairment scores. In the Movement ABC normalisation for ages 4–5, total impairment scores of 17 or more correspond to the bottom 5th percentile. At 3.5 years, 9.1% of controls and 22.5% of hyperopes performed below this criterion. At 5.5 years, 1.5% of controls and 7.0% of hyperopes scored below this criterion.

GRIFFITHS TEST (LONGITUDINAL GROUPS) At 3.5 years, the controls' mean total score was higher for all scales taken together as well as on 5 of the 6 individual scales, but the differences were not significant. On the single scale administered at 5.5 years, controls again had a higher mean, but the difference did not reach significance.

LANGUAGE (LONGITUDINAL GROUPS) No significant differences were found between the two groups on the vocabulary measures. On the MCDI at 2 years, hyperopes had a mean of 57.1 words, s.d. 21.7, compared with a mean 57.2, s.d. 21.9 in the control group. At 5.5 years, the controls' mean standardised BPVS score was 111.3, s.d. 14.7, compared with the hyperopes' 108.1, s.d. 18.4.

STRABISMUS AND AMBLYOPIA To examine whether the differences between groups in the longitudinal analysis were associated with strabismus and/or amblyopia, strabismic and amblyopic children were taken out of the control and hyperopic groups, and the tests which had previously shown significant differences were re-analysed. On the blocks test at 14 months and the other items in the ABCDEFV at 14 months and 3.5 years, this made only a small difference to the distribution of hyperopes' scores (the proportion gaining full marks changing by +0.7%, -1.4% and -2.1%, respectively); hyperopes still performed significantly worse than controls on these tests. On the Movement ABC total at 3.5 years, the hyperopes' mean z-score rose to -0.17 (compared with the previous -0.23), and was no longer significantly different from the controls' 0.01 (Mann-Whitney U = 1401.5, p = 0.10). On the same test at 5.5 years, the removal of the strabismic and amblyopic subjects improved the hyperopes' mean z from -0.34 to -0.28, but the difference between groups remained significant (Mann-Whitney U = 1125, p < 0.002).

CROSS-SECTIONAL ANALYSIS All tests were re-analysed for the crosssectional groups, including data from children who were excluded from the longitudinal group because they did not attend all follow-ups. Table I indicates the numbers at each age.

The tests at 3.5 and 5.5 years that showed a significant group difference in the longitudinal sample continued to do so in the crosssectional group. However, at 14 months, the group differences did not reach significance: on the ABCDEFV non-blocks tasks, 79% of controls scored full marks, compared with 72% of hyperopes; 64% of controls passed on the block construction compared to 58% of hyperopes.

In this reanalysis, totals for the Griffiths scales at 3.5 and 5.5 years showed significantly better performance in the control group than the hyperopic group. At 3.5 years, controls passed a mean 24.2 out of the 29 tests, compared with the hyperopes' 22.8 (Mann-Whitney U = 5882, p < 0.02). When individual subscales were compared, two of the subscales at 3.5 years, 'Performance' and 'Personal-Social', showed significantly better performance by controls. On the 'Performance' scale at 5.5 years, controls performed significantly better than hyperopes (Mann-Whitney U = 3652, p < 0.05) with 55% of controls passing all six tests compared with 41% of hyperopes. It should be noted that the 'Performance' subscale includes visuospatial manipulative tasks requiring similar skills to ABCDEFV and movement ABC items, and that many items in the 'Personal/Social' subscale (e.g., dressing/undressing, undo/fasten buttons, helps to lay the table) have similar requirements.

The cross-sectional re-analysis of the British Picture Vocabulary Scale at 5.5 years also showed a significant difference (U = 4548, p < 0.02) between controls (mean 111.6, s.d. 13.9) and hyperopes (mean 106.3, s.d. 17).

Discussion Children identified at infant screening with significant hyperopic refractive errors show consistently poorer performance on a range of visuocognitive and visuomotor tests up to age 5 years, compared to control children without significant refractive errors. These differences are relatively small, but consistent over the age range in this group who constitute about 5% of the infant population. The general features of the results on the longitudinal group are confirmed in the cross-sectional analysis. This analysis is based on a larger group but does not represent the same set of individuals at each age, and so may be subject to some selection biases, e.g. children brought in after an absence because parents are concerned about their development.

The results in the longitudinal group do not appear to reflect simply a general delay in development, since they contrast with the tests of linguistic competence (vocabulary), and measures of general development from the Griffiths Scales, which showed no difference (although the Griffiths subscales related to 'performance' and 'eye-hand coordination' showed differences that approached significance). It should be noted, however, that none of the tests showed better performance by the children who had been infant hyperopes.

Between the groups as a whole, the differences are reliable but modest. However, they do imply that the hyperopic group contains an increased number of children whose level of impairment is of clinical concern (a six-fold increase in hyperopes compared to controls both in the ABCDEFV tests of visual cognition at 3.5 years, and in the Movement ABC at 5.5 years).

In general, the tests showing a relation to refraction in infancy all broadly involve visual or visuomotor function. However, it is unlikely that the deficits revealed are a simple consequence of blurred vision consequent on poor focussing in hyperopic refractive error. The visual materials involved in all these tests were large and high-contrast; the only case where hyperopic blur might be a limiting factor is the 'black/white cotton test' in the ABCDEFV at 14 months of age. In addition, hyperopic refractive errors in infancy (including hyperopic astigmatism) naturally decrease during the preschool years,^{15,16} and so few of the children in the initially hyperopic group will suffer from any significant blurring of vision in the preschool follow-up tests. In any case, children in the group with significant refractive error beyond three years of age wore any prescribed spectacle correction during testing. It is thus unlikely that any failures on follow-up at 3.5 and 5.5 years were a result of any remaining optical blur. However, our Infant Vision Screening Programmes have shown that hyperopic refractive error in early infancy is associated with higher incidence of strabismus and amblyopia (see, for example, refs. 2-3). Amblyopia, beyond acuity loss, can lead to problems in perceptual organisation and spatial judgements^{17,18} and strabismus is further associated with defective stereo depth vision which might impair a range of visuospatial tasks. Our results, however, show that most of the significant group differences remained when the comparison was restricted to children who showed neither amblyopia nor strabismus. These disorders may make some contribution to the worse performance of the hyperopic group, but they are by no means wholly responsible.

The tests that show a difference between groups can be considered as two clusters, tapping rather different functions. One cluster within the ABCDEFV is broadly visuocognitive, involving perceptual organisation, without an explicit visuomotor component: this includes the embedded figures tests and selecting the correct shape in the shape matching test. The second cluster has both visuocognitive and motor components, with the child required to coordinate and process visual information and link it to a smooth sequence of motor acts. Both fine and coarse motor skills tested in the Movement ABC fall into this category, as does the block construction test (spontaneous stack of two blocks) in the 14 months ABCDEFV. (However, the block construction copying test at later ages did not show a difference between groups, although the control group always showed a higher performance score than the group of hyperopes).

Neurobiological models of developing visual function¹⁹ suggest that the two clusters of tests may be related to a major division in the brain, between the 'ventral' cortical stream leading to the temporal lobe (specialised for object and face discrimination), and the 'dorsal' stream, via the parietal lobe which is involved in controlling actions and in spatial cognition.^{20,21} Since binocular stereo vision is primarily a dorsal stream function,²² the relation of infant hyperopia to strabismus might lead us to expect problems of dorsal-stream function in the group who were hyperopic at screening. However, the two clusters described above would be expected to involve both ventral and dorsal pathways. Dorsal stream deficits^{23,24} and deficits in motor skills²⁵ have been hypothesised to be associated with dyslexia. Our further work with this data set will include analysis of tests of phonological processing that have been shown as predictors of dyslexia, as well as measures of attention (since attentional deficits may well be linked to motor deficits).

We do not know the basis of the correlation between infant refractive error and later visuocognitive and visuomotor failures. Although we do not believe that image blur directly impairs test performance, nor that overt strabismus and amblyopia account for the results, the early (albeit mild) sensory loss due to refractive error may lead to a cascade of effects on later developing brain systems responsible for ability on visuocognitive and visuomotor tasks. Alternatively, hyperopia and poor focussing at age 9 months may themselves reflect aspects of brain function, since the growth of the eye towards emmetropic refraction is under active neural control;^{26,27} thus both refractive status and visuomotor development may be indicators of a common underlying factor.

Whatever the direction of causality, vision screening in infancy can identify a group at greater risk in terms of cognitive and motor performance. Hyperopia should therefore be taken into account as a risk factor in the developmental assessment of young children. It is hoped that this finding may encourage the search for interventions to develop fully these children's skills in the preschool and early school years.

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