# Retrospective investigation of improvements in functional vision for adolescent students with cerebral vision impairments in a specialist residential school and college setting

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**Abstract**

Cerebral and/or Cortical Vision Impairment (CVI) is the leading cause of childhood vision impairment in the Global North. Previous studies have demonstrated that the functional vision of children with CVI can develop over time, but evidence for the effectiveness of interventions is still in its infancy. In this study we retrospectively reviewed student records from a specialist residential school and college in the United Kingdom that had implemented an evidence-based approach to assessment and intervention for adolescent students with CVI called the CVI Range. The outcome of CVI Range assessments were recorded annually over a five-year period, and potential predictor variables such as measures of visual acuity and presence of conditions such as cerebral palsy and seizure disorders were recorded as part of standard practice within the service. A total of 73 annual assessments were analysed from a total of 24 students between the ages of 9 and 25 years old. We used a mixed model for repeated measures approach to reveal a significant fixed effect of time on functional vision that equated to a linear increase of 0.78, 95% CI (0.60, 0.97) in CVI Range Rating 2 for each year of participation on the programme. The mixed effects models also revealed significant interindividual differences in functional vision, which could be partly explained by a significant negative effect of acuity and by a joint positive effect of nystagmus and time, but not by age. These findings demonstrated that significant improvements in functional vision are still possible for students with CVI long after the accepted sensitive period of neuroplasticity in the visual cortex. Further studies incorporating research designs appropriate for evaluating complex interventions are required to determine which individual and contextual characteristics are valid and reliable predictors of improvements in functional vision for young people with CVI.

*Keywords*: adolescent, cerebral vision impairment, CVI Range, habilitation, functional vision, rehabilitation, student

## Introduction

In the contemporary context, Cerebral and/or Cortical Vision Impairment (CVI) is considered an umbrella term for a spectrum of visual difficulties associated with neurological conditions that cannot be attributed to disorders of the eye or anterior visual pathway (Sakki et al., 2018). Individual visual, perceptual, and attentional functions can be independently and uniquely affected by the cause, location, and extent of brain injury or neurodivergence; leading to a vast combination of impairment types and severity (Dutton & Bax, 2010; Ortibus et al., 2011; Zihl & Dutton, 2015). Causes of CVI include perinatal hypoxic-ischemia, asphyxia, cranial trauma, infections and inflammation such as meningitis and encephalitis, metabolic disorders, hydrocephalus, stroke, brain tumour, developmental brain differences, and genetic differences (Bosch et al., 2014; Good, et al., 2001; Huo et al., 1999; Khetpal & Donahue, 2007, Wilton et al., 2021). Additionally, widespread brain injury and developmental brain differences can lead to co-occurring difficulties such as cognitive, motor, or other sensory impairments; even other neurological conditions such as seizure disorders (Huo et al., 1999; Khetpal & Donahue, 2007). As such, CVI is an extremely heterogenous medical condition; although efforts towards a classification framework based on ophthalmological and neuropsychological characteristics are ongoing (Sakki et al., 2020; Philip et al., 2023).

CVI is currently the primary cause of low vision for children in the Global North and is becoming increasingly recognised in the Global South (Solebo & Rahi, 2014). The most recent prevalence study in the United Kingdom has indicated that, on average, one child in a mainstream primary school classroom of thirty pupils will have CVI-related vision difficulties (Williams, Pease, Warnes et al., 2021). The trend of increasing prevalence appears to be partly due to at-risk groups becoming more likely to survive brain injury due to modern advances in healthcare, and partly because there has been a significant reduction in other preventable causes of vision impairment (Good et al., 2001; Ozturk et al., 2016; Rahi et al., 2003; Tunay et al., 2020). It could also be argued that the range of impairments covered by the umbrella term of CVI has increased as our

understanding of perceptual impairments not associated with changes in visual acuity has changed over the last two decades. CVI has a significant negative impact on quality of life irrespective of severity of impairment, visual acuity, or cognitive ability (Sakki, 2018). Additionally, vision impairment incurred prenatally or during infancy can have a significant negative impact on development and learning (Lueck & Dutton, 2015; Sonksen & Dale, 2002, Williams et al., 2011). In practice, the impact CVI can have on cognitive, social, and motor skill development makes the differential diagnosis between CVI and developmental conditions such as Autism and Developmental Coordination Disorders complex (Chokron & Dutton, 2016; Chokron et al., 2020; Lueck et al., 2023).

In some cases of CVI, functional vision may spontaneously improve during early development although it is not clear whether this is due to widespread neuroplastic reorganisation or delayed maturation of extant neuroanatomy (Bennet et al., 2020; Hoyt, 2003; Martin et al., 2016; Matsuba & Jan, 2006). The evidence-base for treatment options for CVI is still developing so holistic support, extended curricula, and environmental adaptations at home and at school remain the mainstay for these children and young people (Ben Itzhak et al., 2021; Ben Itzhak et al., 2022; Chorna et al., 2017; Clark et al., 2021; Ciman et al., 2018; Delay et al., 2022; Ivanov et al., 2018; Kooiker et al., 2020; McDowell & Budd, 2018; McLinden et al., 2016; Overbeek et al., 2022; Pilling & Little, 2020; Salihodžic et al., 2018; Waddington et al., 2015; Waddington & Hodgson, 2017; Waddington et al., 2018; Williams et al., 2014; Williams, Pease, Goodenough et al., 2021). Effective information sharing between parents and carers, clinical professionals, and educational professionals is thought to be crucial for habilitation (Goodenough et al., 2021; Hyvärinen et al., 2012; McDowell, 2020b; Pease et al., 2020). Additionally, evidence from children and young people with amblyopia has indicated that earlier interventions (e.g., with perceptual training) are more effective, but that different visual cognitive functions have different sensitive periods of development and neuroplasticity (Cooper & Mackey, 2016; Siu & Murphy, 2018). Planning support and interventions for someone with CVI requires an individualised assessment of their abilities and aspirations (McDowell, 2021). However, the complex interaction between the impairment, personal factors (e.g., comorbidities,

developmental age, etc.), and environmental factors (e.g., familiarity with activities, complexity of setting, etc.) creates substantial challenges for assessment.

Surveys have been developed as population screening tools to aid early identification of unrecognised CVI (Ben Itzhak et al., 2020; Gorrie et al., 2019). After screening, a more comprehensive assessment of basic visual sensory functions may include measures of visual acuity, visual field sensitivity, contrast sensitivity, colour vision, stereovision, form and motion coherence, light adaptation, and ocular motility. Neurocognitive tests can be used to evaluate higher order visual perceptual and attentional functions. Individual components of the visual cognitive system can be evaluated successfully in some children with CVI, but this is dependent on developmental age (McDowell, 2020a; Vancleef et al., 2020a, 2020b; Waddington et al., 2020). Additionally, methods such as remote eye tracking and standardised parental questionnaires have been developed for early detection of CVI in children too young or otherwise unable to communicate (García-Ormaechea et al., 2014; Kooiker et al., 2014; Ben Itzhak et al., 2023). However, children's visual abilities during everyday activities may differ from those observed during standardised assessments administered in a clinical setting. While it is necessary to understand the root-causes of CVI to plan effective interventions (Ravenscroft & Lueck, 2020), it is also necessary to evaluate how a child with CVI functions during daily vision-related activities by conducting a functional vision assessment (FVA).

A FVA should focus on the impact vision impairment has on meaningful vision-related activities that a child is expected to attempt regularly, such as basic and instrumental activities of daily living, communication, and orientation and mobility (Colenbrander, 2010). A FVA should be person-centred, co-rated when plausible, and administered across contrasting times, locations, and routines as fluctuations in personal and environmental factors can cause functional vision to vary (Deverell, 2011, 2016). Common methods of administering a FVA with children include comprehensive history taking involving families and carers, unstructured observations of the child

behaving naturally in different surroundings, and direct assessment of pertinent vision-related activities; all with multidisciplinary professional support (Lueck 2004; Lueck & Dutton, 2015). Standardised questionnaires have been developed to collect patient reported measures of functional vision that can be compared across individuals or experimental groups, although these often include more general measures of participation and quality of life (Elsman et al., 2017; Gothwal et al., 2012; Khadka et al., 2010; Tadić et al., 2013). While many tools and approaches to administering a FVA have been developed in practice, few have been validated for use with children and young people with CVI.

The CVI Range is one framework for assessing functional vision and planning interventions that has been validated as a reliable tool for administering a FVA specifically for children and young people diagnosed with CVI, over the age of nine months (Newcomb, 2009, 2010; Roman-Lantzy, 2007, 2018, 2019; Roman-Lantzy & Lantzy, 2010). A modified version of the assessment is currently being evaluated for clinical research purposes (Chang et al., 2022). In a simple sense, the framework provides a series of descriptors that indicate steppingstones from lower functional vision to higher functional vision, which are measured on two ordinal scales from 0 to 10. Like many curriculum frameworks, the CVI Range is outcome-based, and descriptors can be used to measure progress and set targets in an education setting. The CVI Range has strengths and limitations that depend on the context of its use. However, there is an expectation that planning habilitation sessions to support the development of functional vision will involve a student-centred and multidisciplinary approach that includes the use of additional assessment tools to account for limitations. A strength of the CVI Range in the context of this study is the ability to provide a reliable and comprehensible outcome measure that can track progress in the development of functional vision over time.

Mensah et al. (2019) published a retrospective cohort study indicating that multidisciplinary and intense rehabilitation provided in an education setting for students with acquired brain jury could produce improvements in functional vision as measured on the CVI Range. Their study focused

on changes in functional vision observed in a group of fourteen children and young people (5-21 years old) with acquired brain injury over a one-year period. They concluded that observed improvements in functional vision were due to participants receiving 10-15 hours of intense multidisciplinary rehabilitation each week while attending the specialist school.

The aim of the study presented in this paper was to retrospectively investigate the development of functional vision in adolescent students with CVI and additional support needs, who were attending a specialist residential school and college in the United Kingdom. This retrospective case series was investigated over a five-year period, during which time CVI Range assessments had been conducted annually. Our primary research aim was to determine whether improvements in functional vision continued after the first year for adolescent students with CVI. As such, our initial hypotheses were that final CVI Range assessment scores would be significantly higher than baseline, and those differences would vary as a function of the number of months between baseline and final assessment dates. We also hypothesised that changes in functional vision could be multifactorial, accounting for differences in age, aetiology, baseline measures on the CVI Range, general health, and therapeutic and educational interventions. As such, our secondary research aim was to explore the relationship between measures of potential predictor variables that were recorded as part of the normal school and college practice over that period with measures on the CVI Range.

## Methods

### Ethics statement

This study was a retrospective analysis of existing data retrieved from school and college records of students over an approximately five-year period before the disruption to education services caused by the global COVID-19 pandemic. Exact dates have not been disclosed to support the maintenance of student confidentiality. The study was reviewed and approved by Plymouth Marjon University Research Ethics Panel (ref: EP176), and separately by the school and college's board of directors prior to sharing the data. Data was not retrieved for students who had not given

consent (or assent with parental consent) to the school and college to share their data with external professionals. Data retrieval and analysis adhered to professional codes of practice and all legal requirements, including compliance with General Data Protection Regulations. Data anonymity has been maintained and data cannot reasonably be made un-anonymous through data linkage to other datasets.

**Student datasets**

Datasets were identified for inclusion if the educational record indicated that a student either had a diagnosis of CVI confirmed from a medical professional or had impaired binocular visual acuity (≥0.5 Logarithm of the Minimum Angle of Resolution; logMAR) with concurrent visual processing difficulties that were suspected to be the result of CVI. Educational records only indicated whether a historical diagnosis of CVI had been given and did not indicate the test procedures used to develop the diagnosis. A total of 35 sets of student data were identified from the school records on the basis that they met the inclusion criteria and the student had completed at least one CVI Range assessment. 7 students had not given their consent to share their data so 28 datasets were shared with the research team.

17 students had a confirmed diagnosis of CVI from a medical professional, and 11 had a suspected or working diagnosis of CVI. The ages of students ranged from 9 years and 8 months to 21 years and 4 months at their baseline CVI Range assessment, *M* = 16 years and 11 months, *SD* = 2 years and 11 months. 16 students identified as male, and 12 identified as female. All 28 students had multiple disabilities, impaired binocular visual acuity, and a neurological condition that was either congenital, perinatal, or acquired before the age of 2 years. Student records also indicated the absence or presence of visual field impairment, strabismus, nystagmus, cerebral palsy, and seizure disorders. Descriptive statistics of these variables and their relationship with CVI Range assessment scores are presented in the Results section.

**"Cortical Visual Impairment Range" ("CVI Range") outcome measures**

A full-time teaching assistant had been trained to conduct CVI Range assessments and generate reports for the multidisciplinary team working with students. The teaching assistant demonstrated competency in conducting the assessments through attaining the former international Perkins-Roman CVI Range Endorsement. 28 students completed one baseline CVI Range assessment, 24 of those students completed two consecutive annual assessments, 18 of those students completed three assessments, and 7 of those students completed four assessments. Only datasets where students completed more than one annual CVI Range assessment were included in the analysis, giving a total $N = 73$ observations.

A thorough description of the CVI Range assessment procedure can be found in Roman-Lantzy (2007). In brief, the aim of each assessment was to triangulate the presence of up to ten behavioural characteristics and evaluate the impact of them on daily activities. The ten behavioural characteristics included but were not limited to: difficulties or preferences in visual attention towards visually complex materials, colour, movement, specific areas of the visual field, or visual novelty. The triangulation was based on three techniques: i) a questionnaire and interviews with parents or carers, ii) a structured face-to-face assessment of the young person's visual functioning, and iii) informal observation of the young person behaving naturally in different environments.

The CVI Range assessment results in two outcome measures: Rating 1 and Rating 2. CVI Range Rating 1 (CVIRR1) can be defined as a general measure of the impact of CVI on a child's functional vision and is measured on an ordinal scale ranging from 1 to 10. CVI Range Rating 2 (CVIRR2) can be defined as a more specific measure of the presence of certain behavioural characteristics that are associated with CVI. CVIRR2 is measured on a series of ten Likert items that each contain five ordinal categories scored from 0 to 1. The Likert items are summed to give a total CVIRR2 score ranging from 0 to 10 on a Likert-type scale. Each CVI Range rating can be more broadly categorised into three developmental phases of functional vision. Ratings between 0-3 can be classified as Phase 1, between 4-7 as Phase 2, and between 8-10 as phase 3.

**Practice setting**

The multidisciplinary team based at the school and college consisted of qualified teachers of the vision impaired, classroom teachers, teaching assistants, healthcare assistants, habilitation (or orientation and mobility) specialists, a braille teacher, occupational therapists, physiotherapists, speech and language therapists, music therapists, nurses, and a clinical psychologist. The team also had assessment information provided by a visiting orthoptist twice per month and a low vision aid specialist twice per year. Hours spent in education, habilitation or therapy were arranged according to students' individual needs and scheduled according to an individualised education, health and care plan. In the sense that most of these professionals were situated in the same offices and worked collaboratively (e.g., when goal setting) on a day-to-day basis, this service delivery model could be considered an interdisciplinary as opposed to multidisciplinary or transdisciplinary team model (Ogletree et al., 2001).

As the evidence-base for functional vision interventions was limited, practitioners relied on systematic observations of students' strengths, difficulties, interests, and dislikes during class, leisure, and residential time to provide functional information to guide goal setting and intervention. Broadly speaking, the type of interventions used were dependent on which phase of the CVI Range the student was functioning in. Students functioning in phase 1 were given the opportunity to develop their functional vision during very short but regular out-of-classroom interventions in well-controlled sensory environments. The intended goal of these interventions was to develop consistent visual attention to simple and/or familiar moving 3D objects. Salient visual characteristics of objects such as colour, shape, and size could be taught during this phase. Students functioning in phase 2 would still require some environmental modifications to consistently attend to objects such as a quiet corner in the classroom where visual and other sensory distractions were minimised. The intended goal of adaptations and interventions in phase 2 was for students to learn to use their vision to do something meaningful such as guide their reach towards an object of interest or play an

adapted computer game using switches. Students functioning in phase 3 could be introduced to more complex teaching materials and activities. The intended goal of adaptations and interventions in phase 3 was to enable students to slowly refine visual cognitive skills (e.g., scanning strategies, identifying salient features) so that they could be used to support meaningful daily activities such as communication, and orientation and mobility in the increasingly complex multisensory environments they would encounter during adulthood.

**Statistical analysis**

Taking the sum of a substantial number of ordinal variables can create an approximation of a continuous variable, when measuring an underlying psychological construct (Johnson & Creech, 1983; Norman, 2010; Sullivan & Artino, 2013; Zumbo & Zimmerman, 1993). We found the distribution of CVIRR2 scores was not statistically significantly different to Normal using the Shapiro-Wilk test, $W(28) = .953$, $p = .238$. Additionally, the distribution of the difference in CVIRR2 scores between baseline and final assessments was not statistically significantly different to Normal, $W(24) = .950$, $p = .271$. As such, we analysed CVIRR2 scores using more familiar parametric methods, and analysed CVIRR1 scores using more statistically appropriate non-parametric methods. To test for significant improvements in CVIRR1 and CVIRR2 between the baseline and final assessment, we used a paired-samples binomial sign test and a paired-samples *t*-test respectively.

Datasets were not balanced as not all students completed the maximum four annual assessments, and there was some variation in the exact timing of annual assessments (e.g., due to scheduling issues). The advantages of using linear mixed-effects models (LMMs) over repeated measures ANOVA when analysing longitudinal data with missing observations have been discussed in relevant previous studies (e.g., Walker et al., 2019). Given the unbalanced datasets, we constructed LMMs to analyse the changes in CVIRR2 over time and to explore the potential effects of predictor variables. We also constructed cumulative link mixed models (CLMMs) to analyse the changes in CVIRR1 over time. These analyses were conducted using R (R Core Team, 2022) with

RStudio (Posit Team, 2023), and the lme4 (Bates et al., 2015), ordinal (Christensen, 2022), performance (Lüdecke et al., 2021), and EMAtools (Kleiman, 2021) packages.

Datasets contained hierarchical data in which repeated measurements (i.e., level 1) were clustered within students (i.e., level 2). Duration (measured in years that students had participated in the habilitation programme) was the only level 1 predictor as it varied between repeated measurements. Level 2 predictors varied between students but not between repeated measurements, and were operationalised as: Acuity (measured in logMAR at baseline assessment), Age (measured in years at baseline assessment), Cerebral palsy (present = 1 or absent = 0), Assigned sex (male = 1 or female = 0), Nystagmus (present = 1 or absent = 0), Seizure disorder (present = 1 or absent = 0), Strabismus (present = 1 or absent = 0), Therapist contact (measured in hours per year), and Visual field impairment (present = 1 or absent = 0).

Centring predictors involves rescaling by subtracting the mean, which can aid in the interpretation of intercepts and intercept variances estimated using multilevel regression. Grand-mean centring uses the mean of the predictor calculated from the full sample. Cluster-mean centring uses the mean of the predictor calculated from within each cluster. The level 2 predictors that were measured on a continuous scale (i.e., Acuity, Age, and Therapist contact) were grand-mean centred. Duration was cluster-mean centred to generate the level 1 predictor: $\text{Duration}_1$. The cluster-means were then reintroduced and grand-mean centred as the level 2 predictor: $\text{Duration}_2$. This essentially separated the initial predictor “Duration” into level 1 (i.e., within student) and level 2 (i.e., between student) variance.

Model 1 was constructed to determine the effect of Duration on CVIRR1. A CLMM was fitted by the Laplace approximation approach to the repeated measures of all recorded CVIRR1 scores after excluding three missing observations ($n_{CVIRR1}$ = 70 of 73 observations). The model included $\text{Duration}_1$ and $\text{Duration}_2$ as fixed effects, with a random intercept for individual students and a random slope for $\text{Duration}_1$. Likelihood ratio tests of the proportional odds assumption were

calculated for a reduced version of this model that included the fixed effects but did not include random effects. There was no evidence of nominal or scale effects for the fixed effect of $Duration_1$ or $Duration_2$. As such a proportional odds model was fitted to Model 1, i.e., a CLMM with a logit link function.

Model 2 was constructed to determine the effect of Duration on CVIRR2. A LMM was fitted by the maximum likelihood approach to all recorded CVIRR2 scores after excluding two missing observations ($n_{CVIRR2}$ = 71 of 73 observations). The model included $Duration_1$ and $Duration_2$ as fixed effects with a random intercept for individual students and a random slope for $Duration_1$.

Model 3 was constructed to explore the effect of the level 2 predictors on CVIRR2. A LMM was fitted by the maximum likelihood approach to CVIRR2 scores. The model included $Duration_1$ and $Duration_2$ as fixed effects with a random intercept for individual students and a random slope for $Duration_1$. All level 2 predictors were also included in the model as fixed effects, to explore their potential contribution to the interindividual differences in the CVIRR2 intercept. Interaction terms between each level 2 predictor and $Duration_1$ were also included in the model to explore their potential contribution to the individual differences in the slope of $Duration_1$.

In all cases, an unstructured covariance matrix was selected to model the correlations between random slopes and intercepts. Intraclass correlation coefficients (ICCs) and pseudo-R-squared values were calculated using the framework proposed by Nakagawa et al. (2017). Cohen's d effect sizes were estimated using the Satterthwaite approximations to degrees of freedom.

## Results

### Outcome measures at the baseline and final assessment

A paired-samples sign test indicated that CVIRR1 scores significantly improved, $S$ = 0, $N$ = 23, $p$ < .001, Cohen's $h_2$ = 1.57, 95% CI (0.78, 1.57); from their baseline, *Mdn* = 5, *Minimum* = 2, *Maximum* = 7; to their final assessment, *Mdn* = 7, *Minimum* = 3, *Maximum* = 9.

A paired-samples *t*-test indicated that CVIRR2 scores significantly improved, $t(23) = 8.17$, $p < .001$, Cohen's $d = 1.10$, 95% CI (0.66, 1.52); from their baseline, $M = 5.17$, $SD = 1.61$; to their final assessment, $M = 6.99$, $SD = 1.69$.

**Descriptive statistics of candidate predictor variables for CLMM and LMMs**

Mean age at the time of baseline CVI Range assessments was 16 years and 11 months, *SD* = 2 years and 11 months. 57% of students identified as male and 43% identified as female. 83% of the cohort had strabismus, 43% had cerebral palsy, 43% had a seizure disorder, 34% had nystagmus, and 17% had a visual field impairment.

Median baseline visual acuity was 0.98 logMAR, 95% CI (0.78, 1.20). 21 of 24 datasets included visual acuity measures at the time of the first two annual CVI Range assessments, and 18 of 18 datasets included acuity measures at the time of the first three assessments. Visual acuity did not significantly differ on average over one year, *Mdn* = 0 logMAR, 95% CI (−0.02, 0.10); or over two years, *Mdn* = 0 logMAR, 95% CI (−0.30, 0.23).

The mean duration that students participated on the habilitation programme was 2 years and 3 months, *SD* = 1 year and 0 months. On average, students participated in 129 hours, *SD* = 32 hours; of contact time with therapists and habilitation specialists on campus each academic year.

**Changes in outcome measures as a function of duration on the habilitation programme**

Table 1 summarises the results of the CLMM for CVIRR1 (Model 1). We found a significant fixed effect of $\text{Duration}_1$, $\hat{\beta} = 5.13$, 95% CI (1.60, 8.67), $p = .004$. Essentially, one year of participation on the habilitation programme decreased the threshold coefficients by 5.13 log units, multiplying the odds for a higher CVIRR1 score by 169, OR = 169, 95% CI (4.95, 5830).

Nakagawa's intra-class correlation coefficients were calculated for Model 1, $\text{ICC}_{\text{LMM}} = .683$; $\text{ICC}_{\text{LMM(adj)}} = .957$; indicating that 68% of the variance in CVIRR1 was explained by interindividual differences. Nakagawa's marginal and conditional pseudo-R-squared values were calculated, $R^2_{\text{GLMM(m)}}$

= .287; $R^2_{GLMM(c)}$ = .969; indicating that 29% of the variance in CVIRR1 was explained by the fixed effects of Model 1. This model was compared with a null model that contained only an overall intercept as a fixed effect and an intercept for each student as a random effect. A log-likelihood ratio test demonstrated that Model 1 was a more likely fit than the null model, $\chi^2(4) = 76.0$, p < .001.

Table 2 summarises the results of the LMM for CVIRR2 that included Duration only as a predictor variable (Model 2). We found a significant fixed effect of $Duration_1$, $\hat{\beta}$ = 0.78, 95% CI (0.60, 0.97), $p < .001$; and a significant fixed effect of $Duration_2$, $\hat{\beta}$ = −1.18, 95% CI (−2.19, −0.13), $p = .023$. Essentially, for each additional year of participation on the habilitation programme, CVIRR2 increased by 0.78 for individual students. Additionally, students who had participated on the habilitation programme for one year longer than the average duration across the cohort had a CVIRR2 that was 1.18 lower than the average CVIRR2 across the cohort, indicating that students with lower CVIRR2 on average participated for longer on the programme.

Nakagawa's intra-class correlation coefficients were calculated for Model 2, $ICC_{LMM}$ = .618, 95% CI (.354, .766); $ICC_{LMM(adj)}$ = .906, 95% CI (.790, .950); indicating that 62% of the variance in CVIRR2 scores was explained by interindividual differences. Nakagawa's marginal and conditional pseudo-R-squared values were calculated, $R^2_{LMM(m)}$ = .318, 95% CI (.181, .481); $R^2_{LMM(c)}$ = .936, 95% CI (.883, .965); indicating that 32% of the variance in CVIRR2 was explained by the fixed effects of Model 2. This model was compared with a null model that contained only an overall intercept as a fixed effect and an intercept for each student as a random effect. A log-likelihood ratio test demonstrated that Model 2 was a more likely fit than the null model, $\chi^2$ (4) = 76.9, p < .001.

**Factors associated with changes in outcome measures over time**

Table 3 summarises the results of the LMM for CVIRR2 that included the full list of candidate predictor variables at level 2 (Model 3). Model 3 included Duration as a fixed effect at level 1 and level 2, each candidate predictor as a fixed effect at level 2, interaction terms for each candidate predictor with $Duration_1$, a random intercept, and random slope for $Duration_1$. We found a

significant fixed effect of $\text{Duration}_1$, $\hat{\beta}$ = 1.23, 95% CI (0.64, 1.83), $p < .001$; and a significant fixed effect of Acuity, $\hat{\beta}$ = −1.35, 95% CI (−2.31, −0.38), $p = .009$. Essentially, CVIRR2 for individual students increased by 1.23 for each year of participation on the habilitation programme, after adjusting for simultaneous linear changes in the other predictors. Additionally, CVIRR2 decreased by 1.35 as acuity increased by 1.0 logMAR across the cohort, after adjusting for simultaneous linear changes in other predictors. We found a significant joint effect of Nystagmus × $\text{Duration}_1$, $\hat{\beta}$ = 0.50, 95% CI (0.12, 0.89), $p = .012$; and a significant joint effect of $\text{Duration}_2$ × $\text{Duration}_1$, $\hat{\beta}$ = −0.45, 95% CI (−0.89, −0.00), $p = .047$. Essentially, CVIRR2 increased by an additional 0.50 for every year of participation on the habilitation programme for students with nystagmus when compared to students without nystagmus, after adjusting for simultaneous linear changes in other predictors. Additionally, for students who participated on the habilitation programme one year longer than the average duration across the cohort, the increase in CVIRR2 each year was 0.45 lower than the average increase across the cohort, after adjusting for simultaneous linear changes in other predictors. The joint $\text{Duration}_2$ × $\text{Duration}_1$ effect may indicate diminishing returns on long-term participation in the habilitation programme.

Nakagawa's intra-class correlation coefficients were calculated for Model 3, $\text{ICC}_{\text{LMM}}$ = .184, 95% CI (.002, .181); $\text{ICC}_{\text{LMM(adj)}}$ = .759, 95% CI (.032, .844); indicating that 18% of the variance in CVIRR2 scores was explained by interindividual differences not included as fixed effects in Model 3. Nakagawa's marginal and conditional pseudo-R-squared values were calculated, $R^2_{\text{LMM(m)}}$ = .758, 95% CI (.752, .927); $R^2_{\text{LMM(c)}}$ = .942, 95% CI (.923, .977); indicating that 76% of the variance in CVIRR2 was explained by the fixed effects of Model 3. This model was compared with Model 2 to explore the contribution of candidate predictor variables to the interindividual differences in CVIRR2 and the interindividual differences in the slope of $\text{Duration}_1$ (on CVIRR2). A log-likelihood ratio test demonstrated that Model 3 was a more likely fit than Model 2, $\chi^2$ (19) = 38.2, p = .006. However, the Bayesian Information Criterion (BIC) was much higher for Model 3, BIC = 253.74; than for Model 2, BIC = 210.93; and the Akaike Information Criterion (AIC) was only slightly lower for Model 3, AIC =

194.91; than for Model 2, AIC = 195.09; indicating Model 3 was perhaps not as parsimonious as Model 2.

## Discussion

The study findings demonstrated that functional vision significantly increased between baseline assessments and final assessments for adolescent students participating in the CVI Range habilitation programme, and that the magnitude of improvements increased in relation to the duration that students participated. In effect, CVIRR2 increased linearly by 0.78 for each year of participation on the programme, and the odds that CVIRR1 would improve after each year of participation was 169 to 1, when other predictor effects were not considered. Without a control group and experimental design, we are unable to infer that these improvements were caused by participation in the habilitation programme alone, as confounding factors such as natural development and placebo effects almost certainly contributed to observed improvements. Intra-class correlation coefficients demonstrated that 62-68% of the variance in functional vision measures was explained by interindividual differences. We found individual traits that were significant predictors of functional vision and of the rate of change in functional vision over time. For example, CVIRR2 decreased linearly across participants by 1.35 as visual acuity increased by 1.0 logMAR, and the rate of change in CVIRR2 increased by 0.50 per year for students with nystagmus when compared to those without. Notably, some individual traits that we might have expected to have a significant effect on the rate of change in CVIRR2, such as age, did not. Finally, pseudo-R-squared values demonstrated that the explained variance of fixed effects improved from 32% when considering a model that contained duration alone to 76% when considering a model that included all candidate predictors, although such a model was not parsimonious and contained very few individual traits that had a statistically significant fixed effect on functional vision.

It is critical to consider that the age of these students ranged from 9 years and 8 months at the start of the programme to 24 years and 1 month at the end of the programme, long after the

sensitive period of early postnatal life when the developing visual system is highly neuroplastic (Siu and Murphy, 2018). As such, natural development of visual function seems an unlikely explanation for the recorded improvements in functional vision, and we found no evidence of improvements in visual acuity over the course of the programme to support such a theory. Additionally, people with CVI may use a range of sensory modalities (e.g., sound, touch, etc.) to compensate for their vision impairment but, while using these complementary strategies is important for the young person and may improve more general measures of function and participation, it is unclear whether developing skills in other modalities would transfer to improvements in functional vision itself. Instead, improvements in functional vision seem most likely to be associated with the natural development of higher order visual cognitive functions (e.g., visuospatial processing, object recognition, visual imagery, etc.) or with learning and developing metacognitive compensatory strategies (e.g., systematic scanning techniques, concept development, organisational skills, etc.).

Roman-Lantzy & Lantzy (2010) reported improvements in functional vision over time from children attending a specialist clinic that was delivered as an extension to neonatal intensive care unit follow-up programmes at the time. The authors evaluated data from a cohort of 77 individuals aged between 2 months and 13 years at the time of their first assessment and observed that the average amount of time for individuals to improve from phase 1 to phase 3 on the CVI Range was 3.7 years, regardless of age. Extrapolating from our own findings we would instead expect to observe an improvement of 2.9 on CVIRR2 for adolescent students over a 3.7-year period, a somewhat reduced improvement when compared with Roman-Lantzy et al.'s observation. This difference in findings may reflect a real age effect that is only observable over a wide range from infants to young adults or may reflect a difference in context between the two services. Mensah et al. (2019) reported significant improvements in functional vision for a cohort of 27 students aged between 5 years and 21 years at the time of their first assessment, observing that CVI Range scores improved by approximately 1.18 over a one-year period. This improvement was again larger than expected from our own findings and may reflect the inclusion of different personal characteristics in Mensah et al.'s

cohort (i.e., younger in age and brain injury acquired later in life) or different contexts between the two services (e.g., intensity of therapy).

It is perhaps not surprising that visual acuity is a significant predictor of functional vision for students with CVI. However, our findings would predict a difference of only 2.0 in CVIRR2 between students with a visual acuity of 0.3 logMAR and those with 1.8 logMAR, defined as "no vision impairment" and "blindness" respectively by the International Classification of Diseases, 11$^{th}$ edition (World Health Organisation, 2019). More surprising is the finding that the functional vision of students with nystagmus would improve at a faster rate than for those without nystagmus, as we might expect comorbid medical conditions such as ocular or oculomotor impairments to be barriers to improving functional vision. However, there is emerging evidence that perceptual training for adolescents with nystagmus can improve visual function and functional vision (Daibert-Nido et al., 2021; Huurneman & Goossens, 2021). These findings require further investigation, especially given that Jacobsen et al. (2009) have demonstrated that periventricular leukomalacia often results in comorbid CVI and nystagmus.

One of the most significant limitations of this study is the lack of a control group to confidently determine which changes in outcome measures are due to the active components of the habilitation programme and which are due to other factors such as natural development or placebo effects that haven't been controlled for. An experimental research design has been a common element missing in previous habilitation studies due to the recognised ethical, operational, and contextual challenges associated with conducting randomised controlled trials (RCTs) to test long-term complex interventions (Tarquinio et al., 2015). A collaborative strategy for overcoming the challenges and limitations of RCTs is required to evaluate habilitation programmes for young people with CVI. We believe our findings contribute to a phased approach to determine the personal and contextual characteristics that are predictive of improvements in functional vision for students with CVI, but we recognise that different epistemological approaches are required to fully evaluate complex

interventions. While we may not be able to definitively confirm efficacy of the CVI Range framework for assessment and intervention with this data alone our findings do demonstrate that functional vision for adolescent students can significantly improve over time, and based on our findings strongly recommend that adolescent students are not excluded from vision habilitation services based on age alone.

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**References**

Bates, D. Maechler, M., Bolker, B., and Walker, S. (2015). Fitting linear mixed-effects models using lme4. *Journal of Statistical Software*, *67*(1), 1-48. doi: 10.18637/jss.v067.i01.

Ben Itzhak, N., Franki, I., Jansen, B., Kostkova, K., Wagemans, J., and Ortibus, E. (2021). An individualized and adaptive game-based therapy for cerebral visual impairment: Design, development, and evaluation. *International Journal of Child-Computer Interaction*, *31*, 100437. doi: 10.1016/j.ijcci.2021.100437.

Ben Itzhak, N., Franki, I., Jansen, B., Kostkova, K., Wagemans, J., and Ortibus, E. (2022). Usability and user experience of an individualized and adaptive game-based therapy for children with cerebral visual impairment. *International Journal of Child-Computer Interaction*. Advance online publication. doi: 10.1016/j.ijcci.2022.100551.

Ben Itzhak, N., Kooiker, M., Pel, J., and Ortibus, E. (2023). Including visual orienting functions into cerebral visual impairment screening: Reliability, variability, and ecological validity. *Research in Developmental Disabilities*, *132*(3), 104391. doi: 10.1016/j.ridd.2022.104391.

Ben Itzhak, N., Vancleef, K., Franki, I., Laenen, A., Wagemans, J., and Ortibus, E. (2020). Visuoperceptual profiles of children using the Flemish cerebral visual impairment questionnaire. *Developmental Medicine and Child Neurology*, *62*, 969-976. doi: 10.1111/dmcn.14448.

Bennett, C., Bauer, C., Bailin, E., Merabet, L. (2020). Neuroplasticity in cerebral visual impairment (CVI): Assessing functional vision and the neurophysiological correlates of dorsal stream dysfunction. *Neuroscience & Biobehavioral Reviews*, *108*, 171-181.

Bosch, D., Boonstra, F., Willemsen, M., Cremers, F., and de Vries, B. (2014). Low vision due to cerebral visual impairment: Differentiating between acquired and genetic causes. *BMC Ophthalmology*, *14*, 59. doi: 10.1186/1471-2415-14-59.

Chang, M., Roman-Lantzy, C., O'Neil, S., Reid, M., and Borchert, M. (2022). Validity and reliability of CVI Range assessment for Clinical Research (CVI Range-CR): A longitudinal cohort study. *BMJ Open Ophthalmology*, *7*(1), e001144. doi: 10.1136/bmjophth-2022-001144.

Chokron, S., and Dutton, G. (2016). Impact of cerebral visual impairments on motor skills: Implications for developmental coordination disorders. *Frontiers in Psychology*, *7*, 1471. doi: 10.3389/fpsyg.2016.01471.

Chokron, S., Kovarski, K., Zalla, T., and Dutton, G. (2020). The inter-relationships between cerebral visual impairment, autism and intellectual disability. *Neuroscience and Biobehavioral Reviews*, *114*, 201-210.

Chorna, O., Guzzetta, A., and Maitre, N. (2017). Vision assessments and interventions for infants 0-2 years at high risk for cerebral palsy: a systematic review. *Pediatric Neurology*, *76*, 3-13.

Chowdhury, M., and Turin, T. (2020). Variable selection strategies and its importance in clinical prediction modelling. *Family Medicine and Community Health*, *8, e000262.* doi: 10.1136/fmch-2019-000262.

Christensen, R. (2022). Ordinal---regression models for ordinal data. R package version 2022.11-16. https://cran.r-project.org/package=ordinal.

Ciman, M., Gaggi, O., Sgaramella, T., Nota, L., Bortoluzzi, M., and Pinello, L. (2018). Serious games to support cognitive development in children with cerebral visual impairment. *Mobile Networks and Applications, 23*, 1703–1714.

Clark, R., Williams, C., and Gilchrist, I. (2021). Oculomotor control in children with special educational needs (SEN): The development and piloting of a novel app-based therapeutic intervention. *Health and Technology*, *11*(*1*), 919-928.

Colenbrander, A. (2010). Assessment of functional vision and its rehabilitation. *Acta Ophthalmologica*, *88*(2), 163-173.

Cooper, E., and Mackey, A. (2016). Sensory and cognitive plasticity: implications for academic interventions. *Current Opinion in Behavioral Sciences*, *10*, 21-27.

Daibert-Nido, M., Pyatova, Y., Markowitz, M., Taheri-Shirazi, M., and Markowitz, S. (2021). Post audio-visual biofeedback training visual functions and quality of life in paediatric idiopathic infantile nystagmus: A pilot study. *European Journal of Ophthalmology*, *31*(6), 3324-3331.

Delaney, H., and Maxwell, S. (1981). On using analysis of covariance in repeated measures designs. *Multivariate Behavioral Research*, *16*, 105-123.

Delay, A., Rice, M., Bush, E., and Harpster, K. (2022). Interventions for children with cerebral visual impairment: a scoping review. *Developmental Medicine and Child Neurology*, *65*(4), 469-478.

Deverell, L. (2011). O&M environmental complexity scale. *Vision Rehabilitation International*, *4*(1), 64-77.

Deverell, L. (2016). *Functional vision research: Measuring vision-related outcomes in orientation and mobility – VROOM* (Doctoral dissertation). University of Melbourne, Melbourne, Australia. Retrieved from: https://minerva-access.unimelb.edu.au/handle/11343/116129.

Dutton, G., and Bax, M. (Eds.). (2010). *Visual Impairment in Children Due to Damage to the Brain*. London, United Kingdom: Mac Keith Press.

Elsman, E., van Nispen, R., Maria, G., and van Rens, B. (2017). Feasibility of the participation and activity inventory for children and youth (PAI-CY) and young adults (PAI-YA) with a visual impairment: a pilot study. *Health and Quality of Life Outcomes*, *15*, 98. doi: 10.1186/s12955-017-0677-x.

García-Ormaechea, I., González, I., Duplá, M., Andres, E., and Pueyo, V. (2014). Validation of the Preverbal Visual Assessment (PreViAs) questionnaire. *Early Human Development*, *90*(10), 635-638.

Good, W., Jan, J., Burden, S., Skoczenski, A., and Candy, R. (2001). Recent advances in cortical visual impairment. *Developmental Medicine and Child Neurology*, *43*(1), 56-60.

Goodenough, T., Pease, A., and Williams, C. (2021). Bridging the gap: parent and child perspectives of living with cerebral visual impairments. *Frontiers in Human Neuroscience*, *15*, 689683. doi: 10.3389/fnhum.2021.689683.

Gorrie, F., Goodall, K., Rush, R., and Ravenscroft, J. (2019). Towards population screening for cerebral visual impairment: Validity of the five questions and the CVI questionnaire. *PLoS One*, *14*(3), e0214290. doi: 10.1371/journal.pone.0214290.

Gothwal, V., Sumalini, R., Bharani, S., Reddy, S., and Bagga, D. (2012). The second version of the L. V. Prasad-functional vision questionnaire. *Optometry and Vision Science*, *89*(11), 1601-1610.

Heinze, G. Wallisch, C., and Dunkler, D. (2018). Variable selection – A review and recommendations for the practicing statistician. *Biometrical Journal*, *60*(3), 431-449.

Hoyt, C. (2003). Visual function in the brain-damaged child. *Eye (London)*, *17*, 371-386.

Huo, R., Burden, S., Hoyt, C., and Good, W. (1999). Chronic cortical visual impairment in children: aetiology, prognosis, and associated neurological deficits. *British Journal of Ophthalmology*, *83*(6), 670-675.

Huurneman, B., and Goossens, J. (2021). Broad and long-lasting vision improvements in youth with infantile nystagmus after home training with a perceptual learning app. *Frontiers in Neuroscience*, *15*. doi: 10.3389/fnins.2021.651205.

Hyvärinen, L., Walthes, R., Freitag, C., and Petz, V. (2012). Profile of visual functioning as a bridge between education and medicine in the assessment of impaired vision. *Strabismus*, *20*(2), 63-68.

Ivanov, I., Kuester, S., MacKeben, M., Krumm, A., Haaga, M., Staudt, M., Cordey, A., Gehrlich, C., Martus, P., and Trauzettel-Klosinski, S. (2018). Effects of visual search training in children with hemianopia. *PLoS One*, *13*(*7*), e0197285. doi: 10.1371/journal.pone.0197285.

Jacobson, L., Flodmark, Y., and Ek, U. (2009). Visual and perceptual characteristics, ocular motility and strabismus in children with periventricular leukomalacia. *Strabismus*, *10*(2), 179-183.

Johnson, D., and Creech, J. (1983). Ordinal measures in multiple indicator models: A simulation study of categorization error. *American Sociological Review*, *48*, 398-407.

Khadka, J., Ryan, B., Margrain, T., Court, H., and Woodhouse, M. (2010). Development of the 25-item Cardiff visual ability questionnaire for children (CVAQC). *British Journal of Ophthalmology*, *94*, 730-735.

Khetpal, V., and Donahue, S. (2007). Cortical visual impairment: Etiology, associated findings, and prognosis in a tertiary care setting. *Journal of American Association for Pediatric Ophthalmology and Strabismus*, *11*(3), 235-239.

Kleiman, E. (2021). EMAtools: Data Management Tools for Real-Time Monitoring/Ecological Momentary Assessment Data. R package version 0.1.4. https://cran.r-project.org/package=EMAtools.

Kooiker, M., van der Linden, Y., van Dijk, J., van der Zee, Y., Swarte, R., Smit, L., van der Steen-Kant, S., Loudon, S., Reiss, I., Kuyper, K., Pel, J., and van der Steen, J. (2020). Early intervention for children at risk of visual processing dysfunctions from 1 year of age: a randomized controlled trial protocol. *Trials*, *21*(1), 44. doi: 10.1186/s13063-019-3936-9.

Kooiker, M., van der Steen, J., and Pel, J. (2014). Reliability of visual orienting response measures in children with and without visual impairments. *Journal of Neuroscience Methods*, *233*, 54-62.

Lüdecke, D., Ben-Shachar, M., Patil, I., Waggoner, P., and Makowski, D. (2021). performance: An R package for assessment, comparison and testing of statistical models. *Journal of Open Source Software*, *6*(60), 3139. doi:10.21105/joss.03139.

Lueck, A. (2004). *Functional vision: A practitioner's guide to evaluation and intervention*. New York, New York: American Foundation for the Blind Press.

Lueck, A., and Dutton, G. (2015). *Vision and the Brain: Understanding Cerebral Visual Impairment in Children*. New York, NY: American Foundation for the Blind Press.

Lueck, A., Harpster, K., Hartmann, E., Tracy, H., Jones, M., Legge, R., Kran, B., Ravenscroft, J., and Bauer, C. (2023). Toward removing barriers in the evaluation, diagnosis, and care of individuals with cerebral visual impairments. *Policy Insights from the Behavioral and Brain Sciences*, *10*(2), 296-307.

Martin, M., Santos-Lozano, A., Martin-Hernández, J., López-Miguel, A., Maldonado, M., Baladrón, C., Bauer, C., and Merabet, L. (2016). Cerebral versus ocular visual impairment: The impact on developmental neuroplasticity. *Frontiers in Psychology*, *7*, 1958. doi: 10.3389/fpsyg.2016.01958.

Matsuba, C., and Jan, J. (2006). Long-term outcome of children with cortical visual impairment. *Developmental Medicine and Child Neurology*, *48*(*6*), 508-512.

McDowell, N., and Budd, J. (2018). The perspectives of teachers and paraeducators on the relationship between classroom clutter and learning experiences for students with cerebral visual impairment. *Journal of Visual Impairment and Blindness*, *112*(*3*), 248-260.

McDowell, N. (2020a). A pilot study of the Austin Playing Card Assessment: detect and find the degree of visual perceptual difficulties related to clutter. *British Journal of Visual Impairment*, *38*(*2*), 118-136.

McDowell, N. (2020b). Power is knowledge: empowering parents of children with cerebral visual impairment. *Disability and Society.* doi: 10.1080/09687599.2020.1751586

McDowell, N. (2021). A review of the literature to inform the development of a practice framework for supporting children with cerebral visual impairment (CVI). *International Journal of Inclusive Education*. Advance online publication. doi: 10.1080/13603116.2020.1867381

McLinden, M., Douglas, G., Cobb, R., Hewett, R., and Ravenscroft, J. (2016). 'Access to learning' and 'learning to access': Analysing the distinctive role of specialist teachers of children and young people with vision impairments in facilitating curriculum access through an ecological systems theory. *British Journal of Visual Impairment*, *34*(*2*), 177-195.

Mensah, N., Savage, R., Camejo, M., Kramer, S., Lozada, A., McAllister, T., Romanelli, L., Sanchez, L., Schneider, L., and Donohue, P. (2019). Does multidisciplinary and intense rehabilitation in a post-acute brain injury school produce positive outcomes? *JSM Pediatrics and Child Health*, *4*, 1-11. Retrieved from: https://www.jsmcentral.org/Pediatrics.

Nakagawa, S., Johnson, P., and Schielzeth, H. (2017). The coefficient of determination $R^2$ and intra-class correlation from generalized linear mixed-effecs models revisited and expanded. *Journal of the Royal Society Interface*, *14*(134), 20170213. doi: 10.1098/rsif.2017.0213.

Newcomb, S. (2009). *The reliability of the CVI Range – a functional vision assessment for children with cortical visual impairment*. (Doctoral dissertation, University of Maryland, College Park, United States). Retrieved from: https://drum.lib.umd.edu/handle/1903/9123.

Newcomb, S. (2010). The reliability of the CVI Range: A functional vision assessment for children with cortical visual impairment. *Journal of Visual Impairment and Blindness*, *104*(10), 637-647.

Norman, G. (2010). Likert scales, levels of measurement and the "laws" of statistics. *Advances in Health Sciences Education*, *15*(5), 625-632.

Ogletree, B., Bull, J., Drew, R., and Lunnen, K. (2001). Team-based service delivery for students with disabilities: Practice options and guidelines for success. *Intervention in School and Clinic*, *36*(3), 138-145.

Ortibus, E., de Cock, P., and Lagae, L. (2011). Visual perception in preterm children: what are we currently measuring? *Pediatric Neurology*, *45*(1), 1-10.

Overbeek, M., Stokla-Wulfse, M., Lievense, P., Kruithof, Y., Pilon, F., and Kef, S. (2022). A mixed-methods study into the effect of a psycho-educational programme for children with cerebral visual impairment (CVI). *British Journal of Visual Impairment*. Advance online publication. doi: 10.1177/02646196221139779.

Ozturk, T., Er, D., Yaman, A., and Berk, T. (2016). Changing trends over the last decade in the aetiology of childhood blindness: a study from a tertiary referral centre. *British Journal of Ophthalmology*, *100*(*2*), 166-171.

Pease, A., Goodenough, T., Roe, J., Rogers, S., and Williams, C. (2020). Perspectives of primary school staff who work with children with additional needs: Insights may help to improve support for visually impaired children. *British Journal of Visual Impairment.* Advance online publication. doi: 10.1177/0264619620935948.

Philip, J., Huurneman, B., Jansonius, N., Cillessen, A., and Boonstra, F. (2023). Childhood cerebral visual impairment subtype classification based on an extensive versus a limited test battery. *Frontiers in Neuroscience*, 17. doi: 10.3389/fnins.2023.1266201.

Pilling, R., and Little, S. (2020). Evaluation of the role of the colour tent in vision stimulation for children with complex disabilities and cerebral visual impairment: A feasibility study. *British Journal of Visual Impairment*, *38*(1), 104-114.

Posit Team. (2023). RStudio: Integrated development environment for R. Posit Software, PBC, Boston, MA. https://www.posit.co/.

R Core Team. (2022). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. https://www.R-project.org/.

Rahi, J., Cable, N., and British Childhood Visual Impairment Study Group. (2003). Severe visual impairment and blindness in children in the UK. *Lancet*, *362*(9393), 1359-1365.

Ravenscroft, J., and Lueck, A. [John Ravenscroft]. (2020, May 7th). *Conversations about CVI with Professor Amanda Lueck* [Video file]. Retrieved from https://media.ed.ac.uk/media/Conversations+about+CVI+with+Professor+Amanda+Lueck./1_6tfb8wnk/160793481.

Roman-Lantzy, C. (2007). *Cortical Visual Impairment: An Approach to Assessment and Intervention*. New York, New York: American Foundation for the Blind Press.

Roman-Lantzy, C., and Lantzy, A. (2010). Outcomes and opportunities: A study of children with cortical visual impairment. *Journal of Visual Impairment and Blindness*, *104*(10), 649-653.

Roman-Lantzy, C. (2018). *Cortical Visual Impairment: An Approach to Assessment and Intervention* (2nd edition). New York, New York: American Foundation for the Blind Press.

Roman-Lantzy, C. (2019). *Cortical Visual Impairment: Advanced Principles*. Louisville, Kentucky: American Printing House Press.

Sakki, H. (2018). *Development of a new classification system and proposed assessment protocol for childhood cerebral visual disorders*. (Doctoral dissertation, University College London, London, United Kingdom). Retrieved from: https://discovery.ucl.ac.uk/id/eprint/10063803.

Sakki, H., Bowman, R., Sargent, J., Kukadia, R., and Dale, N. (2020). Visual function subtyping in children with early-onset cerebral visual impairment. *Developmental Medicine and Child Neurology*, *63*(3), 303-312.

Sakki, H., Dale, N., Sargent, J., Perez-Roche, T., and Bowman, R. (2018). Is there consensus in defining childhood cerebral visual impairment? A systematic review of terminology and definitions. *British Journal of Ophthalmology*, *102*(4), 424-432.

Salihodžic, H., Zilberburg, K., Chakhmaghi, N., Güldenpfennig, F., Fikar, P., and Ganhör, R. (2018). LightSight: A Dice to Meet the Eyes. In *DIS '18 Companion: Proceedings of the 2018 ACM Conference Companion Publication on Designing Interactive Systems, May 2018*, 331–334. doi: 10.1145/3197391.3205399.

Siu, C., and Murphy, K. (2018). The development of human visual cortex and clinical implications. *Eye and Brain*, *10*, 25-36. doi: 10.2147/EB.S130893

Solebo, A., and Rahi, J. (2014). Epidemiology, aetiology and management of visual impairment in children. *Archives of Disease in Childhood*, *99*(4), 375-379.

Sonksen, P., and Dale, N. (2002). Visual impairment in infancy: Impact on neurodevelopmental and neurobiological processes. *Developmental Medicine and Child Neurology*, *44*(11), 782-791.

Sullivan, G. and Artino Jr., A. (2013). Analyzing and interpreting data from Likert-type scales. *Journal of Graduate Medical Education*, *5*(4), 541-542.

Tadić, V., Cooper, A., Cumberland, P., Lewando-Hundt, G., Rahi, J., and Vision-related Quality of Life Group. (2013). Development of the functional vision questionnaire for children and young people with visual impairment: the FVQ_CYP. *Ophthalmology*, *120*(12), 2725-2732.

Tarquinio, C., Kivits, J., Minary, L., Coste, J., and Alla, F. (2015). Evaluating complex interventions: Perspectives and issues for health behaviour change interventions. *Psychology and Health*, *30*(1), 35-51.

Tunay, Z., Ustunyurt, Z., and Idil, A. (2020). Causes of severe visual impairment in infants and methods of management. *Eye (London)*. Advance online publication. doi: 10.1038/s41433-020-1101-z.

Vancleef, K., Janssens, E., Petré, Y., Wagemans, J., and Ortibus, E. (2020a). Assessment tool for visual perception deficits in cerebral visual impairment: Development and normative data of typically developing children. *Developmental Medicine & Child Neurology*, *62*(1), 111-117.

Vancleef, K., Janssens, E., Petré, Y., Wagemans, J., and Ortibus, E. (2020b). Assessment tool for visual perception deficits in cerebral visual impairment: Reliability and validity. *Developmental Medicine & Child Neurology*, *62*(1), 118-124.

Waddington, J., Linehan, C., Gerling, K., Hicks, K., and Hodgson, T. (2015, April). Participatory design of therapeutic video games for young people with neurological vision impairment. In: *CHI '15 Proceedings of the 33rd Annual ACM Conference on Human Factors in Computing Systems*. Paper presented at CHI 2015: The ACM CH Conference on Human Factors in Computing Systems, Seoul Republic of Korea (pp.3533-3542). New York, NY: ACM Press.

Waddington, J., and Hodgson, T. (2017). Review of rehabilitation and habilitation strategies for children and young people with homonymous visual field loss caused by cerebral vision impairment. *British Journal of Visual Impairment*, *35*(3), 197-210.

Waddington, J., Linehan, C., Gerling, K., Williams, C., Robson, L., Ellis, R., and Hodgson, T. (2018). Evaluation of Eyelander, a video game designed to engage children and young people with homonymous visual field loss in compensatory training. *Journal of Visual Impairment and Blindness*, *112*(6), 717-730.

Waddington, J., Pickering, J., and Hodgson, T. (2020). The Table-top Visual Search Ability Test for Children and young people: Normative response time data from typically developing children. *British Journal of Visual Impairment*, *39*(2), 117-130.

Walker, E., Redfern, A., and Oleson, J. (2019). Linear mixed-model analysis to examine longitudinal trajectories in vocabulary depth and breadth in children who are hard of hearing. *Journal of Speech, Language, and Hearing Research*, *62*(3), 525-542.

Williams, C., Northstone, K., Sabates, R., Feinstein, L., Emond, A., and Dutton, G. (2011). Visual perceptual difficulties and under-achievement at school in a large community-based sample of children. *PLoS ONE*, *6*(3), e14772. doi: 10.1371/journal.pone.0014772.

Williams, C., Northstone, K., Borwick, C., Roe, J., Howard, S., Rogers, S., Amos, J., and Woodhouse, J. (2014). How to help children with neurodevelopmental and visual problems: a scoping review. *British Journal of Ophthalmology*, *98*(1), 6-12.

Williams, C., Pease, A., Goodenough, T., Breheny, K., Gaunt, D., Sinai, P., and Watanabe, R. (2021). Improving outcomes for primary school children at risk of cerebral visual impairment (the CVI project): protocol of a feasibility study for a cluster-randomised controlled trial and health economic evaluation. *BMJ Open*, *11*(5), e044830. doi: 10.1136/bmjopen-2020-044830

Williams, C., Pease, A., Warnes, P., Harrison, S., Pilon, F., Hyvarinen, L., West, S., Self, J., Ferris, J., and CVI Prevalence Study Group. (2021). Cerebral visual impairment-related vision problems in primary school children: a cross-sectional survey. *Developmental Medicine and Child Neurology*. Advance online publication. doi: 10.1111/dmcn.14819.

Wilton, G., Woodhouse, R., Vinuela-Navarro, V., England, R., and Woodhouse, M. (2021). Behavioural features of cerebral visual impairment are common in children with Down syndrome. *Frontiers in Human Neuroscience*, *15*, 673342. doi: 10.3389/fnhum.2021.673342.

World Health Organization. (2019). *Internation statistical classification of diseases and related health problems* (11th ed.). Retrieved from: https://icd.who.int/browse11/l-m/en#/http%253a%252f%252fid.who.int%252ficd%252fentity%252f1103667651

Zihl, J., and Dutton, G. (2015). *Cerebral Visual Impairment in Children*. Vienna, Austria: Springer-Verlag.

Zumbo, B., and Zimmerman, D. (1993). Is the selection of statistical methods governed by level of measurement? *Canadian Psychology*, *34*, 390-400.

**Table 1**

*Mixed-effects ordinal regression of Duration on CVI Range Rating 1*

| Effect | Estimate | 95% CI | | *p* | *SE* |
|---|---|---|---|---|---|
| | | *LL* | *UL* | | |
| Fixed effects | | | | | |
| $Duration_1$ (years) [a] | 5.13 | 1.60 | 8.67 | .004 | 1.80 |
| $Duration_2$ (years) [b] | −0.27 | −5.29 | 4.76 | .917 | 2.57 |
| Random effects | | | | | |
| $\tau_{00}$ | 7.93 | | | | |
| $\tau_{11}$ | 3.02 | | | | |
| $\rho$ | .99 | | | | |
| Threshold coefficients | | | | | |
| CVIRR1 ≤ 3 | −13.6 | −20.9 | −6.37 | | 3.71 |
| CVIRR1 ≤ 4 | −9.94 | −16.2 | −3.63 | | 3.22 |
| CVIRR1 ≤ 5 | −6.87 | −12.6 | −1.09 | | 2.94 |

| | | | | |
|---|---|---|---|---|
| CVIRR1 ≤ 6 | −2.91 | −7.52 | 1.70 | 2.35 |
| CVIRR1 ≤ 7 | 2.79 | −1.07 | 6.65 | 1.97 |
| CVIRR1 ≤ 8 | 11.2 | 4.18 | 18.2 | 3.57 |
| CVIRR1 ≤ 9 | 15.5 | 5.34 | 25.7 | 5.19 |

*Note*. A logit cumulative link mixed model was fitted with the Laplace approximation, allowing the $\text{Duration}_1$ effect on CVIRR1 scores to vary for each student randomly. Fixed effect and threshold coefficient estimates are presented on the log odds scale. $\tau_{00}$ = deviance of random intercept, $\tau_{11}$ = deviance of random slope, $\rho$ = correlation of random intercepts and slopes. *SE* = standard error of estimates.

[a] Cluster-mean centred level 1 predictor.

[b] Grand-mean centred aggregate (level 2) predictor.

**Table 2**

*Mixed-effects linear regression of Duration on CVI Range Rating 2*

| Effect | Estimate | 95% CI | | *p* | *SE* | *d* |
|---|---|---|---|---|---|---|
| | | *LL* | *UL* | | | |
| Fixed effects | | | | | | |
| Intercept | 5.98 | 5.42 | 6.55 | <.001 | 0.28 | |
| $Duration_1$ (years) [a] | 0.78 | 0.60 | 0.97 | <.001 | 0.09 | 3.72 |
| $Duration_2$ (years) [b] | −1.18 | −2.19 | −0.13 | .023 | 0.49 | −0.93 |
| Random effects | | | | | | |
| $\tau_{00}$ | 1.29 | 0.98 | 1.80 | | | |
| $\tau_{11}$ | 0.33 | 0.18 | 0.52 | | | |
| $\rho$ | .62 | .10 | .97 | | | |
| $\sigma$ | 0.45 | 0.35 | 0.59 | | | |

*Note*. A random intercept and slope model was fitted with maximum likelihood, allowing the $\text{Duration}_1$ effect on CVIRR2 scores to vary for each student randomly. $\tau_{00}$ = standard deviation of random intercept, $\tau_{11}$ = standard deviation of random slope, $\rho$ = correlation of random intercepts and slopes, $\sigma$ = residual standard deviation. *d* = Cohen's d effect size calculated using the Satterthwaite approximation to degrees of freedom.

[a] Cluster-mean centred level 1 predictor.

[b] Grand-mean centred aggregate predictor.

**Table 3**

*Mixed-effects linear regression of candidate predictor variables on CVI Range Rating 2*

| Effect | Model 3 | | | | | |
|---|---|---|---|---|---|---|
| | Estimate | 95% CI | | *p* | *SE* | *d* |
| | | *LL* | *UL* | | | |
| Fixed effects | | | | | | |
| Intercept | 5.87 | 4.47 | 7.28 | <.001 | 0.69 | |
| $Duration_1$ (years) [a] | 1.23 | 0.64 | 1.83 | <.001 | 0.29 | 1.81 |
| $Duration_2$ (years) [b] | −0.66 | −1.49 | 0.16 | .114 | 0.40 | −0.66 |
| Acuity (LogMAR) [c] | −1.35 | −2.31 | −0.38 | .009 | 0.47 | −1.16 |
| Age [c] | 0.10 | −0.08 | 0.28 | .257 | 0.09 | 0.48 |
| Assigned sex [d] | −0.20 | −1.07 | 0.67 | .649 | 0.43 | −0.19 |
| Cerebral palsy [e] | 0.98 | −0.10 | 2.06 | .077 | 0.53 | 0.76 |
| Nystagmus [e] | 0.33 | −0.57 | 1.23 | .458 | 0.44 | 0.31 |
| Seizure disorder [e] | 0.27 | −0.67 | 1.21 | .565 | 0.46 | 0.24 |

| | | | | | | |
|---|---|---|---|---|---|---|
| Strabismus [e] | −0.79 | −2.38 | 0.80 | .320 | 0.78 | −0.42 |
| Therapy (hours per year) [c] | 0.00 | −0.01 | 0.02 | .744 | 0.01 | 0.14 |
| Visual field impaired [e] | 1.00 | −0.11 | 2.13 | .079 | 0.55 | 0.75 |
| $Duration_2$ (years) [b] × $Duration_1$ [a] | −0.45 | −0.89 | −0.00 | .047 | 0.22 | −0.74 |
| Acuity (LogMAR) [c] × $Duration_1$ [a] | −0.31 | −0.71 | 0.08 | .121 | 0.19 | −0.74 |
| Age [c] × $Duration_1$ [a] | 0.03 | −0.04 | 0.10 | .358 | 0.03 | 0.36 |
| Assigned sex [d] × $Duration_1$ [a] | −0.32 | −0.75 | 0.10 | .128 | 0.21 | −0.59 |
| Cerebral palsy [e] × $Duration_1$ [a] | −0.08 | −0.56 | 0.37 | .725 | 0.23 | −0.21 |
| Nystagmus [e] × $Duration_1$ [a] | 0.50 | 0.12 | 0.89 | .012 | 0.19 | 1.06 |
| Seizure disorder [e] × $Duration_1$ [a] | 0.36 | −0.05 | 0.77 | .088 | 0.20 | 0.74 |
| Strabismus [e] × $Duration_1$ [a] | −0.61 | −1.25 | 0.01 | .058 | 0.31 | −0.88 |
| Therapy (hours per year) [c] × $Duration_1$ [a] | −0.00 | −0.01 | 0.00 | .327 | 0.00 | −0.39 |
| Visual field impaired [e] × $Duration_1$ [a] | −0.16 | −0.67 | 0.35 | .539 | 0.25 | −0.24 |
| Random effects | | | | | | |
| $\tau_{00}$ | 0.72 | 0.53 | 1.02 | | | |

| | | | |
|---|---|---|---|
| $\tau_{11}$ | 0.19 | 0.02 | 0.34 |
| $\rho$ | .50 | −.76 | 1.00 |
| $\sigma$ | 0.42 | 0.34 | 0.55 |

*Note*. Model 3: A random intercept and slope model was fitted with maximum likelihood, allowing the $\text{Duration}_1$ effect on CVIRR2 scores to vary for each student randomly and allowing all candidate level 2 predictors to interact with the $\text{Duration}_1$ fixed effect on CVIRR2 scores. $\tau_{00}$ = standard deviation of random intercept, $\tau_{11}$ = standard deviation of random slope, $\rho$ = correlation of random intercepts and slopes, $\sigma$ = residual standard deviation. *d* = Cohen's d effect size calculated using the Satterthwaite approximation to degrees of freedom.

[a] Cluster-mean centred level 1 predictor.

[b] Grand-mean centred aggregate predictor.

[c] Grand-mean centred level 2 predictor.

[d] 0 = female, 1 = male.

[e] 0 = absent, 1 = present.