Contents lists available at ScienceDirect



Research in Developmental Disabilities

journal homepage: www.elsevier.com/locate/redevdis



Eyes on CVI: Eye movements unveil distinct visual search patterns in Cerebral Visual Impairment compared to ADHD, dyslexia, and neurotypical children

Marinke J. Hokken^{a,b,*}, Niklas Stein^c, Rob Rodrigues Pereira^d, Ingrid G.I.J.G. Rours^d, Maarten A. Frens^a, Johannes van der Steen^a, Johan J. M. Pel^a, Marlou J.G. Kooiker^{a,b}

^a Erasmus MC, department of Neuroscience, Molewaterplein 40, 3015 GD Rotterdam, the Netherlands

^b Royal Dutch Visio, Amersfoorstestraatweg 180, 1272 RR Huizen, the Netherlands

^c University of Münster, Insitute of Psychology, Fliednerstr. 21, 48149 Münster, NRW, Germany

^d Medical Centre Kinderplein, Rotterdam, Metroplein 88, 3083 BB Rotterdam, the Netherlands

ARTICLE INFO

Keywords: Cerebral visual impairment Visual search Eye-tracking ADHD Dyslexia

ABSTRACT

Visual search problems are often reported in children with Cerebral Visual Impairment (CVI). To tackle the clinical challenge of objectively differentiating CVI from other neurodevelopmental disorders, we developed a novel test battery. Visual search tasks were coupled with verbal and gaze-based measurements. Two search tasks were performed by children with CVI (n: 22; mean age (SD): 9.63 (.46) years) ADHD (n: 32; mean age (SD): 10.51 (.25) years), dyslexia (n: 28; mean age (SD): 10.29 (.20) years) and neurotypical development (n: 44; mean age (SD): 9.30 (.30) years). Children with CVI had more impaired search performance compared to all other groups, especially in crowded and unstructured displays and even when they had normal visual acuity. Indepth gaze-based analyses revealed that this group searched in overall larger areas and needed more time to recognize a target, particularly after their initial fixation on the target. Our gaze-based approach to visual search offers new insights into the distinct search patterns and behaviours of children with CVI. Their tendency to overlook targets whilst fixating on it, point towards higher-order visual function (HOVF) deficits. The novel method is feasible, valid, and promising for clinical differential-diagnostic evaluation between CVI, ADHD and dyslexia, and for informing individualized training.

What this paper adds

This study improves our understanding of the distinct visual search patterns of school-aged children with Cerebral Visual Impairment (CVI). By combining verbal and gaze-based parameters, we gained insight into the underlying processes influencing the impaired visual search performance in children with CVI: 1) Children with CVI benefit more from structured and uncrowded visual materials, 2) Children with CVI often don't recognize the target during the first fixation, 3) Visual search impairments were found even when these children had normal visual acuity. These patterns were not only different than those of neurotypical children, but also than children with ADHD and dyslexia. This finding is particularly valuable for addressing the differential-

* Corresponding author at: Erasmus MC, department of Neuroscience, Molewaterplein 40, 3015 GD Rotterdam, the Netherlands. *E-mail address:* m.hokken@erasmusmc.nl (M.J. Hokken).

https://doi.org/10.1016/j.ridd.2024.104767

Received 1 February 2024; Received in revised form 10 May 2024; Accepted 22 May 2024

Available online 10 June 2024

^{0891-4222/© 2024} The Authors. Published by Elsevier Ltd. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).

diagnostic challenges that are frequently encountered in clinical practice. The present insights are clinically significant and can, besides improving diagnostics, guide the development of individualized training and compensation strategies. The incorporation of non-verbal and non-hand motor parameters is feasible, valid, and promising for future assessments in preverbal children and those with motor disabilities. This inclusive approach may facilitate earlier diagnostics and interventions for children with CVI.

Data Availability

The authors do not have permission to share data.

1. Introduction

Cerebral Visual Impairment (CVI) is an umbrella term for visual deficits arising from abnormal brain development or brain damage to the retrochiasmatic pathways and/or cerebral structures (Sakki et al., 2017). These visual deficits can include lower-order visual function (LOVF) deficits such as decreased visual acuity or visual field impairments and/or higher-order visual function (HOVF) deficits such as deficits in visual selective attention (VSA), visual spatial processing, visual identification, visual memory, visuomotor processing or visual processing speed (Zuidhoek et al., 2015). Given its heterogeneous nature, the diagnostic process of CVI ideally is comprehensive and multidisciplinary. It consists of different stages, starting with screening and referral to specialized diagnostic centres, where ophthalmic or orthoptic LOVF and neuropsychological HOVF examinations are performed (Boonstra et al., 2022; Ortibus et al., 2019). Due to overlapping and comorbid behavioural characteristics, it is currently challenging to distinguish CVI from other neurodevelopmental disorders (Chokron & Dutton, 2023). This challenge impacts most stages of multidisciplinary CVI diagnostics.

The first stage of CVI diagnostics, the screening and referral of children with potential CVI, is generally based on parent-reported daily problems and symptoms. A key characteristic of CVI are daily visual search problems, e.g. struggling to find visual details (a toy or a parent) in cluttered and noisy environments (a toy box or a busy crowd) (Bennett, 2018; McKillop & Dutton, 2008). However, these visual search problems and other visual problems have also been reported in children with ADHD or dyslexia (Hokken et al., 2024a). In turn, it is known that many children with CVI have behaviour and/ or reading problems that resemble those of children with ADHD or dyslexia (Chokron & Dutton, 2016; Chokron & Dutton, 2023; Dutton, 2009). Altogether, the overlap in daily symptoms complicates the referral of children with potential CVI, especially because diagnoses such as ADHD and dyslexia are often more well-known by parents, teachers, and health care professionals.

In the next stage of CVI diagnostics (i.e., the LOVF and HOVF assessments) differentiating between CVI, ADHD and dyslexia remains a common challenge. Importantly, visual search deficits have been reported and observed in children with CVI even when LOVF's, such as visual acuity, are of normal or near normal levels (Chandna et al., 2021; Zhang, 2022; Manley et al., 2022). This emphasizes the importance of HOVF assessments in CVI diagnostics. More specifically, visual selective attention (VSA) is a HOVF that is believed to play an essential role in the visual search for a target among distractors (Wolfe, 2016; Theeuwes, 1993; Theeuwes, 2010). VSA refers to the selection of relevant visual elements within a visual scene, whilst effectively excluding the elements that are irrelevant. The selected elements are further processed and stored in working memory (Desimone & Duncan, 1995; Cotton & Ricker, 2022), facilitating the decision-making process whether the selected visual information contains the target. VSA is typically evaluated using visual search tasks, both in clinical neuropsychological HOVF assessments and in scientific research. In such tasks, children are asked to search for a target amongst distractors in either real-world scenes or abstract displays of, e.g., symbols or shapes. Visual search performance, traditionally graded in terms of speed and accuracy in finding a target, varies with search task design and demands. Two design types have been identified: Feature search tasks, where targets and distractors differ by a single distinguishing feature such as shape or colour, and conjunction search tasks, where distractors share a feature with the target (Wolfe, 2016). It has been demonstrated that conjunction search demands a serial exploration of the scene as it requires to focus on each of the separate visual elements, and therefore more cognitive effort. As a result, conjunction search performance is more affected by task demands such as set size (Treisman & Gelade, 1980). Previous studies showed that patients with CVI, aged 7 to 23 years, were slower and less accurate than neurotypical peers in finding a target on both feature search tasks (Bennett, 2018; Manley et al., 2022; Zhang, 2022; McDowell, 2020; McDowell & Butler, 2023) and conjunction search tasks (Manley et al., 2023). In this group, increased set size and decreased spacing between visual elements in the search display had a detrimental effect on visual search performance (Bennett, 2018; Manley et al., 2022; Zhang, 2022). However, we found through systematic literature review that not only children with CVI, but also children with ADHD and dyslexia may have altered performance on visual search tasks (Hokken et al., 2022). It is important to note three things. First, different search tasks were performed in the three groups. To our knowledge, a direct comparison between children with CVI, ADHD and dyslexia has not been reported. Second, only two studies regarding visual search performance in children with CVI could be included, partially because of the focus on school-aged children between 6 and 12 years. This reveals a need for more research regarding visual search performance in school-aged children with CVI. Third, visual search performance was graded with accuracy and reaction time. These two parameters only report the output of the visual search process, but do not give insight in underlying strategies and patterns during search. More sensitive and distinctive parameters are needed to better discriminate between the search performance of children with CVI, ADHD and dyslexia.

A promising trend in recent years is that researchers have increasingly been exploring the additional value of eye tracking-based parameters during visual search tasks. Such a gaze-based approach may overcome the previously mentioned difficulties in assessing

visual search by revealing underlying mechanisms of visual search functions. Previous eye tracking studies using visual search tasks found prolonged reaction times to and larger search areas before first target fixations in children and young adults with CVI compared to neurotypical peers (Manley et al., 2022; Zhang, 2022; Manley et al., 2023). A recent case study showed that the first target fixation of children with CVI during visual search may be coincidental (Hokken et al., 2024b), which could indicate a lack of conscious recognition of the target (Zihl & Dutton, 2016). Therefore, exploring search strategies and patterns over the total search time could improve our knowledge and discrimination of impaired visual search performance in children with CVI and other neurodevelopmental disorders. In addition, using the reaction time of gaze fixations, rather than verbal responses or hand motor actions, broadens the assessment's inclusivity, by making it suitable for preverbal children and children with Cerebral Palsy (Schenk-Rootlieb, 1994; Uggetti et al., 1996; Pagliano et al., 2007). To this end, we assembled a novel eye tracking-based battery to assess visual attention (Erasmus Eye tracking Visual Attention; EEVA), and now present results on newly developed Conjunction search and Scene search tasks as part of this test battery.

The current study set out to explore whether gaze-based visual search characteristics of children with CVI can be differentiated from those of children with ADHD, dyslexia, and neurotypical children. We hypothesize that the newly developed search tasks are valid compared to conventional search tasks and expect that gaze-based search parameters and patterns can distinguish the groups.

2. Materials and methods

2.1. Participants

We recruited children with CVI, ADHD, dyslexia, and neurotypical children between the age of 6 and 12 years. The children with CVI were recruited through Royal Dutch Visio, a rehabilitation centre for blind and visually impaired people. Information on their demographic, ophthalmic and neurologic characteristics was extracted from the medical records. The children with ADHD were recruited via Medical Centre Kinderplein and AlleskITs, both Dutch mental health care institutions. We recruited children with dyslexia via Regionaal Instituut Dyslexie (RID), a centre of expertise for learning disabilities. In all clinical groups, children had previously been diagnosed by experienced health care professionals. Neurotypical children were recruited through siblings of the clinical groups and co-workers at Erasmus MC and Royal Dutch Visio. Exclusion criteria for all children were a visual acuity below 0.1 decimal, a visual field smaller than 30 degrees, an autism spectrum disorder (ASD) diagnosis, a verbal intelligence below 70 or having a double diagnosis. Prior to participating in the study, the parents or caretakers provided written informed consent. The study was approved by the Medical Ethical Committee of the Erasmus Medical Center Rotterdam (MEC-2020–0680) and adhered to the tenets of the Declaration of Helsinki (2013) involving research with human subjects.

2.2. Procedure and experimental setup

The experiment had a duration of 45 to 60 min in total and took place in a noise-free environment. During the eye tracking-based assessment, participants were seated approximately 60 cm away from a 22.5 or 24-inch screen. The screen resolution was set to 1920



Fig. 1. Design of the Conjunction Search Task (A) and the Scene Search Task (B). Search displays of the Conjunction Search Task were created by the researchers. Search displays of the Scene Search Task were stock images purchased online.

by 1080 pixels or 1920 by 1200 pixels, depending on the screen size. A remote eye-tracker (Tobii Pro X3–120) was attached to the centre of the bottom of the screen to record the eye movements. The eye-tracker registered the movement of both eyes at 120 Hz and automatically compensated for movements of the head. The EEVA-S battery, consisting of several visual search tasks, were presented on the screen in a predetermined order. Each task started with a standardized nine-point calibration procedure. Participants were instructed to minimize head movements as much as possible during the experiment. If significant head movements occurred, the researcher could decide to restart the calibration.

2.2.1. Verbal comprehension index

The verbal intelligence of the participants was evaluated by two subtests of the Dutch version of the Wechsler Intelligence Scale for Children (WISC-V-NL): Similarities and Vocabulary (Wechsler, 2017). The subtest scores enabled the computation of the participants' Verbal Comprehension Index (VCI), which was normalized for age.

2.2.2. Pen-and-paper search task

The standardized pen-and-paper visual search task *Map Mission* of the Test of Everyday Attention for Children (TEA-Ch) battery, commonly used in clinical diagnostic settings, was included to assess VSA the conventional way. Participants were instructed to circle as many symbols of restaurants as possible on a map of Philadelphia. Search efficiency was assessed by counting the number of correctly circled symbols within one minute (Manly, 1999).

2.2.3. EEVA-S battery: Erasmus Eye tracking tasks for Visual Attention - Search

This battery consists of several VSA tasks, of which two are analysed in the current study (Fig. 1). In the *Conjunction Search Task* participants were instructed to identify a red circle that was surrounded by red squares and blue circles. The target circle was first presented in the left upper corner for 1.5 s, followed by the search display. During the trials, the researcher observed whether the participant fixated in the target area. Participants were asked to say Yes when they had found the target. The researcher then pressed the space bar. The task consisted of eight trials, divided into three conditions of scene complexity: display pattern (Structured or unstructured), background (white or illustrated) and set size (low or high) (Fig. 1a).

The *Scene Search Task* involved five search displays that were drawings of real-world scenes, including a bus station, supermarket, bedroom, school desk and traffic situation (Fig. 1b). Participants were verbally asked to search for a specific item, at the same time the name of the item was presented on the screen. Next, the search display was shown. Participants were asked to verbally respond with Yes' when they had found the target, at which point the researcher pressed the space bar.

2.3. Gaze data processing and analysis

During the search tasks, the percentage of gaze points on the screen was calculated per trial as a measure of overall attention toward the screen. Visual search performance was measured by calculating the search time and the percentage of the screen area the participant had searched in, e.g. visual search area. A fixation filter was applied on the combined eye tracking signal from both eyes using an algorithm proposed by Engbert and Kliegl (2003). We determined the first target fixation by the start of the first fixation in the area with a radius of 2,5 degree around the target, i.e., the target area. A valid target fixation had to exceed a length of 150 ms in the target area. Fixations up until 300 ms after trial start were not considered for analyses, to exclude trials in which participants unintentionally started their search in the target area without perceiving it. Consecutive target fixations within 400 ms were counted as one consecutive target fixation. All in all, we calculated three different time intervals, presented in Fig. 2: 1) Verbal response time (VRT) was defined as the time from the start of the trial until the participant verbally indicated the target had been found (represented by a key press). 2) Visual search time (VST) was then defined as the search time until first target fixation. Recognition and decision time (RDT) was defined as the time between the first target fixation and the VRT.

To measure the searched area in each trial, we drew a circle with a radius of 2,5 degree around each detected fixation and marked all pixels within as visited, to determine the proportion of visited pixels as a percentage of the display during both the visual search time (VST area) and during recognition decision time (RDT area).



Verbal Response Time

Fig. 2. Time-based response parameters of the EEVA Conjunction Search Task and Scene Search Task.

2.4. Statistical analysis

Pearson correlation coefficients were computed to assess the linear relationship between age, VCI, visual acuity and search tasks performance, quantified by the number of found targets in Map Mission and the VRT of the eye tracking tasks.

An analysis of variance (ANOVA) was conducted to compare groups on descriptive factors and outcomes from both paper-pencil and eye tracking search tasks. For eye tracking outcomes, time intervals were log-transformed and a Welch's ANOVA with Games-Howell corrections for unequal variances was performed, as tests for normality and homogeneity of variance indicated violations of assumptions. Effect sizes were reported as partial eta squared and statistical significance was set at p < .05. Differences in search tasks outcomes between the children with ADHD with and without use of medication were calculated.

3. Results

3.1. Group characteristics

Twenty-two (22) children with CVI (M = 9.63 years ± 0.46 SD; 11 girls) 32 children with ADHD (M = 10.51 years ± 0.25 SD; 12 girls), 28 children with dyslexia (M = 10.29 years ± 0.20 SD; 14 girls) and 44 neurotypical children (M = 9.30 years ± 0.30 SD; 22 girls) participated. Twelve (12) children with ADHD (37.5 %) had taken medication prior to the assessment, 20 children with ADHD (62.5 %) did not.

Table 1 presents the characteristics of the participant groups. Age significantly differed between the groups (F = 3.55, p = 0.017, $\eta^2 = .08$): Children with ADHD were significantly older compared to the neurotypical children (p = .024). No age-differences between the other groups were found. Thus, further analyses comparing children with ADHD and neurotypical children should be interpreted with caution. An overall significant effect was found for verbal intelligence (F = 3.55, p = .016, $\eta^2 = .08$). However no between group differences were found in post-hoc comparisons. The mean verbal intelligence is clinically interpreted as 'average' for all groups. No differences across the groups were found with respect to sex ($\chi^2 = 1.31$, p = .73).

Table 2 summarises the neurologic and ophthalmic examinations of the CVI-group. Half of the children were born preterm. Neurologic findings were heterogeneous. Visual sensory deficits were present in the minority of the CVI group. For example, only 23 % of the children with CVI had a reduced or suboptimal visual acuity. However, all participating children with CVI had HOVF deficits, assessed by a neuropsychologist. Parents of the children with ADHD, dyslexia and neurotypical children did not report any eye problems nor visits to an ophthalmologist.

3.2. Visual search performance and gaze-based outcomes

The performance on the three search tasks significantly correlated with each other (Map Mission and Conjunction Search Task (r = -4.66, p < .001), Map Mission and Scene Search Task (r = -.355, p < .001), Conjunction Search Task and Scene Search Task (r = .450, p < .001)). Age significantly correlated with Map Mission (r = .55, p < .001) and Conjunction Search Task (r = -.366, p < .001) performance, but not with performance on the Scene Search Task (r = -.164, p = .065). VIQ did not correlate with Map Mission (r = .111, p = .216) and Conjunction Search Task (r = -.26, p = .029).

3.2.1. Map mission

The groups significantly differed on the Map Mission search task, both in the raw scores (F = 18.39, p = <.001, $\eta^2 =.31$) and agerelated norm scores (F = 21.16, p = <.001, $\eta^2 =.34$). Children with CVI found significantly less targets ($M = 12.23 \pm 1.56$) compared to all other groups (M > 23.11), even when corrected for age (all p's < .001). Children with ADHD found more targets ($M = 28.44 \pm 1.55$) compared to neurotypical children ($M = 23.11 \pm 1.26$, p = .039). However, this finding was likely the result of the earlier mentioned age-differences, as no difference was found between children with ADHD and neurotypical children on age-related norm scores (ADHD: $M = 9.38 \pm .47$; neurotypical: $M = 9.15 \pm .39$, p = 1.00).

3.2.2. Conjunction search

3.2.2.1. Gaze data quality. All participants completed the task and gaze-data were successfully recorded for all participants on at least two trials. Of the eight trials of the task, gaze-data was successfully recorded in all trials for 81.75 % of the participants, and in seven out of eight trials in 14.28 % of the participants. With respect to the percentage of gaze samples on the screen was 81.19 (2.14)% for the

Group characteristics.

	CVI (n = 22)	ADHD (n = 32)	Dyslexia (n = 28)	Neurotypical (n = 44)
Age: M(sd)	9.63 (.46)	10.51 (.25)	10.29 (.20)	9.30 (.30)
Girls: N (%)	11 (50 %)	12 (37.5 %)	14 (50 %)	22 (47.8 %)
VCI: M(sd)	96.41 (2.60)	97.19 (2.35)	102.18 (2.13)	104.48 (1.61)

Table 2	
Clinical data of the	CVI participants ($n = 22$).

	Ν	%
Birth		
Preterm	11	50 %
Term	11	50 %
Neurologic Findings		
Cerebral Palsy	4	18 %
Epilepsy	3	14 %
Hydrocephalus	1	5 %
Periventricular Leukomalacia (PVL)*	1	5 %
Pregnancy/birth complications	12	55 %
Genetic disorder	1	5 %
Traumatic Brain Injury	2	9 %
Brain Tumour	1	5 %
Other/unspecified	3	14 %
Ophthalmic Findings		
Visual Acuity		
Reduced (0.1 – 0.3 decimal)	2	9 %
Suboptimal (0.4 – 0.7 decimal)	3	14 %
Normal (0.8 > decimal)	17	77 %
Visual field restrictions	1	5 %
Nystagmus	1	5 %
Strabismus		
Exotropia	2	9 %
Esotropia	5	23 %
Neuropsychological Findings	22	100 %
HOVF deficits		

(*) please note that for most participating children, no brain imaging data were available. Because of the high prevalence of preterm children and children with pregnancy/birth complications, we suspect a higher percentage of PVL in the participating children than reported. Also note that factors are not mutually exclusive.

CVI-group, 83.91 (1.61)% for the ADHD-group, 89.11 (1.20)% for the dyslexia-group and 86.78 (1.11)% for the neurotypical group. A significant group difference was found (F = 4.26, p = .009, $\eta^2 = .096$). Children with dyslexia spend more time looking at the screen compared to children with CVI (p = .016). No differences between the other groups were found.

3.2.2.2. Conjunction search performance. Fig. 3 presents the group differences in verbal and gaze-based parameters on the Conjunction Search Task. The groups significantly differed with regard to VRT (F = 19.89, p < .001, $\eta^2 = .057$), VST (F = 4.97, p = .002, $\eta^2 = .016$), RDT (F = 22.39, p < .001, $\eta^2 = .069$), VST area (F = 4.12, p = .007, $\eta^2 = .011$) and RDT area (F = 9.59, p < .001, $\eta^2 = .031$). No difference was found between the groups on FTF duration (F = .717, p = .542, $\eta^2 = .003$) (Fig. 3d).

With respect to reaction times, children with CVI had a significantly longer VRT (M= 12.05 ± 1.66 s) compared to all other groups (all *M*'s < 6.47 s; all *p*'s < .001). Children with ADHD had a shorter VRT (M= 4.09 ± .23 s) compared to children with dyslexia (M= 6.46 ± .56 s; p = .006) and neurotypical children (M= 6.47 ± .43 s; p < .001; Fig. 3a). Children with CVI had longer VST (M= 5.55 ± .64 s) compared to children with ADHD (M= 2.82 ± .18 s; p = .002; Fig. 3b). RDT was significantly longer for children with CVI (M= 6.50 ± 1.37 s) compared to all other groups (all M's < 2.79 s; all p's < .001). Children with ADHD (M: 1.27 ± .15 s) had shorter RDT compared to children with dyslexia (M= 2.79 ± .47 s; p = .004) and neurotypical children (M: 2.70 ± .34 s; p < .001; Fig. 3c).

With respect to search areas, we found that children with CVI had larger VST areas (M= 23.38 ± 1.44 %) compared to children with ADHD (M= 19.26 ± .78 %; (p = .011; Fig. 3e). RDT areas were larger in children with CVI (M= 12.89 ± 1.80 %) compared to all other groups (all M's < 7.01 %; all p's < .024). Children with ADHD had smaller RDT areas (M= 3.82 ± .64 %) compared to neurotypical children (M= 6.85 ± .84 %; p = .024; Fig. 3f).

3.2.2.3. Conjunction search task demands. Fig. 4 shows group differences in reaction times for the different task demands. Significant group differences were found for pattern (F = 4.12, p = .010, $\eta^2 = .206$) and set size (F = 10.193, p < .001, $\eta^2 = .121$). A trend for background was found as well (F = 2.50, p = .069, $\eta^2 = .110$). More specific, children with CVI showed a larger difference in VRT ($M = 7.84 \pm 2.17$ s) between an unstructured and a structured display compared to all other groups (all M's < .81 s; all p's < .023; Fig. 4c). A larger VRT difference depending on set size was observed for children with CVI ($M = 12.72 \pm 3.83$ s) compared to all other groups (all M's < 6.80 s), but this was not statistically significant. We found a smaller difference for children with ADHD ($M = 3.10 \pm .34$ s) compared to children with dyslexia ($M = 6.80 \pm .96$ s; p = .006) and neurotypical children ($M = 6.53 \pm .78$ s; p < .001; Fig. 4a). Finally, whilst there is a larger difference between a white and an illustrated background for children with CVI ($M = 6.5 \pm 2.85$ s) compared to all other groups (all M's < 1.72 s; Fig. 4c), this effect did not reach statistical significance.

M.J. Hokken et al.



Fig. 3. Results on verbal and gaze-based parameters in children with CVI, ADHD, dyslexia, and neurotypical children on the conjunction search task. (***) = p < .001, (**) = p < .01, (*) = p < .05.

3.2.3. Scene search

3.2.3.1. *Gaze data quality.* All participants completed the task and gaze data were successfully recorded for all participants on at least two trials. Of the five trials of the task, gaze data was successfully recorded in all trials for 84.92 % of the participants, and in four out of five trials in 11.90 % of the participants. With respect to the percentage of gaze samples on the screen was 80.76 (2.82)% for the CVIgroup, 83.34 (1.89)% for the ADHD-group, 87.86 (1.04)% for the dyslexia group and 87.87 (1.39)% for the neurotypical group. Whilst a significant group difference was found (F = 3.01, p = .034, $\eta^2 = .082$), post-hoc analyses showed no specific group differences with respect to the percentage of gaze samples towards the screen (all p's > .111).

3.2.3.2. Scene search performance. Fig. 5 shows the group differences in verbal and gaze-based parameters on the Scene Search Task. Overall, the groups significantly differed in VRT (F = 18.859, p < .001, $\eta^2 = .128$), RDT (F = 16.39, p < .001, $\eta^2 = .118$), VST area (F = 2.93, p = .034, $\eta^2 = .016$) and RDT area (F = 7.431, p < .001, $\eta^2 = .063$). No differences were found for VST (F = 1.865, p = .136, $\eta^2 = .014$; Fig. 4b) and first target fixation duration (F = .295, p = .829, $\eta^2 = .001$; Fig. 5d).



Fig. 4. The effect of task demands (i.e. visual scene complexity): (A) set size, (B) display pattern or (C) display background on VRT for children with CVI, ADHD, dyslexia, and neurotypical children. (***) = p < .001, (**) = p < .01, (*) = p < .05.

Regarding reaction times, children with CVI had significantly longer VRT (M= 14.84 ± 1.79 s) compared to the other groups (all M's < 7.09 s; all p's < .001; Fig. 5a). Longer RDT were found in children with CVI (M= 8.00 ± 1.30 s) compared to all other groups (all M's < 3.77 s; all p's < .001; Fig. 5c).

Regarding search areas, children with CVI had a larger VST area (M= 22.65 ± 1.95 %) compared to neurotypical children (M= 16.75 ± .77; p = .040; Fig. 5e). Finally, larger RDT areas were found in children with CVI (M= 16.27 ± 2.56 %) compared to all other groups (all M's < 10.35 %; all p's < .001; Fig. 5f).

3.3. Effect of visual acuity and ADHD medication

Fig. 6 presents the relation between visual acuity and performance on the three search tasks. We found a significant positive correlation between visual acuity and performance on all visual search tasks (Map Mission: r = .421, p = .051 (Fig. 6a), Conjunction Search Task: r = -.565, p < .001 (Fig. 6c), Scene Search Task: r = -.541, p = .013 (Fig. 6e)) in the CVI group. This indicates that a higher visual acuity corresponded with better visual search performance in children with CVI. However, when the CVI group was divided into children with a normal visual acuity (>.7 decimal) and children with a suboptimal or reduced visual acuity (<.7 decimal), no group difference was found on Map Mission (CVI VA <.7: $M = 11.13 \pm 1.31$, CVI VA >.7: $M = 12.61 \pm .59$; p = .841), Conjunction Search (CVI VA <.7: $M = 19.75 \pm 5.54$, CVI VA >.7: $M = 9.86 \pm 1.19$; p = .346), and Scene Search (CVI VA <.7: $M = 14.36 \pm 13.87$, CVI VA >.7: $M = 15.81 \pm 1.92$; p = .408). In addition, both groups performed significantly worse compared to children with ADHD, dyslexia, and neurotypical children on all three search tasks (all p's < .009; Figs. 6b, 6d, 6f).

Search performance in children with ADHD was not significantly affected by medication at the time of testing (Map Mission: F = .088, p = .77, η^2 = .003, Conjunction Search Task: F = .082, p = .78, η^2 = .003, Scene Search Task: F = .033, p = .86, η^2 = .001).

4. Discussion

The present study examined and quantified visual search differences between school-aged children with CVI, ADHD, dyslexia, and neurotypical children. This study contributes to the existing body of literature by confirming that children with CVI show considerable worse visual search performance compared to neurotypical children (Bennett et al.; McKillop & Dutton, 2008; Manley et al., 2022; Zhang, 2022; McDowell, 2020; McDowell & Butler, 2023; Manley et al., 2023; Hokken et al., 2024b), but more importantly, that the weaker visual search performance also holds compared to children with ADHD and dyslexia. To illustrate: in comparison to all the other groups, children with CVI located half of the targets in the paper-pencil search task and required twice the amount of time to locate the target on the gaze-based search tasks.

4.1. Visual search impairments in children with CVI

Our gaze-based approach, that went beyond conventional measures as accuracy and (verbal) search time, uncovered a specific need amongst children with CVI for a longer recognition and decision time (RDT) compared to their peers. One possible explanation is that children with CVI need more time to formulate a verbal response after fixating on the target area. However, our findings suggest otherwise, because the search area after the first target fixation (RDT area) was significantly larger in children with CVI. It seems that children with CVI continued searching in the display after they looked at the target area, a pattern we also observed in a case study (Hokken et al., 2024b). Thus, children with CVI may not consciously perceive the target during their first fixation in the target area, despite similar initial fixation durations in the target area as the other groups. Opposed to this search performance after the first target fixation did not discriminate CVI from other groups. This is in contrast with prior research where children with CVI needed more time and a larger area before fixating on the target (Bennett, 2018; Manley et al., 2022; Zhang, 2022). Several methodological factors may explain this discrepancy. The previous studies included older participants and

M.J. Hokken et al.



Fig. 5. Results on verbal and gaze-based parameters in children with CVI, ADHD, dyslexia, and neurotypical children on the scene search task. (***) = p < .001, (**) = p < .01, (*) = p < .05.

employed a longer first target fixation threshold of 450 ms to ensure that their participants successfully identified the target. Also, in these studies a feature search task (e.g., the toy box task) was used, in which a target is more salient compared to the distractors (pop-out effect). Therefore, a longer VST and larger VS areas are more atypical and arguably a characteristic of participants with CVI. On the contrary, in the presented EEVA search tasks the target does not stand out, and therefore all children may need more time and a larger search area before locating the target.

Notably, the performance on the introduced EEVA search tasks was found to correlate with the conventional Map Mission task. This finding validates the EEVA tasks by demonstrating their efficacy in measuring VSA, because Map Mission was designed to evaluate this attention function (Manly, 1999). In addition, besides the understanding that larger search areas in children with CVI imply greater visual difficulty in locating a target, analysing the search areas provides a more detailed insight into VSA. VSA can be expanded over a large area of the visual scene by integrating multiple visual elements simultaneously (global VSA) or limited within a small area, by focussing on one specific visual detail at a time (local VSA). Global VSA, needed when the target stands out or groups of targets are viewed simultaneously, comes with fewer eye movements. Local VSA is deployed when every element of the display needs to be searched, resulting in more eye movements and thus larger search areas (Theeuwes, 2010; Stigchel et al., 2009). In this light, our



Fig. 6. Correlation between Visual Acuity (VA) and search performance for children with CVI (A, C, E) and group comparisons children with CVI (with and without normal visual acuity), ADHD, dyslexia, and neurotypical children on the three search tasks (B, D, F). (***) = p < .001, (*) = p < .01, (*) = p < .05.

findings indicate that children with CVI tend to employ more local search strategies across the search display compared to other groups. This aligns with literature proposing that children with CVI encounter difficulties in perceiving multiple visual elements simultaneously, i.e. lacking visual overview, both during search tasks and in daily life (Dutton et al., 2004; Philip & Dutton, 2014; Pehere et al., 2020). Moreover, the finding that children with CVI more often miss the target whilst fixating on it, implies potential deficits in local VSA, i.e. overlooking visual details, which is also supported by daily experiences (Lam et al., 2010; Bennett, 2018; McKillop & Dutton, 2008). These insights provide a first step towards mapping specific VSA deficits in children with CVI.

4.2. Effect of task and child characteristics on search performance

Consistent with existing research, children with CVI experienced more difficulty with visual search compared to the other groups when the set size of the search display increased (Bennett, 2018; Manley et al., 2022; Zhang, 2022). In addition, the influence of display pattern and background on search time seemed more pronounced in children with CVI compared to the other groups. Whilst the latter has been observed in clinical practice, this has not yet been reported in literature. These findings highlight the discriminative potential of including different task conditions in screening for CVI. They also show the need for adaptions in the (school) materials of children with CVI, given that they seem to benefit more from calm and structured visual presentations.

In addition to task conditions, some participant characteristics influenced search performance. Within our CVI cohort, all children had HOVF deficits whilst 23 % of those children also had a visual acuity below 0.7 decimal. A decreased visual acuity had a negative impact on search performance, suggesting that children with both LOVF and HOVF deficits may experience more pronounced search difficulties in comparison to the CVI population with normal acuity levels. Importantly, however, the children with CVI with normal visual acuity levels also had significantly more search difficulties compared to children with ADHD, dyslexia, or a neurotypical development. This demonstrates the complexity of visual search impairments in CVI beyond the influence of visual acuity alone. Further investigations into the interplay of specific visual deficits within the CVI spectrum are necessary to improve our understanding of the visual search challenges they encounter in daily life.

Our findings are in line with literature showing that conjunction search performance continues to develop until approximately 12 years of age (Donnelly et al., 2007; Woods et al., 2013). Scene search performance, as expected, did not improve with age in our cohort. Moreover, a higher VIQ was associated with a better scene search performance. This finding may be related to the semantic cue provided by naming the target in the scene search task, as opposed to a visual cue presented by displaying the target in the conjunction search task.

4.3. Findings and implications for ADHD and dyslexia

The children with ADHD and dyslexia did not perform worse than the neurotypical children. Instead, Conjunction Search performance was significantly better in children with ADHD compared to neurotypical children. It might be explained by a small but significant difference in age (ADHD 10.51 years > neurotypical 9.30 years). This is also reflected in the results from Map Mission and Scene Search, where no differences were found between children with ADHD and neurotypical children when age did not correlate with performance. It might be that the relative short duration of the tasks improved the performance of children with ADHD. The children with dyslexia mostly show difficulties in finding linguistic stimuli (Hokken et al., 2022), which might explain the absence of search impairments in the present study. A factor that should be considered is that children with overlapping diagnoses were excluded in our study. More research into comparing children with comorbid CVI, ADHD and dyslexia is needed.

4.4. Towards more inclusive and comprehensive CVI diagnostic methods

Although in most children with CVI the risk factors arise around birth or in their first year, these children are often not assessed until six years of age because neuropsychological HOVF assessments are time-consuming and require verbal and motor responses. Integrating eye tracking within neuropsychological assessments, and the feasibility of our tasks in a diverse group of children, are important first steps towards designing non-verbal and non-hand motor screening methods. These gaze-based methods are promising for early screening, and consequently for early interventions, of CVI in young children and children with motor disabilities, such as Cerebral Palsy. In addition, both in young and older children with CVI, gaze-based methods offer new insights in their patterns and strategies during visual search tasks. The possibility to rewatch the gaze-based videos of the search performance with parents or teachers often results in better understanding of the daily difficulties of the child with CVI. For professionals, these results might help achieving a more tailored design of training sessions and compensatory strategies.

4.5. Study strengths and limitations

Important strengths of this study are the direct comparison and discrimination of children with CVI with other clinical groups, and the better understanding of the impaired visual search performance of children with CVI that has been gained. Another strength is the feasibility of the novel tasks as we were able to collect eye movement data for all participants.

The most pressing limitation of this study concerns the generalizability of the results. First, the relatively small sample size, particularly in the CVI group, limits the statistical power of observed group differences. Although the results on the novel gaze-based search tasks are highly promising for differential diagnostic evaluation, more research with larger cohorts is needed to confirm these observational results. Second, the exclusion of children with overlapping diagnoses might limit the generalizability of the study

findings because in clinical practice children often present with comorbid conditions, e.g., CVI and ADHD (Philip & Dutton, 2014; Chokron & Dutton, 2023; Huo et al., 1999). Excluding these children therefore does not fully capture the complexities encountered in real-world diagnostic screenings. Third, the two screen-based tasks used in our study were quite short, which is a great advantage, but all task conditions were only tested once and in a fixed order. Even though the results are promising, future research with these tasks should include more trials and in a random order. Lastly, the scene search tasks had some ecological validity because it involved daily scenes. However, the extent to which these finding generalize to real-world situations remains an open question. Advanced technical developments, such as head-mounted wearable eye trackers, now enable the use of eye tracking in daily situations of the child, which can gain even more insight in their daily visual strategies.

When examining visual attention or processing deficits in children, it is crucial to be aware of potential overlooked cases of CVI in each clinical group. In addition, awareness of the high prevalence of oculomotor impairments in children with neurological conditions associated with CVI, such as hydrocephalus or Cerebral Palsy (Aring et al., 2007; Bayar et al., 2021; Fazzi et al., 2012), is essential when conducting eye tracking studies that have a focus on CVI. Here, we attemted to address both points by screening for oculomotor abnormalities in all groups and for potential CVI within our ADHD and dyslexia groups. However, we cannot completely rule out that either factor could have affected our results and they should be considered in future studies.

5. Conclusion

In this study we investigated the visual search challenges of school-aged children with Cerebral Visual Impairment (CVI) compared to children with ADHD, dyslexia, and neurotypical children. Using a novel gaze-based test battery, we revealed that children with CVI had more visual search impairments, especially when task demands increased and even when their visual acuity was of near normal levels. Particularly striking was that children with CVI did not always consciously perceive the target during fixations in the target area. Higher Order Visual Functions (HOVF), in particular Visual Selective Attention (VSA) deficits, seem to play a crucial role. Despite heterogeneity in results and limitations in sample size, the current findings are an important contribution to the ongoing conversation about the shared and distinct visual characteristics of children with CVI, ADHD and dyslexia. The presented novel and validated search tasks are therefore promising tools for differential diagnostic evaluation. Finally, as groups did not only differ on verbal, but also on gaze-based parameters, eye-tracking tasks have potential for non-verbal and non-hand motor assessments, for example within CVI screening in young children or children with motor disabilities.

Funding

This work was supported by the Visio Foundation [grant number OI0399066].

CRediT authorship contribution statement

Marinke J. Hokken: Writing – review & editing, Writing – original draft, Visualization, Project administration, Methodology, Investigation, Funding acquisition, Formal analysis, Conceptualization. Niklas Stein: Writing – review & editing, Writing – original draft, Software, Formal analysis. Rob Rodrigues Pereira: Writing – review & editing, Resources. Ingrid G.I.J.G. Rours: Writing – review & editing, Resources. Maarten A. Frens: Writing – review & editing, Supervision, Resources. Johannes van der Steen: Writing – review & editing, Supervision, Resources, Funding acquisition. Johan J.M. Pel: Writing – review & editing, Supervision, Resources, Methodology, Conceptualization. Marlou J.G. Kooiker: Writing – review & editing, Supervision, Methodology, Funding acquisition, Conceptualization.

Declaration of Competing Interest

All authors declare that they have no conflicts of interest.

Data Availability

The authors do not have permission to share data.

Acknowledgements

This study is part of research project '*visual selective attention and processing deficits in children*', which is a collaboration between the department of Neuroscience, Erasmus MC and Royal Dutch Visio. We thank all children and their parents who participated in the study, as well as Medical Centre Kinderplein, RID and AlleskITs for their collaboration. We also thank Kirsten Muller for her help in data collection.

References

Aring, E., Andersson, S., Hård, A. L., Hellström, A., Persson, E. K., Uvebrant, P., Ygge, J., & Hellström, A. (2007). Strabismus, binocular functions and ocular motility in children with hydrocephalus. *Strabismus*, 15(2), 79–88.

Bayar, S. A., Ozturker, Z. K., Oto, S., Gokmen, O., & Sezer, T. (2021). Pattern of oculomotor and visual function in children with hydrocephalus. Journal Français d'Ophtalmologie, 44(9), 1340–1348.

Bennett, C. R., et al. (2018). Assessing visual search performance in ocular compared to cerebral visual impairment using a virtual reality simulation of human dynamic movement. in Proceedings of the Technology, Mind, and Society, 1–6.

Boonstra, F. N., Bosch, D. G., Geldof, C. J., Stellingwerf, C., & Porro, G. (2022). The multidisciplinary guidelines for diagnosis and referral in cerebral visual impairment. Frontiers in Human Neuroscience, 16, Article 727565.

Chandna, A., Ghahghaei, S., Foster, S., & Kumar, R. (2021). Higher visual function deficits in children with cerebral visual impairment and good visual acuity. *Frontiers in Human Neuroscience*, 15, Article 711873.

Chokron, S., & Dutton, G. N. (2016). Impact of cerebral visual impairments on motor skills: Implications for developmental coordination disorders. Frontiers in Psychology, 7, 1471.

Chokron, S., & Dutton, G. N. (2023). From vision to cognition: potential contributions of cerebral visual impairment to neurodevelopmental disorders. Journal of Neural Transmission, 130(3), 409–424.

Cotton, K., & Ricker, T. J. (2022). Examining the relationship between working memory consolidation and long-term consolidation. *Psychonomic Bulletin & Review, 29* (5), 1625–1648.

Desimone, R., & Duncan, J. (1995). Neural mechanisms of selective visual attention. Annual Review of Neuroscience, 18(1), 193-222.

Donnelly, N., Cave, K., Greenway, R., Hadwin, J. A., Stevenson, J., & Sonuga-Barke, E. (2007). Visual search in children and adults: Top-down and bottom-up mechanisms. *Quarterly Journal of Experimental Psychology*, 60(1), 120–136.

Dutton, G. N., Saaed, A., Fahad, B., Fraser, R., McDaid, G., McDade, J., & Spowart, K. (2004). Association of binocular lower visual field impairment, impaired simultaneous perception, disordered visually guided motion and inaccurate saccades in children with cerebral visual dysfunction—a retrospective observational study. *Eye*, 18(1), 27–34.

Dutton, G. N. D. M. (2009). 'Dorsal stream dysfunction' and 'dorsal stream dysfunction plus': A potential classification for perceptual visual impairment in the context of cerebral visual impairment? *Developmental Medicine and Child Neurology*, *51*(3), 170.

Engbert, R., & Kliegl, R. (2003). Microsaccades uncover the orientation of covert attention. Vision Research, 43(9), 1035–1045.

Fazzi, E., Signorini, S. G., La Piana, R., Bertone, C., Misefari, W., Galli, J., Balottin, U., & Bianchi, P. E. (2012). Neuro-ophthalmological disorders in cerebral palsy: ophthalmological, oculomotor, and visual aspects. Developmental Medicine & Child Neurology, 54(8), 730–736.

Hokken, M. J., Krabbendam, E., van der Zee, Y. J., & Kooiker, M. J. G. (2022). Visual selective attention and visual search performance in children with CVI, ADHD, and Dyslexia: a scoping review. Child Neuropsychology, 1–34.

Hokken, M. J., van der Žee, Y. J., van der Geest, J. N., & Kooiker, M. J. (2024a). Parent-reported problems in children with Cerebral Visual Impairment: Improving the discriminative ability from ADHD and dyslexia using screening inventories. *Neuropsychological Rehabilitation*, 1–21.

Hokken, M. J., Stein, N., Kooiker, M. J., & Pel, J. J. (2024b). A novel gaze-based visual search task for children with CVI: A twin study. British Journal of Visual Impairment, 02646196241247973.

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

Lam, F. C., Lovett, F., & Dutton, G. N. (2010). Cerebral visual impairment in children: A longitudinal case study of functional outcomes beyond the visual acuities. *Journal of Visual Impairment & Blindness, 104*(10), 625–635.

Manley, C. E., Bauer, C. M., Bex, P. J., & Merabet, L. B. (2023). Impaired visuospatial processing in cerebral visual impairment revealed by performance on a conjunction visual search task. British Journal of Visual Impairment, 02646196231187550.

Manley, C. E., Bennett, C. R., & Merabet, L. B. (2022). Assessing higher-order visual processing in cerebral visual impairment using naturalistic virtual-reality-based visual search tasks. *Children*, 9(8), 1114.

Manly, T., et al. (1999). The test of everyday attention for children (TEA-Ch). Bury St Edmunds, UK: Thames Valley Test Company.

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

McDowell, N., & Butler, P. (2023). Validation of the Austin Assessment: A screening tool for cerebral visual impairment related visual issues. *Plos One, 18*(11), Article e0293904.

McKillop, E., & Dutton, G. N. (2008). Impairment of vision in children due to damage to the brain: A practical approach. British and Irish Orthoptic Journal, 5.

Ortibus, E., Fazzi, E., & Dale, N. (2019). Cerebral visual impairment and clinical assessment: The European perspective (October). In *In Seminars in pediatric neurology* (Vol. 31, pp. 15–24). WB Saunders, (October).

Pagliano, E., Fedrizzi, E., Erbetta, A., Bulgheroni, S., Solari, A., et al. (2007). Cognitive profiles and visuoperceptual abilities in preterm and term spastic diplegic children with periventricular leukomalacia. *Journal of Child Neurology*, *22*, 282–288.

Pehere, N. K., Dutton, G. N., & Mankad, K. (2020). Simultanagnosia as a cause of visual disturbance following posterior reversible encephalopathy syndrome (PRES): a case report. Indian Journal of Ophthalmology, 68(1), 254.

Philip, S. S., & Dutton, G. N. (2014). Identifying and characterising cerebral visual impairment in children: A review. *Clinical and Experimental Optometry*, 97(3), 196–208.

Sakki, H. E., Dale, N. J., Sargent, J., Perez-Roche, T., & Bowman, R. (2017). Is there consensus in defining childhood cerebral visual impairment? A systematic review of terminology and definitions. *British Journal of Ophthalmology*.

Schenk-Rootlieb, A. J. F., et al. (1994). Cerebral visual impairment in cerebral palsy: Relation to structural abnormalities of the cerebrum. *Neuropediatrics*, 25(02), 68–72.

Theeuwes, J. (1993). Visual selective attention: A theoretical analysis. Acta Psychologica, 83(2), 93–154.

Theeuwes, J. (2010). Top-down and bottom-up control of visual selection. Acta Psychologica, 135(2), 77-99.

Treisman, A. M., & Gelade, G. (1980). A feature-integration theory of attention. Cognitive Psychology, 12(1), 97–136.

Uggetti, C., Egitto, M. G., Fazzi, E., Bianchi, P. E., Bergamaschi, R., Zappoli, F., & Lanzi, G. (1996). Cerebral visual impairment in periventricular leukomalacia: MR correlation. *American Journal of Neuroradiology*, 17(5), 979–985.

Van der Stigchel, S., Belopolsky, A. V., Peters, J. C., Wijnen, J. G., Meeter, M., & Theeuwes, J. (2009). The limits of top-down control of visual attention. Acta psychologica, 132(3), 201–212.

Wolfe, J. M. (2016). In Attention. Visual search (pp. 13-73). Psychology Press.

Woods, A. J., Göksun, T., Chatterjee, A., Zelonis, S., Mehta, A., & Smith, S. E. (2013). The development of organized visual search. Acta psychologica, 143(2), 191–199.

Zhang, X., et al. (2022). Assessing visuospatial processing in cerebral visual impairment using a novel and naturalistic static visual search task. Research in Developmental Disabilities, 131, Article 104364.

Zihl, J., & Dutton, G. N. (2016). Development and neurobiological foundations of visual perception. in Cerebral Visual Impairment in Children: Visuoperceptive and Visuocognitive Disorders.

Zuidhoek, S., Hyvärinen, L., Jacob, N., & Henriksen, A. (2015). In A. Hall Lueck, & G. N. Dutton (Eds.), Assessment of Functional Vision: Assessment of Visual Processing in Children with CVI, in Vision and the Brain: Understanding Cerebral Visual Impairment in Children. American Foundation for the Blind Press.