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Visual perception in children and adolescents with visual impairments, 2nd edition

Edited by
Jeff C. Rabin, Corinna M. Bauer and Karen Harpster

Coordinated by
Raelyn Ottenbreit

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Visual perception in children and adolescents with visual impairments, 2nd edition

Topic editors
Jeff C. Rabin — University of the Incarnate Word, United States
Corinna M. Bauer — Massachusetts General Hospital, United States
Karen Harpster — Cincinnati Children's Hospital Medical Center, United States

Topic coordinator
Raelyn Ottenbreit — University of the Incarnate Word, United States

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Table of contents

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Jae Kun Shim,
University of Maryland, College Park,
United States

*CORRESPONDENCE
Corinna M. Bauer
✉ cbauer@mgh.harvard.edu

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Editorial: Visual perception in children and adolescents with visual impairments

Corinna M. Bauer[1,2]*, Jeff C. Rabin[3] and Karen Harpster[4,5]

[1]Massachusetts General Hospital, Boston, MA, United States, [2]Harvard Medical School, Boston, MA, United States, [3]Rosenberg School of Optometry, University of the Incarnate Word, San Antonio, TX, United States, [4]Cincinnati Children's Hospital Medical Center, Cincinnati, OH, United States, [5]University of Cincinnati, Cincinnati, OH, United States

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Editorial on the Research Topic
Visual perception in children and adolescents with visual impairments

Introduction

The use of functional vision to perceive and interact with one's surroundings and the outside world can be compromised in individuals with visual impairments. Thus, visual impairment—whether due to ocular causes, cerebral visual impairment (CVI), or both—may negatively affect one's activities and participation within the domains of the International Classification of Function (Deramore Denver et al., 2016). Therefore, receiving appropriate services, interventions, and/or supports to gain veridical access to the environment is essential for those with visual impairments. The articles in this Research Topic focus on these unique challenges and highlight how appropriate accommodations and interventions may improve meaningful access and quality-of-life.

As outlined by McDowell et al., there remain a number of challenges regarding our understanding of the visual and perceptual dysfunctions and challenges that are experienced by those with CVI. Consequently, they often go unrecognized, resulting in children and/or adults with CVI-related behaviors who may receive inappropriate diagnostic labels and ineffective educational, vocational habilitation, and/or therapeutic interventions that might not be tailored to one's unique needs. This review underscores the necessity of fully characterizing and diagnosing the visual capacities and challenges of the individual, coupled with assistance, accommodations and interventions needed for an individual with CVI to obtain meaningful access to educational or vocational materials.

Characterizing at-risk populations

CVI (i.e., visual impairment and/or dysfunction secondary to brain injury, damage, malformation, or malfunctioning of the brain's visual processing centers) is increasingly being recognized as a common sequalae across various clinical populations, including individuals with genetic disorders and conditions. In their article, Boatwright et al. presents data from a case series of three individuals with Down Syndrome (DS) and CVI with specific challenges in visual function as well as functional vision of the ventral stream, dorsal stream, and attention accompanied by subcortical changes and abnormal EEGs. While ocular conditions are well-documented in the DS population, CVI is only recently being considered for this population. As such, the CVI diagnosis may come later in life, as was the case for the three cases presented by Boatwright et al.. The authors note that receiving the CVI diagnosis for these families validated their concerns regarding their children's vision use. Earlier detection in such patients can indeed improve cognitive functioning required to optimally develop and interact with the world around them.

Supporting children with brain-based visual impairments

Jakubowski et al. invited family members and specialists who work with children with brain-based visual impairments to participate in an online survey. The survey was completed by educators, rehabilitation staff, clinicians, and family members. Results showed that although caregivers felt like undervalued members of the team, Professionals designated them as the principal and most credible informants regarding information about their child's visual status. There was also consensus that transdisciplinary clinics and inter-professional care were impactful and provided client centered care. Finally, respondents felt that CVI was underdiagnosed, and more formal training was needed to advance the field and provide adequate support to those with brain-based vision impairment.

Methods for evaluating functional vision

Currently, a large body of research involving CVI focuses on the use of surveys and structured history taking, such as the Flemish CVI Questionnaire (Ortibus et al., 2011), the CVI (or Insight) Inventory (Ghahghaei et al., 2021; Fazzi et al., 2007), and the Teach CVI Screening Tool (Teach CVI Partnership, 2017). There is tremendous value in these approaches as they can provide invaluable information regarding the daily challenges in the functional use of vision (Dutton et al., 2010). They can also provide insight into the aspects of visual functioning that are not affected for an individual with CVI. Research reported by Martin et al. applied Rasch analysis to the Teach-CVI Screening Tool in order to evaluate the relationship between person and item measures from the Likert responses. Their results suggest that the tool is well targeted (i.e., relative absence of floor and ceiling effects) and reflects a single underlying construct, namely "the impact of CVI on visual ability" (Martin et al.).

In addition to research involving questionnaires and surveys, there are also concerted efforts by many in the field of vision research, and CVI in particular, to more rigorously characterize visual ability in those with or at risk for CVI (Mooney et al., 2021, 2018, 2020; Kooiker et al., 2016; Hokken et al., 2024, 2025; Bennett et al., 2018a,b; Pamir et al., 2024; Ben Itzhak et al., 2021, 2025; Bauer et al., 2023; Walter et al., 2025). Most recently, approaches implement eye tracking-based methods, which do not rely on a verbal or manual response and, as such, may be more widely applicable to the spectrum of abilities in individuals with CVI. Among these is the manuscript in this Research Topic by Matsunaga et al., which outlines their proposed method for using AI-enabled image saliency analysis to evaluate whether children with or suspected of CVI gaze at similar image features.

Two aspects of CVI frequently studied are selective visual attention impairments and dorsal stream dysfunction. In the article by Hokken et al., they specifically tested global visual selective attention via Gestalt, Navon, and Kanizsa figures in school-aged children. These tasks require the ability to integrate visual elements in order to perceive a whole visual representation. Children with CVI demonstrated global impairments on all tasks compared to neurotypical children as well as children with ADHD or dyslexia. While the authors suggest their findings support the dorsal stream dysfunction hypothesis, the data also reflect underlying impairments in mid-level visual perceptual dysfunctions, such as contour integration and figure-ground segmentation. However, it has yet to be determined if mid-level and dorsal stream dysfunctions can be disentangled in CVI, particularly as dorsal stream dysfunction may manifest as simultanagnostic vision (inability to see more than a single object at one time) and/or restriction of visual attention to small portions of the visual scene. These difficulties are highlighted by three cases in the article by St Clair Tracy et al.. Based on the reported experiences, the authors generated a series of simulations for simultanagnostic vision, demonstrating how the functional field of view may be impacted by the presence of visual clutter. The cases also illustrate the potential emotional responses to complex environments as well as the potential benefit of mindfulness exercises. Additional research is required to fully appreciate how conscious vision strategies and mindfulness exercises impact the ability to expand the dynamic functional field of view and patient wellbeing.

Impact of visual impairments on quality of life and wellbeing

Visual impairment may have an impact on the level of satisfaction and participation in daily activities of teenagers. In the qualitative study of 25 teenagers with either visual or motor impairments, Veldhorst et al. conducted semi-structured interviews which revealed 13 themes, many of which were shared between those with visual and motor impairments. Notably, visual impairment here was based on visual acuity and/or visual field restriction and did not specifically extend to teenagers with cerebral visual impairment (CVI), and the cause of motor impairment is not specified. However, motor impairments were classified based on the GMFCS. Overall, the teenagers in both groups reported satisfaction with the level and amount of participation in leisure activities, with notable differences in barriers to participation. Increasingly visual dysfunctions are being reported and recognized in individuals with cerebral palsy and developmental coordination disorder, with visual impairment occurring over three times more frequently than in the general population (Lo Cascio, 1977; Fazzi et al., 2007, 2012; Duke et al., 2020; Chokron and Dutton, 2016). However, participants with co-occurring visual and motor impairments were excluded from this study. Thus, it remains unclear how barriers and facilitators related to activities and participation are affected in this population.

Improving outcomes—evidence from persons with lived experience

Understanding CVI based on the lived experience of individuals with CVI and their caregivers can be extremely impactful and provides invaluable insight. Bennett et al. emphasized that visual perception experiences and the development of concepts are different for each individual with CVI. They described how individuals with CVI use compensatory skills and strategies to perceive objects, access their environments, and socialize. Duesing et al. describe three case studies of individuals with early onset CVI, and interventions used to improve visual perception abilities and the understanding of the world around them. The authors describe experiences and strategies using various approaches, including self-taught compensatory strategies, sensory substitution, and augmentation techniques, to improve meaningful access to their environment. Importantly, because CVI manifests differently in each person with CVI, intervention approaches need to also be uniquely tailored to each patient and should be regularly evaluated for efficacy (Lueck et al., 2019, 2023).

Future directions—concluding thoughts

Although CVI research is advancing, the current body of evidence remains limited. There are many novel ideas and strategies still in their emerging phasea. In particular, studies related to early detection and early intervention are needed, facilitated by greater inter-disciplinary careRigorous intervention studies employing control groups are essential for advancing scientific knowledge and informing best practices in clinical care. Additionally, to strengthen the field, there is a pressing need for the development and validation of comprehensive outcome measures, both objective and subjective, across the spectrum of abilities and challenges, particularly those that assess not only clinical gains but also functional outcomes in daily activities and participation. As outlined by Jakubowski et al.—we still do not know enough.

Author contributions

CB: Writing – original draft, Writing – review & editing. JR: Writing – review & editing. KH: Writing – original draft, Writing – review & editing.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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EDITED BY
Karen Harpster,
Cincinnati Children's Hospital Medical Center,
United States

REVIEWED BY
Linda Lawrence,
Independent Researcher, Salina,
United States
Nicola McDowell,
Massey University, New Zealand

*CORRESPONDENCE
Carlijn Veldhorst
✉ carlijn.veldhorst@ru.nl

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Participation of teenagers with vision or motor impairments in leisure activities: a qualitative study

Carlijn Veldhorst[1]*, Marjolein Wijnen[2], Sabina Kef[3], Mathijs P. J. Vervloed[1] and Bert Steenbergen[1]

[1]Behavioural Science Institute, Radboud University, Nijmegen, Netherlands, [2]De Kleine Prins Onderwijsgroep, Utrecht, Netherlands, [3]Faculty of Social and Behavioural Sciences, University of Amsterdam, Amsterdam, Netherlands

Purpose: Participation in everyday life activities is important for the development of children and is an important topic in rehabilitation practices. This qualitative study aimed to unveil the perspectives and experiences of teenagers with vision impairments (VIs) or motor impairments (MIs) regarding their participation in leisure activities.
Materials and methods: 13 teenagers with VIs and 12 teenagers with MIs (age range: 11–15 years) participated in this study. Data were collected using semi-structured interviews. Verbatim transcripts were analyzed following the steps of the phenomenological approach.
Results: A total of 623 significant statements were identified and assigned to 221 meaning units. Finally, 13 themes emerged. Teenagers with VIs and MIs shared many experiences and perspectives regarding participation in leisure activities.
Conclusions: Teenagers reported that they can sufficiently indicate what they experience as pleasant and sufficient considering their participation, as well as the challenges they encountered, such as their impairment, limited transport possibilities, or concerns from parents. Overall, teenagers with VIs or MIs were generally satisfied with the degree and frequency of their participation in leisure activities and felt sufficiently involved.

KEYWORDS

participation, leisure activities, vision impairment, motor impairment, teenager, qualitative study, disability

Introduction

Participation in everyday life activities is vital for children's development, psychosocial wellbeing, and competence in physical skills (1, 2). Therefore, it is also an essential goal in rehabilitation services for children with disabilities (3–7). However, children with disabilities face challenges when it comes to participation (2). For instance, children with vision impairments (VIs) have lower participation rates compared to a population-based reference group (8), and children with cerebral palsy tend to participate less in leisure activities than children without motor impairments (MIs) (9). Also, Williams et al. (10) found a negative relation between the extent of impaired functioning and participation among children with intellectual disabilities. This paper seeks to unveil the perspectives and experiences of teenagers with VIs or MIs, without additional disabilities, regarding their participation in leisure activities using a phenomenological approach.

Participation, as defined by the World Health Organization (WHO), involves engaging in life situations that are age-dependent and aligned with an individual's developmental possibilities (11). Some researchers argue that this definition is not comprehensive enough (2, 12, 13). While the term "involvement" suggests a social dimension, participation operationalizations predominantly focus on attendance, that is, simply being present in activities with others. Imms et al. (12) proposed that participation is a combination of both attendance and involvement. While attendance involves the diversity of activities and the act of "being there," involvement concerns the experience and emotions associated with participation, such as feelings of engagement, social connection, belonging, and motivation. In addition, participation has reciprocal relationships with factors both internal to the individual (ability to express preferences, a sense of self, and competence in performing activities) and external factors (environment and context). In terms of Imms et al. (12), "Context is personal, considered from the perspective of the person participating, and relates to the people, place, activity, objects, and time in which participation is set. […] Environment is external, and refers to the broader, objective social and physical structures in which we live" (p. 20). Future participation is determined by the above-mentioned factors, which at the same time are influenced by past participation experiences.

The list of factors internal to the individual is not exhaustive and varies for each individual (12). Factors such as independence, mobility, self-concept, and peer relations are often considered crucial for participation (2, 14–16). However, people with impairments have to deal with restrictions in leisure activities, as they often depend upon others to participate in activities and need assistance to attend activities due to compromised mobility skills (17, 18). Although help from others and special technologies, such as braille apps for blind children (19) or exoskeletons for children with motor disabilities (20), can alleviate mobility challenges, they do not eliminate the individual's frustration with dependence. Relying on others for mobility only partly solves the participation problem by facilitating presence, but at the same time it can also enhance feelings of dependency, incompetence, and lowered self-esteem (15).

Rehabilitation services play a significant role in encouraging participation of children with disabilities by relying on professional expertise (21) and parental input (2). Still, solely relying on professional and parental perspectives is not enough because they may not necessarily align with the perspectives of the children themselves (21, 22). Children are capable of being involved in their own rehabilitation process (23), and their first-hand experiences can help them gain a deeper understanding of what participation means for them, both practically and emotionally (24, 25). This study addresses precisely this by using a phenomenological approach (26) focusing specifically on children's experiences without the influence of preconceived assumptions from researchers, parents, or rehabilitation professionals (27). Unlike other approaches, the phenomenological approach refrains from theoretical assumptions found in questionnaires (28) and avoids comparisons with control groups or established norms (24). As our research questions concern lived experiences of teenagers regarding participation in leisure activities, the phenomenological approach is pre-eminently appropriate because it can study *what* participants experience and *how* they have experienced it. In this way, authentic meaning can be given to the phenomenon "participation" (26).

There is already an abundance of recent studies describing barriers and facilitators to participation across different groups, including adults with vision impairments (14, 16), children on the autism spectrum (25), and heterogeneous groups with and without developmental disabilities (2, 29). Alongside the phenomenological approach, a unique element of this study is its focus on two specific groups—namely, participation experiences in leisure activities of teenagers with VIs or MIs, without additional disabilities. Different types of disabilities may have varying implications for participation (30). However, such heterogeneity can also exist within a single type of disability (31, 32). Cross-disability research can contribute to earlier and more tailored care for children with disabilities and their families (33), especially because effective interventions require a multidimensional approach. In this study, both groups are limited in their mobility and independence, which may impact their attendance and therefore participation. For children with MIs, these challenges with locomotion has predominant neurologic or motor causes (34), while for children with VIs, the difficulties are likely related to problems with wayfinding, orientation, and obstacle detection (35). The nature of the impairment is not the same in both groups. Yet, children with VIs or MIs sometimes exhibit slow and influential movements and therefore share some experiences with regard to mobility, although the origin of the motor problems might differ (36, 37). However, following the family of participation-related constructs (fPRC) framework by Imms et al. (12), participation is determined not only by physical competences but also by factors such as personal preferences and self-concept. These factors may overlap across disability groups.

Emphasizing the various aspects of participation (12) and investigating the meaning of participation for children with disabilities (3) can assist in prioritizing their needs in early intervention and rehabilitation practices (23, 26). Therefore, this paper aims to uncover teenagers' perspectives and experiences regarding participation in leisure activities. To gain insight into independence and mobility, the first research question is: (1) How do teenagers with VIs or MIs experience independence and mobility? To further understand participation, the following research questions are posed: (2) What leisure activities do teenagers with VIs or MIs participate in? and (3) How do teenagers with VIs or MIs experience their participation in leisure activities?

Material and methods

Participants

Participants, aged 11–15 years and fluent in Dutch or English, were recruited via convenience sampling. Group 1 included teenagers with vision impairments (VIs) from the two Dutch rehabilitation centers for people with VIs and were classified as having severe or moderate low vision based on criteria of the

TABLE 1 Demographics of the participants ($N = 25$).

Participant[a]	Age (year; months)	Sex	Degree of vision impairment[b]	Degree of motor impairment[c]
V1	14; 5	Boy	Moderate	—
V2	13; 9	Boy	Moderate	—
V3	13; 2	Girl	Moderate	—
V4	13; 4	Boy	Moderate	—
V5	15; 4	Girl	Severe	—
V6	15; 0	Girl	Moderate	—
V7	14; 11	Girl	Moderate	—
V8	13; 7	Boy	Severe	—
V9	14; 8	Boy	Moderate	—
V10	13; 4	Boy	Moderate	—
V11	13; 5	Girl	Moderate	—
V12	13; 7	Boy	Moderate	—
V13	13; 6	Boy	Moderate	—
M1	12; 11	Girl	—	5
M2	11; 6	Boy	—	1
M3	11; 9	Girl	—	2
M4	11; 8	Boy	—	4
M5	12; 7	Girl	—	1
M6	12; 4	Girl	—	2
M7	12; 3	Boy	—	2
M8	14; 9	Girl	—	1
M9	14; 9	Boy	—	2
M10	13; 4	Girl	—	2
M11	13; 1	Boy	—	2
M12	14; 3	Girl	—	5

[a] V refers to group 1 (vision impairment); M refers to group 2 (motor impairment).

[b] Moderate = visual acuity between 5/100 and 30/100 or a field of vision between 10° and 30°. Severe = visual acuity is less than 5/100 or a field of vision less than 10°.

[c] For consistency, the degree of motor impairment is based on the Gross Motor Function Classification System (39), varying from limited coordination (1) to manual wheelchair in all settings (5).

World Health Organization (38). Moderate low vision was considered a visual acuity between 5/100 and 30/100 or a field of vision between 10° and 30°. Teenagers were categorized as having severe low vision when their visual acuity was less than 5/100 or their field of vision was less than 10°. Teenagers who met the inclusion criteria received information about the study, including a flyer and a link to a video clip with audio details. Group 2 consisted of teenagers with a medical diagnosis of a MI and was classified based on the Gross Motor Function Classification System (39). They were recruited from a special education school for children with MI in the Netherlands. Teenagers who met the inclusion criteria received information, including a flyer and a short presentation about the study in class. In case of interest, additional information was offered. Both teenagers and parents were required to provide consent for participation. In both groups, the presence of evident multiple disabilities, which is more than one impairment, each independently leading to disabilities, was an exclusion criterion.

Participants' demographics are displayed in Table 1. The sample included 25 teenagers, divided into group 1 with VIs ($n = 13$) and group 2 with MIs ($n = 12$). Group 1 consisted of five girls and eight boys, with a mean age of $m = 14.08$ ($sd = 0.76$, range 13–15). Group 2 consisted of seven girls and five boys, with a mean age of $m = 13.00$ ($sd = 1.15$, range 11–14). The specific type of disability was highly varied across both cohorts, such as oculocutaneous albinism and retinopathy of prematurity in group 1 and cerebral palsy and developmental coordination disorder in group 2.

Procedure

Participants were interviewed individually by the first author, second author, or student researchers in face-to-face or online video meetings[1]. All participants provided consent for audio recordings of the interviews. Parents provided demographic information about the participants. The interviews started by asking what the participant did last weekend and on a "typical" day in a week. Follow-up questions were asked to gain a deeper understanding of the children's experiences. Examples of questions are as follows: "What do you like about [the activity]?" and "What does being independent mean to you?." As often as possible, the participants were asked to give examples of what they told to concretize the participants' answers without prompting them for specific answers. This was especially important to enhance the validity of the participants' responses (40). Interviews

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[1] Due to COVID-19 restrictions, in some cases, we were restricted to plan online video meetings to perform the interview instead of a face-to-face meeting.

lasted from 13 to 45 min ($m = 21$ min). In total, 25 interviews were conducted. The study was performed in accordance with the ethical standards outlined in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. Approval was granted by the Medical Ethical Committee of Eastern Netherlands, Nijmegen, the Netherlands (No. NL74630.091.2 0) for group 1 and the Ethics Committee Social Sciences Radboud University, Nijmegen, the Netherlands (No. ECSW-2021-140) for group 2.

Data analysis

Interviews were transcribed verbatim, pseudonymized, and then analyzed using the steps of the phenomenological approach in ATLAS.ti (version 23). This method facilitates an authentic understanding and interpretation of the phenomenon of "participation" as experienced by the target group itself (26) and ensures a systematic and in-depth analysis of the data (41). The first step in the phenomenological approach included "bracketing," meaning excluding the researcher's own feelings and perceptions, to enhance data validity (26). In addition, all authors' backgrounds and contributions were stated in reports (42). The first author was trained as an elementary school teacher and educationalist, with practical experience in teaching children from both regular primary and secondary schools. The second author was also trained as an elementary school teacher, specializing in pedagogical sciences, and currently works at a school for children with motor impairments. The third author holds a PhD in pedagogical sciences. Her research addresses the social-emotional development of adolescents and young adults with vision impairments. The fourth author also holds a PhD in pedagogical sciences. His research addresses the development of young children with vision impairments or multiple disabilities. The fifth author, trained in human movement sciences, holds a PhD in social sciences. One of his research projects focuses on sports and movement of children with motor impairments. In step 2, the first and second authors identified important statements from all participants pertaining to research questions, resulting in 623 significant statements. This was done in a research question-driven way, meaning that we specifically looked for statements related to the research questions. In step 3, the first and second authors assigned general meanings to the significant statements, forming "meaning units" that remained faithful to the original meaning conveyed by the participant. Significant statements with the same tenor were assigned to one meaning unit. The first and second authors created meaning units separately, yielding 221 meaning units after reaching a consensus through discussion. In the fourth step, related meaning units were integrated into themes to develop an in-depth description of the phenomenon. Again, the first and second authors performed this step individually, followed by discussion and consensus. Initially, 17 themes were formed. Subsequently, the third and fourth authors participated in steps 3 and 4, checking the meaning units and verifying the corresponding themes. Based on their judgment, some meaning units were rearranged within the themes, resulting in the final 13 themes. The full methodology is available at the Open Science Framework (https://osf.io/rmuwz/?view_only=ead81624f34045779ba12837e1670ba9).

Results

In total, 13 themes emerged from the interview, as displayed in Figure 1. To illustrate the process of the phenomenological approach of forming meaning units from statements and subsequently categorizing meaning units in themes, Table 2 presents examples of significant statements and their meaning units that form a theme. The answers of the teenagers to the question regarding types of leisure activities were not used as significant statements for forming meaning units but clustered in Table 3 to give an overview of the leisure activities.

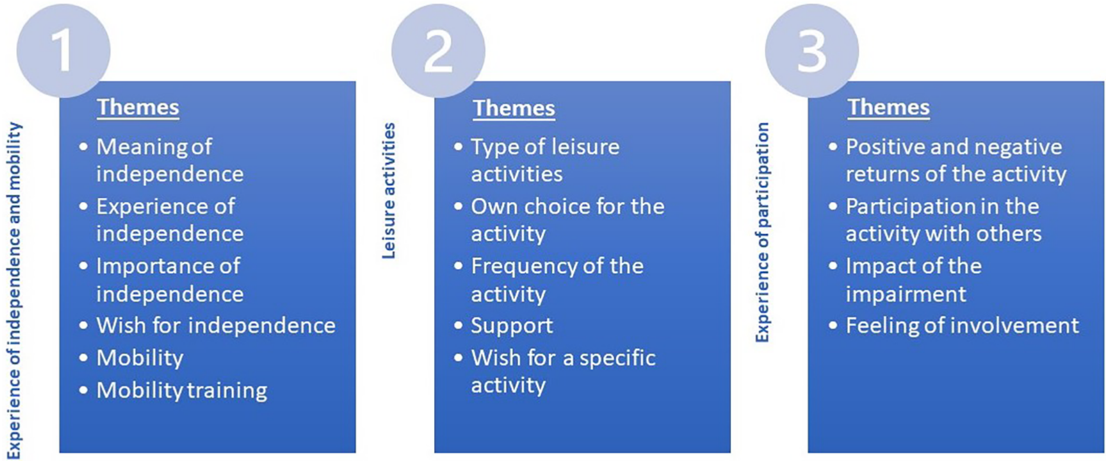

FIGURE 1
Themes related to the research questions.

TABLE 2 Examples of significant statements and their formulated meaning units that together form a theme[a].

Significant statement	Meaning unit	Theme
"*Is it also important for you to be independent?* I don't know. Maybe yes, maybe no."	Not sure about the wish to be more independent	Importance of independence
"Because I'm usually alone and then, uhm, I have to, so if someone arrives or something, I have to be able to stand up for myself and things like that."	Because you are often alone, it is important to be able to stand up for yourself	
"Because, for the future it [independence] is very important."	Independence is becoming more important with age	
"Uhm, yes eventually I also have to be able to do it independently. So then eventually I have to learn it a little bit."	Independence is becoming more important with age	
"But I do think it's important to, if you learn that too, because later on I also have to do things independently and then it's already useful to learn it a bit myself."	Independence is becoming more important with age	
"It is important because I am also getting older. And then I have to be able to do things myself and not have my mother cutting my nails when I'm thirty, I have to learn that myself too."	Independence is becoming more important with age	
"…Pretty important, because you are in grade 6 and then you have to be pretty independent, in my opinion."	Independence is becoming more important with age	
"*Does that come with age do you think?* Yes. I do think that comes roughly with age. Yes. *When you are a bit older, that you become a bit more independent.* Yes."	Independence is becoming more important with age	
"*How important is it for you to do things outside independently?* Important"	Independence is important	
"Just out alone? *Yes, how important do you think that is?* Somewhat important"	Independence is important	
"I think that's [independence] pretty important."	Independence is important	
"*How important is it for you to be independent? To do things outside the home?* Not really very important so far. *Yes, okay, and why is it not very important so far?* Because I'm not out of the house very often	Independence is less important if you do not come outside that often	
"Uhm, yes, I don't like help. Preferably, yes preferably I just want to do everything myself. And arrange things myself, so yes, and I get bored really quickly. I'm really someone who, preferably I'd be working until 11 o'clock right now, so to speak. I just like doing things. Not to be bored."	Independence is important to do things by yourself	
"Ehm, yes because, it is easier if you can do everything yourself rather than always needing help."	Independence is important to do things by yourself	
"It's also important to do things yourself, so yes, I do think it's important."	Independence is important to do things by yourself	
"Um yes, that's um, I'm glad for example that I don't have much trouble with my sight that I can do all that independently. But for the rest yes, I don't really think about it either. *No, and then why is that so important to you?* Um, yes because, it is easier if you can do everything yourself than always needing help."	Independence is important to do things by yourself	
"Pretty important, because I don't always find it necessary for my parents to know everywhere I want and would go."	Independence is important because parents do not need to know everything	
"It's important though. Because you can't ask someone all the time of you, do this, do that."	Independence is important because people cannot always help you	
"Pretty important. Because it's not always possible to bring someone with you, you have to be able to do it yourself once."	Independence is important because people cannot always help you	
"Quite important because people can't always pay attention to you"	Independence is important because people cannot always help you	
"When I go to college, I hope I'll just go home by train, But I already know how to travel by train, so That's not so exciting anymore either."	During student life you need to travel by train, but I already know	

[a]Meaning units and themes were based on significant statements of both teenagers with VIs and teenagers with MIs.

Research question 1: independence and mobility

The first research question explored how teenagers with VIs or MIs perceived independence and mobility. Based on the interviews, we identified six different themes that answered the first research question: meaning of independence, experience of independence, importance of independence, wish for independence, mobility, and mobility training (see also Figure 1).

Theme 1: meaning of independence

Without exception, every teenager described independence as being able to function without help from others and doing things on their own. For instance,

> "Just being able to do things by yourself, and eeh, arranging things by yourself, and doing things by yourself, you know." (V9).

According to the teenagers, independence was clearly related to autonomy, including

> "Well, that you can live your own life well and yes, be able to do everything yourself. That you don't need the help of others, yes." (V7).

> "That for a while, you don't have people telling you what to do." (M1).

TABLE 3 Participation in leisure activities.

	Teenagers with vision impairments	Teenagers with motor impairments
Home-based		
Various individual activities	Drawing, coloring, puzzles (1)	Drawing (2)[a]
	Knitting (1)	Playing or walking with dogs (2)
		Going to animals around the house (1)
		Having a walk (1)
		Taking a bath (1)
		Laze around (1)
Schoolwork	Making homework (10)	Making homework (2)
Digital entertainment	Watching TV (2)	Watching TV (3)
	Watching YouTube (1)	Watching YouTube (1)
	Using the smartphone (1)	Using the smartphone (2)
	Watching a movie (1)	Using the tablet (1)
Gaming	Gaming (with peers) (5)	Gaming (with peers) (1)
	Gaming (company not mentioned) (2)	Gaming (company not mentioned) (1)
		Gaming (sometimes alone, sometimes with peers) (1)
		Gaming (alone) (2)
Home-based activity with the family	Playing games with siblings (1)	Playing games with the parents (4)
	Watching a movie with the household family (1)	Drinking tea with mum (1)
	Helping dad with the pavement around the house (1)	
Outdoor		
Job	Working in the garden (summer) and warehouse (winter) (1)	Doing jobs/chores for money (1)
	Volunteering (1)	Delivering newspapers (1)
	Working at McDonalds; assisting with gymnastics (1)	Photographing (1)
	Assisting with the open day at school (1)	
Musical activity	Piano lessons; singing lessons (1)	Keyboard lessons (1)
Sport—team	Soccer (3)	Soccer (1)
	Korfball (1)	Dancing class (2)
Sport—individual	Swimming (1)	Swimming (2)
	Fitness (1)	Fitness (1)
	Horse riding (2) Judo (2) Karate (1)	Horse riding (2)
Outdoor activity with the family	Going to a brush court (1)	Celebrating the birthday of the mother in a restaurant (1)
	Going to the beach (1)	Went to a market (1)
	Going to the church (2)	Going to a puppy course (1)
	Visiting the grandparents (3)	Watching the swimming class of the siblings while having a drink in the café (1)
	Staying at an aunt (1)	Shopping new clothes (1)
	Going to a cousin's birthday (1)	Watching trains (1)
	Evening walk or forest walk with the household family (1)	Visiting a family member in a clinic and going to the beach (1)
		Going to a grandparents' birthday (1)
		A walk with the dog and sibling (1)
Cultural activity with peers	Fishing with friends (1)	Girls club (1)
	Scouting (2)	Scouting (1)
	Youth association of the church (1)	
Meeting with friends	Meeting with friends (8)	Meeting with friends (1)
	Shopping with friends (2)	Meeting with a friend outside (1)
		Playing outdoors (1)
		Meeting with one friend (1)
No activity mentioned	Doing nothing because of the cancelation of soccer due to COVID-19 (1)	Doing nothing (1)

[a] The number of participants who mentioned the specific activity is displayed between brackets ().

> "Uhm freedom, so just doing what you want and can and not being dependent on everything." (V6).

Independence was also linked to age, specifically in terms of acting age-appropriately and fostering autonomy from parental influence.

> "That I can do things for myself and things that I should actually be able to do myself, that someone my age should be able to do." (M9).

> "Because you can't always expect someone else [mum] to bring you or go with you." (V6).

Theme 2: experience of independence

Teenagers mentioned that they felt both independent and dependent during their leisure activities.

> "When I go to school, I feel independent. Because I know where I'm going. And sometimes when I go to a place I don't know, I don't feel independent." (V5).

> "Yes, I can just do many things on my own, no need for my parents to help me with that." "So, you don't need help with anything?" "No, well, not nowhere with anything." (V9).

The feeling of independence was sometimes hampered by an overprotective social environment, as participants M11 and M12 mentioned. In addition, a substantial number of teenagers received help. The complexity arises as participants indicated mixed feelings about consistently receiving help. While they expressed a desire to avoid constant assistance, they also valued the presence of peers, creating a nuanced perspective. This is where complexities lie because participants mentioned they do not always want help. Yet, they do not want to do everything independently because the presence of peers is also convivial, as V4 described.

> "Mum is far too worried. She is worried that I have to go to the toilet, and no one is there." (M12).

> "I quite like it when people are involved but I don't like it when everyone asks with everything if I need help." (M11).

> "Uhm, doing things outside the door that is important to me, I also find that convivial. But being independent, it's also convivial with friends around you." (V4).

Theme 3: importance of independence

Many teenagers expressed that independence is important to them. Teenagers believed that attaining independence is essential for their transition into adulthood, as they recognized the need to rely less on continuous support from their family. For example, they stated

> "I think that's important for me because I have to learn it myself at some point, because there can't always be someone with me." (M6).

> "… But I do think it's important to learn too, because later on I also have to do things independently and then it's already useful to learn a bit now." (V2).

> "Suppose someone arrives or something, I have to be able to just stand up for myself and things like that." (M10).

The participants' statements revealed their aversion to the inevitability of dependence on others. While some teenagers believed independence is an innate quality, others opined that it must be taught by the environment.

> "I do think someone will have to teach me then." (M6).

Whereas all teenagers with VIs felt the urge to be independent, this was not the case for all teenagers with MIs. Some MI teenagers did not feel the urge to become more independent in the near future because they did not go outside without adult supervision. For participants M9 and M12, help was always available.

> "Because I am never out on my own." (M9).

> "I never leave home without my mum and dad." (M12)

Theme 4: wish for independence

The statements of a small group of teenagers revealed that in addition to the importance of independence, they had an explicit wish to be more independent, for example,

> "…Because I would like to go by myself once without people." (M1).

> "Uhm, yes, I don't like help. Preferably, yes preferably I just want to do everything myself. And arrange things myself." (V6).

Theme 5: mobility

For participants, the theme of mobility intersected with the theme of independence, as being independently mobile had a major impact on teenagers' feelings of independence. Teenagers described mobility as the ability to move from one location to another; for example, the participants expressed it as

> "Uhm, that I can get to where I need to go at a fine pace that suits me. That you can go to it yourself." (V4).

> Yes, yes that is nice when you are just well mobile that you just, yes, you say just I am going there and then I don't need anything else at all." (V1).

The attainment of independent cycling was emphasized as an important goal. In certain instances, teenagers could only achieve this with the assistance of their parents, like V8. Many teenagers with VIs explained that a lack of overview of the traffic resulted in less mobility, like V7 said.

> "If my friend lives here in the village I cycle there with one of my parents." (V8).

> "Then, if something happens, then I, then eeh yes, then maybe I don't react to it so well. Or then I don't see it so

> well. So then, yes, then sometimes things just don't go so well." (V7).

The dependency on adults for mobility was highlighted in many interviews. This was particularly evident for teenagers with MIs, as their friends often lived further away due to attending schools for special education outside their hometowns. Consequently, these teenagers needed their parents for transportation to engage in leisure time activities.

> "But my father or mother will then take me to the riding school or a meet up with friends." (M11).

Teenagers who moved around independently sometimes used aids that supported independent mobility. For example, teenagers who used wheelchairs benefitted from electric wheel support, which requires less power and therefore could cover greater distances. Teenagers with VIs could make use of a cane. In addition, several teenagers mentioned that they used a navigation app to plan their routes.

Theme 6: mobility training

To enhance mobility, teenagers could follow mobility training initiated by rehabilitation services. It was notable that only teenagers with VIs had specific mobility training during leisure time.

> "Uh yeah, I had a few months ago I had mobility training here at [name rehabilitation center], which is the rehabilitation center here, that big one. And it was like to, to learn how to use my cane, for visually impaired and blind people to walk to school." (V5).

Research question 2: types of leisure activities

The second research question concerned the types of leisure activities attended by teenagers with VIs or MIs. Table 3 provides the leisure activities mentioned by teenagers with VIs and MIs separately. Some participants mentioned more than one activity. Teenagers from both groups reported participating in home-based activities, often involving family members, such as playing board games. Other frequently mentioned home-based activities included those centered around digital entertainment, such as watching television or using smartphones. The most popular activity was gaming, for both groups. In addition to home-based activities, outdoor activities were also listed. Teenagers participated in sports activities, both individual and team sports, scouting, or gatherings with friends. Overall, teenagers with VIs participated slightly more in outdoor activities, such as sports activities, part-time jobs, and gatherings with friends, than their peers with MIs. In addition to the type of activities, we found four themes related to the types of leisure activities (see Figure 1).

Theme 7: Own choice for the activity

Teenagers were asked whether they autonomously selected the leisure activity. The responses revealed that many activities, particularly those at sports clubs, were initiated at an early age, with parents taking the lead. As teenagers grew older, they made independent choices to persist in these activities. When teenagers started with activities later in life, such as having a part-time job, they more frequently expressed having chosen the activity themselves. At times, a rehabilitation organization assisted in selecting an activity, like M8 reported.

> "Yes, she investigated with me what I could do and what my hobbies were, and they then referred me to that place." (M8).

The social aspect was one of the reasons given by teenagers for choosing a particular leisure activity, as interacting with peers and the sociability that comes with the activity held importance for them. Teenagers experienced positive feelings during these leisure activities, such as happiness and fun. In addition to the social aspect, teenagers named relaxation and interest in the activity as essential pillars for their participation. V5 described playing music with the piano as relaxing.

> "Playing my piano is quite relaxing, for instance when I come home from school." (V5).

Several teenagers highlighted enjoyment as a significant factor influencing their activity choices, often associating it with elements like competition or skill enhancement. In addition, they considered their impairment when selecting activities, opting for those that could be performed despite the limitations of an impairment, similar to the experience described by M11.

> "Somehow, I've always had a thing for horse riding, back in the day from childhood. But I only started riding later because my body was a bit worse then anyway." (M11).

Theme 8: frequency of the activity

Participants were asked what they thought about the frequency of the leisure activities. The majority of teenagers believed that their engagement in leisure activities was adequate. Only a few expressed a desire for more frequent participation. Notably, distinctions emerged between teenagers with VI and those with MI. Most teenagers with VI expressed a desire for an increased frequency, citing enjoyment or a preference for leisure over obligations. On the other hand, some teenagers with MI expressed a similar desire for more frequent participation, but the constraints of fatigue resulting from their MI made it unfeasible.

> "I would like to do that more often, but I don't think my body can handle it." (M11).

Theme 9: wish for a specific activity

When being asked what they would like to do when anything would be possible, teenagers referred to a broad range of activities, including low-key and more challenging activities.

> "I would love to go to one of those big water parks and cuddle puppies all day." (M3).

> "One time shopping with my grandmother and my mother together." (V3).

> "Driving a tractor, I would love that." (V11).

> "And eeh, I would like to go skydiving with my friends, my blind friends let's say. Because they think that's really cool. Because they are very curious what kind of feeling that gives because they have no idea what that is. So that seems like a really cool thing to do." (V6).

Talking about their desired activities, both teenagers with VIs and MIs did not mention their impairment. Just a minority was dreaming about activities that they thought were not possible because of their impairment.

> "Soccer. Or basketball, just sports." (M4).

> "What do you want to do if anything would be possible? Walk." (M1).

> "Just doing sports with other kids together, that I can do that too." (V7).

Research question 3: participation experiences in leisure activities

The third research question entailed how teenagers with VIs or MIs experienced their participation in leisure activities. Four themes were distinguished to answer this research question: Positive and negative returns of the activity, participation in the activity with others, impact of the impairment, and feeling of involvement (see also Figure 1).

Theme 10: positive and negative returns of the activity

Almost every teenager indicated that the leisure activity was fun. Other positive expressions were calmness and sociability. The presence of peers could enhance the enjoyment of the activity, M8 reported. The presence of peers is not only related to group activities such as a team sport but also when meeting with friends or during gaming. In the latter, teenagers said they interacted with peers online.

> "Having fun with friends and people you know." (M8).

A sense of autonomy was commonly linked to the leisure activities of the participants.

> "Being free and just doing your own thing." (V2).

> "That no one tells me what to do and also something I can do all by myself." (M1).

Leisure activities were primarily considered as fun. Yet a few teenagers mentioned the downsides associated with the activities. These unfavorable aspects were often attributed to the impairment, such as muscle strain or fatigue.

Theme 11: participation in the activity with others

Only some teenagers with MIs mentioned that they preferred to perform activities alone. However, the majority of the teenagers favored participating in leisure activities with others since it is fun and sociable to partake in them together.

> "I do like it in a group." (V1).

Theme 12: role of the impairment

The role of the impairment in leisure activities recurred in several ways, as already mentioned in theme 10. Particularly, teenagers with MIs mentioned the challenges associated with their impairment, for example, the need for help in daily life and personal care.

> "After school I have to be changed on Sundays I am bathed." (M1).

For teenagers with MIs, fatigue and muscle strain due to overexertion played a part. These teenagers mentioned this in the following statements.

> "I would like to do that more often, but I don't think my body can handle it." (M11).

> "I thought that was enough because I was sore for days afterwards from." (M7).

While most teenagers had predetermined activities during leisure time, some deliberately avoided making plans after school or during weekends. They attributed this decision to fatigue, which they linked to their impairment. Considering school as a draining experience, they felt the necessity to rest, opting to either lie in bed or stay at home, engaging in low-intensity activities, although these activities were not necessarily preferred.

> "Well, the weekend is actually to rest, bit of using the tablet lying in my bed, that I don't have to do much more then. […] My hobby is football, but there I can … I actually can't with my muscles. We tried that but that's not an option." (M4).

Another drawback of the impairment mentioned by only some teenagers with MIs is that they did not participate in leisure activities after school due to a lack of time. This is because they were taken home by a taxi at the end of the school day. As the taxi transported multiple students, going home took much longer than an individual ride home. M5 explained what he does after school.

> "Usually not much, because I get home around four, half past five and then there is not actually much to do." (M5).

Also, teenagers with VIs commented on the setback of their impairment. For example, V8 said he could not always participate in a soccer match.

> "But, at the match, there is then, say, a check on what the score is for how much I can, say, play. Okay, because you have impaired sight? Yes. Okay, so it becomes … You don't always get to play? Well on Tuesdays and Thursdays, yes, but not on Saturdays." (V8).

Nevertheless, it appears that the challenges encountered by teenagers with VIs during leisure activities were not primarily linked to the activities *per se* but rather to the transportation to and from the activities.

> "No, it's just, then we do have to take time into account, because then I have to cycle a bit slower in a larger group.. Uhm, yes sometimes it's a pity, but yes, with my vision impairment it's not really possible sometimes either." (V4).

Theme 13: feeling of involvement

Involvement reflects the extent to which teenagers feel themselves involved in the activities they engage in, as well as their connection to their social environment and their satisfaction with their involvement. All participants indicated that they felt involved in their leisure activities, mentioning several reasons for their involvement. An important factor was the presence of peers, as M11 and V9 described. In addition, V3 indicated that he felt more involved because others showed active involvement and fanaticism.

> "Me and my little brother we always do it together and we also share the money, so I do feel involved in it." (M11).

> "Yes, just because I like it, I have a nice team." (V9).

> "Everyone is involved always in games and so on, including me." (V3).

In addition to the presence of peers, multiple factors contributed to the level of involvement. For example, activities that were specifically focused on the teenager, those requiring a high level of concentration, or those fitting well to their abilities despite their impairment resulted in experiencing a high level of involvement. In addition, teenagers who independently chose their activities experienced a high level of autonomy and fun and therefore felt involved.

> "There are always different activities there and some things I cannot see like reading a map and then there are always other activities I can do then." (V4).

> "No, not at all [hard to do the activity], because she even got enlarged cards." (V6).

> "Yes, that's really a lot of fun. Yes, that um, it's fun, very often I get to choose what we're going to do myself too." (V6).

When the teenagers were restricted in their participation in activities due to their impairment, involvement dropped sharply, V9 explained

> "Uhm, because at uh, on Saturday, for example, I can't always play in the match itself. They check what the score is to see how much I can play." (V9).

Discussion

The current study explored the viewpoints and lived experiences of 25 teenagers with either VIs or MIs concerning their participation in leisure activities, independence, and mobility. Previous research has revealed disparities in perspectives between children and their parents (43) and rehabilitation professionals (44), with children exhibiting a more optimistic outlook than parents and professionals. In addition to insights from professionals, parents (45, 46), and researchers (12, 47) in prior studies, this study focused on teenagers' perspectives. The study employed a phenomenological approach (26) to explore teenagers' shared experiences in leisure activities, avoiding preconceived assumptions (27). The cross-disability approach acknowledged the heterogeneity within the groups (32, 48) and provided insight into the extent and impact of living with a disability; most importantly, it highlighted teens' experiences regarding participation, mobility, and independence. This approach aligns with the rationale of the individual differences approach (49) and enhances rehabilitation practices by directly considering the perspectives of the target population, thus ensuring better alignment with their needs (15). In addition, with this empirical information, conceptual models such as the fPRC framework (12) can be complemented, giving meaning to factors like "preferences" and "the environment."

The first research question focused on elucidating the teenagers' definition of independence and mobility. Without exception, independence was defined as the ability to perform tasks without needing immediate assistance from others. It became evident that, for these teenagers, mobility was intricately linked to independence. The expression of personal preferences aligns with one of the intrinsic factors associated with participation in the fPRC framework (12). That independence is a crucial theme for teenagers with disabilities is also evident from the multitude of themes that have emerged, encompassing various aspects of independence: the meaning of independence, the experience of independence, the importance of independence, and the wish for independence.

The second research question explored the types of leisure activities in which teenagers with VIs or MIs engaged in. Various activities were mentioned, with gaming as the most popular one. As Stone et al. (50) concluded for youngsters on the autism spectrum, the popularity of gaming among teenagers may stem from the opportunity to interact with peers conveniently from home, which fosters social participation (51). The convenience for the target group in this article, children with VIs or MIs, might also related to the fact that games can be played from home, overcoming mobility problems.

The findings of the third research question, how do teenagers with VIs or MIs experience their participation in leisure activities, indicate that both groups generally perceive their participation in leisure activities positively, consistent with the review of Powrie et al. (52). Participants emphasized aspects such as having fun and interaction with peers, which align with the intrinsic fPRC factor of sense of self (12) and mirror results from a study involving adults with VIs, who mentioned fun as the most important motive to participate in runs (53). In line with previous research (54, 55), some participants experienced limited participation due to a direct consequence of the impairment, such as functional limitation, pain, or fatigue. In this respect, these limitations negatively affected the fPRC factor of activity competences (12). Teenagers also cited indirect effects of their impairments, such as limited transportation options or parental concerns, as the reasons why they were hindered in leisure time activities (54), related to the fPRC factor of environment (12).

Although this study primarily focused on many similarities, we noticed some differences in the experiences of the two groups. Specifically, whereas all teenagers with VIs expressed a desire for independence, this sentiment was not uniformly shared by teenagers with MIs. Moreover, diverse barriers to participation in leisure activities were identified. Teenagers with MI highlighted obstacles such as pain, fatigue, muscle strain, and time constraints. In contrast, transportation emerged as the primary concern for teenagers with VIs, which aligns with the findings of Wright et al. (44) and Jaarsma et al. (56). Nevertheless, experiences and perspectives were mostly similar instead of different. On the basis of this study, the nature of the disability seems to matter little for participation in leisure activities.

Implications

The current study shed light on teenagers' perspectives regarding participation as a pivotal component in rehabilitation (3, 5–7). To engage teenagers with VIs or MIs, emphasis on the social dimension is necessary. The identified values commonly associated with leisure activities, such as "fun" and "sociability," highlight the importance of tailoring activities that align with these intrinsic motivations. The current study acknowledges the reciprocal relationship between intrinsic factors and participation (12). Investing in activities that match the specific preferences of these teenagers can make them more appealing overall. Participating in "fun" activities can be used to develop skills and competencies. For example, games can improve motor skill development (57).

Teenagers value autonomy and the ability to express preferences, linking the feeling of independence to decision-making. Notably, self-chosen activities were consistently associated with a sense of enjoyment. This observation aligns with insights from self-determination theory (58), describing that human motivation is driven by the fulfillment of psychological needs including autonomy, competence, and relatedness. The interviews showed that excessive assistance can undermine feelings of autonomy. Their understanding of independence involved the ability to perform tasks unaided. The ability to make personal choices and the experience of autonomy emerged as crucial pillars for fostering participation, as noted by Imms et al. (12) and Saebu et al. (59).

Previous research on adolescents who are deaf or hard of hearing suggests that a combination of online and offline interaction can strengthen existing friendships (60). In the current study, a substantial proportion of the participants identified gaming as their favorite leisure activity, valuing (online) interaction with their friends. Incorporating online games into rehabilitation practices can prove to be a valuable complement, particularly when individuals not only engage independently but also connect with friends online during games, in addition to real-life interactions. The utilization of online games can promote interaction with peers (50) and social participation (51).

This study demonstrates knowledge about what teenagers want and consider necessary regarding independence and participation. It is, therefore, highly essential for stakeholders, including educators, rehabilitation professionals, and researchers to engage in ongoing dialog with teenagers, as also emphasized in the review of Paul et al. (15) on inclusive education. The themes identified in this research provide valuable insights for initiating conversations with teenagers. These dialogs may encompass pragmatic considerations, such as transportation logistics and perceptual dimensions of teenagers, clarifying their conceptualizations and visions.

Limitations

Using convenience samples of children with impairments, as in the present study, carries the risk of sample incomparability. In this study, teenagers with VIs attended regular education alongside peers without impairments, while teenagers with MIs were enrolled in special education, where all students have impairments. This educational setting difference may affect participation levels, as larger and/or regular schools tend to increase participation in activities (61–63). Moreover, positive peer attitudes toward impairments are more common in inclusive education settings (64). Consequently, teenagers with VIs might experience these positive peer attitudes more frequently than teenagers with MIs because they are taught within regular education settings. It is further known from the literature that peer attitudes influence the leisure activities of teenagers with disabilities (65, 66). If teenagers are engaged and encouraged by peers to participate, they might show more and more frequent participation (17, 65). However, current research did not consider the impact of education settings on participation.

The study explicitly involved teenagers with a single disability, either VIs or MIs, deliberately excluding teenagers with both VIs and MIs. This exclusion was intentional, as having multiple disabilities does not simply mean that addressing each disability will resolve all issues (67). In fact, the combination of multiple disabilities often leads to more complex and severe challenges. Children with multiple disabilities generally require more extensive support than those with a single disability, who tend to function more independently (68), and may face fewer limitations in daily activities (69). Thus, the current findings are relevant to children with either VIs or MIs, emphasizing their experiences and potential for participation. The study highlights the value of a cross-disability perspective (70) and promotes the development of more effective, individualized interventions (33). However, the study did not address teenagers with both VIs and MIs. While the current findings of the study provide a foundation for further research, they are not comprehensive enough to fully address the needs of teenagers with multiple disabilities. Given the complexities of multiple disabilities, specific research targeting this population is warranted (67–69) to provide more tailored implications for their care and support.

Furthermore, even with using focused research questions to guide the study and to prevent the focus of the study from becoming too broad (26), this procedure may have the consequence that other information participants shared might have been left out, although it could be important to understand participation in leisure activities (41).

Finally, the phenomenological approach assumes that participants have experienced the phenomenon under investigation (26). While heterogeneity exists among the participant groups, this study and previous research indicated that shared experiences outweighed differences, suggesting common perceptions regardless of impairment cause or severity (48, 71).

Conclusion

The findings of this research described the meaning of independence, mobility, and participation in leisure activities for teenagers with VIs or MIs without additional disabilities. Teenagers can sufficiently indicate what they experience as pleasant and sufficient considering their participation and the challenges they encounter. For example, teenagers with VIs or MIs were generally satisfied with the degree and frequency of their participation in leisure activities and felt sufficiently involved. However, teenagers with impairments were also good at naming factors that hinder participation, such as their impairment, limited transport possibilities, or concerns from parents. The study showed how teenagers experienced participation, which complements the perspective of parents, professionals, and researchers. With this, this research adds a new perspective to the concept of participation, namely, of teenagers with impairments.

Data availability statement

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found here: https://osf.io/rmuwz/?view_only=ead81624f34045779ba12837e1670ba9.

Ethics statement

The studies involving humans were approved by approval was granted by the Medical Ethical Committee of Eastern Netherlands, Nijmegen, the Netherlands (No. NL74630.091.2 0) for group 1 and the Ethics Committee Social Sciences Radboud University, Nijmegen, the Netherlands (No. ECSW-2021-140) for group 2. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

CV: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Resources, Validation, Visualization, Writing – original draft, Writing – review & editing. MW: Data curation, Formal Analysis, Investigation, Writing – original draft. SK: Conceptualization, Formal Analysis, Funding acquisition, Resources, Supervision, Validation, Writing – review & editing. MV: Conceptualization, Funding acquisition, Supervision, Writing – review & editing. BS: Conceptualization, Funding acquisition, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Appendix A

The first author was trained as an elementary school teacher and educationalist. She has practical experience in teaching children from both regular primary and secondary schools and now works as a PhD student. The second author was also trained as an elementary school teacher, specializing in pedagogical sciences. She works at a school for children with motor impairments. The third author holds a PhD in pedagogical sciences. Her research addresses the social-emotional development of adolescents and young adults with vision impairments. The fourth author also holds a PhD in pedagogical sciences. His research addresses the development of young children with vision impairments or multiple disabilities. The fifth author was trained in human movement sciences and holds a PhD in social sciences. One of his research projects focuses on sports and movement of children with a motor impairment. Contributions of all authors are based on the Contributor Role Taxonomy (CRediT) (42).

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EDITED BY
Corinna M. Bauer,
Massachusetts Eye & Ear Infirmary and
Harvard Medical School, United States

REVIEWED BY
Marlou J. G. Kooiker,
Erasmus Medical Center, Netherlands
Linda Lawrence,
Independent Researcher, Salina, KS,
United States
Nicole Ross,
New England College of Optometry,
United States

*CORRESPONDENCE
Nicola McDowell
✉ n.mcdowell@massey.ac.nz

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Hiding in plain sight: children with visual perceptual difficulties in schools

Nicola McDowell[1]*, Helen St Clair Tracy[2], Andrew Blaikie[2], John Ravenscroft[3] and Gordon N. Dutton[4]

[1]Institute of Education, Massey University, Auckland, New Zealand, [2]Infection and Global Health Division, School of Medicine, University of St Andrews, St Andrews, United Kingdom, [3]The Scottish Sensory Centre, Moray House School of Education and Sport, University of Edinburgh, Edinburgh, United Kingdom, [4]Department of Vision Sciences, Glasgow Caledonian University, Glasgow, United Kingdom

Cerebral visual impairment (CVI) is increasingly being recognized as a significant cause of visual difficulties in children, particularly those with typical visual acuity, who nonetheless struggle in educational settings. This narrative review aims to elucidate the nature and impact of visual perceptual difficulties (VPD) associated with CVI in school-aged children, who often remain undiagnosed due to the current erroneous focus on visual acuity as a required diagnostic criterion for CVI. The review synthesizes findings from recent studies, highlighting that up to 3.4% of children in mainstream schools and a higher percentage in special educational settings may experience VPD, which significantly impacts upon their learning and development. The manifestations of VPDs, such as difficulties in motion perception, recognition, and visuospatial processing, are often subtle and can thus be overlooked, leading to misconceptions about the origins of the affected child's abilities and behaviors. The review also discusses the challenges in current diagnostic processes, emphasizing the need for comprehensive history taking and assessments that go beyond standard visual acuity tests. It proposes a multi-faceted approach to identification and support, incorporating both clinical assessments and teacher/parental observations, to better address the needs of affected children. Furthermore, this paper advocates for the inclusion of VPDs in the International Classification of Diseases (ICD 11) to ensure children with these visual issues receive appropriate educational support. By integrating lived experiences of individuals with CVI and the latest research findings, this review underscores the urgent need for awareness and tailored educational strategies designed to support children with VPDs. The findings suggest that without such recognition and intervention, many children with VPDs will continue to "hide in plain sight," facing unnecessary challenges in their educational and social development. The review concludes with recommendations for policy changes and future research directions to improve the identification, classification, and support of children with VPDs within the educational system.

KEYWORDS

cerebral visual impairment, visual perceptual difficulties, children and adolescents, screening and assessment, education support

1 Introduction

Cerebral visual impairment (CVI) is a term used to describe a wide range of visual difficulties due to damage or injury to the visual areas of the brain (Lueck and Dutton, 2015). Unlike visual problems caused by issues with the eyes or the optic nerves (the pathways that carry visual information from the eyes to the brain), CVI arises from dysfunctions in the brain's ability to process what is seen. People with CVI may experience various visual challenges, such as reduced clarity of vision, difficulty seeing in certain parts of their visual field, problems with eye movement control, and/or challenges in recognizing and interpreting visual information (Williams et al., 2021). One common outcome of CVI is visual perceptual difficulties (VPDs). VPDs refer to the brain's struggle to make sense of visual information, which can lead to issues like difficulty recognizing objects or people, understanding spatial relationships, issues with visual attention and processing moving and/or complex visual scenes (Chandna et al., 2021). Despite over a century of awareness about VPDs, with early reports of visual issues observed in soldiers returning from World War I (Riddoch, 1917; Holmes, 1918a; Holmes, 1918b), there is still widespread confusion and misunderstanding surrounding these difficulties. While there have been significant advancements in the past decade which is helping to reduce some of this confusion, much of this information is still being disseminated to professionals working in both the education and health sector. This means that for many, CVI and VPDs are still relatively unknown conditions, which often leads to the mismanagement of children and adolescents with these visual issues. This lack of understanding can be detrimental, especially for those who may struggle in school due to their undiagnosed visual issues related to CVI, which may also lead to inappropriate behavioral diagnostic labelling.

Studies have shown that VPDs are not uncommon in children. For instance, research indicates that about 3.4% of children in mainstream primary schools have visual difficulties related to CVI, even though most have typical visual acuity (Williams et al., 2021). The prevalence is even higher in special schools, where between 23.5 and 58% of children may experience CVI-related visual challenges, often without having a formal diagnosis (Black et al., 2019; Williams et al., 2021). Lueck and Dutton (2015) categorized children affected by CVI into three broad groups: those who attend mainstream schools with typical peers, those who face learning difficulties, and a third group who are the most profoundly affected. Even in this third group, where visual impairments are more easily recognized due to reduced visual acuity, unrecognized VPDs are often a significant part of their challenges (Little and Dutton, 2014). Understanding and addressing VPDs is crucial for providing appropriate support to children with CVI, ensuring they can better engage with their learning environments.

This review aims to explain how to better understand and assist children who struggle with VPDs. By firstly examining both research and personal accounts, we will explore how these difficulties show up in daily life and how they can affect a child's learning and development. We will also discuss the ongoing debate around how CVI is defined, particularly in relation to children in both mainstream and special schools. This is important because many children with these visual issues have typical or near typical visual acuity, yet they still face significant challenges in accessing and interpreting learning material (Tsirka et al., 2020). Given the high number of children affected, this is a critical issue. We will look at how well current systems for assessment, diagnosis, and support are meeting the needs of children with CVI and whether there are better ways to understand and address VPDs. Our goal is to highlight that the current approach may not be sufficient and that more needs to be done to ensure children with CVI are not overlooked. In the discussion section we highlight how drawing on the knowledge from researchers, clinicians, educators and those with the lived experience, and working together, will allow the field to find practical and achievable steps to better identify, and support affected children.

2 Unpacking visual perceptual difficulties

The earliest documented reports of brain related visual difficulties describe a range of visual issues. These include those of the primary visual functions, low visual acuity and visual field deficits (Holmes, 1918a). Reported VPDs include an inability to see more than one of two objects at the same time (simultanagnosia), impaired visually guided movement (optic ataxia), inability to volitionally direct visual gaze (apraxia of gaze), further disturbances of ocular movement, inability to recognize objects, letters, faces, difficulty with reading and writing, disturbances of visual localization and orientation in space and reduced ability to see movement (Bálint, 1909; Holmes, 1918a; Holmes, 1918b; Guzzetta et al., 2009), while recovery of the ability to see only movement after visual brain injury (Riddoch, 1917) has also been described (Arcaro et al., 2019). The work by Goodale and Milner (2013) has been foundational in understanding the visual processing pathways, specifically the theories of the dorsal and ventral streams, often referred to as the 'where' and 'what' pathways. The dorsal stream, responsible for vision for action, runs from the occipital lobe to the posterior parietal cortex at the top of the cerebral hemispheres. In contrast, the ventral stream, which is involved in vision for perception, extends from the occipital lobe to the lower and lateral regions of the cerebral hemispheres in the inferior temporal lobes. More recently the anatomical correlates with the dorsal and ventral stream pathways have been identified, respectively, as being the superior and inferior longitudinal fasciculi (Bennett et al., 2020).

VPDs are often linked to atypical processing in the posterior parietal lobes and are sometimes referred to as dorsal stream dysfunction. While this term is often applied to children experiencing additional reduced acuity and/or visual processing challenges (Dutton, 2009), it's application may be less accurate for those children who present with typical visual acuities, yet still face difficulties in visual processing, such as recognizing faces or objects. The anatomical correlate of the dorsal stream, the superior longitudinal fasciculus, connects the occipital, posterior parietal and frontal lobes. If the occipital lobes are functioning typically, it is therefore, plausible that any dysfunction may arise elsewhere in the pathway, particularly in the posterior parietal regions. This raises the possibility that what is traditionally labelled as dorsal stream dysfunction may, in some cases, involve impairments unrelated to the occipital lobe functions, suggesting a more complex interplay of brain networks. This further supports the view of Merabet and Ravenscroft (2023) that a more nuanced understanding of CVI that goes beyond the traditional application of the two-stream model could provide better insights, especially in cases where visual recognition challenges occur in the absence of evidence of temporal lobe dysfunction or pathology. Future research focusing on the interconnectivity of these networks, probing in depth the nature of the related visual experiences could well prove valuable in refining both diagnosis and the salient support strategies for individuals with CVI.

The posterior parietal lobes can be considered a central hub for VPDs, particularly in children with CVI. Research, such as that by Williams et al. (2021), suggests that children with VPDs most commonly exhibit atypical posterior parietal processing. Affected children fall into different groups based on the severity and extent of their learning difficulties. The first group, as described by Lueck and Dutton (2015), presents with mild difficulties. The second group shows more severe impairments, likely extending to the middle temporal lobes where motion is processed, leading to conditions such as dyskinetopsia or, in severe cases, akinetopsia. The third group, characterized by research from Little and Dutton (2014), exhibits more profound impairments, including reduced visual acuity and narrowed visual fields. All three groups tend to exhibit lower visual field impairments affecting both eyes, which vary in severity, correlating with the extent of the affected brain regions. This impairment occurs because the visual information from the lower visual field travels through the posterior parietal lobes in the form of optic radiations, where it may be disrupted before reaching the occipital lobes. A comprehensive understanding of the varying combinations of CVIs, including VPDs, allows for better-targeted interventions. For instance, group one may experience simultanagnostic vision and optic ataxia with lower visual field impairments, while group two may also exhibit additional dyskinetopsia. Group three, the most severely affected, may additionally suffer from reduced contrast sensitivity and visual acuities. The range of difficulties observed across these groups suggests a CVI spectrum, with increasing severity of visual and learning difficulties correlating with the extent of brain involvement (Table 1). This spectrum approach also opens the door to new measurement strategies for assessing the effectiveness of support, potentially using percentile-based scales.

The research conducted by The Laboratory for Vision Neuroplasticity and others has significantly advanced our understanding of VPDs, particularly concerning motion perception, visual attention, scene complexity, navigation, and recognition difficulties. Children with VPDs often display specific behaviors, such as darting eye movements, slower visual processing in complex scenes, impaired search performance, and difficulty with visually guided movements, which are considered to be due to issues with the dorsal ‘where’ visual pathway (Manley et al., 2023; Bennett et al., 2018; Zihl and Dutton, 2015; Manley et al., 2022; Goodale and Milner, 2013). These challenges, coupled with increased frustration and anxiety (McDowell and Budd, 2018), underscore the need for tailored support based on a clear understanding of the underlying brain processes involved. In summary, while recognition difficulties are common among children with VPDs, they may not necessarily stem from an issue with the ventral ‘what’ visual pathway. Instead, these issues may arise from atypical processing in the posterior parietal lobes. Understanding the specific causes of these difficulties is crucial for providing effective support and ensuring that interventions address the root causes of the problem, not just the symptoms. If not accurately understood, this could lead to the child receiving incorrect or inaccurate support.

TABLE 1 Groups of children with CVI due to posterior parietal lobe involvement.

Groups of children with CVI		
Typical visual acuity		Reduced visual acuity
Working at academic level expected for their age.	Cognitive challenges, behind at school.	Profound visual impairment, often with multiple disabilities
Suggested underlying cerebral visual impairments		
Lower visual field impairment (mild) Simultanagnosia Optic ataxia	Lower visual field impairment (moderate) Simultanagnosia Optic ataxia Dyskinetopsia	Lower visual field impairment (severe) Simultanagnosia Optic ataxia Dyskinetopsia Reduced contrast sensitivity Reduced visual acuity

While it is important to have this scientific/medical understanding of VPDs, in order to truly help children and adolescents, it also seems imperative to deepen this understanding by hearing from those with the lived experience on how these visual issues actually impact them on a daily basis. Fortunately, with this increased focus on CVI and VPD in recent times, more and more children, adolescents and adults are starting to speak up and share their experiences in blogs, news stories, videos and via social media platforms (McDowell, 2024; CVI Scotland, 2024b; ABC News, 2022; Bartiméus, 2019; Koninklijke Visio, 2018; 1News, 2022). In these accounts, people with CVI share specific issues they have at school, such as ‘*I cannot find my friends*’, ‘*I need more time for most activities*’, ‘*I find it hard to follow the line I am reading*’ and ‘*Playing sport is really hard*’ (Bartiméus, 2019). They also describe the impact CVI has on their day to day life, ‘*When there are a lot of people in the room and it is noisy, I have to leave the room*’, ‘*If I have to spend a busy day searching and looking and everything, it gets a bit worse and I have a harder time looking for things*’ (Bartiméus, 2019), ‘*My vision is actually like a puzzle. In fact, everything is. At school it is quite tiring*’ (Koninklijke Visio, 2018), ‘*I can only see one thing at a time and the more stressed I get, the fewer things I can see*’, ‘*Social interactions can be challenging. When people greet me, I cannot see them clearly*’ (ABC News, 2022) and ‘*I am clumsy and I trip over a lot*’ (1News, 2022). They have also shared emotional accounts of the toll CVI has had on their lives, ‘*I always felt dumb*’ (1News, 2022), ‘*All my life, I have been inventing excuses for not having to participate in things. Because of this, I am often not asked to come along anymore*’ (Koninklijke Visio, 2018). ‘*Other kids always think I am blind, or I am half blind. But I'm not. I am not blind*’ (Bartiméus, 2019), and ‘*It was such a relief to learn that I had CVI. It helped me to realize that it is not my fault. There is a reason why things are like they are*’ (1News, 2022). These are typical accounts of CVI that can inform understanding and help guide best practice approaches for supporting children and adolescents with VDP's by hearing first-hand how their CVI impacts their daily lives.

2.1 Defining visual impairment in children and adolescents with visual perceptual difficulties

A significant challenge in supporting children and adolescents with VPDs associated with CVI is the lack of comprehensive visual impairment classification. Traditionally, the International Classification of Diseases (ICD) has primarily focused on visual acuity for classifying vision impairment (World Health Organisation, 2019). This approach often fails to identify individuals with typical or near typical visual acuity but significant VPDs. However, the ICD-11 has made strides in recognizing a broader spectrum of visual impairments. Specifically, it includes the following relevant codes:

- 9C43—Visual impairment with higher visual processing dysfunction
- 9C44—Visual impairment with combined structural and higher visual processing dysfunction
- 9C4Y—Other specified visual impairment
- 9C4Z—Visual impairment, unspecified

These codes represent a significant step forward in acknowledging VPDs and other complex visual impairments that may not be captured by traditional acuity measures. However, for the future, it is clear that specific codes, rather than those labelled "unspecified," will become necessary to secure remuneration for medical and habilitation service providers.

Despite this progress, challenges also persist in the practical application of these classifications, particularly in educational settings. For children and adolescents with VPDs, the lack of a clear, widely recognized diagnosis can still impact their access to educational support and funding. Many education systems continue to rely on more traditional definitions of visual impairment, which may not fully capture the needs of students with VPDs. This gap in recognition and support puts affected children at a disadvantage compared to peers with more easily identifiable visual conditions (such as albinism or nystagmus). The discrepancy can lead to inadequate educational interventions and reduced access to necessary habilitation or rehabilitation services. Moving forward, it is crucial to:

- Increase awareness of the ICD-11codes related to VPDs among medical professionals and educators.
- Develop standardized assessment tools that can accurately identify and classify VPDs.
- Update educational policies to recognize and accommodate students with VPDs, even when traditional visual acuity measures are within typical ranges.
- Agree on a unifying definition of the different types of CVIs so that they can map directly on to the ICD classification system.
- Promote research into effective interventions and support strategies for individuals with VPDs.

By addressing these areas, we can work towards a more inclusive and supportive environment for children and adolescents with VPDs, ensuring they receive the appropriate diagnosis, educational support, and interventions they need to thrive.

2.2 Current challenges in the assessment of visual perceptual difficulties

Another problematic area in the management of children and adolescents with VPD is around the assessment and diagnosis process for CVI. Due to the heterogeneous nature of CVI, the diagnosis is often missed in many children, especially in those with typical or near typical visual acuity (Lowery et al., 2006; Chandna et al., 2021). This was highlighted by McDowell (2020b) in their research looking at the impact that knowledge had on empowering parents of children with CVI. Reports by the 75 parent participants in the research showed it took an average of 3.6 years for a CVI assessment and diagnosis, with the longest length of time being 18 years. Many more stories of lengthy delays in diagnosis, or ongoing fights to get a diagnosis have been shared anecdotally with the authors personally and with others via social media platforms. Ideally, the optimum time for CVI to be identified and diagnosed is in the early years (under 5). However, while some early years intervention programs (for children aged under 5 years old) are aware of CVI many are not. More often than not, a child with CVI and VPDs does not get identified until they have started school, when the difficulties with learning become more apparent in formal learning settings. Another roadblock for many in terms of getting a CVI diagnosis, is the reliance on evidence of injury or damage to the visual brain on neuroimaging, specifically an MRI. However, in one study of 33 children diagnosed with CVI (who all had typical or near typical vision), 23% showed none of the reported MRI findings related to this condition (Chandna et al., 2021), suggesting that up to one quarter of children with CVI may not have evidence of brain injury on neuroimaging and further assessment needs to be undertaken to confirm the diagnosis. Further research needs to be conducted on a larger group of children from all three of the groups outlined by Lueck and Dutton (2015) to confirm these findings.

Even if a child has been identified as potentially having CVI, there is still ongoing debate and little consensus on what further assessments should be conducted. In addition, generally the focus of assessment is to diagnose CVI, which does not allow for evaluation of improvements in visual functioning following interventions or help to identify what specific education support is needed. Assessments covering both purposes would significantly help the management of children with CVI. A number of recent publications have attempted to provide a best practice approach for the assessment and diagnosis of CVI, proposing a multidisciplinary approach that includes medical history and CVI question inventories and questionnaires, ophthalmological and orthoptic assessment, assessment of visual behaviors and direct observation, visual perception tests, ocular movement and posture assessments, neuropsychological assessment, neurodevelopmental tests, neuroradiological evaluation and magnetic resonance imaging, genetic assessment, IQ assessments and clinical electrophysiology (McConnell et al., 2021; Boonstra et al., 2022). However, in many countries, this full battery of assessments would be cost inhibitive and could only be conducted on children or adolescents where it had been predetermined that a CVI diagnosis was very likely. To help with this, Pilling et al. (2022) have proposed a checklist approach for determining whether a child needs a full CVI assessment or not, which can be used by general practitioners or first line optometrists. Within the checklist approach, CVI may be present if two or three of the following criteria are met: (1) presence of risk factor(s), (2) reported or observed atypical visual behaviors, and (3) verifiable visual dysfunction on examination. The authors also outline that CVI is highly likely to be present if all three criteria are met (Pilling et al., 2022). To further support this checklist approach, the authors provide information on what would be classed as high-risk factors, including children who:

- are born premature,
- and/or hypoxic ischemic encephalopathy,
- and/or have cerebral palsy,
- and/or developmental delay,
- and/or down syndrome,
- and/or hydrocephalus.

The authors also outline that atypical visual behaviors may include (but are not limited to) non refractive error reduced binocular visual acuity (presenting as lower visual function impairment), abnormal fixation, visual field deficit or inattention, oculomotor impairments (jerky or smooth pursuits or inaccurate saccades) and VPDs. This checklist approach again highlights the need for effective, accessible validated VPD assessment tools to enable clinicians to confirm the suspected CVI diagnosis following examination.

However, while the majority of children affected by VPDs may present with reported or observed atypical visual behaviors, distinguishing between difficulties caused by CVI and those arising from other conditions such as autism, ADHD, dyslexia, or developmental coordination disorder remains challenging (Chokron et al., 2021). This difficulty is particularly pronounced in young preschool children and those with comorbid conditions, where overlapping symptoms can obscure accurate diagnosis. Therefore, effective methods to identify children with visual issues that specifically differentiate CVI from other conditions are essential. For this, structured clinical history taking emerges as the most effective assessment tool in this context. Unlike standardized questionnaires, it allows for a comprehensive and individualized understanding of the child's visual experiences and behaviors. Chandna et al. (2021) conducted an in-depth evaluation demonstrating the high efficacy of this approach in identifying potential CVI. Their findings highlight that a detailed clinical history can capture subtle visual processing difficulties that may not be evident through observational methods alone, making it unparalleled in its ability to discern CVI-related issues.

While tools like the 5 or 11-question inventory have been used as effective screening processes for identifying children with potential CVI, they have limitations in specificity and may not fully capture the complexity of each child's condition (Gorrie et al., 2019; Chandna et al., 2021). These inventories include specific questions to help identify children with possible lower visual field impairment, a condition that is relatively straightforward to detect and difficult to attribute to other causes. The presence of a lower visual field impairment can indicate additional VPDs, but relying solely on questionnaire responses may not provide the depth of information needed for accurate diagnosis, especially in children with comorbid conditions. Additionally, the L94 Visual Perceptual Battery developed by Ortibus et al. (2009) offers a structured method to assess visual perception in children with CVI. Further studies on the L94 Visual Perceptual Battery have validated its utility, showing that it can effectively identify visual perception deficits in children with complex developmental profiles (Ortibus et al., 2011).

An ongoing challenge is the effective assessment of children who have a combination of difficulties or who are non-verbal. Traditional assessment tools often rely on the child's ability to follow instructions and answer questions, which may not be feasible in this population. This underscores the importance of flexible and adaptable assessment methods. Structured clinical history taking, involving detailed interviews with caregivers and observations of the child's behavior in naturalistic settings, can provide valuable insights when direct communication is limited. In conclusion, while various assessment tools are available, structured clinical history taking remains the most effective method for identifying CVI in children, particularly those with comorbid conditions (Chandna et al., 2024; Dutton and Jacobson, 2001; Philip and Dutton, 2014). It allows clinicians to gather comprehensive information tailored to each child's unique presentation, thereby enhancing diagnostic accuracy and informing appropriate interventions. Incorporating tools like the L94 Visual Perceptual Battery can further support the assessment process by providing evidence-based methods to discern visual processing difficulties, although their limitations should be acknowledged.

Another common method for identifying children with visual issues is through national screening programs, which are often undertaken prior to children starting school at age 5 (Muller et al., 2019). However, as the focus of this screening is mainly on visual acuity, children with VPDs are not identified as having perceptual visual issues and therefore, no further assessment is conducted. This was highlighted by McDowell and Butler (2023) in their research to validate the Austin Assessment (a screening tool for CVI related visual issues). The Austin Assessment was used to screen 271 children in one New Zealand primary school and following further assessment by an ophthalmologist on the children who were flagged in the assessment, 17 were found to have 'a verifiable reason or clinical finding for a positive Austin Assessment result'. In reviewing the children's records and talking with their parents, none had been picked up in the B4 screening process as having a visual issue (New Zealand's vision screening program). This could be easily remedied by simply including CVI screening tools, for example the 11 Question Inventory and the Austin Assessment, in vision screening programs (Chandna et al., 2021; McDowell and Butler, 2023). While further research is needed, initial reports show that together, the 11 Question Inventory and Austin Assessment provide objective and subjective evidence of VPDs that can then be confirmed (or not) following a subsequent CVI specific assessment process (McDowell and Butler, 2023).

2.3 Impact of visual perceptual difficulties on learning and development

The impact VPDs can have on a child's learning and development can be significant, especially if they do not receive early intervention services, as these can impact how a child accesses and processes learning material and experiences. Some of the common challenges experienced by children with VPDs include struggling to stay focused on learning tasks, being easily distracted by competing sensory information in the classroom environment, finding it difficult to copy or to keep up with information being presented in the classroom, being overly sensitive and startling often to movement and noise in the classroom, accompanied by high levels of visual and overall fatigue (Lam et al., 2010; Philip and Dutton, 2014; McDowell, 2021). For many children with CVI, this can then lead to learning difficulties in specific subjects that may require additional learning support in the classroom. While there has been limited research in this area, two studies have shown that the number of children experiencing learning difficulties could be as high as 80% (Williams et al., 2011; McDowell, 2023). Both papers reported that the common learning difficulties experienced by children with CVI included underachievement in reading, math and writing (Williams et al., 2011; McDowell, 2023). These issues are often compounded by disparities in the child's functioning (both visually and cognitively) over the course of the school day. Many external (clutter, noise, movement) and internal (fatigue, hunger, sickness, anxiety) factors can impact on a child's visual abilities, which results in children performing less well in vision related tasks in some situations, as compared with others. This disparity is often misunderstood by the adults around the child, who may incorrectly conclude the that child is uninterested in learning, is lazy or is choosing not to engage in the learning process (Erasmus, 2015). In addition to learning difficulties, children with CVI often experience difficulties with developing and maintaining friendships, high levels of anxiety and an inability to regulate emotions effectively, as well as issues with being able to move around their environment safely and confidently (McDowell and Dutton, 2019; McDowell, 2019; McDowell, 2023).

Together, these issues can greatly impact on a child's physical and mental wellbeing, especially when they see themselves as being different from their peers. In particular, the challenges in developing and maintaining friendships causes significant distress for many affected children. This was highlighted in the research by McDowell (2023) focused on understanding and supporting children with CVI in mainstream classrooms. Both parents and teachers involved in the research reported children sharing with them how lonely they felt and how hard it was for them to not have any friends. Similar experiences have also been shared by both children and adults with CVI, through blogs and social media platforms. The high levels of anxiety and difficulties with emotional regulation can also impact significantly on the lives of both the child and the people around them, especially if the root causes of the anxiety are not well understood. Both parents and teachers have reported children seeming to switch from being relaxed and happy 1 min, to being incredibly distressed and inconsolable in an instant with no apparent reason for the sudden change (McDowell, 2020a; McDowell, 2023). While not reported in the literature, many with the lived experience (children, parents, adults) of CVI have also reported physical health issues such as headaches and digestive issues that they can directly relate to the visual issues (CVI Society, 2024; McDowell, 2024). This is another area that requires further examination and research to ensure appropriate supports can be put into place to reduce these issues.

2.4 Awareness of CVI and visual perceptual difficulties in education settings

In recent times the numbers of children in mainstream education with learning support needs has increased significantly. While it is difficult to determine exact figures (due to differences in identification and reporting between different conditions and countries), in New Zealand, it has been estimated that 15–16% of students have additional learning needs (Wylie and MacDonald, 2020; Bourke et al., 2021). This is compared with between 20 and 30% in England (Bourke et al., 2021) and 37% in Scotland, where numbers have increased from 37,000 in 2007 to 260,000 in 2023, representing over a third of all children in schools in Scotland (Scottish Government, 2023). The reason for this increase is in part due to changes in education policies and better identification and assessment practices for developmental conditions such as autism, attention deficit hyperactivity disorder (ADHD), fetal alcohol spectrum disorder (FASD) and dyslexia (Bourke et al., 2021; Yu et al., 2023; Ministry of Education, 2007; Ministry of Education, 2019). This rise in students with additional needs in mainstream education settings has meant that teachers have had to expand their knowledge base around these conditions, which is often done by engaging in professional learning opportunities (Bonne and Wylie, 2017). While further work still needs to be done in this area, the majority of classroom teachers at least feel confident in working with students who have additional needs who require learning support (Bonne and Wylie, 2017; Wylie and MacDonald, 2020; Nicholson and Dymock, 2015), especially following professional development in specific areas, such as autism (Kossewska et al., 2022), ADHD (Zentall and Javorsky, 2017) and dyslexia (Bell, 2013).

Unfortunately, CVI has not been part of this trend, with children and adolescents with CVI and VPDs likely not included in the prevalence data of students with learning support needs for each country, due to the fact that many are undiagnosed and/or currently do not meet the requisite standards for an atypical visual impairment classification. In addition, CVI is still a largely unknown condition in mainstream educational settings. Recent research by Jayasinghe (2023) found that 98% of primary teachers and 80% of secondary teachers across the UK had never heard of CVI. This is similar to the findings of McDowell (2023) in their research on supporting children with CVI in mainstream classrooms in New Zealand, where all 11 of the classroom teachers involved in the research had never heard of CVI. This lack of knowledge on one condition, yet with a more expanded knowledge base on other developmental conditions is further exacerbating the issue of children and adolescents with VPDs not receiving the support they need. Due to the behaviors children with CVI often display in classrooms settings as a result of their VPDs, for example, difficulties with math and reading, struggling to focus, being easily distracted, difficulties with emotional regulation and social interactions (Lam et al., 2010; Philip and Dutton, 2014; Williams et al., 2011; McDowell, 2023), they are often mistakenly identified as having other conditions such as autism, ADHD, dyslexia and auditory processing disorder (Pawletko et al., 2015; Dutton, 2015).

This has been highlighted in the research conducted by McDowell and Butler (2023) to validate the Austin Assessment. When the list of children identified as potentially having visual issues (due to a positive Austin Assessment result) was presented to the school involved in the research, the school Special Education Needs Coordinator (SENCO) and classroom teachers all reported that they 'knew something was going on with each child'. However, they had not even considered it was the child's vision that was causing the learning difficulties they were seeing in the classroom. Instead, owing to their expanded knowledge base around other developmental conditions, they had attributed the issues to either autism, ADHD, dyslexia or auditory processing disorder. While many children may have these conditions in addition to CVI, as highlighted by Gorrie et al. (2019) in her research looking at the effectiveness of CVI question inventories, which found that virtually all children who potentially had CVI also had been labelled as having other conditions including autism, ADHD, dyscalculia, dyslexia, dyspraxia, ocular visual impairments, deafness, hearing impairment, and intellectual disability. Children with multiple conditions still need appropriate support for their CVI in addition to appropriate interventions of the other conditions.

In contrast, research by Pilling et al. (2022) found that specialist vision teachers, referred to as Teachers of Pupils with Vision Impairment

(TVI) are well aware of CVI and do have some understanding of the visual issues associated with CVI and how to support a child in different education settings. This is likely due to an increase focus on CVI in professional learning spaces in recent years and the creation of web-based resources such as the CVI Scotland website (Ravenscroft et al., 2021). Unfortunately, it seems that this increased knowledge and understanding of CVI amongst TVIs is not being disseminated to mainstream teachers, suggesting that it is not just enough to focus on upskilling TVI's, there also needs to be a targeted approach to teaching mainstream teachers. For children in groups two (who face learning difficulties) and three (who are profoundly affected) of the CVI categories outlined by Lueck and Dutton (2015), due to their additional education support needs, they may already be in a special education placement or receiving additional learning support within a mainstream setting. Depending on the school setting (i.e., a specialized school of the blind and visually impaired compared with a more generalized special school), teachers and support staff may or may not have any awareness and understanding of CVI. This highlights the need for a specific focus on CVI to be a mandatory component of tertiary training programs for Vision and Specialist teachers who work in vision and special school settings.

2.5 Current approaches to supporting children and adolescents with visual perceptual difficulties in the classroom

Until recently, owing to a paucity of research focused specifically on supporting children and adolescents with VPDs in the classroom, many were being supported using approaches that had been shown to be effective for children with ocular visual issues (OVI) (Martin et al., 2016; McDowell, 2021). Examples of this include, enlarging printed material, using magnification devices and having children sit in locations in the classroom that faces them towards cluttered and busy spaces (to bring them closer to the information). While this may help some children with CVI, for others it may make it even more challenging to access the learning material (Martin et al., 2016; McDowell, 2021). Due to the difficulties with visual crowding and clutter, it is important to reduce the amount of visual information a child with visual perceptual difficulties needs to process at any one time. Enlarging the material, or bringing a child closer to the front of the classroom, simply increases the visual information and makes the visual scene even more complex (McDowell, 2024). With this understanding, vision educators are starting to move away from using OVI strategies in the classroom, instead exploring other options that could be more effective for children with VPDs. For example, decluttering learning spaces and material, introducing calm breaks and teaching emotional regulation strategies (McDowell, 2020a; McDowell, 2023; Hokken et al., 2024). However, it is important that this trend is supported by the use of validated, effective approaches that take into account the unique visual considerations of children and adolescents with VPD. Currently, as we have shown, all three of our groups are not getting the support they need in school, to the potential detriment of each affected child.

3 Discussion

While there have been some improvements in recent years, overall, the current approach to supporting children and adolescents with VPDs in schools is not, as a whole, working. This is highlighted by the challenges many children with these visual issues in mainstream classrooms are still experiencing, including learning difficulties, struggles with friendships and social interactions, challenges with emotional regulation and mobility issues (Williams et al., 2021; McDowell, 2023; McDowell, 2019; McDowell and Dutton, 2019). In others, learning is so severely affected that they need special educational placements. While these issues in themselves make navigating the school day incredibly hard for children with CVI, without the right support, this could also lead to additional mental health issues that impact greatly on overall health and wellbeing (Goodenough et al., 2021; University of Bristol Research, 2019; McDowell, 2019). Historically, decisions around the management of children with CVI, from identification to assessment, diagnosis and support have been based on external factors, such as funding (both in health and education), limitations in current systems (in both health and education) and current understandings of CVI and VPD (from the research). This approach needs to be re-evaluated with a more collaborative agenda where different voices in the community, including researchers, practitioners and professionals, parents and children with the lived experience considered. In this way, decisions are based on evidence-based practice, where the intersection between the three circles of evidence (research evidence, practice evidence and lived experience evidence) guides a best practice approach (Bourke et al., 2005). An evidence-based practice approach will also provide an opportunity to bring the (at times) competing views of the health and education systems together to ensure every child and adolescent with VPD around the world has equal opportunity to thrive in their education journey.

3.1 Identification of visual perceptual difficulties

The first step in the management of children and adolescents with VPD is identification. Currently, the main avenue for identifying common visual issues is vision screening, which is commonly done on all newborn babies, with more comprehensive screening on babies at high risk for visual issues (such as those born premature, infants with cerebral palsy, HIE and other neurological challenges). Further screening is done in most countries at around age 4 and in some countries again at age 11 (i.e., New Zealand). Other avenues include identification by medical practitioners and other professionals such as teachers and therapists and identification by parents and family members. While all of these avenues provide ample opportunities for a child's VPDs to be identified early, due to the lack of awareness of CVI across the general public, they are not being utilized effectively. To help with this, frontline medical providers, including general practitioners and optometrists need to have a good understanding of CVI, including the range of visual issues associated with this condition, how they might present, and the common manifestations and behaviors demonstrated by children at different developmental stages. For example, in young children this might present as showing signs of distress in busy, cluttered and noisy environments and not making eye contact with familiar and unfamiliar people. For school aged children, this might present as difficulties with learning to read and write, not learning the same as their peers, difficulties with friendships, avoidance of sporting and physical activity and high levels of anxiety (Williams et al., 2021; Bartiméus, 2019; McDowell, 2023).

In line with this, front line medical providers (GP's and optometrists), should also have access to validated CVI screening tools, so that when visual issues are observed or parents share their concerns about their child's vision, functioning or behavior there is a clear first step in the pathway to identification. For this to be effective however, the screening process needs to include a subjective observation of the child's visual abilities and an objective observation of the child's visual functioning, for instance combining a question inventory with a screening app or tool. To ensure this approach is manageable and easy to integrate into short consultation timeframes, the screening would need to be able to be done in under 5 min and clear next step guidelines outlined for medical providers and parents (Legge, 2024). This combined approach to CVI screening could also be included in vision screening programs for school aged children to expand them from solely focusing on visual acuity, to also including visual perceptual difficulties. This approach has been shown to be effective in one primary school in New Zealand as part of the research to validate the Austin Assessment (McDowell and Butler, 2023). Again, clear next steps would need to be in place to ensure that any child flagged in the vision screening process had a clear pathway for further assessment and possible diagnosis of CVI.

Another avenue for identification, especially in the children whose VPDs are impacting them in an education context but may not be severe enough for a CVI diagnosis (under the current thresholds), is the school itself. Teachers and other learning support staff, such as Special Education Needs Coordinator's (SENCOs) are often the first to observe learning difficulties or different behaviors in children or are the first people parents raise their concerns about their child with. Simple to use, effective screening tools that help to identify potential VPDs could be used in these situations, not for the purpose of obtaining a diagnosis, but instead, for the purpose of guiding education support. For instance, the initial 11 Question Inventory followed by the 39 Question Inventory could be used to determine the main challenges for the child (Dutton et al., 2010; Houliston et al., 1999; Chandna et al., 2021; Hellgren et al., 2020).

If a diagnosis was needed for education funding and support services, the same pathway of being seen by a frontline medical provider (GP, optometrist, pediatric ophthalmologist) could be used. However, for schools to be able to help with identifying children with VPD, they firstly need to be aware that these visual issues exist in the first place. With the increase in children with additional learning needs in recent times, teachers have already had to expand their professional knowledge base to include identifying and supporting children with a range of developmental conditions, including autism, ADHD, dyslexia and auditory processing disorder (Bonne and Wylie, 2017; Wylie and MacDonald, 2020). CVI and VPD could easily be added to this by providing professional development opportunities to teachers and learning support staff on the specific visual issues, how they often present and the impact they have in a learning environment. This approach has been shown to be effective for other conditions, including autism, ADHD and dyslexia (Kossewska et al., 2022; Zentall and Javorsky, 2017; Bell, 2013) and could be just as effective for CVI.

Another group that could help with the identification of VPDs are parents. As the experts on their child, they see the daily impact of the visual issues both at home and at school. This was highlighted in the research by McDowell (2023) on supporting children with CVI in mainstream classrooms. Parents involved in this research shared that they had known something was going on with the child's learning but could not quite put their finger on what the issue was. Many had explored other conditions such as dyslexia, but were unsure about how to go about identifying what was going on (McDowell, 2023). This was similar to the finding by Gorrie et al. (2019) in their research that assessed the effectiveness of two CVI question inventories on children who did and did not have CVI. CVI was indicated in many who had not been identified as having CVI (55 of 431 using one question inventory, 166 of 431 using the other question inventory). However, the vast majority had already been identified as having other conditions, including dyslexia, autism and ADHD. Although the research did not explore how the children had been identified with these conditions, it could be surmised that the increased knowledge about developmental conditions in recent years, led to parents recognizing possible characteristics of these conditions in their children and seeking out further information and support, which in turn led to a diagnosis.

Empowering parents, by providing access to information about CVI and a simple, easy to use, effective CVI screening tool has the potential to significantly improve the identification of children who have these visual issues, which would in turn ensure they receive the support they need. This approach has been shown to be effective for one family in New Zealand, who used the Austin Assessment with their 13-year-old son who had been diagnosed as having learning difficulties. They had been recommended to do further vision assessment; however this was not easily accessible to the family and cost prohibitive (Austin Assessment, 2024). Watching their child do the Austin Assessment and the results shared in the report, helped them to see exactly what was going on with his vision and how to help him at home and at school. Empowering parents in this way, will also ensure parents feel confident in supporting their child and are able to take a central role in any education support plan (McDowell, 2020b).

3.2 Assessment of education needs for children and adolescents with visual perceptual difficulties

From research, it is evident that the number of children with CVI and VPDs is significant, with some estimates indicating it may affect approximately 3.4% of children (Williams et al., 2021). However, it is important to note that this figure is based on a single study, and as yet, there has been no replication. Caution should be taken in how widely we generalize these findings until further research is conducted. Moreover, the number of children affected by CVI is expected to rise, partly due to medical advancements in treating conditions associated with CVI (Good et al., 1994; Chong and Dai, 2014). This issue is not restricted to high-income countries; the increase is also being noted in low-and middle-income countries (LMICs). Factors such as poor maternal care leading to hypoxic–ischemic encephalopathy (HIE) and the increasing number of neonatal intensive care units (NICUs) in urban centers, resulting in more premature neonates with periventricular leukomalacia (PVL), contribute to this rise. However, healthcare systems and education infrastructures in these regions are often overstretched, underfunded, and not fully equipped to manage these challenges. Additionally, babies in LMICs are more likely to be exposed to *in utero* infections and pre-natal brain injury, which can further contribute to the prevalence of CVI (Ssentongo et al., 2021; Paneth, 2021; Houweling et al., 2007; Patial and Swaminathan, 2018).

The growing number of children with CVI has also raised concerns about overburdening already strained public health systems in developed countries (McDowell and Butler, 2023). However, if decisions and policies focus solely on economic factors, there is a risk that some children and adolescents with VPDs could be overlooked, especially when traditional definitions and diagnostic criteria are rigidly applied. It is important to recognize that these more "traditional" definitions, while potentially viewed as strict, are often shaped by diagnostic conventions that leave little room for flexibility. As a result, children who are functioning at a high level, despite their visual impairments, may be missed as they are seen to be coping within the expected norms, although they may not have reached their full potential. One alternative is to focus on educational needs rather than solely on visual deficits when assessing children. By integrating this approach into early intervention programs, it may be possible to ensure children receive the necessary support early in their development, reducing the demand on public education services in the long term. This could also lower the lifetime economic costs associated with ongoing support. However, this approach carries a downside: children who work hard to overcome their visual impairments may be overlooked, as their high performance may mask their struggles. Further consideration is needed on how best to support children with CVI to reduce the impact of this constant 'over functioning.'

To support the assessment of educational needs, an evidence-based approach can be adopted, drawing on the expertise of researchers, practitioners, and families (Bourke et al., 2005). Once a child is identified as potentially having CVI or VPDs, a comprehensive assessment should be conducted to develop an individualized CVI profile. However, this need not require lengthy medical appointments; comprehensive history-taking inventories have proven effective in identifying specific CVIs in children (Hellgren et al., 2020). These inventories rely on parents and, in some cases, the child, to report daily challenges. Since parents observe their children most closely, they can provide valuable insights into their child's functioning (Pueyo et al., 2014). Parents can complete the inventories at their convenience, allowing specialists to review the information before appointments, which can streamline the assessment process. This approach empowers parents to take an active role in their child's education and support plan (McDowell, 2020b). However, while empowering parents can be positive, there is also a risk that providing limited information might lead to overconfidence or unrealistic expectations. A balance is needed to ensure that parents are well-informed but also aware of the complexities of their child's condition.

In addition to history-taking inventories, percentiles can be introduced into CVI assessments to objectively determine where a child sits on the VPD spectrum compared to peers (Hellgren et al., 2020). However, subjective assessments are equally important (Chandna et al., 2021). Currently, there is no easily accessible standardized tool for accurately identifying and classifying VPDs, and developing such tools should be a priority for the research community. Clinicians require access to assessment tools that extend beyond visual acuity tests to ensure they can effectively evaluate children with VPDs. Moreover, the role of vision education and therapy practitioners must be considered. Teachers of the visually impaired (TVIs) play a key role in conducting functional vision assessments (FVAs), which assess a child's visual abilities in real-world settings, such as classrooms, rather than clinical environments. Incorporating validated VPD-specific tools into FVAs would help ensure that children receive appropriate interventions. Regular assessments are needed to determine the best way to present learning materials and optimize learning environments for each child. To implement effective FVAs and clinical assessments, clinicians and practitioners must have a thorough understanding of VPDs, highlighting the importance of including CVI in tertiary training programs.

Classroom teachers also play an important role in assessing a child's educational needs. History-taking inventories can be useful here as well, as teachers may observe different challenges in school compared to those seen at home, where the sensory environment is less complex. This was evident in McDowell (2020a) case study, where a parent observed, "*At home she is like a typical child. She can do almost everything her siblings can do. But when she gets to school, she becomes this hugely disabled child who appears so different to her peers. I just wish the school got to see the child we see at home*." Teachers often notice fluctuations in a child's visual functioning and behavior over the course of the school day (McDowell, 2023) and can provide valuable insights into the accommodations and strategies that might be needed at different times and in varying contexts. A collaborative, multidisciplinary approach that integrates clinical, practitioner, educator, and parental input can create an evidence-based assessment of a child's educational needs. This approach ensures that the child receives the most appropriate support while reducing the burden of lengthy and costly assessments on any one service. By working together, all stakeholders can help minimize learning and developmental challenges, including social, emotional, and physical health issues, ensuring that each child has the best opportunity to succeed in their educational journey.

3.3 Education support

For any education support to be effective for a child or adolescent with CVI and VPDs, it needs to be understood that they will have their own unique presentation of visual issues (even in children with the same underlying cause) (Hyvärinen, 2019). In addition, the impact these visual issues have on them in a classroom environment will depend on personality, life experiences and natural abilities. Traditionally each child would go through a thorough assessment to determine their individual education needs. However, there are a few overarching principles that should be considered when developing individual education plans for any child or adolescent who has VPDs. The first of these is to reduce visual clutter in the learning environment, including the learning material (i.e., worksheets, online material and in books) (Hokken et al., 2024). The importance of this was highlighted by an adolescent with CVI in a blog on effective support, where they explained '*What I tell some of my teachers is to make it as boring as they think they possibly can, and then make it even more boring*', as this approach results in the material being accessible for them (CVI Scotland, 2024b). For many children with CVI, the main barrier in accessing and processing learning material is the amount of visual clutter in their classroom environments. Research by Little and Dutton (2014) and McDowell and Budd (2018) has shown that when this clutter is reduced or blocked out for children with severe CVI (including reduced visual acuity) in special school classrooms, it significantly improved the child's learning, behavior and visual functioning. This approach of reducing visual clutter within the learning environment was also shown to be effective for children with VPD in mainstream classrooms in recent research by McDowell (2023). In this research, classroom spaces, such as surrounding the large TV screen (where all learning material was displayed), the teacher's desk and classroom walls were decluttered for a period of 10 weeks. The teachers involved in the research reported that they noticed improvements in engagement and levels of achievement in the children who had been identified as having VPDs. When the children were asked if they liked the decluttered spaces, their comments included statements such as '*There is less noise in my brain when the classroom is not so cluttered, and it is easier to think*', and '*My head does not hurt as much at the end of the school day when there is not so much stuff in the classroom.*'

Another important overarching principle for supporting children and adolescents with VPD in the classroom is ensuring everyone involved in supporting the child, including the child themselves, understands the visual issues and the impact they have in an education setting. This can be achieved by providing individualized CVI education sessions for parents, teachers and other professionals working with a child with visual perceptual difficulties. This approach has been shown to be effective by McDowell (2020a) and McDowell (2023) in research with children in mainstream education settings. The education sessions were provided after a thorough assessment of the child's education needs had been conducted and included an outline of the specific visual issues the child had, how they impacted them in the classroom and the individualized approaches that were needed to support the child. CVI simulation (CVI-SIM) videos (CVI Scotland, 2024a) were also used to help show what the world might look like for the child. Both parents and teachers have reported that these sessions really helped them to understand what the world looked like for the child (McDowell, 2020a). One teacher even noted '*as soon as I could empathize with what life was like for them in the classroom, suddenly it became very clear how to help*' (McDowell, 2023). The CVI education sessions and sharing of the CVI-SIM videos can also help the child themselves, to understand their own visual issues better. Children have shared with the authors anecdotally that they can identify with the CVI-SIM videos, as they show what they experience in everyday life. Sharing these videos with their teachers helps to ensure the teacher then understands why certain learning activities can be so challenging for them.

Another consideration many with the lived experience of CVI and visual perceptual difficulties are requesting (both adolescents and adults), is the opportunity, where appropriate, to learn blindness strategies from a young age such as braille and Orientation and Mobility techniques (i.e., the use of a mobility cane or guide dog). Currently, most educational approaches introduced for children with CVI require the use of the child's vision (CVI Scotland, 2024b). However, for many, the constant strain and over functioning required to be able to access the learning material equal to their peers with typical vision over the course of the school day causes significant visual and overall fatigue that often reduces their access to their vision (Bartiméus, 2019). This was shared so eloquently at a recent conference by four adults sharing their perspectives in a lived experience panel. All four described situations where the visual scene was so overwhelming, that it was easier to switch off their vision and use their other senses to access information and navigate their surroundings (Andrésdóttir et al., 2024). As one of the panelists described:

> *'I have a limited amount of energy supplies for the entire day. Certain activities take up significant amounts of this supply, leaving me with even less supplies to get me through other activities over the course of the day. Once I have depleted my daily supply, my visual function significantly decreases, and I have to try to get through the rest of the day with even less vision than normal'.*

A simple solution to overcoming this issue and preserving energy levels for activities where vision is the best option, is to teach alternative non visual options to children and adolescents with VPDs as another tool to have in their toolbox. As noted by Yellowstone in their blog series on the CVI Scotland website '*blocking out my vision actually enables me to navigate the school more easily*' (CVI Scotland, 2024b). However, this approach might not work for all children and adolescents with visual perceptual difficulties, highlighting that the main premise for any approach introduced to help in a classroom and school environment needs to be based on individual needs, appropriately matched to each individual child's ability level, needs and goals.

4 Conclusion

While there has been significant progress in expanding the knowledge base around children and adolescents with VPDs, especially in the last 10 years (based on the number of recent publications in this area), there is still a lot of work to do. For effective and lasting change that will ultimately improve the lives of affected children and adolescents, we believe the community, including researchers, clinicians, practitioners and those with the lived experience (individuals with CVI and VPD and their parents) need to work together to make informed decisions around identification, assessment and education support. For these to be effective, we also need policy makers, including the World Health Organization to better understand CVI and VPD and be held more accountable for improving outcomes for children with CVI. While we offered some solutions and approaches for identification, assessment and education support, we have also identified some key areas where further research is urgently needed. These include:

Creating effective, accessible validated VPD assessment tools (for clinical use and in FVA), that not only help to determine initial needs, but also provide a method for assessing the effectiveness of interventions over time. The use of percentiles, as highlighted by Hellgren et al. (2020) could be further explored for this purpose. It is also important that these tools cater for the wide spectrum of children affected, including those with communication limitations.

Further research on the impact of VPD's when not supported effectively in an education context and the correlation with the increase in numbers of children with learning support needs that require additional classroom support.

More research on the impact of VPD's and over functioning (working so hard each day just to access the visual world the same as their peers) on emotional and physical wellbeing and health.

Research on the benefits of the use of non-visual strategies (including braille and O&M strategies) for children and adolescents with CVI.

5 Limitations

The aim of this review is to show that despite progress in recent years in the knowledge base around children and adolescents with VPDs, more work still needs to be done to recognize individual needs and to address them effectively. We believe we have provided a comprehensive overview of this topic, bringing together different schools of thought, outlining the main issues and potential solutions. In the conclusion, we also highlight the current research gaps and what we feel needs to be done to address these. What became apparent as we were writing this paper, is the paucity of research in this area, especially in terms of supporting children with CVI and VPDs in school, in a range of different education settings, hence the reliance on only a few papers. To help overcome this, we have included firsthand accounts from people with CVI and those supporting children and adolescents with CVI (teachers, parents), as we believe it is important that these voices are heard and included in any future work to progress the field.

Author contributions

NM: Conceptualization, Writing – original draft, Writing – review & editing. HS: Conceptualization, Writing – original draft, Writing – review & editing. AB: Conceptualization, Writing – review & editing. JR: Conceptualization, Writing – review & editing. GD: Conceptualization, Writing – review & editing.

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EDITED BY
Corinna M. Bauer,
Harvard Medical School, United States

REVIEWED BY
Melinda Chang,
Children's Hospital of Los Angeles,
United States
Nicola McDowell,
Massey University, New Zealand

*CORRESPONDENCE
Jem Martin
✉ matinj@neco.edu

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Rasch analysis and targeting assessment of the teach-CVI survey tool in a cohort of CVI patients

Jem Martin[1]*, Chris Bradley[2], Barry S. Kran[1] and Nicole C. Ross[1,3]

[1]Department of Specialty, Advanced Care and Vision Science, New England College of Optometry, Boston, MA, United States, [2]Wilmer Eye Institute, Johns Hopkins School of Medicine, Baltimore, MD, United States, [3]Department of Psychology, Northeastern University, Boston, MA, United States

Purpose: Cerebral Visual Impairment (CVI) is the leading cause of pediatric visual impairment. Given the diversity of clinical presentations of CVI, we are interested in whether questionnaires appropriately target the spectrum CVI cases, specifically the Teach-CVI Screening Tool. Rasch analysis is a standard psychometric technique for assessing the targeting of questionnaire items, however this analysis technique has not yet been applied to this questionnaire.

Methods: We performed a retrospective review of clinical CVI cases from the NECO Center for Eye Care at Perkins School for the Blind from January 2016 to December 2022. Electronic medical records were reviewed to identify patients with an ICD-9 or ICD-10 code of CVI or other neurological visual impairment. Age, gender, diagnoses, visual acuity, contrast sensitivity, visual fields, ocular alignment, and Teach-CVI responses were collected. We applied the method of successive dichotomizations, a polytomous Rasch model, to estimate item measures and person measures from the survey. Targeting of questionnaire items to the sample population was explored by comparing estimated item measures to person measures. Multiple linear regression was used to determine which factors influence patient visual ability (i.e., Teach-CVI person measure).

Results: 119 patient records were included, 54% of which were male. The mean age was 8.9 years (SD = 6.12) with a range of 0 to 33 years of age. Mean visual acuity was 0.46 logMAR (SD = 0.40), or 20/57. The majority of patients in the sample had a co-occurring visual disorder in addition to CVI (84%), the most frequent being strabismus (69.9%) or visual field loss (25.3%). Item measures ranged from -2.67 to 1.77 logits (SD = 0.76), with a mean of 0 logit by convention. Estimated person measures ranged from -2.19 to 3.08 logits (SD = 1.10) with a mean of -0.03 logit. The range of item measures covered 93.3% of the person measures, and all person measures, except one, were within one logit of an item measure. Visual measures were not statistically significantly associated with Teach-CVI person measures.

Conclusion: The findings from this study suggest that the Teach-CVI survey is well targeted and an appropriate patient reported outcome measure for CVI.

KEYWORDS

Rasch analysis, cerebral visual impairment, cortical visual impairment, visual development, visual function, visually impaired children

Introduction

Cerebral Visual Impairment (CVI) is the leading cause of visual impairment in children in developed countries (1–3). CVI is currently defined as "a verifiable visual dysfunction, which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment" (4, 5).

While some impairments may be found in the primary visual cortex, CVI is characterized by deficits in visual processing beyond the primary visual cortex (5, 6). These deficits are not easily detected through routine eye examinations, which has led to the development of instruments – some are used to guide the clinical interview (7, 8) while others are used as screening tools to screen for symptoms potentially related to CVI (9, 10).

One such instrument is the Teach-CVI screening tool (11). Created by the Teach-CVI partnership, Teach-CVI has three different levels – three different sets of questions (or items) regarding the frequency of behaviors known to be related to CVI for a given developmental age and/or motor ability. Level 1 has 19 items and is intended for those who are non-ambulatory. Level 2 has 35 items and is intended for individuals with a developmental age between 2 and 6 years old. Level 3 has 45 items and is intended for individuals with a developmental age of between 6 and 12 years old. Responses are on a 4-point Likert scale: (1) never, (2) occasionally, (3) frequently, and (4) always.

Teach-CVI responses are typically analyzed by tallying the number of times a person responded with a 3 or a 4, i.e. a "positive score" – for some items the scale is reversed (i.e., a "positive score" corresponds to a response of 1 or 2) and must be flipped prior to analysis. Analyzing scores in this way is problematic for two reasons. First, a Likert scale is not necessarily an equal interval scale. For example, the difference between 1 and 2 on a Likert scale does not *a priori* represent the same difference in latent trait (i.e., functional vision in the case of Teach-CVI) as the difference between 2 and 3, or between 3 and 4. Furthermore, dichotomizing responses into "positive" and "non-positive" does not utilize the full 4-point Likert scale.

Rasch analysis is a standard psychometric technique used to convert Likert scale responses into an equal interval scale. Rasch analysis estimates item measures and person measures on the same measurement scale, allowing them to be directly compared – item measures represent the magnitude of the underlying latent trait required by the item while person measures represent the magnitude of the latent trait possessed by the person (see (12) for further description of item and person measures in low vision rehabilitation, and (13) for further discussion of measurement with Rasch analysis). Both item and person measures are calculated in units of logit, whereby higher item measures indicate more difficult items, and higher person measures represent having more of the latent trait being measured. In our application, with Teach-CVI, this trait is visual ability. Unlike raw scores or sums or means of raw scores, Rasch analysis estimates person and items measures on the same equal interval scale despite missing data for some person-item combinations.

Rasch analysis has become the preferred psychometric technique for analysis of patient reported outcome questionnaire data in many fields, including low vision rehabilitation (12, 14–16). One reason is because Rasch analysis allows assessment of targeting – how precisely a set of questionnaire items can measure the latent trait in the sample of persons. Mathematically, targeting is assessed by comparing the item measure distribution to the person measure distribution, which ideally should be similar. Targeting is important because person measures can be more precisely estimated the closer they are to item measures. A questionnaire that is not well targeted may have ceiling or floor effects – person measures far above the item measure distribution (a ceiling effect) or person measures far below it (a floor effect), and thus may not be suitable for measuring the latent trait in a particular patient sample. The problem with ceiling and floor effects is that it becomes difficult to discriminate between person measures near the ceiling or the floor. Targeting should be assessed during the development of any questionnaire that uses Likert scales (16). In some instances, questionnaires were widely adopted and utilized before ceiling or floor effects were discovered, making it more difficult to modify the questionnaire afterwards (14, 17–24).

Another psychometric property of questionnaires that should be evaluated is unidimensionality, i.e., whether or not all items measure the same construct (same latent trait). A set of items exhibits unidimensionality if the variance in responses to the items can be explained by one source, the latent variable. Factor analysis has been a common method of assessing the dimensionality of questionnaires, in which the number of factors that explain the variance in responses can be explored. Rasch analysis can also be used to assess unidimensionality through its infit mean square statistic (infit).

In this study, we apply Rasch analysis to Teach-CVI to assess targeting and unidimensionality in a sample of patients with a diagnosis of CVI. This psychometric analysis is necessary to determine if Teach-CVI is potentially suitable for use as a patient-reported outcome measure in CVI patients, in addition to its intended use as a screening tool. We also explore how demographic variables and clinical visual function measures (e.g., visual acuity, visual field and contrast sensitivity) influence the latent trait of visual ability as measured by Teach-CVI person measures.

Methods

Teach-CVI responses were retrospectively analyzed from patients with a diagnosis of CVI that presented to the NECO Center for Eye Care at Perkins School for the Blind, a tertiary clinic serving children with low vision and multiple impairments, from January 2016 to December 2022. Charts were identified through searching by ICD-10 codes for cortical blindness, and common etiologies and comorbidities of CVI such as periventricular leukomalacia and hypoxic ischemic encephalopathy. The list of ICD-10 codes searched included all of the following: H47.61X "Cortical blindness", P91.2 "Periventricular leukomalacia", P91.60 "Hypoxic ischemic encephalopathy." The ICD-9 code for cortical blindness (377.75) was also used. A total of 183 charts fit the search criteria. Of those, 136 had at least one completed Teach-CVI, and of those 119 had a diagnosis of CVI and were included in our study.

Demographic information (age, gender, neurologic history and comorbid diagnoses) and visual measures (visual acuities, contrast sensitivity, presence of strabismus, presence of visual field defect) from the most recent vision examination were collected, as well as Teach-CVI responses. While the Teach-CVI tool has been highly utilized at the NECO Center for Eye Care at Perkins School for the Blind, only those patients for which the complete raw data Teach-CVI forms were available were included. Charts with only summary score information were excluded. In cases where multiple surveys were completed for the same patient, the survey completed most recently by a parent was selected. Those without a diagnosis of CVI were excluded.

Examination approaches varied between patients in order to allow for the maximum engagement from the patient, meaning that testing methods for clinical vision measures were not consistent between patients. Distance visual acuity tests included various electronically displayed formats as well as the Feinbloom chart. Near acuity tests included various near cards as well as recognition tests (such as Teller Cards). Qualitative assessment of acuity included fix and follow and light perception, and had their visual acuity reported as a missing variable. Contrast sensitivity testing utilized the Double Happy cards (25), Pelli Robson (26), Berkeley Blinking Squares (27–29) and MARS cards (30). Table 1 reports the type of visual acuity charts and contrast sensitivity tests used in our sample of CVI patients.

TABLE 1 Clinical tests of visual acuity and contrast sensitivity utilized.

Visual Acuity Test	n (%)
Snellen acuity	38 (31.93%)
Patti pics/Lea symbols	27 (22.69%)
HOTV	7 (5.88%)
ETDRS	5 (4.20%)
FrACT*	2 (1.68%)
Feinbloom	1 (0.84%)
Teller Acuity Cards	35 (29.42%)
Missing	4 (3.36%)
Contrast Sensitivity Test	
Pelli-Robson Card	31 (26.05%)
MARS	3 (2.52%)
Double Happy Cards	76 (63.87%)
Berkeley Blinking Squares Contrast App**	4 (3.36%)
Missing	5 (4.20%)

*The Freidberg Visual Acuity Test (FrACT) digital visual acuity test, (31, 32), https://michaelbach.de/fract/

**Berkeley Blinking Squares Contrast app (27–29); https://apps.apple.com/us/app/berkeley-contrast-squares/id979063261

This study was reviewed and approved by the institutional review board (IRB) at the New England College of Optometry and adhered to the tenets of the Declaration of Helsinki. Requirement for informed consent was waived by the IRB due to the retrospective nature of the study.

Teach-CVI

Teach-CVI has three different levels (i.e., different sets of questionnaire items) that can be administered based on mobility and/or developmental age. The level chosen for completion was determined by the provider at the time of the examination after review of the patient's medical, ocular, and educational records, and after time spent with the patient during the examination.

Survey items are asked on both an ability and disability scale (e.g. "Makes eye contact" is on an ability scale, while "Has difficulties with looking at objects" is on a disability scale). For the purposes of the analysis, responses to items using a disability scale were "flipped" to lie on an ability scale for consistency across items.

Rasch analysis

Teach-CVI responses were analyzed using the method of successive dichotomizations (MSD), which is a polytomous Rasch model that estimates person and item measures on the same equal interval scale regardless of the number of response categories, an improvement over previous Rasch models (33). The R package 'msd' was used for analysis. Responses from all three Teach-CVI levels were simultaneously analyzed, so that items common to different Teach-CVI levels had their item measures estimated from all participants who responded to those items.

To assess targeting the distributions of estimated person and item measures were compared to each other. Ideally, the two distributions are similar so that more items are available to precisely estimate the majority of person measures. To confirm that Teach-CVI measures a single latent trait, we assessed unidimensionality through the person and item infit mean square statistics (infits). The infit compares the observed variance in the responses to the expected, which is 1 if the unidimensionality assumption in Rasch models is satisfied. Most person and item infits should lie between 0.5 and 1.5 for evidence of unidimensionality (34).

Statistical analysis

Multilinear regression was used to determine the influence of different variables on the estimated person measures. These variables were: demographic information (age, gender), clinical visual function (e.g., visual acuity, contrast sensitivity, presence of visual field deficit, presence of strabismus), neurologic diagnoses categorized according to prevalence rates in CVI (35) (specifically, categorized as: “gestational and perinatal inflammation” including periventricular leukomalacia, hypoxic ischemic encephalopathy, hypoxia/anoxia, stroke, hydrocephaly, and meningitis; genetic/metabolic conditions, traumatic, and “other”) and level of impairment (categorized as: diagnosis of CVI only, CVI and other visual diagnoses, CVI and co-occurring physical impairment (e.g., cerebral palsy, hemiplegia), or multiply disabled with CVI and both a visual and physical disability).

Results

Table 2 shows demographic and clinical information of our sample. A total of 119 patient electronic health records were included in the review, 54% of which were male. The mean age was 8.9 years (SD = 6.12) with a range of 0 to 33 years of age. Mean visual acuity was 0.46 logMAR (SD = 0.40), or 20/57, with a range from 20/10 to 20/1,9000 Snellen equivalent. The majority of patients, 48%, had multiple impairments with both visual and physical impairments in addition to CVI. Those with a diagnosis of CVI and another visual diagnosis only made up 36% of the sample, and 6% had a physical disability only in addition to CVI. The most frequent co-existing visual condition was strabismus (69.9%), and the next most common ocular/brain based visual function finding was the presence of a visual field deficit (25.3%). The range of co-occurring ocular and binocular pathologies that were noted included: nystagmus, optic atrophy, binocular vision dysfunction, cranial nerve palsies, retinal conditions including retinopathy of prematurity, retinitis pigmentosa, colobomas, and other specific non-refractive conditions.

Figure 1 compares the distribution of estimated person measures (teal) against the distribution of item measures (grey). Item measures ranged from -2.67 to 1.77 logits (SD = 0.76), with a mean of 0 logit by convention. Estimated person measures ranged from -2.19 to 3.08 logits (SD = 1.10) with a mean of -0.03 logit. The range of item measures covered 93.3% of the person measures, and all person measures, except one, were within one logit of an item measure. This provides evidence that Teach-CVI is relatively well targeted to our CVI patient sample.

TABLE 2 Demographics and clinical information.

	Mean (SD)
Age	9.18 years (6.07)
Visual Acuity	0.46 logMAR (0.40)
Contrast Sensitivity	1.57 logCS (0.46)
	N (%)
Female	54 (45.38%)
Male	65 (54.62%)
Strabismus	83 (69.75%)
Visual field defect*	30 (25.21%)
Teach-CVI Level 1	21 (17.65%)
Teach-CVI Level 2	51 (42.86%)
Teach-CVI Level 3	47 (39.50%)
Visual impairment**	44 (36.97%)
Physical impairment**	7 (5.88%)
Multiply impaired**	57 (47.90%)
CVI only**	11 (9.24%)

*Presence of any visual field loss.

**Visual impairment refers to diagnosis of CVI and another visual diagnosis only, physical impairment for CVI and a physical diagnosis, multiply impaired if the diagnoses included CVI, visual, and physical impairments.

Figure 2 illustrates the infits for item measures and person measures. Item measure infits ranged from 0.60 to 1.85 with a mean (SD) of 1.03 (0.28). Person measure infits ranged from 0.20 to 2.24 with a mean (SD) of 1.02 (0.34). Ideally, the mean infit is 1. Approximately 95% of item measure infits and 90% of person measure infits fell within the desired range of 0.5-1.5 (34) which are indicated by the vertical dashed lines. The person and item infit distributions provide evidence that Teach-CVI measures a single underlying construct (i.e., the instrument is unidimensional).

Figure 3 illustrates the relationship between person measures (y-axis) and visual acuity (x-axis).

The correlation was r = 0.011, and the equation of the best fitting line is $y = 0.3x - 0.18$ (the slope of 0.3 was not significant at p = 0.24). In other words, there is no evidence of a significant relationship between Teach-CVI-person measures and visual acuity in our sample.

Table 3 shows the results of multilinear regression to determine the influence of other demographic, visual, and neurologic variables on Teach-CVI person measures. The reference groups were: CVI only for impairment category, and history of gestational or perinatal inflammation (including PVL, HIE, hypoxia, meningitis, hydrocephaly) for neurologic history. None of the included

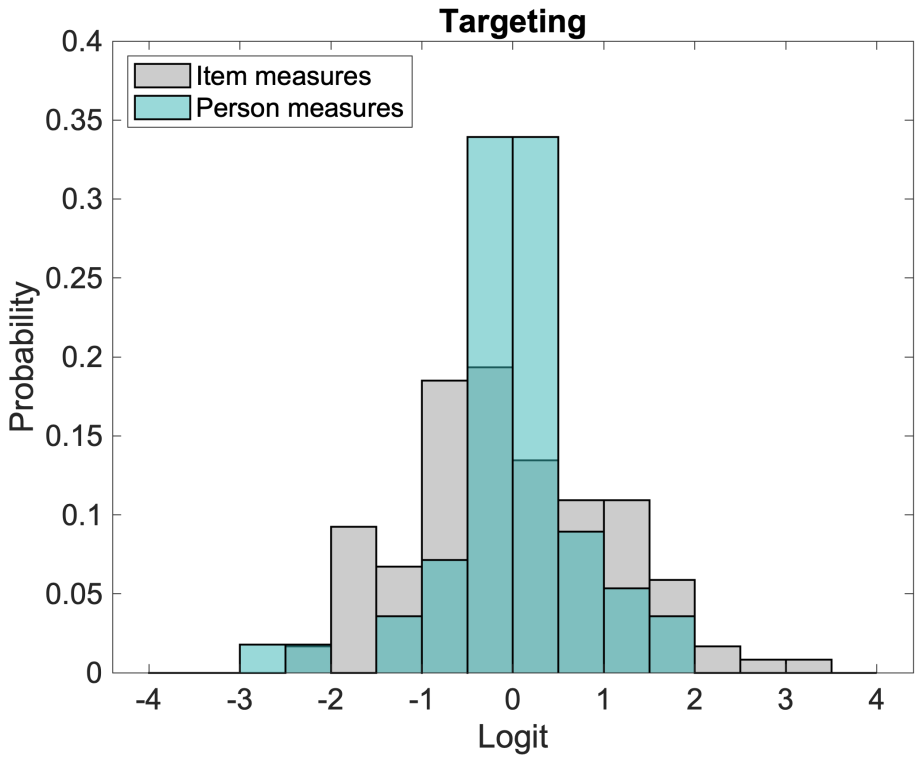

FIGURE 1
Comparison of the distribution of person measures (teal bars) and item measures (grey bars).

variables were found to be significantly associated with the Teach-CVI person measures.

Discussion

This study is the first to explore the psychometric properties of the Teach-CVI instrument through the application of Rasch analysis, offering a novel approach to evaluating visual ability in patients with Cerebral Visual Impairment (CVI). Our findings demonstrate that Teach-CVI is well-targeted to the CVI patient sample, as evidenced by the alignment of estimated person and item measures. The infit statistics provide evidence for unidimensionality of the instrument, which is critical for ensuring that Teach-CVI accurately measures a single construct: the impact of CVI on visual ability.

As anticipated, primary visual cortex (V1) measures of visual function—visual acuity, visual field, and contrast sensitivity—were

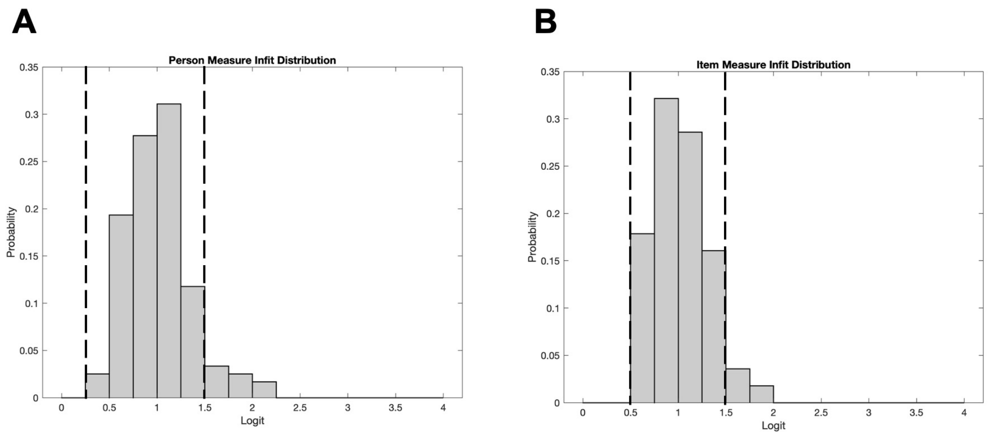

FIGURE 2
(A) Distribution of item measure infits. **(B)** Distribution of person measure infits. Unidimensionality is demonstrated when the data largely falls between 0.5 and 1.5 (indicated by the vertical dashed lines).

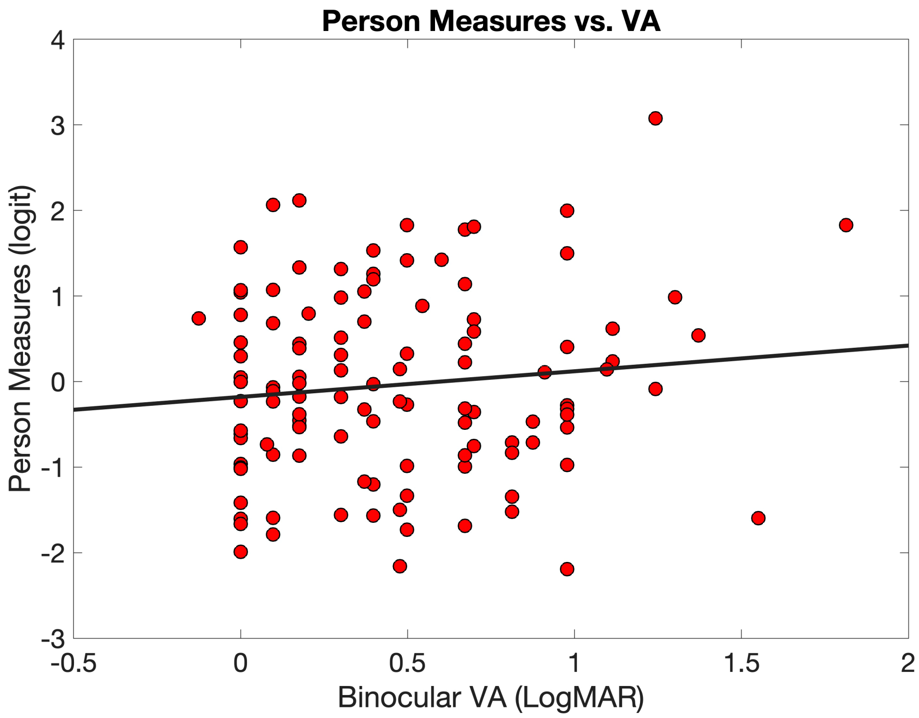

FIGURE 3
Person measures (red circles) plotted against logMAR binocular visual acuity (VA). The equation for the linear regression line (black line) is $y = 0.3x - 0.18$.

not statistically significant predictors of Teach-CVI person measures. This finding is consistent with clinical observations that patients with CVI can often retain good visual acuity but experience profound functional difficulties (5, 6, 36, 37). The disconnect between visual acuity and visual function in CVI has been documented in clinical practice (38, 39), but our analysis provides a more definitive demonstration of this phenomenon. Although visual field defects may be related to occipital lobe damage and therefore can be attributed to CVI, our sample of patients included both ocular based and neurologically based field loss, and therefore was included as a variable to explore whether person measures may correlate with visual field loss independently. While some patients

TABLE 3 Multilinear regression of potential associated factors with Teach-CVI person measures.

	Estimate	Standard Error	t value	Pr(>|t|)
(Intercept)	-0.04	0.56	0.07	0.94
Age	0.01	0.02	0.65	0.51
Contrast Sensitivity (logCS)	-0.17	0.25	-0.68	0.50
Gender: Male	-0.21	0.21	-0.99	0.33
Impairment Category: Visual Impairment	0.40	0.43	0.93	0.35
Impairment Category: Physical Impairment	-0.58	0.57	-1.02	0.31
Impairment Category: Multiply Impaired	0.38	0.45	0.85	0.40
Visual Field Loss	0.09	0.25	0.35	0.73
Presence of strabismus	-0.05	0.28	0.17	0.87
Neurologic History: Genetic/Metabolic	0.30	0.46	0.65	0.52
Neurologic history: Traumatic	0.39	0.67	0.58	0.57
Neurologic history: Other (Prematurity, Epilepsy/seizure disorder)	-0.03	0.25	-0.13	0.90

in our sample presented with 20/20 (0.0 logMAR) acuity and full visual fields, they still reported significant functional impairments, underscoring the importance of tools like Teach-CVI in capturing the true extent of CVI's impact on visual ability. This absence of a relationship provides support for the construct validity of Teach-CVI, as CVI often presents with preserved visual acuity despite significant visual processing deficits. Further, neither neurologic history nor other additional impairments (e.g., physical/motor) were significant, further underscoring CVI's ability to have profound impacts on the individual's functional use of vision regardless of etiology or co-occurring diagnoses.

Other CVI instruments like the Top 11 (6), CVI Questionnaire (40), The Five Questions (10), Dutton CVI Inventory (7), and Paediatric Evaluation of Disability Inventory, Dutch version (41) have been employed for screening and diagnostic purposes but have predominantly relied on raw scores for analyses. Raw score-based methods can be skewed by missing data – one person could respond to only "easy" items, while another could respond to only the "difficult" items, and obtain the same mean raw score. Rasch analysis, in contrast, is robust against missing data and offers more precise person measures. Missing data or "not applicable" responses to any person-item combinations will reduce the precision of the estimated person measure in Rasch analysis, but not the measure itself, with large enough sample size. This property of Rasch analysis is particularly important in the context of CVI, where the heterogeneity of patient experiences may lead to different response patterns.

Previous studies have used methods such as exploratory factor analysis (10, 40) to assess dimensionality, ROC sensitivity analysis to explore sensitivity and specificity for detecting CVI (9) and Cronbach's alpha (10, 41) to assess internal consistency of CVI instruments. However, none of these approaches enable person and item measures to be estimated on the same equal interval scale. Cronbach's alpha in particular is often misinterpreted and misused as a measure of internal consistency (42, 43).

Rasch analysis has numerous applications beyond psychometric validation. It can be used to compare person measures pre- and post-intervention, track longitudinal changes, and perform cross-cultural validation by comparing item measures across different populations. The ability to use the same set of (calibrated) item measures for different studies also enables person measures from different samples to be compared on the same measurement scale. Given its broad utility and widespread adoption in other healthcare sectors (44–51), it is strongly recommended that Rasch analysis be applied to other CVI questionnaires to thoroughly explore their psychometric properties, including targeting.

The Teach-CVI survey, with its flexible, individualized approach, is particularly well-suited for assessing patients across a spectrum of CVI severity. Although originally designed as a screening tool, our study employed Teach-CVI as a guided history, allowing for a deeper understanding of each patient's visual ability. However, there are several limitations, most notably that this was a retrospective study with a sample of patients from a single tertiary clinic. Future studies should aim to include a larger sample from multiple clinics. Additionally, approaches to clinical testing of acuity, contrast, and other measures differed between subjects. However, this was necessary and done to better match the ability of the individual and provide the most accurate measure of their functional vision.

MRI data, which is often used in conjunction with clinical findings and observations to make a diagnosis of CVI, was not available for all patients. This is not unusual given the systemic comorbidities in this patient population. This study also did not further delineate between intellectual and developmental disabilities as this information was not always available to the practitioner. Future studies could implement additional clinical tests to more accurately determine developmental age and level of intellectual impairment or developmental delay, such that the impact of intellectual disability on Teach-CVI person measures could be further explored.

Conclusion

This study represents an important step forward in validating the Teach-CVI screening tool for use as a patient reported outcome measure in patients with CVI. Our results confirm that Teach-CVI is a psychometrically valid instrument that is well-targeted to a sample of CVI patients, and capable of measuring functional vision independently of visual acuity. The application of Rasch analysis has allowed for a more nuanced understanding of the instrument's performance, offering a significant improvement over the traditional raw score-based approach.

While our study is limited by its retrospective nature, small sample size, and variability in clinical testing approaches, it nonetheless introduces a robust method for evaluating CVI-related visual function. Future research should focus on increasing sample size, further refining the Teach-CVI tool, and applying Rasch analysis to other CVI questionnaires. Additionally, longitudinal studies tracking changes in person measures over time could provide valuable insights into the efficacy of interventions for CVI patients.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Institutional Review Board at the New England College of Optometry. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

JM: Writing – original draft, Writing – review & editing, Data curation, Formal analysis, Investigation, Project administration. CB: Writing – original draft, Writing – review & editing, Formal analysis. BK: Writing – original draft, Writing – review & editing. NR: Writing – original draft, Writing – review & editing, Conceptualization, Methodology, Formal analysis, Project administration.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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EDITED BY
Jeff C. Rabin,
University of the Incarnate Word,
United States

REVIEWED BY
Melinda Chang,
Children's Hospital of Los Angeles,
United States
Songül Atasavun Uysal,
Hacettepe University, Türkiye

*CORRESPONDENCE
Marinke J. Hokken
✉ m.hokken@erasmusmc.nl

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Gestalt, Navon and Kanizsa illusion processing in CVI, ADHD, and dyslexia Children with Normal verbal IQ

Marinke J. Hokken[1,2]*, Ymie J. Van Der Zee[2], Rob Rodrigues Pereira[3], Ingrid G. I. J. G. Rours[3], Maarten A. Frens[1], Johannes van der Steen[1], Johan J. M. Pel[1] and Marlou J. G. Kooiker[1,2]

[1]Department of Neuroscience, Erasmus Medical Center, Rotterdam, Netherlands, [2]Royal Dutch Visio, Huizen, Netherlands, [3]Kinderplein Medical Center, Rotterdam, Netherlands

Introduction: Global Visual Selective Attention (VSA) is the ability to integrate multiple visual elements of a scene to achieve visual overview. This is essential for navigating crowded environments and recognizing objects or faces. Clinical pediatric research on global VSA deficits primarily focuses on autism spectrum disorder (ASD). However, in children with cerebral visual impairment (CVI) and other neurodevelopmental disorders (ADHD, dyslexia) similar deficits are reported. The aim of this study was to investigate global VSA performance in children with CVI, ADHD, dyslexia and neurotypical children by combing gaze-based measures with conventional neuropsychological tasks.

Methods: We included children aged 6–12 years with CVI (n = 20), ADHD (n = 30), dyslexia (n = 34) and neurotypical development (n = 37), all with normal verbal IQ. Eye tracking measurements were stepwise introduced within three global VSA tasks: Gestalt Closure (no eye tracking), Navon stimuli (eye tracking-based qualitative analysis) and Kanizsa Illusory Contours (KIC; eye tracking-based quantitative analysis). Verbal responses were compared with non-verbal gaze behavior.

Results: Children with CVI had significantly lower success rates on Gestalt Closure recognition, prolonged verbal response times on Navon stimuli, and decreased verbal and gaze performance on the KIC task compared to all other groups, irrespective of visual acuity. Children with ADHD and dyslexia performed similar to neurotypical children on all tasks.

Discussion: The results suggest a distinct global VSA deficit in children with CVI, which aligns with clinical observations of symptoms in daily life. Incorporating gaze-based analyses provided new information about search strategies beyond verbal answers and made the KIC task more inclusive for children with language and motor disabilities. Assessing global VSA within clinical CVI assessments could improve the differential diagnostic evaluations among children with CVI, ADHD and dyslexia, leading to more personalized treatment approaches.

KEYWORDS

cerebral visual impairment (CVI), attention deficit hyperactivity disorder (ADHD), dyslexia, Kanizsa illusory contours, Navon, Gestalt closure, global visual selective attention

1 Introduction

The visual world around us contains too many elements to process simultaneously. Hence, visual selective attention (VSA) is crucial to avoid visual overload. VSA acts as a spotlight, focusing on specific parts of the visual field for further processing, while other parts remain outside conscious awareness (Posner et al., 1980). According to theories, the size of this 'attentional spotlight' is not fixed, but can be adjusted based on the task at hand. When the attentional spotlight is narrow, known as *local VSA*, individual visual elements are processed in detail. Conversely, when the spotlight is broad, known as *global VSA*, multiple visual elements are processed simultaneously, creating a visual overview that is important for, e.g., segmenting scenes and recognizing objects (Liechty et al., 2003; Förster and Dannenberg, 2010; Förster et al., 2008).

Unlike adults, young children and infants have a local preference as they primarily rely on visual details when exploring a visual scene (Kimchi et al., 2005). The shift to an adult-like global VSA preference typically seems to occur between 5 and 7 years of age (Poirel et al., 2012; Abravanel, 1982; Happé, 1996; Milne and Scope, 2008; Hadad et al., 2010), though some studies suggest it appears earlier (Guy et al., 2013; see for a review Colombo et al., 1991). This developmental shift, however, seems inconsistent in children with neurodevelopmental disorders. These children may retain a local preference, resulting in a fragmented visual processing which hinders their ability to understand larger visual scenes, objects and faces. Lacking a comprehensive view of a visual scene may cause, e.g., difficulties in understanding cause-and-effect relationships and heightened anxiety in crowded scenes such as supermarkets, traffic situations or school yards (Dutton and Jacobson, 2001). Lacking the ability to integrate facial features may lead to misinterpreting social cues and social–emotional difficulties. In children with autism spectrum disorder (ASD), these global VSA deficits have been extensively studied in the context of the weak central coherence theory (Booth and Happé, 2010). Similar daily symptoms have been reported by the parents of children with Cerebral Visual Impairment (CVI), Attention Deficit Hyperacitivy Disorder (ADHD) and dyslexia (Hokken et al., 2024b). Especially in children with CVI, a brain-based visual disorder, global and local VSA deficits are common due to compromised processing within dorsal (i.e., occipital and parietal) cerebral networks (Bennett et al., 2020; Atkinson, 2017). These children often miss important visual details and lack a comprehensive overview over complex scenes, even in the absence of visual acuity or visual field deficits (Atkinson, 2017; Moore and Zirnsak, 2017; Philip and Dutton, 2014; Hokken et al., 2024b). The latter has also been described as simultanagnosia (Chokron and Dutton, 2023). A 12-year-old girl with CVI explained:

> '*I see too many things, too many stimuli around me, and I can't put it all together quickly. My vision is actually like a puzzle. For example, when I see a face, I first see the mouth, and after that little by little, it becomes a whole*' (film fragment adapted from Koninklijke Visio, 2019).

The neuropsychological assessment of CVI, that focusses on a variety of potential impaired visual functions, often includes tasks designed to evaluate global VSA performance. These tasks typically include a global level that must be perceived independently from the local level of the visual scene or object. In other words, the scene or object cannot be easily identified by solely looking at the details one by one, but only when broadening the attentional spotlight to reach visual overview. Examples of well-known conventional tasks are presented in Figure 1. The Kaufman's Gestalt Closure task (Figure 1A) consists of incomplete inkblot pictures (Kaufman and Kaufman, 1983). Navon figures (Figure 1B) consist of global large forms, i.e., letters, that are composed of either congruent or incongruent small local elements (Navon, 1977). Kanizsa illusory contours (KICs;

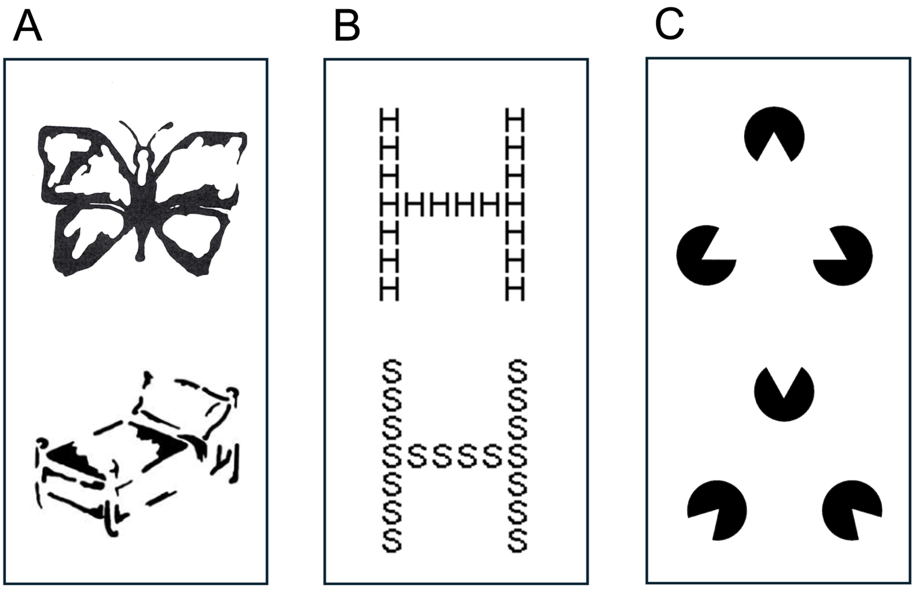

FIGURE 1
(A) Kaufman Gestalt Closure task. **(B)** Navon task: congruent global/local elements (top) and incongruent global/local elements (bottom). **(C)** Kanizsa Illusory Contours: with an illusory contour (top) and without an illusory contour (down).

Figure 1C) consist of local visual elements (Pac-Man) that are placed in such a way that, in target trials, an illusory shape can be distinguished (Kanizsa, 1976).

Although local VSA deficits have been quite extensively reported and analyzed in children with CVI (Bennett et al., 2018; McKillop and Dutton, 2008; Manley et al., 2022; Manley et al., 2023; Zhang et al., 2022; Hokken et al., 2024a), only a handful of studies investigated global VSA. Weaker performance on global VSA tasks was found in children with CVI (van der Zee et al., 2019), and in children with a medical risk of CVI, such as children with Periventricular leukomalacia (PVL; Rešić et al., 2008; Fazzi et al., 2004) and children born very prematurely (<33 weeks of gestation; Datin-Dorrière et al., 2021). Literature on task performance in ADHD and dyslexia is more inconclusive, as a local before global precedence was found for both children with ADHD (Song and Hakoda, 2012; Song and Hakoda, 2015) and children with dyslexia (Franceschini et al., 2017), but other studies did not (Kalff et al., 2002; McAvinue et al., 2012). Taken together, it is challenging to differentiate between CVI, ADHD, and dyslexia in clinical practice based on global VSA task performance.

Another clinical challenge in assessing global VSA is the limitations of the available, conventional tasks. First, many tests are largely observational and lack a standardized method or comprehensive and updated normative data for children. Second, the tests often require a verbal or hand-motor (e.g., mouse click) component, excluding young children and complicating the disentanglement between potential language (naming), motor deficits (pointing) and visual deficits. Third, these tasks result in binary performance measures (normal-abnormal) without insights into potential deviant VSA processes and strategies during task performance. These issues may be overcome by coupling global VSA tasks with eye tracking and analyzing gaze responses during the tasks. These gaze responses are not only non-verbal and non-motor of nature, a proof-of-concept study in children with CVI (van der Zee et al., 2019) also demonstrated how eye tracking can reveal valuable insights about visual attentional processes. Therefore, it is expected that eye tracking may give more detailed insight in global VSA performance and development over age.

The aim of the present study was to investigate global VSA performance in children with CVI, ADHD, dyslexia and neurotypical children by gradually expanding conventional neuropsychological tasks with gaze-based measures. Eye tracking measurements were stepwise introduced within three global VSA tasks: a conventional Gestalt Closure task (no eye tracking), a verbal Navon task (eye tracking-based qualitative analysis) and the non-verbal Kanizsa Illusory Contours task (KIC; eye tracking-based quantitative analysis). In the latter two tasks, verbal responses were compared with non-verbal gaze behavior. We hypothesized that children with CVI performed weaker on all tasks and showed deviant gaze behavior, e.g., slower responses, compared to all other groups.

2 Methods

2.1 Participants

This study took place between January 2021 and August 2022, and included children with CVI, ADHD, dyslexia, and neurotypical children between 6 and 12 years of age were included. Children with CVI were recruited via Royal Dutch Visio, a rehabilitation center for blind and visually impaired people. Demographic, ophthalmic, and neurologic data were extracted from the records. Children with ADHD were recruited via MC Kinderplein and AllesKitz (Dutch mental healthcare centers for children). Parents filled out the Dutch ADHD Questionnaire (ADHD Vragenlijst: AVL) to cluster their daily symptoms (Scholte and Van der Ploeg, 2005). Children with dyslexia were recruited via RID (regional institute for dyslexia). All clinical diagnoses were based on official guidelines and confirmed by experienced health care professionals prior to study inclusion. We excluded children with a combination of the above diagnoses from the study. Neurotypical children, serving as the control group, were selected through siblings of participants with CVI and colleagues of Royal Dutch Visio and Erasmus MC. Exclusion criteria were a verbal IQ, or comprehension index (VCI, from WISC-V), below 70, ASD diagnosis, visual acuity below 0.1 decimal (Snellen equivalent) or a visual field less than 30 degrees. Written informed consent was obtained by parents or caregivers before commencing the study. The study was approved by the Medical Ethical Committee of Erasmus MC Rotterdam (MEC-2020-0680) and complied with the tenets of the Declaration of Helsinki (2013) for research involving human subjects.

2.2 Procedure and experimental set-up

Children underwent a 45- to 60-min assessment in a noise-free room. The assessment consisted of conventional neuropsychological tasks and novel gaze-based tasks, assessed in a fixed order, with breaks provided between tasks when needed, as part of a larger study into visual selective attention processes (EEVA–Erasmus Eye tracking Visual Assessment). A subset focusing on global VSA was included in the present study.

2.2.1 Conventional neuropsychological tasks

2.2.1.1 WISC-V verbal comprehension index

The child's verbal intelligence level was measured using the Similarities and Vocabulary subtests from the Dutch adaptation of the Wechsler Intelligence Scale for Children (WISC-V-NL; Wechsler, 2017). The combined score from these subtests was converted into an age-based norm score that determines the Verbal Comprehension Index (VCI).

2.2.1.2 Gestalt closure task

The Gestalt Closure Task is part of the Kaufman Assessment Battery for Children (K-ABC; Kaufman and Kaufman, 1983). The test has been developed for children between three and 18 years of age and has good reliability ($r = 0.92$). The participants were asked to identify 37 incomplete inkblot images of everyday objects and animals printed on paper. When the participant made four consecutive mistakes, the task was ended. Raw scores of the total number of correctly recognized images were converted into age-based norm scores, with a range of 1 to 19.

2.2.2 Gaze-based tasks

Children were placed in front of a 22.5-inch screen (1920 × 1,080 or 1920 × 1,200 resolution) with a viewing distance of approximately 60 cm. A remote eye tracker was attached to the bottom of the screen to record their eye movements (Tobii Pro X3-120). The eye tracker registered the movement of the eyes at 120 Hz and automatically compensated for movements of the head. A standardized five-point calibration was carried out prior to every task. The children were instructed to minimize movement during the tasks. If they moved, the researcher reminded them to remain still and repositioned them correctly. In case of significant movements, the researcher recalibrated the eye-tracker. Gaze data were processed using Tobii Pro Lab Software and Tobii I-VT fixation filter with a velocity threshold of less than 30 degrees per second to classify fixations and higher than 30 degrees per second to classify saccades.

2.2.2.1 Navon number task

The Navon Number Task (see Figure 2) is a variation on the classic Navon Task (Navon, 1977). Each display showed a large number, i.e., the global level, that was composed of small numbers, i.e., the local level. The task was divided into three sets, all consisting of one practice display and three test displays. In the first set, the participants were given the verbal instruction to name the number that they saw first as quickly as possible (first response set). For the next sets, participants were instructed to name, respectively, the small number (local set) or the large number (global set) as quickly as possible.

Verbal Response Time (VRT) was recorded by the researcher pressing the spacebar directly after the verbal response of the child.

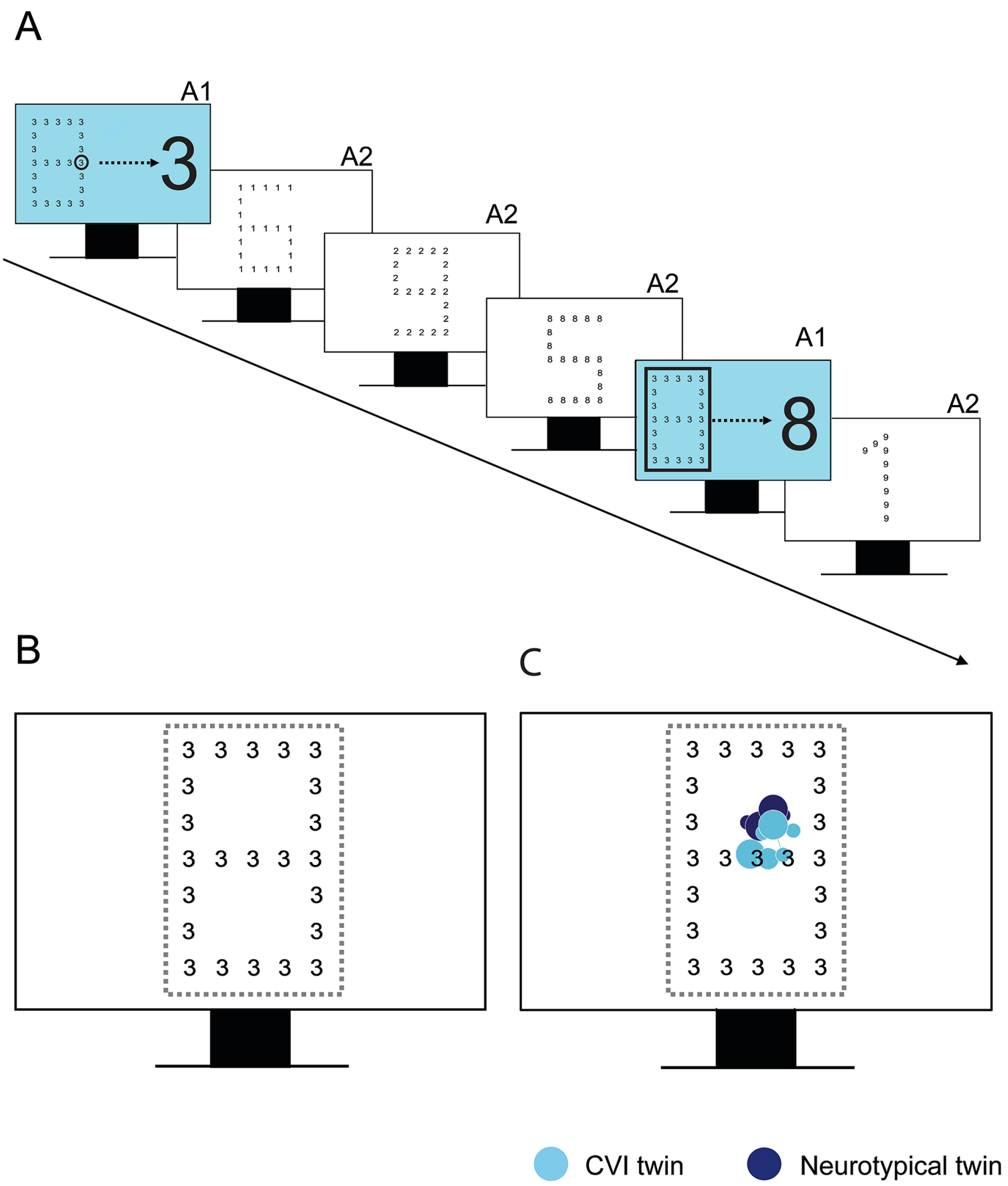

FIGURE 2
(A) Experimental design of the Navon Number Task. For the global and local set, it is explained in practice trials (A1) what level they must report, followed by three test trials (A2). **(B)** Area of interest (AOI) around the Navon stimulus, depicted in yellow. **(C)** Example of gaze behavior in one set of participating twins (12 years of age), one with CVI and one with neurotypical development.

The gaze data were used qualitatively, to check the overall attention for the Navon stimuli. We also analyzed the Time to the First Fixation on the AOI, to investigate whether potential delays in VRT were due to slower fixation on the stimulus or processing deficits. Further analysis of saccades and fixations was not included because of the task set up, in which fixation prior to target presentation was not controlled.

2.2.2.2 Kanizsa illusory contours task

Figure 3 illustrates the Kanizsa Illusory Contour (KIC) Task, inspired by Kanizsa (Kanizsa, 1976). A total of 10 displays included 48 black or black-and-white three-quarter circles (Pac-Man) stimuli in various rotations. In eight target displays, four Pac-Man were arranged to form an illusory square, positioned in one of the four corners of the display. The other Pac-Man stimuli in the display were placed randomly (no illusory square). There were two non-target trials in which no illusory square was presented. No verbal or motor responses were required during the task. Participants did not receive any explicit verbal instructions, except for the request to look at the screen. After calibration, a fixation smiley first appeared in the middle of the screen for 1.5 s. Then, the Illusory Form displays appeared for 5 s. After all displays were shown, three questions were asked to check whether the child perceived the illusory target: (1)

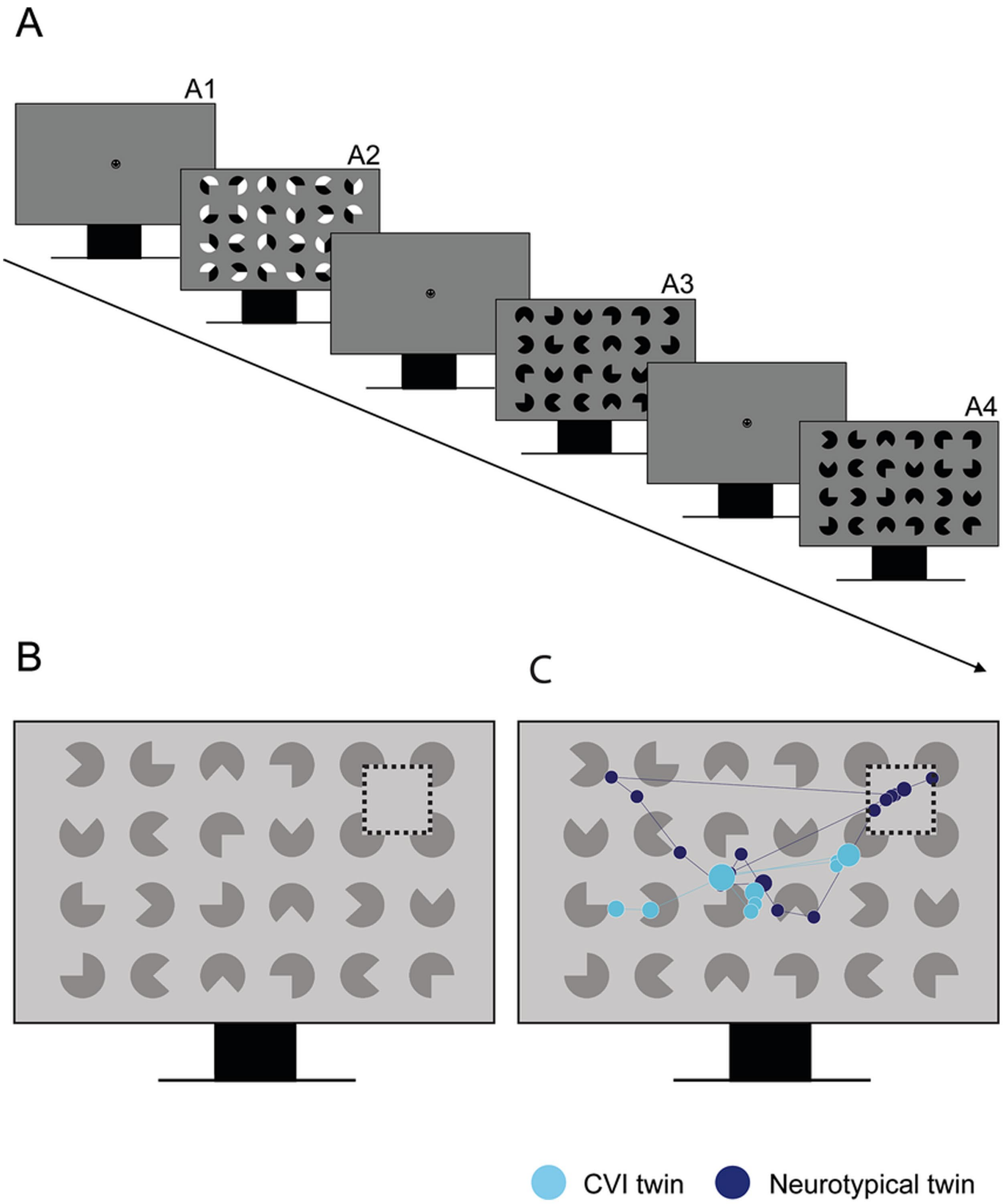

FIGURE 3
(A) Experimental design of the Kanizsa Illusory Contour task. The fixation smiley appears for 1.5 s (A1). The displays consist of black (A3, A4) or black-and-white (A2) Pac-Man stimuli with (A2, A4) or without (A3) an illusory square target. **(B)** Area of interest (AOI) around the Navon stimulus. **(C)** Example of gaze behavior in one set of participating twins (12 years of age), one with CVI and one with neurotypical development.

What did you see? (2) What shape did you see? (3) Did you see a square?

For gaze data analyses, an area of interest (AOI) was created surrounding the four Pac-Man forming the illusory square. Outcome measures were Target Accuracy, i.e., whether the child fixated on the AOI, Time to First Fixation on the AOI, and First Fixation Duration on the AOI. In addition, the Visual Search Area, i.e., the percentage of the total display that the child fixated on, was computed by drawing a circle with a radius of 2.5 degree around each detected fixation and marking all pixels within this circle as visited.

2.3 Statistical analysis

Statistical analyses were carried out using SPSS Statistics package version 29.0.0. After log-transforming all time-based parameters had a normal distribution. The primary analyses focused on comparing all outcome measures per task between all groups, and on comparing the effect of task conditions within groups. An ANOVA was performed for the Gestalt Closure task. We compared the proportions of correct responses on the KIC Task with a chi-squared test. A MANCOVA was conducted for performance on the Navon Number Task and KIC Task, with age as covariate. Additionally, within-group differences were evaluated on task demands using separate paired sample t-tests per group. For all analysis, the alpha threshold was set at 0.05. Effect sizes were quantified using eta squared, with interpretations categorized as small ($d = 0.2$), medium ($d = 0.5$), or large ($d = 0.8$), following Cohen's guidelines (1988).

3 Results

3.1 Group characteristics

In total, 121 school-aged children (6–12 years of age) participated in the study. The groups did not differ in sex ($\chi^2 = 2.17$, $p = 0.55$) and VCI ($F = 1.07$, $p = 0.36$). There was an overall group difference in age ($F = 3.53$, $p = 0.02$, $\eta^2 = 0.08$), but post-hoc tests revealed no significant age differences between the separate groups (all p's > 0.05; Table 1).

Within the CVI group, 11 children (55%) were born prematurely (< 36 weeks). Other associated, not mutually exclusive, neurologic findings were pregnancy or birth complications ($n = 11$, 55%), Cerebral Palsy ($n = 4$, 2%), brain structural abnormality ($n = 4$, 2%), epilepsy (n = 4, 2%), opticus glioma ($n = 1$, 5%), Traumatic Brain Injury ($n = 1$, 5%), Hydrocephalus ($n = 1$, 5%), genetic disorder ($n = 1$, 5%). Two children with CVI (10%) had a reduced visual acuity, i.e., between 0.1 and 0.3 decimal. For three children with CVI (24.5%) the visual acuity was suboptimal: between 0.4 and 0.6 decimal. Visual acuity levels were considered in all three tasks. One child (5%) had hemianopia, and one child (5%) had nystagmus. Strabismus was found in seven children (35%), of which two children had exotropia and five had esotropia.

TABLE 1 Demographic characteristics of the children by group: age, sex and verbal comprehension index of the WISC-V-NL.

	CVI	ADHD	Dyslexia	Neurotypical
N	20	34	30	37
Age	9.5 ± 2.3	10.4 ± 1.4	10.4 ± 1.9	9.4 ± 1.8
Girls (%)	9 (45%)	12 (35%)	16 (53%)	17 (46%)
VCI	96	97	100	104

Within the ADHD group, 25 children (74%) had a (sub) clinical score for Attention deficits, 22 children (65%) for Hyperactivity symptoms and 22 children (65%) for Impulsivity symptoms. Thirteen children with ADHD (38%) took medication prior to testing, 21 children did not. Differences between children with ADHD with and without medication were considered for all three tasks.

3.2 Gestalt closure

Figure 4 shows the Gestalt Closure norm scores per group. Gestalt Closure task performance significantly differed between the groups ($F = 8.04$, $p < 0.001$, $\eta^2 = 0.17$). More specific, children with CVI had significantly lower norm scores on the Gestalt Closure task compared to neurotypical children, children with ADHD and children with dyslexia (all p's < 0.001). There was no difference between neurotypical children, children with ADHD and children with dyslexia (all p's = 1.00).

Within the CVI group, there was no correlation between visual acuity and performance on the Gestalt Closure task ($r = 0.41$, $p = 0.07$). Within the ADHD group, medication did not have a significant effect on performance ($F = 0.00$, $p = 0.99$).

3.3 Navon number task

In Figure 5, the results of the Navon Number Task are shown per group. Age was a significant covariate for Verbal Response Time (VRT) on the first response set ($F = 10.58$, $p = 0.005$, $\eta^2 = 0.41$) and the local set ($F = 13.74$, $p = 0.002$, $\eta^2 = 48$), but not on the global set ($F = 4.41$, $p = 0.05$). Age was not a significant covariate for Time to First Fixation for all three sets (all p's > 0.05; Figure 5B).

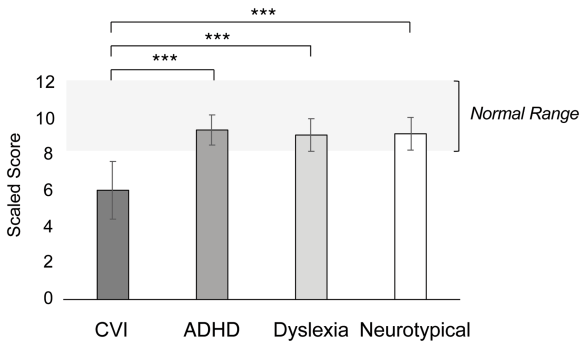

FIGURE 4
Performance on the Gestalt Closures task, in mean scaled scores with error bars indicating ±1 Standard Error (SE). The light gray bar between the scaled score of 8 and 12 refers to the normal, or average, range. ***$p < 0.001$, **$p < 0.010$, *$p < 0.05$.

After controlling for age, there was no group difference in VRT for the first response set ($F = 1.30$, $p = 0.31$), but the groups differed in VRT on the local set ($F = 23.67$, $p < 0.001$, $\eta^2 = 0.83$) and the global set ($F = 7.71$, $p = 0.002$, $\eta^2 = 0.61$): children with CVI responded significantly slower in the local and global set compared to children with ADHD (local set: $p < 0.001$; global set: $p = 0.002$), children with dyslexia (local set: $p < 0.001$; global set: $p < 0.016$) and neurotypical children (local set: $p < 0.001$, global set: $p = 0.008$).

No differences were found between the other groups (all *p*'s > 0.09; Figure 5A). The Time to First Fixation did not significantly differ between the groups for the first response set ($F = 0.77$, $p = 0.53$), local set ($F = 2.04$, $p = 0.15$) and the global set ($F = 0.29$, $p = 0.83$).

For children with CVI, no correlation was found between visual acuity and VRT on the Navon Number Task ($r = -0.29$, $p = 0.24$). Children with ADHD with or without the use of medication did not differ in performance on the Navon Number Task (all *p*'s > 0.05).

3.4 Kanizsa illusory contour task

Figure 6 shows the percentage of correct verbal answers of the children, after the KIC task was completed. These answers did not significant differ between the groups after the first question ('What did you see?': $\chi^2 = 5.23$, $p = 0.16$). However, the groups significantly differed in their response on the second question ('What shape did you see?': $\chi^2 = 10.05$, $p = 0.02$) and the third question ('Did you see a square?': $\chi^2 = 27.71$, $p < 0.001$). Compared to all other groups, a significant larger proportion of children with CVI did not report the square on question 2 (all *p*'s < 0.05) and 3 (all *p*'s < 0.001). No significant differences in verbal responses were found between the other groups (all *p*'s > 0.05).

Figure 7 shows the gaze behavior of the groups on the KIC Task. Age was a significant covariate for Time to First Fixation ($F = 5.03$, $p = 0.03$, $\eta^2 = 0.05$), but not for the other parameters (all *p*'s > 0.05).

After controlling for age, a significant group difference was found for Target Accuracy ($F = 5.72$, $p = 0.006$, $\eta^2 = 0.19$), Time to First Fixation ($F = 3.08$, $p = 0.03$, $\eta^2 = 0.09$) and Visual Search Area ($F = 3.77$, $p = 0.01$, $\eta^2 = 0.10$). Children with CVI fixated significantly less often in the target area than children with ADHD ($p < 0.001$), children with dyslexia ($p = 0.005$) and neurotypical children ($p = 0.01$). There was no difference between children with ADHD, dyslexia and neurotypical children (all *p*'s > 0.85; Figure 7A). Children with CVI were significantly slower in the Time to First Fixation than neurotypical children ($p = 0.04$). No differences between the other groups were found (all *p*'s > 0.18; Figure 7B). Children with CVI had a significantly smaller Visual Search Area than children with ADHD ($p = 0.02$), children with dyslexia ($p = 0.03$) and neurotypical children ($p = 0.02$). No differences were found between children with ADHD, dyslexia and neurotypical children (all *p*'s = >0.05; Figure 7D). The groups did not show significant differences in Target Fixation Durations ($F = 0.59$, $p = 0.62$; Figure 7C).

Within the CVI group, there was no correlation between visual acuity and Target Fixations ($r = -0.181$, $p = 0.446$). Within the ADHD group, medication did not have a significant effect on Target Fixations? ($F = 2.05$, $p = 0.162$).

Figure 8 shows the within-group differences between the different KIC conditions. All groups made significantly fewer target fixations (all *p*'s < 0.001) in the black and white condition compared to the black condition (Figure 8A). Children with CVI had significantly faster Time to First Target Fixation in the black conditions compared to the black-and-white condition ($p = 0.04$) but no differences were found within the other groups (all *p*'s > 0.05; Figure 8B). Within all groups there were no differences between black and black-and-white stimuli in Target Fixation Durations (Figure 7C) and Visual Search Area (Figure 8D; all *p*'s > 0.05).

Children with ADHD, dyslexia and neurotypical children had significantly larger Visual Search Areas in the non-target conditions compared to the target conditions (all *p*'s < 0.05). However, in children

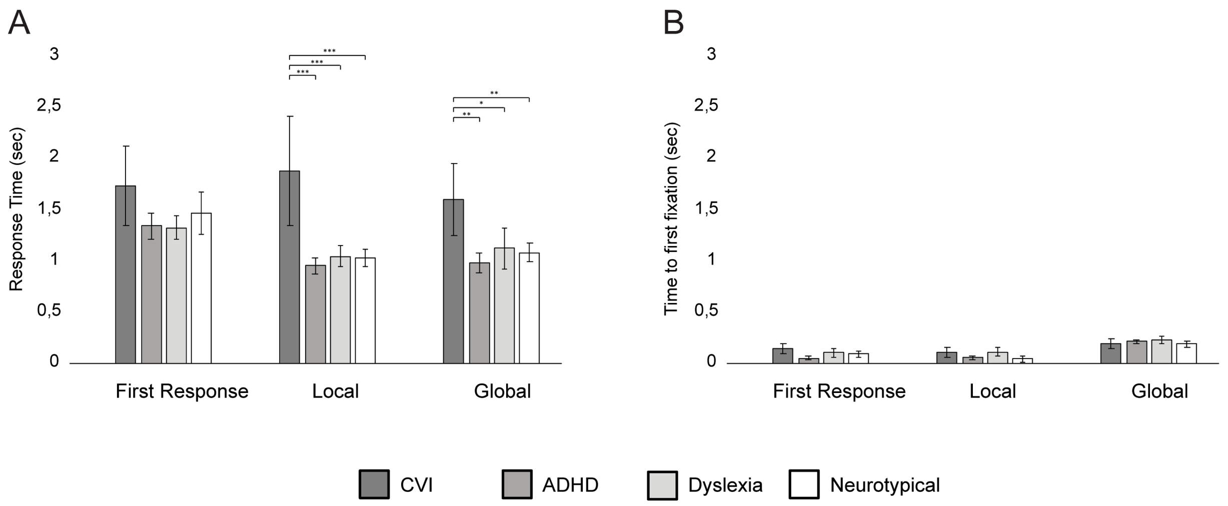

FIGURE 5
Group differences in mean Verbal Response Times **(A)** and Time to First Fixation **(B)** on the Navon Number Task per set, with error bars indicating ±1 Standard Error (SE). ***$p < 0.001$, **$p < 0.010$, *$p < 0.05$.

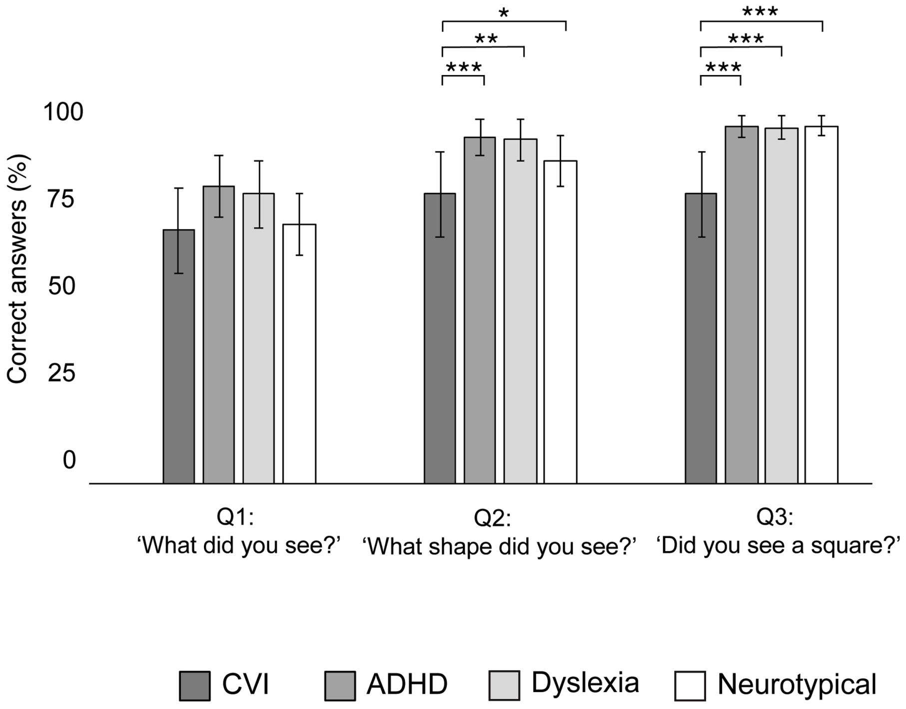

FIGURE 6
Group proportions of mean correct verbal answers (naming the illusory square) after the Kanizsa Illusory Contour Task, with error bars indicating ±1 Standard Error (SE). ***$p < 0.001$, **$p < 0.010$, *$p < 0.05$.

with CVI there was no difference in Visual Search Area between the target and non-target condition ($p = 0.87$; Figure 8E).

4 Discussion

This study examined the global visual selective attention (VSA) deficits that are often reported in the daily lives of children with CVI, but are rarely addressed in scientific research. By comparing children with CVI with their ADHD, dyslexia and neurotypical peers, we found that children with CVI had significant problems with identifying Gestalt Closure pictures, Navon stimuli and Kanizsa Illusory Contours. The gaze-based analyses on two tasks revealed both qualitative and quantitative differences in viewing patterns in children with CVI. In line with previous studies, the impaired performance could not be attributed to visual acuity deficits (Zhang et al., 2022; Manley et al., 2022; Hokken et al., 2024b), suggesting higher order global VSA deficits, potentially due to a dorsal stream vulnerability. More specific, these findings align with the hypothesis that children with CVI have difficulty integrating multiple visual elements into a cohesive whole, which might explain the lack of visual overview they experience in daily life (Chokron and Dutton, 2023; Hokken et al., 2024b).

4.1 Global VSA performance and implications for children with CVI

Although significant group differences were found on all three tasks, each task showed unique insights, benefits and limitations for its use in clinical neuropsychological assessments of children with potential global VSA impairments, such as children with CVI.

The results of the Gestalt Closure task, performed on paper, demonstrated discriminative power: Children with CVI consistently performed below the normative, age-based threshold, unlike children with ADHD, dyslexia, or neurotypical development. As was known up front, this task provides limited insight into the underlying processes and strategies beyond the verbal answer. Therefore, it remains unclear whether children with CVI struggled with naming incomplete figures due to a global VSA deficit, or a broader visual identification deficit. Although such isolated visual identification deficits are not commonly reported in children with CVI (Dutton, 2011; Hokken et al., 2024b), and are unlikely given the participants' average verbal IQ, it is recommended that clinicians compare the results on the Gestalt Closure task with naming tasks that include complete pictures, to ensure correct interpretation of the results.

The results of the Navon Number task, performed on a computer screen with concurrent gaze recordings, showed that children with CVI responded slower to global and local Navon stimuli, but only when the level of analysis (global or local) was prescribed. Qualitative gaze analysis suggested that this delay occurs between stimulus fixation and response, indicating deficits in processing the image or formulating a verbal response. However, further analysis of the cause of this delay, and why this delay is absent when they are free viewing a Navon stimulus, is constrained by the task set-up (see Limitations). Taken together, the Navon Number task requires a refinement of its experimental design before it is a reliable test for clinical practice. Nevertheless, including a Navon task in CVI assessments as an observational tool remains promising, given that it gives valuable

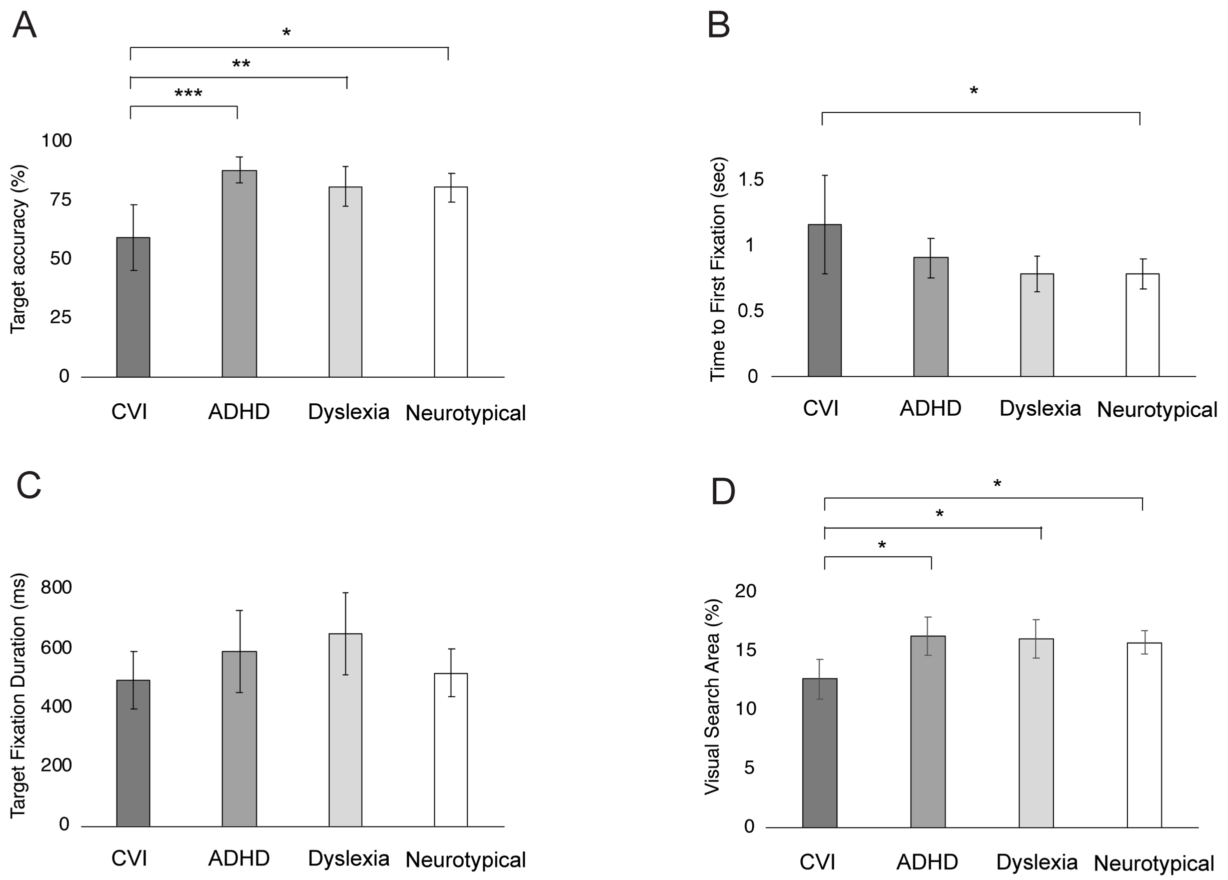

FIGURE 7
Group differences in gaze behavior on the Kanizsa Illusory Contour Task. Bars represent the mean Target Accuracy **(A)**, Time to First Fixation **(B)**, Target Fixation Duration **(C)**, and Visual Search Area **(D)**, with error bars indicating ±1 Standard Error (SE). $^{***}p < 0.001$, $^{**}p < 0.010$, $^{*}p < 0.05$.

insights into whether a child can perceive and switch between local and global levels.

The results of the KIC task, performed on a computer screen with concurrent and detailed gaze recordings, revealed that children with CVI perceived the illusory target less frequently than their peers. Although the first question after the task ('What did you see?') might have been too broad to detect group differences, fewer children with CVI recognized or confirmed the presence of a square in their response to the more specific second ('What shape did you see?') and third ('Did you see a square?') question. These verbal responses align with the measured gaze behavior as they looked less often at the illusory target and were significantly slower in fixating on the black-and-white Pac-Man, indicating greater difficulty with increasing task demands (Bennett et al., 2018; Manley et al., 2022; Zhang et al., 2022; McDowell and Butler, 2023; Hokken et al., 2024a). Drawing from the global VSA theory, children with CVI may perceive the display as a collection of separate elements (Pac-Man elements) rather than a unified illusory shape (Field et al., 1993). As a result, they may have been unaware or uncertain about what to look for and whether it has been found. This could explain the smaller search areas and the lack of expansion of their search when the target was absent. It may also explain the discrepancy with previous studies, which found larger search areas in children with CVI when tasked with finding small details among distractors (Bennett et al., 2018; Manley et al., 2022; Zhang et al., 2022; Hokken et al., 2024a). In those tasks, children with CVI were instructed to search for a specific target but struggled to locate it, causing them to search a larger area of the display. In sum, the KIC Task is promising for clinical CVI assessments, as it has the advantage of enabling analysis of underlying viewing patterns, beyond binary performance outcomes. Additionally, its design, which requires no initial verbal instructions or verbal and motor responses, is particularly inclusive for children with language or motor delays, or for younger children.

4.2 Global VSA performance and implications for children with ADHD and dyslexia

Children with ADHD and dyslexia did not differ from their neurotypical peers in verbal and gaze responses across all three tasks, suggesting an absence of global VSA deficits on a group level. This finding contributes to the ongoing debate within the literature. Although a lack of visual overview in everyday settings was reported in children with ADHD and dyslexia compared to neurotypical

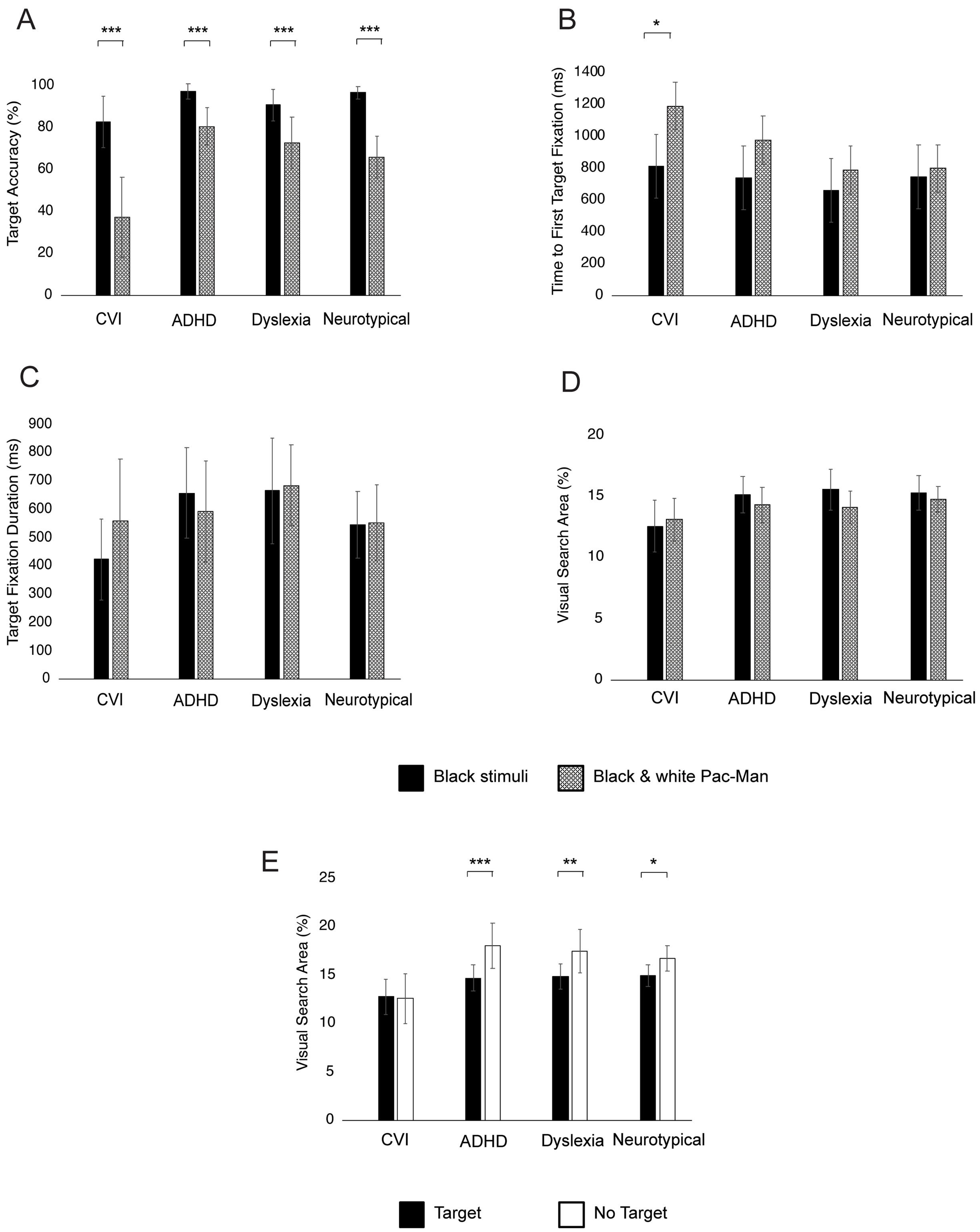

FIGURE 8
Within-group differences between black and black and white Pac-Mans **(A–D)** and between target and no target conditions **(E)**. Bars represent the mean Target Accuracy **(A)**, Time to First Fixation **(B)**, Target Fixation Duration **(C)**, and Visual Search Area **(D,E)**, with error bars indicating ±1 Standard Error (SE). ***$p < 0.001$, **$p < 0.010$, *$p < 0.05$.

children, their visual problem scores were significantly lower compared to children with CVI (Hokken et al., 2024b). Thus, whereas VSA deficits are considered a core deficit in children with CVI, this is not the case for children with ADHD or dyslexia. Within ADHD, attentional inhibition, i.e., difficulty in suppressing attention or responses to stimuli, is a core deficit. Attentional inhibition can lead to difficulties in maintaining overview in dynamic environments (American Psychiatric Association, 2013), which can mimic global VSA deficits. Similarly, the core deficit in children with dyslexia lies in phonological processing (Sanfilippo et al., 2020). Reading development generally progresses from a local, letter-by-letter strategy to global whole-word recognition (Ehri, 2005; Perfetti and Hart, 2002). Consequently, the problems children with dyslexia face with text, word and letter overview, could stem from reading delays, rather than global VSA deficits. Therefore, not all children with ADHD and dyslexia may have global VSA deficits, or their deficits are more subtle and not detected by the tasks used in this study. Further research is needed to clarify the presence of global VSA deficits in these groups.

4.3 Study strengths and limitations

This study is one of the first to examine the global VSA performance of children with CVI and to compare their verbal and gaze outcomes with those of children with ADHD, dyslexia, and neurotypical development. Given the novelty of this research area, we included multiple short tasks, and stepwise introduced eye tracking-based analysis. As a result, the tasks used were brief, consisted of a fixed number of trials (Navon Number Task and KIC task), and uncontrolled fixation positioning (Navon Number Task). Coupled with relatively small sample sizes, along with the exclusion of children with comorbid diagnoses, these factors may limit the generalizability of our findings. Additionally, the current design of the Gestalt Closure and Navon tasks poses limitations for analyzing VSA through eye movements, as both global and local elements fall within the same areas of interest (AOI), and the shift between the local and global elements is related to a mental shift of attention without physical eye movement, i.e., covert visual attention (Bisley, 2011). Future studies could consider either adjusting the task design to better capture scanning behaviors or exploring alternative measures. For example, pupillometry could provide valuable insights as studies on Navon tasks demonstrated that the pupil constricts more during the selection of local elements compared to the global form (Sabatino DiCriscio et al., 2018; Troiani and DiCriscio, 2017). This might be a first step toward a non-verbal and non-motor assessment of how children perceive Gestalt Closure and Navon stimuli.

Another limitation is that children with ASD were not included in this study. Children with ASD often exhibit global VSA deficits (Booth and Happé, 2010) and they share overlapping daily symptoms with children with CVI. For example, both groups have difficulty with visual overview, avoid crowded and cluttered environments, and experience emotion and/or face recognition deficits (Chokron et al., 2020; Fazzi et al., 2019; Kovarski et al., 2021; Lam et al., 2010; Bauer et al., 2023). However, although children with CVI also benefit from predictable visual environments and structure (McDowell and Budd, 2018; Little and Dutton, 2015), in clinical practice it is often observed that they may be able to compensate by using their verbal and social skills and adapt their behavior to the social norm. Future research should include children with ASD to explore specific differences in gaze behavior compared to children with CVI, especially during global VSA or face recognition tasks.

5 Conclusion

In conclusion, this study indicates that VSA deficits in children with CVI involve not only difficulties in locating visual details in crowded environments (local VSA deficits) but also challenges in integrating these details into a unified whole (global VSA deficits). Visual acuity did not affect this global performance, which supports the dorsal vulnerability theory of visual attention deficits in children with CVI. These findings highlight the complexity in higher order visual deficits in children with CVI, both in daily life and in clinical assessments. It further emphasizes the need that tailored CVI interventions address their inability to maintain visual overview.

While further research is necessary, this study offers new insights for improving differential-diagnostic assessments of global VSA in clinical practice. The incorporation of eye tracking in one task provided new information of visual strategies beyond traditional performance measures and enhances the inclusiveness for children with language and/or motor disabilities.

Data availability statement

The datasets presented in this article are not readily available because the authors do not have permission to share data. Requests to access the datasets should be directed to m.hokken@erasmusmc.nl.

Ethics statement

The studies involving humans were approved by Medisch Ethische Toetsings Commissie (METC), Erasmus MC. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

MH: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Software, Visualization, Writing – original draft, Writing – review & editing. YZ: Supervision, Writing – review & editing. RP: Resources, Writing – review & editing. IR: Resources, Writing – review & editing. MF: Resources, Supervision, Writing – review & editing. JS: Funding acquisition, Resources, Supervision, Writing – review & editing. JP: Methodology, Resources, Supervision, Writing – review & editing. MK: Funding acquisition, Methodology, Supervision, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Implications of cerebral/cortical visual impairment on life and learning: insights and strategies from lived experiences

Rachel G. Bennett[1]*, Marguerite E. Tibaudo[2], Ellen C. Mazel[3] and Nai Y.[4]

[1]Perkins School for the Blind, CVI Center, Watertown, MA, United States, [2]Perkins School for the Blind, Deafblind Program, Watertown, MA, United States, [3]Perkins School for the Blind, Educational Programs, Watertown, MA, United States, [4]Consultant, San Francisco, CA, United States

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EDITED BY
Corinna M. Bauer,
Massachusetts Eye and Ear Infirmary and Harvard Medical School, United States

REVIEWED BY
Heather Bortfeld,
University of California, Merced, United States
Elizabeth Hartmann,
Lasell University, United States

*CORRESPONDENCE
Rachel G. Bennett
✉ rachel.g.bennett@perkins.org

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1 CVI: definition, prevalence, and impact

Cerebral/cortical visual impairment (CVI) is a type of vision problem caused by issues in the brain, not in the eyes themselves. In CVI, the brain has trouble processing visual information due to dysfunction in the visual pathways, which are responsible for interpreting what we see (Kong et al., 2012; Sakki et al., 2018). CVI impacts both higher-order visual processing (e.g., recognition) and lower-order visual processing (e.g., tracking, field deficits, acuity) (Bennett et al., 2020; Fazzi et al., 2007; Pilling et al., 2023). CVI can occur at any stage of life, congenital or due to injury, from many causes, and has a spectrum of manifestations (Bauer et al., 2023; Oakes et al., 2024). It often coexists with ocular conditions, cerebral palsy, developmental delay, epilepsy, neurodevelopmental and genetic conditions (Boonstra et al., 2022; Bosch et al., 2016; Fazzi et al., 2007; Olson et al., 2021; Pilling et al., 2023; Tinelli et al., 2020). CVI impacts visual function and functional vision, and both must be considered in assessment and diagnosis efforts (Pilling et al., 2023).

CVI is the leading cause of pediatric visual impairment that may affect up to 1 in 30 children in mainstream classrooms—yet it remains underdiagnosed (Kong et al., 2012; Teoh et al., 2023; Williams et al., 2021). A recent US prevalence study found at least 180,000 kids with CVI or likely CVI, but less than 20% have a CVI diagnosis (Perkins School for the Blind & McKinsey and Company, 2023). CVI can manifest in ways similar to autism, ADHD, learning disabilities, or other developmental conditions, and therefore often gets missed (Chokron et al., 2021). Children with CVI are at a higher risk of developmental disorders affecting cognitive, learning, motor, and social functioning, and overall reduced health-related quality of life outcomes (Collart et al., 2024; Chokron et al., 2021).

While every lived experience is different, there are common CVI behaviors and traits (Table 1) and emerging study of strategies that may support some with CVI (reduced clutter, use eyes or ears, or wait time) (Chokron et al., 2021; Pilling, 2023). Centering the lived experiences of people with CVI is paramount to understanding how CVI affects daily life, the diverse ways CVI manifests, and the varied compensatory skills used for access. With impacted visual attention and recognition, individuals with CVI are likely to develop a unique representation of what things are, who people are, and the actions, events, and meanings that bring people and things together (Treisman, 2006). Providers and educators need to develop and implement individualized instruction and accommodations that match how the person with CVI perceives and accesses their world.

TABLE 1 Perkins 16 CVI visual behaviors.

Visual attention
Visual recognition
Impact of clutter and crowding
Sensory integration and impact on vision
Impact of motion
Visual field abilities
Impact of color
Form accessibility
Visual guidance of upper limbs
Visual guidance of lower limbs
Access to people
Impact of light
Response interval
Visual curiosity and distance viewing
Appearance of the eyes
Movement of the eyes

From Mazel et al. (2021). "Understanding the CVI Visual Behaviors." CVINow.org.

2 Lived experiences of individuals with CVI

In the past several years, more people with CVI are sharing their stories, which continues to widen our understanding of what CVI is and how it affects everyday life. The authors, whose combined professional and personal backgrounds include TVI, deafblind specialist, special educator, person with CVI, and parent of a child with CVI, reviewed 29 sources (online articles, interviews, videos, and blogs) where 23 individuals with CVI discussed their lived experiences and the compensatory skills (e.g., tactile, auditory, visual) used to access their world. The guiding questions for the review were: How does CVI affect access to learning, navigation, and social interaction? What are strategies and compensatory skills people with CVI use to access these areas? How does CVI impact the perception of objects, people, landmarks, and events?

Using the framework of the Perkins 16 CVI visual behaviors (Table 1), the authors chose statements that reflected manifestations of these behaviors. From there, the authors grouped statements by primary visual behavior knowing that other visual behaviors play a role. For example: "Can't find my parents in a crowd" is primarily access to people, and also reflects clutter, sensory integration (crowd noise), and difficulty with visual attention and recognition. Lastly, the authors grouped the statements into the themes of CVI's impact on learning, navigation, and socialization. Another theme emerged from these accounts—CVI's toll on mental and physical health.

These accounts are a small sample and by no means fully represent the spectrum of CVI experiences—yet they can provide a window into understanding the diverse ways CVI manifests.

2.1 Learning

Many factors interrupt visual access to learning for individuals with CVI (Chokron et al., 2021; Zhang et al., 2022). A commonly reported issue was that vision is inconsistent or unreliable. People with CVI shared how difficult it can be to interpret objects and how sometimes they can't trust their vision and are never fully certain what an object is—for example, mistaking a "rough, cold, stiff blanket" for a beloved cat (Nai, 2021b). One stated that "using vision steals all cognitive resources," so they can only look without using other senses or use another sense without vision (Bennett, 2022d). A middle schooler with CVI stated that his body needs to be in a supportive position to better access his vision and his biggest challenge is when there is "complex visual information, auditory information, or both" (Seif and Bennett, 2024). An adult with CVI described that when she's fatigued, her visual field reduces, she can't recognize what she was just looking at, and she "goes almost blind" (Andrésdóttir et al., 2021). Others detailed how multicolored items "play tricks" on their eyes, clutter deeply impacts item recognition, or they can "recognize things only if they are highly familiar" (Bennett, 2021, 2022e).

In a recent study using images, CVI participants were less accurate in identifying images, required more processing time, and showed significantly greater visual search areas and number of fixations per image (Manley et al., 2023). Several with CVI shared challenges with another two-dimensional form: print text. For example, "Reading is really tiring," "I can't focus on the words on the page," and "When I read [print] it feels like my eyes are being pulled out from their centers" (Bennett, 2022b; Marquardt, 2023a). Another individual could not perceive distinct letters and had to rely on the tactile memory of a word shape, so after 5–15 min of this process, visual fatigue set in (Lane-Karnas, 2023). One adult with CVI noted that she cannot read sheet music because "there is too much visual clutter on the page" (Andrésdóttir et al., 2021).

Individuals with CVI reported a range of compensatory skills and strategies to access their learning, including using real objects and manipulatives to support concept development, color-coding to follow math equations or multiple pieces of visual information at once, Braille and tactile graphics, audiobooks, and text-to-speech, and reducing visual clutter (Bennett, 2022a, 2023b; Marquardt, 2023b; Nai, 2022; Seif and Bennett, 2024). Many with CVI naturally create their own workarounds and compensatory skills. Professionals working with students with CVI should support and teach compensatory skills that are aligned with the student's sensory channel use (i.e., vision, hearing, tactile) revealed through formal and informal assessments.

2.2 Navigation

Individuals with CVI reported that navigating environments is difficult, unsafe, and often scary. An unfamiliar place can be imperceivable without the support of compensatory skills.

In cluttered, crowded, and busy environments, some individuals with CVI described they "can't see," "can't find my way" and reported feelings of being overwhelmed and highly stressed (Nai, 2021c; Rastogi-Wilson and Bennett, 2021; Bennett, 2022a,b).

Individuals with CVI commented on difficulty perceiving motion, and that moving cars, bikes, and people are "blurry" (Brossart, 2024; Hamilton et al., 2019). Several used cars as examples: "I can't see moving cars," "I'm just seeing blur because they are moving too fast," "I can't tell the speed of the car," and "I first see the door of the car, then the handle, the window, and only after that, I see the entire car" (Bennett, 2022b; Hamilton et al., 2019; Royal Dutch Visio, 2018).

Individuals with CVI reported difficulty perceiving depth and surface changes, which impacts their ability to navigate steps, sidewalks, shadows, and patterned surfaces. One described that "patterned carpets are a nightmare," another revealed that escalators cause "anxiety attacks," and one noted that shadows looked like "holes" or "drop-offs" (Andrésdóttir et al., 2021; Bennett, 2022b; Nai, 2021c).

Individuals with CVI recounted the disruptive effect of temperature, mainly how heat can cause vision to shut down and affect other sensory and body systems. A hot day can cause "zero vision days," reduced visual field, blurred vision, vision to be "white and washed out," feeling "sick, fatigued, and dizzy," and reduced ability to process auditory stimuli (Bennett and Caruso, 2021; Marquardt, 2024). People with CVI encounter temperature problems most likely in uncontrolled outside environments, which may lead to issues with safety and the ability to orient and navigate.

Individuals with CVI reported using various compensatory strategies to support safe navigation, such as previewing unfamiliar places, relying on landmarks to orient, using Google Maps features to plan routes or zoom in on landmarks at a destination, memory of familiar routes, a white cane for tactile detection of obstacles and surfaces changes, a guide dog, and human guide (Andrésdóttir et al., 2021; Bennett, 2022a; Brossart, 2024).

2.3 Socialization

Individuals with CVI have difficulty attending to and recognizing faces and understanding facial expressions (Bauer et al., 2023). A young adult with CVI described they can perceive "everything about the person except their faces," while another stated that when people greet them, "I can't see them clearly" (ABC News Australia, 2022). One individual expressed that it's not just a passive lack of seeing and recognizing faces; they also experience "faces as twisted and contorted," and the only way to listen is to "tune out visually" (Nai, 2021d).

Individuals with CVI have varied experiences around visual social aspects that can cause social breakdowns, anxiety, and isolation. The impact of clutter, crowding, and noise only increases the difficulties of finding people and accessing conversation and social cues (Bennett, 2022c). One stated "multiple conversations taking place simultaneously creates extra clutter, overwhelming my brain" (Bennett, 2022a). Another explained that "people disappear right in front of me" (Brossart, 2024). Some reported difficulty perceiving and processing another person's emotions and social cues (McDowell, 2019).

Individuals with CVI use compensatory skills to identify people, such as recognizing people by their hair color or distinct features (e.g., glasses, beard) (Liu et al., 2016). **One** individual said, "My mom could change her hair and I couldn't recognize her at all" (Hamilton et al., 2019). Some may also rely on auditory information, the sound of footsteps or the recognition of voices, rather than the visual information that faces provide. In social settings, some with CVI reported they arrive when it's not busy, find "quiet corners," and avoid discussions in large groups (McDowell, 2019).

2.4 Mental and physical health

A thread that weaves throughout each of the above areas is CVI's impact on mental health, stress, anxiety, emotional wellbeing, and physical health. Visual impairments "carry a heavy psychological burden," where people with vision loss have an increased risk of anxiety and depression (Klauke et al., 2023, p. 3). Children with CVI are at risk for health-related quality of life impairments that encompass psychosocial health, and physical, emotional, social, and school functioning (Collart et al., 2024).

In a panel discussion with four adults with CVI at the Perkins 2024 CVI Conference, a key theme was how CVI was so much more than a visual impairment. All shared that CVI is a "full body experience" that can include migraines, nausea, fatigue, and chronic stress, which can have a long-term impact on health (Baskin, 2024). One adult advocated, "If you look at the medical information about CVI, it's not there: It's a visual condition. In reality, we are experiencing full-body medical issues related to CVI" (Baskin, 2024). One described CVI's impact on daily energy, where people without CVI wake up with 20 spoons, but with CVI you might only start with 10 spoons (Baskin, 2024). Another shared, "Sometimes, I just have to go lay down and be horizontal so that my brain can work" (Baskin, 2024).

Some with CVI discuss the stress and anxiety they live with. In the statements reviewed, many described CVI's emotional toll in daily moments with words like "frightened," "flight or fight," "anxious," "stressful," "overwhelming," and "scary." One adult explained, "CVI has left a hole in my life and identity, as understanding the visual world, in part, integrates a big part of your identity and how you relate to the world and people within, touching every aspect of your life" (Bennett, 2022a).

3 Discussion

First-hand accounts from individuals with CVI illustrate the diverse presentations of CVI and how visual access is inconsistent due to many factors (e.g., environment, noise, clutter, temperature, fatigue, stress). Vision use can be difficult, disrupting access to learning, safe navigation, and social interactions, which may cause increased stress, anxiety, and isolation, and very real effects on health and wellbeing.

People with CVI shared a range of compensatory skills used for access to education and daily activities that included auditory, kinesthetic, tactile, and visual skills. There is no one-size-fits all approach. Professionals working with people with CVI must always use a person-centered, holistic approach, and consistently consider the impact of the environment, task, learning materials, and context. Comprehensive assessments (CVI, functional vision, learning media, assistive technology, orientation and mobility), ongoing observation, and collaboration among team members are foundational for implementing accessible instruction and services that support learning, safe navigation, social inclusion, and general wellbeing.

With CVI emerging as a common condition in children, we must listen to people with CVI to inform future areas of research and interventions. Some specific avenues to explore are (1) how CVI impacts quality of life outcomes and the interventions that can help improve these outcomes, (2) non-visual medical issues and long-term health effects of living with CVI, (3) strategies and accommodations to reduce fatigue and anxiety, (4) environmental factors that support access to learning (e.g., lighting, reduced clutter, sound levels), (5) the impact of assistive technology and compensatory techniques on daily functioning, (6) the effect of multisensory instructional approaches and dual media literacy on learning outcomes, (7) how to support inclusion in social contexts, or (8) large prevalence studies to capture the heterogeneity in the CVI population, including associated conditions and socioeconomic risk factors.

4 Limitations

The sample of first-hand accounts is small and does not fully represent the heterogeneous CVI population or spectrum of manifestations. This group also included individuals with CVI who can share their stories. The authors acknowledge that a large population of those with CVI have complex needs that may limit their ability to convey their experiences. This review exemplifies the need to further analyze the lived experiences of individuals with CVI to uncover the themes, strategies, and challenges for these individuals through a qualitative and mixed-method analysis.

The authors focused primarily on the visual perception of three-dimensional objects, people, and events. Emerging research and individuals with CVI discuss the difficulty of perceiving two-dimensional visual targets, which was briefly mentioned in this article. CVI's impact on the perception of two-dimensional visual targets is a broad topic requiring further analysis. CVI also impacts many visual processing areas (visualization, visuospatial), and further study is needed to ensure individuals with CVI receive holistic and high-quality assessment and educational programming.

Author contributions

RB: Conceptualization, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. MT: Conceptualization, Investigation, Methodology, Writing – original draft, Writing – review & editing. EM: Conceptualization, Investigation, Methodology, Writing – original draft, Writing – review & editing. NY: Investigation, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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EDITED BY
Karen Harpster,
Cincinnati Children's Hospital Medical Center,
United States

REVIEWED BY
Arvind Chandna,
Smith-Kettlewell Eye Research Institute,
United States
Terry Schwartz,
Cincinnati Children's Hospital Medical Center,
United States

*CORRESPONDENCE
Helen St Clair Tracy
✉ hsct1@st-andrews.ac.uk

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Emulation of the subjective experience of visual dorsal stream dysfunction: a description of three in depth case studies

Helen St Clair Tracy[1]*, Nicola McDowell[2], Gordon N. Dutton[3], John Ravenscroft[4], Isobel Hay[5] and Andrew Blaikie[1]

[1]Infection and Global Health Division, School of Medicine, University of St Andrews, St Andrews, United Kingdom, [2]Institute of Education, Massey University, Auckland, New Zealand, [3]Glasgow Caledonian University, Glasgow, United Kingdom, [4]Moray House School of Education and Sport, The Scottish Sensory Centre, University of Edinburgh, Edinburgh, United Kingdom, [5]NHS Dumfries and Galloway, Dumfries and Galloway, United Kingdom

These case studies explore the subjective visual experiences of individuals with cerebral visual impairment (CVI), specifically dorsal stream dysfunction (DSD) characterized by simultanagnosia. Through three in-depth case studies, this work documents the challenges these individuals face when navigating cluttered environments. The individuals were asked to describe their visual experiences while watching videos of varying complexity, with the future aim of creating a simulation of simultanagnosia. This process revealed a dynamic constriction of their attentional visual fields as scene complexity increased, and vice versa. Notably, the volunteers experienced a phenomenon where their vision could "get stuck" on specific items, with an apparent concurrent reduction in their ability to perceive and describe visual information as visual clutter increased. These consistent observations indicate that the symptoms of simultanagnosia are not simply limited to perceiving one or two objects at a time but can vary dynamically in response to environmental complexity. They enhance our understanding of how DSD impacts visual search and perceptual experiences, prompting us to propose the term "simultanagnostic vision" to describe this more nuanced and dynamic manifestation of CVI. The results are critical for developing effective interventions and optimizing support strategies for individuals affected by DSD, particularly children at sensitive developmental stages. Furthermore, we recommend deeper investigation into how different visual processing streams in the brain interconnect and influence each other, which may open new avenues for targeted therapeutic interventions.

KEYWORDS

cerebral visual impairment, CVI, simultanagnosia, simultanagnostic vision, dorsal stream dysfunction

1 Introduction

Our vision is constructed through the dynamic interplay of multiple interconnected brain regions that process visual information from our surroundings, engaging over 40% of the brain's capacity (Hubel, 1995). Although specific regions are associated with particular aspects of visual processing, these regions operate in highly interconnected networks (Bennett et al., 2021). Consequently, damage to or dysfunction within the brain can frequently impair vision, leading to cerebral visual impairment (CVI). A widely used definition of CVI describes it as a 'verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any co-occurring ocular impairment' (Sakki et al., 2018). However, this definition offers limited insight into the subjective visual experiences of individuals affected by CVI. According to the ICD-11 classification, CVI is categorized under "Specific visual dysfunctions" (9D92), which "refer to functional deficits in higher cerebral centers. Such dysfunctions may exist with or without visual impairment of the eyes and the lower visual system" (World Health Organization, 2024). The brain's image processing is highly complex, with different regions responsible for specific visual functions, making the outcomes of brain injuries or dysfunctions highly varied and likely unique to each individual. This poses a critical challenge: how do we come to understand how affected individuals perceive their worlds, and how can we use this information to provide effective support as a sequel to diagnosis?

Vision serves four primary functions: guiding movement, enabling social interaction, facilitating access to information, and promoting learning. This understanding is particularly important when teaching children (Hyvärinen and Jacob, 2011). For those with CVI, barriers to learning include low visual acuity, reduced contrast sensitivity, restricted visual fields, impaired movement perception (Pamir et al., 2021) and limited perceptual capacity, especially in terms of parallel processing, related to reduced nerve fiber numbers in the dorsal visual stream pathways (Bennett et al., 2019).

The dorsal stream processes visual data from the occipital and middle temporal lobes, relaying it to the posterior parietal lobes and the interlinked subcortical pathways to enable the visual guidance of movement (Glickstein et al., 2011). This process is typically non-conscious, allowing actions such as running upstairs without active thought (Milner and Goodale, 2006; Goodale and Milner, 2013). In contrast, the ventral stream relays visual information from the occipital lobes to the temporal lobes, facilitating recognition by matching incoming image data with stored memories (Milner and Goodale, 2006; Goodale and Milner, 2013). Both streams are processed in higher cerebral centers, and dysfunctions in either can lead to "perceptual visual dysfunction" (Pehere and Dutton, 2021). However, these functions are not stand-alone processes. The vertical occipital fasciculi (VOF), comprise key white matter pathways, linking the bilateral dorsal (underpinning visual guidance of movement and spatial attention) and ventral (recognition) visual streams, thought to play a key role in integration of these distinct visual functions, in those with typical vision (Yeatman et al., 2014). The connectivity of both VOFs enables integration of visual information, supporting coherent perception and visual memory. Disruptions in the VOF can impair these functions, emphasizing the importance of viewing visual processing as an interconnected network rather than isolated pathways (Bauer and Merabet, 2024). How these interconnected streams are affected when one of the visual centers is not processing typically is not well understood and may vary uniquely among individuals. Therefore, the complexity and individuality of such impairments make it essential to fully ascertain each person's CVI etiology and pattern of visual affectation to provide optimal support.

Dorsal stream dysfunction (DSD), often seen in those with CVI, can result in multiple visual impairments, such as optic ataxia, where individuals struggle with using vision to accurately guide their movements, rendering everyday activities like writing and eating difficult (Vighetto and Salmon, 2007). DSD can also interfere with finding objects in cluttered environments, leading to slower and less precise visual search (Bennett et al., 2020; Zhang et al., 2022). Balint syndrome is the most severe variant of this condition (Gillen and Dutton, 2003). Unilateral parietal lobe pathology can also cause hemi-inattention, where individuals become less aware of one side of their visual field, particularly when the right parietal lobe pathology leads to left sided inattention, with severe cases resulting in visual neglect (Vighetto and Salmon, 2007). While CVI-related impairments in visual acuity, contrast sensitivity, and visual field are well understood, DSD and its effects remain less well recognized, despite detailed reports of simultanagnosia in adults (Michel and Henaff, 2004). Simultanagnosia, an element of DSD, manifests as difficulty perceiving more than one or two objects in a visual scene at once (Hokken et al., 2024), and in severe cases, an inability to simultaneously perceive the parts of a single object (Coslett and Lie, 2008). Simultanagnosia is typically described by the challenges it causes, but questions remain about its underlying nature and how it affects vision at a subjective level. Understanding this is crucial for providing adequate support.

To better understand simultanagnosia, we collaborated with three adults with formally diagnosed DSD to ascertain the nature of their visual experiences and from this, to create a video simulation. By in-depth analysis of what they described seeing during real time observation of photos and videos and discussing their responses with them, we aimed to visually replicate their descriptions, with the aim of helping others affected by this condition to better understand the nature of their vision. This paper presents their accounts, how they contributed to our understanding of simultanagnosia, for the development of a simulation to depict their experiences. We also discuss public responses to the simulation, which was made freely available online. The following narrative outlines the process that led to new insights into simultanagnosia, opening avenues for future research, potential diagnostic methods, and even potential therapeutic options. Additionally, the project prompted revisions in how CVI can best be explained, particularly the importance of distinguishing between the ventral and dorsal pathways in CVI and the need to understand how other pathways and regions in the brain are affected and react when visual areas are not processing typically. The events are presented in chronological order to reflect the unfolding nature of this inquiry.

2 Background

In 2016, although it was generally assumed that clutter and complexity negatively impacted children with CVI, there was limited published evidence, with only one small case study available (Little and Dutton, 2015). An email discussion between a CVI expert, an adult with acquired CVI, and the mother of a child with CVI explored the nature of CVI, particularly simultanagnosia, from their unique perspectives. It emerged that the adult with CVI found environments with less clutter not only easier to navigate but also conducive to better visual perception. This observation had not been previously described and prompted our group to informally test this concept framework.

To investigate further, we initially asked the adult with acquired CVI to view photographs and later expanded the inquiry to include two additional adults with CVI, including DSD, viewing videos. The three participants, anonymized as Cases 1–3, have all been the subject of previously published findings.

2.1 Case 1 medical history

As a 16-year-old, Case 1 initially presented with an acute intracerebral hemorrhage caused by the rupture of a left occipital arteriovenous malformation (AVM). Emergency neurosurgical intervention was performed, resulting in the removal of her left occipital lobe. Post-operatively, Case 1 developed right homonymous hemianopia due to the damage sustained by her occipital lobe. In addition, she experienced a transient episode of left-sided hemiparesis, which resolved shortly thereafter. Following recovery from surgery, Case 1 experienced persistent challenges with visual perception. She struggled to cope in crowded environments and had significant difficulties processing complex visual scenes. These symptoms are consistent with the dysfunction of visual processing pathways associated with the left occipital lobe damage. This published case (McDowell and Dutton, 2019), highlights the long-term visual and cognitive sequelae resulting from the hemorrhagic event and subsequent surgical intervention.

2.2 Case 2 medical history

Case 2 was diagnosed with sagittal suture stenosis and scaphocephaly at age 3 months. A follow-up at a tertiary center at age 3, including a brain MRI, found normal development. However, Case 2's mother recalls that as a pre-schooler, he "…could not remember or retain information about shapes." At age 5, pediatric examination identified severe generalized hypotonia with normal muscle power and noted issues with social communication with peers. Neurological and genetic investigations, including MRI of the brain and spine, as well as peripheral nerve conduction tests, revealed no cause for hypotonia. At age 11, Asperger's syndrome (ICD-10 criteria) was diagnosed after multidisciplinary assessment. An initial ocular examination at age 12 was within normal limits (unaided VAs of 6/9 OD and 6/24 OS, corrected 6/6 OD and 6/9 OS) with automated visual field assessments being normal. Mild left optic nerve pallor was noted. At age 13, tertiary neurological assessment identified Balínt syndrome, and recommendations were made for support as a child with severe visual perceptuomotor disability. At 14 years of age, Case 2 developed severe anxiety and depression, which required medication. At age 15, follow-up ocular examination found stable visual acuities although constricted visual fields were noted on perimetry. No retinal signs were noted. On review at age 17 the clinical findings were stable. Appendix 2: Poster: "Visual and Motor Deterioration in Asperger's Syndrome," is a case report on Case 2. Case 2 has featured in previous published research further documenting assessment tests (Hay et al., 2016).

2.3 Case 3 medical history

Case 3 was born at 38 weeks' gestation with a normal birth weight, but exhibited floppiness at birth, which persisted for several days. She also manifested unexplained cyanotic episodes within the first 72 h of life. Her motor development was delayed; she sat with support at 8 months, crawled at 1 year, and walked independently at 14 months. Throughout early childhood, she experienced frequent trips and falls. As a young child she developed a habit of tugging on her parents' clothes while walking, a behavior that persisted into teenage years, indicative of lower visual field impairment. During infancy, she often used her left hand while her right arm remained at her side. Toys were placed in her right hand to encourage its use. By 8–9 months of age, she could finger-feed independently, but by her late teens, she continued to struggle with using cutlery, tying shoelaces, and folding clothes. In primary school, Case 3's handwriting difficulties and motor clumsiness were noted. A Test of Visual Perceptual Skills assessment revealed significant challenges with visual discrimination and closure. It was also apparent that she recognized shapes by tracing their outlines with a finger rather than relying on vision alone. Pediatric examination revealed general hypotonia and ligamentous laxity, though no significant neurological impairment was found. At the age of 7 she was referred for multi-disciplinary assessment for possible social-communication disorder due to difficulties with peer relationships and a dependence on routines. A diagnosis of autism spectrum disorder (ASD) was made. By the age of nine, her behavior was indicative of severe visual field restriction. She often tripped over low-lying objects, bumped into doorframes, and knocked food off her plate. She also had difficulty with facial recognition. Despite these symptoms, ocular examination was normal, but perimetric examination confirmed severe visual field constriction, subtending only five degrees. However, MRI scans of the brain and anterior visual pathways were normal. At 13 and 14 years old, serial hand movement examinations using slow-motion video identified bilateral optic ataxia, which was most pronounced in the left (dominant) hand and left hemifield. This was consistent with bilateral, or predominantly right parieto-occipital dysfunction; Balínt syndrome was subsequently confirmed by tertiary neurological opinion. Case 3 has featured in previous published research further documenting assessment tests (Hay et al., 2020).

3 Methodology

To examine the potential impact of crowding and clutter on visual perception, photographs were initially taken in various settings with differing levels of complexity. This was followed by the creation of videos, beginning in a small shop that was closed for filming. Visual attention is mapped across the entire body, not just the eyes and head, and is continuously re-mapped as individuals move through their surroundings. This process is essential for avoiding collisions and ensuring accurate visual guidance for reaching (Reed et al., 2007). Replicating this body-centric perspective in videos required the development of a novel method that, to our knowledge, had not been previously attempted. Our initial approach used two synchronized GoPro cameras mounted in a 3D-printed harness; however, this setup produced footage that was unstable and visually disorienting.

A simpler and more effective technique was developed by holding a digital camera at navel height with both hands, keeping the arms close to the body for stability. This method allowed the camera to move naturally with the individual, achieving the desired body-centered perspective. Using this approach, additional videos were recorded in both cluttered and uncluttered environments. From several hours of footage, six short clips were selected to represent a range of settings with varying levels of complexity and movement, enabling an assessment of the impact of these factors on visual perception.

This study was conducted as a qualitative exploration rather than adhering to a formal research protocol; accordingly, the methodology was not standardized. Instead, the design of the sessions described below was guided by the team's expertise. The methods applied evolved organically, adapting in response to feedback from the volunteer participants. This flexible approach is exemplified in the findings from the following three case studies.

4 Data

4.1 Case 1

Case 1 is an academically high-achieving professional woman, who is open, and good-humored, with a love of sports and outdoor activities. She acquired CVI while at school, which led to considerable difficulties that required resilience and determination to overcome.

As an adult with acquired simultanagnosia, Case 1 underwent diagnosis and engaged in a self-developed program of "conscious vision strategies" (Appendix 1). These exercises were designed to transform her unconscious visual processing into a more conscious experience. By gradually honing this approach, she developed the ability to better recognize and understand the nature of her visual perceptions. This process revealed that her visual experience varied dynamically, with the extent of what she could perceive fluctuating, in relation to the visual complexity of her surroundings.

Case 1 gradually realized that as the visual complexity of her environment increased, her visual field became more restricted. In 2016, to evaluate this phenomenon and create a simulation for the launch of a CVI website, Case 1 was provided with photos of varying complexity, including images from inside shops and open countryside. She was asked to describe what she could see ("say what you see") in each image. Her qualitative responses confirmed that her vision varied with the complexity of the scene: the more complex the visual environment, the fewer details she was able to perceive and describe.

Case 1, when viewing the image of the field on the left (see Figure 1), remarked that she could "*study the buildings at the back of the image and they become clearer*" and felt that she had seen "*every element in the photo: grass, hedges, trees, buildings, and sky.*" In contrast, when looking at the photo on the right, Case 1 noted, "*I was straight away drawn to the round hoops in the middle. If I tried to move my gaze away, it quickly got drawn back to them. A bit like a magnet, I guess.*" Her responses suggest that while Case 1 could perceive the entire scene in the simpler image, her attention was involuntarily fixed on specific elements in the more complex image, limiting her overall visual awareness. Full responses to these and other photos are available on the University of St Andrews School of Medicine website (University of St Andrews, 2024). What was not seen by Case 1 in the images was covered with a fog filter (Figure 2).

Reviewing the two images and the impact of simultanagnosia on vision, it became evident that visual clutter significantly impaired Case 1's ability to perceive the scene. In 2018, Case 1 expressed a desire to further explore her perception of moving images in videos and help others with CVI better understand their vision. Before creating the videos, Case 1 walked through a busy gift shop, using a video camera to capture its surroundings, while explaining the challenges she faced. She reported feeling as though her "*eyes were darting around uncontrollably*," making it impossible to form a cohesive picture of her environment. She noted that these darting eye movements were primarily horizontal and explained that she "*preferred horizontal lines*," finding spaces without vertical lines "*much calmer.*" This real-time recording provided valuable insight into how Case 1 navigated cluttered spaces and highlighted the nature of her struggle to maintain visual coherence amidst the complexity, along with a resulting sense of stress and confusion.

To develop a video simulation of simultanagnosia, University of St Andrews Schools of Medicine and Computer Science partnered with the NGO CVI Scotland. Six short videos depicting everyday scenarios with varying degrees of complexity and movement, aiming to simulate simultanagnosia in a dynamic context, were created (Table 1 and Figure 3). The method of asking affected individuals to "say what you see" was once again employed to capture real-time descriptions of their visual experiences.

Case 1's subjective visual challenges with visual crowding and search were initially explored via email. After viewing the videos, Case 1 provided written descriptions of what she was able to perceive. She found certain videos difficult to watch, at one point feeling startled. In one instance, she described seeing only part of a shop assistant's smiling face, remarking, "*Cannot read expression*" while noting that the person, "*does not seem happy*." During a beach video, Case 1 initially stated, "*I can see everything,*" but when a group of people having a picnic appeared, she fixated on that scene and could

FIGURE 1
Two of the photographs used by CVI Scotland when attempting to create a simulation of simultanagnosia.

FIGURE 2
The application of a filter to cover areas that were not perceived, demonstrating an initial attempt at simulating simultanagnosia. Note that while the content inside the hoops was also not perceived, at that stage the significance of this was not yet recognized, and thus it was not initially covered.

TABLE 1 The six videos used to create the simulations, along with their duration (in seconds) and a brief description of each.

Video title	Length (seconds)	Content
Shop (A)	69	Moving through a university gift shop, with toys, clothes and trinkets.
Shop (B)	32	Static position in the same shop, looking at shirts, moving towards the end, approached by someone.
Driving (C)	77	From the passenger seat position, moving (30 mph) through an open part of a town with parks and houses.
Driving (D)	44	From the passenger seat position, slow / stopped in traffic, build up.
Beach (E)	31	Walking on a beach, passing a small group having a picnic and some dogs playing.
Beach (F)	29	Walking on an open beach, passing some people playing football with a dog.

The references (A–F) correspond to the images labeled A to F in Figure 3.

only see one dog, even though there were two. Once the film moved beyond the picnic, Case 1 remarked, "*Lovely open beach, I can even see the seagulls*," which were small and in the far distance. The results aligned with Case 1's previous experiences with photographs: as the visual complexity increased, she could perceive fewer items. To simulate this experience, a "fogging" filter was dynamically applied, to cover the areas of the image that had not been reported (Figure 4).

When the duplicate videos with the fog filter were sent to Case 1 for feedback, she commented, "*Some of the videos seem extreme, and I feel I can see much better than that. But then if I think about it and put myself in the different environments shown in the videos, they are pretty accurate.*" She particularly appreciated how the beach videos captured the constant fluctuation in her vision and felt the shop videos accurately depicted situations she had repeatedly experienced. Case 1 noted, "*I especially like how the filter highlights fast movement and how it reduces what you are seeing.*" A close family member, who had also viewed the videos, felt they accurately reflected the shifts in Case 1's visual experiences, noting changes in anxiety levels and behavior in stressful environments, such as shops, compared to more open spaces like the beach. These findings suggested a dynamic element to simultanagnosia that, at the time, had neither been researched nor documented. The team were eager to share these novel insights with the broader CVI community but, mindful that the findings were based on one individual's experience, sought input from two other adults with simultanagnosia as an element of their CVI diagnoses.

Cases 2 and 3 (Sections 4.2 and 4.3) were asked to watch the same videos in person, with their real-time responses being recorded for analysis. Both the volunteers were seated in quiet, uncluttered rooms and viewed the videos on a laptop positioned at eye level, 30 cm away. The videos were silent, and they were asked to naturally narrate what they could see without making any effort to search for details.

However, the findings from Cases 2 and 3 indicated that they may be more severely affected by simultanagnosia than Case 1, who appeared to perceive more, possibly due to years of practicing conscious vision strategies. Case 1 had not previously viewed the videos in the same "say what you see" manner as Cases 2 and 3, so she was asked to watch one of the videos in this way for comparison. With her agreement, this session was video recorded for review and analysis. The *Shop (A)* video was selected.

Shop (A): At the start, the pace of the video was too fast for Case 1, and she physically pulled back from the screen, similarly to Case 3. Her vision became "stuck" on a teddy bear, and even after the video moved on, she was unaware that the teddy bears were no longer present. When later asked, "*When you cannot see things, what's that experience like?*" she responded,

> "*It's almost like an anxiety response. I wanted to see what was there, yet I could not see it, so I got anxious about it. I got frustrated, and the emotions kicked in, which meant that I stopped looking at anything else. I could see the camera was still moving, but I was thinking about the teddy bears. I wasn't responding to the information that was coming from my eyes… I wasn't even aware of that. It's almost like it had switched off that sense.*"

FIGURE 3
A representative still image from each video, showcasing the typical content and complexity level. The references **(A–F)** align with the video descriptions in Table 1.

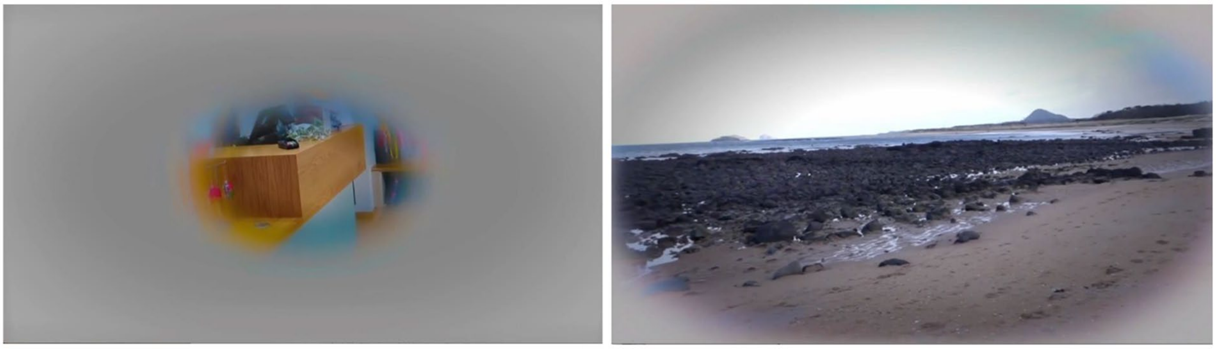

FIGURE 4
Images from the Shop (A) and Beach (C) videos with the added simultanagnosia simulation filter, showing the greater area of vision described on the open beach.

She found it challenging to stay focused on the task and had no memory of previously watching the video. Case 1 noted that she believed her visual function had improved over time, but was surprised by how difficult she found it to view the video. She explained that her fixation on the teddy bears was likely because she had liked teddy bears as a child. Interestingly, in this "live" review, she saw considerably less than she had during their original assessment of the video. The number of items correctly identified by Case 1 is documented alongside the results from Cases 2 and 3, with a comparative analysis presented in Table 2.

This illustrates the complexity of simultanagnosia and how personal emotional responses and past preferences can influence visual perception, especially in visually complex environments. It also suggests that even with conscious vision strategies, the dynamic nature of simultanagnosia continues to present significant challenges.

For the purposes of this manuscript, as Case 1 initially reviewed only one video, she agreed to view the remaining videos. In 2019, when Case 1 first viewed Shop (A) (Table 1), her performance did not stand out compared to Cases 2 and 3. However, upon re-watching the same videos 5 years later, the difference was significant (see Appendix 3). When Case 1 re-watched Shop (A) in 2024, she reported seeing 21 items, a substantial increase from the 6 items she noted in 2019. During this period, Case 1 continued practicing her conscious vision strategies, which may well have contributed to their improved visual perception. The fact that Case 1 had previously seen the video does not fully explain the 15 additional items she was able to identify, suggesting that these exercises have indeed been beneficial. While reviewing previous findings, Case 1's experience of getting "stuck" on the teddy bears in Shop (A) was revisited, and the possibility of palinopsia was raised. Palinopsia is a phenomenon where visual images persist after the object has been removed from view (Bender and Feldman, 1976). Different types of palinopsia include seeing what was there before as either a vivid image, a "ghostly" translucent presence, or a trailing effect.

4.2 Case 2

Case 2 is an academically successful young man who is warm, friendly, and good-humored. He does not make eye contact during conversation because it is difficult for him, and he is unable to

TABLE 2 Number of items identified by Cases 1–3 and the comparison across the six videos and one of the videos, Shop (A) with the simultanagnosia simulation filter added.

Number of items seen							
Video	Shop (A)	Shop (B)	Driving (C)	Driving (D)	Beach (E)	Beach (F)	Shop (A) + Filter
Length (sec)	69	32	77	44	31	29	69
CVI Case 1	6	–	–	–	–	–	–
CVI Case 2	5	6	8	6	9	8	5
CVI Case 3	7	5	13	5	8	6	5
Comparison	33	14	19	43	16	16	26

recognize faces. To navigate his surroundings, he has developed a self-taught visual search technique, scanning different points to mentally map his environment. Despite this, he often bumps into people or objects, not realizing they are there, and struggles to locate items that have been put away. Case 2 was asked to view the same videos as Case 1, and his responses are outlined below in the order he watched the videos. Table 2 provides the number of items he described in each video, and Appendix 3 contains his full verbatim responses.

Beach (D): He identified two people, one dog (out of two), and a football, each when they appeared in the center of his gaze. He missed elements in the peripheral areas, such as a hill in the background and birds in the sky.

Driving (E): He did not notice a large traffic light at the beginning but was aware of the traffic. Peripheral objects, including people, houses, trees, a wall, lamp posts, road signs, a bright blue bin, and additional traffic lights, were not observed. However, when trees and a large white building appeared in the center, they were recognized, though other nearby structures, including church spires, were missed.

Shop (A): He noticed teddy bears and clothing but could not specify details, though he was able to read 'University of St Andrews' on a T-shirt. After 40 s, he requested the video be stopped, stating, "*It's because it's moving so much. I usually rely on looking at different points and making a mental map; it's hard if it keeps moving all the time.*"

Beach (C): He saw two people and recognized them as the same from the previous video. He missed a line of trees on the right and only noticed a hill when it briefly appeared in the center, saying, "*There appears to be a sort of mountain type object.*"

Driving (F): He saw buses, cars, and one of three church spires in the center, along with traffic lights, but nothing in the periphery. People, shops, buildings, and trees on either side were missed.

Shop (B): Similar to the Shop (1), he recognized clothing but could not specify details, though he again read 'University of St Andrews' on a T-shirt. Despite a pause, he missed clothes hangers in the center of the screen. He noticed the assistant's torso briefly on the side but missed a woman in the center, a man behind her, and the assistant gesturing for attention.

Next, he was asked to view *Shop (A)* again with the simultanagnosia simulation filter. He approached it as before, narrating what he saw.

Shop (A) with Simultanagnosia Simulation Filter: He identified bears, clothes, and later additional items, such as the cash register. With the filter, he was able to watch the entire video, remarking, "*I can see everything.*" When asked to compare the filtered and unfiltered versions (Figure 4), he said, "*not that much difference to be honest.*" Upon pausing the video at the end, he acknowledged the grey fogging effect, noting, "*my mind maps images, so at some point during filming I've looked away from the center … there is a large grey color that cut out a large portion of the screen.*" He confirmed he only noticed the filter when looking away from the center, and when asked to focus on the paused image, he said he could see "*a bit of it [the grey fog] on the edges when I look at the center, but only slightly.*"

These observations reinforce the dynamic nature of his visual experience and how simultanagnosia affects his ability to perceive complex scenes (see Figure 5).

When asked how he was feeling, Case 2 responded, "*I am feeling slightly mentally fatigued.*" Upon explaining that their descriptions suggested fluctuations in the area of his field of vision, he reflected, "*I'm not sure if I just do not notice it, but I do not tend to notice my field of view changing. I think that's because it's sort of gradual if it does… I do not notice it varying.*" This response suggests that while there may be dynamic changes in his field of vision, these shifts are not readily perceptible to him, even though the findings indicate variations based on visual complexity.

4.3 Case 3

Case 3, a warm, communicative, and intelligent young woman, struggles socially in crowded environments and quickly becomes tired and overwhelmed by excessive visual complexity and noise. Busy places cause significant discomfort, and crossing roads safely presents challenges. She relies heavily on familiar routines and environments she has memorized for navigation. Case 3 was asked to view the same videos as Cases 1 and 2. Her responses to each video are outlined below, with additional discussion points. The number of items described in each video is listed in Table 2, and their full verbatim responses are included in Appendix 3.

Beach (D): Case 3 identified two people, two dogs, and a green football. However, she did not notice elements in the peripheral areas, such as the hill in the center left or the trees in the center right.

Shop (A): As the video was being set up, a slightly blurred, still image of shop displays appeared on the screen (Figure 6). Upon seeing this image, Case 3 startled, grimaced, said "*ugh*" and moved her head away from the screen. This reaction mirrored a similar response from Case 1, who, when viewing a photo of supermarket displays, commented, "*My first response was to pull away from the photo, as it was too much information, and my automatic reaction was to try to get away from it*" (University of St Andrews, 2024).

Once the video started, Case 3 described seeing teddy bears and children's clothing, though she could not specify details. After long pauses, she later mentioned seeing more clothes (unspecified), the tills, and the sales assistant. These reactions suggest that visual complexity, particularly in cluttered environments like shops, can

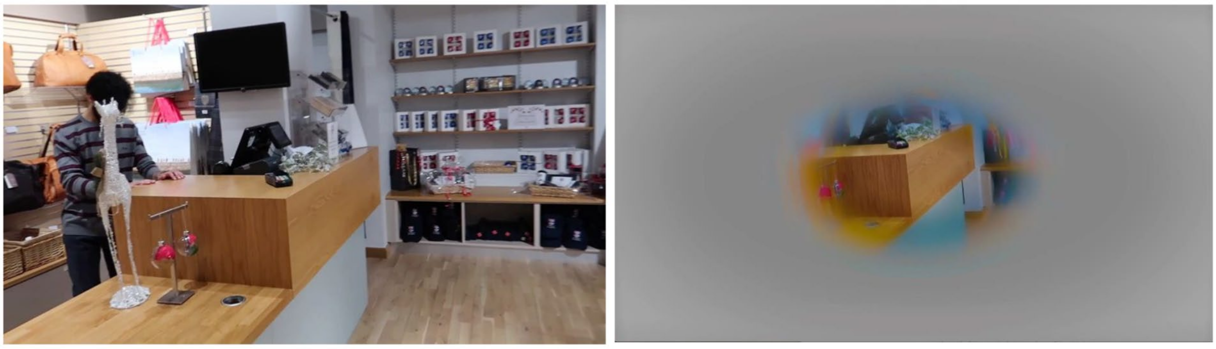

FIGURE 5
The right image is the still image from the end of the Shop (A) video. The right image is the same picture with the filter applied.

be overwhelming and cause a strong aversive response, reinforcing the need for carefully managed environments to reduce visual overload in individuals with simultanagnosia.

Driving (E): Case 3 noticed the traffic lights at the beginning of the video but missed houses, trees, and cars on the sides, though she did observe the bus as it moved into the center of her field of view. She also reported seeing moving cars and a person with a dog to the side but failed to notice another person in the same position a few seconds later. After a pause, she noticed a pink van and, towards the end, some houses and the traffic lights again. Aside from the person and dog, Case 3 did not perceive anything in the peripheral areas.

Beach (A): Case 3 reported seeing rocks, sand, a black Labrador dog, a wheelchair, one person (though there were two), a red setter dog, some trees, and the sea. However, she did not notice the hill in the background. These observations further highlight Case 3's challenges in perceiving peripheral details and their tendency to focus on central elements, missing key objects or people in more complex scenes.

Driving (F): Case 3 observed one double-decker bus, though there were two next to each other, and noticed a bright blue car on the far left, which stood out due to its color. She mentioned "some buildings" and "traffic" but was not specific. She missed key elements such as people, three church spires, a bus stop, shops, and traffic lights on both the left and right of the screen. Across all videos, the only peripheral items Case 3 noticed were a person and a dog in *Driving (E)* and the blue van in *Driving (F)*. When asked, Case 3 mentioned she was a dog lover and is particularly drawn to bright colors, especially pink. After viewing this video, she requested a break that lasted 15-min before continuing.

Shop (B): Case 3 noticed t-shirts and bright red and blue baseball caps but missed grey and navy-blue caps on either side. She also did not see a shirt being removed, the assistant walking towards the camera and gesticulating, or three people in the shop, including the assistant, during a second appearance. These responses emphasize how personal preferences, like a fondness for dogs and bright colors, can influence what Case 3 perceives in cluttered, complex environments. It also highlights her difficulty in noticing more neutral or less distinct items in the visual periphery.

Following this, as with Case 2, Case 3 was invited to view the simultanagnosia simulations the team had been developing, and she agreed (Figure 4).

Beach (D) with Simultanagnosia Simulation Filter: Case 3 saw more background elements (rocks, sand, and trees) that had not been visible previously. She also recognized the dog, two people, another dog, and a green ball as before, but still did not notice the hill. When asked about the filter, Case 3 commented that the video felt "more narrowed down" but that she was "not really aware there was less to see." In the discussion afterward, Case 3 remarked that with the filter, it seemed like there was more to see, not less.

Shop (A) with Simultanagnosia Simulation Filter: Although Case 3 did not notice many additional items (mentioning a teddy bear, some baby clothes, a pink jumper, some displays, and a till), she commented that the scene appeared less cluttered with the filter.

Shop (B) with Simultanagnosia Simulation Filter: Case 3 was able to read 'University of St Andrews' on a top when viewing the video with the filter. Without the filter, she could only see "t-shirts" in the same video.

4.4 Comparison (adult without CVI)

Although it was clear that Cases 1–3 were experiencing visual perception far from typically, the definition of 'typical' vision had not yet been established for comparison. To address this, one of the team members, who has no visual impairments and had not previously seen the videos, agreed to watch the same videos in the "say what you see" manner for comparison. They also gave permission for the session to be video recorded for review and documentation purposes. This provided the team with an initial reference point for typical visual perception under similar conditions, helping to further explore and contrast the unique visual challenges experienced by individuals with simultanagnosia.

Table 2 presents the number of distinct items correctly identified by Case 1, Case 2, Case 3, and the comparison, across six videos, as well as one video with the simultanagnosia simulation filter. This comparison provides a quantitative measure of the differences in visual perception between the cases and someone with typical vision. The full verbatim responses from the comparison participant are available in Appendix 3 for detailed reference. This table helps highlight how each individual's visual experience differs, particularly in complex or cluttered environments, and provides a clearer understanding of how simultanagnosia affects visual recognition and naming of objects.

FIGURE 6
This still image, displayed on a laptop screen, was sufficient to provoke a startle response in Case 3, who grimaced and instinctively moved their head away from the screen. Despite the slight 'softening' effect of the blur, the image remained difficult for Case 3 to visually process, as indicated by her reaction.

At this stage, it was deemed inappropriate to ask anyone else to view the videos without obtaining ethical clearance, given that the content could be stressful and challenging for viewers. However, a small internal team, including another adult with simultanagnosia and a parent, was asked for feedback, and they confirmed that the findings resonated with their own experiences.

5 Discussion

The initial aim of the project was to explore the feasibility of accurately simulating the visual perception challenges associated with DSD, particularly in relation to simultanagnosia. These difficulties have been characterized by problems such as locating familiar objects, like identifying a known individual in a crowd or finding a toy in a toybox (Bennett et al., 2018, 2020). It became evident that, while participants could only name one object at a time, when viewing videos with less visual clutter, they could perceive a broader area of the scene. By contrast, in more complex environments, such as the *Shop (A)* video, the area of vision where they could see appeared to be significantly reduced. This limitation made it more challenging to locate and identify familiar objects, resulting in prolonged task completion times, along with increased fatigue and stress.

Visual neglect, a condition marked by a lack of visual attention in areas where there is typically a normal visual field, has been described as 'a neurological disorder characterized by a deficit in attention to stimuli on one side of the body' (Ting et al., 2011). This condition is most commonly associated with one-sided impairment following a stroke (Linden et al., 2005). Zihl and Dutton (2015) further elaborate that "after acquired brain injury, the field of attention can be altered in an even more dramatic way, with part of the field of attention potentially missing."

In developing the simulations, the observed visual areas suggest a form of attentional visual field loss, though affecting the periphery rather than one side. Additionally, the extent of peripheral attentional field loss appeared to fluctuate for the same individual, depending on the visual complexity of the video, indicating that this type of attentional visual field loss is dynamic. The negative impact of visual complexity on individuals with CVI is now well-documented. For instance, Bennett et al. (2018) described the difficulty in finding a toy in a toybox when distractions were added, and McDowell and Butler (2023) found that increasing complexity made shape-matching tasks more challenging. Both studies demonstrated that children with CVI struggled significantly more with these tasks than control groups. Moreover, the three case studies consistently showed that reducing visual complexity not only improved access to the visual field but also fostered greater emotional comfort. This aligns with recent findings (McDowell, 2023) and suggests that visual difficulties associated with DSD can be either exacerbated or alleviated by changes in environmental complexity (McDowell and Butler, 2023).

The concept of a simultanagnostic 'window' of vision within the attentional dynamic peripheral visual field can be likened to a camera's shutter-controlled aperture, dynamically opening across the field of vision. A similar phenomenon has been previously described. Holmes (1918), in his account of Case 2, observed that the patient 'could not be certain of the position in space of objects which he could see' and experienced 'bilateral inferior hemianopia,' suggesting optic ataxia and potential visual neglect in the lower visual field. This implies that Case 2 may have had Balint Syndrome, a severe form of DSD (Gillen and Dutton, 2003). Holmes also noted that 'the borders of his blind portion were indefinite,' which could be explained by the variability of this 'window'. Milner and Goodale (2010) described 'central sparing' where a small window of vision is available in the center of the visual field, in patients with optic ataxia. They suggested that this might occur because, in the absence of a functioning dorsal stream, the ventral stream compensates, relying on central vision. They observed "some cases of central 'sparing' during real-time reaching in optic ataxia patients." It raises the question of whether this central sparing might instead represent the simultanagnostic window of vision reported by Cases 1–3.

Zihl and Dutton (2015) similarly observed in Balint Syndrome that 'bilateral posterior brain injury can cause bilateral restriction of the field of attention, sparing, depending on the degree of severity, only a small area for visual information processing.' This phenomenon may also align with the concept of the 'window' of vision. Jacobson et al. (2002) found that in children born prematurely with bilateral posterior parietal brain injury, some had normal outer limits of the peripheral visual field but struggled with peripheral attention in crowded environments, consistent with our case study findings. Most recently, Hokken et al. (2024) used eye-tracking to map the gaze of CVI participants in increasingly complex scenarios, with findings consistent with our observations. In our study, while the 'window' of vision was predominantly central, it occasionally extended, particularly when a stimulus 'popped out.' For example, Case 3, a dog lover, was automatically drawn to a dog in the periphery. This 'pop-out' effect, where attention is involuntarily drawn to a particular visual stimulus amid competing stimuli, has been previously described (Adler and Orprecio, 2006).

When asked about the 'shape' of their window of vision, Case 1 explained that it matched the shape of whatever she was directly observing. For instance, when looking at a teddy bear, the window was teddy bear-shaped, rather than a circle with the bear in the center. She could only see the teddy bear itself, with nothing visible around it. Similarly, when looking at her splayed hand, she only saw the hand, with no perception of the space around it or between the fingers.

Figure 7 illustrates how Case 1 perceived the hoops: she not only failed to notice the surrounding environment but also overlooked the toys hanging within the center of the hoops. This observation suggests that our initial simulations, which used circular and oval "windows" to represent the visual experience, may require refinement to more accurately capture the true nature of their vision. Notably, this differs from Figure 2, reflecting the evolution of our understanding for the ongoing refinement of our simulations.

Case 1 described her experience when viewing the display with the hoops, noting, "*If I tried to move my gaze away, it quickly got drawn back to them. A bit like a magnet.*" This observation resonates with findings by Jackson et al. (2009), who identified a 'competing' element in a patient with simultanagnosia. Their patient struggled significantly more when multiple letters were presented simultaneously, compared to when letters were shown sequentially and overlapped, even for brief periods. It raises the question of whether Case 1's difficulty in shifting gaze from the hoops could represent a more extreme form of competing simultanagnosia, where the competing elements are so overpowering that they become visually disabling. This could also offer insight into why some profoundly disabled children with CVI are drawn to stare at bright lights (CVI Scotland, 2024a). If this is indeed a manifestation of competing simultanagnosia, it may serve as a potential method to identify the condition in individuals who may not have the capacity to follow instructions or answer questions, offering a new diagnostic tool for this population.

Cases 1–3 all found the shop videos, particularly *Shop (A)*, to be the most challenging, causing significant stress and fatigue. Case 2 had to stop *Shop (A)* at 40 s for a break, suggesting that environmental complexity affects not only visual processing but also overall well-being. This observation aligns with McDowell's (2023) study, where children with suspected CVI reported anxiety and difficulty regulating emotions in complex environments. It remains unclear whether emotional states, such as stress or anticipating a difficult situation, further impair visual performance. The need for breaks in Cases 2 and 3 suggests that both the complexity of the environment and emotional factors may influence visual functioning.

The responses to the driving video were notable. Despite the complexity of the environment and the speed of movement, which paralleled the challenges of the shop videos, none of the volunteers reported the same level of fatigue. The muted colors in the driving video may have contributed, but it is also possible that the simulation of being in the car with someone else driving allowed the participants to relax, as their automatic alert systems may have been temporarily suspended. This aligns with the idea that emotional state plays a key role in optimizing visual experiences. Further research is needed to determine whether reduced stress improves visual performance, but the findings suggest that challenging environments may be easier to tolerate when someone else is in control, such as being pushed in a wheelchair through a busy airport. This could also explain the popularity of activities like horse riding for individuals with CVI (CVI Scotland, 2024b).

When asked about her experiences during silent periods, Case 3 responded with "*I do not know*," while Case 1 described it as if her

FIGURE 7
Photo of hoops with what was not seen removed, including the objects in the center of the hoops, suggesting their window of vision in this instance was 'hoop shaped'.

'vision sense' had switched off. This highlights the paradox of anosognosia, where individuals are often unaware of their own impairments and view them as normal, rendering the condition disabling yet asymptomatic. However, in this study, Cases 1–3 were able to articulate their difficulties because they had received detailed explanations of their visual impairments years earlier, allowing them to recognize and navigate their challenges.

When viewing the video with a 'fog' filter, all three with DSD found the experience much easier. Case 3 described the fogged video as less cluttered, although she was unaware of the fog itself, which covered over 80% of the screen at times. The number of items she could see remained nearly the same (Case 2: 5 items with and without the filter; Case 3: 7 items without the filter, 5 with), but the task was perceived as less demanding, suggesting that reducing visual complexity alleviates demand on the brain.

Hypothesizing the mechanism behind these observations, we can conceptualize each visual nerve fiber as contributing a voxel; numerous smaller voxels in the central visual field and fewer, larger ones in the periphery. As the demands on the posterior parietal mapping system exceed its limited capacity, the smaller central fibers may continue functioning, while the larger peripheral fibers may temporarily shut down, possibly as a mental compensatory strategy. This results in progressively less peripheral visual information reaching the brain as clutter increases, potentially rendering the individual unable to perceive anything meaningful. This could explain the dynamic narrowing of the attentional visual field, the 'pop-out' effect beyond the central visual area, and why adding a filter reduces visual stress by lowering the system's cognitive load. Given that reducing visual input through a fog-like filter appears to lessen the demand on the mapping system, it is worth considering whether spectacles with peripheral masking could potentially help children with CVI. This approach could be particularly useful in low-movement environments, such as classrooms or while watching television, where unnecessary visual clutter could be minimized, thereby reducing the negative impact of incoming unnecessary clutter and complexity.

The challenges observed in these case studies, along with findings from Hokken et al. (2024), suggest that the peripheral field of vision in individuals with simultanagnosia is dynamic, often significantly impairing vision, as demonstrated by all three cases. This dynamic nature possibly affects the formation of visual memories. For instance, Case 2's inability to recognize faces or facial expressions is a common issue in CVI (Bauer et al., 2023). This difficulty may stem from an inability to perceive an entire face, as many children with CVI anecdotally report that they can only see parts of a face. Further research is required to better understand this phenomenon. This differs from prosopagnosia, a facial recognition disorder resulting from temporal lobe dysfunction, which is typically associated with ventral stream impairment. Although prosopagnosia and CVI-related facial recognition difficulties may co-occur (Thomas et al., 2009). Simultanagnosia and its potential complexities might contribute to facial recognition deficits through mechanisms distinct from those involved in ventral stream dysfunction. Prosopagnosia has also been reported in cases of Balint syndrome (Rosselli et al., 2001), suggesting overlapping but not necessarily identical patterns of impairment.

Deficits in encoding the spatial coordinates of facial features in developmental prosopagnosia (Barton et al., 2003) bear similarities to the deficits reported for simultanagnosia concerning the binding of visual attributes of objects (Robertson et al., 1997). However, it is important to recognize that these similarities do not necessarily imply direct causality between dorsal stream dysfunction and ventral stream processes. Rather, these deficits may reflect challenges in inter-stream connectivity and integration within broader visual processing networks, which involve both vertical and horizontal connections (Robertson et al., 1997).

While simultanagnosia may contribute to impairments in facial recognition through disruptions in visual attention and spatial integration, current evidence does not support a direct influence of dorsal stream dysfunction on ventral stream function. This underscores the need for further research to elucidate how distinct visual pathways interact and compensate for each other, particularly in cases where processing within a specific region deviates from typical functioning. Future studies focusing on the interaction between spatial and object pathways could offer insights into the neural mechanisms underlying these co-occurring deficits.

In clinical practise, CVI can be associated with isolated pathology within the posterior parietal lobes, while the temporal lobes remain unaffected. This pattern suggests the possibility of isolated DSD. For instance, in their review, Dutton and Jacobson (2001) describe how lesions in the posterior parietal lobes can lead to deficits in spatial processing and consequently, visual attention, without impairing ventral stream functions such as object recognition. Similarly, Pehere et al. (2018) in their overview of the neural pathways involved in CVI, also describe instances in which parietal lobe damage was associated with deficits in spatial awareness and visual attention, while also sparing ventral stream functions. Michel and Henaff (2004) further explore simultanagnosia resulting from posterior parietal lesions, demonstrating that such damage disrupts the ability to perceive multiple objects simultaneously, a key function of the dorsal stream, while preserving ventral stream capabilities like object recognition. In contradistinction, persistent ability to see movement and move accurately through three dimensional space, despite bilateral loss of the occipital and temporal lobes (the Riddoch phenomenon), potentially highlights the nature of dorsal stream function when the middle temporal lobes (serving movement perception) and posterior parietal lobes remains intact (Arcaro et al., 2019). These studies underscore a clear distinction between the roles of the dorsal and ventral streams in visual processing. Damage specific to the posterior parietal lobes can lead to impairments in spatial and attentional processing while preserving ventral stream functions and vice versa. Understanding this distinction is crucial for accurately identifying and supporting individuals with CVI, particularly those with isolated DSD, as it provides a foundation for more tailored interventions based on the type and location of the underlying neural damage or dysfunction.

Understanding simultanagnosia reveals it to be a highly nuanced and individualized condition. One emerging insight is the role of the 'pop-out' effect, which may be influenced by personal preferences. For example, Case 1 was particularly drawn to teddy bears and the color blue, while Case 3 preferred bright colors like pink and had a strong affinity for dogs. This suggests that recognizing the individual's preferences may offer opportunities to minimize distracting "pop-out" stimuli or, conversely, leverage them to aid in locating objects. Research from Hokken et al. (2024), using eye-tracking technology, further connects the adverse effects of environmental complexity to the dynamic nature of the attentional visual field in simultanagnosia. By testing vision with specific targets rather than relying on naturalistic descriptions, as seen in tests like the Birthday Party Test (de Vries et al., 2022), the Hokken et al. approach offers a more controlled way to study these visual processing challenges. Combining both approaches, targeted and naturalistic, may provide deeper insights into the complexities of simultanagnosia.

In 2020, the University of St Andrews and CVI Scotland shared some of the findings, including the video *Beach (C),* along with a carefully explained description of the exploratory nature of the findings and their limitations. A short video summarizing the findings was also part of this resource (University of St Andrews, 2024). The public's response to this material, was overwhelmingly positive. The short video, in particular, seemed to resonate with many viewers, including adults who identified CVI in themselves after watching it. Examples of community feedback included: "*OMG! You nailed it! That's exactly what it's like. Bravo!*," "*At last, I can show everyone my child's world so they can understand better… now I can show them!!!*," "*LOOK!!! …this is what it would feel like to walk in my shoes*." The success of this process laid the groundwork for a subsequent clinical trial to test these theories, including the creation of a control group.

Currently, there are no standardized measurements for simultanagnosia or other higher visual processing difficulties associated with CVI, despite the likelihood that the severity of these impairments varies between individuals. It may be feasible to develop measurements targeting the various elements that comprise simultanagnosia. For example, the "window of vision," which appears to be dynamic, could be quantified as a percentage of the total visual field, potentially capturing its range from largest to smallest. Such measurements could allow for tracking improvements over time. In the case of Case 1, vision seemed to improve through their 'conscious vision strategies', but it remains unclear whether the visual field itself expanded, she better tolerated difficulty, or gained more control over the dynamic aspects or 'pop-out' effects.

An inability to locate a pointed-out target can manifest as a child appearing unable to move their eyes toward it, a condition known as apraxia of gaze, an element of dorsal stream dysfunction (DSD). Traditionally, apraxia of gaze and simultanagnosia have been described as separate components of DSD. However, considering findings from studies where individuals were asked to identify a target in a visual scene (Bennett et al., 2018, 2020; Hokken et al., 2024), it is plausible that apraxia of gaze may be a consequence of simultanagnosia rather than a distinct visual impairment. If a person is unable to perceive a target, they cannot move their gaze toward it. This could explain instances where gaze becomes 'stuck,' as seen with Case 1 and the hoops, suggesting that apraxia of gaze might be an effect of simultanagnosia's interference with visual processing, rather than a separate condition.

Identifying the underlying cause of these difficulties is crucial to ensuring that interventions target the cause of the visual impairments rather than its symptoms. Identifying the underlying cause of these difficulties is crucial to ensuring that interventions target the cause of the visual impairments rather than just their symptoms. A CVI question inventory, validated by MacIntyre-Beon et al. (2012) and recently described using the acronym "CVI-I" for Cerebral Visual Impairment Inventory by Chandna et al. (2024), was designed to capture a range of visual behaviors associated with CVI in children.

Although the 2012 paper does not explicitly differentiate between types of CVI, the inventory provides insights into various CVI-related visual processing challenges, which have helped clinicians recognize different profiles of CVI presentations in practice.

Without concrete metrics, it is difficult to assess progress or identify where adjustments to support strategies are needed. Although the Hellgren et al. (2020) study identified visual perceptual challenges in preterm children rather than specifically diagnosing CVI, it used a control group and the CVI-I (MacIntyre-Beon et al., 2012; Chandna et al., 2024) to compare visual function across 39 areas. This approach could be adapted to develop percentage measurements for simultanagnosia, allowing for better differentiation of symptoms and treatment outcomes. Recent work by Hay et al. (2020) extended this methodology, demonstrating a direct correlation between total inventory scores and standard VMI scores for a subset of children with CVI followed over 6 years. This study used age-inappropriate configural disruption in copied drawings as evidence of "local capture" (Dalrymple et al., 2007, 2009; Karnath et al., 2000) and employed VMI standard scores to quantify the severity of simultanagnosia. Furthermore, studies specifically focusing on CVI diagnoses using validated inventories provide valuable insights. For instance, Ortibus et al. (2011) examined the screening value of a CVI questionnaire, while van Genderen et al. (2012) focused on diagnosing CVI in children with good visual acuity. Tsirka et al. (2020) explored the clinical application of the Insight Inventory in CVI and its effectiveness in guiding tailored habilitation strategies. New findings from Chandna et al. (2021) emphasized higher visual function deficits in children with CVI who have good visual acuity, a theme further explored in their 2024 study, which highlighted that these deficits can manifest independently of visual acuity measures (Chandna et al., 2024). These studies collectively underscore the need for validated tools to accurately identify and address the diverse presentations of CVI, particularly in cases where symptoms are not solely linked to reduced visual acuity.

6 Conclusion

The case studies presented in this work highlight the dynamic and individualized nature of dorsal stream dysfunction (DSD) in individuals with cerebral visual impairment (CVI). Our findings suggest that the symptoms of simultanagnosia extend beyond the traditionally understood limitation of perceiving one or two objects at a time. Instead, we observed a dynamic constriction of the attentional visual field, which fluctuates based on the complexity of the environment. This nuanced understanding of simultanagnostic vision underscores the need for a more refined approach to both diagnosis and intervention.

The concept of a dynamic "window" of visual attention, influenced by both internal factors (such as anxiety) and external complexity, presents new avenues for targeted therapeutic strategies. Interventions that reduce visual clutter and complexity, or leverage personal preferences, may help optimize visual functioning and reduce fatigue. These insights could be particularly beneficial for children at sensitive developmental stages, where early and effective interventions are crucial.

Furthermore, our findings highlight the importance of exploring how different visual processing streams in the brain interconnect and compensate for each other. Understanding these interactions may provide deeper insights into the variability of CVI symptoms and open pathways for developing more tailored support strategies. Future research should focus on the neural mechanisms underlying these dynamic visual experiences and explore potential interventions, such as adaptive visual filters or environmental modifications, to enhance quality of life for individuals with DSD.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

Ethical approval was not required as the activities described were not initially conducted as a research study but rather as part of a practical application or demonstration. Consequently, the activities fell outside the scope of research requiring ethical approval under local legislation and institutional guidelines. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

HS: Conceptualization, Investigation, Methodology, Visualization, Writing – original draft, Writing – review & editing. NM: Conceptualization, Investigation, Methodology, Writing – review & editing. GD: Conceptualization, Investigation, Methodology, Writing – review & editing. JR: Conceptualization, Investigation, Methodology, Writing – review & editing. IH: Conceptualization, Investigation, Methodology, Writing – review & editing. AB: Conceptualization, Methodology, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2024.1496811/full#supplementary-material

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EDITED BY
Corinna M. Bauer,
Massachusetts Eye & Ear Infirmary and
Harvard Medical School, United States

REVIEWED BY
Gerard Dumancas,
North Carolina Agricultural and Technical
State University, United States
Kathleen Vancleef,
Durham University, United Kingdom

*CORRESPONDENCE
Melinda Y. Chang
✉ melinda.y.wu@gmail.com

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Method for assessing visual saliency in children with cerebral/cortical visual impairment using generative artificial intelligence

Kate Matsunaga[1], Kleanthis Avramidis[2], Mark S. Borchert[1,3], Shrikanth Narayanan[2] and Melinda Y. Chang[1,3]*

[1]Keck School of Medicine, University of Southern California, Los Angeles, CA, United States, [2]Viterbi School of Engineering, University of Southern California, Los Angeles, CA, United States, [3]Division of Ophthalmology, Department of Surgery, Children's Hospital Los Angeles, Los Angeles, CA, United States

Cerebral/cortical visual impairment (CVI) is a leading cause of pediatric visual impairment in the United States and other developed countries, and is increasingly diagnosed in developing nations due to improved care and survival of children who are born premature or have other risk factors for CVI. Despite this, there is currently no objective, standardized method to quantify the diverse visual impairments seen in children with CVI who are young and developmentally delayed. We propose a method that combines eye tracking and an image-based generative artificial intelligence (AI) model (SegCLIP) to assess higher- and lower-level visual characteristics in children with CVI. We will recruit 40 CVI participants (aged 12 months to 12 years) and 40 age-matched controls, who will watch a series of images on a monitor while eye gaze position is recorded using eye tracking. SegCLIP will be prompted to generate saliency maps for each of the images in the experimental protocol. The saliency maps (12 total) will highlight areas of interest that pertain to specific visual features, allowing for analysis of a range of individual visual characteristics. Eye tracking fixation maps will then be compared to the saliency maps to calculate fixation saliency values, which will be assigned based on the intensity of the pixel corresponding to the location of the fixation in the saliency map. Fixation saliency values will be compared between CVI and control participants. Fixation saliency values will also be correlated to corresponding scores on a functional vision assessment, the CVI Range-CR. We expect that fixation saliency values on visual characteristics that require higher-level processing will be significantly lower in CVI participants compared to controls, whereas fixation saliency values on lower-level visual characteristics will be similar or higher in CVI participants. Furthermore, we anticipate that fixation saliency values will be significantly correlated to scores on corresponding items on the CVI Range-CR. Together, these findings would suggest that AI-enabled saliency analysis using eye tracking can objectively quantify abnormalities of lower- and higher-order visual processing in children with CVI. This novel technique has the potential to guide individualized interventions and serve as an outcome measure in future clinical trials.

KEYWORDS

cortical visual impairment, cerebral visual impairment, eye tracking, generative artificial intelligence, functional vision assessment

1 Introduction

Cerebral/cortical visual impairment (CVI) is a leading cause of pediatric visual impairment in developed countries, and is increasingly diagnosed in developing nations due to improved care and survival of children who are born premature or have other risk factors for CVI, such as hypoxic–ischemic encephalopathy (Chang and Borchert, 2020; Chang and Borchert, 2024; Kong et al., 2012; Chong et al., 2019; Pehere et al., 2019). Although the United States does not currently have a registry of children with visual impairment, a recent study from Denmark found that the prevalence of visual impairment was 1.6 per 1,000 children <18 years of age, with CVI accounting for 36% of children with visual impairment in 2022 (Kessel et al., 2024). In India, CVI was diagnosed in 44% of young children with profound visual impairment seen at a tertiary eye center (Pehere et al., 2019).

CVI encompasses a wide range of visual impairments due to structural and/or functional brain abnormalities that affect visual pathways in the developing brain (Chang et al., 2024). While children with CVI may have comorbid ocular conditions, the visual dysfunction is worse than expected for the degree of ocular pathology (Chang et al., 2024; Sakki et al., 2018). Causes of CVI include hypoxic–ischemic encephalopathy in children born at term, seizures with epileptic encephalopathy, prematurity with periventricular leukomalacia, hydrocephalus, meningoencephalitis, trauma, metabolic and genetic disorders, among others (Chang and Borchert, 2020; Chang et al., 2022; Huo et al., 1999). A wide variety of genetic disorders have been associated with CVI, including Down syndrome, Bosch Boonstra Schaaf optic atrophy syndrome, tuberous sclerosis, Angelman syndrome, and many others (Chang and Borchert, 2020). In the case of these neurogenetic syndromes and other causes of CVI, there may be overlap with other neurodevelopmental conditions such as autism spectrum disorder (ASD). However, the recent working definition of CVI from the National Institutes of Health (NIH) CVI Workshop specifies that "while CVI may be comorbid with other neurodevelopmental disorders, CVI is not primarily a disorder of language, learning, or social communication" (Merabet et al., 2017). Although children with ASD are known to have abnormal visual social attention which can be measured with eye tracking (Falck-Ytter et al., 2013), children with CVI have additional deficits of visual function and processing that cannot be attributed to ASD alone.

Due to involvement of visual pathways in the brain, children with CVI often exhibit visual characteristics that differ from those with purely ocular disorders (Chang and Borchert, 2020; Jan et al., 1987). Similar to children with ocular causes of visual impairment, children with CVI may have abnormalities of lower-order visual function (aspects of vision that are processed in the visual pathway from the eye to the primary visual cortex, such as visual acuity and contrast sensitivity) (Good, 2001; Good et al., 2012). However, children with CVI are unique in that they may have impairments of higher-order visual processing (localized to visual association areas in the brain) (Dutton et al., 1996). Examples include abnormalities of dorsal stream processing leading to dysfunction in visuospatial orientation and complex motion perception, as well as disorders of recognition such as prosopagnosia (difficulty with recognizing faces) (Chang and Borchert, 2020; Dutton et al., 1996, 2006; Ahmed and Dutton, 1996; Chandna et al., 2021a, 2021b; Bauer et al., 2023; Dutton, 2009; Ortibus et al., 2012). Children with CVI also have challenges with visual search and recognition of objects against complex backgrounds (Jan et al., 1987; Manley et al., 2022). While lower-order visual deficits may be elicited by standard pediatric ophthalmologic examination techniques, diagnosis of higher-order visual deficits often necessitates neuropsychological assessment, which generally requires children to have a developmental age of at least 3 years in order to have the cognitive ability to understand and respond to questions about visual perception.

Currently, there is no objective test that measures the diverse visual deficits that occur in CVI and is applicable to young, developmentally delayed children (Chang and Borchert, 2021). We propose a method to assess visual attention to lower- and higher-order visual characteristics in children with CVI by utilizing eye tracking technology combined with artificial intelligence (AI) for saliency analysis. Eye tracking has previously been used to assess visual acuity in young children with CVI, as well as visual search in older individuals with CVI (Chang and Borchert, 2024; Manley et al., 2022). Additionally, eye tracking has previously been used in children with CVI to assess visual orienting functions, especially reaction time to fixation when presented targets of interest such as cartoon faces and moving dots (Ben Itzhak et al., 2023). In the present study, we will assess eye tracking patterns using saliency analysis, a computer vision technique to determine which aspects of an image attract a viewer's attention. Saliency analysis in older teenagers and young adults with CVI has demonstrated that visual search patterns are primarily driven by bottom-up, low-level visual features (including color, orientation, and intensity), rather than top-down, higher-level features that are dependent on semantic associations between words and objects (Walter et al., 2024). Because the previous study used saliency models that combine multiple features, there remains a question of which individual visual characteristics are specifically affected in CVI, and to what degree. For this study, we will use an image-based generative artificial intelligence (AI) model, SegCLIP (Luo et al., 2023) to generate saliency maps that highlight individual features of interest (Figure 1) (Avramidis et al., 2024). By comparing eye tracking patterns in children with CVI and age-matched controls to these saliency maps, we will quantify differences in visual attention to lower- and higher-order visual characteristics between the two groups.

The goals of this study are (1) to assess whether eye tracking combined with AI-generated saliency models can quantify differences in visual attention to lower- and higher-order visual characteristics in children with CVI and (2) to correlate eye tracking measures to corresponding scores on the CVI Range-CR functional vision assessment (Chang et al., 2022).

2 Methods

This study has been approved by the local institutional review board (IRB) and will adhere to the Declaration of Helsinki and the US Health Insurance Portability and Accountability Act of 1996. Informed consent will be obtained from the parent or legal guardian of all participants.

2.1 Participants

Children with CVI and age-matched controls between the ages of 12 months and 12 years will be recruited from our pediatric neuro-ophthalmology clinic and a web-based recruitment service

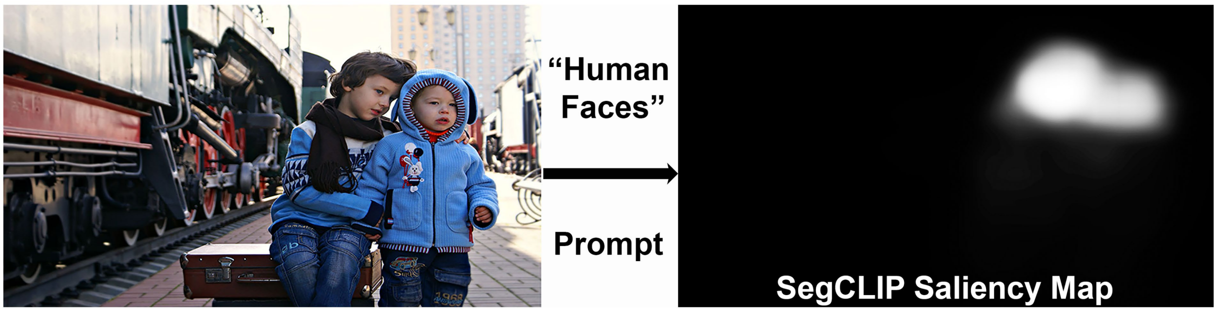

FIGURE 1
Example of a "human faces" saliency map generated by prompting the SegCLIP generative artificial intelligence (AI) model. The saliency map highlights the faces of the children in the image (Reproduced from Victoria Borodinova via https://www.publicdomainpictures.net/en/index.php, licensed under CC0).

(Buildclinical.com). The CVI group will include children who are developmentally delayed and unable to participate in standard neuropsychological assessments of visual perception. The minimum age is 12 months because some practitioners do not diagnose CVI until this age in order to confirm that the child does not have delayed visual maturation (Good et al., 1994). We will include children with a chronological age up to 12 years so that our sample is representative of the age range of children with CVI diagnosed in the community; challenges with measuring higher-order visual processing deficits may lead to CVI being diagnosed late or unrecognized (Williams et al., 2021). Diagnosis of CVI will be based on reduced visual function with a normal eye exam (with the exception of mild optic atrophy) in the context of neurologic pathology affecting structure or function of the optic radiations, primary visual cortex, and/or visual association areas. A neurologic diagnosis will be made in conjunction with a pediatric neurologist based on neurologic examination, neuroimaging, and/or genetic testing, as appropriate. We will allow patients with mild optic atrophy to enroll in the study due to the high proportion of children with CVI in our clinic with this ocular comorbidity (approximately 30%). However, we will perform subgroup analysis to determine whether the inclusion of these patients impacted the study results.

Exclusion criteria for the CVI group will include photosensitive epilepsy, any oculomotor abnormality that would preclude accurate assessment of afferent visual function based on direction of eye gaze (e.g., oculomotor apraxia, the presence of which will be assessed clinically), and binocular visual acuity worse than 3 cycles per degree (cpd) based on preferential looking testing during eye tracking. We chose this threshold for visual acuity because Fourier analysis of the visual stimuli in our experimental protocol revealed that greater than 99% of each image was represented by spatial frequencies above 3 cpd.

The control group will include children with no known neurologic, neurodevelopmental, or visual disorder, other than corrected refractive error. The presence of neurologic and neurodevelopmental disorders will be determined based on medical history, and children born prematurely will be excluded. Controls will be required to have a normal screening eye exam (including age-normal visual acuity) in order to participate.

2.2 Sample size calculation

In our preliminary studies, the saliency characteristic with the least difference between groups was orientation. Fixation saliency values were 48 in the CVI group and 43 in the control group, with a standard deviation of 8. Using these values, with a power of 80% and alpha of 0.05, the sample size required to demonstrate a significant difference between groups is 80 (40 per group). We will aim to recruit 100 participants to account for up to 20% attrition.

2.3 Materials and equipment

2.3.1 Eye tracking

We will use an EyeLink® 1,000 Plus eye tracker (SR Research, Ontario, Canada) to record the direction of eye gaze as X and Y coordinates of each eye at 500 Hz. The Eyelink software automatically identifies fixations and saccades based on the following parameters. Fixations are defined as periods when the gaze is stable within 0.1 degree for at least 100 ms. Saccades are detected when velocity exceeds 30 degrees/s and acceleration exceeds 8,000 degrees/s^2 over an amplitude of greater than 0.1 degree.

During the eye tracking experiment, participants will view a series of still images including naturalistic and cartoon pictures of landscapes, animals, and people displayed on a 24-inch computer monitor. These images are interspersed with psychophysical stimuli to assess other visual characteristics, including visual acuity. Visual acuity will be measured using preferential looking during eye tracking, which we have previously found to be reliable and valid in children with CVI (Chang and Borchert, 2024).

2.3.2 Functional vision assessment (CVI range-CR)

A subset of participants with CVI will undergo functional vision assessment with the CVI Range-CR, a standardized version of the CVI Range developed for clinical research purposes and conducted by a trained neuropsychologist (Chang et al., 2022). Because the CVI Range-CR is only available in English, this assessment will only be performed in children from English-speaking families. Additionally, because the CVI Range-CR assessment is substantially longer than eye tracking (approximately 1 h), some participants who consent to eye tracking are expected to be unable or unwilling to complete the CVI Range-CR assessment. Based on our initial recruitment experience, we expect that approximately 50% of participants who complete eye tracking will be eligible and consent to undergo the CVI Range-CR. This assessment is conducted in a room with a standard configuration, including specific materials placed on the walls (Figure 2) and others used for interactions with the child (Table 1). The CVI Range-CR consists of an interview of parents/caregivers and direct assessment while a child is performing activities to elicit the ability to use vision in everyday life. Children are scored on 10 CVI characteristics, which results in two scores via the "Across-CVI Characteristics Assessment Method" (Score 1) and "Within-CVI Characteristics Assessment Method" (Score 2, Supplementary material). For the purposes of this study, we will use these overall scores as well as scores on individual items of the "Within-CVI Characteristics Assessment Method" (Supplementary material).

2.4 Data acquisition

2.4.1 Eye tracking

During eye tracking, participants will sit (alone, in a stroller, or in a parent's lap) 60 cm from a computer monitor with the eye tracking camera attached at the bottom of the screen. The room lights will be off, and blackout curtains will be placed on the windows. Families will be instructed to avoid distracting the child by speaking, unless necessary to direct gaze to the screen. A target sticker will be placed at the center of the child's brow to facilitate recording. Eye tracking will be performed binocularly, with participants wearing their habitual spectacles. In patients with strabismus and a consistent fixation preference, we will record from the fixating eye. However, if a child has strabismus with alternating fixation, we will patch one eye to prevent switching fixation. After three-point calibration, the eye tracking experiment will commence. The visual stimuli described above (Materials and Equipment section) will be shown on the computer monitor and the participant's direction of eye gaze will be tracked for a total of 10 min. No instructions will be given to the child other than to continue to watch the screen and try to avoid head movement. During the eye tracking session, a warning sound will be played if the eye gaze is detected off the screen. Experimenters will attempt to redirect gaze to the screen verbally or using a toy if needed, and will reposition the head if necessary to regain tracking of the eyes.

2.4.2 CVI range-CR

The CVI Range-CR functional vision assessment will be performed and scored by a neuropsychologist, as described above.

2.5 Data analysis

2.5.1 Saliency maps

In order to assess eye gaze to lower- and higher-order visual characteristics, we will use AI to generate saliency maps of the stimulus images. Compared to other saliency maps that have been used in CVI (Walter et al., 2024), a unique feature of our study is that we will use generative AI to create maps that highlight a single feature of interest, rather than multiple features. Thus, we can assess the degree to which individual lower- and higher-level visual characteristics are impacted in children with CVI. SegCLIP, the generative AI model used in this study, consists of a text and image encoder (Dutton, 2009). It can take free language prompts and an input image to highlight specific regions within the image in the form of saliency maps (Figure 1) (Avramidis et al., 2024). The resulting grayscale maps are then smoothed with a Gaussian filter and normalized such that each pixel is assigned an intensity value from 0 to 255. SegCLIP can also perform differential saliency analysis wherein two prompts with opposite attributes can be inputted in order to enhance the accuracy and nuances of the saliency prediction. Each prompt will initially generate its own saliency map. The subtraction of the two maps will generate the final saliency map used for the

FIGURE 2
Standard room configuration for the CVI Range-CR functional vision assessment. Reproduced from Chang et al. (2022), with permission from BMJ Publishing Group Ltd.

TABLE 1 Materials used for direct assessment in the CVI Range-CR.

Flashlight with red light
Brightly colored strings of beads of various colors ("Mardi gras" beads)
Translucent neon Slinky toys
Clear plastic colored blocks
A dozen plastic animals (approximately 1″ height)
Two stuffed animals (10″ height): red Elmo and yellow Big Bird
Musical toy (e.g., Fisher-Price BeatBo)
Black tray
Colorful patterned fabric
Picture book (see description in text)
Light box
CVI Complexity Sequence Cards (American Printing House, Louisville, KY)
iPad with apps (Tap-N-See Now, CVIHumanFace)
Handheld mirror (8.5×11″)
Dimmable floor lamp

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differential analysis (Avramidis et al., 2024). Each saliency map will be manually checked for accuracy to ensure appropriate areas are highlighted.

The saliency maps that are currently planned include: color (red, yellow, green, and blue); visual field maps (central vs. peripheral screen, upper vs. lower screen, and right vs. left screen); luminance or intensity; contrast; orientational patterns (prompted as "bars" or "stripes"); background; depth; animals; human faces; human bodies; movement; and complexity.

Additionally, we will create saliency maps using DeepGazeII, a saliency model that predicts salient object fixations using a deep neural network pretrained on object recognition via the SALICON dataset, which includes 10,000 images annotated by humans (Kümmerer et al., 2016). The DeepGazeII model predicts the fixations of (presumably) typically developing adults with high accuracy (AUC 0.88). Thus, we will use DeepGazeII saliency maps as an indicator of typical adult visual attention.

2.5.2 Comparison of CVI and control participants

Fixation maps of CVI and control participants will be combined with saliency maps to calculate fixation saliency values, as described below. Subsequently, fixation saliency values will be compared between CVI and control participants.

For each image viewed during each eye tracking recording session, fixations will be identified by the eye tracking software as per the above specifications (gaze within 0.1 degree for a minimum of 100 ms). The SR Research Eyelink software uses a heuristic filter to reduce noise (Stampe, 1993). Fixations will be discarded if they are located at least 20% out of the stimulus image range. Trials without valid fixations will be discarded. If both eyes are recorded, the position of the two eyes will be averaged to calculate a composite fixation position. The center of each fixation will be mapped to the corresponding pixel location in the saliency map of interest, and the intensity of this pixel will be designated the fixation saliency value (Figure 3). We will then calculate the average fixation saliency value per image for each participant. Finally, for each saliency characteristic, we will compare fixation saliency values in CVI participants and controls using Mann–Whitney tests. We will also perform multivariate regression to assess the effects of age and neurologic and ophthalmologic comorbidities on fixation saliency values. We will perform subgroup analysis excluding participants with ophthalmologic conditions such as optic atrophy and nystagmus to assess the impact of these factors on our results. We will also perform subgroup analysis by both chronological and developmental age.

2.5.3 Correlation of CVI fixation saliency values to CVI range-CR scores

In children with CVI, we will use Spearman's correlation coefficient to evaluate the relationship between fixation saliency values and scores of corresponding items on the CVI Range-CR in children with CVI. Specifically, fixation values on the color saliency maps will be correlated to the "color preference" item on the CVI Range-CR. Similarly, fixation values on the visual field saliency maps will be correlated to the "visual field preference" item of the CVI Range-CR. Fixation values on the complexity saliency map will be correlated to scores on the "difficulties with visual complexity" CVI Range-CR item. We will also correlate fixation values on the luminance saliency map with the "need for light" item on the CVI Range-CR. Finally, we will correlate fixation saliency values on the DeepGazeII saliency maps (which indicate visual attention in typical adults) to overall CVI Range-CR scores (Score 1 and Score 2). Correlations will be performed using the whole dataset as well as subgroups divided by age.

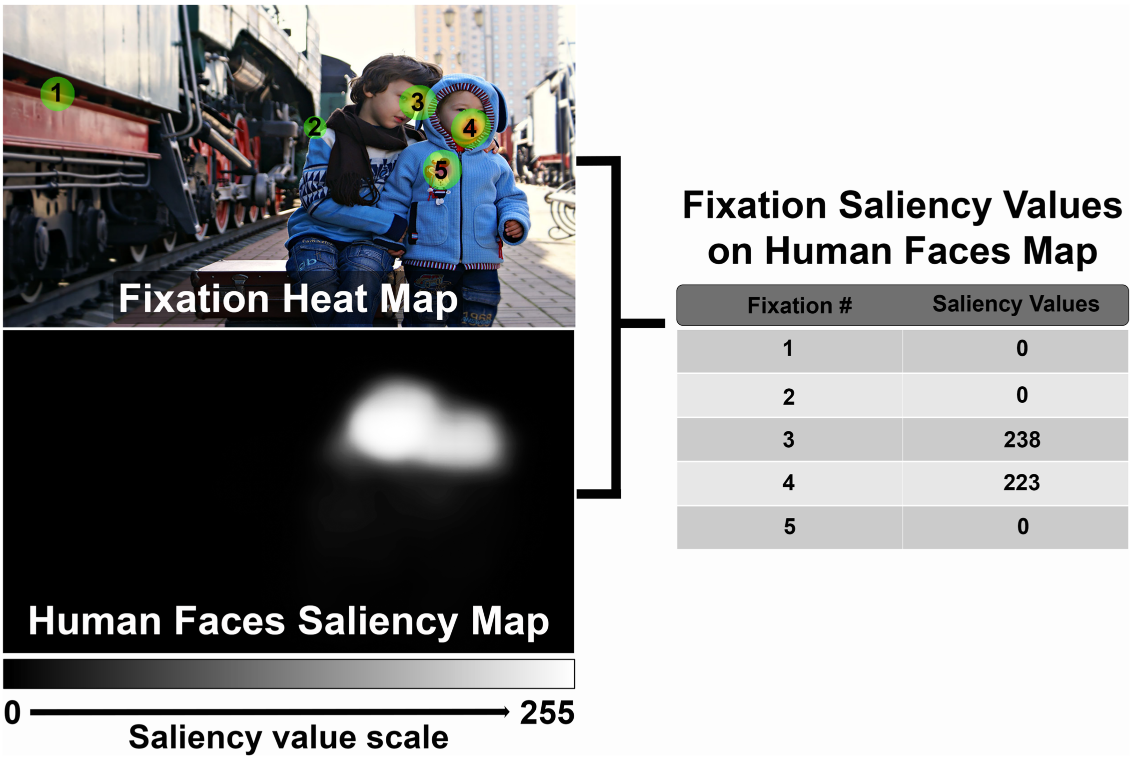
3 Anticipated results

3.1 Saliency maps

We anticipate that by applying adequate prompting engineering, we will be able to generate saliency maps using SegCLIP that accurately represent the visual features that we are investigating. Our preliminary saliency maps are pointing toward this direction (Figure 1). We will continue to refine our prompts to generate appropriate maps for the 12 visual characteristics selected for this study.

3.2 Comparison of CVI and control participants

We expect that our data will demonstrate significant differences between children with CVI and controls. Specifically, we expect that children with CVI will have lower fixation saliency values for visual characteristics that require higher-order visual processing, including still depictions of motion, two-dimensional representations of depth, human faces and bodies, animals, and visually complex scenes. Our preliminary data suggest that fixation saliency values on the "depth" saliency map are significantly lower in children with CVI compared to controls (Figure 4). We also expect that children with CVI will exhibit greater attention to lower-order visual characteristics, indicating greater reliance on visual features that do not require higher-level processing. Specifically, we anticipate that children with CVI will have higher fixation saliency values on maps of color (especially red and yellow), contrast, orientation,



Fixation Saliency Values on Human Faces Map

Fixation #	Saliency Values
1	0
2	0
3	238
4	223
5	0

FIGURE 3
Example demonstrating method to calculate fixation saliency values. See text for full description (Reproduced from Victoria Borodinova via https://www.publicdomainpictures.net/en/index.php, licensed under CC0).

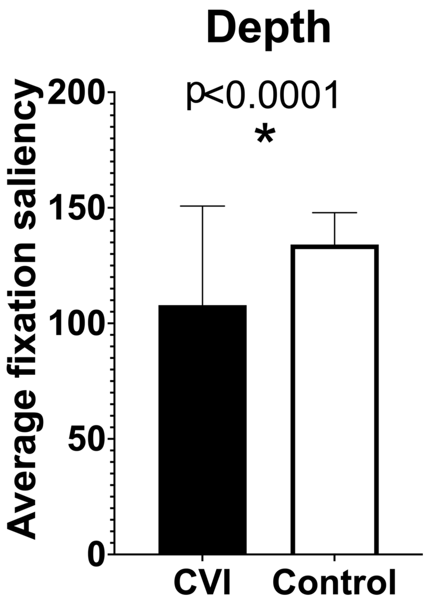


FIGURE 4
Preliminary results comparing fixation saliency values in children with cerebral/cortical visual impairment (CVI) and age-matched controls on "depth" saliency maps.

and luminance. Due to eccentric gaze preference and possible visual field defects, children with CVI are also expected to have higher fixation saliency values on peripheral visual field maps. We also anticipate that children with CVI will have difficulty identifying the subject of each image, so we expect that they will have higher fixation saliency values on the background saliency map. On subgroup analysis by age, we hypothesize that older children may have less severe abnormalities in fixation saliency values compared to younger children, based on prior studies reporting improvement in visual behavior over time in children with CVI (Handa et al., 2018).

3.3 Correlation of CVI fixation saliency values to CVI range-CR scores

In children with CVI, we anticipate that fixation saliency values will correlate significantly to scores on corresponding items on the CVI Range-CR. Further, we expect that fixation values on the DeepGazeII saliency map will demonstrate significant correlations to the CVI Range-CR overall scores (Score 1 and Score 2).

3.4 Advantages

Our saliency modeling approach to evaluate eye fixations has multiple advantages. The approach is quantitative, objective, and scalable, since eye tracking can be performed by technicians or research assistants in approximately 15 min (depending on the child's cooperation). This is in contrast to functional vision assessments that require trained personnel and may last an hour or more. Because the children are not required to do any task other than watch the computer monitor (free viewing), the method is applicable to young, developmentally delayed children with CVI who are cognitively or physically unable to participate in neuropsychological testing. Additionally, this method takes advantage of generative AI, which can adapt and learn complex or higher-level patterns to generate predictions for subtle features that might otherwise be overlooked (Kümmerer et al., 2016). Therefore, the saliency maps that we generate for this project may improve and become more discriminative of CVI patients compared to controls over time. To ensure that the models remain representative of salient features, we can monitor updates in generative models, re-training and re-evaluating by experts as needed. Additionally, this method is scalable and can incorporate any number of image stimuli for any feature of interest, avoiding costly annotation procedures that would otherwise be done manually.

3.5 Limitations

The primary limitation of eye tracking methods in children with CVI is that a minimum level of visual acuity is required to view the stimuli on the computer monitor. We will only include patients with binocular visual acuity of at least 3 cpd, based on Fourier analysis of the images in our experimental protocol (see Participants section above). Thus, our study may not be applicable to children with worse visual acuity (i.e., grating acuity lower than 3 cpd). Since we will exclude participants with oculomotor apraxia, our results may also be inapplicable to children with this condition. Oculomotor apraxia has been reported in 15% of children with CVI (Huo et al., 1999). Additionally, the visual stimulus and testing conditions involving two-dimensional images on a computer monitor in a laboratory may not fully simulate real-life situations. Furthermore, we will use the CVI Range-CR to validate our eye tracking findings, but the validity of CVI Range-CR is still under investigation (Chang et al., 2022). We chose to compare eye tracking to CVI Range-CR scores since there are no other direct assessments for children with CVI that quantify the lower- and higher-order visual characteristics evaluated in this study and are applicable to our population of young, developmentally delayed patients. When there is no gold standard reference test, comparison to other relevant clinical tests or characteristics is an acceptable alternative for preliminary validation (Rutjes et al., 2007).

Another limitation relates to the use of generative AI to create the saliency maps. These AI models can "hallucinate" and generate outputs that may be inaccurate. Therefore, the generated maps require human review to confirm that they are consistent with the visual characteristics that are prompted. Careful attention to prompt engineering and some degree of trial-and-error may be required to generate accurate saliency maps for all characteristics.

Furthermore, we will screen typically developing control participants for neurologic and neurodevelopmental conditions based on birth and medical history. We cannot exclude the possibility that some of these children may have undiagnosed neurodevelopmental conditions such as autism or attention deficit hyperactivity disorder (ADHD). However, they will have a screening eye exam to confirm normal visual function.

Finally, patterns of eye tracking are indicative of visual attention, and we will infer deficits of visual processing when there is decreased visual attention to features that require specific levels of visual processing. However, since we anticipate that all of our CVI participants will be non-verbal or minimally verbal, we will be unable to confirm whether decreased attention is due to abnormalities of visual perception or other factors. For example, children with autism exhibit decreased visual attention to social stimuli on eye tracking (Chita-Tegmark, 2016), which is believed to be related to social rather than visual deficits. Future eye tracking studies in individuals with CVI with greater communication abilities may help to clarify the interpretation of our results.

3.6 Potential pitfalls and alternate approaches

Eye tracking in patients with strabismus requires some modifications. A trained examiner must determine the fixating eye and further assess whether fixation alternates, in order to determine which eye to record from and whether monocular occlusion is necessary (in the case of alternating fixation). Monocular occlusion may not be possible in some children with CVI. If monocular occlusion is needed, we will first attempt this using an adhesive eye patch. If the child wears glasses, occlusion of one lens is an option. If they cannot tolerate this, we will request that that parent cover one eye with a hand, being careful not to allow peeking. If these measures fail, then the child will be excluded from our study.

Furthermore, nystagmus may interfere with accurate detection of gaze direction. Nystagmus in CVI patients generally occurs only in the presence of anterior visual pathway disease (Whiting et al., 1985). Since the only intraocular comorbidity that we are allowing for this study is optic atrophy, we anticipate that only a minority of participants will have nystagmus. Our experience is that if nystagmus amplitude and frequency are low enough to enable calibration, then we can accurately assess the location of fixations during the eye tracking recording (the timing of fixations and saccades, however, will not be reliable). Therefore, we will include patients with nystagmus who are able to complete calibration in this study. We will perform subgroup analysis excluding participants with nystagmus to determine the impact of nystagmus on saliency results.

4 Discussion

Eye tracking with interpretation enabled by AI-generated saliency maps has the potential to serve as a quantitative and objective metric of attention to lower- and higher-order visual characteristics in children with CVI. Fixation saliency values may potentially be used in the future for longitudinal assessments and guidance of personalized interventions. For example, if a color preference is identified based on high fixation saliency values on a certain color saliency map, then the family may be suggested to use objects of this color to encourage visual behavior. Additionally, because this technique is scalable due to minimal time and personnel requirements compared to other methods of visual assessment in CVI, eye tracking is an ideal candidate to serve as an outcome measure in future multi-center clinical trials to identify evidence-based medical treatments. Finally, successful application of eye tracking combined with generative AI for visual assessment in CVI could lead to adoption of this technique in other neurodevelopmental disorders.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by Children's Hospital Los Angeles IRB. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

KM: Writing – original draft, Writing – review & editing. KA: Conceptualization, Methodology, Writing – review & editing. MB: Conceptualization, Writing – review & editing. SN: Conceptualization, Methodology, Supervision, Writing – review & editing. MC: Conceptualization, Funding acquisition, Investigation, Methodology, Resources, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Generative AI statement

The authors declare that no Gen AI was used in the creation of this manuscript.

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Supplementary material

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‘We don’t know nearly enough’: an online survey exploring perspectives of specialists who support children with brain-based visual impairments

Josephine Sabrina Jakubowski[1], Eloise May[2], Rebecca Findlay[3,4], Nicola McDowell[5], Samantha K. Simkin[3,4] and Lisa M. Hamm[3]*

[1]Department of Medicine, Queen’s University, Kingston, ON, Canada, [2]School of Nursing, University of Auckland, Auckland, New Zealand, [3]School of Optometry and Vision Science, University of Auckland, Auckland, New Zealand, [4]Blind and Low Vision Education Network New Zealand (BLENNZ), Auckland, New Zealand, [5]Institute of Education, Massey University, Auckland, New Zealand

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EDITED BY
Melinda Chang,
Children’s Hospital of Los Angeles,
United States

REVIEWED BY
Zahide Pamir,
Bilkent University, Türkiye
Mohammad Mofatteh,
Queen’s University Belfast, United Kingdom

*CORRESPONDENCE
Lisa M. Hamm
✉ l.hamm@auckland.ac.nz

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Introduction: Children with brain-based visual impairments (some of whom have a diagnosis of Cerebral Visual Impairment, or ‘CVI’) represent a growing and underserved population within vision services. These children often have more complex needs than those with ocular visual impairments and benefit from specialist support from multiple disciplines. This study aimed to understand the perspectives of these specialists in terms of their goals, views on collaboration, and understanding of the term ‘CVI’.

Methods: We invited a range of specialists who work with children with brain-based visual impairments, including educators, rehabilitation staff, clinicians, and family members, to complete an online survey between April 2023 and April 2024.

Results: The analysis included 94 respondents: 51 educators, 30 rehabilitation staff, 7 clinicians, and 6 family members. Respondents shared common goals of connecting with the child (87/94, 93%) and fostering their learning and development (82/94, 93%). However, respondents also noted some specific and divergent goals, which can be at odds with each other. Professional staff frequently identified family members as the most valuable source of information about their child’s vision (36/88, 41%), though family members expressed feeling under-valued. Transdisciplinary clinics were highlighted as a helpful model to provide quality child-centered care. Of the 73 professional staff who reported being familiar with the term ‘CVI’ (73/88, 83%), most (61/73, 84%) thought it was underdiagnosed, but respondents had different perspectives on what a diagnosis meant. Only 73% of professionals familiar with CVI reported receiving formal training about it.

Discussion: The varied goals and different perspectives on CVI create challenges to providing cohesive support for children with brain-based visual impairments. Increasing the availability of complementary formal training across disciplines and adopting transdisciplinary models of care are promising approaches to improve the quality of services.

KEYWORDS

brain based visual impairment, CVI, cerebral visual impairment, parent perspectives, survey, transdisciplinary care

1 Introduction

When low vision originates within the eye, it typically limits the visual information accessible to a child, manifesting as reduced visual acuity, diminished contrast sensitivity, or restricted visual fields. When visual challenges originate in the brain, these foundational elements of vision may be limited, but impairments can also alter visual attention, how visual information is interpreted and how it is integrated with other sensory inputs (Lueck et al., 2019; Philip and Dutton, 2014). These challenges are harder to measure and articulate, so they can lead to misunderstandings, and necessitate different approaches to low vision support (Brown et al., 2024; McDowell, 2023; McDowell and Budd, 2018). Although strategies and frameworks are being developed to understand what children with brain-based visual impairments see, and how to best support these children (McDowell, 2023; Pilling et al., 2023; Zatta and Willems, 2024), there are not yet standardized guidelines (Pilling et al., 2023).

Terminology is also an area of ongoing debate (Chang et al., 2024; Costa, 2024; Kran et al., 2019; Pilling et al., 2023; Sakki et al., 2018), with 'cortical' and 'cerebral' commonly used, both shortened to 'CVI'. Here, we use the term 'brain-based visual impairments' broadly to include children who struggle to process visual information but may not have a formal diagnosis, and 'CVI' more specifically to refer to a diagnosed condition. Recent collaborative initiatives to standardize terminology (Chang et al., 2024) and develop assessment protocols (Zatta and Willems, 2024) are essential and timely, given the growing impact of brain-based impairments on childhood low vision and blindness. In 2000, CVI was the cause of 50% of severe visual impairment and blindness in UK children, rising to 61% by 2015 – a trend echoed in other high-income countries (Teoh et al., 2023).

Brain-based visual impairments rarely occur in isolation; children may have had congenital or acquired brain injuries, neurological malformations, and/or genetic conditions, resulting in co-occurring motor, communication and cognitive challenges (Philip and Dutton, 2014). Whether the manifestations of CVI are co-occurring with, critical components of, or causative factors for other neurodevelopmental conditions is another area of ongoing debate (Chokron and Dutton, 2023; Chokron et al., 2020). Given the co-occurring challenges, children living with brain-based visual impairments benefit from the support of a range of specialists, including ophthalmologists, pediatricians, rehabilitation therapists, educators, community support networks and their families. Each specialist requires a comprehensive understanding of each child's strengths and challenges, as well as how they use their vision day to day to reach their goals, however approaches to understanding what a child sees can be very different (Bennett et al., 2019; Silveira, 2019).

In this study we sought to explore perspectives of specialists who support children with brain-based visual impairments. We aimed to understand (1) the goals specialists have when trying to understand what children with brain-based visual impairments see, (2) perceptions about the flow of information between specialists, and (3) how specialists understand the term 'CVI'.

2 Methods

2.1 General

The cross-sectional survey was conducted online using Qualtrics (Qualtrics, 2022; Utah, USA), with recruitment (by email invitation) between April 2023 and April 2024. Qualtrics allows for IP address checking to prevent reporting of duplicate responses. The study is reported using relevant items of CROSS (Sharma et al., 2021) and STROBE (Von Elm et al., 2009) guidelines.

2.2 Pre-testing

The survey was piloted by multiple stakeholders with different roles, including caregivers of children with brain-based visual impairment, educators, rehabilitation and clinical staff, and researchers. Feedback was incorporated into the survey iteratively. Key changes included a broadening of scope (resulting in findings presented across two manuscripts) and modifying terminology to reflect international differences.

2.3 Participants

We used purposive, convenience sampling by emailing a project summary and survey link to leadership at relevant organizations within authors' networks, requesting that it be shared further within their organizations as appropriate. These organizations included parent groups for children with visual impairments, educators of visually impaired children, vision rehabilitation service providers, as well as clinical collaborators. This indirect recruitment strategy precluded reporting of the total number of participants invited to share their perspectives.

When potential participants accessed the survey, they were provided with information about the study, inclusion criteria, and associated definitions. To meet inclusion criteria, participants must have supported at least one child (0–18 years) with suspected or known brain-based visual impairment in a professional or personal capacity. Respondents self-identified as meeting these criteria and provided consent before accessing survey questions. No identifying information was collected, but specific role, country and free text answers were removed from data to preclude identification.

2.4 Data collection

The questions emerged from stakeholder input. A full list of questions for this report is included as a Supplementary material.

2.5 Defining terms

In the information provided before the survey, participants were informed that we used 'brain-based' visual impairments broadly to encompass a variety of terms, and specifically noted that a child did not require a formal diagnosis of CVI for a specialist to participate. Within the survey, we specified that we use 'brain-based' to include

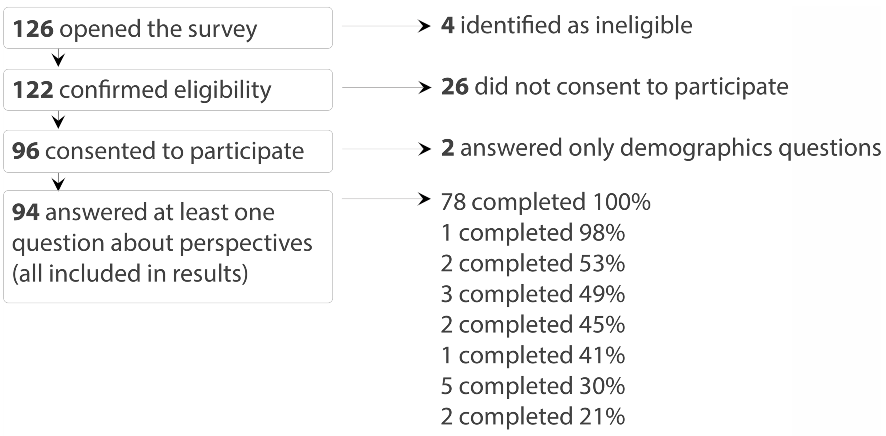

FIGURE 1
Participation among respondents commencing the survey.

a known or suspected neurological origin, 'visual' to include a wide range of visual abilities (not limited to visual acuity), and 'impairment' to refer to challenges impacting the child when both eyes are open. Respondents were asked to describe the children they interacted with (including whether they had a CVI diagnosis) and shared their perspectives on the meaning and implications of the term CVI.

2.6 Predictors/potential confounder/effect modifiers

We used the respondent's self-identified role to contextualize responses, grouping specific roles into the broader categories of family, education, rehabilitation, and clinical staff.

2.7 Analysis

2.7.1 Quantitative

We report descriptive statistics for all quantitative questions. Single-answer and multi-select questions are presented by response frequency. Five-point Likert scales were used to quantify agreement with statements, ranging from 'strongly agree' (coded as 2), to 'strongly disagree' (coded as −2), with neutral responses coded as zero. These codes were used to calculate the mean and standard deviation for relevant questions.

2.7.2 Qualitative

For the free-text questions, similar responses were grouped together, and themes were generated iteratively. Key themes were highlighted with selected quotes.

3 Results

3.1 Respondent characteristics

Of the 126 individuals who opened the online survey, 122 indicated they were eligible to participate. Ninety-six respondents consented to take part in the survey, and 94 completed questions beyond the demographics section and were included in the analysis. Of these 94 participants, 78 (83%) completed the entire survey (Figure 1).

TABLE 1 Demographic information of 94 respondents included in analysis.

Country	*n*	%
Aotearoa, New Zealand	78	83%
United States of America	7	7%
Canada	6	6%
Australia	3	3%
Role		
Education	51	54%
Rehabilitation	30	32%
Clinical	7	7%
Family	6	6%
Setting		
School	64	68%
Home	16	17%
Clinic	12	13%
Other	2	2%

Table 1 summarizes basic demographic information of the 94 respondents. Most were based in Aotearoa, New Zealand (78/94, 83%), with the remainder in the United States, Canada, and Australia. Most respondents came from an education background (51/94, 54%), including specialized teachers for vision, teaching assistants, and classroom teachers, with specialized teachers for vision the most selected role (43/94, 46%). Those who identified as rehabilitation staff (30/94, 32%) mainly included physiotherapists and occupational therapists. Only seven respondents reported a clinical role (including optometrists, ophthalmologists and pediatricians), and six were family members of children with brain-based visual impairment.

Of the 87 respondents who answered questions about severity, only 10 said they mostly supported children with mild visual impairments (10/87, 11%), the rest mostly supported children with moderate (49/87, 56%) or severe (27/87, 31%) visual processing challenges. Consistent with the more severe visual challenges, most respondents reported interacting with children who diagnosed with CVI (85/87, 98%), and co-occurring motor (76/87, 87%), cognitive (76/87, 87%), and communication (72/87, 83%) challenges.

Families reported daily interactions with children, while rehabilitation professionals typically reported weekly interactions, and educators mostly reported monthly interactions (consistent with a higher representation of specialized teachers for visually impaired children than classroom teachers/teaching assistants). In contrast, clinicians reported annual interactions. The duration of each individual interaction corresponded with its frequency: family members spent full or half days, educators and rehabilitation staff spent several hours, and clinical professionals reported spending about an hour per interaction.

3.2 Goals

Respondents were asked why it was important to understand what children with brain-based visual impairments see. They were provided five options and asked to select all that applied, followed by a request to select which goal was most important to them. Respondents were also provided a free text option to expand on their goals.

Most respondents selected multiple answers, with the two most common goals being "to better understand, or connect with a child" (87/94, 93%) and "to help the child develop an aspect of vision or wider development" (82/94, 87%). When asked to choose which goal was most important among selected options, educators and rehabilitation staff most frequently prioritized supporting learning and development (Educators: 25/51, 49%, Rehabilitation staff: 12/30, 40%), clinicians most frequently aimed to set expectations for a child's future visual capacity (4/7, 57%), and family members most frequently reported wanting to understand what their child sees to better connect with them (4/6, 67%).

Free-text responses revealed more specific and divergent goals, with clear alignment to professional role. For example, some classroom teachers emphasized the need to support access to educational curriculum. Specialized teachers for vision focused on supporting families by providing accurate information about what the child sees. Occupational therapists noted the importance for functional planning. Speech-language therapists reported wanting to understand vision to provide suitable communication options. One respondent articulately observed that these more divergent priorities can be at odds with each other, and highlighted the importance of collaboration to overcome tension resulting from differing goals:

> *"It is really important for speech language therapists to collaborate more with low vision specialists, as our priorities are often at odds when we are implementing [communication] systems for children with complex communication needs and CVI. We need to work together to find compromises." – Rehabilitation staff*

Respondents shared common goals of connecting with a child and supporting their development. However, free text responses revealed more divergent practical goals, which can create tension. Where goals are at odds, collaboration was highlighted as a crucial tool. Working together can help refocus specialists on shared goals and navigate compromises to achieve the best outcomes for children.

3.3 Collaboration

Most of our respondents (71/92, 77%) felt like they already worked as part of a collaborative interdisciplinary team, and the value of these networks was frequently highlighted in their comments. For instance, one respondent emphasized the positive impact on families:

> *"…we are working truly in a transdisciplinary model which is very exciting - as it strongly includes the whānau [family, broadly defined] and keeps the child at the centre." – Education staff*

Despite this high level of perceived collaboration among our respondents, on average, our respondents strongly agreed that they would like to collaborate with other specialists more (Mean 1.6 ± StDev 0.7), and respondents were, on average, neutral to the statement *"I do not think I have the time/resources to collaborate with other specialists more"* (Mean 0.0 ± StDev 1.3). Although overall motivation to collaborate more was stronger than reported time/resource constraints, these practical constraints were still cited as barriers in the free text, particularly in more rural or isolated areas:

> *"In the national team I am part of there is wonderful transdisciplinary sharing and collaboration. In my regional hospital role, I feel that there is further need to develop this cross disciplinary working model and while there is interest to do this time and resource gets in the way, as well as developing new ways of working. I think this is especially true for small centres where people have to be more generalist in their roles (like optometrists and ophthalmologists not being specifically paediatric, and paediatricians not being developmental etc)." – Clinical staff*

Respondents were asked about information flow, including sources and recipients of information. They were provided a list of specialists and asked to select all that they received information about a child's vision from, followed by a request to select which specialist provided the most useful information about what a child sees. Respondents were then asked to select all the specialists they provided information to about what a child sees. There was also a free text option to expand on information sharing.

On average, respondents reported receiving information from eight different specialists (StDev±5), most commonly including the family (78/94, 83%), and the child's ophthalmologist (71/94, 76%). Respondents reported providing information to seven different specialists (StDev±5), mostly commonly the family (79/94, 84%), and the child's classroom teacher (69/94, 73%). Family members were unique in that they reported providing more information than they received about their child's vision.

Across all respondents, family members were highlighted most frequently as the most valuable source of information (37/94, 39%). By specialist group, educators (25/51, 49%) and clinicians (4/7, 57%) most frequently reported family as the most valuable source of information about what a child with brain-based visual impairment sees.

The groups reported to provide the most valuable information about what a child with brain-based visual impairment see (family, low vision specialists and specialist teachers for visually impaired students), revealed in free text that they were uncertain if their contributions were valued. Specialist teachers for vision noted spending considerable time writing reports on how children with brain-based impairments use their vision daily yet were provided little feedback about the value this information held for other specialists. Likewise, families reported lack of feedback about the value of their insights, one family member articulated their hope in this domain:

> *"I would like to be more involved and be listened to more and be treated as a person who provides valuable information about my child." – Family member*

Taken together, most of our respondents reported feeling part of a collaborative interdisciplinary team, and they wanted to further invest in these teams because the value for the family was clear. However, setting up this way of working was highlighted as a challenge, especially outside urban where resources are stretched and there is less domain specific expertise. Importantly, those who are providing most of the information to the network of specialists aren't getting the feedback that it is valued, which has the potential to erode these valuable collaborative networks.

3.4 Understanding of CVI

Seventy-eight respondents (78/94, 83%) reported they were familiar with the term 'CVI'. Among those who were, most (69/78, 90%) believed a CVI diagnosis was important, mostly for accessing services (40/69, 58%) but also to help with understanding/identity (29/69, 42%). There was broad consensus that CVI is currently under-diagnosed (64/78, 82%). However, opinions on what constitutes a diagnosis of CVI varied. Respondents were divided on whether CVI should be considered a collection of 'symptoms'/'manifestations' (31/78, 40%), or whether a diagnosis should reflect something atypical about the health of the visual system (31/78, 40%), the remaining 20% offered other interpretations of what CVI. Twenty-two percent of respondents thought that evidence or history of damage in the brain was required for a CVI diagnosis (17/78, 22%) while only 12% believed that reduced visual acuity was required for a diagnosis (9/78, 12%). Clinical professionals, who are responsible for diagnosing CVI, were more likely to view the diagnosis as indicative of the visual system's health and to require reduced visual acuity and evidence of neurological deviation. These differing perspectives on CVI likely contribute to tension among specialists.

> *"CVI is not 'diagnosed' in most cases…it would be helpful if ophthalmologists have more up to date knowledge around CVI and provide it in reports if possible." – Education staff*

Of the 73 professional staff who were familiar with the term CVI, 53 (73%) reported having formal training on CVI, this equates to 60% of all professional staff included in our analysis. Respondents detailed informal ways they learned about CVI, including websites (notably the CVI Scotland website), social media and organic learning from other professionals and interactions with children living with brain-based visual impairments. Respondents employed within the education sector appeared to have the more opportunities for updated professional development on CVI, which was valued by those receiving it. However, this highlighted a knowledge gap between clinical and educational staff.

> *"It often seems that others, even vision professionals, know less about CVI than we do, and we don't know nearly enough!" – Education staff*

Overall, respondents tended to agree that a diagnosis of CVI was important for appropriate service provision (69/78, 90%), and it was underdiagnosed (64/78, 82%). Appropriately increasing the number of diagnoses is difficult when specialists have different ideas of what should constitute a CVI diagnosis, and education about CVI is largely informal, and often self-directed.

4 Discussion

4.1 Key results summary

Promoting a child's development and finding meaningful ways to connect with them were common overarching goals of the specialists surveyed; people caring for children with brain-based visual impairments want them to thrive. Many of our respondents felt they were part of collaborative teams, and specifically highlighted the power of transdisciplinary teams for providing quality services for children with brain-based visual impairments. However, several barriers to quality care were identified.

The logistics of professionals from different disciplines working together can be limited by time and resource constraints. In settings where this infrastructure exists, moving from multi-disciplinary to trans-disciplinary requires strong interpersonal rapport. Our data highlights some factors that could be barrier to these collaborative relationships. First, specialists have different specific goals, which can be at odds with one another. Second, those providing information about how a child uses their vision day to day do not always feel that their contributions are valued. Finally, specialists have different ideas about what a diagnosis of CVI means. These interpersonal factors can erode the collaborative relationships that are so key to transdisciplinary work.

The responses highlighted the need for enhanced formal training about CVI. If core elements of this training are consistent across disciplines, it would help align perspectives. Opportunities for formal teaching about the goals, responsibilities, and values of other disciplines, and how they can work together, would lay the groundwork for integrated support, where all members of multi-disciplinary teams are valued for the expertise they bring.

4.2 Connection to wider literature

The need for more formal training about brain-based visual impairments is aligned with other research. Studies that quantify training time in professional programs suggest training about CVI is limited. Castleberry et al. (2021) quantified CVI education in Ophthalmology and Optometry courses and found that only about half of programs (48%, 23/48) offer formal didactic CVI lectures, most only including 1–2 h. They found clinical instruction on CVI was sporadic and trainees typically first encounter CVI in pediatric eye care clinics and low vision clinics. The limited training about CVI has been reflected in generally poor knowledge in survey studies (Maitreya et al., 2018; Stanimirov et al., 2021). Similarly, Harpster et al. (2022) found a significant gap in formal CVI education for occupational therapists and teachers of students with low vision managing CVI.

Although there is a long way to go, awareness of CVI among diagnosing clinicians has improved over time. In a 2010 survey, parents reporting needing to fight to get a diagnosis (Jackel et al., 2010), while in a 2019 follow-up survey, parents reported the diagnostic process was easier and more timely. However, general knowledge was still lacking. In 2019, parents reported being given very little information about what the diagnosis means (Jackel, 2019). Similarly, interviews with parents in 2023 still indicated professional staff had poor knowledge about CVI (Oliver et al., 2023). Survey data from 2021 showed that parents who receive information at time of diagnosis report better rapport with care providers and feel more empowered to advocate for their child (McDowell, 2021), demonstrating the importance of clear information.

Increasing the hours of training within discipline is important for families, but developing a complementary curriculum across disciplines would help take the onus off the family to integrate information from different specialists. It would also help specialists understand the goals, responsibilities and values of specialists in other roles, promoting realistic expectations and better collaboration. The term 'transdisciplinary' was raised by several respondents in our survey to refer to meaningful collaborative networks centered around the child. While 'multidisciplinary' refers to a group that includes members from a variety of disciplines and 'interdisciplinary' refers to when the concepts from each discipline are integrated into a new whole, 'transdisciplinary' refers to collaborative networks that transcend disciplinary boundaries (Choi and Pak, 2006).

This transdisciplinary collaboration has been an important concept in early intervention for a long time (reviewd by King et al., 2009). There are known costs to implement transdisciplinary services in terms of managerial and team resources, however the benefits for children, families, and staff are considerable (King et al., 2009). The value of collaboration between health and education specifically is also recognized in the wider literature, with tangible steps for implementation. For example Burgess et al. (2015, using school immunisation as a case study) highlight that meaningfully integrating approaches requires substantial time, high levels of trust, and a willingness to share turf. The concept of transdisciplinary work is also highlighted in the CVI literature, though not always by name. Interviews with parents and children with CVI highlighted a central challenge for parents is integrating information between the specialists, and learning how to use this information to advocate for their child (Goodenough et al., 2021). Putting the onus on the service providers to communicate integrated information would reduce this pressure on families who already face substantial challenges.

4.3 Limitations

A strength of this study was the diverse range of specialists invited to participate, including family members, educators, rehabilitation, and clinical staff. Our recruitment was most successful within a blind and low vision education organization in Aotearoa, New Zealand. This organization generally works with children who have visual impairments meeting criteria for support, and they prioritize holistic service and collaboration between disciplines. Consequently, our results reflect an education-centric perspective, and we have limited input from clinicians and families. Our results also reflect the experience of working with children who have more severe impairments, and those who have some experience working as part of an inter- or trans- disciplinary team. This group may not fully represent the broader specialist community. Although our findings may underestimate the challenges faced by specialists outside such well-integrated networks, our data have the benefit of providing insights into the potential of transdisciplinary collaboration.

4.4 Future directions

Summarizing how different specialists respond to the same questions about brain-based visual impairments has highlighted some shared goals, as well as potential tensions between disciplines. This could be further elucidated by specifically sampling groups of specialists to compare perspectives quantitatively. As work is done to develop guidelines, and professional curriculum across disciplines (Pilling et al., 2023), continued research is key to knit together cohesive and complementary information and approaches.

5 Conclusion

Appropriately diagnosing, understanding, and supporting children with brain-based visual impairments is complex and requires a wide range of specialists to work together. These specialists tend to have different specific goals, responsibilities, and understanding of what CVI is, and these differences can lead to tensions between disciplines. Although clinical staff reported the value of family member's and educators' perspectives, families and educators do not always feel that their input is valued, and can question whether clinicians are taking their concerns about the children they support seriously.

Families and specialist teachers for vision feel the burden of educating themselves and others about brain-based visual impairments, despite often feeling uncertain, and not having decision-making power related to access to services. To alleviate this burden and utilize all specialists' desire to see children develop and thrive, clear guidelines and more cohesive education about brain-based visual impairments is needed. Improving knowledge about the condition and about the roles, responsibilities and value of other specialists, fostering cross-disciplinary care networks, and providing a context where interactions can become transdisciplinary, are potential pathways to higher quality care for children with brain-based visual impairments.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by University of Auckland Human Participants Ethics Committee (reference number: UAHPEC21919). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

JJ: Writing – original draft, Writing – review & editing, Formal analysis, Investigation, Methodology. EM: Writing – review & editing, Methodology. RF: Writing – review & editing, Conceptualization. NM: Writing – review & editing, Conceptualization. SS: Writing – review & editing, Conceptualization. LH: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2024.1510812/full#supplementary-material

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Sensory substitution and augmentation techniques in cerebral visual impairment: a discussion of lived experiences

Stephanie L. Duesing[1]*, Katie Lane-Karnas[2], Sebastian James Adam Duesing[3], Mae Lane-Karnas[2], Nai Y[4] and Arvind Chandna[5,6]

[1]Independent Researcher, Naperville, IL, United States, [2]Independent Researcher, Calais, VT, United States, [3]Independent Researcher, Chicago, IL, United States, [4]Independent Researcher, San Francisco, CA, United States, [5]Smith-Kettlewell Eye Research Institute, San Francisco, CA, United States, [6]Alder Hey Children's Hospital, Liverpool, United Kingdom

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EDITED BY
Jeff C. Rabin,
University of the Incarnate Word,
United States

REVIEWED BY
Amanda Lueck,
San Francisco State University, United States
Linda Lawrence,
Independent Researcher, Salina, KS,
United States
D. Luisa Mayer,
New England College of Optometry,
United States

*CORRESPONDENCE
Stephanie L. Duesing
✉ stephduesing@gmail.com

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Pediatric vision loss due to cerebral visual impairment (CVI) is an urgent public health issue, demanding evidence-based (re)habilitation and educational strategies. As with other neurodiverse populations, research on CVI needs to be directly informed by the lived experiences of those affected—children, adults, and their families. In this paper, three individuals with early-onset CVI and two parents discuss sensory substitution and augmentation developed in childhood in the absence of early identification of CVI, and they detail the important impact of the empowering, professionally taught non-visual skills–such as braille, orientation and mobility training, and assistive technology–which were acquired later. Efforts to improve visual perception ability and understanding of the visual world, both effective and ineffective, were made through traditional, professionally administered vision therapy, self-taught coping strategies, and from intensive arts participation. The authors discuss the strategies they use to leverage senses other than vision to achieve their daily life, educational, social, and career goals. Nonvisual skills training effective in those with ocular blindness, though received later in life after the delayed diagnosis, proved to be indispensable for these authors' who have CVI access to all aspects of independent life. It is our hope that these personal experiences may encourage research into how traditional nonvisual skills training used for the ocularly blind, as well as sensory substitution and augmentation techniques, may be used to develop evidence-based multidisciplinary interventions; improved academic and independent life skills; multisensory educational and therapeutic interventions; and successful integration into the community for all CVIers.

KEYWORDS

CVI, sensory substitution, cerebral visual impairment, O&M, sensory augmentation, braille, orientation and mobility, verbal mediation

Introduction

Cerebral visual impairment (CVI) is the number one cause of severe visual impairment/ blindness in children in higher income countries (Solebo et al., 2017; Chong and Dai, 2014; Williams et al., 2021; West et al., 2021; Teoh et al., 2021; Kessel et al., 2024), and the prevalence is increasing worldwide (Pehere et al., 2019; Aghaji et al., 2015; Ozturk et al., 2016). CVI can result from several different causes but commonly occurs due to adverse events in the prenatal or perinatal period, which lead to brain injury affecting processing of vision. CVI is characterized by challenges with visual recognition and visually guided behaviors of action – also termed higher visual function deficits or visual perception deficits (HVFDs; VPDs) and variable loss of visual acuity and visual field defects (Fazzi et al., 2007; Philip and Dutton, 2014; Boot et al., 2010; Zihl and Dutton, 2015; Chandna et al., 2021a, 2021b, 2024). HVFDs can impact those affected so significantly that their CVI informed clinician reports their functional vision is at the level of legal blindness, even in the case of typical VA (Kran et al., 2019). The definition and scope of the term "blindness" has been the subject of debate for over a century with recommendations from as early as 1893 (Elementary Education [Blind and Deaf Children] Act) to extend it beyond visual acuity and visual field to include "too blind" to perform everyday activities and a special dispensation for those "too blind to be able to read the ordinary school books" (Smith and Paton, 1915); a critique of visual acuity as a sole measure of blindness (Harman, 1926; Ray et al., 2016); the limited inter-relationship between visual function and functional vision (Colenbrander, 2009) and recent debates on criteria of blindness specific to CVI (Kran et al., 2019; McConnell et al., 2021; Pilling et al., 2023). In this paper, we incorporate the National Federation of the Blind's functional definition, where the term" blind "refers to anyone who must devise alternative techniques to do anything which he would do with sight if he had normal vision regardless of the cause of the vision loss (Jernigan 1984, 1995).

CVI is poorly understood, often underdiagnosed, diagnosed late and, at worse, misdiagnosed because severity of vision impairment continues to be defined by VA, which may be normal in CVI (van Genderen et al., 2012; Chandna et al., 2021a, 2021b, 2024). In addition, most clinicians and low vision professionals lack awareness of CVI (Maitreya et al., 2018; Pilling, 2022; Oliver et al., 2023). This ignorance is made worse by a lack of reliable tools in routine clinical practice to assess HVFDs though such tools are being evaluated in research (Philip and Dutton, 2014; Lueck et al., 2019; McConnell et al., 2021). Referral for (re)habilitation is historically based on low VA and reduced visual field, and outcomes are typically measured by VA (World Health Organization (WHO), 2022). Parents may notice symptoms but are hard pressed to find low vision professionals who have medically and scientifically accurate knowledge of CVI, and who can tease out what symptoms are due to CVI and what symptoms are due to other conditions. According to the National Institutes of Health (NIH), because CVI often coexists with various medical conditions and learning disabilities, it can be difficult to determine which specific condition is causing a patient's difficulties and this challenge highlights the importance of prioritizing CVI in the diagnostic process (Chang et al., 2024; Gordon et al., 2024). In the US, more than 155,980 young people who have CVI are currently undiagnosed (CVI Center, 2023) and living without non-visual skills training (Appendix), making access to a CVI informed, multidisciplinary team crucial.

Prior to identification of CVI, those who have the condition are unaware of their visual perceptual problems as they did not develop the higher visual functions and are therefore unaware that neurotypical, sighted people have those abilities (Goldenberg et al., 1995; Heilman, 1991; Celesia and Brigell, 2005). This results in underdiagnosis and, worse, a misdiagnosis (Lueck et al., 2019; Chokron and Dutton, 2023), with one in thirty mainstream-educated children having at least one CVI-related vision problem (Williams et al., 2021).

The critical importance of listening to and understanding patient experiences has been reported for visual impairment (VI) (Douglas, 2006; Robertson et al., 2021; Lupón et al., 2018). Qualitative interviews investigating CVI families' experiences have emphasized the importance of more work in this area (Jackel et al., 2010; Goodenough et al., 2021; Oliver et al., 2023). Understanding the lived experiences of CVIers who have awareness of the challenges with HVFDs and who have developed successful multisensory strategies to deal with HVFDs in CVI is paramount for understanding of the condition; clinical and research direction; habilitation planning; and to provide context for families making decisions for their children.

In this paper, two adults (Sebastian Duesing, Nai) and one teenager (Mae Lane-Karnas) who have CVI, two parents (Stephanie Duesing, Katie Lane-Karnas), and their clinical partner (Arvind Chandna) discuss what VPDs exist and how the authors who have CVI independently and non-consciously developed sensory substitution and augmentation techniques to learn and engage in daily life. These coping strategies for learning and surviving as blind people in a visual world, without any appropriate non-visual skills training, were developed in early childhood, in two cases through intensive participation in the arts. Before medical diagnosis of CVI, Duesing, Lane-Karnas, and Nai all experienced ineffective outcomes in regard to improving CVI symptoms with efforts to improve functional vision in vision therapy. These three authors all later pursued training in and use non-visual skills in daily living. Lane-Karnas and Nai struggled with reading print and their challenges are described and discussed in each section.

Diagnosis of CVI

Though it is now recognized that CVI typically has an early onset due to adverse events at or around time of birth, the CVI diagnoses of these authors were not gained until ages 18, 16, and 12 (Nai, Duesing, and Lane-Karnas respectively), and until identification of CVI, these authors, their families, and their healthcare providers had no awareness of CVI.

Nai shares their experience, "Starting at age 12, I was diagnosed with strabismus along all three axes: A-pattern exotropia, heterotropia, and incyclotropia. I was also diagnosed with several oculomotor conditions that are often seen in CVI patients: both accommodative and convergence insufficiencies with a very low AC/A ratio, ciliary spasms, and dysfunction of saccadic, smooth pursuit, and fixation eye movements. At the time, my oculomotor conditions were not understood as potential indicators of an underlying neurological visual disability. Nor was the history of adverse events to my visual development tied to my clinical presentation at the time: being born 3 months premature, incubated for a full year, malnourished in the orphanage, subjected to ongoing deliberate head trauma that was intentionally covered up, and significant visual deprivation due to uncorrected high hyperopia until my first cycloplegic eye exam at the age of 12. Additionally, I had not yet been diagnosed with autism or my genetic disorder, which both show higher rates of sensory processing issues. It was not until I had my first accidental head trauma at 18 due to tripping on a large pile of trash I did not see while walking down a steep sidewalk and being thrust head-first into a brick wall that I was finally diagnosed with cortical blindness based on my MRI. It was only once I studied CVI as a blindness professional that I recognized my lived experiences in descriptions of CVI traits such as visual agnosia, simultanagnosia, and near-total lack of visual imagery. Finally, it is not until discussing my case in detail with CVI-literate eye doctors and researchers that I was able to re-analyze and retroactively understand my CVI holistically. It has only been now, as a blindness professional, that retrospectively, I realize that before I was diagnosed at age 18, I was forced to operate with much less usable vision than many of my ocularly-blind clients who easily qualified as legally blind and benefited from blind services. It was blindness skills that empowered and freed me, not vision therapy or being forced to try to 'see.'"

Duesing discovered his CVI at the age of 15. It took 15 months, countless doctor's appointments, and two research studies before he finally received his comprehensive CVI diagnosis. This paper published by Pamir et al. (2021) was the first scientifically documented instance of an individual with CVI, Sebastian Duesing, using verbal mediation to process his vision. In the paper, fMRI data revealed that Duesing can see or not see with his eyes wide open–at will. The Pamir paper describes how Duesing has no conscious perception of sight when he is not using verbal mediation. He spends most of his waking hours fully blind unless he is actively using his extremely limited vision to do a visual task, such as reading or working on an art project. Duesing has normal acuity and severe CVI. He was diagnosed with CVI, including prosopagnosia, object agnosia, topographical agnosia, and simultanagnosia at the New England Eye Low Vision Clinic at the Perkins School for the Blind. The only things that Duesing can see and recognize like a neurotypically sighted person can are words, letters, numbers, and simple shapes. Thus, Duesing always had visual access to numeracy and literacy because he can visually recognize symbols, even though he cannot see and recognize faces, places, objects, or biological forms like a typically sighted person. Duesing was born full term and his CVI is caused by brain damage due to an adverse birth experience. His parents were told that he had a 9 on his APGAR score and the parents were led to believe that he was perfectly healthy. Duesing had all normal developmental milestones and was identified as extremely intellectually gifted as a kindergartner, 2 years before the gifted program at his school began. Duesing developed minor acuity problems when he was about 8 years old. He began wearing glasses at that time and was seen by an eye care practitioner every year after. At no time before the Duesing family discovered his CVI at age 15 did any of Duesing's medical, educational, or eye care professionals raise any concerns about his vision.

Lane-Karnas was diagnosed with convergence and accommodative insufficiency by their optometrist at age 11, then diagnosed with CVI at age 12 at New England Eye Low Vision Clinic at Perkins, with follow up at Boston Children's Hospital. Diagnoses include simultanagnosia, prosopagnosia, optic ataxia, visual agnosias (letter, form, and topographic), and oscillopsia. All of Lane-Karnas' pediatrician eye screenings had been normal and their acuity is normal. Professionals consulted throughout their childhood never suggested Lane-Karnas' difficulties could be related to impaired vision. Their providers' best understanding is that their CVI is attributed to birth history, which includes oxygen loss.

Sensory substitution and augmentation strategies self-developed during childhood

An undiagnosed young child with CVI has no way to ascertain that their visual perception of the world is different from others (Chokron et al., 2021). As young children, these authors who have CVI were unable to make sense of a visually unfamiliar world without appropriate non-visual skills training because they could not see typically. In response, starting in early childhood, Duesing (Pamir et al., 2021), Lane-Karnas, and Nai all self-developed sensory substitution and augmentation strategies to interpret the world non-visually. We describe sensory augmentation as the creation of a novel sense or enhancing an existing one (e.g.: echolocation as creating a novel skill; increasing our sensitivity to sound frequencies as enhancement of an existing one) and sensory substitution as the use of an alternative intact sense for an impaired sense (e.g.: the use of a non-visual alternative such as hearing or touch as a means of obtaining information from the environment). For example, using touch to feel the dots in order to read braille when the written letters cannot be seen is an example of substituting the sense of touch for the sense of vision. Sensory substitution and augmentation are often closely related. Both could also be delivered or assisted with devices. These three authors' use of non-visual coping techniques extended beyond the traditional "five senses," using proprioceptive, haptic, and kinesthetic senses. The proprioceptive sense is the internal knowledge of the position and movement of the body in space. Haptic memory relates to the sense of touch, and kinesthetic sense is muscle memory–the ability to sense the position and movement of the body. Duesing, Lane-Karnas, and Nai all have a need for movement and touch to develop proprioceptive, haptic, and kinesthetic memories in the absence of, or severely reduced, ability to form visual memories.

People who have CVI often have difficulty visually recognizing faces, objects, environments, forms, and letters (Farivar, 2009). In addition to these visual agnosias, Duesing, Lane-Karnas, and Nai experience a lack of visual imagery in the mind's eye. They all use multisensory, semantic, and verbal compensation strategies to substitute for a lack of visual recognition and visual memory. While there is debate in psychology and neuroscience literature about the interplay of visual recognition, visual imagery, and visual memory (Dulin et al., 2008), these three CVIers articulate lived experience of how they cope with these visual deficits. According to Nai, "I used semantic buttressing (processing and retaining a nonlinguistic symbol through its linguistic meaning) to sidestep my inability to visualize or form mental images. For example, I passed visual memory tests by encoding a series of shapes by the sounds they represent in various writing systems, since I have never been able to hold nonlinguistic information in my mind's eye or visual memory." Similarly, Duesing uses verbal memory to substitute for lack of visual memory: "I simulate recalling visual imagery by memorizing verbal descriptions of the things I see." For example, his cat's characteristics are orange/stripes/yellow eyes. When Duesing thinks those words to himself, he gets a momentary glimpse of what his cat looks like. This is a momentary flash of an image with no retention in visual memory. Duesing says, "I get an idea of it." Duesing continues, "I also recall motions or gestures made by people or animals as kinesthetic memories, remembering the movement via a mental construct of the feeling of performing that movement." Duesing uses an enormous, memorized taxonomy of verbal descriptors to identify people, places, and things. Duesing says, "As a young child, drawing helped me practice the skill of identification by salient characteristics, for example, by repeatedly drawing, painting, coloring, and sculpting characters I like. I learned to memorize the features that made them distinguishable.

The same skills that allowed me to replicate images of those characters helped me to match people to the characteristics by which I knew them in real life." Duesing spent thousands of hours of his childhood drawing, painting, and sculpting beginning in late infancy.

Lane-Karnas expands on the challenge posed by lack of visual mental imagery in combination with visual agnosias, describing how it impacts visual memory: "I cannot make visual memories except for colors, and I cannot visually recognize faces, places, objects, or letters, so every time I see something, it is like seeing it for the first time. But I have to remember things, and I can still make memories, so I've learned to leverage my other senses to make non-visual memories of visual things. I will use tactile memories of texture, and shape, the imagined feeling of tracing it with my fingers, a distinctive smell, any connected storylines or emotions, and color. My non-visual memory of a chicken consists of the feel of the texture of its feathers; the shape of a chicken in my arms; the specific movement their head makes that I hold as a haptic memory. Then I pull up many storylines of interactions I've had with them, and information I know about them."

Because Duesing, Lane-Karnas, and Nai live with topographical agnosia (impaired to no ability to navigate using visual recognition and visual memory of landmarks and routes) they each self-developed haptic and proprioceptive methods of understanding and mapping familiar environments. Duesing explains, "I depend on proprioception-based memories to navigate familiar spaces. Like the way that others can visualize the layout of a room, I have non-visual memories of the physical feeling of moving around in a space, from which I form image-less 'maps' of the positions of walls, doors, furniture, and other objects after I encounter them or maneuver around them physically."

Nai uses kinesthetic logging to memorize information, which they describe as memorizing environmental information based on how it feels in the muscles, joints, and proprioceptive nerve spindles. Nai states, "The information 'sticks' more effectively that way, such as by having spatial information mapped by touch on my back; or repeating instructions to myself in tactile sign language; or by walking routes a few times to encode the proprioceptive information into my nerve spindles. It is helpful to have others spatially map things on my back, especially for new routes or any geographical information, but I also map on my own arm or thighs when processing verbal information too, to better imagine whatever is being described, especially if the spoken content includes verbal step-by-step instructions. I learned the back mapping technique directly from the North American DeafBlind community's emerging tactual language, called ProTactile. It involves the whole body. Grammar is both spatial and kinesthetic. You sign hand-over-hand, or tactile. I was first introduced to it in 2013 at a workshop at Gallaudet University, and I continue to use it throughout my college education and career. I also use it successfully with in-person CVI students now."

Lane-Karnas identifies with the haptic and proprioceptive methods of navigating described by Duesing and Nai, depending on the environment: "When I am navigating familiar spaces with my vision, I use mainly a strategy like Duesing described, avoiding obstacles by remembering exactly how it feels in my body to walk around them. When I am navigating unfamiliar spaces or using my cane, I use a strategy more similar to Nai's, where I imagine drawing a map of my walking paths on my own body and then add tactile details that my cane is giving me, in lieu of a spatial–visual memory."

Severe visual perception deficits with no awareness of visual impairment can cause trauma. Lane-Karnas explains, "All these things I was doing, I thought were normal, I was unaware other people did not do these things. I had no explanation as to why everything was so much harder and more stressful for me, or why I was constantly more tired, angry, and sad than my friends." Duesing states, "I always knew that other people were much less freaked out about going places than I was, but I had no idea why." Nai adds about their own traumatic childhood experience with going undiagnosed, "Since any complaints about being unable to see were met with severe punishments, I had no choice but to develop my coping strategies entirely non-consciously. Being undiagnosed meant I did not have the context or framing to even think of myself as having a legitimate form of blindness. It was only at age 18, when I qualified for services, that I was finally able to consciously think about the non-visual skills I had spent a lifetime developing."

Print reading modifications and CVI

Fluent reading ability is crucial to success in society academically, socially, and functionally. Prior to CVI diagnosis, both Lane-Karnas and Nai had severe visual perceptual problems with reading print despite normal visual acuity. Lane-Karnas struggled with reading print and by 12 years old had materials extremely modified by their homeschooling parent in the following ways: limit of 5–15 min of daily print engagement, use of a tinted typoscope, reduced complexity, increased spacing, blue backlit screen, specific sans serif font, and total absence of distraction. Lane-Karnas notes that this "never made a substantial enough difference for me to be at a functional literacy point where I could live a successful life. I was using every other means at my disposal to try to remember something I could not make a visual memory of. I developed touch-memories for the outside shapes of words, by imagining tracing the exterior shape, which I could not see inside of. I was imagining trying to feel the outside of the word, and then using context clues and prediction as well as storylines in my own head to figure out what was going on. And it was practically impossible to maintain because I was holding it in my short-term memory. I was not able to get to a functional reading point; these strategies were not making me literate and were fatiguing to an extreme extent."

Duesing prefers a 10-point font because he has simultanagnosia, and the small size of the font allows him to see two or three letters at a time. Duesing's clinically measured, "detection" visual field is full, however his functional field is markedly constricted. He has only a tiny patch of acuity in the center of his visual field where the only things he can recognize like a typically sighted person are words, letters, numbers, and simple shapes. The surrounding area of his visual field is like a dense, colorful fog – a mostly useless blur of light, color, and motion. "Additional print reading modification beyond the requirement of a 10-point font has been largely unnecessary for me. Print reading is one of the relatively few visual tasks I can do easily and comfortably."

Without diagnosis of CVI in childhood, Nai lacked alternatives to print and suffered. "Trying to look at print was like trying to remain perfectly still and motionless on a boat in the middle of the sea. It made me nauseous and dizzy and exhausted beyond words. This constant sensation of a rocking boat threw a wrench into any hope of comprehension. The motion of the text made it so hard to see the letters that all of my attention was focused on trying to decode each individual letter. I had difficulty visually discerning similar shapes, such as an a from an e, an r from an n, etc. I was so drained from trying to see the letters that I could not follow the story. By the time I got to the bottom of the page, I would not know what the top said. There was no brain space left for tracking the story. All of it got used up on seeing. Looking at the letters. Piecing them together. Trying to follow and track the lines….I would try and try, and I just could not anymore. When I hit that wall, I resorted to self-harm."

Vision therapy and CVI

Before CVI diagnosis, Duesing, Lane-Karnas, and Nai undertook vision therapy administered by licensed optometrists in an attempt to improve/cure functional vision through improving oculomotor function and focus using eye exercises. Several licensed optometrists claimed that they could cure Duesing's CVI and specifically stated that they could cure his visual agnosias and simultanagnosia, which is, of course, impossible. Vision therapy had no effect on his CVI. He did experience an improvement in his convergence insufficiency, an oculomotor problem that developed from a concussion unrelated to his CVI that occurred during his sophomore year of high school. Duesing reflects, "Vision therapy had no effect on my CVI symptoms; it was a frustrating and exhausting experience with no significant benefit."

Nai remembers, "Vision therapy addressed the oculomotor conditions I had that accompanied CVI but did not improve any of my visual processing issues, as my eye care team was unaware of this deeper layer of visual disability at the time. I developed extreme awareness and conscious control of my intra- and extraocular muscles and over-compensated for accommodation/convergence insufficiency by contracting these muscles hard in short spurts. When I tried my best to approximate horizontal alignment/convergence, my vertical and torsional deviations would skyrocket and trigger hours of extraocular eye muscle cramps and accommodative spasms, which were extremely painful and uncomfortable."

Lane-Karnas's vision therapy was undertaken to address accommodative and convergence insufficiency. After vision therapy, they still could not move their eyes while balancing on one foot, had difficulty talking and moving their eyes, and could not keep print in focus to read a paragraph. They comment, "Like Nai and Duesing, vision therapy did not help my CVI. It did not solve my problems with visual recognition and chronic visual fatigue, among other CVI challenges, and it left me exhausted and incapacitated." Thirty minutes of vision therapy and 5–20 min of vision-based academic tasks daily resulted in a state of chronic visual fatigue and nervous system exhaustion that accumulated into long-term burnout and could not be relieved with rest.

Sensory substitution and augmentation through non-visual skill training

After a CVI diagnosis, Duesing, Lane-Karnas, and Nai sought training in and currently utilize various non-visual skills and tools, including braille, occupational therapy for navigation, orientation and mobility with a white cane, and access technology. Nai uses a white cane and both text-to-speech software and braille displays on multiple devices (phone and both work-issued and personal laptops) on a daily basis in their professional, advocacy, and personal life. Nai says, "I learned these skills through blind rehab services after my acuity and other low-order processes [visual acuity, visual field, color, and contrast] plummeted as an adult after my accidental head trauma, allowing me to qualify as legally blind. I wish I had had these same strategies when I had the higher-order visual processing issues with perfect acuity."

Prior to Duesing's discovery of his CVI, Duesing could not drive, independently navigate on foot, or travel by public transportation. He therefore had no way to safely go to college, begin a career post high school, or live independently. In 2017, Duesing discovered that he had CVI at the age of 15 when he was a sophomore in high school. At that time, no one on Duesing's educational team had ever heard of CVI or received any training in how to educate or teach orientation and mobility (O&M) skills to a person with CVI. Because Duesing was unsuccessful in accessing those life-saving services from his school O&M instructor, and because the family was unsuccessful at finding a private O&M instructor, and because he had only 2 years before he planned to go to college, the Duesing family had to think creatively to find other solutions.

For Duesing the solution to not receiving a timely CVI diagnosis and access to O&M was privately sourced occupational therapy (OT) for navigation, which insurance paid for, combined with traditional O&M at a non-profit institution which happened more than a year later. For Duesing, OT and O&M were complimentary: he received 3 days per week of OT instruction in navigation over a period of 4 months. In OT, Duesing learned how the address system is organized in a major city; how to use Google Maps on his phone to navigate a major metropolitan city using public transit and on foot, which are skills he could never have received from his suburban school district without missing upwards of 5 h per day of instructional time. Later, Duesing was able to attend a private, 1 week O&M "boot camp" at a guide dog training facility where Duesing received instruction on traveling with a white cane and crossing busy intersections using only his non-visual senses. Duesing notes, "Occupational therapy for navigation and O&M training had a massive impact on my ability to successfully and confidently navigate the world. I do not use a white cane most of the time, but I carry it with me as a back-up, and I am glad to have the option to use it when I want or need it. It taught me how to effectively synthesize the visual skills I do have with a variety of non-visual skills."

Duesing, a professional ontologist, was a high-performing academic student. He has always been able to access print visually. He says, "I wish that I had the opportunity to learn braille when I was younger, as I think it would have been useful to have a non-visual reading method as an alternate option so that I could engage with text in non-visual ways when I'm low on energy to use for vision. Though I could still learn braille now, I wish I would have had the opportunity to develop fluency with it as a child" (Silverman and Bell, 2018).

After Lane-Karnas was diagnosed with CVI at 12, they began braille instruction which improved literacy and math understanding and retention. Lane-Karnas writes, "Because visual information is so draining and hard to remember, I have learned non-visual ways to get necessary information I use every day, such as braille, a screen reader, and a long white cane. These tools greatly reduce the amount of visual fatigue in my life, leaving me more able to be happy, and more able to do visual things that I choose to do, like art. Reading print as a 12-year-old I would continually meltdown, cry, and be chronically dysregulated. Now, using braille, access tech, and non-visual learning, I can attend instruction in homeschool, classes, independent study, and tutoring that can range from

30 min to 2 h multiple times a day. I will still be fatigued but am able to quickly recover and pace my day such that I am able to engage in hobbies, time with friends, and family activities. I am also better able to retain and engage with the non-visual information."

Optimizing conceptual understanding using sensory substitution and augmentation

Duesing, Lane-Karnas, and Nai describe how their vision as a sensory input is often replaced or informed by other senses. Their use of non-visual senses is consistently misinterpreted by sighted observers, who presume a task is being done visually. Nai explains: "My brain is naturally hyper-kinesthetic to the point my physical therapists have regularly commented I have exceptionally sensitive interoception (internal sensations of the body). I have relied heavily on my interoception as a substitution for visually guided reach throughout my whole life. For example, I know where to reach for the knife on the rack by how the angles feel in my joints; it appears to the outside observer like I am focusing on a visual target, but I am actually guided by the sensation in my muscles, tendons, and bones. I also learned American Sign Language by feeling angles in my fingers, hands, wrists, and arms because I could not see the signs."

Lane-Karnas is a visual artist and since late infancy has spent hours a day drawing, painting, or sculpting. They have no visual memory of anything they have ever seen or created, and due to simultanagnosia, have never seen their own art in its entirety. Lane-Karnas references using verbal, kinesthetic, and touch modalities to support their art. "When I want to learn how to draw a new thing, it takes a lot of intentional practice, repetition, and visually studying the thing before I can consistently draw it. I'll add words and storylines to all the elements of the thing so I can remember it in enough detail to draw it accurately, and then break those elements down into shapes and encode the movement of drawing the shapes into my hand so I can consistently draw it the same. This process has the added benefit of really strengthening my memory of the figure of the thing and giving me a firmer understanding of what it is. It also helps to identify common visual patterns and expectations which I can use to more easily visually identify the thing." As a younger child Lane-Karnas sought any representations of the human form that they could touch: dolls, posable figures, sculptures, mannequins, and bones, spending endless hours posing and playing with them, drawing the human form, and deconstructing clothing to learn how it is constructed around the body. "Having a tangible, tactile interaction with something helps me to cement what the thing even is; it lets me become clearer on how it is constructed, built, or functions, and have the knowledge to much more accurately reflect what it is through visual arts."

Duesing's internal experience of tasks presumed to be entirely visual are not at all so: "Activities that other people often think of as inherently visual tasks, like playing video games or drawing, are multisensory experiences to me. Muscle memory and proprioceptive understandings of shapes and patterns fill the void left by my absence of visual memory and my visual processing. Though the outcomes of those so-called visual tasks are approximately the same for me and those who do perform them visually, the processes by which we reach those outcomes are very different." Duesing, who has object agnosia, uses haptic memory for tasks like distinguishing ordinary objects that he has frequently held in his hands. "When I am looking for a piece of produce in the grocery store, I cannot always rely on everything to be labeled, which is normally my first-line strategy for identifying objects in a store, because I can read visually. If, for instance, I'm looking for an apple, I imagine the feeling of holding an apple in my hand, and I look for things that fit the approximate shape and size that I 'feel.' If I look at a pineapple, I know that's not the item I'm looking for, because, without touching the fruit, I can feel in my hand that it's too big. If I look at a banana, I know that it's not what I'm looking for, because I can feel that it is too small, even though I have not touched it. Most things can be ruled out like this. I only lean on looking for more vision-intensive characteristics like texture or color when I cannot rule things out by shape and size, like telling an orange from an apple."

Like Lane-Karnas, Duesing is also a visual artist, and reiterates, "Things that seem like visual tasks do not have to be. I find that drawing things requires a lot of study, practice, and repetition. Because I have no visual memory of, e.g., people, I cannot rely on a memory bank of experiences of looking at people in order to draw them. I can represent humans fairly accurately in my art because firstly, I've spent a lot of time studying the canon of proportions of the human body, and secondly, I use a lot of reference images for pretty much everything I draw in order to get body positioning, lighting, and perspective right. I also have to work with a small area of useful vision, so I find it easiest to work very small during the initial parts of the drawing process in which I'm determining larger-scale features like composition, poses, and proportions. By "working small," I mean things like doing thumbnail sketches on a small section of the canvas, zoomed far out, as the first step in the drawing process, and trying to get composition/pose/proportions looking good before I worry about representing any detail at all."

Unlike Duesing and Lane Karnas, Nai found trying to participate in the visual arts to be extremely frustrating. Nai says, "Visual arts were not helpful to me for interpreting the world. It was a torture I endured because of school requirements." The sensory substitution and augmentation techniques utilized by one CVIer may be entirely inappropriate for another due to the unique challenges presented by CVI and their different life experiences.

Discussion

Despite advances in describing the manifestations of CVI in academic, research, and clinical literature, the voices of individuals affected by CVI remain largely unheard in peer-reviewed literature. Listening to patients' lived experiences has been linked to improved healthcare outcomes. Studies by Coulter (2012) and Edbrooke-Childs et al. (2016) show that patient involvement in decision-making leads to better treatment results, increased satisfaction, and more effective care. Systematic reviews by Gleeson et al. (2016) and Bate and Robert (2007) further highlight that incorporating patient feedback into service improvement enhances both clinical outcomes and overall patient wellbeing (Bate and Robert, 2007; Coulter, 2012; Edbrooke-Childs et al., 2016; Gleeson et al., 2016). This is true for children with VI regarding improved healthcare outcomes. Robertson et al. (2021) and Lupón et al. (2018) show that involving families in care decisions enhances emotional and social well-being. Lanza et al. (2024) emphasizes the value of integrated assessments that consider these experiences. Douglas, (2006) demonstrate how understanding the broader impacts of VI leads to more effective support (Douglas, 2006; Lanz, 2024; Lupon, 2018; Robertson et al., 2021). Incorporating these perspectives into care results in better overall outcomes for children with VI.

The significant impact of CVI on patients and their families is just starting to be addressed through the direct voices of CVIers in the literature. The importance of listening to CVIers and their families has been highlighted in semi-structured interviews (Jackel et al., 2010; Falkenberg et al., 2020; Goodenough et al., 2021; McCarty and Light, 2023; Oliver et al., 2023). An approach to visual, emotional, and behavioral needs of the CVI population has been suggested based on a literature review (McDowell, 2023). CVIers have expressed their challenging experiences in support websites (e.g.: CVI Scotland; Perkins School for the Blind). However, until very recently, the direct written experiences of people who have CVI have been missing in the peer-reviewed literature, which has contributed to the existing challenges in underdiagnosis, misdiagnosis, and a lack of CVIer-led intervention methods in the management of CVI. In this paper we demonstrate the value of incorporating direct, self-written perspectives into the published literature in order to advance understanding of CVI.

This paper details self-developed sensory substitution and augmentation strategies, as well as the essential need for non-visual skills training in three CVIers with HVFDs. Multisensory intervention has been used successfully for people who have no residual vision and for people with severe visual acuity and/or visual field impairments (Gori, 2015; Purpura et al., 2017; Morelli et al., 2020). Successful implementation of braille reading has been described in a CVIer with good visual acuity (Lane-Karnas, 2023), and recommended by clinicians (Kran et al., 2019) and educators (Lueck and Dutton, 2015), but it is not routinely available to CVIers as part of the (re)habilitation process, especially when visual field and visual acuity are not impaired to significant levels. In addition, CVIers need to be assessed for functional vision, including their abilities with visually guided behaviors, with particular attention paid to the concerns of the CVIers themselves and/or their families. Clinically administered evaluations of visual function (acuity and field) frequently do not meet criteria for access to non-visual skills training such as braille, O&M, and access technology. The NIH's emphasis on the importance of an individualized approach for the educational, vocational, recreational, and social needs of CVIers (Chang et al., 2024) is strongly illustrated by the heterogeneous needs of these authors who have CVI.

Lacking typical visual perception, the authors recount how proprioceptive, haptic, and kinesthetic senses are crucial for their ability to form non-visual memories in order to do tasks and to navigate familiar environments. Duesing, Lane-Karnas, and Nai detail the importance of O&M training and OT for navigation to learn to navigate in unfamiliar environments in order to live independently, along with other interventions, to mitigate psychological harm from years spent being blind in a world without non-visual skills. CVI visual impairments that prevented fluent print reading, such as letter agnosia and oculomotor issues, and undergoing ineffective vision therapy, further emphasize the importance of developing non-visual skills, such as with braille and access technology. The authors explain how non-visual learning methods effectively improved their academic, social, and personal learning that had been impossible to achieve using visual means.

In this paper, individuals with CVI describe their experiences with vision therapy (eye exercises intended to promote better use of both eyes together as a team). Duesing most likely benefited from vision therapy for his post-concussion convergence insufficiency (Scheiman et al., 2017) during his teenage years. However, the paradigms of behavioral vision therapy (VT) instituted for other symptoms and signs, as experienced by all three authors with CVI, were ineffective in addressing their visual perceptual and oculomotor challenges. The authors underwent VT during childhood during the years when the literature questioned its efficacy as a treatment; reviews of studies on VT for convergence insufficiency had reported minimal benefits (Rawstron et al., 2005; Barrett, 2009), while Ciuffreda (2002) suggested convincing lab-based models for background evidence and recommended further research. At no point in their treatment were these controversies from the published literature research discussed with these three authors who underwent vision therapy, or their families. In later years, Scheiman (2018) provided a comprehensive review toward establishing an evidence-based foundation for vision therapy, particularly in the treatment of convergence insufficiency, while also advocating for continued scientific inquiry to strengthen the field's credibility in areas where the evidence is still mixed or lacking. A Cochrane meta-analysis reported with moderate certainty evidence that pediatric patients with convergence insufficiency are 5 times more likely to show fewer symptoms and better outcomes after office-based vision therapy with home reinforcement compared to placebo therapy (Scheiman et al., 2020). However, this review did not address the efficacy of vision therapy in individuals with CVI. Recently, Flaherty et al. (2024) critically assessed behavioral vision therapy for children with reading difficulties, including dyslexia, noting limited evidence supporting its effectiveness. While some improvements in visual processing were observed, they emphasize the need for more rigorous, evidence-based research to confirm its impact on reading outcomes. None of the studies in the past or recently, had children with CVI. Research into the causes of commonly occurring near focusing and eye movement difficulties in CVI is ongoing, with no proven cause or treatment. While VT may be beneficial for some disorders, the authors recommend prospective controlled trials to determine its efficacy for CVI, as current evidence does not support its clinical use in this population.

In this article, important and unique information is presented by CVIers themselves about self-developed compensatory strategies that were developed without the knowledge of the parents. For example, for two of the authors of this paper, their intense, lifelong interest in the arts helped them to develop a richer understanding of the visual world without any parental or educational awareness of what was driving these CVIers' passionate pursuit of the arts. There is increasing evidence that effective technology and interventions are best developed and implemented with the involvement of the blind and visually impaired (Conradie et al., 2015; Magnusson et al., 2018) and should not be led only by sighted observers or by simulations of visual impairment (Qiu et al., 2024). We provide information which strongly suggests that CVIers and their families should be involved in all aspects of research to improve clinical understanding of CVI and its impacts on education and (re)habilitation. This collaborative effort is essential to developing and implementing a multisensory, integrative approach that is founded on the autonomy and choice of the affected CVIer based on their personal interests and abilities rather than on inaccurate assumptions made by typically sighted outside observers.

Currently, there are at least 180,449 individuals in the US aged 0–22 with diagnosed or likely CVI, with a diagnostic rate under 14% (White Paper CVI Perkins). Additionally, there is a lack of evidence-based, scientifically valid training in CVI for low vision professionals (Harpster et al., 2022; Lueck et al., 2023), as well as a significant shortage of teachers of the visually impaired (TVIs) and O&M specialists (Schles and Chastain, 2023; Randles, 2020). This shortage, made worse with few with expertise in CVI, is affecting service delivery for young individuals diagnosed with CVI. As the number of CVI diagnoses increases, the demand for low vision services, coupled with the limited availability of trained professionals, underscores the need for solutions to address the growing gap in service provision. For example, while the use of OT for teaching independent travel in combination with O&M training with a white cane for CVIers may be novel in the United States, the NIH has recently affirmed the importance of cross-specialty cooperation due to the interdisciplinary effects on individuals with CVI (Chang et al., 2024). Furthermore, the current definition of legal blindness, which relies solely on measures of acuity and visual field, frequently limits the ability of CVIers of all ages to access non-visual learning and (re)habilitation services (Kran et al., 2019). As more people with CVI are diagnosed, the large numbers of people needing low vision services and the lack of providers is cause for concern and raises the need for creative solutions to help this overlooked and vulnerable population. According to the NIH, there is a lack of adequate transition planning to adult care for the majority of people with any form of vision impairment and providing necessary and often life-saving support for CVIers across their lifetimes is of the utmost importance (Chang et al., 2024). For these reasons, it is especially important to consider how late diagnosed individuals with CVI who are aging out, or have already aged out of public school, will access non-visual skills training that are typically accessed by ocularly blind children in the school setting.

Conclusion

Every person who has CVI has a unique experience of their own functional vision (Lueck and Dutton, 2015). Having firmly established the prevalence of HVFDs in CVI at all levels of acuity loss (Ortibus et al., 2011; Chandna et al., 2021a, 2021b; Sakki et al., 2021), the need is great in the medical and education fields to understand common visual perceptual differences in their HVFD spectrum and severity as a heterogenous group as well as for each individual. Direct dialogue with CVIers and their parents uncovers rich details of shared and disparate experiences living with CVI; and of sensory augmentation and substitution that improves understanding of the world in the absence of typical vision. Sensory substitution and augmentation techniques employed by the three blind authors of this paper are invisible to the typically sighted observers and are therefore impossible to identify through observation of behaviors alone, illustrating the essential need for dialogue between CVIers themselves and parents, medical, and scientific professionals. As CVIers begin to share their firsthand experiences with sensory substitution and augmentation techniques used to understand the visual world, consideration must be given to how these stories can be used to help accurately describe the CVI experience; plan research on effective multisensory educational and therapeutic interventions; achieve an accurate diagnosis; and ensure individualized, accessible education and habilitative services.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

Ethics statement

Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

SLD: Writing – original draft, Writing – review & editing. KL-K: Writing – original draft, Writing – review & editing. SJD: Writing – original draft, Writing – review & editing. ML-K: Writing – original draft, Writing – review & editing. NY: Writing – original draft, Writing – review & editing. AC: Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Appendix

Access technology: When AT is mentioned, it is meant to include AAC devices, braille displays/notetakers, and all means of audio-based tools such as screen readers, dictation software, audio books.

Agnosia, visual: decreased or absent ability to visually recognize or identify faces (prosopagnosia), shapes, objects, symbols, environments (topographical agnosia).

CVIer: The term "CVIer" was first coined by one of the authors, Nai, and its use has become accepted and spread in professional and informal settings over the last several years. This reflects and respects our community's growing sense of self and acceptance, and our understanding of belonging within the larger neurodiversity paradigm.

Kinesthetic logging (term introduced by Nai): memorizing environmental information based on how it feels in the muscles, joints, and proprioceptive nerve spindles. It is different from touch because it is not the sensation of the atmosphere on the surface of the skin, but the feeling inside the body relative to the environment.

Non-visual skills: skills such as braille, O&M, access technology, etc. (or as blindness rehabilitation professionals call" blind skills").

Semantic buttressing (term introduced by Nai): processing and retaining a nonlinguistic symbol through its linguistic meaning. For example, interpreting a circle as the letter O, a triangle as the capital Greek letter delta Δ, and a square as the Chinese character 口 (k ǒ u, "mouth").

Sensory augmentation: creating a novel sense or enhancing an existing one (e.g.: echolocation as creating a novel skill; increasing our sensitivity to sound frequencies as enhancement of an existing one).

Sensory substitution: the use of an alternative intact sense for an impaired sense (e.g.: the use of a non-visual alternative such as hearing or touch as a means of obtaining information from the environment). For example, using touch to feel the dots in order to read braille when the written letters cannot be seen is an example of substituting the sense of touch for the sense of vision. Sensory substitution and augmentation are often closely related. Both could also be delivered or assisted with devices.

Typoscope: a tool that obscures all lines of text on a page except for the one line the reader is focusing on.

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EDITED BY
Jeff C. Rabin,
University of the Incarnate Word,
United States

REVIEWED BY
Linda Lawrence,
Independent Researcher, Salina, KS,
United States
Nicola McDowell,
Massey University, New Zealand

*CORRESPONDENCE
Elizabeth Boatwright
✉ boatwrights2@mac.com

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Cerebral/Cortical visual impairment (CVI) in Down syndrome: a case series

Elizabeth Boatwright[1]*, Rudaina Banihani[2,3], Ilse Willems[4], Kathleen Lehman[5,6], Ellen Mazel[4], Hannah Mark[7], Mike Wong[8], Silvia Vietzman[8], Arvind Chandna[8,9] and Gena Heidary[10] for DSMIG DS+CVI Workgroup

[1]Independent Internal Medicine-Pediatrics Physician, Educator, and Researcher, Colorado Springs, CO, United States, [2]DAN Women & Babies Program, Department of Newborn & Developmental Pediatrics, Sunnybrook Health Sciences Centre, Toronto, ON, Canada, [3]Department of Pediatrics, University of Toronto, Toronto, ON, Canada, [4]CVI Center, Perkins School for the Blind, Educational Programs, Watertown, MA, United States, [5]Institute on Human Development and Disability, University of Washington, Seattle, WA, United States, [6]Department of Psychiatry & Behavioral Science, University of Washington School of Medicine, Seattle, WA, United States, [7]Department of Social and Behavioral Sciences, Yale School of Public Health, New Haven, CT, United States, [8]SeeLab, Smith-Kettlewell Eye Research Institute, San Francisco, CA, United States, [9]Translational Clinical Eye Research Centre, Alder Hey Children's Hospital, Liverpool, United Kingdom, [10]Department of Ophthalmology, Boston Children's Hospital and Harvard Medical School, Boston, MA, United States

Cerebral/Cortical visual impairment (CVI), a brain-based visual condition, is a leading cause of childhood blindness and low vision but remains underdiagnosed in individuals with Down syndrome (DS). This report presents three case studies of adolescents with DS and CVI (DS + CVI), illustrating how CVI can manifest alongside the developmental, cognitive, behavioral, and social profiles of individuals with DS. We describe comprehensive ophthalmological evaluations, assessment for visual perceptual deficits with a screener questionnaire, functional vision assessments, and neuroimaging (when available). Through a detailed retrospective examination of these cases, we explore the complex interplay between CVI and DS, emphasizing how CVI-related challenges—such as difficulties with visual attention, spatial perception, processing, and navigation—are often misattributed to DS alone or to other more commonly recognized co-occurring conditions in DS. Diagnostic overshadowing, coupled with a lack of standardized screening tools, has led to delayed diagnoses and missed opportunities for intervention. Our findings highlight the importance of recognizing CVI in individuals with DS using reliable tools for assessment of functional vision to better appreciate the effect on their diverse developmental outcomes, and to incorporate CVI into our understanding of the DS phenotype. These case reports underscore the need for further research to determine the prevalence and impact of CVI in DS and advocate for the development of tailored screening protocols and evidence-based interventions to support individuals with DS + CVI across the lifespan.

KEYWORDS

Cerebral/Cortical visual impairment (CVI), Down syndrome (DS)/Trisomy 21, Down syndrome and Cerebral/Cortical visual impairment (DS + CVI), visual attention, Top-11 Higher Visual Function Question Inventory (HVFQI), functional vision assessment (FVA), case report, visual perception

Introduction

Cerebral/Cortical visual impairment (CVI) is a brain-based visual impairment and the leading cause of childhood blindness and low vision in the United States (Chang et al., 2024), with a rising global prevalence (Teoh et al., 2021; Pehere et al., 2019). CVI is often linked to early brain injuries, including perinatal hypoxia, neonatal complications, and genetic conditions such as Down syndrome (DS) (Lehman et al., 2024; Pehere et al., 2018; Wilton et al., 2021). While CVI is increasingly recognized in other neurodevelopmental conditions, it remains under-identified in DS despite its potential impact on development (Wilton et al., 2021; Chokron and Dutton, 2016; Dale and Sonksen, 2002; Hatton et al., 1997; Mosca et al., 2015).

DS is a complex neurogenetic condition caused by the duplication of all or part of chromosome 21, making it the most common chromosomal anomaly (Mai et al., 2019). Developmental outcomes in DS vary widely (Frank and Esbensen, 2015; Winders et al., 2018), shaped by a dynamic interplay of co-occurring medical (Bull et al., 2022; Baksh et al., 2023; Amr, 2018; Breslin et al., 2014; Laws and Hall, 2014; Rako et al., 2021), ophthalmologic (Jain et al., 2023), and neurodevelopmental conditions, including autism spectrum disorder (ASD) (Richards et al., 2015; Fidler et al., 2022) and attention-deficit/hyperactivity disorder (ADHD) (Ekstein et al., 2011). Advances in research have deepened our understanding of these conditions (Antonarakis et al., 2020), and evidence-based care guidelines have improved life expectancy and quality of life (Bull et al., 2022; Tsou et al., 2020). Our understanding of the strengths and challenges in DS continues to evolve (Fidler, 2005; Schworer et al., 2022). Recognizing variability in medical, behavioral, social, and cognitive profiles is crucial for developing tailored interventions (Fidler and Nadel, 2007; Onnivello et al., 2023a). Despite improvements in care, individuals with DS exhibit a broad range of developmental and cognitive trajectories (Baumer et al., 2024; Channell et al., 2021), suggesting that additional unrecognized factors contribute to these differences.

We propose that CVI is one such overlooked factor. Currently, CVI is absent from DS screening guidelines despite its potential to significantly affect developmental, cognitive, and social outcomes.

This retrospective case report examines three adolescent females with DS and CVI (DS + CVI), analyzing long-term medical, developmental, and behavioral data. Clinical records were reviewed, including ophthalmological evaluations, neuroimaging (when available), structured screening tools for visual perceptual deficits, and functional vision assessments. We reconstruct each diagnostic trajectory, emphasizing early neonatal, medical, and developmental factors contributing to delayed CVI recognition. We highlight diagnostic overshadowing, where CVI-related difficulties—such as impaired visual attention, processing, and navigation—were misattributed to DS or other co-occurring conditions. By analyzing these cases, we illustrate how CVI manifests in DS and advocate for systematic CVI screening and integration into routine clinical care to improve developmental outcomes.

Case reports

Case 1

Born at term, this female infant had APGAR scores of 8 and 8 at one and 5 min. She required oxygen supplementation for mild respiratory distress during a 10-day stay in the neonatal intensive care unit (NICU), during which time she was treated briefly with phototherapy for transient hyperbilirubinemia. A hemodynamically stable patent ductus arteriosus (PDA) was identified but resolved spontaneously. No significant perinatal complications were recorded (Table 1).

From infancy, delays in visual-motor integration were apparent, and inconsistent reaching behaviors and limited eye-hand

TABLE 1 Medical history.

Medical issue	Case 1	Case 2	Case 3
Pregnancy	Uneventful, no prenatal diagnosis	Prenatal diagnosis of DS and VSD	Complicated with oligohydramnios, prenatal diagnosis of DS and CAVC
Delivery	NSVD, APGARS 8/8	NSVD, APGARS 8/9	C/S, APGARS unknown
Gestational age (GA)	41 weeks	39 weeks	36.5 weeks
Birth weight—SGA (<3rd percentile)	No	No	No
Congenital heart disease (CHD)	PDA (no surgery)	VSD (no surgery)	CAVC (surgical repair age 2 months, uneventful)
PPHN	Present	None	None
Evidence of NE	None	None	None
Neonatal hyperbilirubinemia	Yes, phototherapy 2 days	Yes, phototherapy 10 days	None
Complex eye conditions	Yes	Yes	Yes
Hearing Problems	Fluctuating conductive hearing loss	Conductive & sensorineural hearing loss	Fluctuating conductive hearing loss
	Eustachian tube dysfunction, PE tubes	Eustachian tube dysfunction, PE tubes	Eustachian tube dysfunction, PE tubes
	No augmentative devices	Bilateral hearing aids	FM system at school
Thyroid disease	None	Hyperthyroid	None
Hip instability	Yes, treated with PT	No	Yes, multiple surgeries

PPHN, persistent pulmonary hypertension of the newborn; NE, neonatal encephalopathy; CAVC, complete atrial-ventricular canal; VSD, ventricular septal defect; PDA, patent ductus arteriosus; FM, frequency modulation; PE, pressure equalizing.

coordination persisted; at age 3 year, she was not using hands and eyes together. Gross motor skills showed progressive delays (Figure 1). Hip instability contributed to persistent difficulty with gait. Early assessments confirmed delays across cognitive, language, and motor domains, with persistent fine motor challenges such as difficulty reaching, holding objects or writing. Socially, she engaged well in near-gaze interactions but displayed variable joint attention. Language development slowed, particularly after stopping sign language, but she does rely primarily on spoken language, often using scripted phrases and echolalia. At age 14, she underwent a multidisciplinary ASD evaluation and was diagnosed with autism (Table 2).

Ophthalmologic evaluations revealed significant nystagmus by 2 months (Table 3A). She has been treated for strabismus with surgery at age 8 and though she wears corrective lenses for her high myopia, her most recent best-corrected binocular visual acuity (VA) is limited at 20/80 (monocular testing was difficult). She was diagnosed with CVI at 4 months due to lack of visual fixation not explained by ocular findings. She received vision services through school, but her parents and school were led to believe that her CVI had resolved by middle school. Reevaluation at age 19 using the TeachCVI Screener #2 (TeachCVI Project, 2017; Martin et al., 2024) was prompted by ongoing functional vision concerns and confirmed persistent CVI-related deficits (Table 3B). While MRI was not performed, functional assessments revealed significant challenges with clutter, motion, and depth perception. Behavioral patterns such as mirror aversion, difficulty visually following another's pointing finger to distant objects, and sensitivity to loud sounds became more pronounced by adolescence.

Presently, focused CVI accommodations, including the use of contrasting backgrounds, color coded visual schedules, and clutter reduction, have enhanced her ability to navigate both academic and social environments. Behavioral interventions emphasizing multisensory learning have further supported her development, although she struggles to process information from visual, auditory, and motor channels at the same time. Academic skills are minimal and life skills have been emphasized at school.

*** Milestone ranges taken from:** https://doi.org/10.1111/jir.12580

*All cases are 80-90% independent but need help with orientation and fasteners. **Case 1 has not yet achieved this skill. ***None of the three cases have independendly achieved this skill. †Case 1 has no recorded snipping. ††Cases 1 and 2 do not consistently trace.

****Milestone ranges taken from:** https://doi.org/10.1111/jir.12176

Figure 1 Key

▭ DS Norms
▲ Case 1
● Case 2
■ Case 3

FIGURE 1
Developmental trajectories.

TABLE 2 Summary of Developmental Diagnoses and Standardized Assessments.

Case	ASD Diagnosis*	ASD Diagnostic Tools Employed	ADHD Diagnosis*	ADHD Diagnostic Tools Employed
1	Yes	GARS-3; ASRS; ADOS-2	Not assessed	N/A
2	No	ADOS-2	Not assessed	N/A
3	Not assessed	N/A	Yes	VADRS, Parent and Teacher Forms

*Diagnoses were made based on DSM-5 criteria through multidisciplinary team assessment.
ASD, Autism Spectrum Disorder; ADHD, Attention Deficit Hyperactivity Disorder; ADOS-2, Autism Diagnostic Observation Schedule, Second Edition; ASRS, Autism Spectrum Rating Scales; DSM-5, Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition; GARS-3, Gilliam Autism Rating Scale, Third Edition; VADRS, Vanderbilt ADHD Diagnostic Rating Scale; N/A, Not Applicable.

Case 2

This female infant was born at term after a pregnancy complicated by a prenatal diagnosis of ventricular septal defect (VSD), with APGARS of 8 and 9 at one and 5 min. Her neonatal course included transient tachypnea of the newborn (TTN) and mild hyperbilirubinemia, requiring a 7-day NICU stay for observation and treatment. She stabilized quickly and was discharged without complications to continue phototherapy as an outpatient for a total of 10 days (Table 1).

Developmental milestones revealed early gross motor skills in normal limits for DS, with progressive delays (Figure 1). Visuomotor challenges became evident during infancy, with limited reaching and grasping. While she initially learned baby signs, verbal language development slowed after sign language use was discontinued, leading to delays in communication noted by age 3. Early assessments confirmed global developmental delays. Socially, she was interactive but struggled with joint attention and sustained eye contact. She later displayed strong phonics-based reading skills but struggled to read multiple words on a page and had difficulty tracing and writing. Additional medical conditions include mild hearing loss, hyperthyroidism, obstructive sleep apnea, and migraines (Table 1). By age 10, concerns about social behavior and repetitive movements led to an ASD evaluation, which ruled out the diagnosis (Table 2).

Ophthalmologic findings included hyperopia (Table 3A). Esotropia was managed with bifocals. At the most recent follow up, best corrected binocular VA is 20/50 (monocular testing was difficult). Behavioral observations highlighted difficulty navigating stairs and hesitating at thresholds, and heightened sensitivity to sound and motion which interfered with group activities and transitions.

Persistent functional vision concerns prompted CVI evaluation at age 15.5. Functional visual assessments using The Perkins CVI Protocol (CVI Center at Perkins School for the Blind, 2024) confirmed significant deficits in visual attention, clutter, motion, glare, and depth perception (Table 3B). Brain MRI showed increased susceptibility in the basal ganglia regions (Table 4).

Currently, CVI-specific interventions, such as the use of slant boards, dimmed overhead lighting with task lighting, realistic photographs, a 3D schedule system, and multisensory learning aids have improved her academic engagement. Structured routines, deaf-blind supports, and cane use have reduced her anxiety and improved her participation in group settings.

Case 3

Born preterm at 36.5 weeks via cesarean section due to decreased amniotic fluid, this female infant was vigorous at birth (APGARS unknown). Prenatal imaging identified a complete atrioventricular canal (CAVC), which required surgical repair at 2 months. Her neonatal course was otherwise uneventful, with a brief 2-day NICU stay for monitoring.

From infancy, developmental delays were apparent, particularly in visuomotor skills. Developmental milestones revealed early gross motor skills in normal limits for DS, with progressive delays (Figure 1). Language development progressed steadily due to consistent use of multimodal communication methods. Early assessment confirmed significant cognitive, motor, and language delays. Socially, she demonstrated a preference for humor and verbal engagement, often incorporating lines from songs or movies into conversations. Additional medical conditions included congenital heart disease requiring repair, hip instability requiring surgery, obstructive sleep apnea, fluctuating hearing loss, and migraines. Behavioral challenges included difficulty navigating crowded environments, difficulty with transitions, and noncompliance and aggressive behavior in a busy classroom. She was asked to leave her inclusive school setting after 4th grade, after aggressive behaviors including hitting, yelling, and pushing over tables accelerated. At age 13, she was diagnosed with ADHD, which improved with pharmacological treatment.

Ophthalmologic evaluations revealed exotropia with significant myopic astigmatism OD and mild hyperopia OS indicating significant anisometropia but no amblyopia. Exotropia was surgically corrected at age 7.5. At the most recent follow up, her best corrected binocular VA is 20/30 (20/40 monocular VA).

CVI evaluation at age 15 confirmed significant visual processing deficits, including clutter and navigation difficulties. Brain MRI findings indicated increased susceptibility in the globus pallidus, and functional assessments corroborated CVI-related impairments.

Today, CVI-specific accommodations, including the use of backlighting, clutter-free environments, realistic photographs, and cane use have improved her ability to navigate both academic and social contexts.

Diagnostic assessments across the three cases

The diagnosis of CVI in the three cases was established through a systematic and multidisciplinary approach, incorporating birth history,

TABLE 3 **Visual function and functional vision.**

(A) Ophthalmologic history and evaluation

Eye exam, ophthalmologic conditions	Case 1	Case 2	Case 3
Glasses Rx	−8.00 + 3.25 × 65 OD, −7.25 + 3.00×77 OS	+3.75 + 2.50 × 82 OD, +4.25 + 2.50 × 104 OS	−2.50 + 2.00 × 015 OD, +0.75 sphere OS
Best corrected binocular acuity	20/80	20/50	20/30
Strabismus	+Esotropia surgery age 8 years	+Esotropia managed with bifocals	+Exotropia surgery age 7.5 years
Nystagmus	Present	Absent	Absent
Cataracts	+Mild (not visually significant)	None	None
Abnormality of retina or optic nerve	None	None	None
CVI diagnosis	Age 4 months	Age 15.5 years	Age 15 years

(B) Select CVI visual behaviors evaluated by functional visual assessments (FVA)

Visual behavior	Case 1, age 19 years*	Case 2, age 15.5 years**	Case 3, age 15 years**
Visual attention	Brief glances	Brief glances	Brief glances
Visual recognition	Recognizes a set of familiar simple 3D objects; 2D is mostly inaccessible	Recognizes familiar and simple 3D; can recognize certain simple colored photographs	Recognizes familiar 3D objects and realistic colored images and photographs. Recognizes sight words by overall shape of the word
Impact of clutter and crowding	Clutter interferes with visual attention, finding objects	Clutter interferes with visual attention, finding objects; best with 1–2 objects at a time	Clutter interferes with visual attention, finding objects; best with 1–2 objects at a time
Visual field abilities	Lowered head, lowering accessible field	Decreased right, upper fields	Right stronger than left field, decreased use of lower field
Impact of color	Relies on color coding to recognize	Relies on color coding to recognize; drawn to bright colors	Relies on color coding to recognize; drawn to bright colors
Visual guidance of upper limbs	Inconsistently looks while reaching	Inconsistently looks while reaching	Inconsistently looks while reaching
Access to people	Brief eye contact	Uses visual and auditory cues to identify people	Brief eye contact; misidentifies facial expressions
Impact of light	Frequently stares at light sources; backlighting supports visual attention; sensitive to glare	Frequently stares at light sources; prefers low ambient light and task lighting; very sensitive to glare and bright light (can trigger migraine)	Sometimes stares at light sources; backlighting supports visual attention; sensitive to glare (can trigger migraine)
Impact of motion	Motion in the environment can be distracting and overwhelming; small movements draw attention	Motion in the environment can be distracting and overwhelming; often sits in busy environments; small movements draw attention	Motion in the environment can be distracting and overwhelming; small movements of cursor draws attention
Sensory integration and impact on vision	Difficulty looking and listening simultaneously; noises in the environment can be startling and upsetting	Difficulty looking and listening simultaneously; noises in the environment can be startling and upsetting	Difficulty looking at an item she's holding; noises in the environment can be startling and upsetting
Support of compensatory skills	Benefits from auditory and verbal cues, color coding, prediction, and memory	Benefits from auditory cues, color coding, and tactile cues	Benefits from auditory and verbal cues, color coding, and tactile skills

*Assessed with: https://www.teachcvi.net/screening-tools

**Assessed with: https://www.perkins.org/our-work/cvi/the-perkins-cvi-protocol/

(C) Comparison of higher order visual function screening results: top-11 questions*

Top-11 questions	Case 1	Case 2	Case 3
1. Does your child sometimes have difficulty seeing something that is pointed out in the distance?	5	5	4
2. Does your child find uneven ground difficult to walk over?	5	5	5

(Continued)

TABLE 3 (Continued)

(C) Comparison of higher order visual function screening results: top-11 questions*			
Top-11 questions	Case 1	Case 2	Case 3
3. Does your child bump into things when walking and having a conversation?	1 (very cautious, stops talking to walk)	3 (usually does not walk & talk)	5
4. Does your child have difficulty walking downstairs?	3	5	5
5. After being distracted does your child find it hard to get back to what they were doing?	3	4	4
6. Does your child have difficulty finding a close friend or relative who is standing in a group	2	4	5
7. Does your child find copying words or drawings time-consuming and difficult	5	5	4
8. Does your child trip at the edges of pavements going down?	2	2 (stops)	3
9. Does your child find inside floor boundaries difficult to cross?	2	4	5
10. Does your child have difficulty seeing scenery from a moving vehicle?	2	5	4
11. Does your child look down when crossing floor boundaries?	5	5	5
*Doi: 10.3389/fnhum.2021.711873			

Likert scoring rubric (note adjusted scores compared to 2021 paper).
Questions answered by parents have a choice of answers with scores: never—score 1; rarely—score 2; sometimes—score 3; often—score 4; always—score 5.

TABLE 4 Imaging and EEG findings.

Diagnostic tools	Case 1	Case 2	Case 3
MRI findings	Not performed	Mildly small brainstem Increased susceptibility in globus pallidus and substantia nigra	Pons hypoplastic, mildly hypoplastic inferior vermis Increased susceptibility in globus pallidus R > L
EEG findings	Not performed	Diffuse slowing, no epileptic foci	Diffuse encephalopathy, nonspecific etiology

comprehensive eye examinations, assessment with semi-structured question inventory screener, functional vision assessments, and neuroimaging findings where available (see discussion in Pilling et al., 2023). We used the validated Top-11 CVI Screener and HFVQI-51 semi-structured interview (Chandna et al., 2024), completed by parents of the cases, to assess higher visual function deficits. Table 3 provides a comparative analysis of CVI-related behaviors across the cases, illustrating distinct yet overlapping profiles.

Diagnostic assessments revealed a shared constellation of challenges consistent with CVI (Fazzi et al., 2007; Boot et al., 2010; Philip and Dutton, 2014; Sakki et al., 2018), including visual perceptual deficits and visually guided motor behavior delays, leading to challenges with functional vision. All three cases showed both visual function deficits (decreased VA, decreased visual fields, oculomotor impairment), as well as functional vision deficits of the ventral stream (e.g., route finding and orientation, recognition) and dorsal stream (e.g., visually guided movement, motion perception) (Chang et al., 2024). All three had difficulty integrating sensory information (auditory, visual, tactile at one time), to varying degrees. This sensory sensitivity occasionally led to episodes of visual overwhelm, manifesting as shutdowns or vasovagal reactions, sometimes triggering migraines. Compensatory behaviors, such as trailing hands along walls or hesitating before navigating unfamiliar spaces, reflected attempts to adapt to a visually unpredictable world.

Notably, all three cases scored in the high-severity and spectrum of CVI, exceeding normative thresholds established for neurotypical individuals and aligning with findings from other CVI cohorts without DS. The mean severity scores for each case were 2.4, 3.27, and 3.45 (numerical scale: never = 1; rarely, sometimes, often, always = 5) and significantly higher than the mean severity score from normative data (see below). Because of high severity scores on the Top-11 screener, all three cases were evaluated with the detailed HVFQI-51, a structured 51 question inventory (Chandna et al., 2021). Severity scores (SD) for HVFQI-51 for the three cases were: 3.39 (0.18); 3.05 (0.77); 3.04 (0.49), values considerably different from the normative cohort (1.43 S.D. 0.49 cohort $n = 120$) (Chandna et al., 2024).

Although there were similarities in functional visual profiles, the three cases had unique variations and varying levels of severity. These findings align with prior reports emphasizing the heterogeneity of CVI-related sensitivities (Philip and Dutton, 2014).

Neuroimaging findings by MRI scans did not show clear lesions in afferent visual pathways, but did reveal differences in basal ganglia structures, specifically the globus pallidus and substantia nigra. EEG findings in both cases indicated nonspecific slowing.

Discussion

The findings across the three cases emphasize the intricate interplay of developmental, behavioral, and functional challenges associated with CVI in individuals with DS. Each of the three cases had progressive developmental delays compared to DS norms, as visualized in Figure 1, yet each case had a unique developmental trajectory, underscoring the individualized effect of CVI in DS amidst the varied factors influencing overall development. These cases demonstrate the confusing overlap between DS and CVI, ASD, and ADHD (Chokron and Dutton, 2023), as well as the challenge of distinguishing ASD in VI (Molinaro et al., 2020). Functionally, challenges with visual perception and visually guided motor behavior impacted activities of daily living, navigation, leisure activities, and independence. Because of diagnostic overshadowing and lack of awareness of CVI, the complex challenges faced by the three cases were easily attributed to DS alone, concurrent medical conditions, or co-occurring neurodevelopmental conditions like ASD and ADHD, highlighting current challenges with diagnosing CVI in DS.

Clear vision is fundamental to achieving optimal developmental and functional outcomes in individuals with DS, particularly given their strengths as social learners and visual processors (Rosenbaum et al., 2024; Rosenbaum and Gorter, 2012). A heightened awareness of DS + CVI is important as early identification of CVI in DS may lead to early intervention and individualized accommodations that might improve the trajectory of visual development and potentially impact overall development, fostering greater independence and well-being for individuals with DS + CVI across their lifespan.

All three cases had complex ocular conditions common in DS (Jain et al., 2023) and were followed by ophthalmology from birth, but CVI was missed in two cases until the teen years, due to lack of awareness, as well as the lack of clearly defined CVI risk factors, screening guidelines, and diagnostic criteria that would prompt a CVI evaluation in DS. These cases share some common elements in their birth histories, clinical ophthalmologic exams, and imaging studies that may serve as a basis for further study of factors increasing vulnerability to CVI in DS.

One key observation is that none of the cases had classic risk factors for CVI at birth (Chang et al., 2024), but all three birth histories included medical events common for babies with DS, such as brief NICU stays, oxygen supplementation, and treatment for hyperbilirubinemia. While these medical factors are not traditionally recognized as CVI risk factors, emerging evidence outside the DS population suggests that even mild hypoxia (James et al., 2019; Nagy et al., 2021; Li et al., 2022) and mild hyperbilirubinemia in term infants (Amin et al., 2019; Hou et al., 2014) may have subtle but lasting effects on visual pathways in the brain, and both are common in neonates with DS.

In addition, these cases had clinical ophthalmologic findings of subnormal visual acuity (VA) and visual perceptual deficits that persisted despite traditional corrective measures of their complex ocular conditions like strabismus and nystagmus. Complex ocular conditions are common in DS (Santoro et al., 2024) but also occur in CVI (Fazzi et al., 2007), and the fact that the complicated visual profiles of these cases did not fully account for the observed functional vision deficits in these cases is a hallmark feature of brain-based visual impairment. Subnormal VA is common in DS, and may serve as a clinical finding that should prompt further questioning and evaluation for CVI.

MRI findings in two cases align with established neuroimaging patterns in DS alone, and include basal ganglia anomalies, which are more prevalent in DS than in the general population (Santoro et al., 2024; Takashima and Becker, 1985; Lee et al., 2013). While linked to depth perception, motor coordination, and contextual learning challenges in non-DS populations (Milardi et al., 2019; Paprocka et al., 2020), their functional significance in DS remains unclear.

These cases suggest that a CVI diagnosis benefits individuals with DS + CVI, their families, and their care teams. According to parents, the CVI diagnosis validated their concerns about their teen's vision, provided them with a new framework for understanding their teens' behavior, increased their empathy and patience, and provided new tools to support their loved ones. Anecdotally, therapeutic interventions targeting each teen's CVI-specific needs have improved functionality, reduced anxiety, and increased engagement in learning environments in all three cases. However, further research on CVI accommodations would help clarify the effectiveness of interventions.

Despite its retrospective nature (reliance on parental recall for early developmental milestones and behaviors, incompleteness of records, and variability of diagnostic tools), this case report provides compelling evidence for further research into CVI's prevalence, spectrum, and impact within the DS population. The cases presented are teenagers, which may limit the generalizability of findings to younger children, and a case study by nature will not capture the breadth of presentations of CVI in DS.

However, together with recent publications describing nuanced profiles of development in DS (Onnivello et al., 2023a; Onnivello et al., 2023b), cognition in DS (Channell et al., 2021) and autism symptoms in DS (Fidler et al., 2022), these cases raise the possibility that CVI and ocular visual impairments may influence the broader DS phenotype in ways previously unrecognized. To help clarify the role of visual function deficits in DS, prospective studies of development in DS which consider the impact of vision and studies that identify and quantify higher visual function deficits in the larger DS population are necessary. A detailed multidisciplinary assessment of ASD and/or ADHD-like challenges in DS, particularly in familiar versus novel environments, is essential to accurately differentiate between vision-related behaviors and those associated with the neurodevelopmental disorders. Modifications to existing diagnostic tools to account for visual impairments may improve accuracy in these populations (Williams et al., 2014). Incorporating CVI assessments within the already established ophthalmology screening protocols recommended by the AAP may improve early identification, facilitate timely interventions, and enhance developmental outcomes.

Future research should focus on refining CVI-specific screening tools tailored to the DS population and validating interventions for CVI and the long-term impact of CVI accommodations on functional outcomes and quality of life. Additionally, conducting cohort studies involving individuals with DS both with and without CVI, using standardized CVI semi-structured questionnaire interviews such as the Top-11 CVI Screener and HFVQI-51 could provide critical insights into the prevalence, spectrum and severity of HVFDs in this population. These studies should include neuroimaging analyses, including functional MRI (fMRI) (Hirsch et al., 2015), to identify potential biomarkers, anatomical, and neurological differences that may contribute to visual processing challenges, alongside looking at early neonatal, medical, and developmental characteristics contributing to CVI + DS versus DS alone.

Data availability statement

The datasets presented in this article are not readily available because they are the property of SeeLab, Smith-Kettlewell Research Institute. Permission must be granted by AC, via the corresponding author. Requests to access the datasets should be directed to arvind.chandna@alderhey.nhs.uk.

Ethics statement

Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

EB: Conceptualization, Investigation, Project administration, Supervision, Visualization, Writing – original draft, Writing – review & editing. RB: Conceptualization, Writing – original draft, Writing –review & editing. IW: Conceptualization, Writing – original draft, Writing – review & editing. KL: Conceptualization, Writing – review & editing. EM: Writing – review & editing. HM: Visualization, Writing – review & editing. MW: Data curation, Formal analysis, Validation, Visualization, Writing – review & editing. SV: Investigation, Writing – review & editing. AC: Investigation, Methodology, Supervision, Writing – original draft, Writing – review & editing. GH: Conceptualization, Supervision, Writing – review & editing.

Group members of DSMIG DS+CVI Workgroup

Elizabeth Boatwright, Independent Internal Medicine-Pediatrics Physician, Educator, and Researcher, Colorado Springs, CO, United States; Rudaina Banihani, DAN Women & Babies Program, Department of Newborn & Developmental Pediatrics, Sunnybrook Health Sciences Centre, Toronto, ON, Canada, Department of Pediatrics, University of Toronto, Toronto, ON, Canada; Ilse Willems, CVI Center, Perkins School for the Blind, Watertown, MA, United States; Kathleen Lehman, Institute on Human Development and Disability, University of Washington, Seattle, WA, United States, Department of Psychiatry & Behavioral Science, University of Washington School of Medicine, Seattle, WA, United States; Arvind Chandna, SeeLab, Smith-Kettlewell Eye Research Institute, San Francisco, CA, United States, Translational Clinical Eye Research Centre, Alder Hey Children's Hospital, Liverpool, United Kingdom; Gena Heidary, Department of Ophthalmology, Boston Children's Hospital and Harvard Medical School, Boston, MA, United States.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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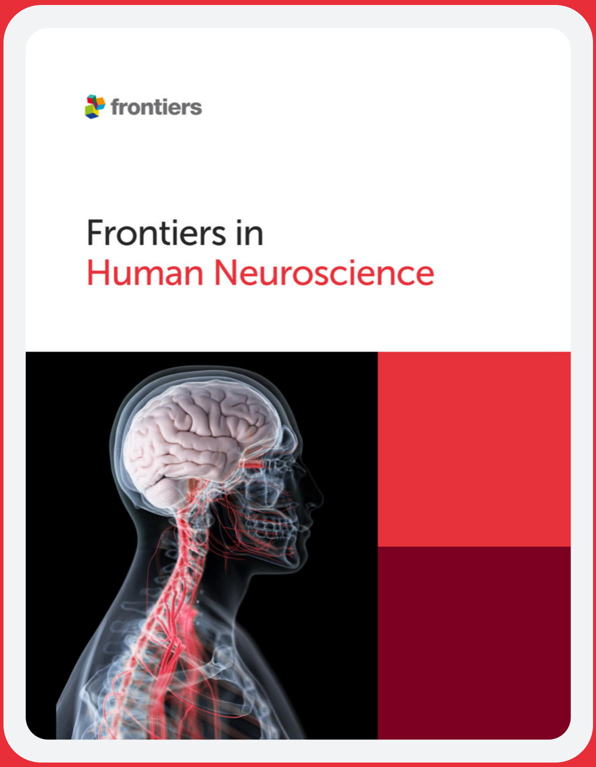

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