

# Impact of Visual Stimulation Therapy in the Management of Cerebral Visual Impairment in Paediatric Cerebral Palsy

## Impacto da Terapia de Estimulação Visual na Abordagem do Défice Visual Cerebral em Crianças com Paralisia Cerebral

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### ABSTRACT

**INTRODUCTION:** Cerebral visual impairment (CVI) is a common yet underdiagnosed cause of visual dysfunction in paediatric cerebral palsy (CP) for which there is not an approved therapy available. The aim of our study was to present our experience in CVI management in paediatric CP subjects involved in a visual stimulation therapy (VST) program.

**METHODS:** A group of paediatric CP patients with severe CVI was selected and submitted to a VST program between February 2022 and September 2023. Median number of VST sessions was 1 per week, conducted over an average follow-up period of 12 months. Visual Assessment Scale - Cerebral Visual Impairment in persons with Profound Intellectual and Multiple Disabilities (VAS CVI-PIMD) was applied for standardized and objective assessment of two parameters: 1) visual functioning level (VFL), and 2) total characteristics of CVI. Assessment was conducted by the same multidisciplinary team of occupational therapists and an ophthalmologist. VAS CVI-PIMD was re-measured after the intervention period to evaluate the effect of VST in the VFL, characteristics, and severity of CVI. A questionnaire was also administered to parents. A related samples Wilcoxon Signed Rank test was used to compare initial pre-VST scores with post-VST scores.  $p$ -values  $<0.05$  were considered statistically significant.

**RESULTS:** Eight children with CP and severe CVI (age range, 1 - 14 years) were selected. Initial median VFL was 3, corresponding to an exogenous attention system. After the VST program, 7/8 subjects improved at least one level, reaching a median VFL of 4 corresponding to a basic recognition and active visual attention system ( $p=0.014$ ). Total number of CVI characteristics in each patient decreased from a median score of 9 on the initial evaluation to a median score of 6 after the follow up period ( $p=0.013$ ). Parent questionnaire revealed a perception of improvement in eye-hand coordination skills and social functioning in everyday life.

**CONCLUSION:** In the present cohort, visual rehabilitation with appropriate stimuli for each stage of visual development leads to measurable gains in the visual performance of children with CVI.

**KEYWORDS:** Cerebral Palsy; Photic Stimulation; Vision Disorders; Vision, Low.

## RESUMO

**INTRODUÇÃO:** O déficit visual cerebral (DVC) é uma causa subdiagnosticada de disfunção visual em crianças com paralisia cerebral (PC) para a qual não existe uma terapia cientificamente aprovada. O nosso objetivo foi apresentar a nossa experiência na abordagem do DVC em crianças com PC submetidas a terapia de estimulação visual (TEV).

**MÉTODOS:** Um grupo de crianças com PC grave e DVC foi submetido a um programa de TEV entre fevereiro de 2022 e setembro de 2023. A mediana do número de sessões foi de 1 por semana, durante uma média de 12 meses. A escala *Visual Assessment Scale - Cerebral Visual Impairment in persons with Profound Intellectual and Multiple Disabilities* (VAS CVI-PIMD) foi utilizada para a avaliação padronizada de dois parâmetros: 1) nível de função visual (NFV) e 2) número de características do DVC. As avaliações foram realizadas pela mesma equipa multidisciplinar, constituída por terapeutas ocupacionais e um oftalmologista. A VAS CVI-PIMD foi re-aplicada após o período de intervenção para averiguar o efeito da TEV no NFV, número de características e gravidade do DVC. Também foi aplicado um questionário aos cuidadores. Utilizou-se o teste de Wilcoxon para mostras emparelhadas para comparar os scores pré-TEV com os scores pós-TEV. Valores  $p < 0,05$  foram considerados estatisticamente significativos.

**RESULTADOS:** Foram selecionadas oito crianças com PC e DVC grave (idades entre os 1 e 14 anos). O NFV médio inicial foi de 3, correspondendo a um sistema de atenção exógeno. Após o programa de TEV, 7 das 8 crianças melhoraram em pelo menos um nível alcançando um NFV médio de 4, que corresponde a uma percepção básica e sistema de atenção visual ativa ( $p=0,014$ ). A mediana do número de características do DVC diminuiu de 9 para um valor de 6 após a intervenção ( $p=0,013$ ). O questionário dos pais demonstrou uma percepção de melhoria na coordenação olho-mão e no funcionamento social na vida quotidiana.

**CONCLUSÃO:** Na coorte apresentada, o treino de visão com estímulos apropriados a cada etapa do desenvolvimento visual permitiu ganhos objetiváveis no desempenho visual dos pacientes com DVC. A TEV deverá fazer parte do programa de reabilitação a par das restantes terapias.

**PALAVRAS-CHAVE:** Baixa Visão; Distúrbios da Visão; Estimulação Visual; Paralisia Cerebral.

## INTRODUCTION

The spectrum of visual problems in paediatric cerebral palsy (CP) is extremely wide, ranging from peripheral pathologies such as refraction disorders and strabismus, to a problem of central origin which is cerebral visual impairment (CVI).<sup>1</sup> CVI, or cortical visual impairment, is defined as a confirmable visual dysfunction caused by malfunctioning of the retrogenic visual pathways, and that cannot be attributed to concomitant ocular impairment or anterior visual pathways disorders.<sup>2,3</sup>

It is estimated that approximately 30% of children with CP simultaneously have CVI.<sup>4</sup> CVI can be very difficult to diagnose. These children usually have limited verbal and cognitive abilities, and symptoms of CVI and degree of visual function are very heterogenous among them.<sup>5</sup> The co-existence of CP and CVI is due to the anatomical proximity of lesions in motor pathways, such as periventricular leukomalacia, to visual pathways.<sup>1,6</sup> As such, in children with CP, the severity of visual impairment frequently correlates with the severity of motor deficits.<sup>6</sup>

Neurobehavioral traits of children with CVI are different from those with exclusively ocular disorders.<sup>7</sup> Some

characteristics of paediatric CVI include: impaired visual attention, due to damage to the occipito-parietal pathway; sparing of colour vision, due to the bilateral hemispherical representation of colour perception; impaired contrast sensitivity, affected by the spatial frequency of the stimulus; increased latency of saccades and fixations; impaired recognition of faces, shapes and objects, due to damage to the occipito-temporal pathway; preferential view of objects at closer range, to decrease the crowding effect; and paradoxical light gazing, which was suggested to result from thalamic injuries.<sup>7-9</sup>

CVI severely impacts all aspects of children's development, including their ability to learn and perform daily life activities. In fact, children with both CP and CVI develop more slowly in the domains of self-care, mobility and social functioning than children with only CP.<sup>4</sup> In spite of the significant impact of visual impairment, functional therapy programs currently available do not take into account the presence of CVI and there is no evidence-based functional therapy for children with both CP and CVI.<sup>4</sup>

Previous studies<sup>10</sup> report some degree of visual recovery over time particularly in young children with CVI. However, this response is not always complete and frequently

these children do not reach a reasonable vision function level even after a long period of time.<sup>5,10</sup> In a retrospective study, Malkowicz *et al* proved that visual stimulation programs are able to improve a brain-injured child's ability to see significantly more when compared with a child that is not receiving visual stimulation therapy at all.<sup>5</sup> It now becomes clear, more than ever, the importance of a multidisciplinary approach in management of these group of children and the need to integrate the aspects from CVI in routine functional therapy to provide the best possible care to this group of children.<sup>4</sup>

The present work aims to present our experience in CVI management in paediatric CP patients involved in an intensive visual stimulation therapy (VST) program, to evaluate whether a customized, multidisciplinary visual rehabilitation approach facilitates the process of visual restoration by aiding the establishment of the necessary neuronal connections for CVI recovery.

## METHODS

### PARTICIPANTS

We recruited a cohort of paediatric patients with CP that were referred to Cerebral Palsy Association of Coimbra (APCC) by medical specialists performing a visual impairment screening.

APCC is a non-profit treatment centre located in Coimbra, Portugal, that provides a variety of services to children with cerebral palsy.

Inclusion criteria were diagnosis of CP, age range between 6 months and 18 years, and presence of CVI. CVI diagnosis in children with profound intellectual and multiple disabilities (PIMD) was based on the neuropsychological and functional definition proposed by Roman and colleagues.<sup>9</sup> According to their definition, the three criteria to determine CVI in individuals with PIMD are: 1) an eye examination that does not fully explain the child's visual abilities performed by an ophthalmologist, 2) a history or presence of neurological issues, in spite of normal brain imaging studies, and 3) the presence or behavioural or visual responses of CVI, such as those reported in the Introduction section.<sup>7-9</sup> Children presenting with severe visual deficits related to anterior visual pathway abnormalities (globe, retina and anterior optic nerve) were excluded.

This study was conducted in accordance with the Declaration of Helsinki. Informed consent was obtained from the legal representative of all participants before data collection.

### INTERVENTION

Visual stimulation sessions were individually tailored to the subject's initial visual function, namely in the assignment of specific stimuli, stimuli intensity, and session duration. The average duration of the Visual Stimulation Program was 12 months. Patients enrolled in weekly visual stimulation sessions for the entire program that lasted an average of 30 to 40 minutes, depending on the level of

cooperation from each patient. Each child had individual treatment goals, according to their degree of visual impairment and severity of CP.

The goal of the sessions was a stepwise stimulation towards progressively more complex stimuli. All the sessions were conducted in newly designed visual stimulation room, equipped with different materials of increasing visual complexity, including: glowing materials under ultraviolet light, lights and lighting material (such as a projector wheel), material with bright colours, high-contrast materials and everyday life materials (Fig. 1).



Figure 1. Visual stimulation room (A) and objects used in visual stimulation sessions: materials with bright colours and glowing under ultraviolet light (B), high contrast materials (C, D) and lighting material (E).

The sessions also included a period Snoezelen therapy, which were conducted in the Snoezelen room located at Cerebral Palsy Association of Coimbra (Associação de Paralisia Cerebral de Coimbra, APCC) (Fig. 2). This multi-sensory room is composed by a set of multi-sensory equip-

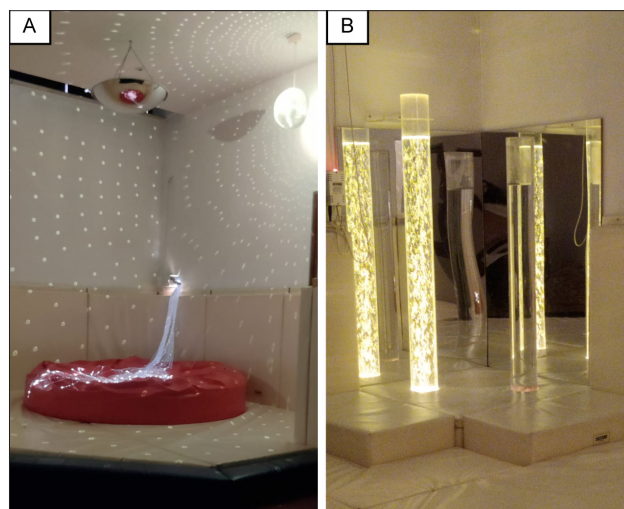
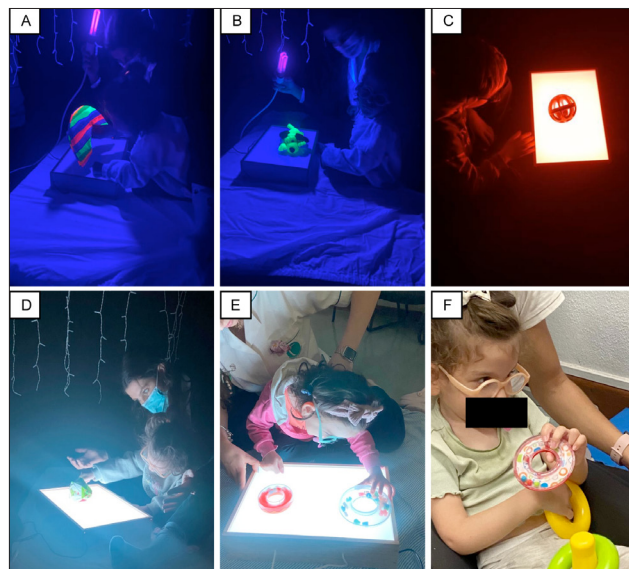


Figure 2. Snoezelen room: fibre optic bundle, revolving mirror ball and bean bag chair (A), multi-colour bubble tube (B).



ment, such as a multi-colour bubble tube, revolving mirror ball and colour wheel, fibre optic bundle with changing colours, magic glow panel (fluorescent board that glows under a bright light) and a bean bag chair.

Sessions were always performed by a trained occupational therapist (Figs. 3A to 3C).



**Figure 3.** Example of VST sessions conducted in the visual stimulation room (A, B, C). Figure D illustrates patient 1 first VST session, using lighting material and material with bright colours. A stepwise progression towards more complex stimuli was achieved during sessions (E). At the last VST session, the patient demonstrated improved eye-hand coordination and already manipulating daily life materials (F) under regular illumination.

## STUDY DESIGN

Our study was conducted between February 2022 and September 2023. It was a pretest-posttest quasi-experimental study that evaluated a targeted, individually defined visual rehabilitation program designed to improve the visual performance of children with CP and CVI conducted for an average period of 12 months in each of the patients.

Prior to the initiation of the VST sessions, Visual Assessment Scale - Cerebral Visual Impairment in persons with Profound Intellectual and Multiple Disabilities (VAS CVI-PIMD) was measured by the same Ophthalmologist before and after the intervention period to evaluate the effect of VST in the visual function, characteristics, and severity of CVI. A VAS CVI-PIMD questionnaire was also administered to caregivers on several aspects of everyday visual tasks, which was also re-assessed after the intervention period.

## OUTCOME MEASUREMENT INSTRUMENTS

A Portuguese version of the standardized Visual Assessment Scale - Cerebral Visual Impairment in persons with Profound Intellectual and Multiple Disabilities (VAS

CVI-PIMD) was used to measure the pretest and posttest outcome measures of the rehabilitation program (Appendix A). This version was developed by a low vision specialist ophthalmologist and trained occupational therapists in vision rehabilitation and is highly comparable to the original VAS CVI-PIMD in terms of word length, syntactical complexity and lexical difficulty, allowing the evaluation of all the original parameters in Portuguese speakers.

VAS CVI-PIMD is a visual scale that provides an objective and standardised assessment of the visual development of children with multiple disabilities, which monitors the development and allows the possibility to register progress on the visual behaviour of the child and measure the effect of the intervention in the area of visual rehabilitation.

The VAS consists of three parts:

### 1. Visual functioning level (VFL)

Gives information about visual development that is partly related to cognitive development. There six levels of visual functioning are:

Level 1 – Blind. Visual functioning is completely absent at this level, and the patient does not show any sign of visual reaction, even when in a visual stimulation room. This level is very rarely found in children with CVI.

Level 2 – Profound visual impairment and functional blindness. At this level, children show targeted visual behaviour and react to light stimuli in a darkened room through short fixation of the eyes and minimal pursuit.

Level 3 – Severe visual impairment and exogenous attention system. At this level, there is no search for the visual stimuli in an active way. Instead, visual stimuli are only perceived when introduced nearby and in the direction of gaze.

Level 4 – Moderate visual impairment and basic recognition and active visual attention system. At this level, the patient actively searches for visual stimuli of interest from daily life, can engage in some degree of eye contact and there is basic spatial recognition.

Level 5 – Mild visual impairment with extended visual recognition. At this level, patients show visual curiosity about their environment and are visually alert. There is already recognition of well-known people through vision only, without auditory clues, and fixation, pursuit and shifting gaze are well developed.

Level 6 – Normal visual functioning. This level is comparable to the visual development typically observed in a 24-month-old child.

### 2. Characteristics of CVI

The nine characteristics that are linked to CVI include: 1) absence of visual curiosity, 2) averting gaze while reaching or handling, 3) superficial gaze and brief visual behaviour, 4) variable visual behaviour, 5) unable to simultaneously use vision along with other senses, 6) tiring gaze, 7) improved recognition and visual behaviour with greater familiarity, 8) auditory over visual preference, and 9) staring into light sources.

The total score of these characteristics allows the classification of the presence and the degree of CVI.

### 3. Questionnaire for caregivers

A questionnaire was also provided to parents/caregivers, as they have the best insight into the daily visual functioning of these patients. The questionnaire collected the caregiver's information about visual behaviour, eye-hand coordination, social functioning, preference of other senses, orientation and mobility, recognition of familiar objects, and reaction to light (Appendix B).

### STATISTICAL ANALYSES

Data was analysed using the Statistical Package for Social Sciences (SPSS), v.29 software (Chicago, IL). The non-parametric Related Samples Wilcoxon signed rank test was used to compare pre- and post-treatment scores in VFL and CVI severity. A significance level of  $p < 0.05$  was used to detect significant differences. Descriptive statistics was used to describe all the variables assessed in VAS-PIMD, including nominal variables in the caregiver questionnaire, and median with range for categorical variables.

### RESULTS

Eight children with CP and CVI ( $n = 2$  males and  $n = 6$  females) with mean age 8 years old (standard deviation 4.31) participated in a VST program with weekly sessions for an average time of 12 months (range, 9-19), with a median number of VST sessions of 49.

Table 1 provides the characteristics of all children included in the study. The most common ethologic factor of CP and CVI was prematurity.

#### VFL

Related Samples Wilcoxon signed rank test indicated

that the median post-treatment VFL was statistically significantly higher than the median pre-treatment VFL ( $Z = -2.456$ ,  $p = 0.014$ ) (Table 2). Median VFL before the start of the VST was 3 (range, 2-4), corresponding to an exogenous attention system. After the VST program, median VFL achieved was 4 (range, 3-6) corresponding to a basic recognition and active visual attention system (Table 3). 7 of 8 patients showed improvement in at least one level (Table 3): one child improved 3 levels, one improved 2 levels, and 5 improved one level. The only patient that maintained the same pre-treatment value had missed the weekly VST sessions for the previous 3 months before the post-VST VAS-PIMD was applied.

### CVI CHARACTERISTICS

Related Samples Wilcoxon signed rank test indicated that the median post-treatment results were statistically significantly lower than the median pre-treatment results ( $Z = -2.536$ ,  $p = 0.011$ ) (Table 2). Median number of CVI characteristics before the initiation of VST program was 9 (range, 7-9), decreasing to 6 (range, 3-8) after the intervention period. All patients improved at least one characteristic (Table 3): two improved 6 characteristics, three improved in 5 characteristics, two improved in 4 characteristics, and one improved in only 1 characteristic. Of note, this was also the child with no improvement in VFL, due to an irregular attendance to VST sessions for the previous three months.

Fig. 3D illustrates one of our patients at the beginning of the VST program, where she had an initial VFL of 3 and 9 total characteristics of CVI, corresponding to a severe visual impairment. At this level, VST included mostly materials under ultraviolet light, lighting material and material with bright colours. She went through the VST with progressively more complex stimuli (Fig. 3E). Fig. 3F illustrates the same patient in the last session of our program, where she now had a VFL of 5 and 3 total characteristics of CVI, corre-

**Table 1.** Description of the study's participants ( $n=8$ )

Participant	Age (years)	Gender	Presence of Epilepsy	CVI etiology	GMFCS
1	4	Female	Yes	Prematurity	IV
2	6	Female	Yes	Prematurity	V
3	14	Female	Yes	Hypoxic-ischemic encephalopathy	V
4	10	Female	Yes	Prematurity	III
5	10	Female	Yes	Prematurity	V
6	12	Male	Yes	Prematurity	IV
7	7	Male	Yes	Perinatal ischemic stroke	III
8	1	Female	No	Perinatal haemorrhagic stroke	III

CVI - cerebral visual impairment, GMFCS - gross motor function classification system.

**Table 2.** Comparison between pre-VST and post-VST VFL and CVI severity

VFL and CVI Severity	Pre-VST median (min-max)	Post-VST median (min-max)	p-value (95%CI)	Z-score
VFL	3 (2-4)	4 (3-6)	<b>0.014</b> (0.500; 2.000)	-2,456 <sup>a</sup>
CVI characteristics	9 (7-9)	6 (3-8)	<b>0.011</b> (-4.500; - 2.000)	-2,536 <sup>b</sup>

a - based on negative ranks; b - based on positive ranks.

CVI - cerebral visual impairment, VFL - visual functioning level, VST - visual stimulation therapy.

Participant	VFL (pre-post-VST) difference	CVI severity (pre-post-VST) difference	Number of VST sessions
1	(3-5) 2	(9-5) 4	39
2	(2-3) 1	(9-3) 6	65
3	(3-4) 1	(9-4) 5	43
4	(3-6) 3	(7-6) 1	39
5	(2-3) 1	(9-3) 6	65
6	(3-4) 1	(9-4) 5	43
7	(4-4) 0	(9-4) 5	69
8	(3-4) 1	(8-4) 4	30

CVI - cerebral visual impairment, VFL - visual functioning level, VST - visual stimulation therapy.

sponding now to a moderate visual impairment according do VAS-PIMD, and was able to maintain eye-hand coordination while manipulating everyday life materials under regular illumination.

### CAREGIVER QUESTIONNAIRE

This was a subjective questionnaire that was provided to parents/caregivers on their perception of the evolution of visual behaviour in several domains. Among the 8 questionnaires, in general, a perception of greater improvement was noted in the areas of eye-hand coordination skills and social functioning in everyday life.

### DISCUSSION

Our results showed that VST significantly improves the basic visual skills of children with CP and concomitant CVI. In VFL assessment, 7 of 8 patients improved at least one level, reaching an average basic recognition and active visual attention system. Also, all patients improved in at least one CVI characteristic, and caregivers reported significant improvement in eye-hand coordination skills and social functioning after the institution of the VST program. Now, more than ever, we understand that all children with CVI benefit from interventions that allow them to overcome functional limitations and maximize their visual development.<sup>2</sup>

The most common cause of CP and CVI found in our subjects was prematurity, followed by perinatal cerebral vascular accident and hypoxic-ischemic encephalopathy. This in agreement with past studies, which state that CVI is most frequent in the following conditions: hypoxic-ischemic encephalopathy, peripartum stroke, periventricular leukomalacia, infection, chromosomal and metabolic abnormalities, and traumatic brain injury.<sup>9,11</sup> It is estimated that around 80% of paediatric CP is caused by insult *in utero*, with subsequent injuries to the developing brain.<sup>12</sup> The prevalence of CP in developed countries is about 2-3 per 1000 live births, however, it significantly increases to 40-100 per 1000 live births in children born prematurely below 28 weeks' gestation.<sup>12</sup>

Our results showed that visual functioning levels were both clinically and statistically significantly influenced and

improved by a weekly VST conducted by occupational therapists trained in visual rehabilitation. Prior to the beginning of the sessions, the level of visual impairment was on average classified as severe by the VAS-PIMD, meaning these children would only react to strong stimuli in broad daylight and when the stimulus was shown in the field of vision, make very short incidental eye contact, have a limited range of vision up to an arm's length, and have short fixation and limited visual pursuit. After the VST, visual functioning level increased to an average of moderate impairment, in which patients actively search for visual stimuli of interest, develop object permanence, and recognise up to ten daily objects and familiar faces.

In a study conducted in 2003, Hoyt *et al*<sup>10</sup> evaluated the recovery of 96 children with CVI without any visual stimulation program for an average period of 5.9 years, and concluded that more than half of children did not improve whatsoever when visual stimulation was not received. This raises the obvious question of whether a visual stimulation program improves the visual abilities of children with CVI. In this same study,<sup>10</sup> among the group that did receive a visual stimulation program, only 1 of them showed no improvement. In another study by Malkowicz and colleagues,<sup>5</sup> 21 children with CVI were submitted to an intensive visual stimulation program, with the sessions performed at the participant's home after parents were given the proper instruction in the visual stimulation procedures. They found that 20 out of 21 children had a significant improvement after 4 to 13 months on the program.<sup>5</sup> Our results are in accordance with both of these works. Indeed, we found that the VST program significantly affects the development of these children's visual function, as well as significantly and positively influences the course of CVI. Despite our limited sample, we suggest that the progress found in our cohort resulted from the visual intervention and not only due to natural evolution of the visual performance in children with CP.

Visual information plays a vital role in the overall development of a child.<sup>13</sup> An increase in the visual skills of children with CP leads to an increased ability to communicate, interact with the outside world, and participate in activities of daily life.<sup>13</sup> However, these gains do not necessarily correspond to measurable objective gains in medical-diagnostic parameters such as visual acuity.<sup>13</sup> Our study



also evaluated the influence of VST in the patient's interaction with the outside world by the uniform VAS-PIMD questionnaire that was answered by caregivers, which gives the best insight into daily visual functioning of these children. With the obvious limitation of being a subjective questionnaire, the caregivers noted that the aspects that had the greatest improvement were those involved in eye-hand coordination tasks, which often include directed movements towards a desired object, and social functioning in everyday life, namely making eye contact and distinguishing between known and unknown people without the use of auditory clues.

There are several explanations to why children with several degrees of CVI seem to have some improvement over time. First, it is possible that the normal maturation process of the visual system that occurs in normally developing children allows the emergence over time of the residual visual potential present in these brain-damaged children.<sup>10</sup> Second, it might be that the damage of the visual cortex is only incomplete in children with CP, and thus the residual visual function is generated by the residual intact visual cortex.<sup>10</sup> However, the most currently accepted theory attributes the apparent natural visual improvement to a mechanism of adaptive neuroplasticity, by which neuronal networks of the developing brain would be able to amplify its visual function by activating, modulating, and strengthening visual signals.<sup>6,10,14</sup> However, further studies are needed for clarification on this matter.

Stimulation programs are based on the premise that repeated stimulation will positively reinforce and elicit favourable responses in the child's visual behaviour.<sup>15</sup> Such behaviours are, for example, fixation, pursuit, and eye-hand coordination. Visual rehabilitation is defined as all interventions aimed at restoring visual abilities, improving visual functioning, and helping patients to cope with their visual disabilities.<sup>15</sup> It encompasses several techniques that are sometimes erroneously used as synonyms: vision stimulation and visual stimulation or training.<sup>15</sup> Vision stimulation is aimed at improving the visual system by enhancing and modifying its anatomy and physiology.<sup>15</sup> In contrast, visual stimulation consists in promoting the development of children with visual impairment through the use of visual material, aiming to improve their visual functioning.<sup>15</sup> One example of visual stimulation technique is Snoezelen. Snoezelen derives from the blending of two Dutch words, "snuffelen" (meaning "to explore") and "doezelen" (meaning "to relax"). This term is used to describe a method of controlled sensory stimulation in a non-threatening environment involving all sensory systems.<sup>16</sup> Kwok *et al.* had previously proposed seven functions of the Snoezelen room: relaxation, self-confidence development, self-control achievement, exploration encouragement, rapport establishment with care takers, enjoyment provision, choice promotion, attention span improvement, and reduction of challenging behaviours.<sup>17</sup>

VST seem to promote visual recovery by providing the injured brain with a chance to form appropriate new con-

nections by increasing the frequency of daily instances of random stimulation.<sup>5</sup> The neurobiological basis which explains VST results seems to be through the strengthening of synaptic connections via repetitive activation of neuronal networks.<sup>14</sup> VST sessions purposefully stimulate neurons to generate action potentials, which further promotes the release of neurotrophic factors from the post-synaptic membrane that will stimulate pre-synaptic synapse formation and improve neurotransmission.<sup>14</sup>

There are some limitations to our study which should be noted. First, we did not include a control group of patients that did not receive VST, since withholding a beneficial intervention from some of the study's participants would not only be unethical but also potentially harming. As such, we cannot exclude that other factors besides our intervention may be influencing our results. The second limitation of our study is the reduced size of our sample ( $n = 8$ ). Thus, we cannot exclude an overestimation of the magnitude of the associations we encountered. Finally, we made no correlation between the improvement in visual skills and structural abnormalities of the brain of our patients. Thus, it would be interesting for further studies to address this association with a larger sample.

## CONCLUSION

CVI remains a significant therapeutic challenge for children with CP. Our work suggests that visual rehabilitation with appropriate stimuli for each stage of visual development could potentially lead to measurable gains in the visual performance of children with CVI. Since 85% of the information we gather from the world is acquired through sight, visual stimulation approaches must become an integral part of the routine rehabilitation programs conducted in children with CP.

## CONTRIBUTORSHIP STATEMENT / DECLARAÇÃO DE CONTRIBUIÇÃO:

IF: Writing, literature research and editing of the manuscript.

AR, MC, IM, LA, CS, JM, CP: Review and supervision of the manuscript.

All authors approved the final version to be published.

IF: Redação, pesquisa bibliográfica e edição do manuscrito.

AR, MC, IM, LA, CS, JM, CP: Revisão e supervisão do manuscrito.

Todos os autores aprovaram a versão final a ser publicada.

Appendix A. Portuguese VAS-PIMD score

Nome	
Idade	
Datas de observação	
Nome e posição do Observador	

Nível 1- Déficit total de visão/ cegueira				Sim	Não	Outro/n.a.
1.1 Nenhuma reação aos estímulos visuais						

Nível 2- Deficiência visual profunda / cegueira funcional				Sim	Não	Outro/n.a.
2.1 Reage a estímulos de luz numa sala escura por meio de um comportamento de aparência direcionado						
2.2 Reage acalmado ou olha numa direção geral a fortes estímulos visuais numa sala normalmente iluminada						
Comportamento de aparência geral e competências visuais:						
2.3 Fixação muito curta (menos de 1 contagem)						
2.4 Capacidade mínima para a procura visual (nem sempre ocorre)						

Nível 3- Comprometimento grave / Sistema de atenção exógena				Sim	Não	Outro/n.a.
3.1 Reage a fortes estímulos visuais em plena luz do dia, com comportamento visual direcionado						
3.2 Pode perceber um estímulo visual quando é oferecido no campo de visão, não procura ativamente por estímulos visuais						
3.3 Mostra atenção visual em plena luz do dia quando acionado por:						
A. Movendo objetos ou crianças						
B. Um estímulo auditivo						
3.4 Faz contato visual acidentalmente						
3.5 É capaz de mostrar preferência por determinados estímulos, sem indicação clara de reconhecimento						
Comportamento de aparência geral e competências visuais:						
3.6 O alcance da visão é limitado, até o comprimento do braço						
3.7 Fixação curta (1-2 contagens)						
3.8 Procura visual limitada						

Nível 4- Comprometimento moderado/ percepção básica e sistema de atenção visual ativo				Sim	Não	Outro/n.a.
4.1 Procura ativamente por estímulos visuais (que sejam de seu interesse)						
4.2 Olha para os objetos da vida cotidiana de uma forma interessada, sem atenção ou quase nenhuma atenção aos detalhes						
4.3 Segue visualmente um brinquedo que cai no chão (permanência do objeto)						
Reconhecimento:						
4.4 Reconhece 1 a 10 objetos diários, por exemplo, o copo, pano de limpeza ou colher e reage adequadamente, sem entrada auditiva ou tátil						
4.5 Reconhece rostos familiares sem estímulos auditivos (alerta elevado, sorriso, alcance, etc.)						
4.6 Reconhece o brinquedo (favorito), sem entrada auditiva e reage de forma adequada						
4.7 Reconhece áreas quando, mais ou menos por acidente, entrar nelas (orientação espacial básica)						
Comportamento de aparência geral e competências visuais:						
4.8 Olhando para uma distância de aproximadamente 1 metro, as crianças em movimento são seguidas até 2-3 metros						
4.9 Perseguição visual está presente						
4.10 Mudança de olhar está presente						
4.11 Faz contato visual regularmente						

Nível 5- Comprometimento leve/ reconhecimento visual estendido				Sim	Não	Outro/n.a.
5.1 Alerta visual: monitoriza ativamente os arredores visualmente						
5.2 Alguma atenção aos detalhes, por exemplo, vê migalhas na mesa						
5.3 Procura contato visual à distância (além de 50 cm)						
Reconhecimento visual / atenção seletiva						
5.4 Reconhece mais de 10 objetos						
5.5 Reconhece objetos e crianças familiares em fotografias que não estão muito preenchidas						
5.6 Pesquisa um determinado objeto entre uma quantidade limitada de objetos						
5.7 É capaz de se orientar em ambientes familiares						
Comportamento de aparência geral e competências visuais:						
5.8 A distância de observação é de até 2-3 metros, se a acuidade visual permitir						
5.9 Usa a visão na comunicação (reage a imitações e gestos)						
5.10 Tenta obter uma visão geral enquanto olha ativamente ao redor de uma sala						
5.11 Fixação, busca e deslocamento do olhar estão bem desenvolvidos, o varrimento pode ter começado						

Nível 6- Não existe comprometimento/ funcionamento visual normal (apropriado para a idade de desenvolvimento de 24 meses)				Sim	Não	Outro/n.a.
Reconhecimento visual / atenção seletiva						
6.1 Procura ativamente os brinquedos favoritos que não são visíveis (sinal de memória visual)						
6.2 Quando um objeto é apontado na distância a criança vai vê-lo						
6.3 Reconhece objetos através de pistas visuais (por exemplo, reconhece uma bicicleta ao ver apenas o guidador)						
6.4 Mostra atenção conjunta (por exemplo, faz contato visual e mostra um brinquedo, aponta para um objeto ou traz para mostrar)						
6.5 Imitar comportamento (por exemplo, acenar, sorrir, franzir o nariz)						
6.6 Compreende objetos / crianças / ações em pictogramas						
6.7 Está interessado em detalhes (por exemplo, em livros ricamente ilustrados). É capaz de procurar um objeto rapidamente (varrimento visual).						

CVI: características				Sim	Não	Outro/n.a.
1. Sem curiosidade visual						
2. Desviando o olhar ao alcançar ou manusear objetos						
3. Comportamento visual curto e fugaz						
4. Comportamento visual variável						
5. Incapacidade de usar a visão simultaneamente com outros sentidos, como ouvir ou tocar						
6. Olhar é cansativo						
7. A familiaridade proporciona um melhor comportamento visual e / ou reconhecimento						
8. Prefere o auditivo ao visual						
9. Olhando para as fontes de luz						
Número total de características do CVI						

Nível de funcionamento visual	Número total de características	Diagnóstico
2 e 3	3-9	CVI Severo
4-6	6-9	CVI Severo
4-6	3-5	CVI Moderado
4-6	0-2	Sem CVI

Número total de características do CVI \ Nível de função visual	0	1	2	3	4	5	6	7	8	9
	2									
3										
4										
5										
6										



Appendix B. Portuguese caregivers' questionnaire

Comportamento visual e interesses						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	tem interesse visual no seu ambiente					
2.	movimentos vagos ou olhar perdido					
3.	adota uma posição da cabeça incomum ao olhar					
4.	olha para fontes de luz, como por exemplo, uma lâmpada ou uma janela					
5.	brinca com a luz, por exemplo, movendo os dedos					
6.	tem preferência por materiais de alto contraste e brilhantes					
7.	responde principalmente ao movimento de crianças' objetos					
8.	olha para cima quando alguém entra numa sala					
9.	mostra interesse visual em objetos					
10.	vê televisão / computador / tablet					
11.	vê pequenos objetos como migalhas, granulado ou missangas					
12.	aproxima-se dos objetos para vê-los melhor					
13.	mostra comportamento visual variável (às vezes percebe objetos e às vezes não)					
14.	nota a mudança no ambiente ou nas outras crianças					
	Outros:					
Informação adicional:						

Coordenação olho-mão (apenas para ser preenchida se as competências motoras permitirem)						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	revela movimentos dirigidos quando quer agarrar um objeto					
2.	dirige a mão e tenta agarrar o objeto					
3.	agarra em pequenas coisas como migalhas ou missangas de uma maneira direcionada					
4.	agarra em objetos do tamanho de uma banana					
5.	agarra em objetos com a mão aberta					
6.	agarra objetos usando o polegar e o dedo indicador (preensão em pinça)					
7.	desvia o olhar quando quer agarrar em algo e, em seguida, usa apenas o sentido do tato para pegá-lo					
	Outros:					
Informação adicional:						

Funcionamento social						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	faz contato visual ao perto					
2.	faz contato visual à distância					
3.	segue as crianças a caminhar pela sala					
4.	dá a impressão de olhar através de si					
5.	distingue crianças conhecidas das desconhecidas					
6.	imita expressões faciais e / ou gestos					
7.	reconhece uma criança familiar (sem a ajuda de reconhecimento de voz)					
8.	é assustado por um som inesperado					
9.	fica assustado se alguém se aproxima dele sem avisar					
10.	responde a expressões faciais (sem som)					
11.	reconhece crianças sem o uso de som / voz					
	Outros:					
Informação adicional:						

Outros sentidos						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	prefere estímulos auditivos, ouve antes de olhar					
2.	tem forte preferência por atividades auditivas, jogos com som, música ou outros sons (caseiros)					
3.	pesquisas tateando para encontrar um objeto pelo toque					
4.	usa o toque para explorar objetos					
5.	cheira objetos e / ou comida					
	Outros:					
Informação adicional:						

Orientação e mobilidade						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	está interessado em explorar o ambiente					
2.	tropeça ou cai em coisas regularmente					
3.	move-se hesitantemente quando há diferenças de cores no piso					
4.	muitas vezes hesita em prosseguir quando há curvas e degraus					
5.	mostra que reconhece um lugar familiar					
6.	consegue orientar-se em ambientes bem conhecidos					
	Outros:					
Informação adicional:						

Reconhecimento						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	Tem uma clara preferência por certas cores. Em caso afirmativo, que cores?					
2.	É capaz de reconhecer objetos					
3.	É capaz de nomear objetos					
4.	Faz escolhas entre dois objetos (quando apenas mostrados visualmente)					
5.	Consegue escolher um objeto solicitado de uma pilha de objetos					
6.	Reconhece objetos em fotografias/imagens					
7.	Reconhece família e/ou cuidadores em fotografias					
8.	Consegue combinar imagens (por exemplo, jogo de memória)					
9.	Pode encontrar um objeto se estiver em uma superfície visualmente ocupada (por exemplo, mesa cheia, toalha de mesa ocupada)					
10.	Segue um objeto caído visualmente					
11.	procura um objeto caído					
12.	responde adequadamente a gestos familiares de um adulto					
	Outros:					
Informação adicional:						

Reações à luz						
	Ele/Ela	Nunca	Raramente	Frequentemente	Sempre	N/A
1.	Fecha os olhos com a luz brilhante e não tolera o brilho do sol					
2.	Procura e parece preferir áreas bem iluminadas					
3.	Procura e parece preferir áreas mais escuras					
	Outros:					
Informação adicional:						

## RESPONSABILIDADES ÉTICAS

**Conflitos de Interesse:** Os autores declaram a inexistência de conflitos de interesse na realização do presente trabalho.

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**Proteção de Pessoas e Animais:** Os autores declaram que os procedimentos seguidos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínquia revista em 2013 e da Associação Médica Mundial.

**Proveniência e Revisão por Pares:** Não comissionado; revisão externa por pares.

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