Original Article

Visual Function Classification System for children with cerebral palsy: development and validation

Giovanni Baranello, [1,2,✉]
Email giovanni.baranello@istituto-besta.it

Sabrina Signorini, [3]

Francesca Tinelli, [3]

Andrea Guzzetta, [4]

Emanuela Pagliano, [1]

Andrea Rossi, [5]

Maria Foscan, [1]

Irene Tramacere, [6]

Domenico M M Romeo, [7]

Daniela Ricci, [8]

On Behalf of the VFCS Study Group

Riccardo Zanin,

Elisa Fazzi,

Giovanni Cioni,

Eugenio Mercuri,

AQ1

AQ2

[1] UO Neurologia Dello Sviluppo, Fondazione IRCCS Istituto Neurologico Carlo Besta, Milan, Italy

[2] The Dubowitz Neuromuscular Centre, UCL NIHR GOSH Biomedical Research Centre, Great Ormond Street Institute of Child Health, London, UK

[3] Centre of Child Neuro-Ophthalmology, Unit of Child Neurology and Psychiatry, IRCCS Mondino Foundation, Pavia, Italy

[4] Department of Developmental Neuroscience, IRCCS Stella Maris Foundation, Pisa, Italy

[5] Child and Adolescent Neurology and Psychiatry Unit, Children Hospital, ASST Spedali Civili of Brescia, Brescia, Italy

[6] Department of Research and Clinical Development, Scientific Directorate, Fondazione IRCCS Istituto Neurologico Carlo Besta, Milan, Italy

[7] Pediatric Neurology Unit, Fondazione Policlinico Universitario A Gemelli IRCCS, Rome, Italy

[8] National Centre of Services and Research for Prevention of Blindness and Rehabilitation of Visually Impaired, Rome, Italy AQ3

Correspondence to Giovanni Baranello, UO Neurologia dello Sviluppo, Fondazione IRCCS Istituto Neurologico Carlo Besta, Via Celoria 11, Milan 20133, Italy. E-mail: giovanni.baranello@istituto-besta.it

Aim

To develop and validate the Visual Function Classification System (VFCS), which was created to classify how children with cerebral palsy (CP) use visual abilities in daily life.

Method

The process of development and validation of the VFCS involved four phases: (1) drafting of the five levels from the analysis of literature and clinical experience; (2) validation of constructs and revision of the levels for meaningfulness of the concepts, using nominal group process; (3) refinement by international Delphi survey; (4) assessment of interrater reliability among professionals and with caregivers, and of test–retest reliability.

Results

Five nominal groups involved 29 participants; 65 people completed the first round and 51 the second round of the Delphi Survey. Construct validity was demonstrated within an expert group and external validation through several stakeholders, with the involvement of patients and families to ensure meaningfulness of the concept. Discussions continued until consensus was reached about the construct and content of the five levels. Participants in the reliability study included 29 professionals, 39 parents, and a total sample of 160 children with CP (mean age [SD] 6y 6mo [3y 4mo]; median 5y 7mo, range 1–19y). Absolute interrater agreement among professionals was 86% (weighted κ=0.88; 95% confidence interval [CI] 0.83–0.93). Test–retest reliability was high (weighted κ=0.97; 95% CI 0.95–0.99). Parent–professional interrater reliability on 39 children was moderate (weighted κ=0.51; 95% CI 0.39–0.63).

Interpretation

The VFCS has been appropriately constructed and provides a reliable system to classify visual abilities of children with CP both in clinical and in research settings.

Additional members of the VFCS Study Group can be found in the Acknowledgements.

Abbreviations

ASD Autism spectrum disorder
CFCS Communication Function Classification System
EDACS Eating and Drinking Ability Classification System
ICF International Classification of Functioning, Disability and Health
MACS Manual Abilities Classification System
VFCS Visual Function Classification System

What this paper adds

- The Visual Function Classification System is a valid and reliable system.
- It classifies visual abilities of children with cerebral palsy in clinical and research settings.
- At a clinical level, it can be used to harmonize communication among professionals and identify patients' strengths and weaknesses.
- In research settings, it can be used to stratify patients, define natural history evolution, and interpret intervention studies.

Some degree of visual impairment is reported to affect up to half of the children with cerebral palsy (CP), either as a consequence of the brain damage itself, or because of the involvement of peripheral visual structures, or as a combination of both.[1, 2, 3, 4, 5, 6, 7] This is also clearly stated in the new definition of CP, which suggests that the visual–perceptual disorders, along with the motor disorder, are an integral part of the clinical picture of CP.[8] Although motor impairment is a requirement for the diagnosis of CP, the so-called associated disorders, such as disturbances of sensation, perception, cognition, communication, and behaviour, may impact daily life of children with CP even more than the motor disorder per se.[8, 9, 10] Surprisingly, it is still common that the assessment of vision relies only on the ophthalmological examination, and only rarely on the functional evaluation of different aspects of visual abilities, namely how the child performs in vision-related activities.[9, 10, 11, 12, 13] Therefore, children with CP often start rehabilitation programmes without specific information on their visual functioning available to

their therapists.[14, 15] The World Health Organization's International Classification of Functioning, Disability and Health (ICF) highlighted the importance of evaluating the functional consequences of all health states.[16, 17] Over the past 20 years there has been an increasing awareness of the importance to classify the way the impairments underlying CP affect activity and participation.[18] Accordingly, new classification systems for gross motor, manual, communicative, and eating/drinking abilities have been developed.[19, 20, 21, 22] All these classifications emphasize the need to describe children's abilities rather than their impairments, focusing on how they typically behave in real-world situations. For this, the levels of abilities in the current Visual Function Classification System (VFCS) were derived empirically to describe usual performance rather than best capacity in daily life.[23]

Although vision impairment may markedly affect the quality of life of a child with a developmental disorder, no system is currently available to classify levels of visual functioning analogous to the existing functional classification systems for children with CP.[24, 25] For this reason, a group of experts in the field of CP and visual disorders, with the contribution of parents and patients, set out to develop a new classification system for visual function in children with CP. The aim of this study was to develop and validate the VFCS, which was created to classify how children with CP use visual abilities in their daily life, focusing on activity and participation as described in the ICF.

Method

The process of development and validation of the VFCS was similar to that used for the development and validation of the classification systems already available for CP, namely the Gross Motor Function Classification System (GMFCS),[19] the Manual Abilities Classification System (MACS),[20] the Communication Function Classification System (CFCS),[21] and the Eating and Drinking Ability Classification System (EDACS),[22] and involved four phases. In the first phase, a development team drafted an initial version starting from the analysis of the literature[1, 2, 3, 4, 5, 6, 10, 11, 12, 14, 15] and the clinical experience of participants. The first draft was then examined and revised using several iterations of a nominal group process. Two rounds of an online Delphi survey further revised the VFCS (Appendix S1, online supporting information), until agreement about the content and wording was reached. The fourth phase assessed interrater reliability among professionals and with caregivers, as well as test–retest reliability. Ethical approval for this research was granted by the institutional review board at the Fondazione IRCCS Istituto Neurologico Carlo Besta, Milan, Italy, and accepted by the other institutional partners.

Phase 1: drafting the VFCS

Participants

Fifteen participants from five groups of people with experience with CP, visual impairment, and assessment participated in this phase: an adult with CP, two

parents of children with CP, one educator, eight child neurologists, two therapists, and an ophthalmologist.

Procedures

The participants discussed and developed the first draft of the VFCS, on the basis of reviews of the literature on visual abilities in children with CP and the participants' experiences in real-life situations. Participants shared their ideas through a series of five meetings, three conference calls, and subsequent e-mails, discussing and suggesting revisions. One important step of this phase was the dynamic process of construct development of the classification, starting from the conceptual background. A hierarchical algorithm model was used to define five levels of visual abilities with reference to the key features of 'independence' and 'external support'. Independence referred to the consistent and successful use of vision in vision-related daily activities without adaptations, other sensory modalities, or external support. External support referred to the need for assistance and/or adaptations of the environment to perform vision-related daily activities. Each element of the VFCS was evaluated for usefulness and clarity of wording.

The initial development process ended when the participants were satisfied with the VFCS draft. The draft was then evaluated by the nominal groups in the next phase.

Phase 2: nominal groups

Participants

This process involved 29 participants from Italy. Their backgrounds are reported in Table 1.

Table 1 Backgrounds of participants in nominal group process and Delphi surveys

Background	Nominal group, *n*=29 (%)	Delphi survey round 1, *n*=65 (%)	Delphi survey round 2, *n*=51 (%)
Individuals with CP	5 (17)	8 (12)	4 (8)
Parents of individuals with CP	6 (20)	12 (18)	6 (12)
Therapists (including physiotherapists, occupational therapists, orthoptists)	7 (26)	15 (23)	12 (23)
Educators	3 (10)	12 (18)	9 (18)
Neurologists	5 (17)	8 (13)	11 (21)
Ophthalmologists	2 (7)	9 (14)	7 (14)
Others	1 (3)	1 (2)	2 (4)

CP, cerebral palsy.

Procedures

The nominal group process facilitates discussions among individuals with the purpose of creating a consensus opinion.[26] Five nominal group rounds, involving the various stakeholders, were held in different locations in Italy. Participants examined the content of the VFCS and suggested changes. Participants received by e-mail the last version of the VFCS at least 1 week before the nominal group meeting, and were invited to prepare comments and suggestions to be discussed in the meeting. Each group was given the opportunity to discuss the VFCS during a 6-hour time block. The first and last authors participated in all the group meetings to promote equality of participation among participants; facilitate discussion; ask questions; move between topics; restate participants' comments, suggestions, and ideas; and provide structure during the meeting. The purpose, content, appropriateness, consistency, and clarity of the VFCS draft were discussed, and participants were encouraged to suggest changes. Each group was asked to vote on the various suggestions until consensus was reached.

Phase 3: Delphi surveys

Participants

Sixty-five international participants across five continents, with recognized experience in CP, including parents, individuals with CP, and professionals, were recruited to take part in the Delphi survey. All participants from round 1 were invited to participate in round 2, and 37 of the original 65 completed round 2. Fourteen additional participants were invited to participate in round 2. See Table 1 for participants' characteristics.

Procedures

Delphi surveys provide a structured method for stakeholders to provide anonymous feedback.[27, 28] The purpose of the surveys was to reach a large number of individuals with expertise in vision and CP, to evaluate the clarity, conciseness, consistency, and usefulness of the VFCS draft. Delphi surveys were conducted in rounds until a predefined 80% target agreement was reached on all closed-ended questions. The round 1 Delphi survey, with 18 questions similar to those used in the nominal groups and the development of some of the other classification systems in CP, was completed in a paper-based format (questions are listed in Appendix S1). A second Delphi survey (round 2) was held for the three questions that did not reach the targeted 80% agreement. Open-ended responses were analysed and taken into account for possible changes to the VFCS.

Participants were asked to examine the content of the VFCS represented using both open-ended questions and questions with possible answers on seven-point Likert scales (1, strong disagreement; 4, neither agree nor disagree; 7, strong agreement).

Phase 4: reliability

Participants

Participants in the reliability study included 29 professionals, 39 parents, and 160 children with CP (mean age [SD] 6y 6mo [3y 4mo]; median 5y 7mo, range 1–19y). Other demographic information for the children and young people included sex, GMFCS level, CP subtype, presence of intellectual disability, epilepsy, and autism spectrum disorder (ASD) and/or behavioural problems (Table 2).

Table 2
Demographics of children (*n*=160) with cerebral palsy (CP) included in reliability studies

Age	
Range	1–19y
Median	5y 7mo
Mean	6y 6mo (SD 3y 10mo)
Sex: males/females	85/75
GMFCS level (*n*, %)	
I	46 (29)
II	28 (17)
III	22 (14)
IV	24 (15)
V	40 (25)
CP classification according to SCPE, *n* (%) (predominant limb involvement)	
Spastic unilateral	12 (7.5)
Spastic bilateral	126 (79)
Diplegia	69 (43)
Quadriplegia	57 (36)
Dyskinetic	21 (13)
Ataxic	1 (0.5)
Comorbidities, *n* (%)	
Intellectual disability	
Mild	23 (14)

Moderate	38 (24)
Severe	25 (16)
Epilepsy	48 (30)
ASD/behavioural problems	21 (13)

GMFCS, Gross Motor Function Classification System; SCPE, Surveillance of Cerebral Palsy in Europe; ASD, autism spectrum disorder.

Procedures

To test interrater reliability among professionals, 15 pairs of professionals involved in a child's rehabilitation programme and/or health care service scored visual abilities of the same child (in the total sample of 160 children with CP) independently. As clearly indicated in the VFCS instructions provided to raters, the classification should be made by someone who knows the child and their abilities in daily vision-related activities. To assess test–retest reliability (weighted κ), the 29 professionals reclassified a subset of 132 children at least 2 weeks after the first classification, being sure that health conditions had not changed in the meantime. To test interrater reliability between professionals and parents, a subset of 39 children were rated by pairs of professionals and one parent (handled separately as pairs, except for one child evaluated by one professional and one parent). Absolute agreement, and the extent to which agreement exceeded chance (κ), were calculated between paired independent observers. Agreement can be considered as moderate (κ=0.41–0.60), substantial (κ=0.61–0.80), and almost perfect (κ=0.81–1.00).[29, 30] Kendall's τ was calculated to examine the association between VFCS and levels in the GMFCS, MACS, and CFCS (one level of each classification was available for each child). No data on the EDACS levels were collected, as it is still not used regularly by professionals.

Univariate and multivariate analysis of variance were used to assess the relationship between the VFCS and the presence of intellectual disability, epilepsy, and ASD/behavioural problems.

Results

Phase 2

Phase 2 provided initial external validation of the VFCS. The nominal group process used feedback from group participants leading to a dynamic revision of the VFCS. The modifications receiving the most votes from that nominal group were incorporated into the next revision. This process continued until the last nominal group reached a consensus about the concepts and simply suggested wording changes.

An important result of this phase was the clear description of distinctions between levels with examples of vision-related activities, self-initiated strategies, and environmental adaptations to perform activities. Another

substantial result of this phase was the creation of a VFCS level identification chart in the form of a decision-tree algorithm (Appendix S2, online supporting information), to facilitate visually and analytically – by means of simple and alternative questions – the identification of the VFCS level for each child (Appendix S3, online supporting information).

Phase 3

More than 80% of participants agreed on 15 of the 18 questions representing the content of the VFCS in round 1, selecting a score of 5 or more on each Likert scale. On the basis of scores and comments/suggestions received in round 1, changes were made to the VFCS revising the definitions of terms used and providing a clearer layout of information presented. The three questions on which agreement was lower in round 1 (possible overlap/difficulty in distinction between two levels; clarity of wording for level II and for level III) received more than 80% agreement in round 2 after amending the wording, clarifying distinctions between levels and giving examples. Participants were concordant in highlighting the usefulness of the VFCS both in clinical and in research settings, particularly for identifying the level of visual abilities and limitations of children with CP, for harmonizing communication among professionals, and for planning interventions and supports or adaptations to promote activity and participation. The focus on the recognition of visual functioning, which could be otherwise overlooked in children with CP, and the possibility to integrate it with the other levels of functioning with the GMFCS, MACS, CFCS, and EDACS, was considered particularly relevant.

Building on the experience of a wide group of experts and the involvement of patients and families, this phase enabled the researchers to achieve construct validity and demonstrate meaningfulness of the concept of the five levels of the VFCS.

Phase 4

Table 3 shows the results of the reliability studies. Absolute interrater agreement among professionals was 86% (weighted κ=0.88, indicating almost perfect agreement; 95% confidence interval [CI] 0.83–0.93). In the few situations where disagreement was observed, it was by only one level. Test–retest reliability among professionals was almost perfect (weighted κ=0.97; 95% CI 0.95–0.99). Parent–professional interrater reliability was moderate (weighted κ=0.51; 95% CI 0.39–0.63). When professionals and parents did not agree, the score only differed by one level; when this was the case, in all but one case parents rated their child one level higher, namely more able, than the professional. Parent–professional reliability did not significantly change with the increasing age of the children. There was a moderate positive correlation between the VFCS and the levels in the GMFCS, MACS, and CFCS (Kendall's τ=0.42 and 0.46, p<0.01 with the GMFCS; 0.42 and 0.44, p<0.01 with the MACS; 0.43 and 0.44, p<0.01 with the CFCS for the two professionals respectively, on the basis of 136 children evaluated).

Table 3 Reliability measures associated with use of the VFCS

Professional 1 VFCS classification, *n*	**Professional 2 VFCS classification, *n***					
	I	**II**	**III**	**IV**	**V**	**Total**
I	**19**	3	0	0	0	22
II	7	**36**	4	0	0	47
III	0	2	**50**	5	0	57
IV	0	0	1	**29**	0	30
V	0	0	0	0	**4**	4
Total	26	41	55	34	4	160
Professional interrater reliability on 160 children, 29 professionals involved (weighted κ=0.88; 95% CI 0.83–0.93). Absolute agreement 86%						
Time 1 VFCS classification, *n*	**Time 2 VFCS classification, *n***					
	I	**II**	**III**	**IV**	**V**	**Total**
I	**43**	2	0	0	0	45
II	2	**79**	0	0	0	81
III	0	2	**83**	2	0	87
IV	0	0	1	**44**	0	45
V	0	0	0	0	**6**	6
Total	45	83	84	46	6	264
Test–retest reliability by professionals on 132 children, 29 professionals involved (weighted κ=0.97; 95% CI 0.95–0.99). Absolute agreement 97%						
Professional VFCS classification, *n*	**Parent VFCS classification, *n***					
	I	**II**	**III**	**IV**	**V**	**Total**
I	**20**	2	0	0	0	22
II	14	**10**	0	0	0	24
III	0	17	**8**	0	0	25
IV	0	1	3	**2**	0	6
V	0	0	0	0	**0**	0
Total	34	30	11	2	0	77
Parent–professional interrater reliability on 39 children, 29 professionals, and 39 parents involved (weighted κ=0.51; 95% CI 0.39–0.63). Absolute agreement 52%						

Values in bold type indicate agreement between observers. VFCS, Visual Function Classification System.

The one-way analysis of variance showed significant direct associations between the VFCS level and the presence of intellectual disability ($p<0.01$), epilepsy ($p<0.01$), and ASD/behavioural problems ($p=0.02$). The corresponding multivariate analysis showed a significant association between the VFCS level and the presence of intellectual disability ($p<0.01$), a borderline association with the presence of epilepsy ($p=0.05$), and no significant association with the presence of ASD/behavioural problems ($p=0.90$).

Discussion

The VFCS was developed empirically following a step-by-step process to classify visual abilities of children with CP in everyday life. Visual abilities refer to how the child uses vision purposefully to see, direct gaze, recognize, interact with the environment, and explore it.[1, 2, 24, 25] In fact, while measures of visual impairment, at the level of the eye or brain, are valuable and necessary in the diagnosis and management of children with CP, they do not provide information on how visual dysfunction affects daily life, and do not take into account children's abilities. Although some children with CP may show limited levels of functioning and restricted participation due to their visual impairment, others can rely on their visual abilities to overcome their limitations due to motor impairment.[24, 25]

The VFCS was developed focusing on the ICF activity and participation concepts, regardless of the body structure and functions underlying the visual disorder.[23] Key factors included how well and how often visual function is used to perform activities, the environments where it is used, the need for support, or the predominant use of other sensory modalities to perform vision-related activities.

Similar to other classification systems already available to classify gross motor, manual, communicative, and eating/drinking abilities in children with CP (Table 4), we started from the analysis of the literature and from clinical experience, progressively moving to collect a wide range of expert knowledge and opinions worldwide, involving different stakeholders at each step. Specifically, the involvement of patients with CP, their primary caregivers, and practitioners was crucial to ensure that all domains of visual ability that are meaningful to children with CP would be included.[24] This was an evolving process aimed at demonstrating construct validity as well as concept meaningfulness of the five levels of the classification.

Table 4
General summary headings for the GMFCS, MACS, CFCS, EDACS, and VFCS

Level	GMFCS	MACS	CFCS	EDACS	VFCS

I	Walks without limitations	Handles objects easily and successfully	Sends and receives with familiar and unfamiliar partners effectively and efficiently	Eats and drinks safely and efficiently	Uses visual function easily and successfully in vision-related activities
II	Walks with limitations	Handles most objects but with somewhat reduced quality and/or speed of achievement	Sends and receives with familiar and unfamiliar partners but may need extra time	Eats and drinks safely but with some limitations to efficiency	Uses visual function successfully but needs self-initiated compensatory strategies
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and/or modify activities	Sends and receives with familiar partners effectively, but not with unfamiliar partners	Eats and drinks with some limitations to safety; there may be limitations to efficiency	Uses visual function but needs some adaptations
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations	Inconsistently sends and/or receives even with familiar partners	Eats and drinks with significant limitations to safety	Uses visual function in very adapted environments but performs just part of vision-related activities
V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions	Seldom effectively sends and receives, even with familiar partners	Unable to eat and drink safely – tube feeding may be considered to provide nutrition	Does not use visual function even in very adapted environments

GMFCS, Gross Motor Function Classification System; MACS, Manual Abilities Classification System; CFCS, Communication Function Classification System; EDACS, Eating and Drinking Ability Classification System; VFCS, Visual Function Classification System.

The VFCS can be used by parents and caregivers, and by all professionals working with children with CP (health care, school, and rehabilitation professionals). It is of interest that analysis of the interrater and intrarater reliability of the VFCS showed almost perfect agreement between and within professionals, while reliability between professionals and parents was moderate. Of note, parents tended to score their child as having better visual abilities than

professionals, consistent with what has been found in the reliability studies of other classification systems for CP, but the difference was usually not more than one level. During meetings with parents or within the expert group, it clearly emerged how parents tended to score the VFCS level considering their child engaged in less demanding activities, while professionals tended to base their scores on more challenging situations, often requiring support or adaptations, such as during school or outdoor activities. This discrepancy between parents' and professionals' judgement of a child's ability is a common finding in clinical practice, and becomes more apparent with age and increasing social and environmental challenges. Examples have been added in the chart 'Distinctions between levels' (Appendix S3). It has been agreed that more clarifications should be provided on what is expected from a child according to the level of abilities when the VFCS is presented to users during workshops, scientific meetings, and training sessions.

In the present validation work, the reliability of the VFCS was shown to be easily used from the age of 1 year, and was not found to correlate with age. However, only a small proportion of children were younger than 3 years (median age 5y 7mo). Further studies will assess the feasibility of the VFCS in the youngest population, as well as the stability of the levels over time starting from the first years of life.

Similar to the GMFCS, MACS, CFCS, and EDACS, the VFCS is not intended as an assessment tool and does not explain the underlying factors contributing to the level of visual functioning shown by the child. Indeed, within the conceptual framework of the ICF, the level of disability can be influenced by a host of factors, including comorbidities such as intellectual disability, behavioural and social-interaction problems, as well as environmental factors such as barriers to receiving care and lack of resources. Interestingly, intellectual disability, epilepsy, and ASD/behavioural problems were all associated with the VFCS level; however, the presence of intellectual disability was the strongest predictor of the children's visual abilities. This is not surprising, given the influence of cognition on visual abilities (and vice versa), which is obvious in the first years of life and becomes more evident with age. However, it is important to highlight once more that the goal of the VFCS (and the other classification systems for children with CP) is to classify what the children actually do, irrespective of the underlying reasons.

Furthermore, it is of interest that the correlation between the VFCS and the other classification systems was only moderate, further highlighting that children with more severe limitations in motor or communicative functions can have better levels of visual ability and vice versa. This suggests that a correct characterization of the various functional domains is essential for the correct development of rehabilitation programmes tailored to the child's many abilities and needs.

One of the main challenges of a classification system is to discriminate meaningfully among different degrees of functional abilities.[18, 23] Indeed, appropriate interventions to minimize limitations due to vision impairment and to maximize abilities and participation in vision-related daily activities cannot be provided to children with CP and their families in the absence of valid and reliable tools.[24] We are aware that the effort to condense all key visual functions into five levels may be limiting and challenging; however, our data support the use of the VFCS as a reliable and easily applicable tool, both in clinical and in research settings, and further validity data will follow.

One possible limitation of the VFCS is that raters might find it hard to identify the usual performance in daily activities, as opposed to the best or the worst one. Indeed, the rater should know the child well and classify them according to the VFCS level that best describes the visual abilities in daily life. Specific training in the use of the VFCS, validation in different languages and, possibly, in other target populations of children with developmental disabilities are being planned and will provide further evidence of the feasibility of the clinical implementation of the VFCS. Another possible limitation of the present study is that the vast majority of the patients in the sample had spastic forms of CP, as assessed in tertiary referral centres. Further studies will be needed to confirm the reliability of the VFCS in different subtypes of CP and in primary health care settings. Our intention is that this first report on the VFCS will be followed by a series of studies designed to confirm its validation in community services and with primary health care providers, as well as in wider cohorts of patients with non-spastic CP.

As part of the development and validation process of the VFCS, the opinion on the usefulness of the VFCS was widely shared by the participants in the study. Similar to the other classification systems for children with CP, a wide agreement was found about the VFCS being an important contribution to the diagnosis and assessment of CP, and an important aid to stratify people participating in natural history as well as intervention studies. The implementation of a classification system for visual functioning could be very helpful in highlighting the importance to detect and take into account the existence of visual difficulties in a child with CP, as these could be (and actually often are) otherwise under-recognized by caregivers and professionals dealing with the child. Additionally, it can enable policymakers and health systems to plan more accessible services and assisted living facilities for the population affected by CP.

In conclusion, this new 5-level classification system of visual functioning for children with CP may on the one hand address the need to harmonize communication among professionals and between families and professionals; and on the other, it may allow clinicians and researchers to better define treatment and prognosis according to the specific level of (dis)ability of that child.

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Supplementary Material

Appendix S1: Delphi questions

Appendix S2: VFCS level identification chart

Appendix S3: VFCS levels

Appendix S4: List of participants who contributed to the development of the VFCS

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