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An international clinical perspective on functioning and disability in adults with cerebral palsy

ICF Core Set for Adults with CP Study Group; Noten, Suzie; Rodby-Bousquet, Elisabet; Limsakul, Chonnanid; Tipchatyotin, Suttipong; Visser, Fay; Grootoonk, Anneke; Thorpe, Deborah E; van der Slot, Wilma Ma; Selb, Melissa

Published in: **Disability and Health Journal**

DOI: 10.1016/j.dhjo.2022.101318

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version Publisher's PDF, also known as Version of record

Publication date: 2022

Link to publication in University of Groningen/UMCG research database

Citation for published version (APA):

ICF Core Set for Adults with CP Study Group, Noten, S., Rodby-Bousquet, E., Limsakul, C., Tipchatyotin, S., Visser, F., Grootoonk, A., Thorpe, D. E., van der Slot, W. M., Selb, M., & Roebroeck, M. E. (2022). An international clinical perspective on functioning and disability in adults with cerebral palsy. *Disability and Health Journal, 15*(3), [101318]. https://doi.org/10.1016/j.dhjo.2022.101318

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Disability and Health Journal 15 (2022) 101318



Contents lists available at ScienceDirect

Disability and Health Journal

journal homepage: www.disabilityandhealthjnl.com

Original Article

An international clinical perspective on functioning and disability in adults with cerebral palsy



Disability and Health Journal

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ARTICLE INFO

Article history: Received 31 October 2021 Received in revised form 8 March 2022 Accepted 9 March 2022

Keywords: Cerebral palsy Adults Functioning Clinical study ICF

ABSTRACT

Background: This international, multi-center cross-sectional study is one of the preparatory studies in the development of the International Classification of Functioning, Disability and Health (ICF) Core Sets for adults with cerebral palsy (CP) to describe their functioning and health.

Objective: To identify the most common problems in functioning of adults with CP presenting in healthcare services, and facilitating and hindering environmental factors, using the ICF as a reference. *Methods:* Participants were adults with CP who visited healthcare services in the Netherlands, Sweden, Thailand, and the United States. Structured interviews were performed using an adapted version of the generic ICF checklist 2.1a (106 categories) to rate the participant's functioning and the impact of environmental factors. Descriptive statistics were used for frequency analysis.

Results: In total, 101 participants were included, of whom 69 without intellectual disability (mean age \pm SD of 38.4 \pm 14.7 y; 85.5% with spastic type of CP; Gross Motor Function Classification System (GMFCS) levels I–V) and 32 with intellectual disability (mean age \pm SD of 25.0 \pm 6.4 y; 71.9% with spastic type of CP; GMFCS levels I–V). A total of 104 ICF categories in the ICF checklist were frequently present in adults with CP: 27 body functions, 4 body structures, 53 activities and participation, and 20 environmental factors.

Conclusions: The most common problems of adults with CP presenting in healthcare services are diverse and highly prevalent. The study results add the clinical perspective on relevant categories of functioning to the basis for developing the ICF Core Sets for adults with CP.

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Cerebral palsy (CP) describes a group of permanent developmental disorders affecting movement and posture and leading to activity limitations.¹ Worldwide, CP occurs in 2–2.5 per 1000 live births.^{2,3} Presently, the majority of persons with CP are adults.⁴ Although the number of studies on adults with CP has been increasing over the past years,⁵ research still has limited focus on

https://doi.org/10.1016/j.dhjo.2022.101318

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adults as compared with children and youth. In previous studies, adults with CP reported problems in several areas of functioning: on average 65%-70% have pain,⁶ 42% are unable to walk, and only 39% are employed.⁷

The functioning of adults with CP depends on the fetal or early brain damage and is also influenced by personal characteristics and available resources or facilitative environmental factors.⁸ The wide range of problems in functioning experienced by adults with CP relates to different clinical needs.⁹ Although many outcome measures are available for adults with CP,⁵ systematic assessment of functioning and disability and comparison of an individual to CPspecific references is hampered by a lack of standardization.⁷ Therefore, efforts are needed to standardize assessments in clinical and research settings in order to optimize interventions for adults with CP worldwide. The development of ICF Core Sets (International Classification of Functioning, Disability and Health) for persons with specific health conditions can facilitate these efforts.^{10,11}

The ICF reflects a comprehensive biopsychosocial model of functioning and disability and encompasses the components body functions (b), body structures (s), activities and participation (d), and contextual factors, including environmental, (e) and personal factors (pf). The ICF provides a framework to describe these aspects of functioning within these components, starting with chapter or first-level categories (e.g. b2 sensory functions and pain). Each chapter consists of second-level categories (e.g. b280 sensation of pain), which can be subdivided in more specific third- (e.g. b2801 pain in body part) or fourth-level categories (e.g. b28016 pain in joints). The implementation of the ICF in research and clinical practice is challenging since it contains over 1400 categories. To facilitate the practical application of the ICF, the ICF Research Branch and the World Health Organization (WHO) created an evidence-based methodology to develop ICF Core Sets for specific health conditions. ICF Core Sets are standardized, internationally accepted tools and include shortlists of ICF categories that are most relevant for describing functioning of persons with a health condition.^{12,13} ICF Core Sets support the standardization of the assessment and reporting in clinical practice and in research. Currently, over 30 ICF Core Sets are available, including the one for children and youth with CP.¹⁴ Since functioning and health outcomes of persons with CP deteriorate with age¹⁵ and participation areas relevant for adults may be different than for children, for example, housing and employment,¹⁶ there is a need for ICF Core Sets for adults with CP.

This study is one of the preparatory studies to develop ICF Core Sets for adults with CP that reflect the diverse perspectives of professionals and adults with CP and their family. The present study adds the clinical perspective on functioning of adults with CP to the other perspectives applied in a literature review (researcher's perspective),⁵ a qualitative study (patient's perspective),¹⁷ and an expert survey (health professional's perspective).¹⁸

This clinical study aims to identify the most common problems as well as facilitative and hindering environmental factors in adults with CP who are visiting a clinical setting in different parts of the world. Along with the other preparatory studies the present results generated an aggregated list of "candidate" second-level ICF categories to serve as the basis for a structured consensus process to decide on the ICF Core Sets for adults with CP.

Methods

Study design

An international multi-center cross-sectional study was performed in adults with CP with and without ID in the Netherlands, Sweden, Thailand, and the United States. For adults with CP with ID, caregivers participated as proxies in the study, and where possible, the respective person with CP was present and answered for him or herself. We performed face to face interviews; telephone interviews or video calls were used for those participants included during the COVID-19 pandemic, to resolve logistical issues or if preferred by the participant; no technical issues were reported. This study was approved by the medical ethics committee of Erasmus MC University Medical Center Rotterdam (MEC-2018-1126) and the participating centers. Participants or their caregivers signed informed consent prior to the interview.

Participants

Participants were recruited from four countries, representing three WHO regions. Participating healthcare centers were Amsterdam University Medical Center (UMC), Libra Rehabilitation & Audiology, Eindhoven, De Hoogstraat Rehabilitation, Utrecht, UMC Groningen in the Netherlands, in Sweden, Prince of Songkla University, Songkhla in Thailand, and The University of North Carolina at Chapel Hill, in the United States. Additionally, information was sent out to coordinators for Cerebral Palsy Follow-Up Programmes (CPUP) in all 21 healthcare regions in Sweden, and the study was advertised through the website of their CPUP, the family forum and social media for the CP association.

A consecutive sample was included in the study of individuals diagnosed with CP, at least 18 years of age, able to communicate (or their caregiver) in the country's native language, who had visited the healthcare center in the last 3 years. Adults with CP were considered having an ID if a health professional diagnosed the person as having an IQ level of less than 70 or a developmental age below 12 years old.¹⁹ Participants were enrolled in the study between July 2019 and July 2020.

Tailored version of the ICF checklist 2.1a

Health professionals and/or researchers performed structured interviews by using a tailored version of the ICF checklist 2.1a.¹¹ The first part of this checklist consisted of CP-related and demographic information on age, sex, type of CP,²⁰ Gross Motor Function Classification Scale (GMFCS) level,²¹ marital status, if the person has children, living status, educational level, and working status. The person's physical, mental and emotional health in the previous month was scored on a five-point scale ranging from very good to very bad. The next part contained second-level ICF categories of functioning that were scored on a five-point scale indicating the extent of a problem. The generic ICF checklist 2.1a contains 123 slevel ICF categories that are considered important by the WHO¹⁰; this was adapted for the present study to ensure its specificity to functioning of adults with CP. First, the generic ICF checklist was extended with those ICF categories addressed in more than five percent of the studies based on the information from the systematic review on outcomes in adults with CP,⁵ one of the previous preparatory studies for the ICF Core Sets. Second, five experts on CP in adulthood (MK, ER-B, SH, ST, MR) were consulted to include or exclude items from the extended list to ensure that the most essential categories for adults with CP were included in the CPtailored checklist, and less relevant categories were omitted. They weighed the relevancy of categories based on their expertise and on the relative frequency in which a category was studied according to the systematic review,⁵ and aimed to reduce the number of items to a feasible checklist for the interviews. The resulting tailored version of the ICF Checklist 2.1a consisted of 106 ICF categories (28 body functions, 4 body structures, 54 activities and participation and 20 environmental factors, see Supplementary data). The checklist used

the official translations of the ICF categories that are available in Dutch, Swedish and Thai. Interviewers completed the ICF eLearning tool (http://www.icf-elearning.com/) and studied the ICF browser (http://apps.who.int/classifications/icfbrowser/) to get familiar with the structure and application of the ICF. A user guide was created to assure quality of scoring across the centers.

Data collection

In the interviews the extent of problems was rated using the ICF qualifiers: 0) No impairment or difficulty, 1) mild impairment or difficulty, 2) moderate impairment or difficulty, 3) severe impairment or difficulty, and 4) complete impairment or difficulty.²² Environmental factors were rated as barriers (from mild to complete; 1 to 4), facilitators (from +1 to +4), or no barriers or facilitators (0). Additional scoring options were qualifier 8 "not specified," which was used in case there was insufficient information to specify the severity of the impairment and 9 "not applicable," in case it was inappropriate to apply a particular code. A visual scale was used during the interviews to clarify the qualifiers. Additionally, items that were not included in the checklist, but were considered important during an interview, for example, societal attitudes, were included and rated. In the last part, personal factors that impact a person's functioning were documented. No communication challenges were reported during the interviews. All participants finished the interview process in one session; participants were given a break when needed. In case of doubt about the scoring, interviewers contacted the first author (SN).

Data analysis

Descriptive statistics were used to characterize the study sample and to analyze the frequency of problems (score ≥ 1 for problems and barriers and $\geq +1$ for facilitators); scores of 8 and 9 were excluded from the frequency analyses. Also frequencies of personal factors were counted. Frequencies of problems in adults with and without ID were compared. ICF categories that were reported in at least 20% of the adults with CP without ID or of those with ID were identified as common problem, facilitator, or barrier.¹³ Those were considered relevant and included in the list of candidate categories provided for the subsequent consensus process, as recommended by the guideline.¹³

Results

Participants

In total, 101 participants were included, 69 without ID (mean age \pm SD of 38.4 \pm 14.7 y; 38 female; 59 with spastic type of CP; GMFCS levels I–V) and 32 with ID (mean age \pm SD of 25.0 \pm 6.4 y; 15 female; 23 with spastic type of CP; GMFCS levels I–V). Table 1 shows the characteristics of participants.

Relevant areas of functioning

Physical health and mental and emotional health were scored moderate or good by more than 90% of the adults with CP. In total, 104 of 106 ICF categories of the tailored version of the ICF checklist 2.1a were identified in more than 20% of adults with or without ID: 27 body functions, 4 body structures, 53 activities and participation, and 20 environmental factors. The 20% threshold was not met for ICF categories heart functions (b410) and listening (d115). Frequencies of all second-level categories in adults with CP with or without ID are presented in the Supplementary data. When comparing results between countries, two categories would not

Table 1

Demographic and cerebral palsy-related information of adults with cerebral palsy with or without intellectual disability.

| Participant characteristics | Adults without intellectual | Adults with intellectual |
|---------------------------------------|-----------------------------|--------------------------|
| | disability $(n = 69)$ | disability $(n = 32)$ |
| Age in years, mean \pm SD, range | 38.4 ± 14.7, 18-79 | 25.0 ± 6.4, 19-53 |
| Sex, n (%) | | |
| Male | 31 (44.9) | 17 (53.1) |
| Female | 38 (55.1) | 15 (46.9) |
| Type of CP, ³⁰ n (%) | | |
| Spastic | 59 (85.5) | 23 (71.9) |
| Dyskinetic | 6 (8.7) | 0 (0.0) |
| Ataxic | 1 (1.5) | 2 (6.2) |
| Mixed | 3 (4.3) | 7 (21.9) |
| CP distribution, n (%) | | |
| Unilateral | 12 (17.4) | 5 (15.6) |
| Bilateral | 57 (82.6) | 27 (84.4) |
| GMFCS level, ²¹ n (%) | | |
| Ĩ | 9 (13.0) | 1 (3.1) |
| II | 20 (29.0) | 10 (31.2) |
| III | 26 (37.7) | 3 (9.4) |
| IV | 12 (17.4) | 6 (18.8) |
| V | 2 (2.9) | 12 (37.5) |
| Marital status, n (%) | (= (= = =) | |
| Single | 47 (68.2) | 32 (100.0) |
| In a relationship | 4 (5.8) | 0 (0.0) |
| Married/living with partner | 15 (21.7) | 0 (0.0) |
| Divorced/separated | 3 (4.3) | 0 (0.0) |
| Having children, n (%) | 14 (20.3) | 0(0.0) |
| Living status, n (%) | 20 (40 C) | 0 (0 0) |
| Living independently | 28 (40.6) | 0(0.0) |
| Living with partner | 15 (21.7) | 0(0.0) |
| Living with parents | 19(27.5) | 21 (65.6) |
| Living with others | 2 (2.9) | 3 (9.4) |
| Living in residential care | 1 (1.5) | 6(18.8) |
| Spacial education in (%) | 4(3.6) | 2(0.2) |
| Special education, II (%) | 21 (50.4) | 19 (59.4) |
| No advisation | 1 (1 4) | 8 (25.0) |
| Flomontary school | 1(1.4) | 8 (25.0) 18 (56.2) |
| Lower secondary education | Z (2.9) 6 (8.7) | 10(30.2) |
| Higher secondary education/high | 16 (23.2) | 3 (9.4) |
| school | 10 (25.2) | 5 (5.4) |
| Advanced vocational education | 12 (17.4) | 0 (0.0) |
| Higher education | 32 (46.4) | 0 (0.0) |
| Working status, n (%) | | |
| Full-time employment/self employed | 24 (34.8) | 0 (0.0) |
| Part-time employment | 10 (14.5) | 0 (0.0) |
| Non-paid employment | 2 (2.9) | 0 (0.0) |
| Supported employment | 0 (0.0) | 7 (21.9) |
| Unemployed | 13 (18.8) | 20 (62.5) |
| Student | 11 (15.9) | 1 (3.1) |
| Receiving benefit grants | 8 (11.6) | 4 (12.5) |
| Retired | 1 (1.5) | 0 (0.0) |
| Country | 00 (01 0) | e (ee t) |
| The Netherlands | 22 (31.9) | 9 (28.1) |
| Sweden | 28 (40.6) | 2 (6.3) |
| Inailand | 9(13.0) | 21 (65.6) |
| USA | 10(14.5) | U (0.0) |

have been covered if only European countries were included, that is, family relationships (d760) and religion and spirituality (d930). Items that were not included in the checklist, but were considered important during an interview, were limited.

Body functions

The most commonly identified chapters in body functions were mental functions (b1) and neuromusculoskeletal and movementrelated functions (b7). The top 3 second-level categories in adults without ID were muscle tone functions (b735, 97.1%), gait pattern functions (b770, 90.5%) and vestibular functions (b235, 89.9%), and in adults with ID, gait pattern functions (b770, 100.0%), vestibular functions (b235, 96.9%) and intellectual functions (b117, 96.9%). The proportion of adults with CP with and without ID with problems in the ICF categories of body functions is presented in Fig. 1.

Body structures

In the body structures component, most commonly reported problems were structure of lower extremity (s750, no ID 84.1%; ID 62.5%) and structure of trunk (s760, no ID 59.4%; ID 54.8%). Adults without ID frequently mentioned the structure of upper extremity (s730, 66.7%) and adults with ID additional musculoskeletal structures related to movement, including bones, joints and muscles (s770, 60.0%).

Activities and participation

In activities and participation, the most frequently reported chapter was mobility (d4). In adults without ID, the most common ICF categories were walking (d450, 89.7%), moving around (d455, 89.1%), lifting and carrying objects (d430, 78.3%), and maintaining a body position (d415, 75.4%). In adults with ID, difficulties in activities and participation were highly prevalent; 30 of the 54 activities and participation were reported in at least 75% of them (Fig. 2). Common difficulties experienced by adults with ID were higher

education (d830, 100.0%), walking (d450, 96.9%), doing housework (d640, 96.9%) and solving problems (d175, 93.8%).

Environmental factors: Facilitators and barriers

The majority of the environmental factors were mentioned as facilitators (20 ICF categories); 7 barriers were mentioned. The chapters products and technology (e1), and support and relationships (e3) were commonly mentioned as facilitators. The chapter services, systems and policies (e5) was reported as a facilitator as well as a barrier.

The most often indicated facilitators in adults with and without ID were products and technology for personal indoor and outdoor mobility and transportation (e120, no ID 93.7%; ID 83.9%) and immediate family (e310, no ID 89.7%; ID 93.8%). Highly prevalent facilitators were products and technology for personal use in daily living (e115, 87.5%) and friends (e320, 86.4%) in adults without ID, and health professionals (e355, 87.5%) and personal care providers and personal assistants (e340, 84.0%) in adults with ID.

The most common barriers in adults with and without ID were design, construction and building products and technology of buildings for public use (e150, no ID 56.5%; ID 51.6%) and transportation services, systems and policies (e540, no ID 50.0%; ID 45.2%). Additional prevalent barriers were general social support services, systems and policies (e575, 44.2%) in adults without ID and housing services, systems and policies (e525, 42.1%) in adults with ID.



Fig. 1. Proportion of adults with cerebral palsy with and without intellectual disability (%) with problems in the ICF categories of body functions.



Fig. 2. Proportion of adults with cerebral palsy with and without intellectual disability (%) with difficulties in the ICF categories of activities and participation.

Personal factors

In total, 152 and 99 personal factors influencing daily life functioning were mentioned by adults without ID and with ID, respectively. A broad range of personal factors was indicated, with frequently mentioned factors such as perseverance, patience, positive, and educational status in adults without ID, and positive, educational status and socioeconomic status in adults with ID.

Fig. 3 shows the ICF framework with the second-level categories most frequently mentioned in at least 75% of the participants (with or without ID) as well as the most commonly mentioned personal factors.



Fig. 3. International Classification of Functioning, Disability and Health (ICF) framework including the most frequent second-level categories in \geq 75% of the adults with cerebral palsy with or without intellectual disability (ID) and commonly mentioned personal factors. Underlined categories: commonly mentioned in adults with and without ID; categories in normal font: commonly mentioned in adults without ID; categories in Italics: commonly mentioned in adults with ID. Environmental factors above 75% were all facilitators, barriers did not meet the 75% level.

Discussion

This international, multi-center cross-sectional study gave us insight into the most common problems in functioning among adults with CP presenting in healthcare facilities. The clinical sample showed a similar distribution for the type of CP and proportion of GMFCS levels I–III to those reported in clinical follow-up studies in adults with CP, except for an underrepresentation of adults with CP functioning at GMFCS level I.^{15,23} The broad range of problems in functioning and contextual factors in this study are in line with the impairments and limitations which are common in CP irrespective of age group, addressing both physical and mental issues.^{7,15} The present results add to the evidence that the prevalence of these problems is very high in adults with CP who visit

healthcare services, with many impairments and difficulties in activities and participation being present in more than 3 out of 4 persons without ID, and even more in those with ID.

The most common impairments mainly address neuromusculoskeletal and movement-related functions, such as gait pattern and muscle tone but also balance, which are known issues in CP, especially in adulthood.^{24,25} In participants with ID, speech functions and also several mental functions, such as intellectual functions, higher-level cognitive functions, energy and drive, emotional functions and memory functions, were highly prevalent. Notably, also adults without ID often reported problems in several mental functions, for example, energy and drive, emotional functions, attention and sleep (over 40%). For body structures, problems referred to structures related to movement, mainly in the upper and lower extremity and trunk, which are known problems in adults with $\mbox{CP.}^{26}$

Regarding activities and participation, frequently occurring problems address mobility and domestic life activities. Also, problems in other domains were frequently reported by adults without ID, such as handling stress (domain of general tasks and demands), caring for body parts (self-care domain) and acquiring or keeping a job (employment domain). The prevalence of difficulties was often higher in adults with ID compared with adults without ID, emphasizing the amount of difficulties they experience. The present results underline the clinical experience of professionals that many adults with CP have a broad spectrum of impairments and disabilities, reflecting the long-term consequences and disability burden of this condition at adult age in many areas.^{15,18} Strikingly, although a high frequency of functioning problems is reported by adults with CP, routine follow-up and rehabilitation care for this population is limited.^{9,27,28}

Environmental factors were mentioned more often as facilitators than as barriers, suggesting that the physical, social and attitudinal environment is experienced by adults with CP as supportive. Support from products and technology for mobility and transportation, such as wheelchairs, and support from immediate family were seen as important. When considering ID, support from health professionals and personal assistants was relevant for adults with ID while support from friends and products and technology for personal use were reported more frequently by adults without ID. These results may be the reflection of the specific needs of the sub-population of adults with CP. For example, the need for support from health professionals and personal assistants may be more prominent in adults with CP with ID due to their lower level of independence as compared to those without ID. Although more facilitators were mentioned than barriers, adults with CP did report some barriers, predominately accessibility of public buildings, transportation services and housing services. Participants highlighted problems with many stairs or doorsteps, the lack of fully accessible vehicles for transportation and finding an appropriate place to live. Additionally, adults without ID indicated problems with general social support services, because of requirements that must be met and changing regulations regarding eligibility for assistance.

The findings on environmental factors emphasize the importance of including environmental support in interventions and follow-up measures, and suggest that there are nevertheless barriers that need remedying.

The findings of the current study are in line with the results from the other three preparatory studies for developing the ICF Core Sets for adults with CP, and the studies together ensure that the most common problems in adults with CP visiting healthcare services are taken into account.^{5,17,18} All four studies identified problems in a broad range of functioning aspects, underscoring the heterogeneity of the CP population, as well as the impact of CP in many areas. Aspects of functioning that were most frequently identified as relevant for adults with CP were pain, muscle tone, intellectual functions, structure of lower extremity, walking, self-care and employment, support from products for mobility and transportation and support from the immediate family. Specifically, the health professionals in the expert survey were among the most aware of the importance of person-environment interactions in adults with CP. The present clinical study confirmed both the facilitative or hampering effects of environmental factors on functioning in adults with CP.

Together with the other preparatory studies, the present clinical perspective generated an aggregated list of "candidate" second-level ICF categories to serve as the basis for the consensus process during which relevant stakeholders decided on the ICF Core Sets for adults with CP.²⁹ These ICF Core Sets encompass the most

relevant aspects of functioning (ICF categories) and will serve as a guideline for the systematic assessment, documentation and reporting on adults with CP in clinical practice and in research. The Core Sets will indicate the most important areas of functioning and assist users by providing a checklist of "what" to measure. Their implementation is expected to significantly contribute to improving the clinical follow-up and healthcare services for adults with CP. The assessments of various areas of functioning can be distributed among members of a multidisciplinary team, each taking on its own area of expertise.

Some limitations should be taken into account when interpreting the results. Since rather small samples of four countries across three WHO regions were included, we must be cautious to generalize the results. Also, the age of this sample was rather young, especially of the adults with ID. Despite additional efforts, we were not able to include older participants. They seem to adhere less often to clinical care, which is not unexpected as the referral from pediatric to adult care has only in recent years increased.²⁷ From followup studies in adults with CP, we know that different problems might arise with age.¹⁵ Furthermore, the established methodology for conducting this preparatory study calls for considering available medical information. However, there was no access to medical records due to privacy reasons; thus, the information was based solely on the interview. Although the clinical information derived from medical records might have enhanced the accuracy of the qualifier rating, the participants (or their caregivers) were well aware of their impairments and disabilities, thus minimizing possible inaccuracies related to self-reporting on functioning. It is also worth noting that some categories of the generic ICF checklist 2.1a were not included in the tailored checklist used in the present study, such as blood pressure and sexual functions, which might have been important for understanding functioning in CP. Nevertheless, those were obtained by other preparatory studies¹⁷ and included in the aggregated list of candidate categories provided for the consensus process to develop the final comprehensive ICF Core Set.²⁹

Conclusions

The study results confirm the heterogeneity of adults with CP, the high frequency of functioning problems they experience and the importance of environmental factors that facilitate their functioning. The study was performed among adults with CP visiting healthcare services, completing the series of preparatory studies that provide the scientific basis for developing the ICF Core Sets for adults with CP in a next step. Such Core Sets can then be implemented in clinical care and research, and will promote standard-ized data collection worldwide.

Presentation

Results of this study were presented at the AACPDM Annual Meeting 2020:

Noten S., Rodby Bousquet E., Limsakul C., Visser F., Tipchatyotin S, de Groot V., Grootoonk A, Konijnenbelt M, Meuzelaar- Kiezebrink F, van der Slot W.M.A., van den Berg-Emons H.J.G., Selb M., Roebroeck M.E. An international clinical study on impairments and disability in adults with cerebral palsy as a base for developing an ICF Core Set, 2020, oral presentation.

Funding

This work was supported by Rijndam Rehabilitation, Rotterdam, the Netherlands. The work of Elisabet Rodby-Bousquet was funded by the Swedish Research Council for Health, Working Life and Welfare (FORTE, grant no. 2018-01468).

Conflicts of interest

None.

Appendix

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.dhjo.2022.101318.

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