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# Assessment tools for evaluating body structure-function and activity in dyskinetic cerebral palsy: a systematic review of instrumented assessments according to ICF-CY

Eda Burç, Cemil Özal, Mintaze Kerem Günel

Department of Physical Therapy and Rehabilitation, Faculty of Physical Therapy and Rehabilitation, Hacettepe University, Ankara, Turkiye

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## ABSTRACT

Dyskinetic cerebral palsy (CP) is one of the most severe forms of CP, characterized by dystonia or choreoathetosis and can be classified into dystonic and choreoathetosis subgroups. The International Classification of Functioning, Disability, and Health-Child and Youth Version (ICF-CY) provides a framework for physical therapists to understand the health, functioning, activity, participation, and impact of dystonia and choreoathetosis. This review aimed to examine the clinical use of ICF-CY tools to assess body structure, function, and activity in children with dyskinetic CP. A systematic search was conducted in June 2024 using PubMed, Embase, Scopus, and Google Scholar databases. The search included terms related to cerebral palsy, dyskinesia, choreoathetosis, dystonia, body structure, function, and activity. After removing duplicates, 11,800 articles remained and 34 met the inclusion criteria. The review found that ICF-CY activity assessments focused primarily on fine-motor, communication, eating-drinking, bimanual fine motor, and speech functions following gross motor function. Some studies have evaluated ICF-CY body structure and function. Most studies used the Dyskinesia Impairment Scale. This review presents evaluations using instrumented assessments as objective outcome measures in patients with dyskinetic CP. Future studies should develop measurements that are applicable outside the laboratory by using new technologies.

**Keywords:** Dyskinetic cerebral palsy, assessment tools, ICF-CY, body structure and function, activity

## INTRODUCTION

Dyskinetic cerebral palsy (CP) is one of the most severe forms of CP.[1] It is a motor disorder characterized by changes in muscle tone and posture, with a variable element of involuntary movement.[2,3] Dyskinetic CP is based on the predominance of dystonia or choreoathetosis; thus, it can be further classified into the dystonic and choreoathetosis subgroups. Dystonia and choreoathetosis often coexist in dyskinetic CP, and the term dyskinetic CP is used when the predominance of dystonia and choreoathetosis is difficult to define.[2-6]

In dystonic CP, involuntary movements and sustained/intermittent muscle contractions occur, causing abnormal twisted posture and repetitive movements in abnormal posture.[5] Dyskinetic CP is the most common definition of dystonia in children, which occurs as a result of hypoxic-ischemic damage to the basal ganglia, thalamus, brain stem, and cerebellum during the prenatal, perinatal, or infancy periods.[5]

In choreoathetoid CP, hyperkinesia and hypotonia co-occur and fluctuations in muscle tone are dominant.[5,7] Choreoathetoid movements are defined as rapid, involuntary, jumpy, and small-amplitude movements that usually involve the distal extremities. Athetosis is an involuntary, discrete, slow, ever-changing, complex, writhing, irregular movement. It is prominent on the distal extremities and face.[5,7] Choreoathetosis appears to be associated with pure thalamic and basal ganglia lesions.[4]

Currently, several scales are used to define the severity of dystonia in dyskinetic CP, such as the Barry-Albright Dystonia Scale (BADS), Dyskinesia Impairment Scale (DIS), Burke-Fahn-Marsden Dystonia Rating Scale Movement (BFM-M), hypertonia assessment tool (HAT), and Unified Dystonia Rating Scale (UDRS).[7-13] The DIS also assesses choreoathetosis and dystonia.[7] Clinical scales are often used in conjunction with questionnaires such as The pediatric evaluation of disability inventory (PEDI), Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD), and quality of upper extremity skills test (QUEST), which determine performance-based outcome measures.[14-16] In addition, individualized outcome measures such as the Goal Attainment Scaling (GAS) and the Canadian Occupational Performance Measure (COPM) are used to evaluate treatment outcomes in patients with dyskinetic CP.[17-23]

**Corresponding Author:** Eda Burç, eda_burc_1994@hotmail.com

Functional classification systems play an important role in the management of children and youth with CP and in distinguishing the characteristics of functional abilities, and can assist in setting goals and planning management. The most commonly used classification systems for CP are the gross motor function classification system (GMFCS), manual ability classification system (MACS), communication function classification system (CFCS), eating and drinking ability classification system (EDACS), Viking Speech Scale (VSS), and visual function classification system (VFCS).[24-31] Although dyskinetic CP is the second largest group of CP in the entire CP population, only a few studies have investigated the functional classification of dyskinetic CP. In addition, it is important to establish a comprehensive functional profile to develop targeted interventions for dyskinetic CP.

With increasing attention paid to limitations in function, activity, and participation in neurodevelopmental disability, function-based classification schemes have recently emerged.[32] The gross motor function classification system (GMFCS) addresses gross motor functional capacity and classifies children along a specific functional trait, particularly as it relates to the core functions of individual ambulation.[24] It is psychometrically appropriate in terms of reliability and validity, enables the identification of a child's skills and needs, and allows for clear and concise communication between healthcare professionals.[33] Unlike the traditional subtype classification scheme, it provides information about the correct prognosis and the need for eventual ambulation status when applied early in life.[34] The number of publications citing the GMFCS is increasing every year, and this classification system has become internationally accepted in research and clinical use for clear communication among health professionals about gross motor functioning in children.[35]

The International classification of functioning, disability, and health (ICF) is a classification system created by the World Health Organization (WHO) in 2001 to establish a standard language and common framework for describing health and health-related conditions.[36] After this classification system was established for adults by the WHO in 2001, the International classification of functioning, disability and health-child and youth version (ICF-CY), which deals with children and young people and considers growth and development, was published in 2007.[37] The ICF-CY provides a useful framework for physical therapists to better understand health, functioning, activity, participation, contextual factors, and the impact of dystonia and choreoathetosis. ICF contributes to a comprehensive understanding of dyskinetic CP and allows therapists to manage it effectively.[32] Moreover, evaluating the body functions, structures, and activity levels of children and adolescents with dystonia and choreoathetosis within the framework of the ICF-CY may provide more effective clinical management of dyskinetic CP.[7]

Currently, there is no consensus on most applicable, reliable, and valid tools used for the evaluation of choreoathetosis and dystonia in children with dyskinetic CP.[38] This systematic review aimed to describe and critically examine the rate of clinical use of tools reported to assess body structure function and activity under ICF-CY for clinical types of dystonia and choreoathetosis in children and adolescents with CP. Additionally, we aim to provide an overview of the parameters that can be derived from these measurement tools.

## METHODS

### Search Strategy

A systematic search was conducted in June 2024 using four electronic databases: PubMed, Embase, Scopus, and Google Scholar. The search strategy included the following blocks.

**Diagnosis:** terms related to cerebral palsy,

**Movement disorder:** Terms such as dyskinesia, choreoathetosis, and dystonia,

**Body structure, function, and activity:** Terms related to body structure function and activity.

The detailed search strategy for each database is provided in Supplementary Table 1.

**Table 1. Inclusion and exclusion criteria defined in line with the PICOS design framework**

| | Definition | Inclusion/Exclusion criteria |
|---|---|---|
| **Participants** | Dyskinetic CP, patients aged 2-24 years | • The study sample or a significant number of subjects (50% minimum) are represented in the study population or in a sub-study population analyzed separately.<br>• Because the definition of dyskinetic CP is not always clear, studies describing "dystonia and choreoathetosis ocurring with CP" were included. |
| **Intervention** | Instrumented Measurements to assess body structure-function and activity | • Studies in which body structure function and activity evaluations were performed and the results of these evaluations were specified were included.<br>• In studies using video recordings, computerized analysis techniques that required observational video scores were also included. |
| **Comparison** | No control or comparison group was required | • Studies that compared the effect of the intervention assessed with a clinical test, control group, or methods were included. If there was no comparison, the method was presented in the review. |
| **Outcome** | ICF-CY categories | • Body structure function and activity categories were included in the ICF-CY categories.<br>• Other ICF-CY categories (for example, mental function, sensory function, pain, or self-care) were excluded. |
| **Study design** | Original research studies and and participants and presenting sufficient knowledge of the methodology are included | • There are no restrictions on the type of studies: technical reports, case studies, case control studies, and intervention studies, etc.<br>• Articles published in languages other than English were also excluded.<br>• Only full-text articles were selected for both abstract and full-text articles published using the same data/methodology. |

PICOS: Participants, intervention, comparison, outcome, study, CP: Cerebral palsy, ICF-CY: International classification of functioning, disability and health-child and youth version

### Inclusion Criteria

The inclusion criteria were established using the PICOS framework.

**Participants:** Studies involving individuals diagnosed with cerebral palsy.

**Intervention:** Studies examining movement disorders (e.g., dyskinesia, choreoathetosis, dystonia).

**Comparison:** Studies that included a control group or comparative intervention.

**Outcome:** Studies reporting body structure function and activity outcomes.

**Study design:** Randomized controlled trials, cohort studies, case-control studies, and cross-sectional studies.

### Selection Process

The search results were imported into EndNote for reference. Duplicate records were removed prior to screening. The initial screening of the titles and abstracts was performed by a single reviewer (EB). The full texts of potentially eligible studies were retrieved and assessed in detail against the inclusion criteria by the same reviewer (EB).

### Data Extraction

Data extraction was performed using a standardized form. The extracted data included the following.

- Study characteristics (author, year, country),
- Participant characteristics (sample size, age, gender, diagnosis),
- Intervention details,
- Outcomes measured,
- Main findings.

### Quality Assessment

The quality of the included studies was assessed using the Cochrane Risk of Bias tool for randomized controlled trials and Newcastle-Ottawa Scale for observational studies. Risk of bias assessment was independently conducted by two reviewers (EB and MG). Discrepancies were resolved through discussion or by consulting with a third reviewer (CO).

### Data Synthesis

Narrative synthesis of the findings was conducted because of the heterogeneity of the included studies. Where possible, a meta-analysis was performed using a random-effects model to account for the variability among studies. Statistical heterogeneity was assessed using the $I^2$ statistic.

### Resolution of Disagreements

Any disagreements encountered during article selection, data extraction, or quality assessment processes were resolved by discussion between the primary reviewer (EB) and the second (MG) and third reviewers (CO).

## RESULTS

Table 1 presents the inclusion and exclusion criteria created using the preferred reporting ıtems for systematic reviews and meta-analyses (PRISMA). After removing duplicate articles from the search results, 11,800 records remained. Forty-six articles meeting the inclusion and exclusion criteria were reviewed for further eligibility assessment, and the full texts were obtained.

In 34 of 46 articles, body structure function and activity evaluation of clinical types of dystonia and choreoathetosis in children and adolescents with CP, children, and young people with dyskinetic CP were included as the main participant group or as a separate subgroup, and the results have been reported accordingly. The descriptions of the included articles are shown in Table 2.

### Function and Disability

Body functions and structures: Body structure function was evaluated in 32.3% of the 34 studies. The Dyskinesia Impairment Scale (DIS) was used in 14.71% of these studies; Barry-Albright Dystonia Scale (BADS) in 11.7%; Burke-Fahn-Marsden Dystonia Rating Scale-Movement (BFM-M) in 15.4%; as intellectual and executive function evaluation criteria, Raven's colored progressive matrices were used in 15.4%, the stop signal task in 15.4%, the Wisconsin card sorting test in 15.4%, the stockings of Cambridge test in 15.4%, magnetic resonance (MR) imaging in 20.5%, pattern/verbal recognition memory task in 7.6%, Benton's facial recognition test in 7.6%, Benton's judgment of line orientation test in 7.9%, The peabody picture vocabulary test in 7.6%, the computer-based instrument for low motor language testing (C-BilLLT) in 5.8%, IQ testing in 5.8%, Movement Disorder-Childhood Rating Scale (MD-CRS 4-18) in 2.9%, The neonatal neuroimaging classification system (NNICS) in 2.9%, spasticity test (SPAT) in 2.9%, Visual Analogue Scale (VAS) in 2.9%, pediatric evaluation of disability inventory (PEDI) in 2.9%.[4,21,39-54] Lexical Verbal Fluency tests in 15.4% Pena-Casanova et al.[55]

### Activity

Activity was evaluated in 34 studies. The following classification systems were used in these articles: GMFCS, 94.1%; MACS, 70.5%, CFCS 32.3%; EDACS, 11.7%; bimanual fine motor function (BFMF) classification, 11.7%; and VSS, 8.8%. The gross motor function measure (GMFM) was used in 5.8% of patients.[4,21,39-43,45,47-65]

### Gross Motor Function Classification System (GMFCS)

In the following studies, 27.9%-100% of the participants were classified as GMFCS V: Sun et al.[56] and Préel et al.[68] and Westbom et al.,[69] and Elze et al.[45] and Bonouvrié et al.,[51] Williams and Pountney,[66] Zouvelou et al.,[67] Bekteshi et al.[61] Vanmechelen et al.[39] and Monbaliu et al.[4] and Knights et al.,[11] and Eek et al.,[21] Carnahan et al.,[63] Monbaliu et al.,[41] Bonouvrie et al.,[42] Battini[54] and Unes et al.[53]

Of the participants, 42.8%-61% had GMFCS IV in the following studies: Shevell et al.,[70] Park et al.,[44] Andersen et al.[65] and Soleimani et al.[60]

In Gimeno et al.,[46] 85.7% of the participants had GMFCS IV or V (equal numbers of patients for each).

**Table 2. Description of included articles**

| Study | Population/setting | Intervention/aim | Body structure and function scales | Activity scales | Results | Design |
|---|---|---|---|---|---|---|
| Sun et al.[56] | Twenty-six participants (28.0%) had dystonic CP, 26 (28.0%) had choreoathetotic CP, and 41 (44.1%) had mixed CP. | Clinical characteristics and functional status of children with different subtypes of dyskinetic cerebral palsy. | | **GMFCS n (%)**<br>I 13 (13.9), II 9 (9.6), III 22 (23.6), IV 23 (24.7), V 26 (27.9),<br>**MACS**<br>I 7 (7.5), II 29 (31.1), III 12 (12.9), IV 18 (19.3), V 25 (26.8)<br>**CFCS**<br>I 19 (20.4), II 34 (36.5), III 17 (18.2), IV 17 (18.2), V 6 (6.4) | Functional classification levels were distributed unequally among the 3 subgroups (p<.01).<br>No significant difference between GMFCS and MACS was found among the 3 subgroups (p>.05).<br>Different subtypes of dyskinetic CP have specific comorbidities, radiological characteristics, and functional attributes according to their etiological factors and brain lesions. Children with dystonic CP have more limited functional status than children with choreoathetotic CP. | Observational study |
| Shevell et al.[70] | Spastic quadriplegia 85 (35)<br>Spastic hemiplegia 77 (31)<br>Spastic diplegia 52 (21)<br>Dyskinetic 16 (7)<br>Ataxic-hypotonic 9 (4)<br>Other 4 (2) | The relationship of cerebral palsy subtype and functional motor impairment: a population-based study. | | **Dyskinetic GMFCS**<br>I 1 (6.2), II 2 (12.5), III 1 (6.2), IV 7 (43.7), V 5 (31.2) | dSP subtype versus GMFCS levels I to III or IV to V was distributed pro-portionally as follows: spastic diplegic, 51/52 (98%) versus 1/52 (2%); spasticquadriparetic, 20/85 (24%) versus 65/85 (76%); spastic hemiplegic, 76/77 (99%)versus 1/77 (1%); dyskinetic, 4/16 (25%) versus 12/16 (75%); other (triplegic orataxic-hypotonic), 10/13 (77%) versus 3/13 (23%).<br>These distributions (propor-tions) all yielded significant (p<0.001) pearson v2values | Observational study |
| Ballester-Plané et al.[57] | Fifty-two subjects with dyskinetic CP (28 males, mean age 24 y 10 mo, SD 13 y) and52 typically-developing controls (age- and gender-matched) completed a comprehensive neuropsychological assessment. | Cognitive functioning in dyskinetic cerebral palsy: Its relation to motor function, communication and epilepsy. | | **GMFCS**<br>I 15 (28.8), II 8 (15.3), III 6 (11.5), IV 11 (21.1), V 12 (23)<br>**BFMF**<br>I 7 (13.4), II 12 (23), III 16 (30.7), IV 12 (23), V 5 (9.6)<br>**MACS**<br>I 5 (9.6), II 10 (19.2), III 17 (32.6), IV 10 (19.2), V 10 (19.2)<br>**CFCS**<br>I 17 (32.6), II 23 (44.2), III 6 (11.5), IV 6 (11.5), V 0 (0) | Dyskinetic CP participants performed worse than controls on all cognitive func-tions except for verbal memory.<br>Milder cases (GMFCS I) only showed impairment inattention, visuoperception and visual memory.<br>Participants with GMFCS IIeIII also showedimpairment in language-related functions. Severe cases (GMFCS IVeV) showed impairmentin intelligence and all specific cognitive functions but verbal memory.<br>CFCS was associated with performance in receptive language functions. Epilepsy was related to performance inintelligence, visuospatial abilities, visual memory, grammar comprehension and learning. | A case control study |
| Préel et al.[68] | The total number of CP cases was 1165 of which 92 had dyskinetic and 540bilateral spastic CP. | Children with dyskinetic cerebral palsy are severely affected as compared to bilateral spastic cerebral palsy. | | **Dyskinetic GMFCS**<br>I 119 6.7%, II 72 3.3%, III 50 5.6%, IV 158 22.2%, V 226 62.2% | Prevalence of dyskinetic CP was 0.16 per 1000 live births. Inparticipants with dyskinetic compared to bilateral spastic CP, there was more frequently an Apgar level less than five at five minutes (22.7% vs. 11.2%) and neonatal seizures (43.5% vs. 28.5%), but less respiratory deficiency, hyperbilirubinaemia and sepsis. Impairmentbased on gross motor function classification was more severe in dyskinetic CP (level III-V90.0% vs. 66.0%). In dyskinetic CP, there was a high rate of reduced development alquotient (68.1%), visual impairment (39.3%) and epilepsy (51.6%).<br>Basal ganglia lesions were more prevalent in dyskinetic compared to bilateral spastic CP (27.7% vs. 12.8%) | Observational study (Cross-sectional) |
| Westbom et al.[69] | Total 343 Dyskinetic (50) | Cerebral palsy in a total population of 4-11 year olds in southern Sweden. Prevalence and distribution according to different CP classification systems. | | **Dyskinetic: GMFCS**<br>I 8 (16), II 4 (8), III 5 (10), IV 14 (28), V 19 (36) | The prevalence of CP was 2.4/1.000 (95% CI 2.1-2.6) in children 4-11 years of age born in Sweden, excluding post-neonatally acquired CP.<br>Children born abroad had a higher prevalenceof CP with more severe functional limitations. In the total population, the prevalence of CP was 2.7/1.000 (95% CI 2.4-3.0) and 48% were GMFCS-level I (the mildest limitation of gross motor function).<br>One third of the children with CP, who were born or had moved into the area after a previous study in 1998, were not in the CPUP database. The subtype classification in the CPUP database was adjusted in the case of every fifth child aged 4-7 years not previously reviewed. | Observational study (Cross-sectional) |
| Ballester-Plané et al.[58] | 25 subjects with dyskinetic CP and 24 healthy controls | Whole-brain structural connectivity in dyskinetic cerebral palsy and its association with motor and cognitive function. | | **Dyskinetic CP, GMFCS**<br>I: 10 (40), II: 3 (12), III: 1 (4), IV: 4 (12), V: 7 (28)<br>**MACS**<br>I: 3 (12), II: 8 (32), III: 4 (16), IV: 3 (12), V: 7 (28) | Grossand fine motor functions correlated with FA in a pathway comprising the sensorimotor system, butgross motor also correlated with prefrontal, temporal and occipital connections. Intelligence correlatedwith FA in a network with fronto-striatal and parieto-frontal connections, and visuoperception was relatedto right occipital connections. These findings demonstrate a disruption in structural brain connectivity indyskinetic CP, revealing general involvement of posterior brain regions with relative preservation of pre-frontal areas. We identified pathways in which WM integrity is related to clinical features, including butnot limited to the sensorimotor system | Randomized controlled trial |
| Laporta-Hoyos et al.[47] | Thirty-nine participants (19 females, median age 21y) with DCP | Brain lesion scores obtained using a simple semi-quantitative scale from MR imaging are associated with motor function, communication and cognition in dyskinetic cerebral palsy. | 1.Intellectual functioning Raven's coloured progressive matrices.<br>2. Four domains of executive functions were assessed:<br>- Attentional control: the stop signal task (Cambridge Cognition, 1999)<br>- Cognitive flexibility: Wisconsin card sorting test (Kongs et al., 2000).<br>-Goal setting: Stockings of Cambridge test (Cambridge Cognition, 1999).<br>- Information processing: Lexical verbal fluency test (Peña-Casanova et al., 2009).<br>-Pattern/verbal recognition memory task (Cambridge Cognition, 1999).<br>-Benton's facial recognition test (Benton, 1994) and Benton's judgment of line orientation test (Benton, 1994)<br>-The Peabody picture vocabulary test third edition MRI | **Dyskinetic cerebral palsy GMFCS**<br>I: 15 (38.4), II: 7 (17.9), III: 3 (7.6), IV: 5 (12.8), V: 9 (23)<br>**MACS**<br>I: 5 (12.8), II: 10 (25.6), III: 12 (30.7), IV: 3 (7.6) V: 9 (23)<br>**CFCS**<br>I: 15 (38.4), II: 16 (41), III: 4 (10.2), IV: 4 (10.2), V: 0 | Brain lesions were most frequently located in the ventral posterior lateral thalamus and the frontal lobe.<br>Gross (B=0.180, p<.001; B=0.658, p<.001) and fine (B=0.136, p=.003; B=0.540, p<.001) motor function were associated with global sqMRI score and parietal involvement. Communication functioning was associated with putamen involvement (B=0.747, p<.028). Intellectual functioning was associated with global sqMRI score and posterior thalamus involvement (B=-0.018, p<.001; B=-0.192, p<.001).<br>Selective attention was associated with global sqMRI score (B=-0.035, p<.001), parietal (B=-0.063, p=.023), and corpus callosum involvement (B=-0.448, p<.001). Visuospatial and visuoperceptive abilities were associated with global sqMRI score (B=-0.078, p=.007) and medial dorsal thalamus involvement (B=-0.139, p<.012), respectively | Randomized controlled trial |
| Laporta-Hoyoset al.[59] | DCP [n ¼52; 24 females, median age 20.5 y: 5mo, interquartile range (IQR) ¼13.75 y: 7mo; GMFCS IeV] | Executive function and general intellectual functioning in dyskinetic cerebral palsy: comparison with spastic cerebral palsy and typically developing controls. | | **Dyskinetic cerebral palsy GMFCS**<br>I: 15 (80), II: 8 (15.3), III: 6 (11.5), IV: 11 (21.1), V: 12 (23)<br>**MACS**<br>I: 5 (9.6), II: 10 (19.2), III: 17 (32.6) IV: 10 (19.2),V: 10 (19.2)<br>**CFCS**<br>I: 17 (32.6), II: 23 (44.2), III: 6 (11.5), IV: 6 (11.5) | Both CP groups had lower intelligence than TDC and performed poorer in almost all EF tasks. Intelligence was higher in DCP than SCP (z ¼2.51, p ¼0.01). Participants with DCP also performed significantly better in goal-setting tasks (z ¼2.27, p ¼0.03) and in-formation processing (z¼2.54, p ¼0.01) than those with SCP. | Cross-sectional study |
| Elze et al.[45] | Children with HMDs, n=161; median age 10y 3mo, range 2y 6mo-21y | Burke-Fahn-Marsden dystonia severity, gross motor, manual ability, and communication function classification scales in childhood hyperkinetic movement disorders including cerebral palsy: a 'Rosetta stone'study. | BFM-M | **Dyskinetic cerebral palsy GMFCS**<br>I: 22 (13.6), II: 20 (12.4), III: 9 (5.5), IV: 21 (13),V: 89 (55.2)<br>**MACS**<br>I: 7 (4.3), II: 21 (13), III: 20 (12.4), IV: 26 (16.1),V: 87 (54)<br>**CFCS**<br>I: 25 (15.5), II: 26 (16.1), III: 36 (22.3), IV: 49 (30.4),V: 22 (13.6) | All four scales were strongly associated (all Spearman's rank correlation coefficientrs >0.72, p<0.001), with worse dystonia severity implying worse function. Secondary dystoniashad worse dystonia and less function than primary dystonias (p<0.001). A longer proportionof life lived with dystonia is associated with more severe dystonia (rs=0.42, p<0.001) | Cross-sectional study |

**Table 2. Description of included articles (continues)**

| Study | Population/setting | Intervention/aim | Body structure and function scales | Activity scales | Results | Design |
|---|---|---|---|---|---|---|
| Bonouvrié et al.[51] | We conducted a multicenter, randomized, double-blind, placebo-controlled trial at the AUMC, located at Free University AUMC, Amsterdam, and the MUMC, Maastricht, the Netherlands. | The effect of intrathecal baclofen in dyskinetic cerebral palsy: the IDYS trial. | MRI the C-BiLLT | **Dyskinetic cerebral palsy GMFCS**<br>IV: 8 (44.4), V: 10 (55.5)<br>**MACS**<br>III: 2 (11.1), IV: 4 (22.2), V: 12 (66.6) | Thirty-six patients were recruited from January 1, 2013, to March 31, 2018. Data for final analysis were availablefor 17 patients in the intrathecal baclofen group and 16 in the placebo group. Mean (standard deviation) GAS T score at3 months was 38.9 (13.2) for intrathecal baclofen and 21.0 (4.6) for placebo (regression coefficient=17.8, 95% confidenceinterval=10.4-25.0, p<0.001). Number and types of (serious) adverse events were similar between groups. | Sistematik review |
| Park et al.[44] | Twenty-three patients with dyskinetic CP (13 males, 10 females; mean age 34 years, range 16-50 years) | Neuroradiological and neurophysiological characteristics of patients with dyskinetic cerebral palsy. | BADS | **Dyskinetic cerebral palsy GMFCS**<br>I: 1 (4.3), II: 9 (39.1), III: 1 (4.3), IV: 11 (47.8), V: 1 (4.3) | Mean BADS was 16.4±5.0 in ambulatory group (GMFCS levels I, II, and III; n=11) and 21.3±3.9 in nonambulatory group (GMFCS levels IV and V; n=12). Twelve patients showed normal MRI findings, and eleven patients showed abnormal MRI findings (grade I, n=5; grade II, n=2; grade III, n=4). About half of patients with dyskinetic CP showed putamen and thalamus lesions on MRI.<br>Mean BADS was 20.3±5.7 in normal MRI group and 17.5±4.0 in abnormal MRI group. VBM showed reduced volume of the hippocampus and parahippocampal gyrus.<br>In DTT, no abnormality was observed in CST, but not in SLF. In MEPs, most patients showed normal central motor conduction time. | Randomized controlled trial |
| Williams et al.[66] | Three participants had dyskinetic quadriplegia, seven had spastic quadriplegia, and one had spastic diplegia. | Effects of a static bicycling programme on the functional ability of young people with cerebral palsy who are non-ambulant. | | **Dyskinetic cerebral palsy GMFCS**<br>V: 3 (100)<br>**GMFM** | Resultsshowed significant improvements in GMFM-66 (p=0.010)and in GMFM-88 dimensions D (Standing; p=0.012) and E (Walking, Running, and Jumping; p=0.011) over theintervention period, but not over the baseline or follow-upperiods. Significant improvements were found in 'cycling' ability for duration of pedalling (p<0.001), speed (p=0.01),and resistance (p=0.001). | Randomized controlled trial |
| Laporta-Hoyos et al.[48] | Thirty-three participants with dyskinetic CP (mean±SD age: 24.42±12.61, 15 female) were age and sex matched with 33 controls | White matter integrity in dyskinetic cerebral palsy: relationship with intelligence quotient and executive function. | 1.Intellectual functioning Raven's coloured progressive matrices.<br>2. Four domains of executive functions were assessed:<br>- Attentional control: the Stop signal task (Cambridge Cognition, 1999). Selective visual and<br>- Cognitive flexibility: Wisconsin card sorting test (Kongs et al., 2000).<br>-Goal setting: Stockings of Cambridge test (Cambridge Cognition, 1999).<br>- Information processing: Lexical verbal fluency test (Peña-Casanova et al., 2009).<br>MRI | **Dyskinetic CP GMFCS**<br>I: 12 (36.3), II: 6 (18.1), III: 3 (9), IV: 5(15.1),V: 7 (21.2)<br>**MACS**<br>I: 5 (15.1), II: 8 (24.2), III: 11 (33.3), IV: 2 (6), V: 7 (21.2)<br>**CFCS**<br>I: 14 (42.4), II: 13 (39.3), III: 2 (6), IV: 4 (12.1),V: 0 | White matter FA was significantly reduced in the CP group in all cerebral lobes, predominantly in regions connected with the parietal and to a lesser extent the temporal lobes. There was no significant correlation between IQ or any of the four executive function domains and WM microstructure in the control group. In participants with CP, lower IQ was associated with lower FA in all cerebral lobes, predominantly in locations that also showed reduced FA compared to controls. Attentional control, goal setting and information processing did not correlate with WM microstructure in the CP group. Cognitive flexibility was associated with FA in regions known to contain connections with the frontal lobe (such as the superior longitudinal fasciculus and cingulum) as well as regions not known to contain tracts directly connected with the frontal lobe (such as the posterior corona radiata, posterior thalamic radiation, retrolenticular part of internal capsule, tapetum, body and splenium of corpus callosum). | Randomized controlled trial |
| Soleimani et al.[60] | Unilateral Spastic 36<br>Bilateral Spastic 125<br>Ataxic 10<br>Dyskinetic 14<br>Unclassified(mixed) 15 | Cerebral palsy: Motor types, gross motor function and associated disorders. | | **Dyskinetic MACS**<br>I: 0, II: 3 (21.4), III: 1 (7.1), IV: 5 (35.7), V: 5 (35.7)<br>**GMFCS**<br>I: 2 (14.2), II: 2 (14.2), III: 0, IV: 6 (42.8), V: 4 (28.5) | During the study period, 200 CP children (103 males, 97 females) aged 4-12 years were seen with an overall male: female ratio of 1.06, with a mean (SD) age of 7.7 (2.4) years.<br>Level IV in MACS classification (23%) and also level IV in GMFCS classification (30.5%) were the more commons.<br>The remaining cases were distributed rather equally to other levels, near to (19-20%) to the MACS classification and (11-24.5%) to the GMFCS classification per level. | Observational study (Cross-sectional) |
| Andersen et al.[65] | Of the 294 children, 96 had the spastic unilateral CP type, 143 the spastic bilateral, 19 the dyskinetic 15 the ataxic type. In 21, the subtype could not be classified by the referring centre. | Cerebral palsy in Norway: prevalence, subtypes and severity. | | **Dyskinetic GMFM**<br>**GMFCS**<br>I- II: 0, III:2 (11), IV: 11 (61), V: 5 (28)<br>**Bimanual fine motor function (BFMF)**<br>I: 0, II: 0, III:1 (5), IV: 4 (21), V: 14 (74) | A total of 374 children with CP were identified with a prevalence of 2.1 per 1000 live births. Detailed information was obtained from 294 (79%) children. Median age at clinical assessment was 6.9 years (range: 1.9-10.2 years). Thirty-three percent of the children had spastic unilateral CP, 49% spastic bilateral, 6% dyskinetic, 5% ataxic CP and 7% were not classified. Severely impaired vision and hearing were present in 5% and 4% of the children, respectively. Active epilepsy was present in 28%, mental retardation in 31% and severely impaired or no speech in 28% children. The most severe impairments in gross motor function were observed in children with low Apgar scores, and the most severe impairments in fine motor function in children born at term, with normal birth weight and low Apgar scores. | Observational study (Cross-sectional) |
| Zouvelou et al.[67] | Fifteen patients manifested with features resembling dyski-netic CP (12 dystonic and 3 choreic), 9 resembled spastic CP, 5ataxic CP and 18 mixed CP. | The genetic etiology in cerebral palsy mimics: The results from a Greek tertiary care center. | | **Dyskinetic GMFCS**<br>I: 3 (20), II: 1 (6.6), III:3 (20), IV: 1 (6.6), V: 7 (46.6) | 31.91% of patients manifested with features resembling dyskinetic CP, 19.14% spastic CP, 10.63% ataxic CP and 38.30% mixed CP. In 23 patients molecular diagnosis was reached and included 5 hereditary SPG in spastic CP mimics; HPRT1, TH, QDPR, DDC in dystonic CP mimics; ADCY5 and NIKX2-1 in choreic CP mimics; ANA1Ain ataxic CP mimics; andSPG, PDHA1, NIKX2-1, AT, SLC2A1 and SPRin mixed CPmimics. In 14 patients, the etiological diagnosis led to specific treatment. | Retrospective study |
| Bekteshi et al.[39] | 53 participants with DCP | Clinical presentation of spasticity and passive range of motion deviations in dyskinetic cerebral palsy in relation to dystonia, choreoathetosis, and functional classification systems. | | **Dyskinetic GMFCS**<br>I: 10 (18.8), II: 5 (9.4), III:5 (9.4), IV: 7 (13.2), V: 26 (49)<br>**MACS**<br>I: 8 (15), II: 5 (9.4), III:6 (11.3) IV: 10 (18.8), V: 24 (45.2) | Spasticity and limited pROM were correlated with dystonia of the upper limbs (0.41<rs<p<.002) and lower limbs (0.31<rs<p<.025), and both functional systems of gross motor (0.32<rs<p<.018) and fine manual abilities (0.34<rs<p<.014). Hypermobility is correlated only with choreoathetosis of the lower limbs (0.44, p=.001). | Cross-sectional study |
| Vanmechelen et al.[39] | Fifty-two subjects with dyskinetic CP | Presence and severity of dystonia and choreoathetosis overflow movements in participants with dyskinetic cerebral palsy and their relation with functional classification scales. | Dyskinesia Impairment Scale | **Dyskinetic GMFCS**<br>I: 9 (17.3), II: 4 (7.6), III:8 (15.3), IV: 9 (17.3), V: 22 (42.3)<br>**MACS**<br>I: 7 (13.4), II: 4 (7.6), III: 8 (15.3), IV: 13 (25), V: 20 (38.4)<br>**CFCS**<br>I: 6 (11.5), II: 18 (34.6), III:20 (38.4), IV: 7 (13.4), V: 1 (1.9)<br>**EDACS**<br>I: 8 (15.3), II: 19 (36.5), III:12 (23), IV: 10 (19.2), V: 3 (5.7)<br>**VSS**<br>I: 7 (13.4), II: 11 (21.1), III:13 (25), IV: 21 (40.3) | Dystonia and choreoathetosis overflow movements were simultaneously present.<br>Median scores of dystonia overflow movements were significantly higher than choreoathetosis overflow movements.<br>Dystonia and choreoathetosis overflow movements were significantly higher in extremities than in the central body.<br>Correlations between dystonia and choreoathetosis overflow movements were fair. Moderate to good correlations were found between dystonia overflow score and gross motor function classification system, manual ability classification system, and eating and drinking ability classification system. | Cross-sectional study |
| Monbaliu et al.[4] | 55 participants | Clinical patterns of dystonia and choreoathetosis in participants with dyskinetic cerebral palsy. | DIS, MRI | **Dyskinetic GMFCS**<br>I: 10 (18.1), II: 5 (9), III:6 (10.9), IV: 7 (12.7), V: 27 (49)<br>**MACS**<br>I: 5 (9), II: 8 (14.5), III:7 (12.7), IV: 11 (20),V: 24 (43.6)<br>**CFCS**<br>I: 6 (10.9), II: 20 (36.3), III:18 (32.7), IV: 8 (14.5), V: 3 (5.4) | Dystonia and choreoathetosis are simultaneously present.<br>Median levels of dystonia (70.2%) were significantly higher than levels of choreoathetosis (26.7%) and both were significantly higher during activity than at rest (both p<0.01).<br>High correlations were found between dystonia levels and GMFCS level (Spearman's rank correlation coefficient, rs=0.70; 95% CI 0.53-0.81; p<0.01) and MACS (rs=0.65; 95% CI 0.47-0.81; p<0.01),and fair correlation with CFCS (rs=0.36; 95% CI=0.11-0.57; p<0.05).<br>No significant correlation was found between choreoathetosis levels and motor classifications.<br>Finally, higher choreoathetosis levels were found in participants with pure thalamus and basal ganglia lesions (p=0.03) than mixed lesions, but not for dystonia (p=0.41) | Cross-sectional study |

**Table 2. Description of included articles (continues)**

| Study | Population/setting | Intervention/aim | Body structure and function scales | Activity scales | Results | Design |
|---|---|---|---|---|---|---|
| Monbaliu et al.[40] | 55 participants | Functional outcomes in children and young people with dyskinetic cerebral palsy. | DIS | **Dyskinetic GMFCS** I: 10 (18.1), II: 5 (9), III: 6 (10.9), IV: 7 (12.7), V: 27 (49) **MACS** I: 4 (7), II: 8 (15), III:7 (13), IV: 12 (22), V: 24 (44) **EDACS** I: 9 (16), II: 18 (33), III:10 (18), IV: 12 (22), V: 6 (11) **CFCS** I: 6 (10.9), II: 20 (36.3), III: 18 (32.7), IV: 8 (14.5),V: 3 (5.4) **VSS** I: 8 (14.5), II: 12 (21.8), III:11 (20), IV: 24 (43.6) | Over 50 per cent of the participants exhibited the highest limitation levels in GMFCS, MACS, and VSS. Better functional abilities were seen in EDACS and CFCS.<br>Moderate to excellent interrelationship was found among the classification scales.<br>All scales had significant correlation (rs=0.65-0.81) with dystonia severity except for CFCS in the young people group.<br>Finally, only MACS (rs=0.40) and EDACS (rs=0.55) in the young people group demonstrated significant correlation with choreoathetosis severity | Cross-sectional study |
| Butler et al.[62] | 7 Dyskinetic cerebral palsy | Temporal-spatial parameters of the upper limb during a reach & grasp cycle for children. | | **MACS** I: 2 (28.5), II: 2 (28.5), III: 3 (42.8) | Heconsistent normative data and the substantial differences between children with CP and controls reflectutility of the reach & grasp cycle for quantitative evaluation of upper limb motor deficits. | Retrospective study |
| Gimeno et al.[46] | Secondary CP dystonia 14 | Evaluation of functional goal outcomes using the COPM following DBS in childhood dystonia. | COPM, BFMDR | **GMFCS** I-III: 2 (14.2), IV-V: 12 (85.7) **MACS** I-III: 1 (7.1), IV-V: 13 (92.8) | DBS improved functional performance, independently of thedystonic phenotype.<br>Improvements in individualized COPM functional goal areas wereseen in the absence of significant changes in BFMDRS scores, highlighting the relativeinsensitivity of impairment scales in this patient group. | Retrospective study |
| Eek et al.[21] | 25 dyskinetic cerebral palsy | Intrathecal baclofen in dyskinetic cerebral palsy: effects on function and activity. | Barry-Albright Dystonia Scale Modified Ashworth Scale plastic goniometer | **Gross motor function measure 88-item version GMFCS** IV: 5 (20), V: 20 (80) **MACS** III: 1 (4), IV: 3 (12), V: 21 (84) **BFMF** III: 1 (4), IV: 6 (24), V: 18 (72) | After ITB in individuals with dyskinetic CP, improvements were found in sitting, communication, and fine motor skills. There was a reduction in dystonia and muscle tone, and pain and sleep improved. | A retrospective cohort study |
| Carnahan.[63] | 51 dyskinetic | Association between GMFCS and MACS in children with cerebral palsy. A population-based study of 359 children. | | **GMFCS** I: 4 (7.8), II: 2 (3.9), III:7 (13.7), IV: 17 (33.3),V: 21 (41.1) **MACS** I: 5 (9.8), II: 4 (7.8), III:5 (9.8) IV: 11 (21.5), V: 26 (50.9) | Gross motor function and manual ability are often discrepant in children with CP, and the patterns seem to vary across the different subgroups based on the predominant neurological findings. To give a complete clinical picture when evaluating these children, both aspects have to be described. The GMFCS and the MACS seem to work well in this context and seem very useful in population-based studies, in health care registers for children with CP, and in clinical practice. | Cross-sectional study |
| Eliasson.[26] | 19 dyskinetic CP | The MACS for children with cerebral palsy: scale development and evidence of validity and reliability. | | **MACS** III:3 (15.7), IV: 4 (21), V: 12 (23.5) | The results demonstrated that MACS has good validity and reliability. The intraclass correlation coefficient between therapists was 0.97 (95% confidence interval 0.96-0.98), and between parents and therapist was 0.96 (0.89-0.98), indicating excellent agreement. | |
| Monbaliu.[67] | 55 Dyskinetic Palsy | The relationship of dystonia and choreoathetosis with activity, participation and quality of life in children and youth with dyskinetic cerebral palsy. | DIS | **GMFCS** I: 9 (17), II: 5 (9), III: 6 (11), IV: 7 (13), V: 27 (50) **MACS** I: 4 (7), II: 8 (15), III: 7 (13), IV: 11 (21),V: 24 (44) **GMFM the FMS the JTT the ABIL-K the LIFE-H** | This cross-sectional study is the first to examine the relationship of dystoniaand choreoathetosis in dyskinetic CP with the level of activity, participation and QOL.<br>The results revealed dystonia has a higher impact on activity, participation and quality of lifethan choreoathetosis. These findings seem to suggest it is necessary to first focus ondystonia reducing intervention strategies and secondly on choreoathetosis. | |
| Dhondt et al.[52] | CP subtype classification was not possible for 26 children (2.3%). Among 1101 children included in subtype-stratified analyses:<br>•Spastic CP: 948 children (86.1%)<br>•Dyskinetic CP: 105 children (9.5%) (including dystonic dyskinesia in 59 and choreo-athetotic dyskinesia in 14 children)<br>•Ataxic CP: 48 children (4.4%) | To report on the prevalence, neuroimaging patterns, and function of children with CP in Belgium for birth years 2007-2012, and identify distinctive risk indicators and differences in outcome between CP subtypes. | IQ testing MRICS the NNICS | **GMFCS** 20 (19.0), 25 (23.8), 11 (10.5), 23 (21.9), 24 (22.9), 2 (1.9) **MACS** 11 (10.5), 16 (15.2), 23 (21.9), 13 (12.4), 24 (22.8), 18 (17.1) **BFMF** 9 (8.6), 20 (19.0), 20 (19.0), 16 (15.2), 19 (18.1) **Missing** 21 (20.0) **VSS** 12 (11.4), 22 (21.0), 22 (21.0), 31 (29.5) **Missing** 18 (17.1) | In total, 1127 children with CP were identified in Belgium. The birth prevalence of overall CP was 1.48 per 1000 live births. The likelihood of dyskinetic CP increases if the child was born to a mother aged ≥35 years, mechanically ventilated, and had predominant grey matter injury, while an increased likelihood of ataxic CP is associated with ≥2 previous deliveries. Children with dyskinetic and ataxic CP are more likely to function with impairments in motor, speech, and intellectual abilities. | Observational study |
| Vanmechelen et al.[78] | Fifty-two subjects with dyskinetic CP were included. | This cross-sectional study aims to investigate the presence and severity of overflow move- ments of dystonia and choreoathetosis in dyskinetic CP and to assess the relationship of overflow movements with functional classification scales. | DIS | **GMFCS** 9 (17.3), 4 (7.6), 8 (15.3), 9 (17.3), 22 (42.3) **MACS** 7 (13.4), 4 (7.6), 8 (15.3), 13 (25), 20 (38.4) **CFCS** 6 (11.5), 18 (34.6), 20 (38.4), 7 (13.4), 1 (1.9) **EDACS** 8 (15.3), 19 (36.5), 12 (23), 10 (19.2), 3 (5.7) **VSS** 7 (13.4), 11 (21.1), 13 (25), 21 (40.3) | Dystonia and choreoathetosis overflow movements were simultaneously present. Median scores of dystonia overflow movements were significantly higher than choreoathetosis overflow movements. Dystonia and choreoathetosis overflow movements were significantly higher in extremities than in the central body. Correlations between dystonia and choreoathetosis overflow movements were fair. Moderate to good corre-lations were found between dystonia overflow score and gross motor function classification system, manual ability classification system, and eating and drinking ability classification system. | Cross-sectional study |
| Bonouvrié et al.[20] | A multi-center prospective cohort study was conducted including 34 non-walking individuals with severe dyskinetic CP | To assess attainment of individual treatment goals one year after ITB pump implantation in individuals with dyskinetic CP. | BADS<br>DIS<br>SPAT<br>VAS<br>PEDI<br>C-BiLLT | **GMFCS IV** 13 (38.2%), V 21 (61.7%) **MACS** III 3 (8.8%), IV 9 (26.4%), V 22 (64.7%) | Seventy-one percent of individuals with dyskinetic CP fully achieved one or more treatment goals. One or more treatment goals were partially achieved in 97% of individuals. Two factors were found to be associated with attainment of goals: DIS score at baseline and the difference in pain score between baseline and follow-up. These two variables explain 30% of the variance in the outcome. | A multi-center prospective cohort study |
| 33. Stewart et al.[43] | Fifty-seven children with dyskinetic CP | To outline the development and examine the content and construct validity of a new tool, the D-FIS, which measures the impact of dyskinesia on everyday activities in children with CP. | BADS<br>D-FIS | **GMFCS** 8 (14), 6 (11), 7 (12), 23 (40), 13 (23) **MACS** 0, 19 (33), 12 (21), 12 (21), 14 (25), 0 **CFCS** 16 (28), 13 (23), 14 (25), 10 (17), 4 (7) **EDACS** 16 (28), 19 (33), 11 (19), 1 (2), 10 (17) | Fifty-seven parents of children [29 males, 28 females, mean [SD] age 11y 8mo (4y 4mo), range 2y 6mo-18y] completed the D-FIS. Correlation between D-FIS and GMFCS was r=0.86 (95% CI: 0.77–0.91, p<0.001); MACS r=0.84 (95% CI: 0.73-0.90, p<0.001); CFCS r=0.80 (95% CI: 0.67-0.88, p<0.001); and EDACS r=0.78 (95% CI: 0.66-0.87). Correlation between D-FIS and BADS was r=0.77 (95% CI: 0.64-0.86, p<0.001). Cronbach's alpha was 0.96. | Observational study |
| Battini et al.[54] | this measurement-focused study was carried out on a cohort of 57 participants with dcp | MD-CRS 4-18 is a tool aimed to evaluate movement disorders in developmental age, validated since 2008 and applied in the literature. | MD-CRS 4-18 | **GMFCS** 8 (14), 1 (2), 3 (5), 12 (21), 33 (58), 8, 1, 3 | This study supports the relevant contribution of MD-CRS 4-18 r to identify the severity of movement disorders in dyskinetic cerebral palsy, as indicated by the higher icc values on index ii compared to previous MD-CRS 4-18 results. SEM and Minimally detectable difference (Mdd) of MD-CRS 4-18 r in dcp were all very low, with sEMs ranging from 0.01 to 0.02 and Mdd from 0.03 to 0.06. | Observational study |
| Unes et al.[53] | The clinical types were spastic (n=159, 74 unilateral, 85 bilateral), dyskinetic (n=43, 34 dystonic, 9 choreoathetotic), ataxic (n=7) and unclassified (n=16) according to the SCPE classification | To investigate the relationships between four functional classification systems in children with CP and parent-interpredicted intelligence level, and the functional status in clinical types of CP. | | **GMFCS** 5 (12), 4 (9), 5 (12), 12 (27), 17 (40) **MACS** 3 (7), 6 (14), 11 (26), 5 (11), 18 (42) **CFCS** **EDACS** | Correlations were found between all functional levels; the strongest were between GMFCS-MACS (r=0.784, p<.001), CFCS-EDACS (r=0.772, p<.001). Strong correlations were found for the IQ-CFCS (r=0.762, p<.001) and IQ-EDACS (r=0.634, p<.001). Correlations were stronger in children with bilateral CP and IQ level <70. | Observational study |

**CP: Cerebral palsy, CI: Confidence interval, MD-CRS 4-18: Movement disorders-childhood rating scale for age 4-18, HMDs: Hyperkinetic movement disorders, BFM-M: Burke-Fahn-Marsden Dystonia Rating Scale-movement, C-BiLLT: Computer-based instrument for low motor language testing, COPM: Canadian occupational performance measure, DBS: Deep brain stimulation, BFMDR: Burke-Fahn-Marsden Dystonia Rating Scale, MRICS: MRI classification system, AUMC: Amsterdam University Medical Center, MUMC: Maastricht University Medical Center, NNICS: Neonatal neuroimaging classification system, DIS: Dyskinesia Impairment Scale, SPAT: Spasticity test, VAS: Visual Analogue Scale, PEDI: Pediatric evaluation of disability inventory, ITB: Intrathecal baclofen, D-FIS: Dyskinetic Cerebral Palsy Functional Impact Scale, MACS: Manual ability classification system, GMFCS: Gross motor function, SPG: Spastic paraplegia genes, VSS: Viking Speech Scale, BADS: Barry-Albright Dystonia Scale, FMS: Functional Mobility Scale, JTT: Jebsen-Taylor hand function test, ABIL-K: ABILHAND-kids questionnaire, LIFE-H: LifeHabits kids, SEM: Standard error measurement**

In the following studies, 35.9%-80% of the prticipants were GMFCS I: Ballester-Plané et al.[57] and Ballester-Plané et al.[58] and Laporta-Hoyos et al.,[47] and Laporta-Hoyos et al.,[48] Bonouvrie.[42]

## Manual Ability Classification System (MACS)

In the following studies, 23.5%-84% of participants had MACS V: Elze et al.,[45] Bekteshi et al.,[61] Vanmechelen et al.,[39] Bonouvrié[51] and Monbaliu et al.,[4] Monbaliu et al.,[40] Butler et al.,[62] Eek et al.[21] Carnahan et al.,[63] Eliasson et al.,[26] and Monbaliu et al.,[41] Bonouvrie,[42] Stewart et al.[43] and Unes et al.[53]

Soleimani et al.[60] and Gimeno et al.,[46] 35.7% and 92.8% of the participants were MACS IV or MACS V, respectively, and they were equal in each study.

In subsequent studies, 17-33.3% of the participants were MACS III, and Laporta-Hoyos et al.,[47] Laporta-Hoyos et al.,[59] Laporta-Hoyos et al.,[48] Dhondt et al.,[52] Stewart et al.[43] and Unes et al.[53]

Sun et al.[56] and Ballester-Plané et al.,[58] and 31.1% and 32% of the participants were MACS II.

## Communication Function Classification System (CFCS)

Elze et al.[45] found that 13.6% of the participants had CFCS IV.

In Vanmechelen et al.,[39] 38.4% of the participants were classified as CFCS III.

In these studies, 36.3%-44.2% of the participants had CFCS II. Sun et al.[56] Laporta-Hoyos et al.,[47] and Laporta-Hoyos et al.,[59] Monbaliu et al.[4] and Monbaliu et al.[40]

A study by Laporta-Hoyos et al.,[48] Stewart et al.[43] and Unes et al.,[53] 28% -44% of the participants were CFCS I.

## Bimanual Fine Motor Function (BFMF) Classification

In Andersen et al.[65] and Eek et al.,[21] 72% and 74% of the participants had BMFM V, respectively.

In Ballester-Plané et al.,[57] Dhondt et al.,[52] 15.2% and 30.7% of participants had BMFM III.

## Eating and Drinking Ability Classification System (EDACS)

Vanmechelen et al.,[39] Monbaliu et al.,[40] Stewart et al.,[43] 33% and 36.5% of the participants were EDACS II, respectively.

## Viking Speech Scale (VSS)

In Vanmechelen et al.,[39] Monbaliu et al.[40] and Dhondt et al.,[52] 29.5% and 43.6% of the participants had CFCS IV, respectively.

## Gross Motor Function Measure (GMFM)

Williams and Pountney,[66] Andersen et al.,[65] and Monbaliu et al.[40] used GMFM as an activity measure in their studies.

# DISCUSSION

Our study provides an overview of instrumented measures used to assess body structure function and activity as part of ICF-CY in children and youth with CP and with clinical types of dystonia and choreoathetosis. The current study can guide researchers and clinicians in making informed decisions regarding evaluations for specific purposes in dyskinetic CP. In dyskinetic CP, there are a number of instrumented classification systems for assessing activity directly. Most body function assessment tools assess involuntary movements that are not directly related to the desired task, expressed as an excessive flow of muscle activation.

## Body Structure and Function

This systematic review identified scales reported to measure dystonia and choreoathetosis in children with CP. All of these scales assess dystonia, but only the DIS assesses choreoathetosis in addition to dystonia. Two scales, the BADS and DIS, were designed primarily to measure secondary dystonia in people with CP. Other scales have been designed to assess primary dystonia or more than one type of movement disorder. Three articles evaluated dystonia and choreoathetosis at rest and during activities.[39,40,66] These assessments were performed using the DIS. This scale allows researchers to identify explosive movements found in dystonia and choreoathetosis and to map them in the body according to clinical patterns. In these articles, explosive movements of dystonia and choreoathetosis were evaluated simultaneously using this scale. Considering the reviewed articles, this scale has broadened our perspective on the structural and functional profiles of individuals with dyskinetic CP. Considering the reviewed articles, the DIS scale has expanded our perspective on the structural and functional profiles of individuals with dyskinetic CP. The DIS enables body structure and function assessment of dystonia and choreoathetosis in dyskinetic CP, together with functional classification systems that assess gross motor skills, upper extremity function, eating and drinking ability, communication, and speech.

In the articles included in our review, the clinical benefits of MRI imaging used in body structure-function assessment in children with dyskinetic CP have been shown to establish reasonable relationships between lesions in regions of the brain other than those that cause dyskinetic CP in general and add more value to clinical feedback.[45,46] However, a large proportion of individuals with dyskinetic CP have severe intellectual disabilities.[62,67] Tests assessing intellectual and executive function in the studies were carefully selected to allow most participants to respond autonomously. In the studies, the results of these tests were transferred to a computer environment and the use of assistive technology for communication was allowed. Participants were encouraged to use an assessment scale appropriate for their degree of disability and the communication devices that they normally use.

The impact of treatment options on different types and severity of dyskinetic CP also needs to be fully investigated. Therefore, the determination of dyskinetic CP and the dominant subclinical type requires clear evaluation and reporting criteria. One of the purposes of this review is to bridge this gap and identify and critically examine tools that assess body structure and function.

## Activity

Most of the methods reviewed in the present study had this in common: they assess manual dexterity and/or ambulation activity during tasks that require some level of understanding of the task instructions. The methods reviewed here provide the full functional profile of individuals with dyskinetic CP, including areas of speech, eating, and drinking, using current classification systems in addition to gross motor, dexterity, and communication classification. In most of the reviewed articles, individuals with dyskinetic CP had severe deficiencies in gross motor function, manual dexterity, and speech production. Therefore, a large number of children and adolescents with dyskinetic CP have been evaluated using only a few instrumentation methods.

The literature clearly demonstrates a high correlation between all CP subtypes that occur with significant frequency and the level of GMFCS (the low number of children with the ataxic-hypotonic variant precludes definitive analyses and statements regarding this subtype).[68] The GMFCS is a particularly valid method for assessing gross motor function, which ultimately reflects the ability to act independently. It is quite unusual and rare for a child with spastic diplegia or hemiplegia to be unable to ambulate independently. Ambulation is an important question asked by parents when their children are first diagnosed. Independent walking, however, is an important determinant of participation, leading to improved individual quality of life.[69,70] Independent ambulation occurs only in a small group of children with spastic quadriplegia or dyskinesia. Therefore, when the neurologic subtype is known, a strong prediction of ambulation-related functional status can be made, emphasizing that the GMFCS is a strong predictor. Conversely, the determined functional status can provide information about which neurological subtypes may occur.

Intervention outcomes for children with dystonia and other hyperkinetic movement disorders are often defined using a disorder-based dystonia rating scale such as the Burke–Fahn–Marsden Dystonia Rating Scale-Movement (BFM-M). However, without additional data, scores on such scales allow only limited inferences about an individual's functional status.[45] The use of functional classification scales such as the GMFCS, MACS, and CFCS provides a common language for describing motor affect severity and functional status of both patients and research participants. These scales require no formal clinician training, are fast, cost-effective, and easy to administer. They help contextualize BFM-M scores and facilitate the interpretation of research results by providing a clearer understanding of the functional abilities of the study participants. These classification systems, which reflect performance in daily life by focusing on function, provide meaningful information beyond etiology and disorder, as recommended by the ICF.[45]

Finer abilities in eating and drinking functions are a rare finding compared to gross motor and upper extremity functions, and this is consistent with the results of communication assessments in studies on dyskinetic CP.[4] However, speech production was not evaluated using the evaluation criteria of the EDACS and CFCS. This can be explained by one of the components evaluated at these scales. Communication function and eating and drinking abilities are supported by changes in the motor development processes. The focus of the EDACS and CFCS is to define an individual's ability to eat and drink safely and perform their daily communication function regardless of the communication method used. Speech production, respiratory control, phonation, and articulation were assessed using the VSS. The VSS, together with scales evaluating communication skills and eating and drinking abilities, provided complementarity to profile oral-motor function and upper/lower motor function in children with dyskinetic CP.

The GMFM, used as a measure of activity in the articles described in this systematic review, has been found to be successful in distinguishing various domains of motor dysfunction in children with CP and with spastic diplegia and athetosis. This criterion provides guidance for the development of children's participation in their functional activities, and therefore, in their activities in individual, family, and social circles.[71] In our literature review, we observed that the GMFM was used as an activity evaluation criterion for individuals with dyskinetic CP.

Body structure function and activity evaluations were performed at the ICF-CY level in the articles reviewed in this review. However, none of the articles evaluated the body structure function and activity in the daily environment of the participants. Therefore, it is controversial whether the results can be generalized to real-life situations. Many children and young people with dyskinetic CP depend on wheelchairs (manual or electric) in daily life. Therefore, it may be useful to evaluate the quality and duration of wheelchair use using instrumented methods in daily life. Evaluating wheelchair performance (or the performance of different controllers for motorized wheelchair mobility) in a virtual environment, as recently reported, is a very interesting option for this group.[72]

In recent years, wearable sensor technologies have been increasingly used to detect certain movements, such as neurological disorders, epilepsy, and stereotypical movement patterns including CP.[73-75] However, no study has specifically investigated dyskinetic CP. Wearable sensors allow the monitoring of dyskinetic movements in daily life outside the laboratory environment. Considering the severity of the disorder, movements may change over time and may be exacerbated by external stimuli such as stress, pain, and noise.[66] For this reason, an evaluation that can be performed for a longer period of time in the daily environment may provide more reliable evaluation results in children and adolescents with dyskinetic CP.[76-79]

## CONCLUSION

This review presents the instrumented measures used as objective outcome measures in patients with dyskinetic CP. Future studies should aim to develop instrumented measurements that can be applied outside the laboratory with new technological developments. This is especially important for severely disabled young adults and children with dyskinetic CP.

## ETHICAL DECLARATIONS

### Referee Evaluation Process

Externally peer-reviewed.

### Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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**Author Contributions**

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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