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Dyskinetic Cerebral Palsy Functional Impact Scale: development and validation of a new tool

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ABBREVIATIONS

BADS	Barry Albright Dystonia Scale
CFCS	Communication Function Classification System
D-FIS	Dyskinetic Cerebral Palsy Functional Impact Scale
EDACS	Eating and Drinking Ability Classification System
GMFCS	Gross Motor Function Classification System
MACS	Manual Ability Classification System

AIM To outline the development and examine the content and construct validity of a new tool, the Dyskinetic Cerebral Palsy Functional Impact Scale (D-FIS), which measures the impact of dyskinesia on everyday activities in children with cerebral palsy (CP).

METHOD D-FIS content was informed by a systematic review of dyskinesia outcome measures, in collaboration with children with dyskinetic CP, parents, caregivers, and expert clinicians. The D-FIS uses parent proxy to rate impact of dyskinesia on everyday activities. Construct validity was determined by examining internal consistency; known groups validity with the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), Communication

Function Classification System (CFCS), and Eating and Drinking Ability Classification System (EDACS); and convergent validity with the Barry Albright Dystonia Scale (BADS).

RESULTS 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% confidence interval [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<0.0001). Cronbach's alpha was 0.96.

INTERPRETATION The D-FIS demonstrates good construct validity and high internal consistency. The D-FIS will be useful for identifying priorities for intervention. It adds to the measurement toolkit for children with dyskinetic CP by addressing functional impact of dyskinetic movements and postures.

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Dyskinetic CP Functional Impact Scale *Kirsty Stewart et al.*

What this paper adds:

- Dyskinetic Cerebral Palsy Functional Impact Scale (D-FIS) assesses the perceived impact of dyskinesia on daily activities in children with cerebral palsy.
- The D-FIS demonstrates good construct validity and high internal consistency.
- The D-FIS is a clinically feasible, family centred tool that fills a current gap in the dyskinetic CP assessment toolkit.

[main text]

Dyskinetic cerebral palsy (CP), one of the most disabling forms of CP,[1,2] is a motor disorder characterized by changes in muscle tone and posture, with varying degrees of involuntary movement.[3] Dystonia and choreoathetosis are the two clinical manifestations of dyskinetic CP, although many children classified as dyskinetic CP also present with spasticity.[4,5] Standardized and accurate measurement of dyskinesia in people with CP is important to objectively quantify the movement disorder, monitor intervention outcomes, ensure practice is based on high quality evidence, and guide future interventions.

A number of scales have been developed to measure dystonia and choreoathetosis severity in children with dyskinetic CP.[6] The majority of scales assess dystonia severity only, taking into consideration its duration, provoking factor, and amplitude. The most recently published Dyskinesia Impairment Scale[7] is the only scale to address both dystonia and choreoathetosis in CP and the Barry Albright Dystonia Scale (BADS)[8] is the only other scale developed specifically for people with CP. All dyskinesia scales are intended as impairment, body function, and structure level assessments according to the International Classification of Functioning, Disability and Health,[9] with some providing limited insight into the impact of the dyskinesia or movement disorder on a small number of broad daily activities. In addition to severity, the importance of assessing the activity and participation domains of the International Classification of Functioning, Disability and Health, considering individual personal, cultural, and environmental factors, has been highlighted.[10–12]

Dyskinesia scales for people with CP can be used for two purposes: to provide a measure of severity at a single time point and to measure change after an

intervention targeting dyskinesia.[6] However, a reduction in dyskinesia severity does not necessarily translate to enhanced function or participation, or improvements in caregiver assistance and quality of life.[13] A clinical tool which will assist in identifying how changes in dyskinesia severity translates to changes in function, activity participation, caregiver assistance, and thus impact quality of life would be a useful adjunct to clinical and research practice.

A number of well-established, psychometrically robust outcome measures currently exist for use with children with CP and with disability generally.[14–21] When examining baseline function and measuring intervention outcomes of children with dyskinetic CP, multiple questionnaires and outcome tools are required to adequately cover all domains of the International Classification of Functioning, Disability and Health,[22] few of which have been developed specifically for children with dyskinetic CP and the unique functional limitations they may experience. This may result in assessment burden and fatigue for families, children, and clinicians. In addition, children with dyskinesia are typically seen in busy clinics where assessment needs to be feasible, focused on functional activities, and enables family priorities for intervention to be identified and measured efficiently and effectively. A single tool to specifically measure the impact of dyskinesia on daily function in children with dyskinetic CP is therefore considered a valuable addition to the existing measures.

The Dyskinetic Cerebral Palsy Functional Impact Scale (D-FIS) was developed to identify the impact of dyskinesia on daily functional activities for children and their caregivers and guide families in identifying daily activities which are their priorities for intervention.

The aim of this paper is to describe the development of the D-FIS and to report on a study evaluating its content and construct validity.

METHOD

Development of the D-FIS

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The need for a new tool to specifically quantify the impact of dyskinesia on daily function in children with CP was identified: (1) by parents of children with dyskinetic CP and children with dyskinetic CP during a qualitative study investigating the lived

experience of dyskinetic CP;[23] and (2) during an Australian Dyskinetic CP symposium in 2015 attended by occupational therapists, physiotherapists, speech pathologists, paediatric rehabilitation physicians, and paediatric neurologists (*n*=115).

Development of the D-FIS as an evaluative tool followed the methodological framework for assessing health indices by Kirshner and Guyatt.[24] The items for the pilot version, aimed at comprehensively covering usual daily activities for children, were generated from: (1) the extensive clinical experience of the research team; (2) recommendations obtained by interview, from parents and caregivers of children with dyskinetic CP, and children and adolescents with dyskinetic CP; and (3) a survey of 115 clinicians with interest and expertise in the management of children with dyskinetic CP attending the national Dyskinesia Symposium.[23] Daily activities and constructs deemed most important to families, children, and clinicians, and that were part of daily routine, formed the item pool. The item pool was further informed by current best evidence in the assessment of dyskinesia,[23] the results of a systematic review of currently available dyskinesia measures,[6] and literature in the areas of quality of life and caregiver burden,[15,20] family centred goal setting,[14] sleep,[18] pain,[16,21] and participation.[17,25]

A 16-item pilot version was subjected to review and feedback to develop and refine the D-FIS. A convenience sample of eight primary caregivers (six females, two males) of children with dyskinetic CP, recruited from an outpatient clinic of the Children's Hospital at Westmead, completed the pilot D-FIS. These primary caregivers reported for six children (mean age 14y 1mo [range 5y 7mo–18y 6mo, SD 4y 10mo], Gross Motor Function Classification System [GMFCS][26] levels: II=1, III=1, IV=3, V=1; Manual Ability Classification System [MACS][27] levels: II=1, III=2, IV=2, V=1; Communication Function Classification System [CFCS][28] levels: I=3, III=1, IV=2; and the Eating and Drinking Ability Classification System [EDACS] levels: I=3, III=1, V=2).[29] This convenience sample broadly represents CP classification levels and the general age range of children with dyskinetic CP for whom this tool is intended. Parents then participated in semi-structured face-to-face interviews to elicit views on the D-FIS and identify revisions. The pilot version of the D-FIS was also emailed to a purposive sample of eight Australian clinicians with specific expertise in the management of children with dyskinetic CP and/or experience in outcome

measurement development to elicit written feedback on the items, wording, scales, and coverage of the D-FIS. Clinicians included paediatric rehabilitation physicians, a CP sleep disturbance expert, occupational, physical, and speech therapists, and a paediatric neurologist. These combined processes resulted in two additional items, inclusion of the Priority Scale, refined content and language of all items and scales, and enhanced usefulness of the questionnaire. The final 18-item D-FIS is under exploration in this study.

All primary caregiver participants recruited for the study were provided with the printed questionnaire and detailed instructions. The front page of the D-FIS explains dyskinesia as it is seen in children with CP. The questionnaire was accompanied, in all cases, by a discussion regarding: (1) their child's specific movement disorders; (2) how to identify dyskinetic movements and postures in their child; and (3) that dyskinetic movements and postures may be only one reason why their child may have difficulty with daily activities. We also reminded families that there may be alternative reasons for a functional difficulty – each item has a 'not applicable' option with the following wording: 'Standing (depending on the item) is difficult but not due to dyskinesia'.

Development of the D-FIS progressed in stages and was approved by the Royal Children's Hospital (RCH HREC: 36129A) and the Sydney Children's Hospitals Network (LNR/18/SCHN/32) Human Research Ethics Committees.

The D-FIS

The final D-FIS consists of 18 items, 16 daily activities and two additional constructs that impact children with dyskinetic CP (pain and fatigue). It takes approximately 10 minutes to complete via parent proxy report (Appendix S1, online supporting information). The D-FIS items are rated on two scales: an Impact Scale and a Priority Scale. Each item is a separate functional activity: Sitting; Standing; Walking; Positioning; Transfers; Other Gross Motor Activities; Fine Motor Activities; Reaching; Use of Technology; Daily Hygiene Activities; Upper Body Dressing; Lower Body Dressing; Feeding; Speech; Sleep; and Leisure Activities. Pain and Fatigue make up the two additional constructs. The Impact Scale rates the impact dyskinesia has on each activity/construct on a 5-point ordinal scale from 0 (no impact), 1 (mild impact), 2 (moderate impact), 3 (severe impact), to 4 (extreme impact). The Impact Scale

ratings also account for functioning in each activity without the assistance of equipment and/or a caregiver and function with the assistance of equipment and/or caregiver. Each activity is briefly described in the tool and examples are provided that span developmental levels. These examples are not intended to be all-inclusive but rather to give parents an understanding of what the activity may cover. The Impact Scale scores are summed, the total score ranges from 0 to 72, and higher scores indicate greater impact of dyskinesia on daily functional activities. The Priority Scale identifies the current priority of each activity for the child and their family from 1 (not a priority) to 4 (highest priority). The purpose of the Priority Scale is to assist families and clinicians with goal identification and ensuring the activity areas most important to children and their families are the focus of intervention.

Construct validity of the D-FIS

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Participants

A convenience sample of primary caregivers (*n*=57) were recruited from outpatients attending the Kids Rehab Department of the Children's Hospital at Westmead between October 2018 and November 2019. Eligible caregivers were those with children aged 2 to 18 years with dyskinetic CP, or dystonia/choreoathetosis and spasticity where dystonia or choreoathetosis was the predominant motor type. Predominance of motor type was determined clinically using the Cerebral Palsy Description Form.[30] Forty-nine (86%) of the 57 primary caregivers were mothers or female carers. Their children were aged between 2 years 6 months and 18 years (mean age 11y 7mo, SD 4y 4mo). Functional classification levels, GMFCS, MACS, CFCS, and EDACS, were assigned for all participants and comorbidities recorded (Table 1). Informed, written consent was obtained from all participating caregivers.

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Construct validity, the degree to which the D-FIS measures the intended construct, was evaluated using known groups methods based on correlation of the D-FIS with the functional levels of the GMFCS, MACS, CFCS, and EDACS, and differences in D-FIS scores across levels within each of these four systems. Each system classifies children across five levels, with level I indicating minimal disability and a high level of independence and level V indicating total dependence on equipment and carers for all daily needs. Differences in D-FIS total scores between ambulant and non-ambulant children according to the GMFCS (ambulant=levels I, II

and III; non-ambulant=levels IV and V) were compared. The hypotheses were that: (1) children in higher GMFCS/MACS/CFCS/EDACS levels (children with lower function) would have higher D-FIS Impact Scale total scores (lower ability in daily functional activities); (2) D-FIS would be strongly positively correlated with the GMFCS and MACS; (3) D-FIS would be moderately correlated with the CFCS and EDACS; and (4) ambulant children would have significantly lower D-FIS scores than non-ambulant children.

Construct convergent validity,[31] the extent to which the D-FIS produces similar results as another well-established tool that measures a related construct (i.e. dystonia severity), was assessed against the BADS. The BADS was selected as it is a clinically useful scale, developed specifically for people with CP, has demonstrated responsiveness to change after interventions, and is used extensively in Australia.[32] The BADS takes approximately 10 minutes to complete which minimizes child and family assessment burden. The BADS scores dystonia on a 5-point criterion based, ordinal severity scale (0–4) across eight body regions. Scores for each body region are summed to a maximum of 32, with higher scores indicating increasing severity of dystonia. The BADS was completed with every child by two of the researchers (KS, JL) who have extensive experience rating dyskinesia in children with CP. The hypothesis was that children with higher BADS scores would have higher D-FIS Impact Scale total scores, and these would be strongly positively correlated.

Data analysis

Spearman's rank correlation assessed the correlation between total D-FIS Impact Scale scores and GMFCS, MACS, CFCS, EDACS, and BADS. Confidence intervals (CIs) for Spearman's rank correlation were calculated, based on Fisher's transformation. Correlation coefficient of $r \geq 0.7$ was considered strong and 0.5 to <0.7 was considered moderate.[33] One-way analysis of variance was used to analyse differences in total D-FIS scores across levels of the classification systems and the difference in D-FIS scores between ambulant and non-ambulant was examined using an independent *t*-test. The D-FIS total score was normally distributed as shown by histograms and box plots and confirmed by the Shapiro–Wilk test and a Normal Q–Q plot. Homogeneity of the variances was demonstrated using Levene's

test. A two-tailed significance level of 0.05 was used in all tests. All data were analysed using Stata version 16.1 (StataCorp, College Station, TX, USA).

Ratings on the Priority Scale, intended to focus clinical intervention on those activities rated most important to families, were reported using descriptive statistics. Priority Scale mean scores are presented for ambulant and non-ambulant children.

RESULTS

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The D-FIS was completed by 57 caregivers for 57 children (see Table 1 for characteristics of the children). Children were distributed across the functional classification levels except MACS level I – a finding similar to other dyskinetic CP cohort studies.[5,34]

Mean total D-FIS Impact Scale scores by classification levels are reported in Table 2. The mean total score for the whole cohort (n=57) on the D-FIS Impact Scale was 40.44 (SD 16.72, range 8–72). Increasing functional severity levels on the GMFCS, MACS, CFCS, and EDACS demonstrated increased mean D-FIS scores (p<0.001). Spearman's rank correlation for D-FIS Impact Scale total score and GMFCS was r=0.86 (95% confidence interval [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, p<0.001), indicating strong relationships with the D-FIS. Mean total D-FIS Impact Scale scores between ambulant and non-ambulant children were significantly different (respectively n=22, 23.6 [SD 9.8]; n=35, 51.0 [SD 10.1]; p<0.001). The correlation between D-FIS Impact Scale total scores and BADS was r=0.77 (95% CI: 0.64–0.86, p<0.001), indicating a strong relationship. The D-FIS demonstrated good internal consistency (α=0.96).

The mean Priority Scale score for the ambulant, non-ambulant, and total cohorts are presented in Figure 1. In the ambulant cohort, the highest priority activity, identified by parents for their children, was fine motor activities (mean 3.5) followed by leisure, daily hygiene, gross motor, and walking (means 2.7–3). The least prioritized activities were sleep, sitting, and positioning (total means of 1.6–1.8). In the non-ambulant cohort, the activities rated the highest priority were transfers (mean 3.65), positioning, technology use, dressing, and daily hygiene activities

(means 3.3–3.6). The least important activity rated by parents was gross motor activities (mean 2.3), walking, speech, and fatigue (total means of 2.5–2.8).

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DISCUSSION

This study reports the development and initial psychometric properties of the D-FIS, a newly developed tool to identify the impact of dyskinesia on daily activities in children with CP. The D-FIS was intended to be clinically feasible to implement within the confines of specialty CP clinics in busy children's and rehabilitation services. The D-FIS demonstrates good content and construct validity and strong internal consistency in children aged 2 years 6 months to 18 years. Dyskinesia is a complex and disabling movement disorder in people with CP. Tools to measure the severity of the movement disorder have been available for decades, yet no single tool assesses the impact of dyskinesia on the daily functional activities of children with CP nor measures the impact of targeted interventions on those functional tasks, either from a parent/caregiver's or child's perspective. The D-FIS aims to fill this current gap in measurement.

The D-FIS consists of 16 daily functional activities and two additional constructs, pain and fatigue. These additional items, although not functional activities, are impacted by dyskinesia[11,21,35] and have been demonstrated to impact a child's participation across all daily activities. Despite appearing as an additional construct, results indicate they did not impact the internal consistency of the D-FIS.

The D-FIS is able to distinguish between ambulant and non-ambulant children, with a clear gradient of mean total scores across GMFCS and MACS levels. These findings confirm our hypotheses and support the construct validity of the D-FIS. The findings also support the validity of the D-FIS in a heterogenous sample of children in relation to age and severity of presentation. Although analysis of variance identified differences between levels for CFCS and EDACS, small participant numbers in some levels on these classifications mitigated against observing a gradient of increasing scores. Considered together with the strong correlations between D-FIS total scores and each of the functional classification systems, there is good evidence of construct validity. This supports our initial hypothesis that children with CP with more severe dyskinesia demonstrate poorer

function across the activities they complete on a daily basis and require greater physical support from caregivers.

The Priority Scale results demonstrate that activities that require the most physical assistance during care, such as positioning, daily hygiene, and dressing, were amongst the highest priority for the parents of non-ambulant children. Access to technology was also rated as a high priority for this group of children, reflecting the possible reliance on technology for both school-based academic tasks, communication, and as a quiet leisure option. Parents of ambulant children indicated that fine motor activities were the highest priority for their children which is consistent with the knowledge that dyskinesia frequently affects the upper limbs more than the lower limbs in more ambulant children with CP and in particular children with unilateral involvement.[5] We expected that pain, sleep, and fatigue may feature more frequently as a priority for families, given the increasing focus on these factors in the literature and their impact on quality of life.[11] As the D-FIS is a parent proxy questionnaire, the priority scale reflects the parent's perceptions of their child's experiences, particularly in regards to pain, fatigue, and to some extent sleep, and may not be viewed as a higher priority unless impacting family life. As expected, however, they were reported more frequently as a priority in non-ambulant children. Children themselves may rate these factors as a high priority and these factors may become more prominent with increasing age.

There are factors, other than dyskinesia, which may impact the domains of the D-FIS. Parents were found to be very good at understanding their child's difficulties and the reasons why, for example, due to their abnormal movements, their intellectual disability, or low tone making head control difficult hence access to an eye gaze system problematic. We ask respondents for their perception of functional ability attributable to dyskinesia. We do not ask families to discriminate dystonia or choreoathetosis. We have not detected any uncertainty by families in the D-FIS scales which have now been completed as part of the development and validation of the tool. Ultimately, this is a parent proxy tool, which reflects parent perception of their child's ability framed within the context provided by the D-FIS.

The D-FIS authors have found the questionnaire to be responsive and clinically useful in children aged over 3 years. By the age of 3 years, typically

developing children are independent in most D-FIS daily functional activities with emerging independent dressing skills and daily hygiene skills, ensuring that the descriptive scale levels for each activity rate the impact of dyskinesia on that activity rather than risk rating developmental ability.

This study was limited by sample size as recruitment was reliant on clinical availability of families with children with dyskinetic CP attending outpatient clinics. Test–retest reliability is ongoing and, whilst important, is more difficult to determine in a condition such as dyskinetic CP because of the changing nature of the movement disorder and the daily influences of health, environment, mood, and sleep on dyskinesia. At this stage, the D-FIS remains a parent proxy questionnaire. A self-report version is under development.

The D-FIS is valuable as a valid and time-efficient means to identify the impact of dyskinesia on function and to understand family priorities for intervention. Further research is underway to evaluate its responsiveness to change after intrathecal baclofen therapy, deep brain stimulation therapy, and medication trials.

The D-FIS is a newly developed scale that measures the impact of dyskinesia on typical daily functional activities of children with dyskinetic CP. This parent proxy version demonstrates strong validity. Further research is ongoing to determine test–retest reliability and responsiveness to change which will inform use of the D-FIS as an outcome measure. A self-report version will be developed with guidance from children with dyskinetic CP and will provide a more complete picture of the child's function and priorities.

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The authors declare no conflicts of interest with respect to the research, authorship, or publication of this article.

Supporting information

The following additional material may be found online:

Appendix S1: Dyskinetic Cerebral Palsy Functional Impact Scale.

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[Figure legend]

Figure 1: Parent report activity priority for ambulant, non-ambulant, and entire cohort

Table 1: Participant demographics and classification levels of their child with cerebral palsy (CP)

Demographics

Age (y:mo)		
	Range	2:6–18:0
	Mean (SD)	11:7 (4:4)
Sex		
	Male	29 (51)
	Female	28 (49)
Distribution		
	Bilateral CP	46 (80)
	Unilateral CP	11 (20)
GMFCS level		
	I	8 (14)
	II	6 (11)
	III	7 (12)
	IV	23 (40)
	V	13 (23)
MACS level		
	I	0
	II	19 (33)
	III	12 (21)
	IV	12 (21)
	V	14 (25)
CFCS level		
	I	16 (28)
	II	13 (23)
	III	14 (25)
	IV	10 (17)
	V	4 (7)
EDACS level		
	I	16 (28)
	II	19 (33)
	III	11 (19)
	IV	1 (2)
	V	10 (17)
Comorbidities		
	Nil	13 (23)
	Epilepsy	16 (28)

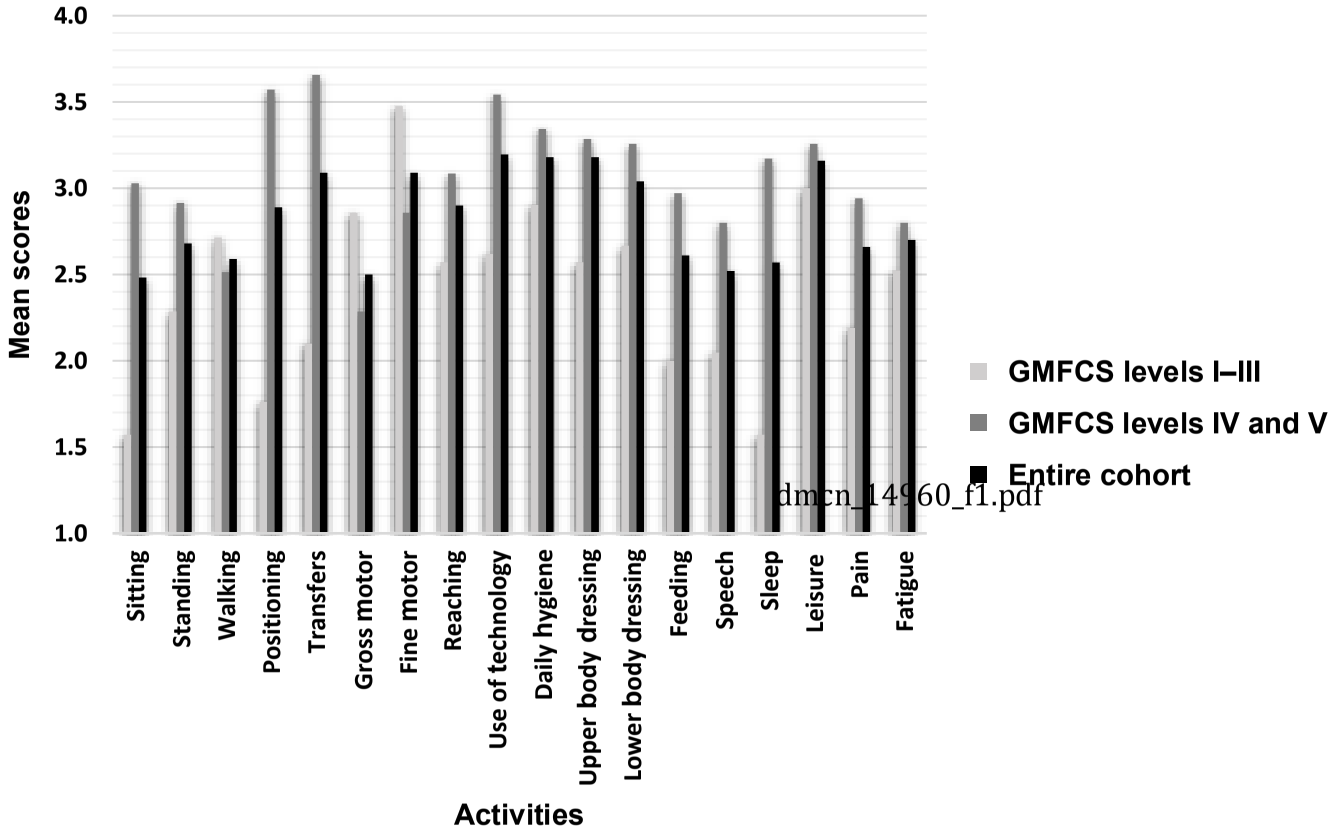
Gastrostomy	9 (16)
Visual difficulties	16 (28)
Hearing impairment	4 (7)
Intellectual disability	23 (40)

Data are *n* (%) unless otherwise stated. GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System; EDACS, Eating and Drinking Ability Classification System.

Table 2: Mean total scores for the D-FIS Impact Scale by classification levels

Level[a]	Total D-FIS Impact Scale scores[b] for each classification system			
	GMFCS	MACS	CFCS	EDACS
I	17.2 (6.2, 8–27)	0	22.4 (10.8, 8–43)	23.1 (11.2, 8–46)
II	22.4 (7.7, 11–39)	22.5 (9.9, 8–47)	36.5 (10.1, 20–50)	38.8 (11.4, 20–61)
III	32.0 (9.8, 19–49)	38.3 (6.8, 31–47)	47.3 (9.5, 30–61)	49.7 (8.3, 30–58)
IV	46.6 (7.4, 31–61)	52.9 (8.3, 38–69)	59.1 (8.6, 44–72)	*n*=1 child, 69
V	58.5 (9.7, 38–72)	55.9 (9.7, 38–72)	54.5 (12.9, 38–69)	58.3 (10, 38–72)
ANOVA	$F(4,52)=45.4$ $p<0.001$	$F(3,53)=46.6$ $p<0.001$	$F(4,52)=25.1$ $p<0.001$	$F(4,52)=22.0$ $p<0.001$

Data are mean (SD, range) unless otherwise stated. [a]Level I represents higher function, level V represents lower function. [b]Higher D-FIS scores indicate lower function. GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System; EDACS, Eating and Drinking Ability Classification System; ANOVA, analysis of variance.



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