Identification and measurement of dystonia in cerebral palsy

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ABBREVIATIONS

BAD	Barry-Albright Dystonia scale
CFCS	Communication Functional Classification System
HAT	Hypertonia Assessment Tool
MACS	Manual Ability Classification System
PMT	Predominant motor type
SCPE	Surveillance of Cerebral Palsy in Europe

AIM To establish the prevalence and severity of dystonia in a population of children with cerebral palsy (CP) with hypertonia assessment and measurement tools.
METHOD A cross-sectional study of 151 children (84 males, 67 females) with CP who were assessed with the Hypertonia Assessment Tool (HAT) and Barry-Albright Dystonia scale (BAD) for identification and measurement of severity of dystonia. HAT dystonia items were assessed for construct and convergent validity.
RESULTS Distribution by predominant motor type (PMT) was: 85% spastic, 14% dyskinetic, and 1% ataxic. Spastic and dyskinetic groups showed widespread evidence of dystonia according to HAT profiles and BAD scores. The dyskinetic PMT group had a higher mean BAD score than the spastic group (difference of 13 units, 95% CI 9.1–16.4). Dystonia severity (BAD score) increased linearly across gross motor ($p<0.001$), manual ability ($p<0.001$) and communication functional levels ($p<0.001$). Divergence was noted in how HAT item six identified dystonia compared to items one and two.
INTERPRETATION The HAT provided an estimate of the prevalence of both spasticity and dystonia in a large CP population, beyond predominant motor type. Dystonia is a common finding in the spastic PMT group, and its severity increases as motor function worsens.

Classification of abnormal tone and movement patterns is critically important in the evaluation of children with cerebral palsy (CP). When classified according to the predominant motor type (PMT), spasticity accounts for up to 80% to 95% of cases in CP populations; dyskinesia (dystonia and/or chorea and athetosis) is observed in 4% to 17%; while ataxia accounts for up to 5% and hypotonia 2%.[1,2] Spasticity occupies a large part of the complex picture of motor impairment and functional limitation[3,4] while dystonia, defined as 'a movement disorder in which involuntary sustained or intermittent muscle contractions cause twisting and repetitive movements, abnormal postures, or both', is less well understood in terms of its impact.[5,6] When the term 'hypertonia' is used in CP it generally reflects the observation that spasticity and dystonia may be present in isolation or in combination, without necessarily defining its components.[5]

In the assessment of hypertonia in CP, spasticity can be elicited by clinical examination by performing passive range of joint motion at varying speeds, in order to identify any velocity-dependent increase in a muscle stretch reflex.[7] A small number of measures provide a practical and reliable method for both identifying and quantifying spasticity.[8,9] Identification and measurement of dystonia provide methodological challenges as by its nature dystonia exhibits variability in presentation, even over a short period, compounded by the influence of external stimuli and emotional state.[10] In addition, limb hypertonia noted during joint range of motion may change during the course of the examination, which only adds to the challenge of classification.[11,12]

Classification systems in CP typically regard spasticity and dystonia as being mutually exclusive findings, when in reality they appear to frequently coexist.[13] When classifying the predominant motor type (PMT) using the Surveillance of Cerebral Palsy in Europe (SCPE) classification tool, once spasticity is readily recognized on the basis of increased tone and pathological reflexes, the decision-making process is ordered such that a child is likely to be classified as having spastic CP.[14,15] Registers may rely on identification of the predominant motor type using consensus-based guidelines rather than standardized assessment tools; this may lead to differences between assessors on what constitutes the 'predominant' pattern.[16] The inclusion of a secondary hypertonia component has been suggested as a means of avoiding this dilemma.[17]

When dystonia is recorded as the PMT, its severity may be such that any features of coexistent spasticity are overlooked. Improving the recognition of dystonia and

situations where spasticity coexists is important not only for promoting a clearer description of tone and movement abnormalities, but also to help tailor appropriate treatments leading to improved outcomes, and recognizing that a 'one-size-fits-all' approach of spasticity management in CP may be ineffective or even harmful in treatment outcomes in CP.[3]

In a previous related study which described the motor profiles of a geographic population of 5-year-old children with CP, 93.2% of the population were coded by PMT as spastic; 3.2% dyskinetic and 3.6% ataxic.[18] When these children were directly examined, 19.4% of the population were noted to have abnormal movements including features of dyskinesia. Abnormal movement frequency increased by Gross Motor Function Classification System (GMFCS) grouping from 7% (level I) to 45% (level V). We questioned whether the recognition of dystonia was at times 'masked' by the fact that most of these children had already been categorized as having spastic PMT.

To date, no study has systematically examined for the concurrent presence of types of abnormal tone and movements in CP populations, beyond determining the predominant motor type in a mutually exclusive fashion, for example spasticity or dystonia. The primary objective of this study was to determine the prevalence of dystonia in a defined CP population using the Hypertonia Assessment Tool (HAT), with the hypothesis that this would be higher than that determined by the PMT. The secondary objectives were to measure the frequency of coexistent spasticity and dystonia in this population, and to assess for associations between the severity of dystonia and motor function.

What this paper adds

- Dystonia is readily identified in cerebral palsy (CP) using the Hypertonia Assessment Tool, regardless of the predominant motor type.
- Spasticity and dystonia frequently coexist in the CP population.
- Severity of dystonia is inversely related to motor function.

METHOD

Setting

This study was undertaken at the Women's and Children's Hospital in Adelaide, Australia. The study was approved by the Human Research Ethics Committee of the Women's and Children's Health Network.

Data collection

All families of children with CP attending paediatric rehabilitation clinics at the Women's and Children's Hospital between 2011 and 2014 were approached with regard to participation in the study. Inclusion criteria included a confirmed diagnosis of CP and age between 2 years and 18 years. Exclusion criteria were the presence of any progressive neurological disorder and children whose age was outside of this range. After informed consent, study participants underwent a comprehensive assessment performed by research physiotherapists. Key demographic data and associated medical history were recorded, including verification of inclusion on the state-based CP register, which captures key data on all children with CP within the state of South Australia.

Hypertonia assessment

The clinical assessment included: (1) determination of predominant motor type (PMT) by application of the SCPE classification method; (2) differentiation of hypertonia by application of the HAT; and (3) measurement of dystonia using the Barry-Albright Dystonia (BAD) scale with video recording. Motor abilities were classified according to the GMFCS, Manual Ability Classification System (MACS); and Communication Functional Classification System (CFCS).

Assessment tools

The SCPE classification tool utilizes a decision-making tree process to determine the predominant motor type of CP.[19] The HAT is a seven-item standardized clinical assessment tool used to differentiate the various types of paediatric hypertonia. The tool contains two spasticity items, two rigidity items, and three dystonia items. A positive score for at least one item of the subgroup confirms the presence of the subtype of hypertonia (spasticity, dystonia, rigidity, or a mixed pattern) in the limb examined.[20,21] Although the HAT is designated for use in children over four years of age, it was applied across the whole population in this study. (The HAT user manual and online tutorial can be accessed at https://research.hollandbloorview.ca/outcomemeasures/hat.) The BAD scale is a five-point, criterion-based ordinal scale for measuring dystonia in CP, with reported validity and reliability (Appendix S1, online supporting information).[22,23] It assesses dystonia in eight body regions. Severity is scored from none to severe, with each body region having specific descriptors for scoring.

Recording abnormal movements

The assessment protocol for the BAD scale was followed and the child was observed in a number of standardized positions using video recording for the purposes of scoring. Positioning was made in supine as per HAT user protocol with videorecording to ensure that all limbs could be observed. Videos were then reviewed by two members of the three-person research physiotherapist team which had over 30 years of combined clinical experience in working with children with neuromotor disabilities. Each assessor reviewed the child's video separately with use of a third reviewer in situations where consensus could not be achieved.

Data analysis

For estimates of proportions, Wilson's 95% confidence intervals were calculated. For investigation of categorical variables, Fisher's exact test was used with standardized effect size of Cramer's V. Comparisons between groups with interval data were undertaken with one-way ANOVA with bootstrapped confidence intervals. Fleiss kappa was used to assess agreement among nominal variables. Polyserial correlations were reported when investigating

associations between interval and ordinal variables. Factorial validity assessment of ordinal outcomes was undertaken with weighted least squares mean and variance adjusted (WLSMV) estimator using MPlus software (MPlus Base Program and Combination Add-On (Version 7.4; Los Angeles, CA: Muthén & Muthén). Statistical analyses were performed in SPSS version 22.0 (SPSS Statistics for Windows, version 22.0; IBM Corp., Armonk, NY, USA). A significance level of 0.05 was used throughout and 95% confidence intervals were reported.

RESULTS

Demographics

One hundred and fifty-one children were recruited into the study from a total of 304 families approached during the study period. The mean age at assessment was 8.8 years (range 2–18y) with 84 males and 67 females (M:F 1.3:1). Most (95%) of the study participants were listed on the state CP register; the remaining 5% resided out of state or had not consented to register enrolment. Sampling bias was investigated as there was information available regarding sex, GMFCS, and PMT classification for non-participants. There was no difference between participants and no-participants for sex (p=0.49, Cramer's V=0.044, 95% CI 0.002–0.16) or GMFCS (p=0.67, Cramer's V=0.09, 95% CI 0.06–0.24). A difference in PMT patterns between the groups was noted (p=0.008, Cramer's V=0.16, 95% CI 0.08–0.27).

Prevalence of dystonia – SCPE classification

According to the SCPE classification for PMT 85.4% of the population were spastic (95% CI 78.9–90.0), 13.9% were dyskinetic (95% CI 9.2–20.0), and 0.7% ataxic (95% CI 0–3.6). No cases with hypotonia were identified. The characteristics of the spastic and dyskinetic groups are compared in Table I. All participants labelled as dyskinetic had features of dystonia on HAT and BAD scores.

Prevalence of dystonia – HAT

Of 604 studied limbs in 151 participants, 540 (89%) were allocated a HAT score. Reasons for lack of score allocation in 11% (n=64) included pain or skin integrity issues limiting the passive range of joint motion and handling, behavioural difficulties, and limited ability to follow commands to complete voluntary tasks, either in young children or those with limited motor ability. Figure 1 highlights the population distribution of HAT profiles for all four limbs. Distinct proportional differences were noted when comparing upper and lower limbs. Agreement among all four limb assessments was very low (k=0.12, 95% CI 0.08–0.17). Although there was little difference in overall proportions between left and right limbs within upper or lower body segments, the agreement statistics of relevant pairs of HAT assessments suggest that a single upper limb HAT evaluation may not be sufficient.

Table II describes the prevalence of dystonia by HAT score between spastic and dyskinetic groups, noting the frequency of dystonia findings in the spastic PMT group, identified either as mixed tone (spasticity and dystonia) or all dystonia. During the early phase of assessment in this CP population, it became apparent that none of the children had a positive score on the HAT rigidity items (5 and 7). As a result these items were not included in subsequent analysis.

Table I: Comparison between spastic and dyskinetic predominant motor type groups

Characteristic[a]	Spastic n=129	Dyskinetic n=21	p value	Effect size (95% CI)
Mean age at assessment, y (SD)[b]	8.8 (5.1)	8.8 (5.1)	0.947	0.08 (−2.2–2.4)
Sex,[c] n (%)				
Males	70 (54)	14 (67)	0.346	0.09 (0.01–0.23)
Females	59 (46)	7 (33)		
Gestation,[c] n (%)				
<37wks	65 (50)	3 (14)	0.002	0.25 (0.1–0.39)
>37wks	64 (50)	18 (86)		
GMFCS level,[c] n (%)				
I	35 (27)	3 (14)	0.03	0.28 (0.15–0.49)
II	36 (28)	3 (14)		
III	24 (19)	2 (10)		
IV	22 (17)	7 (33)		
V	12 (9)	6 (29)		
MACS level,[c] n (%)				
1	41 (32)	0 (0)	<0.001	0.42 (0.31–0.6)
2	40 (31)	2 (10)		
3	23 (18)	6 (29)		
4	12 (9)	9 (43)		
5	13 (10)	4 (19)		
CFCS level,[c] n (%)				
1	69 (54)	2 (10)	<0.001	0.34 (0.24–0.52)
2	19 (15)	3 (14)		
3	12 (9)	6 (29)		
4	17 (13)	6 (29)		
5	12 (9)	4 (19)		

[a]Characteristics of the one child who was ataxic is not described above; [b]independent samples t-test/effect size is age difference; [c]Fisher's exact test significance/standardized effect size is Cramer's V. GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Functional Classification System.

Additional analysis was performed to evaluate measurement properties of the dystonia HAT items, in regards to construct (exploratory factor analysis) and convergence validity (polyserial correlations with BAD scores) as shown in Table III. Factor analysis indicated a lack of unidimensionality of the HAT dystonia scale, reflecting the divergent nature of item 6 compared with the other two items. Item 6 also showed least association with the BAD scale (correlations ranging from 0.01–0.38) while correlations for item 2 exceeded 0.6. Between 19% and 34% of cases were identified as dystonic exclusively on the basis of HAT item 6.

Severity of dystonia

All participants were allocated a BAD score. There was a statistically significant difference between spastic and dyskinetic subgroups with a difference of 13 BAD units (95% CI 9.1–16.4) with the spastic group having a mean BAD

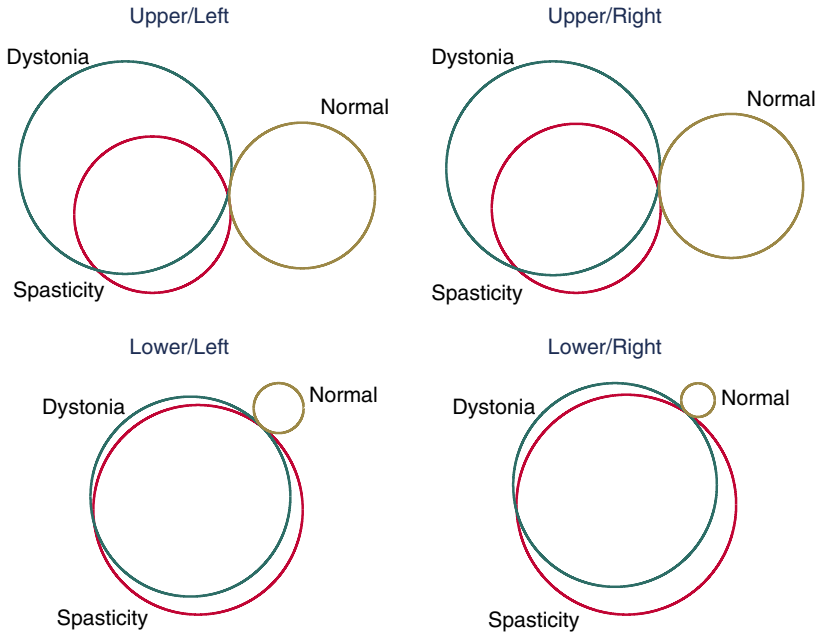


Figure 1: Venn diagrams representing size and relationship of Hypertonia Assessment Tool profiles across population for all four limbs. N.B. overlap between spasticity and dystonia equates to mixed tone. [Colour figure can be viewed at wileyonlinelibrary.com].

Table II: Prevalence of dystonia across study population by Hypertonia Assessment Tool (HAT) score for each limb according to motor type

		Predominant motor type	
HAT score		Spastic	Dyskinetic
Mixed tone (spasticity and dystonia) % (95% CI)	Upper limbs		
	Left	25.7 (18.5–34.4)	50.0 (26.0–74.0)
	Right	32.8 (24.3–42.1)	55.6 (30.8–78.5)
	Lower limbs		
	Left	75.9 (67.0–83.3)	100 (81.5–100)
	Right	72.0 (63.0–79.9)	89.5 (66.9–98.7)
All dystonia (pure dystonia and mixed tone) % (95% CI)	Upper limbs		
	Left	59.3 (50.1–67.9)	94.4 (72.7–99.9)
	Right	62.1 (52.6–70.9)	88.9 (65.3–98.6)
	Lower limbs		
	Left	80.2 (71.7–87.0)	100 (81.5–100)
	Right	76.3 (67.8–83.0)	100 (82.4–100)

score of 4.8 (95% CI 4.0–5.6) compared with dyskinetic group score of 17.8 (95% CI 13.9–21.1) Dystonia severity according to BAD score increased linearly across gross motor (Polynomial contrast $F_{4,145}$=8.2, p<0.001), manual ability (Polynomial contrast $F_{4,145}$=12, p<0.001) and communication functional levels (Polynomial contrast $F_{4,145}$=10.7, p<0.001). Figure 2 shows mean BAD scores with bootstrapped 95% CIs for GMFCS, MACS, and CFCS domains respectively.

DISCUSSION

In this population the headline frequencies of spasticity and dyskinesia according to PMT were similar to those identified in other population-based studies.[1,2,24] Across the population the degree of dystonia increased as motor function worsened, regardless of PMT. Dystonia was readily identified and measured using the HAT and BAD scales, and occurred much more frequently than suggested by the PMT frequency of dystonia. Spasticity and dystonia frequently coexisted, regardless of motor type, with a marked difference noted in the amount of measured dystonia between the spastic and dyskinetic PMT groups. The dyskinetic group had a higher proportion of children with lower gross and fine motor and communication abilities, consistent with other study findings.[25]

To our knowledge, the prevalence of spasticity and dystonia as concurrent hypertonia components has not previously been recorded in population-based studies. The implications of the PMT classification principle that children have either a spastic or dyskinetic motor pattern can be altered by defining hypertonia in this manner. This has clear benefits for treating the motor aspects of CP, as treatments can be tailored and methods utilized which are more likely to address specific tone components. For example, the child with spastic diplegia who also has dystonia and undergoes a selective dorsal rhizotomy will likely experience reduction in spasticity; however, the associated dystonia will not be affected and may continue to impact on function. In the surgical management of the upper limb with wrist flexion deformity, tendon transfer surgery may not be successful if overactivity of antagonistic muscles, in part reflective of dystonia, is not recognized in advance.

Rigidity was not identified in any participant in this study. A finding of true rigidity is rare in paediatric neurological disorders, and is distinct from the frequent description in CP of a joint that has severe limitation of

Table III: Polyserial correlation (with Barry-Albright Dystonia scale scores) and exploratory factor analysis for Hypertonia Assess Tool (HAT) dystonia items

HAT ITEM	Polyserial correlation (95% CI)	*p* value	Exploratory factor analysis[a]		
			Factor 1	Factor 2	Factor 3
L lower limb 6	0.01 (−0.20 to 0.22)	0.904	−0.2	0.0	**0.9**
L upper limb 6	0.38 (0.19–0.55)	0.000	0.4	−0.1	**0.6**
R lower limb 6	0.07 (−0.14 to 0.27)	0.539	−0.2	0.1	**0.9**
R upper limb 6	0.33 (0.13–0.50)	0.002	0.2	0.3	**0.6**
L lower limb 2	0.67 (0.49–0.79)	0.000	0.0	**0.9**	0.1
L upper limb 2	0.66 (0.50–0.77)	0.000	**0.8**	0.1	0.2
R lower limb 2	0.61 (0.42–0.75)	0.000	0.0	**0.9**	0.2
R upper limb 2	0.68 (0.54–0.78)	0.000	**0.8**	0.0	0.3
L lower limb 1	0.41 (0.20–0.59)	0.000	0.0	**0.9**	−0.5
L upper limb 1	0.48 (0.26–0.65)	0.000	**0.8**	0.2	0.0
R lower limb 1	0.45 (0.24–0.62)	0.000	0.3	**0.7**	0.0
R upper limb 1	0.54 (0.35–0.69)	0.000	**0.8**	0.3	0.0

Polyserial correlation: Items 1 and 2 consistently correlated with related limb Barry-Albright Dystonia scale scores, whereas item 6 did not. Exploratory factor analysis: The meaning of the individual factors is inferred from individual items with high loadings. Item results in bold point to an affiliation with a specific factor; in the case of factor 1, items 1 and 2 (left and right upper limbs) showed strong affiliation. [a]Fit statistics: chi-square=66.7, $p<0.001$, Ratio of chi-square to df=2, CFI=0.97, TLI=0.95 SRMR=0.12. While a four-factor solution produced a better fitting model, it reported factor loadings exceeding 1 and more cross-loading items. A three-factor model was retained.

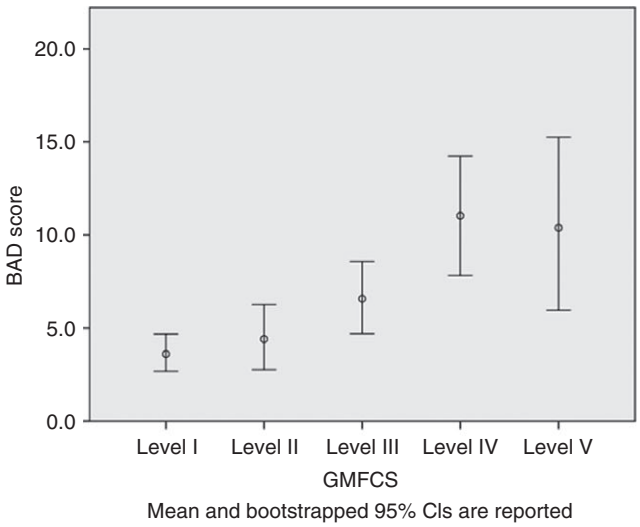
movement due to co-contraction, contracture, or other form of resistance.[5] When the HAT was devised it was intended for use in a range of paediatric conditions associated with hypertonia. Our findings suggest that it is reasonable to remove items 5 and 7 from the HAT when applying this to children with CP.

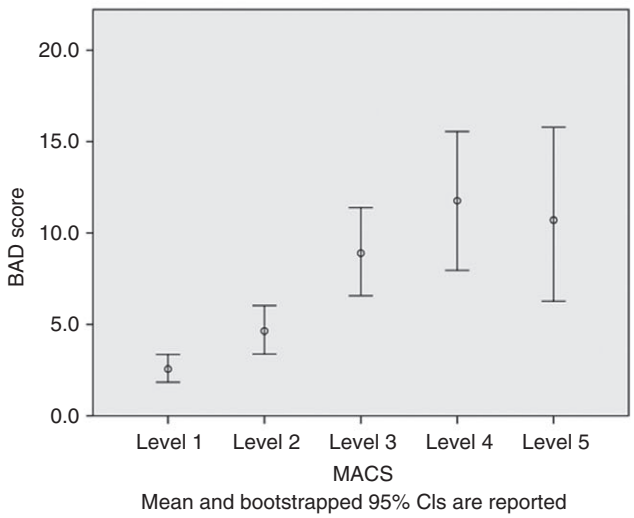
The HAT identified dystonia frequently, often in combination with spasticity, but did not in itself indicate the severity of dystonia or its contribution to function. It acts as an impairment-level or body function-level tool which should be used in combination with other body function-level and activity-level measurement tools to gain an indication of the impact of spasticity and dystonia. While it may appear surprising that dystonia was identified so frequently across the population by the HAT, it should be recognized that HAT items 1 and 2 utilize tactile stimulus and purposeful movement where sensory stimuli and intentional movement are likely to induce dystonic movements and postures; on the other hand, the BAD scale makes use of participant observation without direct interaction.

The relationship between BAD scores and GMFCS, MACS, and CFCS levels as shown in Figure 2 provides perspective on the relationship between degree of dystonia and motor function, and suggests that dystonia plays a greater negative influence on function across the CP population than previously recognized. Although for many children with spastic CP the co-occurrence of dystonia (with a low BAD score) may not overtly impact on motor function, what has not been resolved from this study is whether dystonia may influence motor function within specific subgroups of spastic CP, such as ambulant children with predominant lower limb hypertonia. We did not measure negative motor signs associated with spasticity, such as weakness and poor selective motor control; it is thus possible that other factors associated with hypertonia may account for some of this change in motor function.

Experience with the use of the HAT in CP populations is limited.[20,21] We were interested in how the individual items behaved in relation to each other according to exploratory factor analysis, as a means of further assessing the validity of the HAT. A divergence was noted in how items 1 and 2 and item 6 worked in dystonia identification. One explanation is that item 6 identified a different aspect of dystonia by way of palpation during active limb movement, which can introduce a sensory stimulus in addition to the intentional movement, as well as identify increased tone not observed through the other items. The higher correlation values for items 1 and 2 and BAD score values demonstrate sound convergence validity and the practicality of using both tools for identification and then measurement. Item 6 performed relatively poorly in these validity assessments and may over-represent the prevalence of dystonia. From this perspective, it could be considered for removal from the tool.

The mean BAD score of 17.8 for the dyskinetic PMT group is similar to that estimated in a geographic population of children with dyskinetic CP by Himmelmann et al., and reflects at least a moderate level of severity.[26] We suggest that there may be a spectrum of severity of dystonia across the CP population regardless of predominant motor type; and that part of the process of applying the label of dystonia in classification relies on the clinician's expertise such that a severity threshold is reached in certain cases where the dystonia (often identified but not quantified) is of a high enough level for the clinician to be confident in using this as the predominant motor type. Below this 'cut point' clinicians may be more likely to use a spastic label, as it may be more readily recognizable, regardless whether it really accounted for the greater part of the motor picture and associated impairments. This is one possible area to explore in future research focusing on clinician decision-





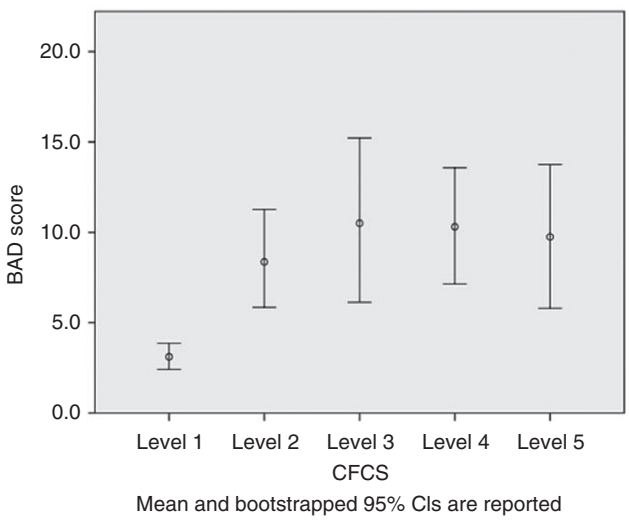


Figure 2: Distribution of Barry-Albright Dystonia scale (BAD) scores (mean, bootstrapped 95% CI) across population for categories of gross motor (Gross Motor Function Classification System [GMFCS]), manual ability (Manual Ability Classification System [MACS]), and communication (Communication Functional Classification System [CFCS]) functional domains.

making in the spastic versus dyskinetic (dystonia) classification issue.

Study participants were recruited from a hospital clinic rather than directly from the state CP Registry, although 95% of participants were listed on it.[27] While this produced a sound participation rate of 50%, there was an over-representation of participants with dyskinetic CP compared with non-participants. This is likely driven by a higher willingness to participate in the study by caregivers whose child has dyskinetic CP or features of dystonia, and who may perceive a benefit from participation.

In conclusion, hypertonia in the CP population can be evaluated by use of the HAT to concurrently identify components of dystonia and spasticity to overcome issues which may result from use of a motor predominance label. Across the whole population, motor function is lower in children with greater levels of dystonia indicating that dystonia may have more influence on function than previously recognized. Dystonia occurs much more frequently than may be assumed by the predominant motor type and this adds to the complexity of motor classification when spasticity is also present. There is a need to further explore situations where both spasticity and dystonia exist in terms of their relative contribution to motor impairment.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Barry-Albright Dystonia scale.

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