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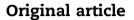
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Upper limb and hand patterns in cerebral palsy: Reliability of two new classifications



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ABSTRACT

Aim: To evaluate the inter- and intra-rater reliability of two previously developed classifications of upper limb and hand patterns.

Method: Two hundred and twelve films of patients with CP (118 of UL postures and 94 of hand tasks; median age 14, 3–46 years) were viewed by 18 examiners from 2 different rehabilitation centers, and one expert who had participated in the design of the classifications. They classed upper limb (3 patterns with sub-types) and hand patterns (2 patterns with subtypes) twice, at 2 months' interval. Inter- and intra-rater reliability were analysed. Results: Intra-rater and inter-rater reliability were very high for upper limb and hand patterns (0.87 < k < 0.92), and high for the subtypes (0.58 < k < 0.68). Examiners stated that both classifications were useful and feasible in clinical practice.

Interpretation: Despite the single, short training session on use of the classifications, agreement between the examiners and the expert examiner was good to high, confirming that these classifications are easy to use and reliable. The classifications proposed here provide homogenous terminology for use in both clinical practice and research, to describe, evaluate and follow-up changes in upper limb and hand patterns in patients with cerebral palsy, particularly those with dyskinesia.

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Cerebral palsy (CP) has been defined as "a group of permanent disorders of the development of movement and posture causing activity limitation(s) that are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain".³

Hypertonia of the upper limb is common in CP. It interferes with both active and passive arm function, leading to disability. It may also cause pain. The identification and treatment of hypertonia is a key aspect of rehabilitation. Local intramuscular injections of botulinum toxin type A (BoNT-A) are an established and well-tolerated treatment for local hypertonia in patients with CP.¹⁷ BoNT-A can be used in both lower and upper limb muscles, and is often combined with splinting and physical and occupational therapy.

Guidelines for BoNT-A therapy state that the specific pattern of hypertonia should be considered when determining the muscles to inject, and that the choice must relate to the goal of treatment.^{18,19} However, the muscles that should be injected to improve a particular spastic or dyskinetic pattern are not simple to determine. A classification system that states the most likely muscles involved in a particular spastic pattern would be useful. Equally, such a classification could be used to evaluate changes over time and following treatment. Existing classifications for subtypes of CP are based on location of lesion, part of the body involved, degree of impairment,⁴ gait patterns in spastic hemiplegia and spastic diplegia,⁵ or function such as the Gross Motor Function Classification System (GMFCS⁶) that evaluates function in sitting and walking. More recently, the Manual Ability Classification System (MACS⁷) was developed to evaluate how children use their hands to handling objects during daily activities. Several classifications have attempted to describe different grips or to classify hand-postures in patients with cerebral palsy,^{8–10} however they do not include all the patterns observed in patients. McConnell¹¹ and Wagner¹² recently highlighted several useful classifications that were first developed for surgical purposes, but are commonly used in clinical practice; for instance the Zancolli¹³ classification for the wrist, and the Matev,¹⁴ House¹⁵ and Corry¹⁶ classifications for the thumb.

Hefter et al.²⁰ proposed a classification of upper limb and hand postures in adults with acquired cerebral lesions that indicates the muscles involved. However, there are no similar classifications for use in patients with CP.

The final goal of any treatment is to improve participation, as defined in the International Classification of Functioning, Disability and Health (ICF – World Health Organization 2001), however many treatments are aimed at the body function and structure levels. Treatment of upper limb and hand deformities in children is important to help normal growth and prevent neuro-orthopedic complications that can lead to pain in adulthood, and potentially impact participation and quality of adult life.²¹ A classification of upper limb and hand deformities would be very useful to evaluate the relationship between improvements in body function and structure, activity and participation, as well as the role of changes in muscle function on improvements or losses of function, through longitudinal studies. A classification system would also be useful in clinical practice to identify predominant patterns of deformity for example in patients with dyskinesia, to facilitate communication between clinicians and for patient follow-up. From a research point of view, such a classification could help to identify homogenous subgroups of patients with CP for investigations as well as to evaluate the effects of treatments such as botulinum toxin injection or surgery.

We previously developed two classification systems, one for the upper limb and one for the hand, based on 100 films of patients with cerebral palsy.²² Separate classifications were developed following an initial study in which we found no correlations between upper limb and hand patterns, meaning that specific upper limb patterns are not always associated with specific hand patterns. Thumb patterns were not included in these classifications since robust classifications already exist,^{14–16} moreover thumb patterns are independent from hand patterns.

The aim of the present study was to evaluate the inter- and intra-rater reliability of classifications of upper limb patterns and hand patterns in patients with CP. The second aim was to evaluate the usefulness and feasibility of such a questionnaire in clinical practice and research, according to clinicians.

1. Methods

1.1. Participants

Patients with CP in our center are systematically filmed during assessments. We selected 212 films of patients to test the reliability of the classifications. The films were selected to cover a wide range of ages and what we assumed would be a wide range of upper limb and hand postures, that is to say that all films were included as far as they were exploitable regarding technical aspects (standardized shots, adequate definition of the image). Upper limb patterns were classified at rest (n = 40), during ambulation (walking or wheelchair) (n = 38) and during activity of the contralateral limb (n = 40). Regarding ambulation, there is some difference between the situations of walking or propelling an electric wheelchair, but there is also some likeness, as both situations allow the observation of tonic syncinesia while mowing. Patients are often complaining about the position of their upper limb while moving - because they sometimes hardly control it, and it can lead to uncomfortable situations, as the hand hitting other people or doorframe for example. This occurs during walking or while propelling an electric wheelchair. We thus chose to define ambulation as a situation of observation, as it was closely connected with patients' complains.

Hand patterns were classified at rest (n = 39) and during grasping activities (n = 55). The examiners were clinicians of different professions from two rehabilitation centers.

1.2. Refining of the classification

The first versions of the classifications²² were presented at national and international conferences (Toxine et MembreSupérieur – Colloque Lyon 2010, SPHERE – Brest 2013) to

collect opinions from professionals and determine interest in such classifications. A preliminary study of validity was also carried out using 45 short films of patients with cerebral palsy that were classified by a group of 8 clinicians including physiatrists, neuropaediatricians, physical therapists and occupational therapists. Results of this preliminary validity study, as well as comments and suggestions from the clinicians were taken into account to refine the wordings and the distinctions between the different upper limb and hand patterns. One of the main conclusions was that, since the same patient could present one type of pattern at rest and another during activity, it was important to state the conditions in which the patterns were classified.

1.3. Evaluation of inter and intra-rater reliability

Different conditions of observation were defined: the global upper limb pattern was classified at rest, during ambulatory activity, and during contralateral activity; the hand pattern was classified at rest and during grasping. One expert (a very experienced occupational therapist who participated in the development of the classification) was considered as goldstandard and classified all videos.

Reliability was then tested by examiners in two different rehabilitation centers run by the Croix-Rouge Française, one in Lyon and one in Paris.

For inter-rater reliability, the examiners all classified the same films. All examiners received a brief introduction to both classifications by the gold-standard expert. They were provided with a leaflet describing the classifications, which they were encouraged to look at thoroughly and regularly. The examiners received a copy of the films, which they were asked to classify within 4 weeks.

To evaluate intra-rater reliability, the same examiners classified the same films twice, at an interval of 2 months. They were sent a copy of all the films for the first classification, and another copy two months later, with all the films renamed and randomly reordered. Examiners were blinded to the classification they had previously assigned.

1.4. Statistics

Intra-rater reliability (test-retest) was analyzed as the percentage agreement between the two classifications done by each examiner.

Inter-observer reliability was determined with intra-class correlation coefficient (ICC) estimated from a multinomial model with mixed effects (a fixed effect on the condition of observation and a random effect on the video was introduced in the model). Its 95% confidence interval was built by the bootstrap method.

Concordance between all examiners and the expert examiner was determined with Fleiss's kappa (k) and its 95% confidence interval.²³ The following levels were determined for interpretation of the kappa²⁴: $k \leq 0.00 =$ disagreement, $0.01 < k \leq 0.20 =$ very weak agreement, $0.21 < k \leq 0.40 =$ weak agreement, $0.41 < k \leq 0.60 =$ moderate agreement, $0.61 < k \leq 0.80 =$ high agreement and $0.81 < k \leq 1.00 =$ almost perfect agreement.

Discordant classifications were described. SAS software was employed for the statistical analysis.

1.5. Evaluation of relevance and feasibility of the classifications

All examiners were asked to complete an anonymous questionnaire regarding their opinions of the classifications. The questionnaire was based on a 5-point Likert scale with 18 items concerning time to complete, relevance, and usefulness in clinical practice and research.

1.6. Ethics

A prior declaration to the CNIL (Commission Nationale de l'Informatiqueet des Libertés) was made to allow computer processing of personal data, and the study was conducted after approval from the ethic committee of Centre des Massues. Informed consent was obtained from the patients or the parents of children.

2. Results

Two hundred and twelve films were selected for the evaluation of reliability, from a group of 106 patients (48 (45%) quadriplegia, 17 (16%) triplegia, 41 (39%) hemiplegia), including 44 (41.5%) girls and 62 (58.5%) boys. Median age of the patients in the films was 14 years (range 3–46 years).

The films were rated by 19 examiners who did not participate in the development of the classification and were thus naïve to it: 8 occupational therapists, 5 physiotherapists, 5 physiatrists and 1 neuropaediatrician. Ages ranged from 25 to 57 (median = 38), and professional experience ranged from 0 to 27 years (median = 13).

2.1. Intra-rater reliability

Reproducibility between the 1st and 2nd observations was very high for all examiners for both upper limb (type I, II and III: mean agreement 89.5%) and hand patterns (Flex or Punching hands: 88.7%). Reproducibility was lower, but still very good, for the subtypes (72.7% for upper limb and 68% for hand).

2.2. Inter-rater reliability

Table 1 presents the global concordance (overall kappa) between the examiners and the expert examiner for each type and subtype of upper limb (a) and hand pattern (b). Table 2 presents the reproducibility between the examiners (ICC) for types and subtypes of upper limb (a) and hand patterns (b). Agreement and reproducibility for both upper limb and hand types was very high in all cases. Agreement and reproducibility for subtypes was always slightly lower, although the kappas showed good agreement. A descriptive analysis of discordant observations was carried out to identify the discordant subtypes (Table 3). Upper limb pattern type Ic was often classed as type Ia or Ib. Type IIb was often classed as type IIa. There was rarely discordance for Type III. Regarding hand patterns, the Total Flex Plus patterns were frequently incorrectly classified as either Total Flex or Simple flex Plus. The Other Flex patterns were consistently correctly classified.

Table 1 — Global concordance (overall kappa) bet upper limb (a); and hand patterns (b).	ween the examiners and the expert exami	iner for types and subtypes of							
Condition	Туре	Subtype							
	Kappa [CI 95%]	Kappa [CI 95%]							
a. Global concordance (overall kappa) between the exam	a. Global concordance (overall kappa) between the examiners and the expert examiner for types and subtypes of upper limb patterns								
At rest (40 videos)	0.91 ^a [0.89; 0.94]	0.68 ^b [0.64; 0.71]							
During contralateral activity (40 videos)	0.89 ^a [0.87; 0.92]	0.58 [0.54; 0.62]							
During ambulation (38 videos)	0.88 ^a [0.85; 0.92]	0.63 ^b [0.59; 0.67]							
Total (118 videos)	0.9ª [0.88; 0.91]	0.64 ^b [0.61; 0.66]							
b. Global concordance (overall kappa) between the exam	iners and the expert examiner for types and subt	types of hand patterns							
Grasping (55 videos)	0.89 ^ª [0.85; 0.93]	0.63 ^b [0.6; 0.66]							
At rest (39 videos)	0.87ª [0.83; 0.92]	0.67 ^b [0.64; 0.71]							
Total (99 videos)	0.92 ^a [0.89; 0.94]	0.65 ^b [0.62; 0.67]							

The agreement is characterized by a Kappa coefficient less than chance agreement if k < 0; slight agreement if 0.01 < k < 0.20; fair agreement if 0.21 < k < 0.40; moderate agreement if 0.41 < k < 0.60; substantial agreement if 0.61 < k < 0.80; almost perfect agreement if 0.81 < k < 0.99. Almost perfect agreement.

^b Substantial agreement.

Table 2 - Interrater reliability of the examiners (not including expert examiner). ICC [CI 95%]^a Condition Level a. Interrater reliability for types and subtypes of upper limb patterns At rest (40 videos) 0.96 type 0.78 subtype 0.91 During contralateral activity type (40 videos) subtype 0.84 During ambulation (38 videos) 0.97 type subtype 0.75 0.958 [0.952; 0.964] Total (118 videos) type subtype 0.704 [0.567; 0.841] b. Interrater reliability for types and subtypes of hand patterns During grasping (55 videos) 0.94 type 0.88 subtype At rest (39 videos) type 0.93 subtype 0.87 Total (94 videos) 0.944 [0.942; 0.946] type subtype 0.878 [0.856; 0.900]

^a ICC = intra-class correlation coefficient estimated from a multinomial model with mixed effects (a random effect on the video was introduced in the model), 95% confidence interval built by the bootstrap method.

For the Punching patterns, the Intrinsic pattern was clearly identified, but there was some confusion between Superficialis and Profundus Punching patterns.

2.3. Relevance and feasibility of the classifications

Eighty-three percent of the examiners stated that the classifications were useful for clinical practice, 83% found them quick to carry out and 70% stated they were easy to understand. Eighty-five percent reported that the classifications provided a precise description of patterns, 89% that they would facilitate communication between professionals, 86% that they would help to orient treatment, and 91% that they would help to determine the muscles involved in pathological patterns.

3. Discussion

Inter-rater reliability was high for both the upper limb and hand types. Agreement for subtypes was lower. Disagreements were explored in a descriptive analysis of concordant and discordant responses (Table 3); Type Ic includes shoulder extension while Type Ia and Ib do not. This was not always identified by the examiners, probably because shoulder extension is difficult to observe in the presence of a large thoracic kyphosis. However this pattern is clinically relevant with regard to treatment. The frequent discordance for Type IIb can be explained by the use of films, which made forearm position difficult to assess, especially when the wrist was completely flexed. For the hand classification, the Total Flex Plus pattern was consistently classified as either Total Flex or Simple Flex Plus. The Intrinsic Punching pattern was clearly identified, but there was some confusion between Superficialis and Profundus Punching patterns. Again, these discrepancies would probably not occur in the presence of patients since the films were sometimes of insufficient quality to determine if the fingers were completely flexed or not, or if there was a swan neck deformity.

These classifications can, and should, be used in different conditions since the patterns may differ depending on the activity. The condition should be specified during the assessment. For example, in the case of dyskinesia, the same patient may display a Type IIc or a Type IIIa pattern, depending on the activity (rest, walking, trying to catch an object etc.), or a Profundus Punching hand alternating with an Total Flex Plus hand during the approach to an object. The need for a clear communication between us about upper limb and hand patterns specifically emerged when treating dyskinetic patients, who often present variable patterns. We used to spend a lot of time describing these different patterns, and discussing about which pattern was the worse for function and for the patient's specific objective. So, before going on with this, we wanted to make it easier and quicker to identify patterns, and that is now allowed by the use of these classifications. But of course this is one step - the first step ... and clinicians will have to

Table 3 – Descriptive analysis of concordances between the examiners and the expert examiner for subtypes, and frequency table for discordant subtypes of upper limb (a) and hand patterns (b); in all conditions of observation.

a. Descriptive analysis of concordances between the examiners and the expert examiner for subtypes, and frequency table for discordant subtypes of *upper limb patterns*, in all conditions of observation combined (at rest, during ambulation and during contralateral activity)

Туре				Subtypes										
Pattern	Concordances	Discordances	Pattern	Concordances	Discordances	Description of discordances								
	N (%)	Ν		N (%)	N	Ia	Ib	Ic	IIa	IIb	IIc	IIIa	IIIb	NA ^a
Ia	325 (90)	36	Ia	232 (71)	93	0 (0)	29 (31)	62 (67)	-	_	_	-	_	2 (2)
Ib	531 (100)	1	Ib	393 (74)	138	116 (84)	0 (0)	17 (12)	-	-	-	_	-	5 (4)
Ic	72 (76)	23	Ic	24 (33)	48	21 (44)	25 (52)	0 (0)	-	-	-	-	-	2 (4)
IIa	196 (86)	32	IIa	147 (75)	49	_	-	-	0 (0)	45 (92)	3 (6)	_	-	1 (2).
IIb	230 (93)	17	IIb	110 (48)	120	_	-	-	87 (73)	0 (0)	32 (27)	_	-	1 (1)
IIc	206 (99)	3	IIc	140 (68)	66	-	-	-	21 (32)	45 (68)	0 (0)	_	-	0 (0)
IIIa	355 (89)	44	IIIa	352 (99)	3	-	-	-	-		_	0 (0)	3 (100)	0 (0)
IIIb	156 (91)	15	IIIb	139 (89)	17	-	-	-	-	-	_	14 (82.4)	0 (0)	3 (18)

b. Descriptive analysis of concordances between the examiners and the expert examiner for subtypes, and frequency table for discordant subtypes of *hand patterns*, in all conditions of observation combined (at rest and during grasping)

Type Subtypes														
Pattern	Concordances	Discordances	Pattern	Concordances Di	scordances	Description of discordances								
	N (%)	N		N (%)	N	Flex Simple	Flex Total	Flex Simple Plus	Flex Total Plus	0	Punching Superficialis	Punching Profundus	NA ^a	MD ^b
Flex Simple	493 (99.8)	1	Flex Simple	383 (77.7)	110	0 (0)	48 (43.6)	53 (48.2)	7 (6.4)	-	-	-	2 (1.8)	0 (0)
Flex Total	447 (98)	9	Flex Total	336 (75.2)	111	78 (70.3)	0 (0)	5 (4.5)	26 (23.4)	-	-	-	2 (1.8)	0 (0)
Flex Simple Plus	152 (100)	0	Flex Simple Plus	131 (86.2)	21	3 (14.3)	1 (4.8)	0 (0)	17 (81)	-	-	-	0 (0)	0 (0)
Flex Total Plus	265 (99.6)	1	Flex Total Plus	147 (55.5)	118	3 (2.5)	47 (39.8)	67 (56.8)	0 (0)	-	-	-	1 (0.8)	0 (0)
Punching Intrinsic	112 (73.7)	40	Punching Intrinsic	95 (84.8)	17	0 (0)	-	-	-	0 (0)	12 (70.6)	4 (23.5)	1 (5.9)	0 (0)
Punching Superficialis	86 (90.5) s	9	Punching Superficialis	52 (60.5) s	34	0 (0)	-	-	-	4 (11.8)	0 (0)	28 (82.4)	2 (5.9)	0 (0)
Punching Profundus	154 (90)	17	Punching Profundus	117 (76)	37	0 (0)	-	-	-	1 (2.7)	29 (78.4)	0 (0)	6 (16.2)	1 (2.7)

^a NA: not answered. Subtype was voluntarily not determined by the examiner.

^b Missing data.

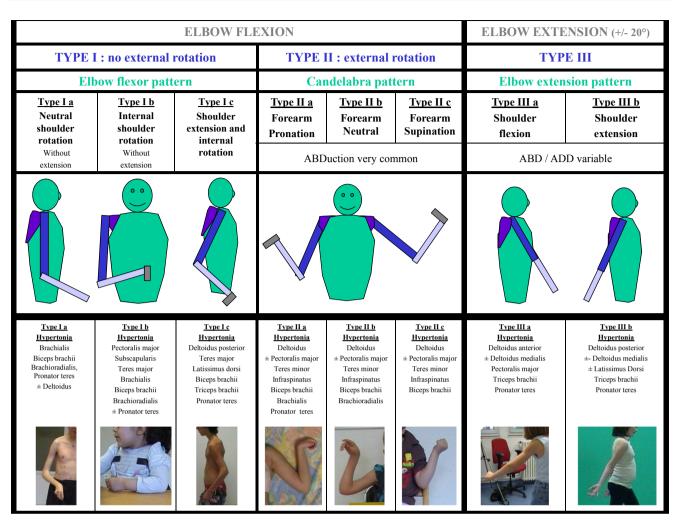


Fig. 1 - Classification of upper limb patterns.

determine which pattern limits function the most, in order to plan treatment.

These classifications focus on the anatomical and muscular status of the upper limb, regardless of type of CP and functional status. Future studies should determine if specific patterns are related to different degrees of impairment of unimanual or bimanual function.

In contrast with functional-types of classifications, which are very important to evaluate the effect of treatment on activity and participation, the classifications presented here can help to determine the targets for treatment. Figs. 1 and 2 state the muscles likely implicated in the different upper limb and hand patterns respectively. Needless to say that these descriptive classifications of patterns are not sufficient to define the treatment. Even if the muscular implications are suggested in the definition of patterns, these suggestions must be confirmed by a rigorous clinical examination. Furthermore, these patterns are not correlated to function - even if it seems that some patterns are more noxious for the successful functional use of both hands together. But it is clear that we do not treat one pattern simply because it exists, but we treat one pattern because it is annoying for one specific patient in one specific situation - just like we do not treat spasticity just

because we find it clinically but we treat spasticity if it is annoying functionally. Difficulty with hygiene and pain seem to be often associated with closed hands (Total Flex pattern or Superficialis and Profundus Punching pattern). Wrist extension and radial deviation increase the difficulty to open the hand, particularly since the finger and thumb flexors are hypertonic. The last phalanxes are pressed against palm, which can cause pain, cutaneous lesions from the nails and even ingrowing nails. Upper limb patterns can interfere with total body posture and balance. This is the case for Type I, which can be associated with a symmetric or asymmetric thoracic kyphosis because of hypertonia of Pectoralis Major. Type II can induce difficulties with positioning in non-ambulant patients who use a seat orthosis. It can also reduce balance in ambulant patients, whose gait is often already unsteady. Type III can also affect global posture, if the patient tries to hide his or her upper limb because of the perception of its appearance.

Our work shows the reliability of the classifications which we developed, but does not claim to be able to show that they will be sensitive to change, in particular in a context of contractures. In case of studies focusing on treatment efficiency (toxin, surgery ...), these classifications can be used to define homogenous groups of patients, but the effect of the

FLEX HANDS (wrist flexion pattern)									
<u>Simple Flex</u> Wrist collapse, finger extension	<u>Total Flex</u> Wrist and finger flexion								
Fingers may be slightly passively flexed, and/or MCP joints may hyperextend on approach to object	PIP and DIP joints are flexed, MCP joints may extend on approach to object								
Hvpertonia No hypertonia or FCU/ Palmar/ FCR, FDS/FDP ?	Hvpertonia FDS/FPD Palmar, FCU, FCR FPL ?								
Simple Flex Plus	<u>Total Flex Plus</u>								
Wrist flexion associated with swan neck deformity or dinosaur hand (exclusion of index during grasping)	Wrist and finger flexion associated with swan neck deformity or dinosaur hand (exclusion of index during grasping)								
HypertoniaNo hypertonia orFCU/ Palmar/ FCR,FDS/FDP ?+ Lumbricales / IO	Hypertonia FDS/FPD Palmar, FCU, FCR FPL ? + Lumbricales / IO								

PUNCHING HANDS (Wrist extension pattern)										
<u>Intrinsic Pu</u>	nching Hand	<u>Superficialis P</u>	unching Hand	Profundus Punching Hand						
Wrist extension MCP flexion, PIP and DIP extension		Wrist extension MCP and PIP flexion, DIP extension		Wrist extension MCP, PIP and DIP flexion						
<u>Hvpertonia</u> ECRB – ECRL – Lumbricales - IO		<u>Hvpertonia</u> ECRB – ECRL - FDS		<u>Hypertonia</u> ECRB – ECRL - FDP	SR)					

Fig. 2 – Classification of hand patterns.

treatment must be measured with other tools, adapted to the objective of the treatment.

Further study is, however required to specifically correlate each classification with clinical examinations, specifying force, spasticity and/or contractures, as did Hefter in his classification of patterns in adults with upper limb spasticity.²⁰ It will be interesting to explore if some patterns are really similar, as they seem to be (in patients with spastic hypertonia), and it will be also interesting to see if patterns specific to our group of cerebral-palsied patients are more linked to dyskinetic hypertonia or to young patients without contractures.

4. Conclusion

The aim of this study was to determine the intra-and interrater reliability of two classifications of upper limb and hand patterns in patients with CP. Both intra and inter-rater reliability were found to be "good" to "almost perfect". Examiners had participated in only a short training session for use of the classifications; however there was good agreement between the expert examiner and the examiners who were naïve to the classifications, confirming that they are easy to use and are reliable. Moreover, the examiners stated that the classifications were easy to use, as well as useful for communication in clinical practice and for research.

Further investigation is needed to confirm the specific muscles involved in each pattern, as well as to determine if the patterns are correlated with functional level and age. However, the classifications proposed here can already be used to describe upper limb and hand patterns in patients with cerebral palsy, particularly those with dyskinesia, in both clinical practice and research.

Conflict of interest

The authors declare no conflict of interest.

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