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Hypertonia Assessment Tool: Reliability and Validity in Children With Neuromotor Disorders

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Abstract

The Hypertonia Assessment Tool is a 7-item instrument that discriminates spasticity, dystonia, and rigidity on 3 levels: item scores, subtype, and hypertonia diagnosis for each extremity. We quantified the inter- and intrarater reliability using Kappa statistics, Gwet's first-order agreement coefficient (both with 95% confidence interval), and percentage agreement for all levels. For validity, we compared the Hypertonia Assessment Tool subtype with the clinical diagnosis provided by the physicians. Two physiotherapists tested 45 children with neuromotor disorders. The interrater reliability (n = 45) of the Hypertonia Assessment Tool subtype was moderate to substantial whereas the intrarater reliability (n = 42) was almost perfect. The Hypertonia Assessment Tool showed good agreement in detecting spasticity. On the contrary, there was a higher presence of dystonia of 24% to 25% tested with the Hypertonia Assessment Tool compared to the clinical diagnosis. Even some individual items showed lower agreement between raters; the Hypertonia Assessment Tool subtypes and diagnosis were reliable. Validity of the Hypertonia Assessment Tool to test spasticity is confirmed, whereas, for dystonia and rigidity, further studies are needed.

Keywords

cerebral palsy, dystonia, spasticity, measurement, psychometric properties

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Treatment of high tone in children with neuromotor disorders depends on the type of impairment.[1] It is known that abnormalities of muscle tone can influence daily activities and movement variability and affect joint mobility, growth, posture, and biomechanical alignment.[2] Assessments that can assess hypertonia and distinguish between hypertonia types such as spasticity or dystonia would be valuable to improve our understanding of the mechanisms leading to deformities and to plan rehabilitative or surgical interventions.[3] Sanger et al[4] defined 3 types of hypertonia: spasticity, dystonia, and rigidity. Spasticity is defined as velocity-dependent resistance to muscle stretch, and resistance to externally imposed movement that rises rapidly above a threshold speed.[5] Dystonia is both hypertonic and hyperkinetic. For the assessor, it is important to differentiate if the movements can be controlled voluntarily or not.[4] Rigidity in children with neuromotor disorders is rather scarce. By the consensus group, rigidity is defined as follows: no velocity-dependent resistance, simultaneous cocontraction, the limb stays in a placed position and does not return to a relaxed one, and no involuntary movement occurs.[4]

Following these definitions, Jethwa et al[6] developed the Hypertonia Assessment Tool (HAT). The Hypertonia Assessment Tool consists of 7 items: items 1, 2, and 6 assess signs of dystonia, items 3 and 4 assess signs of spasticity, and items 5 and 7 assess signs of rigidity. The items are scored as positive or negative. One or more positive scores of 1 hypertonia item confirm the presence of this subtype. Each limb can be observed and receives an individual diagnosis of hypertonia. Jethwa et al[6] found substantial to almost perfect reliability (test-retest and interrater) results for identification of spasticity and moderate reliability for dystonia in children. In their study, only 1 limb per child with known hypertonia was tested. The validity was fair to substantial, with a higher positive agreement for identifying the presence of spasticity and dystonia.

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Because of the absence of rigidity, a high negative agreement was observed. Knights et al[7] found comparable results for reliability and validity of the Hypertonia Assessment Tool subtypes in children with cerebral palsy. Although they investigated the influence of using video recording for scoring the Hypertonia Assessment Tool items, they did not find any additional advantages of using the recordings.[7]

In these previous studies, all 4 limbs of children with cerebral palsy and known hypertonia were tested, and the results were pooled, that is, it remained difficult to assess the validity or reliability for each extremity separately.[6,7] In contrast, we intended to investigate the reliability and validity of the Hypertonia Assessment Tool scoring for each extremity separately. Further, we included children with different neuromotor disorders such as acquired brain injuries and neurologic syndromes, as these could also present signs of hypertonia.

The first step was to translate the Hypertonia Assessment Tool into German.

The aim of our study was to assess the inter- and intrarater reliability of the Hypertonia Assessment Tool in a group of children with different neuromotor disorders for all 4 limbs independently. We investigated this for all 3 levels of the Hypertonia Assessment Tool: (1) the 7 items, (2) the 3 types of hypertonia (Hypertonia Assessment Tool subtypes), and (3) the Hypertonia Assessment Tool diagnosis. As hypertonia can be influenced by different internal and external factors, such as emotional state or environmental factors,[8] we also investigated whether the order of testing influenced the outcome. As a second aim, we investigated the validity by comparing the Hypertonia Assessment Tool subtype with the clinical diagnosis provided by the physicians.

Methods

The Hypertonia Assessment Tool was translated independently into the German language by 2 native German-speaking physiotherapists (first and second author), according to the guidelines of Beaton et al.[9] The consensus version was reread by a study associate, queries were discussed by the authors, and a final consensus version was made (Appendix A, Supplementary Material). This version was back-translated into English by a professional company and was compared to the original English version.

Study Procedure

This psychometric study used a cross-sectional design with repeated assessments to determine the validity and reliability, respectively, of the Hypertonia Assessment Tool. The test was performed 3 times in each child. On day 1, testers A (second author) and B (first author) performed the test in a random order. Each tester was blinded for the results of the other tester. The test was repeated 1 week later by one of the 2 testers at the same time of the day in the same room. We followed the Guidelines for Reporting Reliability and Agreement Studies (GRRAS).[10] Because no criterion standard is available to assess hypertonia, we used the diagnosis of the physician as current praxis to compare the Hypertonia Assessment Tool subtypes for validity. An independent and blinded research associate collected the presence of hypertonia per extremity of the diagnoses provided by the physicians.

Participants

Children were recruited from the Rehabilitation Center for Children and Adolescents of the University Children's Hospital Zurich in Affoltern am Albis. Inclusion criteria were diagnosed as having a neuromotor disorder, aged 4 to 19 years, and able to follow simple instructions (such as closing and opening the eyes) and lie in a supine position for 20 minutes. Participants were excluded from the study if they had received surgery and/or botulinum toxin in the limbs within the last 3 months, changed medication (muscle relaxants) during the measurement period, or if they were not allowed to be moved passively. Because of convenience sampling, we expected to include children with signs of hypertonia as well as with no hypertonia in certain extremities. The COSMIN guidelines for psychometric studies considered sample sizes less than 30 as poor.[11] Therefore, we aimed to recruit more than 30 children.

Measures

Besides the German Hypertonia Assessment Tool version and the medical diagnosis, we included the Functional Independence Measure for children (WeeFIM) mobility and total score for all participants and the Gross Motor Function Classification System (GMFCS) to describe the motor abilities of the children.[12,13] The Gross Motor Function Classification System was developed to describe the ability for ambulation in children with cerebral palsy. Five categories describe clinically meaningful levels of mobility. In clinical practice, the Gross Motor Function Classification System is also applied to children with other neuromotor impairments, such as acquired brain injuries or syndromes, as an indicator of functional mobility.

Raters

The 2 raters (first and second authors) were physiotherapists with 14 and 5 years of practical experience, respectively, in treating children with neuromotor disorders. Both raters had started to use the Hypertonia Assessment Tool clinically 3 months before the study started.

Assessment Procedure

The Hypertonia Assessment Tool was performed according to the German manual of the Hypertonia Assessment Tool (Appendix A, Supplementary Material). During the first week, the 2 Hypertonia Assessment Tool assessments (interrater reliability) occurred in a 1-hour time window, after the lunch break. One week later, half an hour was scheduled to perform the third test (intrarater reliability). The child was in supine position (flat) with slightly bended knees (supported) and a pillow under the head. The limbs were tested in a random order. Item 6 "increased tone with movement of another body part" is always tested in a joint classified as having spasticity (items 3 "velocity dependent resistance to stretch" and/or 4 "presence to spastic catch"). If more than 1 joint of a limb showed spasticity, item 6 was assessed on the more distal joint. The spasticity items 3 and 4 were tested during the same movement.

Statistical Analysis

Statistical calculations were performed with SPSS (IBM SPSS Statistics 19, Chicago, IL), and for the Kappa statistics with the open online platform http://vassarstats.net/kappa.html. For the reliability statistics, we presented the percentage agreement, Kappa coefficient and 95% confidence interval, and maximum attainable Kappa (κ_{max}) as recommended by Sim and Wright.[14] The κ_{max} values represent the greatest possible agreement of a data matrix. As some items might show an

imbalance between the presence or absence of signs of hypertonia, which would negatively affect the Kappa calculation, we applied Gwet's first-order agreement coefficient (Gwet's AC1) using the SPSS syntax found at http://www.ccitonline.org/jking/homepage/Gwet.sps.[15] We used the bootstraps confidence interval overall method for calculating the 95% confidence interval for Gwet's AC1. Kappa coefficients and Gwet's AC1 were interpreted as follows: <0 = poor, 0.0-0.20 = slight, 0.21-0.40 = fair, 0.41-0.60 = moderate, 0.61-0.80 = substantial, and 0.81-1.00 = almost perfect.[16]

To make our results comparable to the previous reliability studies of the Hypertonia Assessment Tool, we provided the prevalence index (prevalence index = agreement of present divided by the agreement of absent), the bias index (bias index = disagreement of absent divided by the disagreement of present), and bias-adjusted Kappa in the Appendices B and C (Supplementary Material).[14]

As moving the body parts of the patient might influence the muscle tone, we also compared the first with the second test (performed on the same day). We calculated the presence of a positive hypertonia sign (prevalence rate) of all items for all 3 subtypes. As a parameter to detect changes, risk differences were calculated as follows: risk difference = prevalence rate first test minus prevalence rate second test.[17] Statistical differences were examined with the McNemar test.

To determine the validity, the Hypertonia Assessment Tool subtypes were compared to the diagnosis by the physicians for each limb separately. We used positive predictive values and negative predictive values to estimate the accuracy of the Hypertonia Assessment Tool assessment. The positive predictive value is used to indicate the probability that in the case of a positive score, the patient really shows the presence of this subtype. The negative predictive value is used to indicate the probability that in the case of a negative score, the patient does not show this hypertonia type. A high result, close to 1, can be interpreted as indicating the accuracy of the Hypertonia Assessment Tool. To reflect the absolute difference, we calculated the risk difference (prevalence rate Hypertonia Assessment Tool subtypes minus prevalence rate clinical diagnosis) again[17] and performed McNemar tests. Alpha was set at 0.05.

Results

Participants

Forty-six children (30 boys, 16 girls) were recruited from August 2013 to July 2015 for this study. One child (Gross Motor Function Classification System level IV; spastic cerebral palsy) performed just one measurement, because of an illness of one of the raters. This child was only included for the validity analysis. The mean age (n = 46) was 11 year 6 months (standard deviation SD 4 years 4 months) range 4 years 2 months to 18 years 10 months. The children had a mean Functional Independence Measure for Children of 18 (standard deviation 11; maximal score 35) in the mobility part whereas 27 used a wheelchair and 19 could walk (with and without walking aids). The total mean Functional Independence Measure for Children was 71 (standard deviation 36; maximal score 126 points). The children were diagnosed with spastic cerebral palsy (n = 21), dyskinetic cerebral palsy (n = 5), ataxic cerebral palsy (n = 5), status after acquired brain injuries (n = 11), or neuromotor disorders caused by different syndromes (n = 4). The Gross Motor Function Classification System levels varied as follows: level I, n = 7; level II, n = 7; level III, n = 14; level IV, n = 9; and level V, n = 9.

The Hypertonia Assessment Tool could be applied to all children. Overall, it took 12 to 20 minutes to test all 4 extremities in 1 child. Three children, with larger cognitive impairments, were not able to perform items 2 and 6. They could not perform a movement of the distal body part, as described in the manual. In these cases, only item 1 could be performed to determine dystonia. Rater A observed rigidity in 1 child and rater B in 3 children, as confirmed by item 5 (equal resistance to passive stretch during bidirectional movement of a joint). No child was positively tested with item 7. The Hypertonia Assessment Tool diagnosis for each limb obtained during the first test is presented in Figure 1.

Interrater Reliability

The percentage agreement, Gwet's AC1, and Kappa values (with 95% confidence interval) for all items, the subtype, and the Hypertonia Assessment Tool diagnosis (Table 1) were presented (n = 45). Slight to fair agreement according to the Kappa values was found for items 4, 6, and 2 on the left side. All other items showed moderate to almost perfect values with varying K_{max}. For the subtypes dystonia and spasticity, all Kappa values were fair to moderate, and for the Hypertonia Assessment Tool diagnosis, moderate for the upper limb and the lower right limb and fair for the lower left limb. As rigidity was rarely diagnosed, Kappa could not be calculated, but Gwet's AC1 values and the percentage agreements were almost perfect. Except for 2 items and 1 subtype, Gwet's AC1 values were similar to or exceeded the Kappa values. As the prevalence and bias index were low ($\leq$0.20), most Kappa values of the subtypes did not differ from the bias-adjusted Kappa. Bias-adjusted Kappa and Kappa values differed only strongly for the left lower limb spasticity (bias-adjusted Kappa 0.60 vs Kappa 0.41) and for the subtype rigidity (bias-adjusted Kappa: 0.91-1.00; Appendix B, Supplementary Material).

Intrarater Reliability

Because of practical circumstances, 2 children could not be measured a second time. Rater A tested 28 children twice, rater B 15. One child had to be excluded because of change of medication; therefore, 42 children were included in this analysis. The Kappa coefficients and Gwet's AC1 values varied from moderate to almost perfect (Table 2). Item 2 showed the lowest Kappa values. The percentile agreements, Kappa values, and Gwet's AC1 values of the subtypes (dystonia and spasticity) were almost perfect (except for Dystonia, right lower limb, according to Gwet's AC1 value). Compared with the interrater reliability findings, Gwet's AC1 values were much closer to the Kappa values for the intrarater reliability. The percentage agreements and Gwet's AC1 values for (absence of) rigidity were almost perfect. For the left lower limb subtype spasticity and all limbs subtype rigidity the prevalence index was higher than 0.50; therefore, higher bias-adjusted Kappa resulted. The bias-adjusted Kappa differed from the Kappa value for left lower limb spasticity (bias-adjusted Kappa 0.81 vs

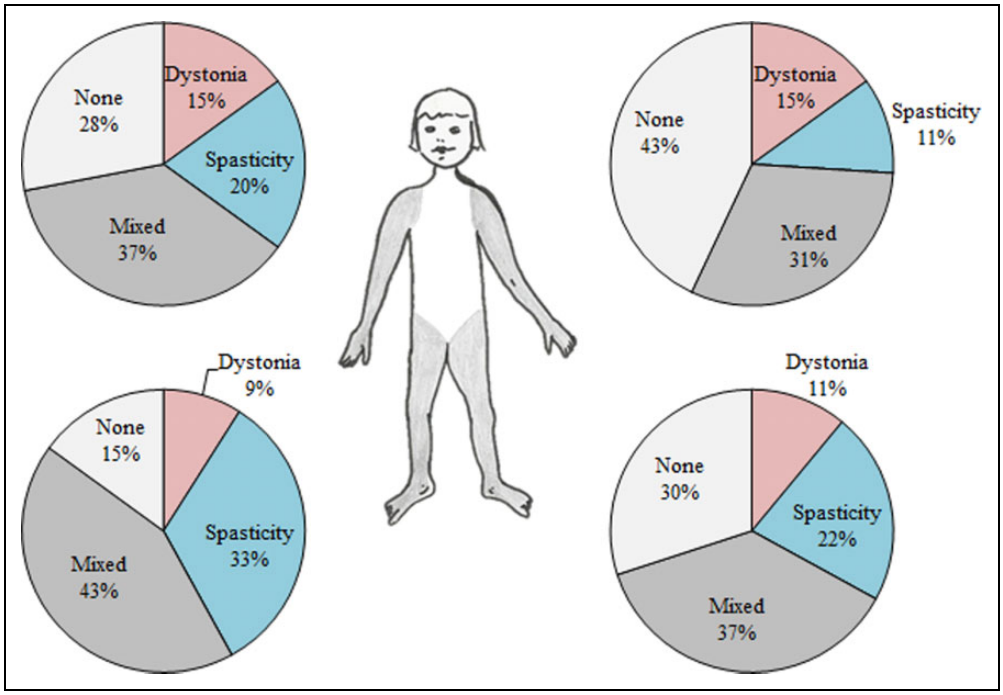

Figure 1. Distribution of the Hypertonia Assessment Tool diagnosis. The distribution of the Hypertonia Assessment Tool diagnosis in all children (n = 46) according to the limbs, measured at the first test situation (rater A, n = 23; rater B, n = 22). Because of no pure rigidity, this diagnosis is missing on the figure (0%).

Table 1. Interrater Reliability of the Hypertonia Assessment Tool (HAT) Items, Subtypes, and Diagnosis.

			Left side				Right side			
HAT	Limb	Hypertonia	% Agr.	AC1 (95% CI)	Kappa (95% CI)	κ_{max}	% Agr.	AC1 (95% CI)	Kappa (95% CI)	κ_{max}
Items	Upper limb	1. Dystonia	87	0.68 (0.42-0.88	0.62 (0.34-0.90)	0.62	76	0.59 (0.55-0.64)	0.43 (0.14-0.72)	0.44
		2. Dystonia	76	0.62 (0.58-0.67)	0.39 (0.06-0.72)	0.39	76	0.64 (0.60-0.70)	0.42 (0.11-0.73)	0.42
		3. Spasticity	80	0.60 (0.56-0.64)	0.61 (0.36-0.83)	0.87	77	0.57 (0.34-0.81)	0.55 (0.31-0.81)	0.64
		4. Spasticity	51	0.37 (0.32-0.41)	0.15 (0.00-0.37)	0.43	80	0.57 (0.53-0.61)	0.29 (0.00-0.70)	0.76
		5. Rigidity	93	0.98 (0.94-1.00)	–	–	96	0.95 (0.91-0.99)	–	–
		6. Dystonia	71	0.50 (0.46-0.56)	0.34 (0.05-0.65)	0.44	69	0.53 (0.49-0.59)	0.31 (0.00-0.62)	0.31
		7. Rigidity	100	1.00 (1.00-1.00)	–	–	100	1.00 (1.00-1.00)	–	–
	Lower limb	1. Dystonia	87	0.79 (0.75-0.85)	0.62 (0.36-0.88)	0.62	77	0.65 (0.60-0.70)	0.42 (0.11-0.74)	0.54
		2. Dystonia	74	0.59 (0.55-0.68)	0.40 (0.12-0.67)	0.62	74	0.59 (0.55-0.64)	0.41 (0.11-0.71)	0.52
		3. Spasticity	84	0.75 (0.71-0.81)	0.59 (0.32-0.87)	0.94	77	0.57 (0.53-0.62)	0.55 (0.31-0.79)	1.00
		4. Spasticity	51	0.03 (0.00–0.07)	0.11 (0.00-0.37)	0.43	62	0.29 (0.25-0.35)	0.22 (0.00-0.51)	0.59
		5. Rigidity	93	0.93 (0.88-0.97)	–	–	96	0.95 (0.91-0.99)	–	–
		6. Dystonia	63	0.41 (0.36-0.46)	0.20 (0.00-0.53)	0.84	73	0.57 (0.52-0.61)	0.43 (0.14-0.72)	0.64
		7. Rigidity	98	0.98 (0.94-1.00)	–	–	100	1.00 (1.00-1.00)	–	–
Subtypes	Upper limb	Dystonia	73	0.46 (0.43-0.51)	0.48 (0.23-0.73)	0.65	80	0.61 (0.57-0.67)	0.60 (0.37-0.84)	0.60
		Spasticity	78	0.56 (0.52-0.61)	0.56 (0.32-0.81)	0.82	80	0.61 (0.57-0.65)	0.60 (0.36-0.83)	0.69
		Rigidity	100	0.98 (0.94-1.00)	–	–	100	0.98 (0.94-1.00)	–	–
	Lower limb	Dystonia	71	0.42 (0.38-0.47)	0.42 (0.16-0.69)	0.96	77	0.56 (0.52-0.61)	0.56 (0.32-0.80)	0.75
		Spasticity	80	0.70 (0.66-0.75)	0.41 (0.05-0.75)	0.93	87	0.74 (0.71-0.79)	0.72 (0.52-0.93)	1.00
		Rigidity	93	0.96 (0.93-1.00)	–		96	0.95 (0.91-0.99)	–	–
Diagnosis	Upper limb		58	0.48 (0.44-0.52)	0.42 (0.23-0.62)	0.79	64	0.57 (0.53-0.61)	0.49 (0.29-0.69)	0.68
	Lower limb		58	0.51 (0.33-0.69)	0.36 (0.14-0.58)	0.83	62	0.54 (0.50-0.58)	0.48 (0.28-0.67)	0.72

Abbreviations: AC1, Gwet's first-order agreement coefficient; Agr., agreement; CI, confidence interval.
[a]Table presents % agreement, Gwet's first-order agreement coefficient, Kappa, and κ_{max} between 2 raters of 2 assessments following each other, n = 45; excluding items 2 and 6, n = 42.

Table 2. Intrarater Reliability of the Hypertonia Assessment Tool (HAT) Items, Subtypes, and Diagnosis.

			Left side				Right side			
HAT	Limb	Hypertonia	% Agr.	AC1 (95% CI)	Kappa (95% CI)	κ_{max}	% Agr.	AC1 (95% CI)	Kappa (95% CI)	κ_{max}
Items	Upper limb	1. Dystonia	83	0.71 (0.67-0.76)	0.62 (0.37-0.88)	0.84	83	0.77 (0.68-0.78)	0.62 (0.37-0.88)	0.84
		2. Dystonia	78	0.61 (0.38-0.87)	0.50 (0.22-0.79)	0.94	78	0.64 (0.59-0.69)	0.50 (0.22-0.79)	0.94
		3. Spasticity	95	0.90 (0.85-0.94)	0.90 (0.78-1.00)	1.00	95	0.82 (0.77-0.86)	0.90 (0.78-1.00)	1.00
		4. Spasticity	88	0.85 (0.80-0.89)	0.67 (0.39.0.94)	0.80	88	0.84 (0.79-0.89)	0.67 (0.39.0.94)	0.80
		5. Rigidity	100	1.00 (1.00-1.00)	–	–	100	0.98 (0.93-1.00)	–	–
		6. Dystonia	95	0.91 (0.76-1.00)	0.90 (0.75-1.00)	1.00	95	0.87 (0.81-0.91)	0.90 (0.75-1.00)	1.00
		7. Rigidity	100	1.00 (1.00-1.00)	–	–	100	1.00 (1.00-1.00)	–	–
	Lower limb	1. Dystonia	86	0.80 (0.76-0.84)	0.58 (0.26-0.89)	0.86	81	0.72 (0.68-0.76)	0.51 (0.20-0.82)	0.88
		2. Dystonia	85	0.77 (0.72-0.81)	0.68 (0.44-0.91)	0.89	85	0.76 (0.72-0.80)	0.69 (0.46-0.92)	0.79
		3. Spasticity	90	0.72 (0.51-0.89)	0.78 (0.57-0.98)	0.89	95	0.82 (0.78-0.86)	0.90 (0.78-1.00)	1.00
		4. Spasticity	86	0.61 (0.57-0.65)	0.71 (0.49-0.92)	0.80	86	0.72 (0.68-0.77)	0.68 (0.44-0-92)	0.89
		5. Rigidity	100	0.98 (0.94-1.00)	–	–	100	0.98 (0.93-1.00)	–	–
		6. Dystonia	95	0.75 (0.70-0.79)	0.79 (0.59-0.98)	1.00	90	0.75 (0.58-0.98)	0.79 (0.44-0.92)	0.89
		7. Rigidity	100	1.00 (1.00-1.00)	–	–	100	1.00 (1.00-1.00)	–	–
Subtypes	Upper limb	Dystonia	95	0.90 (0.77-1.00)	0.91 (0.78-1.00)	0.91	91	0.81 (0.77-0.85)	0.81 (0.64-0.99)	0.91
		Spasticity	95	0.91 (0.77-1.00)	0.91 (0.78-1.00)	0.91	93	0.87 (0.82-0.90)	0.86 (0.71-1.00)	0.86
		Rigidity	100	1.00 (1.00-1.00)	–	–	100	1.00 (1.00-1.00)	–	
	Lower limb	Dystonia	90	0.86 (0.81-0.90)	0.90 (0.77-1.00)	1.00	90	0.77 (0.72-1.00)	0.81 (0.77-1.00)	0.90
		Spasticity	90	0.84 (0.81-0.89)	0.74 (0.52-0.98)	1.00	95	0.91 (0.86-0.94)	0.90 (0.77-0.00)	0.90
		Rigidity	100	1.00 (1.00-1.00)	–		100	1.00 (1.00-1.00)	–	
Diagnosis	Upper limb		95	0.94 (0.89-0.98)	0.94 (0.85-1.00)	0.94	84	0.86 (0.82-0.90)	0.83 (0.69-0.97)	0.83
	Lower limb		88	0.83 (0.79-0.87)	0.83 (0.68-0.97)	0.86	87	0.80 (0.76-0.84)	0.79 (0.68-0.92)	0.86

Abbreviations: AC1, Gwet's first-order agreement coefficient; Agr., agreement; CI, confidence interval.
[a]Table presents % agreement, Gwet's first-order agreement coefficient, Kappa, and κ_{max} of 2 tests of the same rater with 1 week in between; n = 42; except for items 2 and 6 (n = 39).

Kappa 0.74) and the subtype rigidity (bias-adjusted Kappa 0.91-1.00; Appendix C, Supplementary Material).

Influence of Sequence

Rater A tested 23 children in the first test situation, rater B 22. Across the limbs, dystonia showed a prevalence of 30% to 32% in the first test and 24% to 26% in the second test. There was a 4% to 7% less chance (risk difference value) to detect the presence of dystonia in the second test. However, these differences were not significant (upper limbs: right $P = .21$, left $P = .20$; lower limbs: right $P = .16$, left $P = .49$). Spasticity showed a prevalence of 30% to 56% in the first test and 27% to 62% in the second test, that is, a 6% to 7% less chance. For lower limb spasticity on the left side, 6% more present of spasticity was rated in the second test situation. For the right side, spasticity was rated 7% less in the second test, and for the upper limb, 3% (right) and 4% (left) less presence of spasticity was diagnosed in the second test. Again, these differences were not significant (upper limbs: right $P = .66$, left $P = .51$; lower limbs: right $P = .35$, left $P = .36$). Rigidity showed a prevalence of 1% to 5% in the first test, and rigidity was not observed in the second test, by a P value of 1.0 for all limbs.

Validity for Dystonia and Spasticity

Although the prevalence of spasticity was in high agreement when diagnosed by the physician and the Hypertonia Assessment Tool scoring, such high agreement was not found for dystonia (Table 3). For spasticity, the risk difference was very low between 0% and 7% and not significant. Positive predictive value and negative predictive value were high as well. For dystonia, the risk difference of the presence of dystonia was significantly higher with 24% to 26% more signs of dystonia measured by the Hypertonia Assessment Tool than clinically diagnosed (Table 3). Therefore, the positive predictive value of the Hypertonia Assessment Tool subtype of dystonia was low, due to higher testing of positive signs of dystonia with the Hypertonia Assessment Tool, while the negative predictive value was high, due to the high agreement of the Hypertonia Assessment Tool with the negative diagnosed. Rigidity was not included, because of its small prevalence.

Discussion

The aim of this study was to determine the reliability and the validity of the Hypertonia Assessment Tool in children with neuromotor disorders. This is the first study presenting the reliability, for all 3 levels of the Hypertonia Assessment Tool: (1) the 7 items, (2) the 3 subtypes, and (3) the Hypertonia Assessment Tool diagnosis.

The interrater reliability of the subtypes was in line with the original studies of Jethwa and Knights (compare Appendix B: bias-adjusted Kappa, Supplementary Material).[6,7] Reliability for dystonia was even higher in our study than observed by Jethwa et al.[6] On item level, items 4 (presence of a spastic catch) and 6 (increased tone with movement of another body part) showed slight to fair agreement as indicated by the Kappa values. Part

Table 3. Validity Values for Hypertonia Assessment Tool (HAT) Subtypes Dystonia and Spasticity.[a]

		Left side				Right side			
Limb	Hypertonia	PPV	NPV	McNemar	RD	PPV	NPV	McNemar	RD
Upper limb	Dystonia	0.27	0.83	0.01	0.26	0.25	0.85	0.02	0.24
	Spasticity	0.79	0.68	1.00	0.00	0.68	0.89	0.51	0.07
Lower limb	Dystonia	0.30	0.83	0.01	0.26	0.27	0.83	0.01	0.26
	Spasticity	0.79	0.67	0.55	0.07	0.78	0.74	1.00	0.02

Abbreviations: NPV, negative predictive value; PPV, positive predictive value; RD, risk difference.
[a]Positive predictive value, negative predictive value, McNemar test, and risk difference of the HAT subtype spasticity and dystonia were evaluated by the HAT (first measurement) according to signs of hypertonia of the clinical diagnosis; all children n = 46.

of the poor agreement could be due to the calculation of the Kappa statistic, as the Gwet's AC1 values were mostly higher, but still did not exceed moderate levels of agreement. Another explanation could be the subjective interpretation of the spastic catch. Per definition, spasticity should be a clinically observable phenomenon that responds the same degree to a passive movement applied in a particular velocity.[18] But in practice, it has been shown that the interpretation of spasticity varies between testers.[19,20] In a recent study that investigated the reliability of the Australian Spasticity Scale in 22 children with CP (age 1-16 years, 15 children with Gross Motor Function Classification System level I), the interrater reliability was found to be almost perfect for the lower and upper limbs.[21] In our study, we found a poorer reliability. In our study, more children with moderate to severe motor impairments were included. We assume that recognizing a spastic catch might be more difficult in children who are more severely affected, and this might have negatively affected our reliability findings. On the level of the Hypertonia Assessment Tool subtypes, interrater agreement was moderate to substantial.

The intrarater reliability values were higher than the interrater reliability values and varied from substantial to almost perfect for the subtypes. These results indicate that the intersession time of 1 week seems to influence the results less than the scoring of another tester (who performed the assessment immediately after the first assessment). In 3 children with cognitive impairments, items 2 and 6 could not be tested. To keep the Hypertonia Assessment Tool applicable for these children as well, we recommend not deleting item 1 out of the assessment, unlike the recommendation of Knights et al.[7]

The validity of the subtype spasticity seems given, as its presence was consistent with the clinical diagnosis provided by the physician. The prevalence of the subtype dystonia was 24% to 26% higher with the Hypertonia Assessment Tool compared to the clinical diagnosis. It seems to be an underrepresentation in the diagnosis or an overrepresentation of dystonia in the Hypertonia Assessment Tool. Dystonia often occurs in combination with spasticity. As spasticity might mask the symptoms of dystonia, dystonia may remain undetected during clinical routine examinations. By testing with a specific measurement tool like the Hypertonia Assessment Tool, the symptoms are more systematically evaluated. Sanger stated that "it is likely that many children with a primarily spastic clinical picture also have some degree of dystonia" and "dystonia is maybe underreported in children with movement disorders.[18] Gordon et al[21] found in their study that many children diagnosed with spastic cerebral palsy show elements of dystonia too.

We recommend reporting Hypertonia Assessment Tool subtypes for clinical documentation rather than the Hypertonia Assessment Tool diagnoses. On the one hand, the Hypertonia Assessment Tool subtypes showed best inter- and intrarater reliability. On the other hand, clinically relevant information gets lost when solely reporting the Hypertonia Assessment Tool diagnosis. For example, in a child diagnosed as having a mixed diagnosis, it is unclear what or how many hypertonia types the child has. On the German rating sheet, we decided to report only the presence or absence of the 3 types of hypertonia and not the Hypertonia Assessment Tool diagnosis.

Limitations of our study were that for both testers the Hypertonia Assessment Tool was a relatively new assessment tool. Despite their experience in assessing spasticity in children, we expect improved reliability with further training and exchange between Hypertonia Assessment Tool users. Furthermore, interrater reliability might have been poorer if more than 2 raters had performed the Hypertonia Assessment Tool assessments. Also, although we tried to control for external factors that might influence the results (eg, we tested the children at the same time of day in their room), internal factors like mood or distraction were hard to standardize. In a further step, it would be interesting to evaluate if the Hypertonia Assessment Tool would be sensitive enough to detect changes in hypertonia symptoms, for instance, a reduction in symptoms reflecting spasticity after a dorsal selective rhizotomy.

Finally, one methodologic consideration would be the statistical analysis. Especially for the interrater reliability analysis, Gwet's AC1 exceeded Kappa values (except for a few items), showing that Kappa values were influenced by an imbalance between the presence and absence of signs of hypertonia. A huge discrepancy was observed for the rigidity items and subscales, where reliability could not be calculated with Kappa, while Gwet's AC1 showed excellent agreement values. For future (interrater) reliability studies of the Hypertonia Assessment Tool, Gwet's AC1 might be the preferred statistical analysis.

Conclusion

We could show that the reliability of the German Hypertonia Assessment Tool version is comparable to the original one. The low agreement of spasticity item 4 between the 2 testers

indicates the issue of subjective rating of signs of a spastic catch. To enhance the psychometric properties, we recommend improving the standardization of the test execution (eg, hand positioning) and provide training to assessors. For professionals working in pediatric rehabilitation, the Hypertonia Assessment Tool offers a rather well-described assessment tool to distinguish between hypertonia types in children with neuromotor disorders.

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Author Contributions

PM collaborated in designing the study, translated the Hypertonia Assessment Tool, tested participants, performed the analyses, and interpreted the results. PM wrote the first draft of the manuscript and incorporated the feedback from the other authors. VFW collaborated in designing the study, translated the Hypertonia Assessment Tool, tested participants, and assisted with writing the manuscript. JB assisted with the translation process, recruited participants, and interpreted the results. HvH provided senior mentorship in designing the study, advised with statistical analyses and interpretation of results, and provided feedback on the manuscript.

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Ethical Approval

The study was approved by the ethics committee of the Canton of Zurich, complied with the Declaration of Helsinki, and followed the guidelines of good clinical practice. All children (and caregivers) agreed to participate. Parents and adolescents aged 15 years and older signed the informed consent form.

Supplemental Material

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