

1 **Research paper**

2 **Title:** Intra and inter-tester reliability of spasticity assessment in standing position in
3 children and adolescents with cerebral palsy using a paediatric exoskeleton

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29

30 **Running title:** Reliability of the Lokomat spasticity assessment.

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34

35 **Abstract (244)**

36 *Background:* The L-STIFF tool of the Lokomat evaluate the hip and knee flexors and
37 extensors spasticity in a standing position. It moves the lower limb at a controlled
38 velocity, measuring joint resistance to passive movements. Since its reliability in children
39 with cerebral palsy remains unknown, our goal was to evaluate the relative and absolute
40 reliability of L-STIFF in children with cerebral palsy.

41 *Methods:* Reliability was determined in 16 children with cerebral palsy by two
42 experienced therapists. The changes in resistive torque in hip and knee in both flexion
43 and extension were measured. Relative and absolute reliability were estimated using the
44 intra-class correlation coefficient, standard error of measurement and minimal detectable
45 change. Reliability was assessed on three levels: (1) intra- and (2) inter-tester within
46 session, and (3) intra-tester between sessions.

47 *Results:* Intra-class correlation coefficients were moderate to excellent for intra-tester
48 reliability (all $p \leq 0.01$). The standard error of measurement ranged from 0.005 to
49 0.021 Nm/° (i.e., 7 to 16%) and minimal detectable change from 0.014 to 0.059 Nm/°.
50 Inter-tester Intra-class correlation coefficients ranged from 0.32 to 0.70 (all $p \leq 0.01$),
51 standard error of measurement from 0.012 to 0.029 Nm/° (i.e., 6 to 39 %) and minimal
52 detectable change from 0.033 to 0.082 Nm/°. L-STIFF reliability was better during fast
53 and medium movement speeds compared to slow speeds.

54 *Conclusions:* The assessment tool L-STIFF is a promising tool for quantifying lower limb
55 spasticity in children with cerebral palsy in a standing position. However, the results
56 should be interpreted carefully.

57 **Keywords:** Psychometric properties, Muscle tone, Mobility, Cerebral palsy, Lokomat

58 1. Introduction

59 Spasticity is the most common motor disorder in children with cerebral palsy (CP)
60 with a prevalence of 80% to 90%. Spasticity is an involuntary muscle contraction
61 following sensory input induced by muscle stretch and it has been characterized as a
62 velocity-dependent phenomenon [1]. Spasticity results in a greater than normal resistance
63 to externally imposed movements, and this resistance increases with velocity [1,2].
64 Although spasticity is of neural origin [1], there is recent evidence that spasticity involves
65 a structural and functional changes in skeletal muscle [3–5]. It hampers normal muscle
66 lengthening during growth and is thus can contribute to the development of secondary
67 muscle and soft tissue contractures and to skeletal deformation [5–7]. These muscle
68 contractures and skeletal deformities may result in lever arms modifications, which alter
69 joint moments during walking [8].

70 Characterization and quantification of muscle spasticity are important to plan
71 medical or surgery interventions (e.g., repetitive passive mobilization, injection of
72 botulinum toxin or selective dorsal rizhotomy). To do so, reliable clinical tests are
73 required for daily clinical monitoring of muscle spasticity [10,11]. Modified Ashworth
74 Scale (MAS) and Modified Tardieu Scale (MTS) are frequently used in clinical practice
75 to assess muscle response to a quick passive stretch applied by the therapist. The MAS is
76 a 6-point ordinal scale qualifying the resistance of muscles to passive movement [12].
77 Using a single speed measure, the MAS tends to cluster muscles into broad severity
78 categories, thereby limiting its ability to detect response to treatment [13]. The MTS was
79 developed to qualify the resistance to passive stretching at three velocities also on a 6-
80 point ordinal rating scale. The therapist measures two resulting joint angles using a

81 goniometer: the angle of catch at which a muscle response (stretch reflex) is provoked by
82 the fast velocity stretch (R1) and the angle of full passive range of motion obtained with
83 low speed stretching (R2). The difference between these two values (R2-R1) is thought to
84 describe the level of dynamic contracture at the joint [14,15]. However, this assessment is
85 affected by the difficulty of controlling the speed movement during the testing as well as
86 the experience of the examiner [16]. Moreover, several studies pointed out the low inter-
87 tester reliability of the MAS and MTS in children with CP [17]. The above-cited
88 limitations of these clinical tests result in a misestimation of spasticity. Finally, due to the
89 discrepancies between spasticity manifestation in sitting or lying position and how
90 spasticity may contribute to walking limitations, the transfer of clinical assessment to
91 functional limitations remains uncertain [16]. Complementary methods are needed to
92 improve the reliability of spasticity measurements and to assess spasticity in a
93 standardized and functional position.

94 The Lokomat, the most used robotic gait trainer, provides isometric strength (L-
95 Force) and spasticity (L-STIFF) assessment tools [18,19]. L-STIFF tool measures the
96 changes in resistive torque in hip and knee in both flexion and extension during
97 predefined passive movements by moving the joint at a constant velocity. With L-STIFF
98 tools, both the range of motion and the velocity of the movement are controlled. L-STIFF
99 offers continuous data and thus a wider range of possible outcomes which is more
100 adequate for longitudinal follow-ups [20]. Moreover, apart from the patient setup which
101 depends on the examiner, L-STIFF testing execution is not affected by variations usually
102 observed among examiners [18]. For its clinical validation, L-STIFF measurement at
103 different velocity tool was first compared to MAS on patients with central nervous

104 lesions [18,21]. The MAS score correlated with L-STIFF measurement at the highest
105 velocity, only [18,21]. A high stiffness value is assumed to correspond to strong
106 spasticity [21,22]. Afterwards, Schmartz et al., [23] evaluated the intra-tester reliability of
107 L-STIFF in children with CP (n=9) and highlighted excellent reliability (intraclass
108 correlation coefficients (ICCs) = 0.83–0.97). However, studies with larger sample size
109 and additional psychometric properties (i.e., inter-session and inter-tester reliability) are
110 required for the clinical use of L-STIFF. In particular, inter-tester and inter-session
111 reliability are needed for daily clinical follow-up of spasticity. The aim of the present
112 study was to assess intra-and inter-tester reliability of L-STIFF.

113

114 2. Material and methods

115 2.1. Participants

116 The sample size estimation was based on a significance level of 0.05, a power of 0.80,
117 and an ICC-value between 0.60 (fair) and 0.90 (excellent) for both intra and inter-tester
118 reliability analysis. The minimal sample size was 15 for this reliability analysis. In the
119 present study, 16 children and adolescents with CP (mean \pm standard deviation, age:
120 10 ± 3 years (range: 7–19 years), mass: 30 ± 9 kg, height: 131 ± 9 cm, 7 Females and 9
121 Males) were included. Participants had a Gross Motor Function Classification System
122 (GMFCS) level II ($n = 10$) and III ($n = 6$). The inclusion criteria were: (1) children and
123 adolescents with a diagnostic of spastic CP, (2) ability to communicate fear, discomfort
124 or pain, as evaluated by the physiotherapist, (3) understanding simple instructions and,
125 (4) a femur length of 23-35 cm (to fit in Lokomat pediatric orthosis). Children were
126 excluded if they had received: (1) spasticity medication before the assessment, (2)
127 botulinum toxin injections six months prior to the assessment and, (3) an intrathecal
128 baclofen pump or a surgical intervention during the last 12 months. This study was
129 approved by the Research Ethics Board of UHC Sainte-Justine.

130 2.2. Reliability assessments

131 Hip and knee spasticity were assessed by L-STIFF tool using the pediatric version of
132 Lokomat Pro (Hocoma AG, Volketswil, Switzerland). Technical utilization of L-STIFF
133 tool has been previously described [18,21]. Briefly, the participant is lifted above the
134 treadmill (unloading from 100% of his body weight) to be able to freely move hip and
135 knee joints without touching the ground with the feet (see Figure 1).

136

**** Insert **Figure 1******

137

138 Then, the Lokomat performed a controlled displacement of each of the four actuated
139 joints (i.e., left and right hip and knee) in flexion and extension at three velocities
140 (22.5°/s, 45°/s and 90°/s for the hip and 30°/s, 60°/s and 120°/s for the knee). The range
141 of motion was set according to patient capacity and can be reduced for patients with
142 contractures or range of motion limitations. Maximal range of motion is fixed at 46° in
143 the hip and at 55° in the knee joint. The protocol was divided into two measurement
144 sessions to assess inter and intra-tester reliability (see Figure 2).

145

146

**** Insert **Figure 2******

147

148 *Stage 1: Inter-tester & Intra-tester within same session reliability*

149 Each participant was tested independently by two experienced Lokomat users (GG and
150 YC) on the same day. To ensure proper reliability assessment, the order of the two testers
151 was randomized and each tester was blinded to the results obtained by the other. Each
152 participant accomplished a familiarization trial of L-STIFF test followed by two real
153 tests. Participants were asked to relax completely and to avoid any voluntary muscle
154 contraction during the test. During each test, the instruction "3-2-1-Go" was given
155 verbally by the tester. A 2-min rest was given between the two blocks of tests. During the
156 assessments, we did not make noise or talk to keep the patient as relaxed as possible
157 during the test. Then, the participant was taken out of the Lokomat and had a short
158 (15 min) break period. Thereafter, the second tester reinstalled the participant into the

159 Lokomat and repeated the same procedure. Measures acquired during this stage were also
160 used to assess *intra-tester* within same session reliability for each tester since each tester
161 measured the torque twice.

162 *Stage 2: Intra-tester (between days) reliability*

163 In the second part of the protocol, *intra-tester reliability* (between days) was conducted
164 by the tester GG a week later (see Figure 1). Participant was installed into the Lokomat
165 by tester GG who used the same anthropometric settings as in stage 1. Then, participant
166 performed two L-STIFF trials. Overall, L-STIFF tool took about 2.5 min to measure hip
167 and knee flexion and extension torques.

168 **2.3. Statistical analysis**

169 An analysis of variance (ANOVA)-based ICC was used to evaluate the relative reliability
170 of L-STIFF measurements at each measurement speed. To test reliability, ICCs with 95%
171 confidence intervals (two-way random-effect models) were used. ICC scores were
172 compared with the following scale for interpretation of correlation: excellent (1.00 –
173 0.81), good (0.80 – 0.60), moderate (0.41 – 0.60), fair (0.21 – 0.40) and poor (< 0.20)
174 [24]. Standard error of measurement (SEM) and the 95% CI of the minimal detectable
175 change (MDC) were calculated to evaluate L-Stiff absolute reliability [25]. The ICC
176 reflects the degree of consistency of a measurement. The SEM and MDC provide
177 information about the expected trial-to-trial measurement error [26]. As it is recognized
178 that there is a large inter-limb strength difference in children with CP, statistical analysis
179 considered each side independently for all the tests (n=2x16), as done in previous studies
180 [27,28]. The level of significance was set to p -value < 0.05. All statistical analysis was
181 performed with SPSS (version 25.0, IBM SPSS Incorporated, Chicago, IL, USA).

182 **3. Results**

183 *Intra-tester reliability*

184 The median, minimum and maximum values and first and third quartiles of measured hip
185 and knee torques are reported in **Figure 3**. For *intra-tester* (same session), ICCs varied
186 from 0.69 to 0.95, SEM from 0.002 to 0.007, and MDC from 0.009 to 0.021 Nm/° (see
187 Table 1). For *intra-tester* reliability (between days), ICCs ranged from 0.49 to 0.89 (all
188 $p \leq 0.01$), while SEM varied from 0.005 to 0.021 Nm/° (*i.e.*, 7 to 16%) and MDC from
189 0.014 to 0.059 Nm/°. The highest SEM (0.005 to 0.021 Nm/°) and MDC (0.025 to
190 0.059 Nm/°) values were observed at fast velocity (see Table 2).

191

192 **** Insert **Figure 3******

193

194 **Table 1:** Intra-tester (same-session) reliability of L-STIFF measurements in hip and knee flexion and extension (all $p \leq 0.01$)

	ICC			SEM (Nm/°)			MDC (Nm/°)		
	Slow	Medium	Fast	Slow	Medium	Fast	Slow	Medium	Fast
Hip flexion	0.86	0.95	0.92	0.006 (4%)	0.003 (2%)	0.003 (2%)	0.017	0.008	0.009
Hip extension	0.92	0.93	0.95	0.004 (2%)	0.003 (2%)	0.002 (1%)	0.010	0.009	0.006
Knee flexion	0.77	0.82	0.83	0.006 (15%)	0.004 (9%)	0.004 (6%)	0.016	0.0100	0.012
Knee extension	0.87	0.89	0.69	0.004 (8%)	0.003 (6%)	0.007 (9%)	0.011	0.008	0.021

195 Abbreviation: ICC: Intraclass correlation coefficient; SEM: standard error of measurement in units of measure and in percentage (%);
 196 MDC: minimal detectable change.

197

198 **Table 2:** Intra-tester (between days) reliability of L-STIFF measurements in hip and knee flexion and extension (all $p \leq 0.01$)

	ICC			SEM (Nm/°)			MDC (Nm/°)		
	Slow	Medium	Fast	Slow	Medium	Fast	Slow	Medium	Fast
Hip flexion	0.65	0.63	0.58	0.015 (8%)	0.018 (9%)	0.021(10%)	0.042	0.049	0.059
Hip extension	0.49	0.66	0.62	0.021 (11%)	0.015 (8%)	0.017 (8%)	0.059	0.042	0.047
Knee flexion	0.89	0.84	0.84	0.005 (7%)	0.008 (11%)	0.010 (10%)	0.014	0.024	0.029
Knee extension	0.70	0.89	0.88	0.015 (16%)	0.007 (7%)	0.009 (8%)	0.041	0.018	0.025

199 Abbreviation: ICC: Intraclass correlation coefficient; SEM: standard error of measurement in units of measure and in percentage (%);
 200 MDC: minimal detectable change.

201

Inter-tester reliability

202 Inter-tester ICCs ranged from 0.32 to 0.70 (all $p \leq 0.01$) and SEM from 0.012 to
203 0.029 Nm° (*i.e.*, 6 to 39%). For the hip flexors and extensors, fair to moderate inter-
204 tester reliability (ICCs =0.32 – 0.46) was found for movement with slow velocity,
205 whereas moderate to good reliability was found with peak torque measurements at fast
206 velocity (ICCs=0.68 – 0.70). Concerning the knee flexors and extensors, ICCs were
207 moderate for all speeds ranging from 0.43 to 0.55. The highest SEM values were
208 observed at slow velocities for knee flexion and extension (34 and 39%, respectively).
209 Minimal detectable changes ranged from 0.033 to 0.082 Nm° with highest values again
210 observed at slow velocity (see Table 3).

211 **Table 3:** Inter-tester reliability of L-STIFF measurements in hip and knee flexion and extension (all $p < 0.05$)

	ICC			SEM (Nm/°)			MDC (Nm/°)		
	Slow	Medium	Fast	Slow	Medium	Fast	Slow	Medium	Fast
Hip flexion	0.46	0.49	0.68	0.028 (16%)	0.028 (16%)	0.016 (8%)	0.079	0.078	0.043
Hip extension	0.32	0.60	0.70	0.027 (15%)	0.017 (9%)	0.012 (6%)	0.074	0.047	0.033
Knee flexion	0.55	0.50	0.47	0.020 (34%)	0.020 (33%)	0.023 (26%)	0.057	0.054	0.063
Knee extension	0.48	0.54	0.43	0.029 (39%)	0.020 (27%)	0.026 (27%)	0.082	0.054	0.072

212 Abbreviation: ICC: Intraclass correlation coefficient; SEM: standard error of measurement in units of measure and in percentage (%);

213 MDC: minimal detectable change.

214 4. Discussion

215 The present study aimed to evaluate the reliability of L-STIFF tool to assess hip
216 and knee spasticity in a standing position in a group of children and adolescents with
217 spastic CP (GMFCS level II and III). Our main findings were that: **(1)** the most reliable
218 measurement using L-STIFF tool was intra-tester within the same session, followed by
219 intra-tester between sessions, and then inter-tester, and **(2)** L-STIFF reliability was higher
220 during fast and medium movement speeds compared to slow speeds.

221 *L-STIFF relative reliability*

222 In a feasibility study (n=9), Schmartz et al., [23] reported good intra-tester
223 reliability of L-STIFF (ICCs=0.83 – 0.97) in children with CP assessed by one tester in
224 only one session. Our intra-tester ICC values (n=16; same session; ICCs=0.69 – 0.95)
225 were in good agreement with those of Schmartz et al., [23] except for the knee extension
226 at fast speed for which our ICC was slightly lower (ICC=0.69). This reliability
227 differences could be due to the between-participants variability within the study samples
228 (GMFCS II to V in [23] vs. GMFCS II and III in our study), as ICC score is strongly
229 dependent on the between participant variability [29]. More importantly, L-STIFF intra-
230 tester reliability (between days) was overall better (ICCs=0.49 – 0.89) than those found
231 with the MAS (ICCs=0.21 – 0.82) and MTS (ICCs=0.38 – 0.93) in a sample of 18
232 children with CP (GMFCS level I to V) [30]. When measured at different days by the
233 same tester and with the same settings, intra-tester ICCs were good to excellent for the
234 knee flexors and extensors measurements (ICCs=0.70 – 0.89) and moderate to good for
235 hip flexors and extensors (ICCs=0.49 – 0.66). Considering the results of Yam et al., [31],
236 again, inter-tester reliability of L-STIFF (ICCs=0.32 – 0.70) was better than MAS

237 (ICCs=0.27 – 0.56) but slightly lower than the MTS (ICCs=0.55 – 0.74). Compared to a
238 sample of 17 children with CP (GMFCS levels II and III) more similar to ours [31], L-
239 STIFF inter-tester reliability was slightly better than the reliability found with the MTS
240 (ICCs=0.22 – 0.71). Overall, L-STIFF relative reliability is better than MAS and MTS.

241 *Sources of variability and errors*

242 While the L-STIFF execution relies on the machine only, the patient setup into the
243 Lokomat depends on the assessor. For that reason, the L-STIFF intra-tester (within and
244 between sessions) reliability was generally higher than the inter-tester values. As for the
245 isokinetic dynamometer [32], a source of error in L-STIFF measurements could mainly
246 come from the misalignment of axes between participant joints and the Lokomat motors.
247 This point is generally underlined and is the object of recommendations in several studies
248 regarding the use of the isokinetic dynamometer [32,33]. Although the Lokomat is multi-
249 articular, this recommendation is even more applicable for L-STIFF tool. A special care
250 must be given to the installation and alignment of the child into the Lokomat to minimize
251 erroneous spasticity measurement. Above all, standardization and training should be
252 further improved to reduce extrinsic error when different testers perform the
253 measurement. In addition to error due to patient setup, the variability of L-STIFF
254 measurements can be due to intrinsic factors (*e.g.*, abnormal muscle tone change, muscle
255 voluntary contraction). It could also come from the repetitive movement. In particular,
256 multiple studies have highlighted a decrease in resistive torque in patients with spasticity
257 after repeated passive movements by using an isokinetic dynamometer [34,35]. By
258 performing eight passive movements four times at three different speeds over a span of
259 1 hour, muscle stretching or reflex habituation may have occurred. Thence, the absolute

260 reliability should also be investigated and considered in the interpretation of results of L-
261 STIFF.

262 *L-STIFF absolute reliability*

263 Although *relative* reliability addresses the degree to which subjects keep their
264 rank in a sample through repeated measures, *absolute* reliability (*i.e.*, SEM and MDC)
265 informs about the measurement errors in absolute values and the limit for the smallest
266 change that indicates a real clinical improvement following an intervention. Overall, 84%
267 of our SEM values were smaller than 20% of the grand mean of measurements (all intra-
268 tester SEM values are <20%). From a clinical point of view [36], our SEM values seem
269 reasonable and confirm that L-STIFF measurements can be used to detect real changes
270 for a group of children with CP. Our absolute SEM values were overall smaller than
271 Schmartz et al.' results [23], which is probably due to the severity of impairment that was
272 more important in their group of children (including children with GMFCS IV and V). In
273 regards to the effect of anti-spastic treatment reported in literature, L-STIFF
274 measurements can be deemed sensitive enough to differentiate between pre and post
275 treatment interventions [13,23].

276 *Clinical implications*

277 Spasticity clinical scales provided a non-specific and non-controlled testing
278 velocity, which is problematic to evaluate a velocity-dependent mechanism. [21,31]. The
279 advantage of L-STIFF is to assess spasticity at three controlled movement velocities in a
280 standing position. When walking, children with CP move their lower limb joints at
281 various speeds [38,39]. The slow (22.5°/s), medium (45°/s) and fast (90°/s) velocities

282 used in L-STIFF measurement at the hip correspond to values measured at this joint
283 during terminal swing and the pre-swing phase of gait. Such matching velocities may
284 better inform on spasticity implications during gait [16]. Regarding the knee
285 measurements, L-STIFF testing velocities (30, 60 and 120°/s) were overall slower than
286 knee angular velocities during walking [38,40]. Tuzson et al., [41] reported a peak knee
287 angular velocities during fast walking ranged from 100° to 450°/s in patients with CP and
288 were significantly slower than the knee angular velocities of typically developed
289 participants. In the same study, the authors determined spastic velocity thresholds of the
290 knee joint via an isokinetic dynamometer. Knee spastic threshold velocity was on average
291 $175^{\circ}/s \pm 63.8^{\circ}/s$ [41]. Later, Damiano et al., [40] reported a moderate correlation between
292 the assessment of hamstring and quadriceps stiffness at 120°/s and maximum extension
293 velocity ($r = -0.47$ and -0.41 , respectively) and maximum flexion velocity ($r = -0.50$ and $-$
294 0.39 , respectively) during walking at comfortable speed. The results of these studies
295 imply that 120°/s may probably be too low to elicit spastic responses, particularly in
296 patients who only have moderate spasticity [40–42]. L-STIFF testing velocities in the
297 knee are probably more suitable for children with more pronounced spasticity. Finally, L-
298 STIFF allows evaluating spasticity at three different velocities and in standing position, it
299 could be used to refine the relationship between lower limb spasticity and joint
300 biomechanics during walking.

301 *Study limitations*

302 This study had some limitations that need to be considered when interpreting the
303 results. Firstly, the results are valid for children and adolescents with CP with GMFCS
304 levels II and III which limit the results generalizability but correspond to the children and

305 adolescents with CP who benefit the most of gait rehabilitation with the Lokomat.
306 Secondly, cognitive impairment was not formally assessed, while it could be a source of
307 variability when assessing motor function in people with CP [43]. Fourthly, limited
308 intrinsic/extrinsic errors may be introduced by repeated stretch repetitions and the
309 learning effect of the measurement routine. Fifthly, L-STIFF quantifies only the
310 spasticity of hip and knee flexors and extensors while spasticity can affect various lower
311 limb muscles (e.g., hip adductors, plantar-flexors) in children and adolescents with CP.

312 **5. Conclusion**

313 Based on the outcomes of the present reliability study, L-STIFF measurements were
314 more reliable when the same tester, rather than different testers performed the
315 assessment. When different testers perform the measurement, standardization and
316 training should be improved to minimize the extrinsic error as much as possible.
317 Moreover, our SEM and MDC should be systematically used for longitudinal follow-up
318 of spasticity to determine if the change exceeds uncertainties or not. On the whole, L-
319 STIFF of the Lokomat is a promising tool for evaluating lower limb spasticity in children
320 and adolescents with CP in a more functional posture than traditional clinical scales.

321 **Disclosure of interest**

322 The authors declare that they have no competing interest.

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447 **Figure captions :**

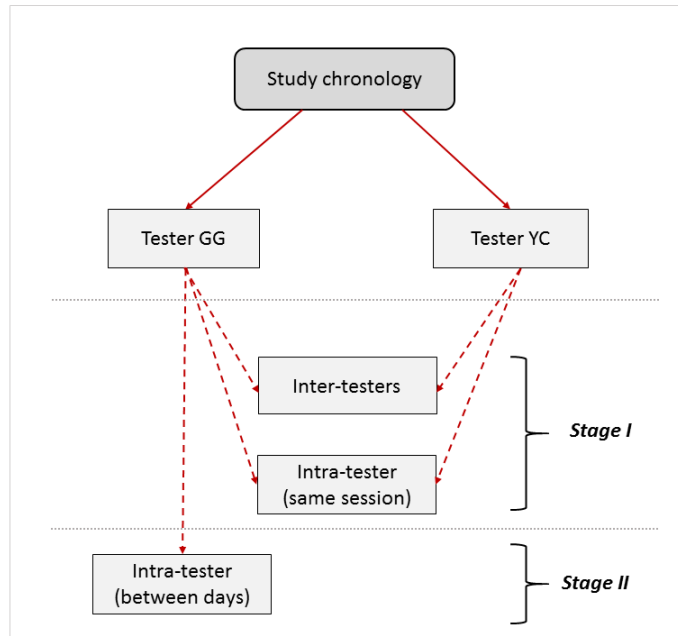


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449 **Figure 1:** Measuring position used for the spasticity assessment using L-STIFF tool.

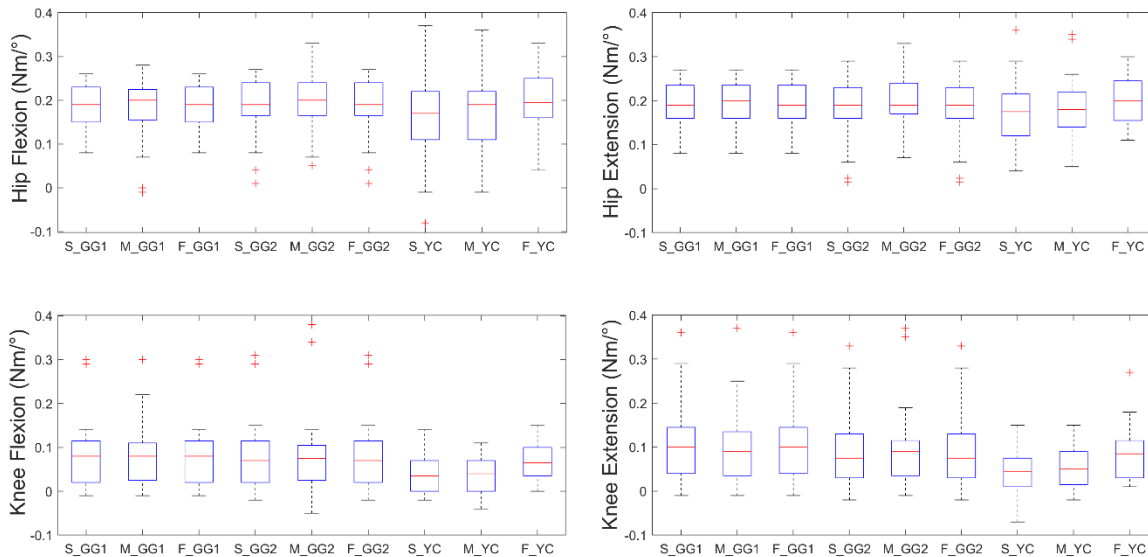
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Figure 2: Schematic illustrating the three aspects of reliability (Inter-tester, intra-tester same session and intra-tester between days) evaluated during the present study.



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Figure 3: L-STIFF measurements box plots with median, minimum and maximum values and 25 and 75 percentiles from tester GG at day1 (GG1), tester GG at day2 (GG2) and tester YC. [Slow (S), Medium (M) and Fast (F)]. The plus signs (in red) represent the extreme values (the outliers).

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