

SUPPLEMENTARY DIGITAL MATERIAL 4

Supplementary Table III.—Measurement properties of the included studies in Charcot Marie Tooth disease: validity.

Articles	Walking test	Type of variables studied	Validity					
			Type of validity	Compared to	Results (95% CI)	COSMIN	Criterion quality and/or criteria rating	Hypotheses testing for construct validity
Padua <i>et al.</i>, 2016⁴⁹	6MWT	6MWD	Construct	(i) 10MWT (ii) CMTNS (iii) plantar flexion as detected by a myometer (iv) dorsi flexion as detected by a myometer	(i) $\rho = -0.54$ (P<0.001) (ii) $\rho = -0.55$ (P<0.001) (iii) $\rho = 0.45$ (P<0.001) (iv) $\rho = 0.49$ (P<0.001)	Very good	NA	?
Mori <i>et al.</i>, 2018⁵²			Construct	(1) CMTNS (2) dorsal and (3) plantar-flexor muscles strength	(1) $\rho=0.11$; P=0.52 (2) $\rho=0.32$; P=0.051 (3) $\rho=0.004$; P=0.98 and correlation between 6MWT and 10MWT: $\rho=-0.63$; P<0,001	Very good	NA	?
Pazzaglia <i>et al.</i>, 2019⁵³			NA	NA	NA	NA	NA	NA
Mori <i>et al.</i>, 2018⁵²	10MWT	10MWT time	Construct	(1) CMTNS (2)dorsal and (3) plantar-flexor muscles strength	(1) 0.26; P=0.084 (2) $\rho = -0.50$; P=0.0017 (3) $\rho=-0.34$; P=0.027	Very good	NA	?
Solari <i>et al.</i>, 2007⁴⁶			NA	NA	NA	NA	NA	NA
Pazzaglia <i>et al.</i>, 2019⁵³			NA	NA	NA	NA	NA	NA
Coghe <i>et al.</i>, 2020⁵¹		(i) velocity (ii) step width (iii) stride length (iv) DST (v) kinematic data	Construct	CMTNS2	(i) $\rho = -0.783$ (P=0.001) (ii) $\rho = -0.248$ (P=0.306) (iii) $\rho=-0.776$ (P<0.001) (iv) $\rho=0.523$ (P=0.022) (v) ρ from -0.004 (p=0.986) for Hip Abduction–adduction to 0.832 (P<0.001) for Knee Flexion–extension	Very good	NA	+
Pazzaglia <i>et al.</i>, 2019⁵²	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)	NA	NA	NA	NA	NA	NA
Padua <i>et al.</i>, 2016⁴⁹	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)	Construct	(i) 10MWT (ii) CMTNS (iii) plantar flexion as detected by a myometer	(i) ρ from -0.08 to 0.10 (P>0.05) (ii) ρ from -0.31 to 0.07 (the most of correlation)	Very good	NA	?

					(iv) dorsi flexion as detected by a myometer	with P>0.05)	(iii) ρ from -0.20 to 0.40 (the most of correlation with P>0.05)	(iv) ρ from -0.23 to 0.35(the most of correlation with P>0.05)			
Ferrarin <i>et al.</i>, 2011⁴⁷	NA (Motion analysis)	(i) CS (1) velocity and (2) double support (3) Mean ankle angle TW– mean ankle angle HW (4) cadence (5)stride length normalized to body height (6) kinematics data (7) kinetics data (ii) RS (1) velocity and (2) double support (3) cadence (4) stride length normalized to body height (5) kinematics data (6) kinetics data	NA	NA	NA	NA	NA	NA	NA	NA	
Guillebastre <i>et al.</i>, 2013⁴⁸	NA (GAITRite of 8.3m)	(1) Velocity (2) double support	Construct	MRC scores (i) dorsal (ii) plantar-flexor muscles	(1) (i) $\rho = 0.49, P<0.05$ (ii) $\rho = 0.50, P<0.05$ (2) (i) $\rho = -0.19, P>0.05$ (ii) $\rho = -0.04, P>0.05$	Very good	NA	+			
Lencioni <i>et al.</i>, 2017⁵⁰	NA (Motion analysis)	(1) Velocity normalized to body height (2) stride length normalized to body height (3) kinematics data (4) kinetics data	NA	NA	NA	NA	NA	NA	NA	NA	

CMT: Charcot Marie Tooth; CMTNS: Charcot-Marie-Tooth Neuropathy Score; HW: Heel Walking; GC: Gait Cycle; ICC: Intercorrelation coefficient; NA: Non Applicable; RS: rapid speed; RSME: Root-mean-square error; SD: Standard Deviation; SEM: Standard Error Measurement; SL: Stride Length; SRM: Standardised Response Mean; ST: Step Test; SWT: Swing Time; TW: Toe Walking; 6MWD: 6-minute walking distance; 6MWT: 6-minute walk test; 10MWT: 10-minute walk test.

Supplementary Table IV.—Measurement properties of the included studies in Charcot Marie Tooth disease: reliability.

Articles	Walking test	Type of variables studied	Reliability		COSMIN*	Quality criteria rating
			Design	Results (95% CI)		
Padua et al., 2016 ⁴⁹ Mori et al., 2018 ⁵² Pazzaglia et al., 2019 ⁵³	6MWT	6MWD	Test-retest	ICCs >0.9	adequate	+
			NA	NA	NA	NA
			NA	NA	NA	NA
Mori et al., 2018 ⁵² Solari et al., 2007 ⁴⁶ Pazzaglia et al., 2019 ⁵³ Coghe et al. 2020 ⁵¹	10MWT	10MWT time (i) velocity (ii) step width (iii) stride length (iv) DST (v) kinematic data	NA	NA	NA	NA
			(i)Inter-rater Intra-rater	(ii) 10MWT (i)ICC=0.97, 95% CI [0.88–0.99]; (ii)ICC=0.96, 95% CI [0.87–0.99]	very good	+
			NA	NA	NA	NA
Pazzaglia et al., 2019 ⁵³	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)	NA	NA	NA	NA
Padua et al., 2016 ⁴⁹	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)		ICCs >0.9	NA	NA
Ferrarin et al., 2011 ⁴⁷	NA (Motion analysis)	(i) CS (1) velocity and (2) double support (3) Mean ankle angle TW– mean ankle angle HW (4) cadence (5)stride length normalized to body height (6) kinematics data (7) kinetics data (ii) RS (1) velocity and (2) double support (3) cadence (4) stride length normalized to body height (5) kinematics data (6) kinetics data	Test-retest (4–6 weeks apart)	(i) (1) ICC=0.95, (2)ICC=0.93 (3) ICC = 0.95 (4) ICC=0.91 (5) ICC=0.96 (6) All variables have ICC > 0.7 except Trunk ROM in sagittal plane (ICC=0.22) and in transverse plane (ICC=0.34) (7) All variables have ICC > 0.8 (ii) (1) ICC=0.78 (2) ICC = 0.72 (3) ICC=0.71 (4) ICC = 0.78 (5) All variables have ICC > 0.7 except Trunk ROM in sagittal plane (ICC=0.27) and in frontal plane (ICC=-0.04) and transverse plane (ICC=-0.49)(6) All variables have ICC > 0.8 except hip positive mechanical work (ICC=0.74)	very good	+

Guillebastre et al., 2013 ⁴⁸	NA (GAITRite of 8.3m)	(1) Velocity (2) double support	NA	NA	NA	NA
Lencioni et al., 2017 ⁵⁰	NA (Motion analysis)	(1) Velocity normalized to body height (2) stride length normalized to body height (3) kinematics data (4) kinetics data	NA	NA	NA	NA

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Supplementary Table V.—Measurement properties of the included studies in Charcot Marie Tooth disease: measurement error, responsiveness, and feasibility.

Articles	Walking test	Type of variables studied	Measurement error			Responsiveness			Feasibility	
			Results (95% CI)	COSMIN	Quality criteria rating	Results (95% CI)	COSMIN	Quality criteria rating	Outcomes	Results
Padua et al., 2016 ⁴⁹	6MWT	6MWD	NA	NA	NA	NA	NA	NA	NA	NA
Mori et al., 2018 ⁵²			NA	NA	NA	NA	NA	NA	NA	NA
Pazzaglia et al., 2019 ⁵³			NA	NA	NA	baseline vs. 12 months: 6MWT p=0.71	inadequate	?	NA	NA
Mori et al., 2018 ⁵²	10MWT	10MWT time	NA	NA	NA	NA	NA	NA	NA	NA
Solari et al., 2007 ⁴⁶			NA	NA	NA	NA	NA	NA	NA	NA
Pazzaglia et al., 2019 ⁵³			NA	NA	NA	baseline vs. 12 months: 10 MWT p=0.21	inadequate	?	NA	NA
Coghe et al. 2020 ⁵¹			(i) velocity (ii) step width (iii) stride length (iv) DST (v) kinematic data	NA	NA	NA	NA	NA	NA	NA
Pazzaglia et al., 2019 ⁵³	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)	NA	NA	NA	baseline vs. 12 months: several SAM outputs demonstrated worsening (P<0.05) and CMTNS too (P<0.001)	inadequate	?	NA	NA
Padua et al., 2016 ⁴⁹	Monitoring during 5 days	Several outputs during the five days monitoring (as activity index)	NA	NA	NA	NA	NA	NA	NA	NA
Ferrarin et al., 2011 ⁴⁷	NA (Motion analysis)	(i) CS (1) velocity and (2) double support (3) Mean ankle angle TW– mean ankle angle HW (4) cadence (5) stride length normalized to body height (6) kinematics data (7) kinetics data (ii) RS (1) velocity and (2) double support (3) cadence (4) stride length normalized to body height (5) kinematics data (6) kinetics data	(i) (1) SEM=3.67m/s (2) SEM=1.11%GC (3) SEM=2.7° (4) SEM=1.99 steps/min (5) ICC=2.16% (6) SEM from 2.31° to 9.36° according to the kinematics variables (ii) (1) SEM = 10.12 m/s (2) SEM = 2.19%GC (3) SEM=5.77 steps/min (4) SEM=6.09% (5) (6) SEM from 2.54° to 6.03°	very good	?	NA	NA	NA	NA	NA
Guillebastre et al., 2013 ⁴⁸	NA (GAITRite of 8.3m)	(1) Velocity (2) double support	NA	NA	NA	NA	NA	NA	NA	NA
Lencioni et al., 2017 ⁵⁰	NA (Motion analysis)	(1) Velocity normalized to body height (2) stride length normalized to body	NA	NA	NA	(1) SRM=-0.55 [%body height/s] for step ascending and -0.43 for step descending (2)	very good	+	NA	NA

height (3) kinematics data (4)
kinetics data

SRM=-0.54 [%body
height] for step ascending
and -0.56 for step
descending (3) from -
0.56° to -0.25° according
to variables in CS and
from -0.53° to -0.25° in
step descending (4) for
step descending and
ascending: from -0.53J to
0.35J

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