

SUPPLEMENTARY DIGITAL MATERIAL 5

Supplementary Table VI.—Measurement properties of the included studies in myotonic dystrophies: validity.

Articles	Subtype of DM	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
Kierkegaard <i>et al.</i>, 2007⁵⁴	DM1	6MWT	6MWD	NA	NA	NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶				NA	NA	NA	NA	NA	NA
Solbakken <i>et al.</i>, 2016⁶¹				Construct	(i)MMT of PEmg (iii)Tmg	(ii) DEmg (ii) (i) $\rho = 0.45$, $P=0.005$ (ii) $\rho = 0.62$, $P<0.001$ (iii) $\rho = 0.67$, $P<0.001$	Very good	NA	+
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				construct	(i) SARA QMT (ii) Hip flexors QMT (iii) ankle dorsiflexors QMT (iv) knee extensors QMT	(i) $\rho=-0.65$; (ii) $\rho=0.51$; (iii) $\rho=0.45$; (iv) $\rho=0.47$	Very good	NA	+
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 6MWD	NA	NA	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	10mWT (CS and RS)	10MWT time	NA	NA	NA	NA	NA	
Hammaren <i>et al.</i>, 2014⁶⁰				Construct	Total lower limb muscle strength	$\rho=-0.705$; $P<0.001$ (CS) and $\rho=-0.665$; $P<0.001$ (RS)	Very good	NA	
Kierkegaard <i>et al.</i>, 2017⁵⁶			10MWT (CS)	NA	NA	NA	NA	NA	
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷			construct	(i) SARA QMT (ii) Hip flexors QMT (iii) ankle dorsiflexors QMT (iv) knee extensors QMT	(a) 100% (b) (i) $\rho=0.65$, (ii) $\rho=-0.45$, (iii) $\rho=-0.43$, (iv) $\rho=-0.36$	Very good	NA		
Knak <i>et al.</i>, 2020⁶⁴			NA	NA	NA	NA	NA	NA	

Knak et al., 2020⁵⁸			Construct	Muscle strength (hip extension, knee extension, and ankle plantar and dorsal flexion)	$\rho = -0.488$, 95%CI -0.719 to -0.256 ($P < 0.0005$)	Very good	NA
Jimenez-Moreno et al., 2019⁶³		Mean acceleration units during the 10MWT	NA	NA	NA	NA	NA
Jimenez-Moreno et al., 2019⁵⁷	DM1	10mW/RT	10mW/RT time	construct (i) SARA QMT (ii) Hip flexors (iii) ankle dorsiflexors QMT (iv) knee extensors QMT	(i) $\rho = 0.55$ (ii) $\rho = -0.51$ (iii) $\rho = -0.47$ (iv) $\rho = -0.32$	Very good	NA
Jimenez-Moreno et al., 2019⁶³		Mean acceleration units during the 10mW/RT	Construct	muscle strength of (i) knee extensors and (ii) hip flexors	(i) $\rho = 0.4$, $P = 0.05$ (ii) $\rho = 0.43$, $p = 0.02$	(ii) NA	NA
Hammaren et al., 2012⁵⁹	DM1	TUG	TUG time	NA	NA	NA	NA
Hammaren et al., 2014⁶⁰			Construct	Total lower limb muscle strength	$\rho = -0.585$; $P < 0.001$	Very good	NA
Kierkegaard et al., 2017⁵⁶			NA	NA	NA	NA	NA
Solbakken et al., 2016⁶¹			Construct	(i) MMT of dorsal extension wrist, dorsal extension ankle, MMT of Hip flexion, knee flexion, knee extension, shoulder abduction, elbow flexion, elbow extension, (iii) MMT of trunk muscle group	(i) $\rho = -0.43$, $P < 0.001$ (ii) $\rho = -0.3$, $P = 0.08$ (iii) $\rho = -0.58$, $P < 0.001$	(ii) ρ Very good	NA
Knak et al., 2020⁶⁴			NA	NA	NA	NA	NA
Knak et al., 2020⁵⁸			Construct	Muscle strength (hip extension, knee	$\rho = -0.313$, 95%CI -0.687 to -0.286 ($P < 0.0005$)	Very good	NA

						extension, and ankle dorsal flexion)			
Kierkegaard et al., 2018 ⁶²					NA	NA	NA	NA	NA
Hammaren et al., 2012 ⁵⁹	DM1	(i)Fo8 and (ii)ST	Fo8 time and number of steps of ST	NA	NA	NA	NA	NA	NA
Hammaren et al., 2014 ⁶⁰	DM1	ST	number of steps of ST	Construct	Total lower limb muscle strength	$\rho=0.610$; $P<0.001$	Very good	NA	
Knak et al., 2020 ⁵⁸				Construct	Muscle strength (ankle plantar and dorsal flexion)	$\rho=0.546$, $P<0.0005$ 95%IC[0.327–0.764]	Very good	NA	
Knak et al., 2020 ⁶⁴				NA	NA	NA	NA	NA	NA
Galli et al., 2012 ⁵⁵	DM1	velocity (CS)	NA	Construct	lower limb muscle strength (MRC rectus femoris)	$\rho=0.62$, $P<0.05$	Very good	NA	
Radovanovic et al., 2016 ⁶⁵	(i)DM1 and (ii)DM2	NA (Motion analysis)	CT, SL, SWT	Construct	MRC scores	(i)with CT during basic walk ($\rho = -0.658$), with CT during dual task ($\rho = -0.579$) and SL in all gait conditions (ρ from 0.625 to 0.726) (ii) with SL during basic walk ($\rho = 0.692$), SL during dual task ($\rho = 0.627$)	Very good	NA	
Montagnese et al., 2020 ⁶⁶	DM2	6MWT	6MWD	Construct	lower limb MMT sum	$\rho=0.492$ $P<0.001$	Very good	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 VII.—Measurement properties of the included studies in myotonic dystrophies: reliability.

Articles	Subtype of DM	Walking test	Type of variables studied	Reliability			
				Design	Results (95% CI)	COSMIN	Quality criteria rating
Kierkegaard <i>et al.</i>, 2007⁵⁴	DM1	6MWT	6MWD	Test-retest (one week apart)	ICC=0,99 (95%CI 0,97-1,0)	very good	+
Kierkegaard <i>et al.</i>, 2017⁵⁶				Intra-rater (Best trial– average of trials)	ICC=1,00 (0,99-1,00)	very good	+
Solbakken <i>et al.</i>, 2016⁶¹				NA	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				NA	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 6MWD	intra-accelerometer reliability	ICC= 0,97 (95% CI 0,95–0,99; P<0,001)	very good	+
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	10mWT (CS and RS)	10MWT time	Test-retest (1 week)	ICC=0,91(CS) ICC=0,94(RS)	very good	+
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶		10MWT (CS)		Intra-rater (Best trial– average of trials)	ICC=0,99 (0,92-1,00)	very good	+
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷			Intra-rater (3 measures)	ICC = 0,99, 95% CI [0,99–0,99]	very good	+	
Knak <i>et al.</i>, 2020⁶⁴			NA	NA	NA	NA	
Knak <i>et al.</i>, 2020⁵⁸			Intrarater reliability	ICC= 0,96 (0,94–0,98)	very good		
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 10MWT	intra-accelerometer reliability	ICC=0,86 (95% CI 0,74–0,93; p=0,003)	very good	+
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷	DM1	10mW/RT	10mW/RT time	Intra-rater (3 measures)	ICC=0,99 (0,98–0,99)	adequate	+
Jimenez-Moreno <i>et al.</i>, 2019⁶³				Mean acceleration units during the 10mW/RT	intra-accelerometer reliability	ICC=0,96 (95% CI 0,93–0,98; P<0,001)	very good
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	TUG	TUG time	Test-retest (1 week)	ICC=0,83	very good	+
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶				Intra-rater (Best trial– average of trials)	TUG=0,98 (0,92-0,99)	very good	+

Solbakken et al., 2016 ⁶¹				NA	NA	NA	NA
Knak et al., 2020 ⁶⁴				NA	NA	NA	NA
Knak et al., 2020 ⁵⁸				Intrarater reliability (1 to 2 weeks)	ICC= 0.68 (0.54–0.79)	very good	-
Kierkegaard et al., 2018 ⁶²				NA	NA	NA	NA
Hammaren et al., 2012 ⁵⁹	DM1	(i)Fo8 and (ii)ST	Fo8 time and number of steps of ST	Test-retest (1 week)	(i)ICC=0.96, (ii)ICC=0.94	very good	+
Hammaren et al., 2014 ⁶⁰	DM1	ST	number of steps of ST	NA	NA	NA	NA
Knak et al., 2020 ⁵⁸				Intrarater reliability (1 to 2 weeks)	0.90 IC95% [0.82–0.94]	very good	+
Knak et al., 2020 ⁶⁴				NA	NA	NA	NA
Galli et al., 2012 ⁵⁵	DM1	velocity (CS)	NA	NA	NA	NA	NA
Radovanovic et al., 2016 ⁶⁵	(i)DM1 and (ii)DM2	NA (Motion analysis)	CT, SL, SWT	NA	NA	NA	NA
Montagnese et al., 2020 ⁶⁶	DM2	6MWT	6MWD	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 VIII.—Measurement properties of the included studies in myotonic dystrophies: measurement error.

Articles	Subtype of DM	Walking test	Type of variables studied	Measurement error		
				Results (95% CI)	COSMIN	Quality criteria rating
Kierkegaard <i>et al.</i>, 2007⁵⁴	DM1	6MWT	6MWD	SEM= 12m (Better of 2 6MWTs); SEM=11m (Better of 3 6MWTs); SEM=9m (Mean of 3 6MWTs)	very good	?
Kierkegaard <i>et al.</i>, 2017⁵⁶				NA	NA	NA
Solbakken <i>et al.</i>, 2016⁶¹				NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 6MWD	NA	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	10mWT (CS and RS)	10MWT time	(i) SEM=0.6s and ME=1.3(CS) SEM=0.4s and ME=0.7(RS)	very good	?
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶		10MWT (CS)		NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				NA	NA	NA
Knak <i>et al.</i>, 2020⁶⁴				NA	NA	NA
Knak <i>et al.</i>, 2020⁵⁸				SEM=0.26s	very good	?
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 10MWT	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷	DM1	10mW/RT	10mW/RT time	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 10mW/RT	NA	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	TUG	TUG time	SEM=0.7s and ME=1.4	very good	?
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA

Kierkegaard et al., 2017 ⁵⁶					NA	NA	NA
Solbakken et al., 2016 ⁶¹					NA	NA	NA
Knak et al., 2020 ⁶⁴					NA	NA	NA
Knak et al., 2020 ⁵⁸					SEM=1.09s	very good	?
Kierkegaard et al., 2018 ⁶²					NA	NA	NA
Hammaren et al., 2012 ⁵⁹	DM1	(i)Fo8 and (ii)ST	Fo8 time and number of steps of ST	(i)SEM=1.7steps and ME=14.4 (ii)SEM=1.5 steps and ME=2.9		very good	?
Hammaren et al., 2014 ⁶⁰	DM1	ST	number of steps of ST		NA	NA	NA
Knak et al., 2020 ⁵⁸					SEM=1.7steps ((±9%)	very good	?
Knak et al., 2020 ⁶⁴					NA	NA	NA
Galli et al., 2012 ⁵⁵	DM1	velocity (CS)	NA		NA	NA	NA
Radovanovic et al., 2016 ⁶⁵	(i)DM1 and (ii)DM2	NA (Motion analysis)	CT, SL, SWT		NA	NA	NA
Montagnese et al., 2020 ⁶⁶	DM2	6MWT	6MWD		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 IX.—Measurement properties of the included studies in myotonic dystrophies: responsiveness and feasibility.

Articles	Subtype of DM	Walking test	Type of variables studied	Responsiveness			Feasibility	
				Results (95% CI)	COSMIN	Quality criteria rating	Outcomes	Results
Kierkegaard <i>et al.</i>, 2007⁵⁴	DM1	6MWT	6MWD	NA	NA	NA	(i) to be able to perform (i) 52/64 participants (ii) 13 (6–two 6MWTs on the same day (1 hour apart) (ii) Median Borg RPE-score	19)
Kierkegaard <i>et al.</i>, 2017⁵⁶				NA	NA	NA		
Solbakken <i>et al.</i>, 2016⁶¹				NA	NA	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				baseline vs. one year visit (1) 1.3s [2.2–0.3] p= 0.009 (2) 0.9s [1.5–0.2] p=0.01 and (3) -36.2 [-19.3– -53.1) P<0.001	inadequate	?	% of participants made 1 trial of 6MWT, 10mWT, 10mW/RT and 30 seconds sit and stand test	100% performed one trial, 80% performed at least a second trial of each test and over 50% completed three trials in those tests required
Jimenez-Moreno <i>et al.</i>, 2019⁶³				Mean acceleration units during the 6MWD	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	10mWT (CS and RS)	10MWT time	NA	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶				NA	NA	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷				baseline vs. one year visit: 1.3s [2.2–0.3] p= 0.009	inadequate	?	% of participants made 1 trial of 6MWT, 10mWT, 10mW/RT and 30 seconds sit and stand test	100% performed one trial, 80% performed at least a second trial of each test and over 50% completed three trials in those tests required
Knak <i>et al.</i>, 2020⁶⁴				change between baseline and 1 year: -0.009 (-0.15; 0.13) p=0.90; AUC between GRS and 10MWT: AUC >0.7	very good	+	NA	NA
Knak <i>et al.</i>, 2020⁵⁸				MCID = 0.69 (13%) with MDD95% = ±0.72 (±12%) [±12%]	very good	?	NA	NA

Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 10mWT	NA	NA	NA	NA	NA
Jimenez-Moreno <i>et al.</i>, 2019⁵⁷	DM1	10mW/RT	10mW/RT time	0.9s [1.5–0.2] p=0.01	inadequate	?	% of participants made 1 trial of 6MWT, 10mWT, 10mW/RT and 30 seconds sit and stand test	100% performed one trial, 80% performed at least a second trial of each test and over 50% completed three trials in those tests required
Jimenez-Moreno <i>et al.</i>, 2019⁶³			Mean acceleration units during the 10mW/RT	NA	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	TUG	TUG time	NA	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2014⁶⁰				NA	NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2017⁵⁶				NA	NA	NA	NA	NA
Solbakken <i>et al.</i>, 2016⁶¹				NA	NA	NA	NA	NA
Knak <i>et al.</i>, 2020⁶⁴				change between baseline and 1 year: 0.35 (0.17; 0.53) p=0,0003; AUC between 0.6 and 0.7	very good	-	NA	NA
Knak <i>et al.</i>, 2020⁵⁸				MCID = 0.92 (11%) with MDD95% = ±1.26 (±26%) [±19%]	NA	NA	NA	NA
Kierkegaard <i>et al.</i>, 2018⁶²				Criterion approach AUC walking = 0.8; 95CI% [0.7–0.9]	very good	+	NA	NA
Hammaren <i>et al.</i>, 2012⁵⁹	DM1	(i)Fo8 and (ii)ST	Fo8 time and number of steps of ST	NA	NA	NA	NA	NA
Hammaren <i>et al.</i>, 2014⁶⁰	DM1	ST	number of steps of ST	NA	NA	NA	NA	NA
Knak <i>et al.</i>, 2020⁵⁸				MCID = 2.87 (16%) with MDD95% = ±4.70 (±26%) [±19%]	very good	?	NA	NA
Knak <i>et al.</i>, 2020⁶⁴				change between baseline and 1 year: -0.06 (-0.75;	very good	-	NA	NA

				0.63) p=0.88; AUC between GRS and ST: AUC<0.7				
Galli et al., 2012 ⁵⁵	DM1	velocity (CS)	NA	NA	NA	NA	NA	NA
Radovanovic et al., 2016 ⁶⁵	(i)DM1 and (ii)DM2	NA (Motion analysis)	CT, SL, SWT	NA	NA	NA	NA	NA
Montagnese et al., 2020 ⁶⁶	DM2	6MWT	6MWD	decrease of 34,16 m inadequate (p=0.003) between visit 1 and 2 (one year)	?		NA	NA

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