Muscle tone assessments for children aged 0 to 12 years: a systematic review

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ABBREVIATIONS

ATNA	Amiel-Tison Neurological Assessment
COMFORTB	COMFORT behaviour scale
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
HINE	Hammersmith Infant Neurological Examination
NNNS	Neonatal Intensive Care Unit Network Neurobehavioral Scale
NSMDA	Neurological Sensory Motor Developmental Assessment

AIM The aim of this study was to identify and examine the psychometric properties of muscle tone assessments for children aged 0 to 12 years.
METHOD Four electronic databases were searched to identify studies that included assessments of resting and/or active muscle tone. Methodological quality and overall psychometric evidence of studies were rated using the COnsensus-based Standards for the selection of health Measurement INstruments checklist.
RESULTS Twenty-one assessments were identified from 97 included studies. All assessments were broad developmental assessments that included muscle tone items or subscales. Most assessments (16/21) were designed for young children (<2y). Four assessments measured resting and active tone and demonstrated at least moderate validity or reliability: the Amiel-Tison Neurological Assessment (ATNA) at term, Neonatal Intensive Care Unit Network Neurobehavioral Scale (NNNS), Premie-Neuro for newborn infants, and the Hammersmith Infant Neurological Examination (HINE) for infants aged 2 months to 2 years. For children over 2 years, the Neurological Sensory Motor Developmental Assessment (NSMDA) assesses resting and active tone but has limited validity.
INTERPRETATION The ATNA at term, NNNS, Premie-Neuro, HINE, and NSMDA can assess resting and active tone in infants and/or children. Further psychometric research is required to extend reliability, validity, and responsiveness data, particularly for older children.

Atypical muscle tone is one of the most common clinical features observed in children with neurodevelopmental disorders. It is a characteristic of many conditions, for example those with early brain injury or cerebral palsy (CP) (2.1 per 1000),[1] genetic conditions such as Down syndrome (1.4 per 1000),[2,3] and children with Developmental Coordination Disorder (5–8 per 100).[4,5] With these populations in mind, the collective prevalence of atypical muscle tone can be estimated to affect about 5 to 8 per cent of the general population. This paper will focus on the intensity attribute of tone, which ranges between hypertonia (atypically high tone) and hypotonia (atypically low tone).[6–9] Persistent hypertonia is problematic because it can restrict movement and lead to secondary impairments such as contracture, pain, limited motor development, and restricted participation.[10–13] Persistent hypotonia produces other issues, such as poor joint stability, poor postural alignment, decreased activity tolerance, and delayed motor skill acquisition.[9,14] Given such a large proportion of the population experiences difficulties with muscle tone, accurate assessment of muscle tone is essential for diagnostic, prognostic, and treatment planning purposes.[15,16] However, no criterion standard muscle tone assessment has been endorsed to date.[17–19]

Both neural and non-neural factors contribute to tone and can produce atypical muscle tone. The neural component of muscle tone represents the tonic stretch reflex, which is generated when a muscle is facilitated by neural commands from cortical and subcortical centres, spinal circuitry, a stretch reflex, or other peripheral inputs.[6,7,20] Brain dysfunction can therefore impact on regulation of muscle tone. For example, damage to the basal ganglia, which normally inhibits descending motor commands, may result in higher muscle tone.[21–24] Damage to the cerebellum, which normally facilitates motor commands,[20,22] may result in low muscle tone. The neural component of muscle tone is highly influenced by internal factors such as an individual's arousal state,[25] and external factors such as sensory stimuli.[26,27] As these factors contribute to the neural drive to muscles, the lowest muscle tone that can be achieved is under anaesthesia when neural drive to a muscle is blocked.[28]

The non-neural component of muscle tone represents the inherent viscoelastic properties or stiffness of muscle

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tissue[6–8] and is contributed to by multiple structures within the musculotendinous unit.[29] For example, the amount of collagen or advanced glycation end product in a muscle's extracellular matrix contributes to the stiffness of the musculotendinous unit in animal models[30–32] and children with CP.[33] In addition, proteins such as titin[32,34] and dystrophin[35] within the muscle cell impact on muscle stiffness. Titin is a giant elastic protein which links the myosin filament and the z-disc within each sarcomere and promotes myosin filament to return to its initial position after stretch.[36] Reduction in titin isoform size has been associated with an increase in muscle stiffness.[34] Dystrophin is a cytoskeletal protein that assists in stabilizing the muscle fibre membrane during contraction and relaxation. A deficiency in dystrophin can lead to altered viscoelastic properties of the muscle tissue including loss of muscle stiffness.[35]

Clinically, atypical muscle tone can be assessed in resting and active states. According to the definition by Lance and McLeod,[37] refined by Sanger et al.,[7] resting muscle tone can be assessed as the 'resistance to passive stretch while a patient is attempting to maintain a relaxed state of muscle activity'[7] and 'the resistance is determined partly by mechanical factors of musculoskeletal structures and partly by the tonic stretch reflex'.[37] To standardize the influence of neural drive, it is essential to control postural load (i.e. by lying down) and other internal and external stimuli (i.e. by testing a child in a quiet alert state without unwanted sensory stimulation).[38] Electromyography monitoring can be used to detect undesired myoelectric activity during testing.[39] In clinical practice it is essential to record the child's state of arousal and ability to relax.[40] Active tone can then be defined as an individual's 'ability'[41] or 'readiness'[18] to respond to environmental demands. Measurement of active muscle tone has been performed by observing how a child engages with and reacts to the environment, especially gravity, or during self-initiated movement, sensory stimulation, or physical facilitation.[18,41,42]

With such a high prevalence of atypical muscle tone in children with disabilities and the lack of an endorsed assessment, there is a need for a psychometric review of existing assessments. This systematic review is required before being able to make suitable recommendations for children of different ages and abilities. The aim of this study was to systematically identify and examine the psychometric properties of muscle tone assessments, assessment subscales, or items designed for measuring resting or active tone in children aged 0 to 12 years. This age range was selected because neural and physical maturity, secondary musculoskeletal changes, and early identification and intervention are important factors to consider when examining muscle tone.

What this paper adds

- This is the first review of muscle tone assessments for children aged 0 to 12 years.
- Twenty-one assessments contain muscle tone items and 16 are for children under 2 years.
- Four assessments are reliable or valid to measure both resting and active tone.

METHOD

Search strategy

A systematic search of articles published between 1st January 2000 and 21st November 2017 was conducted using the computerized databases: PubMed, Embase, MEDLINE, and CINAHL (through EBSCOHost). Search terms used were 'muscle' AND 'tone' OR 'tonus' OR 'tonic' OR 'stiff*'; OR 'neurologic*' AND 'motor' OR 'neuromotor' OR 'neurosensory' OR 'neurodevelopmental' OR 'neurobehavior'; AND 'assess*' OR 'evaluat*' OR 'measur*' OR 'test' OR 'tests' OR 'testing' OR 'examin*'; AND 'child*' OR 'infant*' OR 'neonat*'; AND 'psychometric' OR 'reliab*' OR 'reproducib*' OR 'valid*' OR 'agreement'. Secondary searches involved searches for the name and author/s of assessments that met inclusion criteria, as well as reference lists of included papers.

Inclusion/exclusion criteria

A paper was included in our review if it included an assessment that (1) measured resting or active tone in human skeletal muscle; (2) was quantitative in nature; (3) was designed for clinical assessment and conducted without the need for laboratory equipment; (4) had been utilized in a study involving children aged 0 to 12 years; and (5) had instructions and psychometric properties available for review. A paper was excluded if (1) it was not published in English (2) as a full article in a peer-reviewed journal after the year 2000; (3) it did not have original data for psychometric evaluation; or (4) the assessment was designed to measure spasticity (stretch reflex excitability in response to rapid passive stretch), other movement disorders (e.g. dyskinesia, ataxia), or oral motor tone.

Data extraction and quality assessment

Two of the authors (MG and LMJ) independently reviewed all titles and abstracts, then full text, as required to identify included papers. Disagreements were discussed until a consensus was reached, and if needed a third author (KT) was consulted. Full-text articles, administration manuals, and scoring sheets were sought to examine the eligibility of assessments that measure muscle tone. These were evaluated against the inclusion criteria by two authors (MG and LMJ) to generate the final list of assessments to be analysed for their psychometric properties.

The characteristics of each assessment were extracted using a *CanChild* Outcome Measures Rating Form.[43] This included its primary purpose, target population, clarity of the assessment, requirement of equipment/qualifications/training, and validity and reliability. Methodological quality of studies containing psychometric data was assessed according to the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN).[44] Adequate reliability of COSMIN has been published.[45] We utilized the COSMIN checklist boxes for validity

(including content validity, structural validity, hypotheses testing, and criterion validity) and reliability (including internal consistency and reliability). Each box contains 5 to 18 items that address aspects of design requirement and statistical methods of study. The quality of the studies was rated on the COSMIN checklist with a four-point scale (i.e., 'excellent', 'good', 'fair', or 'poor'). According to the COSMIN guideline, the overall quality score per measurement property was then determined by the lowest rating of any of the items in each box ('worst score counts').

To score the design requirement within COSMIN checklist, several adaptations were applied in our review. For example, rather than using the standard COSMIN description for sample size, the design requirement was rated in relation to the use of a study specific sample size calculation. Appropriateness of criterion measures was rated as excellent if the criterion is a clear standard such as a survival rate or CP diagnosis. Terms used to describe psychometric properties in each article were not always identical with terms described within the COSMIN. The COSMIN taxonomy has been applied in some cases. For instance, reliability was subdivided into interrater-, intrarater- or test–retest reliability, and criterion validity was subcategorized into concurrent- or predictive validity.

Data synthesis

To determine an overall evaluation of psychometric properties of each assessment, the level of evidence was used. This measure was initially developed to rate systematic reviews of clinical trials,[46] but it has also been applied in systematic reviews for measurement properties.[47–49] The level of evidence is scored as 'strong', 'moderate', 'limited', 'conflicting', or 'unknown' (Table I), which are determined by the number of studies and the quality of the studies that could be judged by a COSMIN rating (excellent, good, fair, or poor) and the finding of each study (positive or negative).[50]

RESULTS

The systematic search identified 1169 studies, of which 97 studies met our inclusion criteria. From the 97 studies, 21 assessments for muscle tone were identified. The process of identifying relevant studies was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram[51] (Fig. 1). A list of the assessments that were excluded (n=44) in this process is available online (Appendix S1).

Table I: Criteria of psychometric evidence

Level	Rating	Criteria
Strong	+++ or – – –	Consistent findings in multiple studies of *Good* methodological quality OR in one study of *Excellent* methodological quality
Moderate	++ or – –	Consistent findings in multiple studies of *Fair* methodological quality OR in one study of *Good* methodological quality
Limited	+ or –	One study of *Fair* methodological quality
Conflicting	+/–	Conflicting findings
Unknown	?	Only studies of *Poor* methodological quality

Assessments including muscle tone items

The 21 included assessments were the Apgar, Assessment of Preterm Infant Behavior, Amiel-Tison Neurological Assessment (ATNA) at term, Einstein Neonatal Neurobehavioral Assessment Scale, Hammersmith Neonatal Neurological Examination, Kathmandu Neonatal Encephalopathy Scale, Neurobehavioral Assessment of the Preterm Infant, Neonatal Intensive Care Unit Network Neurobehavioral Scale (NNNS), Premie-Neuro, ATNA from birth to one year, Hammersmith Infant Neurological Examination (HINE), Harris Infant Neuromotor Test, Infant Neurological International Battery, Movement Assessment of Children, Movement Assessment of Infants, Touwen Infant Neurological Examination, ATNA from birth to six years, COMFORT behaviour scale (COMFORTB), INCLEN Diagnostic Tool for Neuro-motor Impairment, Neurological Sensory Motor Developmental Assessment (NSMDA), and the Touwen's neurological examination.

An overall description of the 21 assessments is provided as online supporting information (Appendix S2) and is summarized in Table II. All of the 21 assessments contained muscle tone items or subscales as part of a broad developmental assessment. Most assessments (16 of 21) were designed for infants younger than 2 years (Table II). Eight assessments reported a specific muscle tone domain, including the three ATNAs (at term, from birth–1y, and from birth–6y), the two Hammersmith assessments (Hammersmith Neonatal Neurological Examination and HINE), the Chandler's two assessments (Movement Assessment of Children and Movement Assessment of Infants), and the Touwen's neurological examination (Table II). Fifteen assessments included both resting and active tone items (Assessment of Preterm Infant Behavior, ATNA at term, Einstein Neonatal Neurobehavioral Assessment Scale, Hammersmith Neonatal Neurological Examination, Kathmandu Neonatal Encephalopathy Scale, Neurobehavioral Assessment of the Preterm Infant, NNNS, Premie-Neuro, ATNA from birth–1y, HINE, Infant Neurological International Battery, Movement Assessment of Children, Movement Assessment of Infants, ATNA from birth–6y and NSMDA) (Table III).

Across the 21 assessments, 44 individual muscle tone items (techniques) were identified (Table III). Twenty-six items were identified to assess resting tone. These items used one of four approaches: (1) observation, (2) palpation, (3) passive range of motion (ROM), or (4) resistance to passive movement. The two most commonly identified resting tone items were ROM-knee extension (i.e. popliteal angle) and ROM-shoulder adduction (i.e. scarf sign). Eighteen items were identified to assess active tone. These

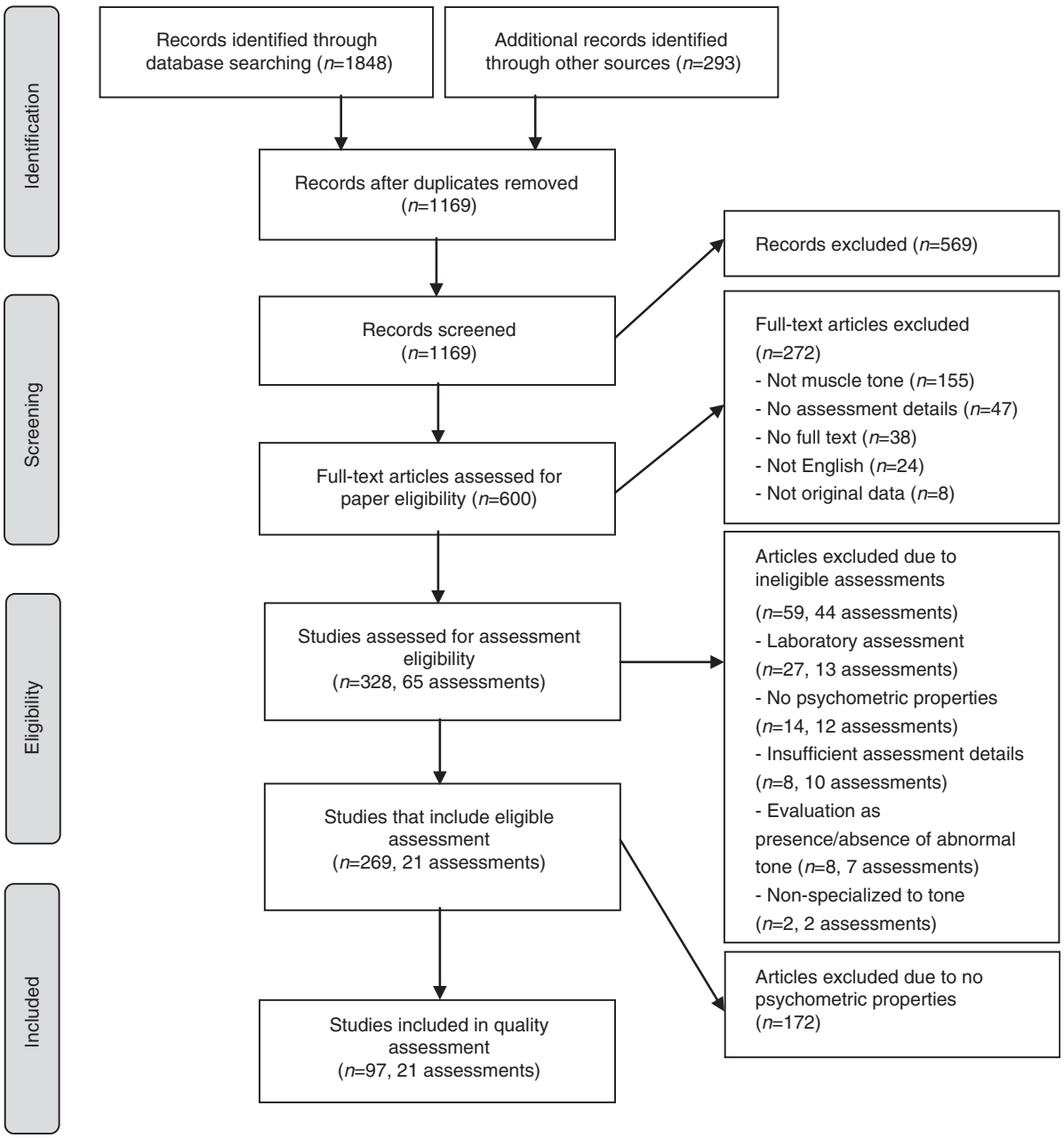

Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). Flow diagram describing selection of relevant papers in the review.

items used one of three approaches: (1) observation, (2) response to gravity, or (3) resistance to movement facilitation. The two most commonly identified active tone items were pull to sit (hands)-forwards and ventral suspension. The 44 items are summarized in Figure 2 according to the construct of muscle tone measured (i.e. resting or active tone), the approach used for measurement and the body area tested.

Evidence of validity

Evidence of validity was identified for 20 of the 21 assessments within 86 of the 97 studies. Based on each study outcome and COSMIN rating (Appendix S3, online supporting information), the level of evidence for validity of individual assessments is determined (Table IV). Validity data for specific tone items or subscales were available for the Apgar, ATNA at term, Einstein Neonatal Neurobehavioral Assessment Scale, Neurobehavioral Assessment of the Preterm Infant, NNNS, Premie-Neuro, ATNA from birth to six years, COMFORTB, HINE, Infant Neurological International Battery, Movement Assessment of Infants, NSMDA, and Touwen's neurological examination (Appendix S3). Seven of the 20 assessments achieved sound psychometric evidence for validity (at least 'moderate'

Table II: Characteristics of included assessments

Assessment	0–1mo	2mo–2y	>2y	Applicable age	Primary purpose	Tone items (total)	Tone domain	Time to administer	Manual (Publish year)	Training requirement	Normative data
Assessments only for newborn infants											
Apgar	•			First few min	Discriminative	1 (5)	N/A[e]	In an instance	Article (1953)	Not stated	Not identified
APIB	•			0–1mo	Evaluative	6[b] (280)	No	60min	Article (1982)	Required 7.5d training	Not applicable
ATNAT	•			37–42wks CA	Discriminative	9[b] (35)	Yes	5min	Article[f] (2002)	Not required	Not identified
ENNAS	•			37–42wks PMA	Evaluative	5 (20)[c]	N/A[e]	30–45min	Book (1977)	Not stated	118 low birthweight infants and 76 full term infants
HNNE	•			37–42wks	Evaluative	11[b] (34)	Yes	Not stated	Book (1999)	Not stated	224 low-risk term newborn infants born at 37–42wks GA
KNES	•			0–24h	Discriminative	1 (7)	N/A	Not stated	PhD thesis (1999)	Not stated	Not identified
NAPI	•			32wks CA[a]-term	Evaluative	7 (41)[c]	N/A[e]	30min +	Book (2000)	Required Reliability test	521 infants born preterm
NNNS	•			30wks GA ≤46–48wks CA	Evaluative	10[b] (115)[c]	No	20min	Article (2004)	Required 5d training	125 full-term healthy newborn infants aged 1–2d 344 healthy newborn infants 1388 cocaine/opiate exposed and control infants aged 1mo
Premie-Neuro	•			23–37wks PMA	Evaluative	8 (24)	N/A[e]	30–60min	Unpublished file (2004)	Recommended CD/DVD	Not applicable
Assessments for infants <2y											
ATNA-0-1	•	•		0–1y	Discriminative	17[b] (55)[c]	Yes	Not stated	Book (1986)	Not stated	Not identified
HINE		•		2–24mo	Evaluative	8 (37)	Yes	5–10min	Book (1999)	Not stated	92 infants at 12mo, 43 infants at 18mo with no known perinatal risk factors (optimality score)
HINT		•		2.5–12.5mo	Discriminative	2 (21)	No	15–25min	Book (2010)	Recommended 2days workshop	412 Canadian infants born at term and weighted greater than 2500g
INFANIB		•		4–18mo	Evaluative	6 (20)	N/A[e]	Not stated	Book (1994)	Not stated	Infants were treated in NICU
MAC		•		2–24mo	Evaluative	3[b] (53)	Yes	30min	Book (2010)	Required Reliability test	Typically developing infants between 2mo and 24mo
MAI	•	•		0–12mo	Discriminative	5[b] (65)	Yes	30min	Book (1980)	Recommended	Not identified
TINE	•	•		1y	Evaluative	1[b] (50)[c]	No	15–20min	Book (1976)	Not required	Not identified
Assessments for children >2y											
ATNA-0-6	•	•	•	0–6y	Evaluative	7[b] (65)[d]	Yes	15min	Book (2001)	Not stated	Not identified
COMFORTB	•	•	•	0–3y	Evaluative	2 (7)	N/A[e]	2min	Unpublished file (unknown)	Not stated	Not applicable
INDT-NMI			•	2–9y	Discriminative	1 (13)	N/A[e]	20–25min	Unpublished file (2014)	Recommended	Not identified
NSMDA	•	•	•	1mo–6y	Evaluative	1 (29)[d]	No	Not stated	Book (2014)	Not stated	Not identified
Touwen			•	4y upwards	Discriminative	9 (263)	Yes	30min	Book (1979)	Recommended DVD	Not identified

[a]CA means conceptional age in this instance; corrected age in all other uses. [b]Some items relating to tone were excluded because of subjectivity of tests chosen. [c]The number of summary items was not counted. [d]The number of items are different according to age groups. [e]Not applicable, meaning that absence/presence for domain is not able to present because of absence of domain in each measure. [f]Manual is not written in English but the article is assumed as a manual since it includes all information that this review considers. APIB, Assessment of Preterm Infant Behavior; ATNAT, Amiel-Tison Neurological Assessment at Term; CA, corrected age; ENNAS, Einstein Neonatal Neurobehavioral Assessment Scale; PMA; postmenstrual age; HNNE, Hammersmith Neonatal Neurological Examination; GA, gestational age; KNES, Kathmandu Neonatal Encephalopathy Scale; NAPI, Neurobehavioral Assessment of the Preterm Infant; NNNS, Neonatal Intensive Care Unit Network Neurobehavioral Scale; ATNA-0-1, Amiel-Tison Neurological Assessment from birth to one year; HINE, Hammersmith Infant Neurological Examination; HINT, Harris Infant Neuromotor Test; INFANIB, Infant Neurological International Battery; NICU, Neonatal Intensive Care Unit; MAC, Movement Assessment of Children; MAI, Movement Assessment of Infants; TINE, Touwen Infant Neurological Examination; ATNA-0-6, Amiel-Tison Neurological Assessment from birth to six years; COMFORTB, COMFORT behaviour scale; INDT-NMI, INCLEN Diagnostic Tool for Neuro-motor Impairment; NSMDA, Neurological Sensory Motor Developmental Assessment; Touwen, modified Touwen's neurological examination.

Table III: Muscle tone items identified from included assessments

			Assessment																					
			Newborn infants									Infants <2y							Children >2y					Total
Muscle tone	Method to measure	Muscle tone item	Apgar	APIB	ATNAT	ENNAS	HNNE	KNES	NAPI	NNNS	Premie-Neuro	ATNA-0-1	HINE	HINT	INFANIB	MAC	MAI	TINE	ATNA-0-6	COMFORTB	INDT-NMI	NSMDA	Touwen	
Resting tone	Observation	Posture-face																		•				1
		Posture-hands																	•[b]					1
		Posture-hip flexion										•												1
		Posture-supine					•[a]			•							•[b]				•[b]			4
		Posture-prone															•[b]							1
	Palpation	Palpation															•				•[b]			2
	Passive ROM	ROM-shoulder flexion											•			•[b]								2
		ROM-shoulder adduction			•			•[b]	•	•	•	•	•		•				•					9
		ROM-pronation/supination											•											1
		ROM-wrist flexion										•												1
		ROM-trunk flexion			•							•							•					3
		ROM-trunk extension			•							•							•					3
		ROM-trunk lateral flexion										•												1
		ROM-hip flexion									•	•			•									3
		ROM-hip abduction										•	•		•	•[b]			•					5
		ROM-knee extension			•	•	•	•[b]	•	•	•	•	•		•				•					11
		ROM-dorsiflexion										•	•		•	•[b]			•					5
	Resistance to passive movement	Resistance-head rotation										•												1
		Resistance-arms		•					•	•														3
		Resistance-legs		•					•	•														3
		Resistance-multiple joints												•[a]				•		•	•[b]	•[b]	•[a]	6
		Recoil-arms		•	•	•	•		•	•	•													7
		Recoil-legs		•	•		•		•	•														5
		Flapping-hands										•									•[b]			2
		Flapping-feet										•									•[b]			2
		Flapping-multiple joints															•							1

Table III: Continued

			Assessment																					
			Newborn infants									Infants <2y							Children >2y					Total
Muscle tone	Method to measure	Muscle tone item	Apgar	APIB	ATNAT	ENNAS	HNNE	KNES	NAPI	NNNS	Premie-Neuro	ATNA-0-1	HINE	HINT	INFANIB	MAC	MAI	TINE	ATNA-0-6	COMFORTB	INDT-NMI	NSMDA	Touwen	
Active tone	Observation	Movement-supine														•[b]	•[b]							2
		Movement-prone														•[b]	•[b]							2
		Movement-hands																	•[b]					1
		Posture-whole body	•																					1
	Response to gravity	Pull to sit (hands)-forward		•		•		•[b]		•	•	•	•		•									8
		Pull to sit (hands)-reverse						•[b]																1
		Pull to sit (hands)-head lag				•	•				•													3
		Pull to sit (shoulders)-forward			•							•												2
		Pull to sit (shoulders)-reverse			•							•												2
		Traction-arms					•				•													2
		Traction-legs					•																	1
		Body righting acting on body			•																			1
		Ventral suspension				•	•		•	•	•		•			•[b]	•							8
		Sitting-head control (flexor)					•					•[b]												2
		Sitting-head control (extensor)					•					•[b]												2
	Resistance to movement facilitation	Resistance-trunk flexion								•														1
		Resistance-while moving																				•[b]		1
		Cuddliness		•																				1

[a]Single items scored using two or more criteria – HNNE: 2 criteria, HINT: 2 criteria, and Touwen: 9 criteria. [b]Multiple items scored together using combined criteria – KNES: 4 items, ATNS-0-1: 2 items, ATNA-0-6: 2 items, INDT-NMI: 5 items, MAC: 6 items, MAI: 4 items, NSMDA: 2 items. APIB, Assessment of Preterm Infant Behavior; ATNAT, Amiel-Tison Neurological Assessment at Term; ENNAS, Einstein Neonatal Neurobehavioral Assessment Scale; HNNE, Hammersmith Neonatal Neurological Examination; KNES, Kathmandu Neonatal Encephalopathy Scale; NAPI, Neurobehavioral Assessment of the Preterm Infant; NNNS, Neonatal Intensive Care Unit Network Neurobehavioral Scale; ATNA-0-1, Amiel-Tison Neurological Assessment from birth to one year; HINE, Hammersmith Infant Neurological Examination; HINT, Harris Infant Neuromotor Test; INFANIB, Infant Neurological International Battery; MAC, Movement Assessment of Children; MAI, Movement Assessment of Infants; TINE, Touwen Infant Neurological Examination; ATNA-0-6, Amiel-Tison Neurological Assessment from birth to six years; COMFORTB, COMFORT behaviour scale; INDT-NMI, INCLEN Diagnostic Tool for Neuro-motor Impairment; NSMDA, Neurological Sensory Motor Developmental Assessment; Touwen, modified Touwen's neurological examination; ROM, range of motion.

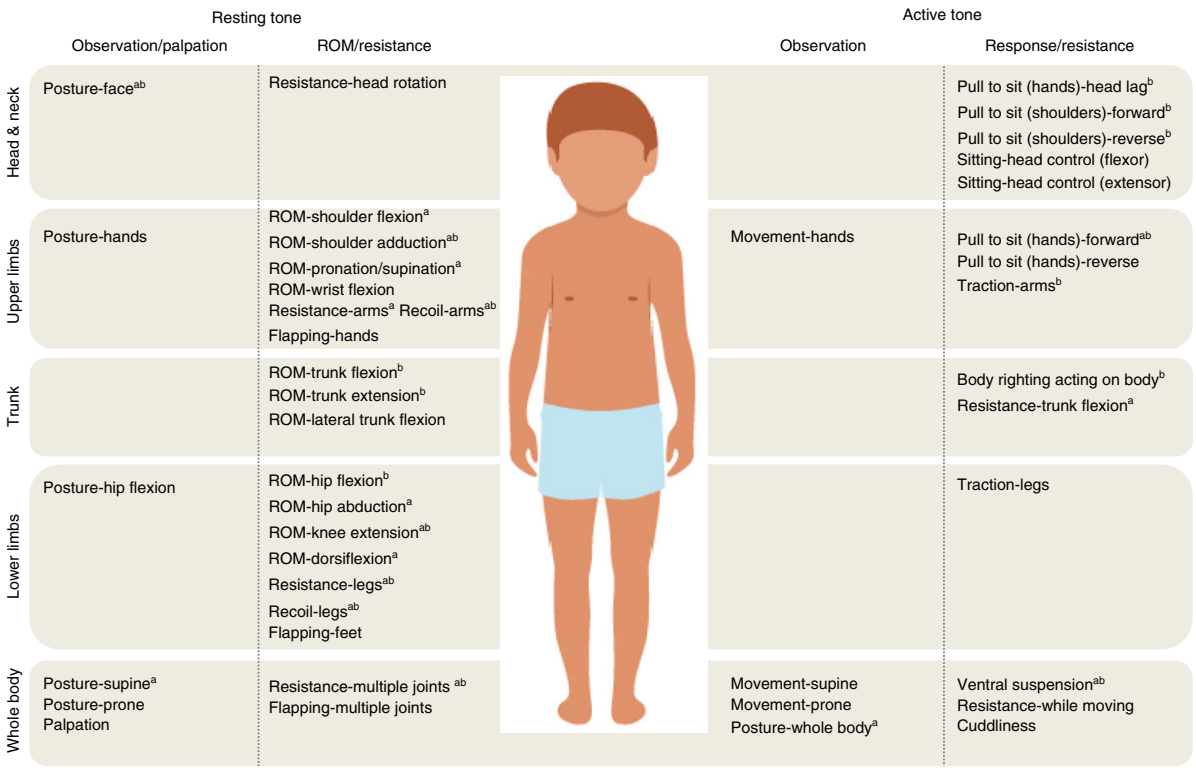

Figure 2: Muscle tone items with sound psychometric properties. [a]Items identified from assessments with 'strong' or 'moderate' positive evidence in validity. [b]Items identified from assessments with 'strong' or 'moderate' positive evidence in reliability. ROM, range of motion. [Colour figure can be viewed at wileyonlinelibrary.com].

evidence with positive result). The NNNS and Touwen's neurological examination achieved 'moderate' construct validity. For concurrent validity, the Harris Infant Neuromotor Test achieved 'moderate' evidence and the COMFORTB reported 'strong' evidence. For predictive validity, the Apgar achieved 'strong' evidence and the NNNS and HINE reported 'moderate' evidence. No assessments achieved sound content validity.

Evidence of reliability

Evidence of internal consistency and reliability was identified for 15 of the 21 assessments within 27 of the 97 studies. Based on each study outcome and COSMIN rating (Appendix S3), the level of evidence for reliability of individual assessments was determined (Table IV). Reliability data for specific tone items or subscales were available for the Apgar, ATNA at term, Hammersmith Neonatal Neurological Examination, Neurobehavioral Assessment of the Preterm Infant, NNNS, ATNA from birth to six years, COMFORTB, Harris Infant Neuromotor Test, Infant Neurological International Battery, Movement Assessment of Children, Movement Assessment of Infants, and Touwen's neurological examination (Appendix S4, online supporting information). Three assessments achieved sound psychometric evidence for reliability (at least 'moderate' evidence with positive result). The COMFORTB achieved 'strong' evidence and the Premie-Neuro achieved 'moderate' evidence of internal consistency. The ATNA at term achieved 'moderate' evidence of interrater reliability. No assessments achieved sound evidence of intrarater reliability or test–retest reliability.

Data synthesis

To suggest assessments for clinical use, two considerations were applied: that the assessment (1) included both resting and active tone items, and (2) reported sound evidence of validity and/or reliability (i.e. at least 'moderate' evidence with positive result). Fifteen assessments included resting and active tone items (Table III). Of the 15 assessments, sound validity or reliability was available for the ATNA at term, NNNS, and Premie-Neuro for newborn infants and the HINE for infants between 2 months and 2 years. No assessments met both criteria for children over 2 years.

Consideration of individual muscle tone items, regardless of assessments, was also made. Nine individual muscle tone items were identified that were reported in at least one assessment with sound validity as well as at least one assessment with sound reliability (Fig. 2, marked as 'ab').

DISCUSSION

This is the first review, to our knowledge, to systematically examine the evidence to identify clinical assessments,

Table IV: Level of evidence (direction +/– and strength +/++/+++) and COSMIN ratings for each included assessment

	Validity								Reliability							
	Content		Construct		Concurrent		Predictive		Internal consistency		Interrater		Intrarater		test–retest	
Measure	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN	+/–	COSMIN
Assessments only for newborn infants																
Apgar			+	1F			+++	3E/2G/2F			?	3P				
APIB			+	1F/1P			+	1F								
ATNAT					+	1F	– –	1G/3F			++	2F				
ENNAS			+	1F	–	1F	– –	2F								
HNNE			?	1P			–	1F							?	1P
KNES							–	1F			?	1P				
NAPI			+/–	4F	–	1F	–	1F			+	1F/1P			+	1F
NNNS			++	1G/1F			++	2F/1P	?	2P						
Premie–Neuro			+	1F/1P			?	P	++	1G	–	1F			–	1F
Assessments for infants <2y																
ATNA-0-1							– –	2F								
HINE			?	1P			++	5F/2P								
HINT	?	1P	+	1F/1P	++	1G/2F/1P	+/–	2F			?	1P	?	1P	+	1F
INFANIB			+/–	3F					+	1F	+	1F				
MAC											?	1P				
MAI			+	1F	–	1F	+/–	6F/1P			–	1F/1P			?	1P
TINE			+	1F			–	1F			+	1F				
Assessments for children >2y																
ATNA-0-6			?	1P	?	1P	–	1F/2P			?	1P				
COMFORTB	?	1P	+/–	3F/2P	+++	1E/2F			+++	1E/2P	+/–	5F			–	1F
INDT-NMI					+	1F										
NSMDA			+	1F/1P	?	1P	+/–	4F/1P								
Touwen			++	1G/1P							?	3P	?	1P	?	2P

COSMIN, COnsensus-based Standards for the selection of health Measurement Instruments; APIB, Assessment of Preterm Infant Behavior; ATNAT, Amiel-Tison Neurological Assessment at Term; ENNAS, Einstein Neonatal Neurobehavioral Assessment Scale; HNNE, Hammersmith Neonatal Neurological Examination; KNES, Kathmandu Neonatal Encephalopathy Scale; NAPI, Neurobehavioral Assessment of the Preterm Infant; NNNS, Neonatal Intensive Care Unit Network Neurobehavioral Scale; ATNA-0-1, Amiel-Tison Neurological Assessment from birth to one year; HINE, Hammersmith Infant Neurological Examination; HINT, Harris Infant Neuromotor Test; INFANIB, Infant Neurological International Battery; MAC, Movement Assessment of Children; MAI, Movement Assessment of Infants; TINE, Touwen Infant Neurological Examination; ATNA-0-6, Amiel-Tison Neurological Assessment from birth to six years; COMFORTB, COMFORT behaviour scale; INDT-NMI, INCLEN Diagnostic Tool for Neuro-motor Impairment; NSMDA, Neurological Sensory Motor Developmental Assessment; Touwen, modified Touwen's neurological examination; E, excellent, G, good; F, fair; P, poor; +, positive result; –, negative result; +++, *Strong*; ++ or – –, *Moderate*; + or –, *Limited*; +/–, *Conflicting*; ?, *Unknown*.

assessment subscales, or individual items used to measure resting or active muscle tone in children 0 to 12 years. We identified 21 assessments containing muscle tone items for children in this age group. Four assessments included both resting and active tone items and demonstrated sound evidence of validity or reliability, including the ATNA at term, NNNS, and Premie-Neuro for newborn infants, and the HINE for infants between 2 months and 2 years. For children up to 6 years, the NSMDA can measure both resting and active tone but further psychometric research is needed. There were no assessments meeting all criteria for children aged 7 to 12 years.

In terms of clinical utility, we identified 21 broad developmental assessments that included muscle tone subscales or items. Most assessments (16/21) were designed for infants younger than 2 years. This reflects the importance of early identification and classification of atypical muscle tone and the need to closely monitor potential muscle tone changes within the first 24 months of life in association with cerebral maturation.[52,53] The relative lack of assessments for older children highlights an area for further test development. Interestingly, we did not identify any assessments that were solely designed to measure muscle tone. This may explain why there is no criterion standard assessment of muscle tone recommended to date,[17] since use of each identified muscle tone item will occur only after a broader developmental assessment has been selected. This finding is in contrast to other movement disorders such as spasticity[54,55] or dyskinesia,[56] for which there are a large number of symptom-specific tools available. Nevertheless, systematic reviews for those tools suggest further psychometric data is urgently needed. Likewise, our findings indicate that research is warranted to develop specific muscle tone assessment/s for infants and children of various ages and to test the validity and reliability of these assessments for clinical populations.

When considering individual tone items, resting tone was assessed using four different methods (Table III). Three of these methods, i.e. palpation, passive ROM, and resistance to passive movement, were consistent with the recommendations for assessing resting muscle tone suggested by Lance and McLeod,[37] and Sanger et al.[7] Passive ROM and resistance to passive movement use a response to muscle length change to assess resting tone. Of the three methods, passive ROM was the most frequently included in the identified assessments, however administration instructions such as the force that an examiner applies to passive movement, reliable measurement of angles, and description of normal motor responses at different ages of testing were somewhat variable.[57] Some assessments included instruction that correctly emphasized the need to establish a 'resting' (and/or quiet alert) state of the child and to use a 'slow' passive movement speed for testing to minimize muscle activation.[53,58] Although no assessment stipulated a specific movement speed, Sanger et al.[7] recommended 3 seconds to complete one passive movement at a joint. Further research is required to determine the influence of resting state and movement speed on resting tone measurement and the possible relationship with other factors such as spasticity.

Active tone items are assumed to reflect both neural and non-neural contributors to muscle tone as these items are conducted using tests that involve muscle activation. Items for active tone were most often reported for head/neck or upper limb assessment (Fig. 2). This may be because active tone items were predominantly identified from assessments for newborn infants, and head control is a key factor to examine early neurological status. Unlike the resting tone items, specific dimensions of active tone were well documented and assessed for individual items. For instance, 'pull to sit' and 'ventral suspension' intend to measure the child's ability to recruit postural muscles (i.e. active postural tone) when required to maintain a body segment against gravity. Whereas, 'movement in supine' and 'movement in prone' assess an ability to maintain the antigravity posture and/or movement.[59] 'Cuddliness' assesses overall muscle tone through an ability to relax active muscles (i.e. reduced neural drive) and return to resting state.[60] These transitional dimensions of active tone are in line with the definition of active tone that relates to an individual's 'ability'[41] or 'readiness'[18] to respond to a change in internal or external demands. Also, these transitional dimensions of active tone emphasize that active tone is an essential element contributing to adequate postural control.[13,61,62]

Clinical assessment of muscle tone is sometimes criticized for its subjectivity and difficulty to detect small changes.[19] Overall our data supports this concern, since COSMIN ratings for the identified assessments were 'poor' or 'fair'. The primary reason was insufficient methodological quality, particularly small sample size (<30) and non-reporting of how missing data were handled.[44] These issues could be overcome by future studies with improved design. From the assessments available, we identified four assessments that measured resting and active tone and that also demonstrated at least moderate psychometric data for clinical use. When infants are born preterm, the NNNS (from 30wks gestational age) has 'moderate' evidence for construct and predictive validity and the Premie-Neuro (from 23wks gestational age) has 'moderate' evidence for internal consistency. For infants at term (birth–1mo), the ATNA at term has 'moderate' evidence of interrater reliability, however careful interpretation is required when predicting future outcomes because of 'moderate' evidence for negative predictive validity. For infants between 2 months and 2 years, the HINE has 'moderate' predictive validity. Muscle tone assessment is emphasized in the HINE and the optimality score (normative data) is provided for that age group. For children over 2 years, no assessments met all criteria as they either assessed only one aspect of muscle tone (COMFORTB, INCLEN Diagnostic Tool for Neuro-motor Impairment, and Touwen's neurological examination) or had limited psychometric evidence (ATNA from birth–6y and NSMDA). At this point, the NSMDA can be suggested for children up to 6 years because it

assesses both resting and active tone and has fair quality studies of construct validity and predictive validity (Appendix S3). This data suggests that since all identified assessments require further psychometric research, valid and reliable assessment of muscle tone in infants and children cannot be ensured at this time and that use of infant results for prognosis should be implemented with caution. In addition, new test development is needed for older children, perhaps with items identified as having promising psychometric data in Figure 2.

There are a few limitations of the data reported in this study. First, each of the muscle tone items identified in this study were contained within broad developmental assessments and so psychometric data of individual items was not always available for analysis (Appendices S3 and S4). Second, our searches were limited to papers in English, so additional data may exist in papers in other languages, or unpublished theses, or conference abstracts. Third, assessments did not always include items for both resting and active tone for all body regions, so data is not available for a holistic muscle tone battery. Finally, assessments showed considerable variability in the terminology used to define muscle tone and in the test items used to measure tone, with misleading application of tests for muscle tone, strength, spasticity, and motor performance in some cases. Our selection of items was based on currently accepted definitions noted in our introduction, however future consensus research is needed to fully define tone and its dimensions, which can then underpin new empirical research on muscle tone assessment.

CONCLUSION

This review identified 21 assessments that measure muscle tone for children aged 0 to 12 years. The majority were designed for infants and young children. Overall, psychometric evidence was insufficient to endorse one assessment. Four assessments measured resting and active tone and had published evidence of at least sound validity or reliability, including the ATNA at term, NNNS, and Premie-Neuro for newborn infants, and the HINE for infants between 2 months and 2 years. The NSMDA can be suggested for children up to 6 years, with a recommendation for further psychometric work. No assessments met criteria for children aged 7 to 12 years. Further psychometric research is needed to develop and test a specific muscle tone assessment battery for infants and children.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Excluded assessments.
Appendix S2: Brief summary of each assessment.
Appendix S3: Validity of included assessments.
Appendix S4: Reliability of included assessments.

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RESUMEN

EVALUACIONES DE TONO MUSCULAR EN NIÑOS DE 0-12 AÑOS: UNA REVISIÓN SISTEMÁTICA

OBJETIVO El objetivo de este estudio fue identificar y examinar las propiedades psicométricas de instrumentos de evaluaciones del tono muscular en niños de 0-12 años.
MÉTODO Se realizaron búsquedas en cuatro bases de datos electrónicas para identificar estudios que incluyeron evaluaciones del tono muscular en reposo y / o activo. La calidad metodológica y la evidencia psicométrica general de los estudios se calificaron utilizando la plantilla de *estándares basados en consenso para la selección de instrumentos de medición.*
RESULTADOS Se identificaron 21 instrumentos en 97 estudios. Todos los instrumentos de evaluación fueron herramientas globales del desarrollo que incluyeron elementos de tono muscular o subescalas. La mayoría de las herramientas (16/21) se diseñaron para niños pequeños (<2 años). Cuatro instrumentos midieron el tono de reposo y activo y demostraron validez o confiabilidad al menos moderada: la evaluación neurológica de Amiel-Tison (ATNA) a término, Neurobehavioral Scale (NNNS) de la Unidad de Cuidados Intensivos Neonatales, Premie-Neuro para recién nacidos y el examen neurológico de Hammersmith (HINE) para bebés de 2 meses a 2 años. Para niños mayores de 2 años, la evaluación de desarrollo motriz sensorial neurológica (NSMDA) revisa el tono de reposo y activo, pero tiene una validez limitada.
INTERPRETACIÓN El ATNA a término, NNNS, Premie-Neuro, HINE y NSMDA pueden evaluar el tono de reposo y activo en bebés y / o niños. Se requiere futuras investigaciones psicométrica para ampliar la confiabilidad, la validez y la capacidad de respuesta, en particular para los niños mayores.

RESUMO

AVALIAÇÕES DO TÔNUS MUSCULAR DE CRIANÇAS DOS 0-12 ANOS: UMA REVISÃO SISTEMÁTICA

OBJETIVO O objetivo deste estudo foi identificar e examinar as propriedades psicométricas das avaliações do tônus muscular de crianças dos 0 aos 12 anos.
METODO Quatro bases de dados eletrônicas foram pesquisadas para identificar estudos que incluíram avaliações do tônus muscular em repouso e/ou ativo. A qualidade metodológica e toda a evidência psicométrica dos estudos foram classificadas usando a check-list do COnsensus-based Standards for the selection of Measurement INstruments (COSMIN).
RESULTADOS Vinte e uma avaliações foram identificadas nos 97 estudos incluídos. Todas foram avaliações do desenvolvimento global que incluíam itens de tônus muscular ou como subescalas. A maioria das avaliações (16/21) foi destinada a crianças menores de 2 anos. Quatro avaliações mediram o tônus em repouso e o ativo, e demonstraram, no mínimo, validade ou confiabilidade moderada: a Amiel-Tison Neurological Assessment (ATNA) para crianças a termo, a Neonatal Intensive Care Unit Network Neurobehavioral Scale (NNNS), Premie-Neuro para recém-nascidos e o Hammersmith Infant Neurological Examination (HINE) para lactentes de 2 meses a crianças de 2 anos. Para crianças com mais de 2 anos, a Neurological Sensory Motor Developmental Assessment (NSMDA) avalia tônus de repouso e o ativo, mas tem validade limitada.
INTERPRETAÇÃO A ATNA para crianças nascidas a termo, NNNS, Premie-Neuro, HINE e NSMDA podem avaliar o tônus de repouso e o ativo de lactentes e/ou crianças. Pesquisas psicométricas adicionais são necessárias para ampliar dados de confiabilidade, validade e responsividade das avaliações, particularmente para crianças mais velhas.