Effectiveness of Stretch Interventions for Children With Neuromuscular Disabilities: Evidence-Based Recommendations

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Purpose: To determine whether casting, orthoses, stretching, or supported standing programs are effective in improving or maintaining body functions and structures, activity, or participation in children with neuromuscular disabilities. **Methods:** A systematic review was conducted using 6 electronic databases to identify Level 1 and 2 studies investigating stretch interventions for children aged 0 to 19 years with neuromuscular disabilities. Interventions were coded using the International Classification of Function and rated with Grading of Recommendation Assessment, Development and Evaluation, the Oxford Levels of Evidence, and the Evidence Alert Traffic Light System. **Results:** Sixteen studies evaluated the effectiveness of stretch interventions. Low-grade evidence supports casting temporarily increasing ankle range of motion, orthoses improving gait parameters while they are worn, and supported standing programs improving bone mineral density. **Conclusion:** There is limited evidence suggesting stretch interventions benefit body functions and structures. There is inconclusive evidence to support or refute stretching interventions for preventing contractures or impacting a child's activity or participation. **Trial Registration:** Prospero CRD42014013807. **(Pediatr Phys Ther 2016;28:262–275)** *Key words: activities and participation, bone mineral density, casting, children and youth, contractures, gait, neuromuscular disabilities orthoses, positioning, quality of life, range of motion, stretching, supported standing programs, systematic review*

INTRODUCTION AND PURPOSE

Contractures, hip pathologies, and spinal malalignments[1-3] are common complications for children with neuromuscular disabilities, including cerebral palsy (CP), muscular dystrophies, and neural tube defects. To address complications and promote independence in these children, considerable therapeutic resources are used such as orthoses, therapy equipment, and therapy time.[4-7] Therapists frequently prescribe and encourage compliance to a variety of stretch interventions including (1) active stretching, (2) passive stretching, (3) prolonged positioning through supported standing, or (4) prolonged stretching through casting and orthoses.[8,9] The clinical rationale for using these interventions is to avoid or defer surgery, decrease complications such as contractures, and promote function.[8,9] Proposed causes of contractures that have been hypothesized include agonist-antagonist muscle imbalance, muscle fiber atrophy, spasticity, static positioning, and structural changes to muscle tendon tissue (eg, the reduction of in-series or in-parallel sarcomeres).[8,10,11] Regardless of the cause, research shows that contractures interfere with activities of daily living, cause pain, sleep disturbance, and increase the burden

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of care.[10] Stretching, positioning, and active movement are proposed to prevent contractures and malalignment by avoiding the reduction of the number of in-series sarcomeres that decreased movement causes.[8] Despite the common practice of prescribing stretch interventions, these clinical rationales have not been validated as there is limited and varied evidence about the actual causes of contractures, the proposed physiological theory of stretching, and the clinical effectiveness of stretching in the human model.[8,10]

For all therapeutic interventions, clinicians need to consider potential benefits and harms to the child and family.[12] This clinical decision is even more important when there is limited evidence to guide practice. Although physical therapists have clinical rationales for the possible benefits of stretch interventions, the possible harmful effects also need to be examined. For example, continuous postural management can have a negative effect on sleep hygiene,[13,14] and assisted stretching is frequently reported as the most common daily activity that causes pain for children with CP.[15] Complying with an intervention that compromises a child's sleep or induces pain can place a significant emotional burden on the child, caregivers, and parents.[13,16]

Considering the routine prescription of stretch interventions and the burden and cost of implementation, a systematic analysis of the efficacy of these interventions is needed. The objective of this systematic review (SR) is to determine whether casting, orthoses, stretching programs, or supported standing programs are effective in improving or maintaining body functions and structures, activity, or participation in children and youth with neuromuscular disabilities.

METHODS

Search Strategies

English language titles were searched from the earliest date available until December 31, 2014, in the following electronic databases: CINAHL, EMBASE/Ovid, EBMR/Ovid, MEDLINE/PubMed, MEDLINE/EBSCO, and Physiotherapy Evidence Database. See the Appendix for the detailed electronic database search strategy. We did not use population-specific search terms (eg, CP and muscular dystrophy) to get comprehensive search results to later limit by inclusion criteria. Preliminary searches did not yield any articles with the same objective of this review. Details of the protocol for this SR were registered on September 19, 2014, on the International Prospective Register of Systematic Reviews (PROSPERO) and can be accessed at: http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42014013807.[17]

Eligibility Criteria

Inclusion. The inclusion criteria of this review were: (a) studies published in peer-reviewed journals appraised as Level 1 or 2 Oxford Centre of Evidence-Based Medicine (OCEBM) levels of evidence;[18] (b) study participants were younger than 19 years and had a confirmed neuromuscular disability; (c) studies contained a stretch intervention; and (d) studies evaluated the effect of stretch interventions on any body structure, body function, activity, or participation provided that there was a primary outcome measure of flexibility. Studies were included if cointerventions involved education and/or other exercise prescription (eg, aquatic therapy, aerobic training, and strength training) as long as one of the interventions was a stretch intervention. These types of cointerventions were included to allow for comprehensive programs that can be fully delegated to members of the child's team (as stretch interventions often are) under the supervision, but not direct treatment, of a physical therapist.

To address studies that included both pediatric and adult subjects, the following was determined a priori to determine study eligibility: (a) individual clinical studies must have 50% or more pediatric subjects, or a mean participant age of less than 19 years; and (b) SRs must have 50% or more studies that met the pediatric criteria, or they must provide subanalyses of the pediatric population.

Exclusion. Exclusion criteria of this review were (a) observational studies and surveys; (b) studies included able-bodied youth or youth with disabilities not considered neuromuscular in nature; (c) studies involved concurrent treatment of other physiotherapeutic interventions directly provided by a physical therapist or other health care provider (eg, acupuncture, Botox, electric modalities, manual therapy, massage, or neurodevelopmental treatment); (d) where casting was used as part of constraint-induced movement therapy; or (e) where recent surgery was done.

Operational Definitions

For the purpose of this review, the definition of a neuromuscular disability is any chronic disease or syndrome that impairs the function of skeletal muscles. This impairment can affect the muscle structure itself and/or the signal sent to the muscle. Examples of neuromuscular disabilities that were considered for review include CP, muscular dystrophies, neural tube defects, spinal cord injuries, spinal muscular atrophies, traumatic brain injury, and other rare neuromuscular diseases. The definition of a stretch intervention is an intervention aimed at maintaining or increasing joint mobility by influencing the extensibility of soft tissues spanning joints.[10] The following were preidentified as possible stretch interventions: bracing, casting, orthoses, positioning programs, self-administered stretches, splinting, stretches by caregivers, and yoga programs. Bracing, splinting, and orthoses were considered to be one treatment category, herein "orthoses," to improve clarity and knowledge translation. Both active and passive range of motion (ROM) and stretch programs were included. For the purpose of this review, flexibility was defined as the ability to move a joint through its complete ROM[19] and could have been measured with a goniometer, through gait analysis or with another valid instrument.

Selection of Studies

One reviewer (JC) screened the titles and abstracts of found articles using the inclusion and exclusion criteria stated earlier. Full-text copies of any study that appeared to meet the inclusion criteria were obtained for further inspection. Two reviewers (JC and GW or RM) independently read each article and recommended inclusion or exclusion. In cases of disagreement, a third reviewer (CH) was consulted and discussion occurred until agreement could be met.

Data Extraction

Two review authors (JC and GW or RM) independently performed data extraction of included studies using internally made data extraction forms. For each study, we collected information on the authors' main conclusions, level of evidence,[18] outcome measures coded by the International Classification of Functioning, Disability and Health (ICF),[20] participant baseline characteristics, sample size, study design, study methods, and type of intervention(s).

Assessment of Risk of Bias and Study Quality in Included Studies

For randomized controlled trials (RCTs), we assessed the risk of bias of individual studies by using a domain-based evaluation recommended by the Cochrane Collaboration[21] because the use of scales for assessing quality or risk of bias is explicitly discouraged.[22] The quality of SRs was assessed using an OCEBM appraisal sheet[23] and recorded on the data extraction form. OCEBM levels of evidence were also assigned during the data extraction process and could be downgraded due to study bias or upgraded because of large effect sizes.[18]

We used the Grading of Recommendation Assessment, Development and Evaluation (GRADE) approach to assess risk of bias across studies.[24] For purposes of SRs, GRADE defines the quality of a body of evidence as high, moderate, low, or very low for a particular outcome measure.[24] Three review authors (JC, GW, and CH) first independently performed the quality of evidence grading for each identified outcome measure. A decision about the final grade assigned was reached through discussion and consensus.

Knowledge Translation

The strength of clinical recommendations was made using the GRADE approach[12] and Evidence Alert Traffic Light System (EATLS)[25,26] to facilitate knowledge translation. A GRADE strength of recommendation is the extent to which one can be confident that the desirable consequences of an intervention outweigh its undesirable consequences.[12] The recommendations are graded as either strong or weak, and in support of or against an intervention for a particular outcome measure.[12] The EATLS rates interventions based on the quality of evidence according to the following criteria: green, go (ie, high-quality evidence supporting the effectiveness of this intervention, therefore use this approach); yellow, measure (ie, low-quality or conflicting evidence supporting the effectiveness of this intervention, therefore measure the outcomes of the intervention when using this approach to ensure the patient's goal is met); red, stop (ie, high-quality evidence demonstrating this intervention is unsafe or ineffective, therefore do not use this approach).[25,26] Three review authors (JC, GW, and CH) independently performed a GRADE strength of recommendation and EATLS rating for each identified outcome measure. A decision about the final grading and strength of recommendations assigned was reached through discussion and consensus. Reporting of SRs followed the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) statement.[27]

RESULTS

Results of the Search

Electronic database and hand searching yielded 24 930 references. After removing duplicates and screening titles and abstracts, 81 studies were eligible for full-text review. There were 10 studies that required a third reviewer to determine eligibility. After inspecting the full reports, 16 articles were included (See Figure 1).

Excluded Studies

Figure 1 provides a summary of reasons for exclusion of studies (See Supplemental Digital Content 1, available at http://links.lww.com/PPT/A106, which lists all articles excluded at the full-text level). The most common reasons for exclusion were lower level of evidence (n = 26), adult population (n = 9), no stretch intervention (n = 8), cointerventions (n = 7), and downgraded due to risk of bias (n = 6). All articles downgraded due to risk of bias were determined to have unacceptable risk of attrition, detection, performance, selection, and other biases. Other biases included confounding cointerventions, poor compliance to intervention or poor reporting of compliance, lack of statistical analyses to determine whether groups were similar at baseline, and sample populations not representative of exposed cohort.

Included Studies

Levels of Evidence and Risk of Bias in Included Studies. Among the 16 included articles, 12 were SRs and 4 were randomized controlled or crossover trials (Table 1).[28-43] Two of the SRs were appraised as Level 1 evidence as they had both high methodological quality and only included RCTs or SRs.[28,29] The 10 other SRs were appraised as Level 2 evidence due to inclusion of lower levels of evidence and/or poor methodology. All of the included RCTs were graded as Level 2 evidence. In general, the methodological quality of included articles was poor. Common methodological weaknesses

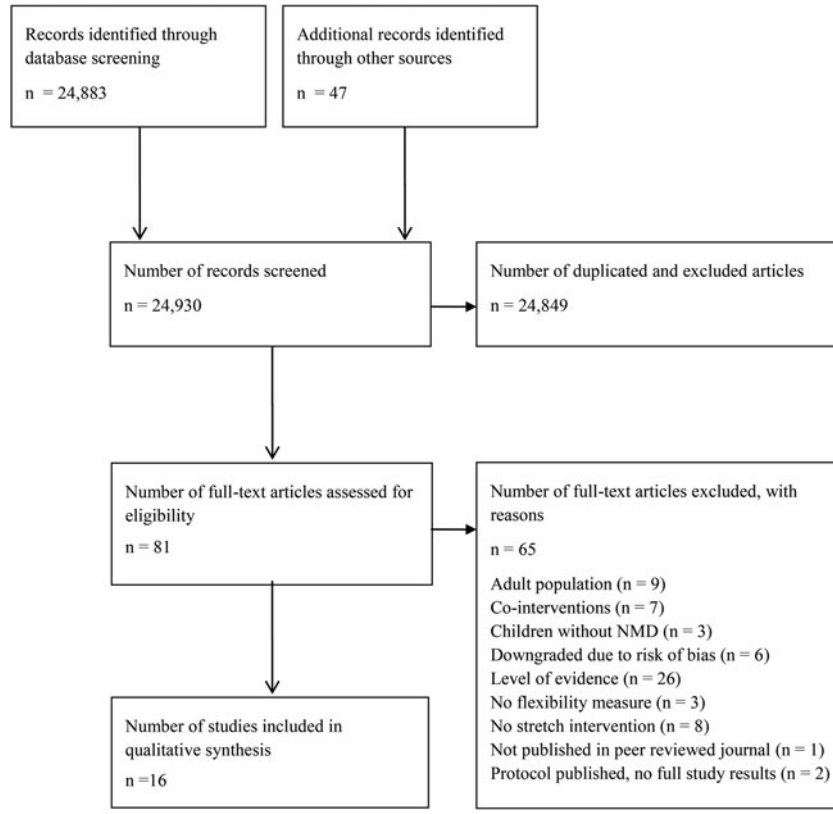

Fig. 1. PRISMA flow diagram.

in the studies included lack of reporting, or inadequate randomization methods, allocation concealment, reporting of dropouts, and controlling for confounding cointerventions (see Supplemental Digital Content 2, available at http://links.lww.com/PPT/107, which summarizes methodological quality of included studies).

Participants

Sample sizes of the included studies ranged from 14 to 1110 (Table 1). The effectiveness of stretch interventions was investigated in the following populations: CP (n = 9), mixed disabilities (n = 4), Charcot-Marie-Tooth (n = 2), and Duchenne muscular dystrophy (n = 1). The age of participants ranged from 20 months to 30 years, with all studies having a median age less than 19 years.

Interventions

The included studies evaluated the effectiveness of casting (n = 5), orthoses (n =10), passive stretching or positioning (n =5), and supported standing programs (n =6). A wide range of casting protocols, orthoses configurations, prescription of stretching programs, and supported standing equipment was evident from the literature. Not all research reports described this in adequate detail to be replicated. Comparison interventions were explicitly mentioned in all included RCTs[30-33]; however, SRs often did not record comparison interventions. Where this information was well reported, comparison groups were often lacking because of study design (eg, case report) (Table 1).

Intervention dosing parameters such as treatment duration, frequency, and intensity were well recorded in all of the RCTs; however, these parameters were inconsistently reported in the SRs. When recorded, casting intervention lasted for 3 to 5 weeks with casting protocols not being well documented.[28,29,34-36] There was a lack of information about the specific orthotic intervention dosing protocols recorded by the SRs.[28,29,37-39] RCTs investigating night splinting required splints to be worn all night[30,32,33] or every other night[31] for a duration of 4 weeks,[32] 6 weeks,[33] 12 months,[31] or 30 months.[30] Passive stretching or positioning dosing reported in the SRs noted that 30 minutes' total stretch program was the most commonly chosen session time, with each stretch typically being held for 30 to 60 seconds and repeated for several repetitions.[35,40,41] One SR noted an average duration of passive stretching or positioning study length to be 8.2 weeks with a mean frequency of intervention to be to 4.5 times per week.[40] Supported standing program dosing was well recorded in 1 SR with the following evidence-based dosage recommended: 5 days/wk positively affects bone mineral density (BMD) (60-90 min/d), hip stability (60 min/d in 30°-60° degrees hip abduction), ROM of hip knee and ankle (45-60 min/d), and spasticity (30-65 min/d).[42] The majority of included studies in the 2 other reviews noted 30 minutes as the common duration of supported standing; however, there was a large variation in study duration from just 1 session to

TABLE 1
Characteristics of Included Studies

Author	Study Design	OCEBM Level of Evidence	Population	Intervention(s)	Articles and Sample Size	Body Structure and Function Outcome Measures	Activity and Participation Outcome Measures
Autti-Rämö et al[28]	SR	1	CP	I: Upper and lower limb casting and orthoses C: Varied (eg barefoot condition, no casting, within participant orthoses comparison)	5 SR (n = 663)[a]	EMG, energy expenditure, gait analysis, muscle tone, muscle strength, quality of movement, ROM	Balance, COPM, functional tasks (sit to stand, stair use, walking), GMFM, grasp, hand function/use, parent perception, Peabody, QUEST, visual motor performance
Blackmore et al[34]	SR	2	CP	I: Ankle serial casting C: No comparison interventions (eg, 10 of the 12 studies had no controls); varied (eg, 2 of the 12 studies: NDT-based physical therapy, physical therapy, and home program)	19 studies (n = 395)	3D gait analysis, passive ankle ROM	None[b]
Effgen et al[35]	SR	2	School-aged children with disabilities	I: Lower extremity casting, orthoses, splints; passive stretching; weight-bearing interventions C: No comparison interventions recorded	15 SR (n = not reported)[c]	BMD, gait parameters, prevention of contracture, ROM, spasticity	Balance, functional task (eg, sit to stand), GMFM, hand function
Figueiredo et al[37]	SR	2	CP	I: Any type of AFO C: Varied (eg, within-group barefoot condition, shoes only, hinged or nonhinged AFO)	20 studies (n = 446)	EMG, energy expenditure, gait kinematics, gait kinetics, ROM	BOTMP, GMFM, GMPM, PEDI
Franki et al[40]	SR	2	CP	I: Passive stretching and weight-bearing interventions C: No comparison interventions recorded	83 studies (n = 660)[d]	Behavioral state, BMD, bowel activity (diary), gait analysis, gait velocity, muscle tone, ROM	ADL (feedback form), endurance (2-min walk test), CRIB, personal feeling of improved daily functioning, PEDI
Hyde et al[30]	RCT	2	DMD	I: Passive stretching combined with use of night orthoses C: Passive stretching	n = 27	Anthropometric measures, hip flexor and tendoachilles contracture, muscle strength	Gower's maneuver, MA, timed physical performance (eg, time taken to run)
Lannin et al[36]	SR	2	Children with neurological conditions	I: Upper extremity casting C: No comparison interventions (eg, 13/23 studies); varied (eg, 10/23 studies: no casting, passive stretching, postcasting follow-up of either cast or orthotic regime, shorter casting duration, traditional therapy)	23 studies (n = 326)	EMG, hypertonicity, ROM	None

(*continues*)

TABLE 1
Characteristics of Included Studies (*Continued*)

Author	Study Design	OCEBM Level of Evidence	Population	Intervention(s)	Articles and Sample Size	Body Structure and Function Outcome Measures	Activity and Participation Outcome Measures
Maas et al[31]	RCT	2	CP	I: Use of a night KAFO C: No KAFO use	n = 28	Ankle-foot dorsiflexion ROM, ankle-foot and knee angle in gait, complaints, orthosis wearing time	GMFM
Montero et al[38]	SR	2	Children with motor disabilities	I: Technical devices including orthoses and supported standing programs C: No comparison interventions recorded	27 studies (n = 664)[e]	BMD, energy expenditure, gait analysis, hip migration percentage, hip and spine x-rays, hip subluxation and dislocation, joint ROM, muscle alignment, muscle strength, perceived exertion, spasticity, transitional movement of sit to stand	Balance, BOTMP, changes in daily activities and posture (questionnaire), checklist of feeding problems, GMFCS, GMFM, GMPM, PEDI
Neto et al[39]	SR	2	CP	I: Articulated AFO C: Rigid AFO	7 studies (n = 120)	EMG, energy expenditure, gait analysis: kinetics, kinematics, and gait parameters	None
Novak et al[29]	SR	1	CP	I: Casting; orthoses; stretching via manual stretching, splinting or positioning; weight-bearing via standing frame C: No comparison interventions recorded	166 studies (n = not reported)[f]	BMD, gait analysis, prevention of contracture, ROM, spasticity	Upper and lower limb function
Paleg et al[42]	SR	2	Children with atypical development, with or without neuromuscular diagnosis including CP	I: Supported standing program C: No comparison intervention recorded or no comparison intervention (eg, 18/30 studies); varied (eg, 12/30 studies: addition of trochanteric girdle to long leg braces set in abduction, dynamic standing, no stander or positioning equipment, standing with hip abduction and extension, straddled weight-bearing, whole body vibration, within-group nonstanding phase)	30 studies (n = 1110)[g]	Functions of the bone as related to BMD, functions of the bone as related to hip stability, functions of the digestive system, mental functions, muscle power functions, muscle tone functions, neuromusculoskeletal and movement-related functions, skin and related functions	Mobility and major life areas (ie, speed of feeding, social interaction, eased burden of care, GMFM)

(*continues*)

TABLE 1
Characteristics of Included Studies (*Continued*)

Author	Study Design	OCEBM Level of Evidence	Population	Intervention(s)	Articles and Sample Size	Body Structure and Function Outcome Measures	Activity and Participation Outcome Measures
Pin[41]	SR	2	CP	I: Passive stretching programs C: No detailed information about comparison interventions (eg, 3/7 studies participants acted as their own control with no record of within-group comparison intervention; 4/7 studies had a comparison intervention but not recorded)	7 studies (n = 133)	EMG, gait analysis, ROM, spasticity	None
Pin[43]	SR	2	CP	I: Static lower or upper body weight-bearing C: No detailed information about comparison interventions (eg, 7/10 studies had a comparison but not recorded)	10 studies (n = 122)	Behavioral state, BMD, EMG, gait analysis, hand posture, hand surface area, muscle tone, ROM	Bayley Scales of Infant and Toddler Development (mental scales), CRIB, grasp and release, Jebsen Taylor Hand Function test, prehension, spontaneous use of hand
Refshauge et al[33]	RCOT	2	Charcot-Marie-Tooth	I: Night ankle orthoses C: No night ankle orthoses	n = 14	Isometric strength, passive ROM	None
Rose et al[32]	RCT	2	Charcot-Marie-Tooth	I: Serial night orthoses for 4 wk, followed by 4 wk of stretching C: No intervention	n = 30	Ankle ROM (lunge test), foot deformity	Balance, falls, mobility (eg, standing up from chair, walking, and stairs), self-reported activity limitations

Abbreviations: ADL, activity of daily living; AFO, ankle-foot orthotic; BMD, bone mineral density; BOTMP, Bruininks-Oseretsky Test of Motor Proficiency; C, comparison; CP, cerebral palsy; COPM, Canadian Occupational Performance Measure; CRIB, Carolina Record of Individual Behavior; DMD, Duchenne muscular dystrophy; EMG, electromyography; GMFCS, Gross Motor Function Classification System; GMFM, Gross Motor Function Measure; GMPM, Gross Motor Performance Measure; I, intervention; KAFO, knee-ankle-foot orthosis; MA, motor ability scale; OCEBM, Oxford Centre of Evidence-Based Medicine; PROM, passive range of motion; QUEST, Quality of Upper Extremity Skills Test; PEDI, Pediatric Evaluation of Disability Inventory; RCOT, randomized crossover trial; RCT, randomized controlled trial; ROM, range of motion, SR, systematic review.
[a] 1/5 studies did not report sample size.
[b] Only studies that had a cointervention of Botox had functional measures.
[c] 6/15 studies on stretch interventions.
[d] 12/83 studies on stretch interventions.
[e] 17/27 studies on stretch interventions.
[f] 11/166 studies on stretch interventions.
[g] 5/30 studies did not report sample size.

9 months of intervention.[35,43] No studies commented on the intensity of the intervention, besides 1 RCT that mentioned that if night splinting interfered with sleeping, participants were to use them during day rest periods instead.[31] It is important to note that although several of the SRs did not record specific dosing parameters, they noted a lack of long-term follow-up in their included studies.[28,29,35,37,40]

Outcomes

ROM, prevention of contractures, BMD, gait analyses, and spasticity were the most studied body function and structure measures (Table 1). Functional mobility assessment (eg, sit to stand) and the Gross Motor Function Measure were the most common activity measures. The Canadian Occupational Performance Measure was the only participation measure identified in any study.

Analysis of the Evidence

The effectiveness of all of the interventions as coded by ICF levels, GRADE quality of evidence, and by the previously mentioned knowledge translation tools is summarized in Table 2. Because of the large heterogeneity and lack of reporting of interventions and outcome measures used in individual studies and SRs, effect-size estimation and meta-analysis of the data were not performed.

Adverse Events

Two RCTs,[31,32] and 2 SRs[4,36] on casting and orthoses reported on adverse events. Adverse events such as bruising and blistering were seen in 13% of subjects who had serial casting,[32] whereas the majority of participants in a study on knee-ankle-foot-orthoses (KAFO) reported frequent pain because of muscle strain and pressure spots, as well as sleep disturbance.[31] Additional complaints of night-time use of KAFOs included hot or sweating legs, itching, cramping, and bed-wetting.[31] The most common adverse events of casting cited from the SRs included skin irritation, skin breakdown, and pain.[34,36]

Casting

Evidence. Consistent but very low evidence supports the use of 3 to 5 weeks of ankle casting for the positive short-term effects that it has on passive ankle dorsiflexion.[28,29,34,35] Short-term improvements in gait parameters such as self-selected pace and stride length following ankle casting have also been noted[29,34]; however, this review did not identify any Level 1 or 2 evidence supporting or refuting long-term benefits on gait and ROM. There is insufficient research on the effectiveness of casting for other lower extremity joints. One SR that assessed the effectiveness of upper extremity casting for children with neurological conditions concluded that there is insufficient high-quality evidence regarding the effect or long-term effects to either support or abandon upper extremity casting.[36] No studies identified in this review included the direct measurement of the effect of casting on activity or participation of children with neuromuscular disabilities.

Clinical Recommendation From the Evidence. Using the EATLS, serial casting for short-term improvement of ankle ROM is rated as a green intervention supported by very low evidence. All other outcome measures have insufficient evidence, thus casting is rated as a yellow intervention for these measures.

Orthoses

Evidence. The most consistent finding among studies identified in this review is that there is very low evidence that ankle-foot orthotic (AFO) devices that restrict plantarflexion improve gait kinematics and kinetics while the device is worn (Table 2).[28,29,35,37,38] One SR that compared articulated and rigid AFOs for children with CP found significant differences in peak dorsiflexion, reduction in double-support time, increase in gait speed, and reduction in energy expenditure with the use of an articulated orthosis.[39] There is both conflicting and insufficient evidence on the effectiveness of orthoses for the prevention of contractures, either by the use of AFOs or by wearing night splints.[28-33,35,37,39] Two randomized trials showed that night ankle splints or KAFOs do not improve ROM in children with CP or Charcot-Marie-Tooth, whereas 1 study found that the expected annual change in tendoachilles contracture for boys with Duchenne muscular dystrophy was 23% less in the night splint and passive stretch group compared with the passive stretch-only group.[30,31,33] One RCT found that at 4 weeks of postserial night casting, the experimental group had significant but small increase in ankle dorsiflexion; however, these effects were not maintained with stretching at 8 weeks.[32] There is also conflicting and insufficient evidence in the studies identified to support the use of orthotics for promoting activity or participation while the device is worn. Two studies reported that wearing a lower extremity device might make functional activities, such as rising up from the floor, more difficult,[28,35] whereas another study showed that orthoses have a positive effect on functional activities related to mobility.[37] Most SRs reported that there is insufficient evidence to support or refute the use of orthoses in improving function,[28,29,35] whereas the majority of randomized trials[30-32] showed no functional difference between experimental and control groups or did not include a functional measure.[33] Several authors mention that the wide variety of lower limb orthoses investigated as well as different terms used for the same orthoses made a systematic evaluation difficult.[28,37,38] Poor compliance and tolerance of night orthoses has also been cited as a limitation in determining the effectiveness of this intervention.[31,32] No high-quality studies that assessed the effectiveness of upper extremity orthoses were found.

Clinical Recommendation From the Evidence. According to the EATLS, orthoses that restrict plantarflexion

TABLE 2

Summary of Findings: Stretch Interventions for Children With Neuromuscular Disabilities

Intervention Outcome[a]	Studies	GRADE Quality of Evidence[b]	GRADE Strength of Recommendations[c]	Traffic Light Action[d]	Comments
Intervention: casting					
Body function: PROM of lower limbs	Autti-Rämö et al[28] Blackmore et al[34] Effgen et al[35] Novak et al[29]	⊕◯◯◯ Very low	Strong for	Green: go	Effective for increasing ankle range in the short term. No evidence on the long-term effects for different joints
Body function: PROM of upper limbs	Autti-Rämö et al[28] Lannin et al[36] Novak et al[29]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute the use of casting
Body function: gait kinetics and kinematics	Blackmore et al[34] Effgen et al[35] Novak et al[29]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Immediate gains in gait parameters (ie, stride length and walking speed) are likely secondary to improvements in ROM; however, the long-term benefits on gait are unknown
Body function: spasticity	Blackmore et al[34] Lannin et al[36] Novak et al[29]	⊕◯◯◯ Very low	Weak against	Yellow: measure	Insufficient evidence to support or refute the use of casting
Activity and participation: functional abilities	Autti-Rämö et al[29] Blackmore et al[34] Effgen et al[35] Lannin et al[36] Novak et al[29]	⊕◯◯◯ Very low	Weak against	Yellow: measure	Insufficient evidence to support or refute the use of casting
Intervention: orthoses					
Body function: PROM and prevention of contracture of lower limbs	Autti-Rämö et al[28] Effgen et al[35] Figueiredo et al[37] Hyde et al[30] Maas et al[31] Neto et al[39] Novak et al[29] Refshauge et al[33] Rose et al[32]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Effective for increasing ankle ROM while wearing the device. There is no evidence to support or refute the long-term benefit of wearing orthoses on ROM. Compliance has been noted as an important factor
Body function: gait kinetics and kinematics	Autti-Rämö et al[28] Effgen et al[35] Figueiredo et al[37] Montero et al[38] Neto et al[38] Novak et al[29]	⊕◯◯◯ Very low	Strong for	Green: go	AFO devices that restrict plantarflexion are effective for improving gait parameters while wearing the device
Activity and participation: lower limb functional abilities	Autti-Rämö et al[28] Effgen et al[35] Figueiredo et al[37] Hyde et al[30] Maas et al[31] Montero et al[38] Neto et al[39] Novak et al[29] Refshauge et al[33] Rose et al[32]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute the use of lower limb orthoses.
Intervention: positioning, range of motion, stretching					
Body function: PROM and prevention of contracture	Effgen et al[35] Franki et al[40] Novak et al[29] Pin[41] Rose et al[32]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute the use of stretching programs. Although there is insufficient evidence, generally studies showed an increase in ROM poststretching or a loss of ROM after stretching stopped
Body function: spasticity	Franki et al[40] Pin[41] Rose et al[32]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute the use of stretching programs

(*continues*)

TABLE 2
Summary of Findings: Stretch Interventions for Children with Neuromuscular Disabilities (*Continued*)

Intervention Outcome[a]	Studies	GRADE Quality of Evidence[b]	GRADE Strength of Recommendations[c]	Traffic Light Action[d]	Comments
Activity and participation: functional abilities	Effgen et al[35] Franki et al[40] Novak et al[29] Pin[41] Rose et al[32]	⊕◯◯◯ Very low	Weak against	Yellow: measure	Insufficient evidence to support or refute the use of stretching programs
Intervention: supported standing					
Body structure: BMD	Effgen et al[35] Franki et al[40] Montero et al[38] Novak et al[29] Paleg et al[42] Pin[43]	⊕◯◯◯ Very low	Strong for	Green: go	Effective to increase lower limb bone mineral density; however unclear whether this prevents pathological fractures
Body function: PROM and prevention of contracture of lower limbs	Effgen et al[35] Franki et al[40] Montero et al[38] Paleg et al[42] Pin[43]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute; however, several studies showed a positive effect on hip range of motion or migration percentage
Body function: spasticity	Paleg et al[42] Pin[43]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Effective in the temporary reduction of lower limb spasticity
Activity and participation: functional abilities	Effgen et al[35] Franki et al[40] Montero et al[38] Paleg et al[42] Pin[43]	⊕◯◯◯ Very low	Weak for	Yellow: measure	Insufficient evidence to support or refute the use of casting

Abbreviations: AFO, ankle-foot orthosis; BMD, bone mineral density; GRADE, Grading of Recommendations Assessment, Development, and Evaluation; PROM, passive range of motion; ROM, range of motion.
[a]Coded with the International Classification of Functioning, Disability and Health.[20]
[b]GRADE specifies 4 quality of evidence ratings (high, moderate, low, and very low) that are applied to a body of evidence. The GRADE quality of evidence rating reflects the confidence that the estimates of the effect are correct.[24]
[c]The GRADE strength of a recommendation is separated into strong and weak. It is defined as the extent to which one can be confident that the desirable effects of an intervention outweigh its undesirable effects.[12]
[d]The Evidence Alert Traffic Light System rates interventions according to the following criteria: Green, go (ie, high-quality evidence supporting the effectiveness of this intervention, therefore use this approach); yellow, measure (ie, low-quality or conflicting evidence supporting the effectiveness of this intervention, therefore measure the outcomes of the intervention when using this approach to ensure the patient's goal is met; red, stop (ie, high-quality evidence demonstrating this intervention is unsafe or ineffective, therefore do not use this approach)".[25,26]

to improve a child's gait while the device is worn is a green intervention. Orthotic use for the prevention of contractures and promotion of activity and participation are yellow interventions.

Positioning and Stretching

Evidence. The most commonly reported outcome measures for positioning and stretching programs were prevention of contractures and ROM (Table 1). Two SRs reported that there is insufficient evidence to support or refute the use of passive ROM[35] or positioning[29] to prevent contractures. One review[29] concluded that manual stretching is ineffective for contracture prevention in the short to medium term (<7 mo) based on a comprehensive and robust meta-analysis; however, this conclusion was based mainly on one review[10] that included mostly adults, only looked at static stretches, and was not able to define or standardize the control condition of usual care. Two SRs identified in this study noted that there is limited or weak evidence to support the effectiveness of passive stretching for improving ROM and spasticity.[40,41] Although there was limited/weak evidence, generally studies showed an increase in ROM poststretching or a loss of ROM after stretching stopped.[40,41] One author concluded that it appeared that sustained stretching of longer duration was preferable to improve range of movements and to reduce spasticity of muscles around the targeted joints.[41] No studies identified by this review reported the effect of positioning, ROM, or stretching programs on the activity or participation of children with neuromuscular disabilities.

Clinical Recommendation From the Evidence. There is insufficient evidence to make any recommendations in regard to the use of positioning or stretching programs; therefore, all are yellow interventions in accordance with the EATLS.

Supported Standing Programs

Evidence. Six SRs evaluated the effectiveness of supported standing programs.[29,35,38,40,42,43] In this review,

1 SR was identified that evaluated the effectiveness of static weight-bearing in children with CP. It found that other than the findings of increased BMD and temporary reduction in spasticity, there is limited evidence supporting the intervention because of weak research methodology.[43] Another study, we identified assessed the effectiveness of standing programs for children with atypical development, concluded that there is evidence that the intervention positively affects BMD, hip stability, ROM of the hip knee and ankle and spasticity.[42] The authors concluded that to see these positive results, standing program dosage should be between 30 and 90 min/d.[42] The most consistent finding reported in all studies we identified is that there is very low evidence that supported standing programs using an external device increase lower limb and vertebral BMD.[29,35,38,40,42,43] There is no evidence that improved BMD prevents pathological fractures.[35,43] Although there is insufficient evidence to refute or support the effectiveness of this intervention for promoting ROM and/or preventing contractures, 2 studies showed a positive effect on hip ROM or hip migration percentage.[38,42] Two SRs we identified also found that there is evidence that supported standing programs temporarily decrease spasticity.[42,43] This review identified no quantitative research on the effect of supported standing programs on the activity or participation of children with neuromuscular disabilities.

Clinical Recommendation From the Evidence. Using the EATLS, supported standing programs using an external device are rated as green interventions for increasing lower extremity BMD. Supported standing programs are yellow interventions for all other outcome measures.

DISCUSSION

Summary of Main Findings

The primary outcome measure for this SR was flexibility. For this outcome measure, the strongest evidence found in this review was for the use of casting to increase passive ankle dorsiflexion in the short-term. Conflicting evidence was found for the use of orthotics to prevent contractures in populations of children with CP, Charcot-Marie-Tooth, and Duchenne muscular dystrophy. Insufficient evidence was found to support or refute stretching/positioning programs or supported standing programs for the increase of ROM or prevention of contractures.

In addition to the flexibility measure, the strongest evidence identified in this review supports the use of orthoses for improving gait kinetics and kinematics while worn, and for supported standing programs for improving lower extremity BMD. The majority of research has focused on assessing short-term outcomes of the lower extremity at the body structure and function level. Of the limited reporting of activity and participation measures, no treatment effect was found when compared with controls.[30-32] For example, although AFOs that restrict plantarflexion have a favorable effect on improving gait kinetics and kinematics, we identified no consistent research to indicate this increases the child's quality of life (Table 2). Treatment interventions varied immensely with a wide range of the following interventions being recorded: casting protocols, orthoses configurations, prescription of stretching programs, and supported standing program equipment. Adverse events were rarely mentioned and when recorded they included skin irritation and pain from casting and orthoses.[31,32,34,36] Only one review[35] has looked at the same breadth of interventions for multiple pediatric disabilities as this current research has presented. The comparable review[35] included lower levels of evidence and no quality assessment was conducted. All other studies have focused on 1 population[28-34,37,39,40,41,43] and/or on 1 or 2 stretch interventions.[28,30-34,36,37,39,41-43] To date, this is the most comprehensive review on all stretch interventions for a large population treated by pediatric physical therapists. In addition, this is only the second review article[29] on pediatric stretch interventions that has used the GRADE approach and the EATLS to categorize treatments based on the quality and strength of evidence. These knowledge translation tools allow clinicians to quickly implement research into practice. This is valuable, as previous research has shown that although pediatric physical therapists have a positive attitude toward evidence-based practice, they routinely self-report that they are unable to implement this information into practice.[44] Although this review confirms the effectiveness of some stretch interventions, it is clear that there is still a large gap between clinical practice, treatment rationales, and the available evidence. More specifically, there is no high-quality evidence to support or refute that stretch interventions can avoid or defer surgery, decrease complications such as contractures, or promote function as reported by pediatric physical therapists.[8,9] The yellow rating of many interventions identified in this review highlights the importance of clinicians using not only the best-available research regarding function and basic science muscle/tendon physiology but also the 2 other tenets of evidence-based practice: clinician experience and patient values.[45,46] For example, clinicians should consider their historical knowledge about how disuse, muscle imbalances, and immobility affect flexibility and function.[45] When the evidence in the literature is unclear, clinicians are encouraged to use outcome measures specific and meaningful to their clients to track the effectiveness of and to modify treatment as necessary.[46]

Limitations

Although this review only included Levels 1 and 2 evidence, 10 of the 12 identified SRs did incorporate lower levels of evidence. There is a possibility that some relevant studies of lower levels of evidence may have been missed were they not included in these reviews, and could have provided additional insight into this research question. However, although case studies or series may have demonstrated more polarized results for the individuals (larger positive or negative effects) compared with groups (because of regression to the mean), it is

unlikely that their results could have been generalized to the population investigated in this review. Inclusion criteria requiring 1 primary outcome measure of flexibility may have also excluded articles that only looked at other measures of body functions and structures or primarily on activity or participation measures. This may mean that other benefits of these interventions, apart from those on flexibility measures, were missed. The large umbrella search strategy, however, should have still found these articles, and only 3 studies were excluded only on the premise of not having a flexibility measure (see Table, Supplemental Digital Content 1, which lists all articles excluded at the full-text level).

Implications for Research and Clinical Practice

Assessment. Future research is needed to verify whether stretch interventions have a clinically significant effect on the activity, participation, and quality of life of children with neuromuscular disabilities. Several of the included studies note the need to use outcome measures of activity and participation that are meaningful for the child.[28,29,32,35,37,38,43] This is significant in that both clinicians and parents identify quality of life as the most important domain to assess in this population.[47] The majority of stretch intervention studies have focused at the body function and structure level, and it is rare to see a study that assessed all ICF levels at once. When functional measurements were assessed, no difference between experimental and control groups were detected.[30-32] The intervention itself might be ineffective for functional changes; however, an alternative explanation is that the activity and participation measures currently designed may not be reliable, sensitive, or valid enough to detect change during the course of intervention(s). For example, in SRs of activity and participation measures in children with CP, many of the tools did not have sound psychometric properties.[48,49] Thus, it is evident from the literature that even if clinicians and researchers start to implement more activity and participation assessment, further development of outcome measures is needed to address the reliability, sensitivity, and validity of these tools.

Treatment. Another observation when functional measures were assessed was that improvements at the body function level did not correlate with improvements in activity or participation.[29,40] A probable explanation for the lack of carryover between ICF levels uses the motor learning principles of bottom-up and top-down interventions. Bottom-up treatment interventions focus on remediating an underlying impairment or motor deficit, whereas top-down treatment interventions typically use a problem-solving approach to motor skill development or task-specific interventions focused on the direct teaching of a skill.[50] Employing an intervention at the bottom (eg, body structure and function level) and assuming that there is overflow[40] or translation upstream to the activities level[29] is not as logical as implementing an intervention aimed at improving activity and participation and then assessing for change at this level. Distinguishing bottom-up and top-down interventions is important as a recent SR found that the majority of green interventions for children with CP were top-down therapy approaches, aimed at improving activities performance.[29] Thus, not only should clinicians and researchers assess functional and quality of life measures with sensitive tools, but functional interventions should be considered in conjunction with interventions targeted at the body structure and function level. For the clinician, assessment and treatment should be organized according to findings and hypotheses about impairments and limitations of the specific individual client in the context of best evidence, clinician experience, and client values.

Research Considerations. Future studies also need to address study design and rigor to improve the quality of the research and thus the applicability of the knowledge into clinical practice. More specifically, studies need to account for dropouts, blind assessors and report on concurrent activities and therapies. Researchers also need to look at long-term follow-up (eg, >1 y) and report on adverse events and safety considerations, as the majority of the included studies did not address these events. This is paramount because the majority of stretch interventions are considered yellow interventions, meaning that there is low-quality or conflicting evidence supporting the effectiveness of the interventions.[25,26] For all interventions, especially where there is insufficient evidence to support or refute its effectiveness, the possible desirable effects of an intervention have to be weighed against its undesirable effects, such as adverse events and burden of care.[12] Possible negative effects of a stretch intervention may include increased emotional burden on caregivers,[13,16] pain,[15,31,34,36] sleep disturbances,[13,14,31] or even a negative physiological effect.[51] Research on other pediatric populations has demonstrated better coreporting of primary outcome physiological measures along with secondary activity and participation outcome measures including adverse events. For example, extensive research has been conducted on the clinical effectiveness of orthoses for adolescent idiopathic scoliosis along with the coreporting of pain, quality of life, psychological issues, and self-image.[52,53] Similar research should be conducted for children with neuromuscular disabilities. In order to perform high-quality clinical trials, researchers should conduct sample size calculations a priori to provide adequate power. This was only mentioned in one of the included studies of this review.[31] The reporting of sample size calculation in physical medicine and rehabilitation research has been identified as being inadequate given current publication guidelines.[54] Where RCTs are unethical or impossible to conduct,[10] long-term prospective cohorts that address the previously identified methodological weaknesses may be useful. Multicenter trials organized by using databases, such as CP registries, can acquire the large sample sizes needed for this type of research. Following a large group of children over a long period, while using sensitive outcome measures and employing top-down interventions, will likely yield the most meaningful evidence.

CONCLUSION

Three green interventions were found: ankle casting for improving passive ankle dorsiflexion, orthoses for improving gait kinetics and kinematics, and supported standing programs for improvement of lower extremity BMD. All other stretch interventions are yellow interventions for a particular outcome measure, meaning that clinicians can continue to implement these interventions but need to use sensitive outcome measures to see whether these interventions have helped the child reach their goal. No red stretch interventions were found, indicating that, at this time, no interventions need to be discontinued on the basis of ineffectiveness or detrimental effects. Further investigation of stretch interventions is warranted because the gap between clinical practice and the lack of clear scientific evidence can have implications that could influence the future allocation and use of pediatric physical therapy services.

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APPENDIX

Search Strategy

The following search strategy was employed along with relevant MESH terms for each database: "stretches" or "stretch" or "stretching" or "range of motion" or "passive range of motion" or "active range of motion" or "positioning" or "casting" or "casts" or "cast" or "splints" or "splinting" or "splint" or "bracing" or "braces" or "brace" or "yoga" or "orthotics" or "orthoses" or "orthotic" (and) "passive range of motion" or "active range of motion" or "joint mobility" or "flexibility" or "flexible" or "pain" or "quality of life" or "spastic" or "spasticity" or "activities of daily living" or "activity of daily living" or "participation" or "contracture" or "contractures."