

## REVIEW ARTICLE

## THE EVIDENCE-BASE FOR BASIC PHYSICAL THERAPY TECHNIQUES TARGETING LOWER LIMB FUNCTION IN CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW USING THE INTERNATIONAL CLASSIFICATION OF FUNCTIONING, DISABILITY AND HEALTH AS A CONCEPTUAL FRAMEWORK

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**Objective:** This systematic review provides an overview of the effectiveness of basic techniques used in lower limb physical therapy of children with cerebral palsy. It aims to support the development of clinical guidelines for evidence-based physical therapy planning for these children.

**Data sources and study selection:** A literature search in 5 electronic databases extracted literature published between January 1995 and December 2009. Studies were evaluated using the framework recommended by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM), which classifies outcomes according to the International Classification of Functioning, Disability and Health.

**Data extraction:** Three independent evaluators rated the strength of evidence of the effects according to the AACPDM levels of evidence classification, and the quality of the studies according to the AACPDM conduct score system.

**Data synthesis:** A total of 83 studies was selected and divided into categories (stretching, massage, strengthening, electrical stimulation, weight-bearing, balance-, treadmill- and endurance training). Interventions targeting problems at body function and structure level generally influenced this level without significant overflow to activity level and vice versa.

**Conclusion:** The more recent studies evaluating strength training mainly demonstrated level II evidence for improved gait and gross motor function. There was limited evidence for specific information on intensity, duration and frequency of training.

**Key words:** cerebral palsy; physical therapy; evidence-based; ICF.

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### INTRODUCTION

Cerebral palsy (CP) describes a group of disorders of movement and posture that cause activity limitations. CP is attributed

to non-progressive disturbances occurring in the developing foetal or infant brain (1, 2).

Physical therapy (PT) plays a major role in the treatment of children with CP. High intensities and frequencies of therapy are reported (3) and varying approaches and techniques are used. In lower limb treatment, basic techniques, such as passive stretching, massage, muscle strengthening and many others, are frequently used. These basic techniques usually target specific problems at the level of body structure and function, such as range of motion (ROM), strength and muscle tone. More complex treatment approaches, such as neurodevelopmental treatment (NDT), Vojta therapy and Petö therapy are generally based on different principles of motor learning and require specific, specialized training.

A structural basis for PT planning is provided by the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) (4–7). The ICF-CY describes a conceptual framework to report the variety of health information (diagnosis and functioning) and thereby delivers a common language and terminology to describe the child's problems in relation to functions and anatomical properties, activity limitations and participation problems. Physical therapists can use the model to guide the selection of measurement tools, both in goal-setting and decision-making processes and to determine meaningful outcomes.

Limited evidence is available on the effectiveness of different PT interventions in CP. A high-quality review by Anttila et al. (8) included both upper- and lower-limb treatment and included randomized controlled trials (RCTs) only. This review found only limited evidence supporting strength training, constraint-induced movement therapy and hippo-therapy.

In general, little consistency is provided on the use of different techniques in clinical practice of lower limb treatment in CP. While interventions at any of the elements of the ICF model may be important, each using different PT strategies and techniques (9), only limited emphasis is put on differentiation of the outcome effects on the different levels of the ICF.

This systematic review aims to summarize the effectiveness of different basic PT techniques used in children with

CP and to differentiate the outcome effects on the different levels of the ICF.

It thereby aims to identify interaction effects between the different outcome levels of the ICF and to explore the possibilities to develop clinical guidelines in PT treatment of children with CP.

METHODS

Search strategy

A systematic, stepwise search of the literature on PT in CP was performed using the following electronic databases: Web of Science, PubMed, Cochrane Library, Physiotherapy Evidence Database (PEDro) and CINAHL. The general search terms used were: “cerebral palsy” and “physiotherapy”, “physical therapy”, “exercise” and “training”. More specific search terms were: “stretching”, “electrical stimulation”, “electrostimulation”, “massage”, “strength”, “treadmill”, “balance” and “weight bearing”.

Inclusion criteria were: original articles published in peer-reviewed journals between January 1995 and December 2009, focusing on PT interventions targeting lower limb treatment in children and adolescents (<18 years) with CP. Only articles written in English were included. Articles including children with different pathologies or targeting upper limb or trunk only were excluded from the study. In addition, interventions using mixed approaches or techniques and post-surgical interventions were not included in the study.

Based on the title and abstracts of the resulting articles, a first selection resulted in 159 articles. In a subsequent step, all articles were then screened by the first author. Articles not fulfilling the inclusion criteria were withdrawn. If the title and abstract did not provide sufficient information to fulfil the inclusion criteria, the full article was checked. In addition, all case-studies, expert opinions and non-systematic reviews were excluded. As a final step, reference lists of all systematic reviews included in the study were searched and missing articles fitting the inclusion criteria were added. The inclusion of more doubtful articles was discussed with a second and third assessor.

A resulting total of 83 studies was included in the study. A flow-diagram of the selection process is shown in Fig 1.

Data collection

The full texts of all selected articles were read. The following data were extracted: type of PT intervention, numbers of patients included, topographic distribution of cerebral palsy, age of patients, type, frequency and duration of intervention, duration of follow-up, evaluation method and timing, summary of the results and conclusion.

Grouping data

The selected articles were grouped according to the type of intervention used. Interventions at the level of body function and structure were stretching (n=5), massage (n=4), electrical stimulation (n=13), strength training (n=25), endurance and physical fitness training (n=10) and weight-bearing (n=7). Interventions at both body structure and function and activity level were balance training (n=6) and treadmill training (n=13). Interventions using treadmill training were considered as a separate group, since mixed training goals, such as gait function, endurance or strength, made these articles difficult to categorize in one of the previous groups.

Classification and rating of the different outcome measures

Classification of outcome measures, rating of level of evidence and scoring of conduct scores was carried out by 3 independent evaluators: 1 PhD physical therapist (CVdB), 1 research physical therapist (IF) and 1 physical therapist MSc student (LVL).

Classification of the outcome measure

All 3 evaluators rated the outcome of the intervention on the level of the ICF: Body structure and function, Activities and Participation, Personal factors, and Environmental factors.

*Body structures* are defined as the anatomical parts of the body, such as organs, limbs and their components. *Body functions* are the physiological functions of body systems (including psychological functions). *Activity* is the execution of a task or action by an individual. *Participation* is involvement in a life situation. *Environmental factors* make up the physical, social and attitudinal environment in which people live and conduct their lives. They can be viewed as facilitators (positive influence) or barriers (negative influence). *Personal factors* are the particular background of an individual’s life and living, and compromise features of the individual that are not part of a health condition. These factors may include gender, race, age and other health conditions (7).

Level of evidence

The same independent evaluators rated the studies according to their level of evidence using the rating system proposed by the American Academy of Cerebral Palsy and Developmental Medicine (AAPDM) (Table I) (10). Level of evidence describes the potential in a research study design to control for factors, other than the intervention, that may affect the observed outcome. In descending order, the designs decreasingly demonstrate that the intervention, and not something else, is responsible for the observed outcome. Level I evidence is the most definitive for establishing causality, with greatest reduction in bias. Level IV evidence can only hint at causality; level V evidence only suggests the possibility (10).

Reliability between the grading of the levels of evidence assigned by the different evaluators was tested in pairs, using an un-weighted Kappa coefficient. The Kappa coefficient varied between 0.608 and 0.919.

In a second step, all discrepancies were discussed. The raters argued the reasons for the score given. In case an agreement could not be found in this way, the score with the highest frequency was chosen (2 out of 3 raters scoring the same level of evidence). This final consensus score was used in the summary tables.

Quality of the studies

The conduct of the study rating indicates the extent to which a study applied the controls possible within the research design. Quality as-

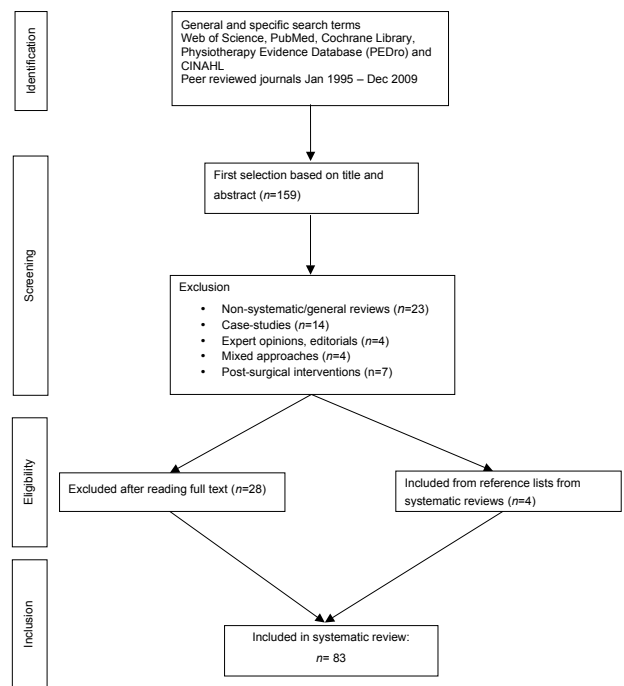


Fig. 1. Selection process.

Table I. Hierarchy of levels of evidence based on research design types (American Academy of Cerebral Palsy and Developmental Medicine)

Level	Intervention (group) studies	Single-subject research design (SSRD)
I	Systematic review of randomized controlled trials (RCTs) Large RCTs with narrow confidence intervals ( $n > 100$ )	Randomized controlled n-of-1 (RCT) Alternating treatment design (ATD) Concurrent or non-concurrent multiple baseline design (MBD) Generalizability if the ATD is replicated across 3 or more subjects and the MBD consists of minimum of 3 subjects, behavior setting. These designs can provide causal interferences
II	Smaller RCTs with wider confidence intervals ( $n < 100$ ) Systematic reviews of cohort studies Outcomes research (large ecologic studies)	Non-randomized controlled concurrent MBD Generalizability if design consists of minimum of 3 subjects, behaviours or settings Limited causal interferences
III	Cohort studies with concurrent control group Systematic reviews of case control studies	Non-randomized, non-concurrent, controlled MBD Generalizability if design consists of minimum of 3 subjects, behaviours or settings Limited causal interferences
IV	Case series Cohort studies without concurrent control group (e.g. historical control group) Case-control study	Non-randomized controlled SSRD with at least 3 phases (ABA, ABAB, BAB, etc) Generalizability if replicated across 3 or more different subjects. Only hints at causal interferences
V	Expert opinion Case study or report Bench research Expert opinion based on theory or physiologic research Common sense – anecdotes	Non-randomized controlled AB SSRD, generalizability if replicated across 3 or more different subjects. Suggests causal interferences allowing testing of ideas

assessment was again performed by the same 3 independent evaluators, using the conduct score system proposed by the AACPD (Table II). For group designs, the conduct of an individual study is judged as “Strong” (yes on 6–7 questions), “moderate” (score 4–5) or weak ( $\leq 3$ ). For single subject designs, the conduct of an individual study is judged as strong (yes on 11–14 questions), moderate (scores 7–10) or weak (score  $< 7$ ). Systematic reviews are also evaluated using this score system, reaching a maximum of 10 points. Inter-rater reliability of the conduct score system was tested resulting in an ICC score of 0.640 for group designs, 0.352 for single subject designs and 0.888 for systematic reviews.

Larger discrepancies were discussed in a similar way to the quality rating score. The answers of the different assessors to the questions were compared and the questions causing the disagreement were traced. Again, the reasons for the scores given were discussed until agreement was found and if no consensus could be found, the score with the highest frequency was chosen. After this discussion, the conduct score was recalculated. These consensus scores were used in the summary tables.

## RESULTS

### Stretching techniques

Two systematic reviews and 3 interventions studies were found that used stretching techniques (Table SI (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)). The intervention studies included one RCT (12) and two single subject designs (11, 13), all with moderate conduct scores. A total of 47 children with CP were included in these studies.

Effectiveness was evaluated on the *level of Body structure* only and demonstrated level II evidence for the positive effect of passive stretching on ROM and spasticity (12, 13). Fragala et al. (11) reported a significant deterioration in passive range of motion (pROM) after non-intervention periods longer than

5 weeks, but no significant change during the intervention periods. A limited positive additional effect was found when stretching was combined with electrical stimulation (12) and heat application (13). In addition, Lee & Ng (13) indicated that sustained stretching of longer duration (minimum 30 s) was preferable to improve ROM and to reduce spasticity of the muscles around the targeted joints.

The mean duration of treatment in the studies was 8.2 weeks (standard deviation (SD) 10.4), using a mean frequency of stretching of 4.5 times per week (SD 2.8).

The 2 systematic reviews each included 7 studies and confirmed the weak evidence on the effectiveness of passive stretching (14, 15).

### Massage

Four studies used different massage techniques in treatment of 125 children with CP (Table SII (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)). On *Body structure and function*, one level II study found a significant positive effect on spasticity, ROM, fine- and gross motor function and cognitive behaviour (16). However, similar effects were reported in the control group and the difference in progress between both groups was not evaluated. One level IV study found no significant effects on pROM and stretch reflexes (17), another study demonstrated significant improvements on Psychological well-being and Satisfaction with Life scales (18). A similar study showed that children enjoyed the relaxing aspects of massage and reported a number of other improvements in their health, such as muscle relaxation, mobility and bowel movements and reduced pain (19).

On *Activity and Participation level*, Hernandez-Reiz (16) found significant positive effects on the Development Programming and

Table II. Assessment of study conduct as proposed by the American Academy of Cerebral Palsy and Developmental Medicine

*Conduct of group design studies*

Were inclusion and exclusion criteria of the study population well described and followed?

Was the intervention well described and was there adherence to the intervention assignment? (for 2-group designs, was the control exposure also well described?) Both parts of the question need to be met to score "yes".

Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?

Was the outcome assessor unaware of the intervention status of the participants (i.e. were the assessors masked)?

Did the authors conduct and report appropriate statistical evaluation including power calculations? Both parts of the question need to be met to score "yes".

Were dropout/loss to follow-up reported and less than 20%? For 2-group designs, was dropout balanced?

Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?

*Conduct questions for single-subject designs*

Was/were the participant(s) sufficiently well described to allow comparison with other studies or with the reader's own patient population?

Were the independent variables operationally defined to allow replication?

Were intervention conditions operationally defined to allow replication?

Were the dependent variables operationally defined as dependent measures?

Was inter-rater or intra-rater reliability of the dependent measures assessed before and during each phase of the study?

Was the outcome assessor unaware of the phase of the study (intervention vs control) in which the participant was involved?

Was stability of the data demonstrated in baseline, namely lack of variability or a trend opposite to the direction one would expect after application of the intervention?

Was the type of SSRD clearly and correctly stated, for example, A-B, multiple baseline across subjects?

Were there an adequate number of data points in each phase (minimum of 5) for each participant?

Were the effects of the intervention replicated across 3 or more subjects?

Did the authors conduct and report appropriate visual analysis, for example, level, trend and variability?

Did the graphs used for visual analysis follow standard conventions, for example x- and y-axes labeled clearly and logically, phases clearly labeled (A, B, etc.) and delineated with vertical lines, data paths separated between phases, consistency of scales?

Did the authors report tests of statistical analysis, for example celeration line approach, two-standard deviation band method, Cstatistic, or other?

Were all criteria met for the statistical analyses used?

*Conduct scores for systematic reviews*

Were the search methods reported?

Was the search comprehensive?

Were the inclusion criteria reported?

Was selection bias avoided?

Were the validity criteria reported?

Was validity assessed properly?

Were the methods used to combine studies reported?

Were the findings combined appropriately?

Were the conclusions supported by the reported data?

What was the overall scientific quality of the overview?

SSRD: single-subject research design.

Videotaped interactions, but Macgregor et al. (17) could not find significant improvements in gross motor function measurement (by the Gross Motor Function Measure; GMFM).

On *Environmental factors*, one level IV study demonstrated improved parent anxious behaviour and depressed mood, stress, self-efficacy and satisfaction with life, together with subjective feelings of their child's well-being and functioning after a 4-month training and support programme involving basic massage (18).

The mean duration of the massage programme was 8.3 weeks (SD 2.9 weeks). Two studies reported a mean training frequency of twice per week, while in the two other studies the frequency was not specified.

*Electrical stimulation*

Of the 13 articles using electrical stimulation, 3 interventions used threshold electrical stimulation (TES, lower level electrical stimulation causing no visual contraction, < 10 mA) (20–22) and 8 intervention studies used neuromuscular electrical stimulation (NMES, higher level electrical stimulation causing visual contraction, > 10 mA) (23–32). An overview of the studies using TES and NMES is given in Tables SIII and IV (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>).

The studies using TES included a total of 59 children. The conduct of these studies varied from moderate (18) to weak (20–22).

On *Body structure and function*, two level II studies based on a randomized controlled design found no significant long-term effect of TES on strength, ROM, deep tendon reflexes or muscle cross-sectional area of the m. quadriceps femoris and m. tibialis anterior (20, 21). Level IV evidence was found for the positive effect of 1 month of TES on ROM and selectivity (22).

On *Activity level*, only level IV evidence was found for standing/hopping on 1 foot (22).

Nevertheless, parents and children rated an important subjective feeling of improvement along with a high feeling of satisfaction (20, 21).

Seventy-eight children with CP were included in the 10 studies evaluating the effectiveness of NMES. On *Body structure and function*, level II effectiveness was mainly demonstrated on strength (27), muscle cross-sectional area (27), ROM at the extremities (26, 12, 28) and the trunk (23) and on spasticity (28, 12, 29). On *Activity level*, two level II studies showed significant better effects of NMES on the quality of gait and walking velocity compared with the control group (24, 28). In a RCT design, Park et al. (23) found significantly greater improvement on the sitting dimension of the GMFM in a group of children receiving electric stimulation of the abdomen and the back extensors compared with a control group. This positive effect on gross motor function could not be demonstrated by van der Linden (25) and Kerr et al. (30).

On *Participation level*, Kerr et al. (30) measured statistically significant improvements on the Lifestyle Assessment Questionnaire and this provided level II evidence.

The studies using TES were rather long-term and had a mean duration of 40.1 weeks (SD 23.9). The duration of the studies using

NMES varied from 3 days to 12 weeks only, with a mean of 6.1 weeks (SD 4.7 weeks). The mean frequency of treatment was 4.95 and 9.37 times per week for TES and NMES, respectively. One RCT compared TES with NMES, but showed no significant difference on body function and structure or on activity level (30).

One study compared the effectiveness of NMES with 15 s isometric voluntary contractions and reported improvements in strength and muscle morphology of the *m. quadriceps femoris*, but not in the *triceps surae* (27). In contrast to NMES, volitional isometric strength training did not significantly influence walking speed.

### Strength training

Twenty intervention studies and 6 systematic reviews evaluated the effect of strength training in children with CP. The studies were subdivided according to the specific modality of strength training used: isotonic strength training, functional strengthening exercises, isokinetic strength training, isometric strength training and mixed forms of strength training.

Table SV (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>) summarizes the 11 studies evaluating the effectiveness of *isotonic strength training*. A total of 128 children with CP was included.

On the *level of Body function and structure*, 7 out of 8 studies evaluating the effect of isotonic strength training on strength registered significant improvements (32, 33, 35, 38, 40–42), including one study demonstrating level II evidence. Other level II evidence was demonstrated on economy of movement (39). Hints of causality or level IV evidence were found on muscle volume (42), energy expenditure (36) and ROM (40). A RCT by Dodd et al. (37) demonstrated a significantly lower self-concept of scholastic competence and social acceptance in a group of 10 children with CP participating in a home-based strength training programme compared with a control group. However, another RCT by Unger et al. (39) reported a significant improvement in the perception of body image of a group of children using circuit training.

On *Activity level*, level II evidence was found for the effectiveness on gait (39, 41) and gross motor function (41).

No studies evaluated the effect of isotonic strength training on *Participation level*.

The studies using isotonic strength training reported only limited description of the resistance used during training. Three studies used a resistance of 65% of the one repetition maximum (RM) and one study used 80% of RM. The mean training frequency was 3.8 times per week, with a duration of 12.4 weeks. All studies using isotonic strength training scored moderate to weak based on conduct scores.

An overview of the different studies using *functional, isokinetic, isometric and mixed strength training* is provided in Table SVI (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>).

Four studies evaluated the effects of functional strength training and included a total of 40 children with CP. Functional strength training often consisted of a home-based training programme, including task-oriented strength training during

functional activities. Effects of functional strength training were demonstrated on *Activity level*: gross motor function significantly improved in one level II study (46). A high-quality RCT by Dodd et al. (44) demonstrated significant effects on muscle strength, but only a trend towards improved gross motor function. Level IV evidence was also recognized for participation in school and leisure activities and self-perception (45).

The studies frequently used 8–10 repetitions and had a mean duration of 5.4 weeks at a frequency of 2.4 times per week.

Level II evidence was also found for *isokinetic strength training* and this on all levels of the ICF (47, 48).

None of the studies using any form of strength training demonstrated an increase in spasticity.

Only 9 studies used a follow-up period, with an average duration of follow-up of 32 weeks. All these studies recognized a decrease in training results at all levels after a non-training follow-up period.

The studies demonstrate strong evidence of all forms of strength training on body structure and function, but to a lesser extent on activity and participation.

Table SVII (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>) provides an overview of the systematic review evaluating the effectiveness of strength training (50–55). The results are confirmed by all except one meta-analytic review on strength training by Scianni et al. (55), stating that muscle strengthening is not effective in children with CP. However, since Scianni also included biofeedback and electrical stimulation under interventions of muscle strengthening the results cannot be compared with the results of this systematic review.

### Endurance training

Eight intervention studies and two systematic reviews evaluated the effect of endurance training for children with CP, with a main focus on aerobic capacity (Table SVIII (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)) (56–65). A total of 108 children were included in the intervention studies. Modalities used varied from walking and running to circuit training, cycling and mixed gymnastic exercises.

On *Body function and structure*, significant level II effects were mainly found for aerobic capacity (56, 60), anaerobic capacity (60), muscle strength (60), agility (60) and oxygen uptake (62). On *Activity level*, level II significance was reported for improvements in gross motor function (60, 62). Improved *Participation* measured by use of the Children's Assessment of Participation and Enjoyment (CAPE) was reported by Verschuren et al. (60) 2007. Endurance training positively influenced quality of life (QoL) measured by the Health-Related Quality of Life (HRQoL) (level II evidence) (63) and self-perception of physical appearance (level IV evidence) (57).

The mean duration of the training programmes was 13.3 weeks (SD 13.1). Four studies used a follow-up period with a mean of 9.3 weeks (57, 59, 60, 63). Weak to moderate conduct scores were found for all interventions.

All studies reported little to no deterioration when comparing the measurements after follow-up with the immediate post-training results. The two systematic reviews on aerobic training (64, 65) both confirm the overall positive effects of endurance training in children with CP. Nevertheless, both reviews report low methodological qualities of the studies and lack of information on the effectiveness on daily activity and participation level.

#### *Weight-bearing*

Of the selected studies, 6 intervention studies and 1 systematic review investigated the effectiveness of weight-bearing activities in children with CP (Table SIX (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)) (66–72). A total of 89 children was included in the studies. Included modalities were standing programmes in different types of walking frames and PT programmes focusing on weight-bearing exercises. The studies were of very low to moderate quality.

On *Body structure and function*, two level II studies demonstrated a positive effect of increased weight-bearing on bone mineral density (66, 69). However, level IV evidence was demonstrated for a positive effect on bone mineral density (68), ROM (67, 70) and bowel activity (71). A positive effect on the behavioural characteristics and personal feeling of improved daily functioning was demonstrated with level IV evidence (68).

On *Activity and Participation level*, two level II studies evaluated the effectiveness of weight-bearing programmes on activities of daily living (ADL), functional performance and walking speed, but this did not yield statistical significance (70, 71).

The duration of the standing programmes was very variable and ranged from 2 weeks to 9 months, with a frequency ranging from 2 to 5 times per week.

#### *Balance training*

Five intervention studies and one systematic review evaluated the effectiveness of different forms of balance training for a total of 43 children with CP, each using different modalities (Table SX (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)) (73–78). Three studies used a moveable platform to train balance, with one study using additional visual feedback (74–76).

On *Body structure and function*, one study targeting specific sitting balance found significant positive effects on trunk control, as well as in the extremities (73).

The studies mainly evaluated on *Activity level* and registered task-specific improvement meaning a reduced centre of pressure (CoP) area and a reduced stabilization time. One level II study also reported a more symmetrical walking pattern after a 6-week training programme and one level III study reported visible improvements in GMFMD (no statistics) after a 5-day training programme. One study using multidimensional random perturbation also evaluated on activity level only and found level II evidence for significant improvement on gross motor function together with an improved mechanical efficiency (77).

The systematic review including 12 studies, reported low-quality experiments providing low levels of evidence on the effectiveness of studies targeting postural control and balance (78).

#### *Treadmill training*

In the last years, treadmill training has become a very popular therapy method in CP. Three systematic reviews and 10 interventions studies (including 61 children) evaluated the effect of treadmill training in children with CP, with different objectives, such as gait rehabilitation, functional improvement and improvement of aerobic capacity (Table SXI (available from <http://www.medicaljournals.se/jrm/content/?doi=10.2340/16501977-0983>)) (79–91). Training modalities varied strongly among the different modalities, such as body weight support, treadmill speed (range 0.25 km/h up to 5 km/h or as fast as possible) and duration (10–30 min).

On the level of *Body structure and function*, weak level IV evidence was reported for positive effects on energy expenditure (85) and H-reflex (87).

Effects of treadmill training in children with CP were mainly limited to *Activity level*. Two level II (81, 83) and 5 level IV studies (82, 84, 85, 87, 88) found significant effects on different gait parameters. One level II (84) and one level IV (88) reported a significant positive effect on gross motor function.

The mean duration of the training periods was 6.4 weeks (SD 5.7 weeks) at a mean frequency of 6.7 times per week (SD 5.21). Three studies used a follow-up period, with a mean of 10.08 weeks. Results at follow-up are concurrent with the results of endurance training and also show very limited deterioration during the follow-up period.

#### *Summary of the results*

Table III provides an overview of the evidence of the different interventions. For this summary table, the most commonly used outcome parameters on the different levels of the ICF were selected.

It compares the number of studies finding statistical significant effects on this parameter to the total number of specific interventions evaluating this parameter. Some level II RCTs did not statistically compare between-group differences in progress. Therefore, in Table III, these studies are assigned to the level IV studies. Although the design of these studies could be rated as level II, statistically significant differences within in the experimental or control group only can be compared with the analysis of a level IV case series.

On the level of *Body structure and function*, the parameters spasticity, ROM, bone mineral density, energy expenditure, strength and muscle morphology were the most commonly used. As demonstrated in Table III, level II evidence was most frequently demonstrated for the effectiveness of all forms of strength training on muscle strength. More conflicting evidence was found for the influence of NMES and endurance training on muscle strength. Weight-bearing positively influenced bone mineral density.

Table III. Overview of the number of studies demonstrating level II, III and IV evidence

Level II evidence			
Body structure and function	<i>Muscle tone</i>	<i>Bone mineral density</i>	<i>Energy expenditure/movement efficiency</i>
	Stretching, 1/1 (12)	Weight-bearing, 2/2 (69, 72)	Isotonic strength training, 1/1 (39)
	NMES, 1/2 (24, <b>28</b> )	<i>Strength</i>	Functional strength training, 1/1 (46)
	TES, 0/2 (20, 21)	NMES, 1/3 (25, <b>27</b> , 30)	Balance training, 1/1 (77)
	<i>ROM</i>	TES, 0/1 (20)	<i>Muscle morphology/cross-sectional area</i>
	Stretching, 1/1 (12)	Isotonic strength training, 1/1 (41)	Isometric strength training, 1/1 (27)
	NMES, 1/4 ( <b>12</b> , 23, 24, 25)	Functional strength training, 1/2 ( <b>44</b> , 46)	NMES, 1/1 (27)
	TES, 0/2 (20, 21)	Isometric strength training, 1/1 (27)	TES, 0/1 (21)
		Endurance training, 1/2 (56, <b>60</b> )	
Activity and Participation	<i>Gait</i>	<i>Gross motor function</i>	<i>Participation</i>
	NMES, 1/3 ( <b>24</b> , 25, 27)	NMES, 1/3 ( <b>23</b> , 25, 30)	Endurance training, 1/1 (60)
	TES, 0/1 (20)	TES, 0/2 (20, 21)	NMES, 1/1 (30)
	Isotonic strength training, 2/2 (39, 41)	Isotonic strength training, 2/2 (39, 41)	
	Isometric strength training, 1/1 (27)	Functional strength training, 1/2 ( <b>44</b> , 46)	
	Treadmill training, 1/1 (83)	Endurance training, 2/2 (60, 62)	
	Balance training, 1/1 (75)	Balance training, 0/1 (77)	
Level III and IV evidence			
Body structure and function	<i>Muscle tone</i>	<i>Bone mineral density</i>	<i>Energy expenditure/movement efficiency</i>
	Stretching, 1/1 (13)	Weight-bearing, 1/1 (74)	Isotonic strength training, 1/2 (34, <b>39</b> )
	NMES, 2/2 (28, 29)	<i>Strength</i>	Treadmill training, 1/1 (85)
	Treadmill training, 0/1 (81)	Isotonic strength training, 6/7 ( <b>32</b> , <b>33</b> , 34, <b>35</b> , <b>38</b> , <b>40</b> , <b>42</b> )	Isokinetic strength training, 0/1 (47)
	Massage, 1/2 ( <b>16</b> , 17)	Functional strength training, 1/1 (43)	<i>Muscle morphology/cross-sectional area</i>
	<i>ROM</i>	Isokinetic strength training, 2/2 (47, 48)	Isotonic strength training, 1/1 (13)
	Stretching, 2/2 (11, 13)	Endurance training, 1/1 (57)	
	TES, 1/1 (23)		
	NMES, 2/2 (28, 29)		
	Isotonic strength training, 1/3 (35, <b>40</b> , 42)		
	Massage, 1/2 ( <b>16</b> , 17)		
	Weight-bearing, 1/3 ( <b>70</b> , <b>71</b> , <b>73</b> )		
Activity and Participation	<i>Gait</i>	<i>Gross motor function</i>	<i>Participation</i>
	NMES, 1/1 (28)	TES, 0/2 (20, 21)	Massage, 3/3 (16, 18, 19)
	Isotonic strength training, 5/7 ( <b>32</b> , <b>34</b> , 35, <b>36</b> , <b>38</b> , <b>40</b> , 42)	Isotonic strength training, 3/4 ( <b>34</b> , <b>38</b> , <b>40</b> , 42)	Treadmill training, 1/2 ( <b>84</b> , 88)
	Functional strength training, 1/1	Functional strength training, 1/1 (43)	<i>Environmental factors</i>
	Endurance training, 1/1 (63)	Isokinetic strength training, 2/2 (47, 48)	Massage, 1/1 (18)
	Treadmill training, 6/7 (79, <b>81</b> , <b>82</b> , <b>84</b> , <b>85</b> , <b>87</b> , <b>88</b> )	Endurance training, 2/2 (61, 63)	Isokinetic strength training, 1/1 (48)
	Isokinetic strength training, 1/1 (48)	Treadmill training, 2/5 (79, <b>81</b> , 84, 85, <b>88</b> )	Weight-bearing, 1/1 (74)
		Massage, 1/2 ( <b>16</b> , 17)	<i>Quality of life</i>
		Balance training, 1/2 ( <b>73</b> , 74)	Treadmill training, 1/1 (86)
			Isokinetic strength training, 1/1 (48)

... /... indicates the number of studies reaching significant treatment effects vs the total number of studies evaluating the effect of the intervention on that specific parameter. In case of conflicting evidence, the references demonstrating significant effects are bold.

ROM: range of motion; NMES: neuromuscular electrical stimulation; TES: threshold electrical stimulation.

On the level of activity and participation, level II evidence for positive effects on different gait parameters were also demonstrated by different forms of strength training. In addition, treadmill training, NMES and balance were methods that significantly improve gait. The same is valid for effectiveness on gross motor function. Limited evidence was available on the most useful methods to improve participation, but this is mainly because studies rarely used participation as an outcome measure.

DISCUSSION

This systematic review evaluated the effectiveness of different basic PT techniques used in the treatment of children with CP. Eighty-three articles were included, comprising 27 RCTs, 16 systematic reviews, 11 single-subject designs and 29 prospective, non-randomized trials. One study demonstrated level I evidence, 44 studies level II, 2 studies level III and 34 studies level IV. Since case-studies and non-systematic reviews were excluded, only 2

studies were included with level V evidence. These numbers demonstrate that, high-quality research is possible and being done.

It should be noted that studies demonstrating high levels of evidence did not necessarily reach high conduct scores. Even well-designed RCTs often showed methodological difficulties, such as a small number of participants, a lack of control over different confounding variables and an inappropriate description of the exercises used in the control group. Statistically, RCT designs often missed an appropriate comparison of between-group differences in treatment effects. On the other hand, some single-subject research designs (SSRDs) and prospective interventions used a relatively high number of participants and reached high conduct scores with an adequate control over confounding variables. For this reason, lower level of evidence designs were also included in this systematic review. Nevertheless, the authors support the AACPD statement that level IV and V studies only hint at causality and the results of these studies should be interpreted very cautiously.

Low to moderate agreement was found for the rating system on the conduct score system and this was specifically a problem for the SSRDs. The evaluators encountered difficulties when scoring the last questions regarding visual and statistical analysis. These questions were obviously more open for interpretation and therefore more subject to individual opinion.

Other weaknesses of the conduct score system was the fact that it did not consider the number of subjects in the studies and also did not take into account that some questions may be more critical than others. The conduct scoring system therefore was found to be valuable in giving a general score for the quality of the interventions, but should also be interpreted cautiously.

Only one study reported adverse effects. Dodd et al's (37) results suggested that participation in a relatively short home-based strength training programme may have an inhibitory effect on the self-concept of children with CP. Despite the inhibitory effect, self-concept in the experimental group remained positive after strength training, suggesting that clinicians should not be overly concerned about the psychological effects of the intervention (37). However, this study does demonstrate that we have to appreciate the fact that children with CP may show different values from their therapists.

A wide variety of frequencies and intensities were used in the intervention studies, and thus the optimal training modalities remain open for discussion. For strength training, many different types of resistive exercises were used and in physical fitness or endurance training, different training heart rates and durations of trainings were used. This indicates a need for structured research comparing different training intensities, which is necessary to develop more specific clinical guidelines.

Strength interventions often do not respect adequate training principles according to the training recommendations by the National Strength and Conditioning Association (92): the age of the children is often not appropriate, the training intensity is often insufficient or the duration of training is too short. This might be another reason why Scianni et al. (55) and Verschuren et al. (93) did not find significant results regarding the effectiveness of strength training.

Another important issue is the fact that a lack of evidence does not always reflect a lack of effectiveness. In particular, the lack of control over confounding variables was often a major problem limiting the studies to demonstrate evidence.

Careful interpretation is advised, considering the large numbers of studies evaluating, for example, strength training in comparison with a much lower number of studies evaluating the effectiveness of massage or stretching. In addition, a higher number of studies evaluated the effectiveness of the level of body structure and function in comparison with the number of studies using outcome parameters on activity and participation level. Older studies in particular often restricted their outcome parameters to the level of body structure and function.

None of the studies differentiated outcome effects according to the age or functional level of the participants. However, many studies use a wide age range and a large variety in functional level in their inclusion criteria. Nevertheless, a young child with mild unilateral CP might benefit from a completely different treatment approach with different techniques from a teenager with severe bilateral CP.

Concerning effects on gross motor function, in particular, it is important to consider the expected outcome related to age and GMFCS level. A younger child with a GMFCS level I or II usually develops more in the natural course of development than an older child with a GMFCS level IV or V (94–96).

A restriction of the ICF model was found in the scoring of the parameter QoL, a problem that was previously reported in the literature (97). The World Health Organization (WHO) defines QoL as "an individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, standards and concerns" (98). The WHO suggests that QoL can be broken down into different domains: physical, psychological, social relationships and environmental. Given the inclusion of personal and environmental factors in its model of functioning and disability, the ICF encompasses all domains that comprise human life and thus impact QoL. The components of functioning and contextual factors can be seen as the various manifestations of a person's QoL. In this systematic review, not many interventions used QoL measures in their outcome parameters. However, to be able to give this discussion a place, the authors have scored these parameters under a separate category of QoL.

This systematic review did not include mixed or eclectic approaches. This decision was based mainly on the methodological limitations of mixed interventions to distinguish the causes of the different reported effects. Nevertheless, the results of this review indicate that, in clinical practice, one might have to combine different techniques and methods in order to have a meaningful outcome.

Summarizing the effectiveness of all these interventions demonstrates that the ICF model provides a good model to evaluate the effectiveness of different physiotherapy interventions for CP. The results reveal very limited interactions between the different levels of the ICF. In general, interventions targeting problems at body function and structure, mainly influence body structure and function with only limited overflow to activity level. Stretch-



ing only significantly improved ROM and spasticity, without evidence of improved ROM during walking or other functional activities. Only for the different forms of strength training and for NMES, effectiveness was also demonstrated on activity level. Likewise, interventions at activity level directly influenced motor functions, but limited evidence was available for direct influence on impairment problems such as strength, ROM or spasticity. It was even more challenging to look for interaction effects of participation and other ICF levels.

This leads to the conclusion that a targeted treatment approach based on a complete and extensive evaluation on all levels of the ICF is necessary to create an appropriate treatment plan. In true efforts to increase independence and to prevent secondary disabilities, the child should be considered as a whole. The PT treatment plan should therefore comprise specific goals and exercises targeting the individual problems specified on each level of the ICF. Specific motor learning strategies might be necessary to integrate the different components of impairment level during functional activities. The effectiveness of different general conceptual approaches will therefore be investigated in a subsequent paper (100). The aim of developing further specific clinical guidelines cannot yet be fulfilled with the present paper. Only a limited number of clinical messages could be deduced. Developing clinical guidelines should, however, be an important extra step, to be performed in a systematic way and based on both literature and expert opinion.

### CONCLUSION

- Interventions targeting problems at body function and structure generally influenced this level without significant overflow to activity level and vice versa.
- Stretching can be useful to improve ROM in children with CP. The stretch should preferably be maintained for a minimum of 30 s.
- Conflicting evidence is available on the effectiveness of electrical stimulation strength. NMES (visual contraction, > 10mA) is preferable.
- Strength training is very effective in improving muscle strength and into a lesser extends to improve gait and motor function. Resistance of 65–80% of one RM seems to be well tolerated by the children. Effects seem to be lost relatively quickly after stopping the programme.
- Depending on the modalities used, treadmill training is beneficial in improving gait and endurance in children with CP. With partial body weight support, treadmill training can be effective in very young children with CP.
- Endurance training is useful to improve aerobic endurance. Effects seem to last minimally as long as the training programme itself. A training heart rate of 75% of the maximum heart rate seems to be well tolerated.
- Massage improves the feeling of well-being in children with CP and their parents.
- Balance training is optimally trained in a task-specific context.
- Weight-bearing is useful to improve bone mineral density in children with CP.

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