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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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ABSTRACT

OBJECTIVES: The primary objective is to systematically evaluate the evidence for the effectiveness of task-specific training (TST) of gross motor skills for improving activity and/or participation outcomes in ambulant school aged children with cerebral palsy (CP). The secondary objective is to identify motor learning strategies reported within TST and assess relationship to outcome.

DESIGN: Systematic review

METHOD: Relevant databases were searched for studies including; children with CP (mean age >4 years and >60% of the sample ambulant); TST targeting gross motor skills; and activity (skill performance, gross motor function and functional skills) and/or participation-related outcomes. Quality of included studies was assessed using standardised tools for risk of bias, study design and quality of evidence across outcomes. Continuous data were summarised for each study using standardised mean difference (SMD) and 95% confidence intervals (CI).

RESULTS: Thirteen studies met inclusion criteria: eight randomised controlled trials (RCT), three comparative studies, one repeated-measures study and one single-subject design study. Risk of bias was moderate across studies. Components of TST varied and were often poorly reported. Within-group effects of TST were positive across all outcomes of interest in 11 studies. In RCTs, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes (one study: Life-HABITS performance SMD = 1.19, 95% CI 0.3 to 2.07, p< 0.001; Life-HABITS satisfaction SMD = 1.29, 95% CI 0.40 to 2.18, p =0.001) while no difference or negative effects were found for gross motor function. The quality of evidence was low-moderate overall. Variability and poor reporting of motor learning strategies limited assessment of relationship to outcome.

CONCLUSIONS: Limited evidence for TST for gross motor skills in ambulant children with CP exists for improving activity and participation-related outcomes and recommendations for use over other interventions are limited by poor study methodology and heterogeneous interventions.

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KEY MESSAGES

What is known about the subject

- Strong evidence for motor interventions involving task specificity for functional mobility in adults post stroke and for upper limb function in children with cerebral palsy exists
- The effectiveness of task-specific gross motor skills training in ambulant school aged children with cerebral palsy has not been systematically evaluated or synthesised

What this study adds

- A low-moderate overall quality of evidence was found for task-specific gross motor skills training for ambulant school aged-children with cerebral palsy
- Limited evidence for task-specific training to improve specific skills performance, functional skills and participation-related outcomes exists
- While clear recommendations for use of task-specific training over other interventions are limited, ways to strengthen the evidence in future studies are identified

Short title: Task-specific gross motor skills training in CP

Key words: cerebral palsy, task-specific training, gross motor skills, activity, participation

ABBREVIATIONS

CP	Cerebral palsy
GMFCS	Gross motor Function Classification System
ICF	International Classification of Functioning, Disability and Health
TST	Task-specific training
AACPDM	American Academy of Cerebral Palsy and Developmental Medicine
SMD	Standardised mean difference
CI	Confidence interval
RCT	Randomised controlled trial
NDT	Neurodevelopmental therapy
GMFM	Gross Motor Function Measure

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PEDI Pediatric Evaluation of Disability Inventory

Cerebral palsy (CP) is an umbrella term used to describe a group of disorders of movement which cause varying degrees of activity limitations[1]. The most widely used means for classifying gross motor function in children with CP is the Gross Motor Function Classification System (GMFCS)[2]. Children classified GMFCS I are able to walk and run but have limitations with speed, balance and coordination whilst children classified GMFCS V are transported in a wheelchair in all settings. Although the focus of the GMFCS is on functional mobility, the realm of gross motor activities undertaken by children is much broader. Gross motor skills involve movement of the large muscles of the limbs or the whole body including tasks such as sit to stand, ball skills, bicycling and walking[3]. Development of gross motor skills underpins functional, play and social activities across childhood and complex movement skills required for sports in older children[3]. In children with CP, limitations in gross motor function increase as GMFCS level increases, however, children at all GMFCS levels (I-V) participate, on average, less in physical activities than their typically developing peers[2]. This is an issue because of the known poor health outcomes in adulthood due to inactivity in childhood[4]. Effective interventions tailored to GMFCS levels and developmental stages are required to improve these outcomes for children with CP.

The International Classification of Functioning, Disability and Health (ICF) has become the common language for clinicians, researchers and families for understanding the effect of CP on the individual and for targeting interventions[5,6]. Where interventions previously focused on remediating limitations at the body structures and functions level, there has been a more recent acknowledgement of the importance of the effect of interventions within the activity and participation domains[7 8]. That is, the carrying out of tasks or skills by the child (activity) and their involvement in life situations (participation)[5]. Clinicians working with children with CP need guidance from evidence synthesis to implement effective means of improving physical skills *and* improve the uptake of these skills in the child's daily life. The historical bias towards impairment-focused motor interventions yielded few effective treatments[6 9 10] thus more functional approaches have emerged.

Task-specific training (TST) involves practice of context-specific tasks where the intervention focuses on the skills needed for a task(s)[11] - there is similarity between the training task and the goal of the intervention. Although Level I evidence exists for TST to improve gross motor activities in adults after stroke[12], the majority of high level evidence for

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interventions involving task specificity in children with CP relates to training of upper limb or fine motor activities[6 13] with limited evidence for training of gross motor skills. TST inherently involves principles of motor learning with components including context, practice and dosage[11]. Other motor learning strategies, such as feedback and task modification, have the potential to optimise TST, however, this has not been systematically studied[14]. Context is a key of TST – it should involve varied components depending on the requirements of the skill, the environment and the function of the child[15]. Moreover, training for a child of higher-level motor function (e.g. GMFCS I-III) should be targeted towards different skills compared to training with a child of lower-level motor function (e.g. GMFCS IV-V). Similarly, children of different ages and developmental stages have varying skill and learning capabilities, and physical demands placed on them by their context[16].

Previous systematic reviews of motor interventions in children with CP have been broad in terms of ages (including infants and children) and motor function (all GMFCS levels included), while interventions under examination have been heterogeneous[17 18]. This lack of specificity necessarily limits generalizability and the ability to draw conclusions around effectiveness. Additional reviews examining more specific foci are required to help guide decision-making about the best intervention for the individual child at a particular time and/or for specific motor goals.

TST may be a promising approach for ambulant children who have specific gross motor skills goals that are relevant to school aged children. However, there has been no systematic review of the effectiveness of task-specific gross motor skills training in this population. The primary aim of this study is to evaluate and synthesise the evidence for the effectiveness of task-specific gross motor skills training in ambulant children aged 4 – 18 years with CP for activity and participation outcomes. The secondary aim of this study is to identify motor learning strategies reported within TST and assess relationship to outcome.

METHOD

Eligibility criteria

Published studies were included if they met all of the following criteria:

1. Level of evidence: Studies categorized as level II-IV using the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) levels of evidence[19]. All group designs were included, as well as studies classified as single subject designs (n of 1) involving over 10 participants per study.

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2. Population: The majority (i.e. > 60%) of the participants represented were ambulant children with CP (GMFCS I-III) and the mean age of sample was four – 18 years.
3. Interventions: TST of gross motor skills where there was similarity between the training task and the goal of the intervention, including those interventions described as involving motor learning strategies/coaching, goal-directed training, activity focused training and/or functional skills training. Any duration or intensity of TST was included.
4. Comparison: Studies comparing TST to another intervention, another type of TST or no intervention.
5. Outcomes: Activity outcomes including; gross motor skill performance (specific to the task being trained or other gross motor task to assess for transferability), gross motor function and functional skills; and participation-related outcomes. Only studies reporting outcomes separately for children with CP.

Exclusion criteria: TST was applied within a combined intervention approach and the influence of TST could not be isolated (e.g. Botulinum Toxin-A, virtual reality, treadmill training, orthoses or robotics), greater than ten percent of the intervention was passive or where the article was not in English.

Search strategy

Relevant articles were identified by searching Medline, EMBASE, CINAHL, Psycinfo, SPORTDiscus and PubMed with all searches limited to articles published in peer-reviewed journals in English but not limited by article type. A search was conducted in June 2016 and search terms were tailored for each database. Reference lists of included studies and related narrative or systematic reviews were also searched. See Appendix 1 for full search strategy for Ovid databases (Medline, EMBASE and PubMed).

Study selection, data extraction and quality assessment

Study selection, data extraction and quality assessment was completed by two authors (RT and CB) independently, with a third author (AS, AH or JM) used to resolve any disagreements. If inclusion was uncertain from abstract, the full text was retrieved.

For included studies, data were extracted using a customised form based on the Cochrane recommendations[20]. Data extracted included: study details (author, year, country, funding), study design, AACPDM level of evidence[19], characteristics of the study sample,

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detailed TST characteristics (including motor learning strategies), comparison intervention characteristics, outcomes measured and associated ICF domain, effects of the intervention and author conclusions. Reported motor learning strategies in all TST, including when studies involved a comparison of two or more TST approaches, were identified using pre-defined motor learning strategy codes based on current literature[21 22]. Authors were contacted for complete data extraction when needed.

Intervention replicability was assessed using relevant components of the Template for Intervention Descriptions and Replication (TIDieR) Checklist[23]. Risk of bias for individual studies was assessed using the Cochrane Risk of Bias Tool[20]. The Grading of Recommendations Assessment Development and Evaluation (GRADE) system, was used to assess the quality of the evidence across outcomes[24].

Data analysis and reporting

Analyses were conducted using Cochrane Review Manager Software RevMan 5.3. Only data from level II studies comparing TST to non-task specific interventions were included in quantitative analysis to ensure comparison was between studies of similar design. Outcomes measured were reported under the ICF domains of body structure and function, activity (skill performance, gross motor function and functional skills) and participation. Continuous data were summarized for each study within outcomes of interest using standardized mean difference (SMD) and 95% confidence intervals (CIs). A SMD of 0.2 was considered small, 0.4-0.6 moderate and 0.8 a large effect size[20]. Given the heterogeneity between studies in tasks and characteristics of the TST and comparison interventions, meta-analysis was not undertaken.

AACPDM level of evidence and study design were used to systematically report the results. Activity outcomes were organised into 3 categories; gross motor skill performance, functional skills and gross motor function whilst participation-related outcomes were grouped together.

RESULTS

Following removal of duplicates, the titles and abstracts of 1247 studies were screened and 145 full-text articles were retrieved for full appraisal (Figure 1). Thirteen studies involving 405 participants met inclusion criteria and underwent qualitative analysis with six of these

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studies involving 237 individuals also included in quantitative analysis. Characteristics of included studies are summarised in Table 1.

The 13 studies included 12 group designs and one single-subject design (level I). The group designs involved eight randomised controlled trials (RCTs) (level II), three comparative studies with concurrent controls (level III) and one repeated measures study (level IV). Eleven studies did not report adverse events as an outcome and two studies reported no adverse events[25 26].

Risk of bias assessment

Risk of bias was low in four studies[14 27-30], unclear in seven studies[14 25 26 31-34] and high in two studies[34-36] leading to an overall moderate risk of bias across the studies (Table 2).

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Table 1: Characteristics of included studies

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Study	Study design	AACPDM LoE	CP n	Age Range, Mean and SD in years	GMFCS levels / proportions	Task-specific training (T)	Comparison (C) or other task-specific training	Activity outcomes			*Participation related outcomes (measure)*	*Other outcomes (ICF domain)*
								Gross motor function	*Functional skills*	*Gross motor skill performance*		
								Outcome measured used				
1. Group research designs												
a. Level II – III studies (Task specific vs comparison)												
Bar-Haim 2010	RCT	II	Total = 78 T = 39 C = 39	Range = 5.5 – 12.2 Total = 8.9 +/- 1.7 T = 8.8 +/- 1.7 C = 8.9 +/- 1.7	T II = 36%, III = 64% C II = 44% III = 56%	Motor learning coaching	Routine physiotherapy - NDT	GMFM	Parent survey			Mechanical efficiency (BSF)
Bleyenheuft 2015	Crossover RCT	II	Total = 24* T = 12 C = 12	T = 8.9 +/- 1.7 C = 8.5 +/- 1.7	T I = 50% II = 50% C I = 42% II = 58%	Hand arm bimanual intensive training including lower extremity (HABITILE)	Routine physiotherapy (in general had NDT while waiting for delayed HABITILE)		PEDI (self-care domain only)	6MWT ABILOCO Kids	Social participation (Life-H Performance and Satisfaction)	Body weight distribution (BSF) Mean step length (BSF) UL function (Act) Adverse events
Declerck 2016	RCT	II	Total = 14 T = 7 C = 7	Range = 7 – 17 T = 8.7 +/- 3.4 C = 11.8 +/- 3.5	T I = 14% II = 86% C I = 29% II = 57% III = 14%	Swimming skills program	Routine physiotherapy			WOTA-2† 1MWT	Adherence/ enjoyment	Pain (BSF) Fatigue (BSF) Adverse events
Grecco 2013	RCT	II	Total = 36* T = 17 C = 18	T = 6.8 +/- 2.6 C = 6.0 +/- 1.5	T I = 47% II = 41% III = 12% C I = 31% II = 50% GMFCS III = 19%	Overground walking	Treadmill training	GMFM	PEDI	6MWT† TUG		Balance (Act)
Ketelaar	RCT	II	Total = 55	Range (total) =	T	Functional	Routine	GMFM	PEDI			

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2001			T = 28 C = 27	2.0 – 7.25 T = 4.5 +/- 1.7 C = 4.7 +/- 1.7	mild = 79%, mod = 21% C mild = 77%, mod = 23%	therapy	physiotherapy - generally NDT or Votja method.					
Kumban 2013	RCT	II	Total = 21 T = 10 C = 11	T = 12.3 +/- 2.6 C = 12.4 +/- 2.4	T I-II = 60%, III = 40% C I-II = 55% III = 45%	Regular PT + sit-to-stand task training	Routine physiotherapy			FTSST†		Balance ability (Act) Functional balance and lower limb strength (Act)
Swe 2015	RCT	II	Total = 30* T = 15 C = 15	Range (total)= NS Total = 13.2 +/- 3.4 T = 13.0 +/- 3.6 C = 13.4 +/- 3.3	T II = 53% III = 47% C II = 67%, III = 33%	Overground walking	Partial weight support treadmill training	GMFM (standing and walking domains only)		6MWT† 10MWT†		
Capio 2015	Comparative study with concurrent controls	III	Total = 24 T = 12 C = 12	T = 6.92 +/- 3.04 C = 7.98 +/- 1.74	T I = 33% II = 58% III = 8% C I = 25% II = 58% III = 17%	Functional motor skills training	Routine physiotherapy or physical education	TGMD- 2			Physical activity (accelerome-ter)	
b. Level II – III (task specific vs task specific)												
Hemayatt - alab 2013	RCT	II	Total = 20 T1 = 10 T2 = 10	Total = 11.6 +/- 1.5 T1 = 11.9 +/- 1.6 T2 = 11.3 +/- 1.4	I-III Proportions not stated	Throwing task + self-control feedback	Throwing task + yoked feedback			Throwing accuracy†		
Hemayatt - alab 2010	Comparative study with concurrent controls	III	Total = 24 T1 = 8 T2 = 8 T3 = 8	Range (total) = 7-15	I = 100%	T1 = dart throwing task + 50% KR T2 = dart throwing task + 100% KR	T3 = Dart throwing task + 0% KR			Throwing accuracy†		
Lowing 2009	Comparative	III	Total = 44	Total = 4.1 +/-	T	Activity	Goal directed	GMFM	PEDI		Goal	

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	study with concurrent controls		T = 22 C = 22	1.4 T = 4.3 +/- 0.7 C = 3.8 +/- 1.3	I = 45% II = 23%, III = 14% IV = 18% C I = 41% II = 23%, III = 22% IV = 14%	focussed individual therapy	functional group therapy				attainment (GAS) in GDT group only	
c. Level IV												
Sorsdahl 2010	Repeated measures design	IV	T = 22*	Range = 2.8 – 9.25 Average age = 5.5	T I = 36% II = 23%, III = 27% IV-V = 14%	Functional family-centred goal directed therapy		GMFM GMPM			Goal attainment (GAS)	Quality of fine motor movements (Act)
2. Single subject research designs												
Thorpe 2002	13 x n=1 studies with random sampling to protocols (A, B or C)	I	N(total) = 13	Range = 6.0 – 12.7 Age = 8.6 +/- 1.89	III = 100%	All protocols = task specific A = 2 x no AI 1 x KP and 5 x KP+CS	B = 3 x no AI, 2 x KP and 3 x KP+CS C = 4 x no AI, 3 x KP and 1 x KP+CS			Change in backward displacement†		

Table 1 notes and abbreviations: * = sample size justified with statistical consideration, † = skill performance outcome is specific to skill being trained LoE = level of evidence, SD = standard deviation, GMFCS = Gross Motor Function Classification System, ICF = International Classification of Functioning, Disability and Health, NDT = Neurodevelopmental therapy, GMFM = Gross motor function measure (-88 or -66), BSF = Body structures and functions, PEDI = Pediatric Evaluation of Disability Inventory, 6MWT = Six minute walk test, ABILOCO = measure of locomotion ability, Life-H = Assessment of Life Habits, Act = activity, WOTA-2 = Water Orientation Test Alyn – 2nd edition, 1MWT = one minute walk test, 10MWT = ten metre walk test, TGMD – 2 = Test of Gross Motor Development – 2nd edition, KR = Knowledge of results, GAS = Goal Attainment Scale, GMPM = Gross Motor Performance Measure, AI = Augmented Information, KP = Knowledge of Performance, CS = cognitive strategies

Table 2: Risk of bias within included studies

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	Study	Selection bias		Performance bias	Detection bias	Attrition bias	Reporting bias	Other bias	Overall bias
		Random sequence generation	Allocation concealment	Participant and personnel blinding	Outcome assessor blinding	Incomplete outcome data	Selective outcome reporting		
Group research designs									
Level II and III studies	**Bar-Haim 2010**	Low	Low	High	Low	High	Low	High	**Low**
	Bleyenheuft 2016	Low	Low	High	Unclear	High	Unclear	Unclear	**Unclear**
	Declerck 2016	Low	Low	High	Unclear	Unclear	Low	High	**Unclear**
	Grecco 2013	Low	Low	High	Low	Unclear	Low	Unclear	**Low**
	Ketelaar 2001	Low	Unclear	High	Low	Unclear	Unclear	Unclear	**Unclear**
	Kumban 2013	Low	Low	High	Low	High	Low	Unclear	**Low**
	Swe 2015	Low	Low	High	Low	Low	Low	Unclear	**Low**
	Capio 2015	High	High	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2013	High	Unclear	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2010	Unclear	High	High	Unclear	Low	Low	Unclear	**Unclear**
	Lowing 2009	High	High	High	High	Low	Low	High	**High**
IV	**Sorsdahl 2010**	High	High	High	Low	High	Low	Unclear	**High**
Single subject design									
I	**Thorpe 2002**	Low	Unclear	High	High	Low	Low	Unclear	**Unclear**

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Level II-III group designs

(i) Task-specific training versus comparison

Participants

Of the 12 group-design studies included, eight involved comparing TST to another intervention (Table 1). These studies included seven RCTs involving 258 children and one comparative study involving 24 children[31]. The mean ages of children ranged from 4.6[33] to 13.2 years[30] . All participants were classified as GMFCS I-III.

Interventions

Six of the eight studies compared TST to routine physiotherapy[25-27 29 31 33]. The target of TST varied; four studies involved training a variety of tasks or movement skills[21 25 31 33], one study focused specifically on swimming[26] and another study trained sit to stand[29]. The content of the "routine physiotherapy" was generally poorly defined; described broadly as neurodevelopmental therapy (NDT)[25 27 33] in three studies, physical education in one study[31] and not reported in two studies[26 29]. The remaining two studies involved over-ground walking compared to treadmill training. In these studies, over-ground walking was deemed the TST, as treadmill training was considered not specific to the gross motor aim of the studies, which was to improve over-ground walking.

Two of the eight studies were directed by the child's goals while four studies reported the TST was driven by motor learning principles[25 27 31 33] (Table 3). Repetitive practice was the most commonly reported motor learning strategy with feedback and task modification involved in four[25 27 28 33] and five[25 27 29 31 33] of these studies respectively (Table 3). The remaining strategies were either not utilised or reported.

Physiotherapists generally conducted interventions within these studies but the focus, dosage, format and setting varied widely. Overall intervention time ranged from three hours[31] to 90 hours[25] over periods ranging from 10 days[25] to six months[33]. Reported settings were largely ecological including schools[27 29 30], a recreation camp[25], a swimming pool[26] and home environments[33]. Format was reported as group-based in two of these eight studies[25 26], otherwise was not stated.

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Table 3: Interventions: Components of task-specific training for included studies

Study	Task-specific intervention/s	Key characteristics of task-specific training									Components of the TIDiER checklist					
		Overall approach		Motor learning strategies reported							Dosage (Total hours)	Setting	Format	Providers	Adherence Reported?	Fidelity Reported?
		Goal-directed	*Motor learning driven*	*Feed back*	*Cogn. strat.*	*Repet. practice*	*Task mod.*	*Environ. mod*	*Obs. learning*	*Dual task learning*						
Group designs																
Bar-Haim 2010	Motor learning coaching	✓	✓	✓		✓	✓	✓		✓	1 hour, 3 days/ week for 3 months (36 hours)	School	NS	Trained PT		
Bleyenheuft 2015	HABITILE		✓	✓		✓	✓	✓		✓	9 hours/day for 10 consecutive days (90 hours)	Recreation camp	Group	Trained PTs, OTs, and PT / OT students	✓	
Declerck 2016	Swimming skills program					✓					40-50 mins, 2 days/week for 10 weeks (15 hours)	Swimming pool	Group	PT and PT students	✓	
Grecco 2013	Overground walking			✓		✓	✓				30 mins, 2 days/week for 7 weeks (7 hours)	NS	NS	Therapist		
Ketelaar 2001	Functional therapy	✓	✓	✓		✓	✓	✓			6 months – intensity or hours NS (NS)	Clinic and home	NS	Trained PTs and parents		
Kumban 2013	Sit-to-stand task training					✓	✓				20 min, 3 days/ week for 6 weeks (6 hours)	School	NS	Child's own PT		
Swe 2015	Overground walking					✓	✓				30 mins, 2 days/ week for 8	School	NS	PTs	✓	

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AACPDM Level III & IV task-specific sys Compilation

											weeks (8 hours)				
	Capio 2015	Functional motor skills training		✓			✓	✓			45 mins, 1 day/week for 4 weeks (3 hours)	NS	NS	Each child's regular PT	✓
Level I & III RCTs specific vs task	Hemayattal-ab 2010	Throwing task with different feedback conditions		✓	✓		✓			✓	2 sessions in 2 days (NS)	NS	NS	NS	
	Hemayattal-ab 2013	Throwing task with different feedback conditions		✓	✓		✓			✓	8 sessions Intensity or hours NS (NS)	NS	NS	Specialist trainers	
	Lowing 2009	Activity focussed individual therapy vs goal-directed therapy	✓ (goal group only)				✓ inc. home program				3 sessions / fortnight for 12 weeks (NS)	Clinic and home / preschool	Group and indiv	PT, OT, SP, teacher and parents	
Level IV	Sorsdahl 2010	Functional family-centred goal directed therapy	✓		✓		✓ inc. home program	✓	✓		3 hours, 5 days/ week for 3 weeks (45 hours)	Clinic	Group	Trained PTs and parents	✓
Single subject design															
Level I	Thorpe and Valvano 2002	Novel motor task learning with different conditions		✓	✓	✓	✓				3 sessions over 5 days (NS)	School	Indiv	PT (also the PI)	

Table 3 abbreviations: TIDiER = Template for Intervention Descriptions and Replication, Cogn. = cognitive, Strat. = strategies, Mod = modification, Environ = environmental, Obs = Observational, HABITILE = Hand Arm Bimanual Intensive Training Including Lower Extremity, PT = Physiotherapist, OT = Occupational therapist, SP = Speech therapist, NS = Not stated, Indiv = individual, Trained = trained in specific intervention protocol, inc = including, PI = Principal investigator

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Outcomes and effects

Outcomes were assessed across ICF domains with all studies reporting outcomes at the activity level, two studies reporting participation-related outcomes and three studies also involving body structure and function outcomes (Table 1). Outcomes were measured at one to three time points (T1, T2 and T3). In general T1 was immediately following the intervention, however, T2 and T3 were varied. T2 was between four weeks and six months following the intervention in the seven studies that measured these outcomes[25-30 33], while when measured, T3 was defined as two[26] or 12 months[33] following the intervention.

Activity outcomes

(i) Gross motor skill performance

Gross motor skill performance was measured in five of the eight studies[25 26 28-30]. Four of these studies measured skill performance specific to the training tasks[26 28-30] (Table 1). Overall, skills and measures were varied. Four studies measured walking performance[25 26 28 30] with three studies utilising the six minute walk test (6MWT)[25 28 30] whilst the timed up and go, one minute walk test, ten metre walk test and the ABILOCO-Kids questionnaire were used in one study each[25 26 28 30]. Swimming performance was measured using the Water Orientation Test Alyn – 2nd edition in one study[26] and sit to stand performance was measured using the five-times sit to stand test in another[29].

Between-group effects of TST on walking performance were mixed (Figures 2-b). Large effects favouring TST on walking performance at immediately following the intervention (T1) were found (ABILOCO Kids SMD = 0.96, 95% CI = 0.11 to 1.81; 6MWT SMD = 0.87, 95% CI 0.02-1.71)[25]. However, large effects favouring the comparison at both T1 (6WMT SMD = 1.47, 95%CI 0.69 to 2.25) and at 4 weeks follow up (6WMT SMD = 1.46, 95%CI 0.69 to 2.24)) were also found[28]. Non-significant mixed effects were found at both time points in the remaining two studies[26 30].

There was a no significant effect for TST compared to the comparison on swimming performance[26] or on sit to stand performance[29]. There was no between-group effect found for TST on sit to stand performance[29] or swimming performance at T1 (SMD = 0.46, 95%CI -0.06 to 1.53) or swimming performance at three months post intervention (SMD 0.34, 95%CI -0.72 to 1.40)[26].

Within-group effects of TST were positive for all skill performance outcomes in each study, except for the GMFCS III subgroup in one study[29].

(ii) Gross motor function

Overall gross motor function was measured in five of the eight studies[27 28 30 31 33] (Table 1). Four studies[27 28 30 33] utilised the Gross Motor Function Measure (GMFM)[37]. Three studies measured all domains of the GMFM[27 28 33] (A: lying and rolling, B: sitting, C: crawling and kneeling, D: standing, E walking running and jumping) while one study measured domains D and E only[30].

In the four studies that measured GMFM domains D and E, no significant effect was found for TST compared to comparison approaches at T1 or at T2 (Figures 3a-d)[27 28 30 33]. Three studies demonstrated no effect of TST at T1 or T2 in either domain[27 30 33] while the fourth study *(Grecco 2013)* showed a large effect favouring the comparison approach for both domains at both T1 (GMFM-D SMD = 1.39, 95%CI 0.62 to 2.16; GMFM-E SMD = 1.97, 95%CI 1.12 to 2.82) and T2 (GMFM-D SMD = 1.32, 95%CI 0.56 to 2.09; GMFM-E SMD = 2.08, 95%CI 1.22 to 2.95) (Figures 3a-d)[28]. Here, the comparison was treadmill training and the TST over-ground walking.

For overall gross motor function (GMFM total score) no significant effects for TST were found (Figures 3e-f)[27 33]. Again, a large effect favouring the comparison was found for overall gross motor function at both time points (T1 SMD = 1.83, 95%CI 1.03 to 2.63; T2 SMD = 1.67, 95%CI 0.87 to 2.48) by *Grecco 2013*[28].

The remaining level III design study reported a significant positive effect of TST on gross motor function as measured by items from the Test of Gross Motor Development - 2nd edition[31].

Within-group effects of TST on gross motor function were positive in all five studies.

(iii) Functional skills

Functional skill outcomes were measured in four of the eight studies[25 27 28 33] (Table 1). Functional skills included; self-care, as measured by the self-care domain of the Pediatric Evaluation of Disability Inventory (PEDI), in three studies[25 28 33]; mobility, as measured by the PEDI mobility domain in two studies[28 33] and a parent questionnaire[27]; and social function as measured by the PEDI social function domain in one study[28].

A large effect (SMD 1.07, 95%CI 0.21 to1.94) favouring TST was found for self-care skills at T1 in *Bleyenheuft 2015*[25]. No significant effects were found for the other two studies measuring self-care at T1 and T2[28 33] (Figures 4a-b).

One study measured mobility skills immediately following (T1) and at six (T2) and 12 months (T3) follow up[33]. There was no effect of TST compared to the comparison at T1 (SMD

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0.34, 95%CI -0.19 to 0.87,) a moderate effect was found at T2 (SMD = 0.58, 95% CI 0.04 to 1.12,) and a moderate-large effect was found at T3 (SMD 0.76 95%CI 0.21 to 1.31, Figures 4c). In contrast, a large effect (SMD 1.32, 95%CI 0.56 to 2.09) favouring the comparison was found for mobility skills immediately following the intervention in the study by Grecco 2013 comparing TST to treadmill training[28] (Figure 4c). No difference in mobility were found between the groups on the parent questionnaire immediately post intervention or a 6 months[27]. Similarly, no between-group difference was found for social function immediately post intervention or at 4 weeks follow up[28].

Within-group effects of TST on all functional skills outcomes were positive in all four studies.

Participation-related

Participation-related outcomes were measured in three of the eight studies[25 26 31] (Table 1). This included measuring social participation using the Life-HABITS performance and satisfaction questionnaire[25], assessing adherence and enjoyment[26] and measuring participation in physical activity using an uni-axial accelerometer[31].

Large effects favouring TST were found for both social participation performance (SMD = 1.19, 95%CI 0.31 to 2.07) and satisfaction (SMD = 1.29, 95%CI 0.40 to 2.18)[25]. Positive effects of TST on weekend physical activity were reported immediately following the intervention[31]. High adherence rates (median = 100%) and enjoyment levels (median = 5 on a five point Likert scale) were reported in the TST group but this data was not collected in the comparison group[26].

(ii) Task-specific versus task-specific

Participants

Three of the 12 group design studies involved a comparison of two or more task-specific interventions including one RCT (level II)[32] and two comparative studies with concurrent controls (level III)[34 35]. These studies involved 88 children with over 90% classified GMFCS level I-III.

Interventions

Two of the three studies[32 34] compared the effect of two TST programs with different feedback conditions on a throwing task (Table 1). The throwing task training was informed

by motor learning principles but was not goal-directed (Table 3). The other study examined the effects of goal-directed training compared to activity-based training only[35]. Repetitive practice was the only motor learning strategy reported in both of these programs. The intensity of the throwing TST in both studies was approximately two hours over two days but setting, format and providers were not reported[32 34]. The other study involved 18 sessions of TST over 12 weeks with parent involvement and was conducted at school, home and the clinic[35]. The goal-directed program involved group and individual sessions while the activity program was individual only.

Outcomes and effects

Gross motor skill performance (specific to the trained task), as measured by throwing accuracy, was the sole outcome assessed in both throwing TST studies[32 34]. This was assessed immediately after the last training session (T1 – acquisition) in both studies, 24 hours later in one [32] (T2 – retention and transfer) and three days later (T2 – retention) in the other[34]. Outcomes were assessed at T1 only in the remaining study: including gross motor function (total GMFM score) and functional skills (all domains of the PEDI) in both groups, and the participation-related outcome of goal attainment, using the goal attainment scale (GAS) in the goal-directed group only[35].

The highest frequency of feedback (100% knowledge of results) within TST was found to improve throwing accuracy significantly more than 50% or no feedback in the acquisition phase (T1). However, in the retention phase (T2), those who received knowledge of results 50% of the time performed significantly better than those who received feedback 100% of the time or no feedback at all[34]. Self-controlled feedback within TST was found to improve throwing accuracy significantly more than yoked feedback at T2 but not at T1[32].

Goal-directed training improved overall gross motor function and functional skills including mobility and self-care more than activity-based training[35]. There was no difference in social function between the groups. Eighty-five percent of goals were attained to an expected or greater than expected level with goal-directed TST[35].

Level IV group design

One repeated measures study involving 22 children across all GMFCS levels (86% GMFCS I-III) was included[36] (Table 1). This study involved goal-directed, group-based and intensive TST conducted by two physiotherapists (Table 3). Measures were assessed at three baseline

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time points, immediately (T1) and 3 weeks (T2) after the intervention. Gross motor function (overall GMFM score), functional skills (mobility, self-care and social function using the PEDI) and goal attainment (GAS) were measured. Positive effects were reported for overall gross motor function at both T1 and T2, and for self-care skills at T2, while 66% of goals were attained to an expected or greater than expected level at T2.

Single subject design

One Level I single subject design study was included[14]. Thirteen children (all GMFCS III) were randomised to one of three feedback protocols for learning to move the Pedalo, an exercise vehicle, backward. The three protocols varied in terms of the following components; no feedback, knowledge of performance and knowledge of performance with a cognitive strategy. Gross motor skill performance (specific to the trained task) was measured using backwards displacement of the Pedalo and was assessed on each trial and on further trials two days following the training (T1). Eight of the 13 participants demonstrated significant improvement at T1. No specific feedback protocol was clearly superior.

Intervention replicability

There was wide variation in reporting of intervention characteristics (Table 3). Four studies reported intervention providers were trained in the intervention protocol[25 27 33 36], five studies reported format of the intervention[14 25 26 35 36] while participant adherence to the intervention protocol was described in five studies[25 26 30 31 36]. No study reported the extent to which the providers delivered the intervention as intended (fidelity).

Quality of the evidence by outcome

The overall quality of evidence was moderate for gross motor skill performance and functional skills and low for gross motor function and participation-related outcomes.

DISCUSSION

To our knowledge, this is the first systematic review to evaluate and synthesise the evidence for task specific gross motor skills training in ambulant school aged children with CP. Given the nature of TST, where the focus is on practice of tasks rather than remediating impairments, and the increasing recognition of importance of child and family-centred

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effects of interventions, this review explicitly focused on activity and participation outcomes. In general, positive within-group effects of TST were reported across outcomes of interest. However, in RCTs where TST was compared to comparison interventions, between-group effects were largely non-significant with the exception of two studies[25 28] reporting large but conflicting effects. Overall, there was moderate quality evidence for conflicting effects of TST to improve specific skills performance and functional skills but low quality evidence showing no difference or negative effects on gross motor function. Any positive effects on these activity outcomes were generally found immediately following TST, with evidence of longer-term retention lacking. For participation-related outcomes, low quality evidence for positive effects of TST was found, again largely related to immediate outcomes.

The secondary aim of this study was to identify motor learning strategies reported in TST and assess any relationship to outcome. The overall poor reporting of motor learning strategies and heterogeneity in interventions and outcomes limited definitive conclusions in relation to this aim. However, this review provides some insight into potentially important characteristics of interventions, in particular; practice dosage, feedback and goals. Unsurprisingly, given its intrinsic relationship to TST, the most consistently reported motor learning strategy was repetitive practice. While reporting of dosage was variable, the largest positive effects of TST were found where dosage was highest[25]. Studies comparing characteristics of TST provide insight into the influence of different feedback conditions and goals on outcomes. Whilst no specific feedback condition emerged as clearly superior in the three studies comparing these[14 32 34], results suggest that the different feedback conditions may influence the phases of task training (acquisition vs retention) differently. Replication of these studies for varying tasks is required to provide further clarity into the role of feedback. Although causal inferences are limited due to lack of randomisation, better activity outcomes were found when the TST was goal-directed rather than activity focussed[35]. These results are consistent with the growing evidence base across the CP literature for interventions that are targeted towards the goals of children and their families[6 16].

Our review found some conflicting results between study findings, with large positive and negative effects sizes found in *Bleyenheuft 2015*[25] and *Grecco 2013*[28] respectively. The TST in *Bleyenheuft 2015*[25] was high in dose and explicitly driven by motor learning principles with strategies including feedback, task and environmental modification and observational learning. It also involved TST of a wide range of gross motor tasks including ball skills, bicycling and walking. By contrast, the TST in *Grecco 2013*[28] dosage of over-ground walking was low in comparison and limited motor learning strategies were

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reported. The comparison treatment in this study was treadmill training without body weight support – training that may be regarded as task-related but was not deemed TST given the primary goal of the study was to improve over-ground walking. Whilst practice dose was equal between the groups in *Grecco 2013*[28], task progression was potentially greater with the use of the treadmill functions. The results of this study suggest that task-related adjuncts, such as treadmill training, may be more beneficial than pure TST for developing (rather than acquiring) gross motor skills, such as walking in this already-ambulant population.

Methodological considerations

Low-moderate quality of evidence found across the outcomes due to significant methodological limitations across and within the included studies. These limitations fit broadly into four categories; (1) sample size (2) sources of bias, (3) intervention replicability and (3) outcome issues.

First, description of sample size calculation was not reported in most studies (Table 1). Wide confidence intervals found for outcome data from Level II-III studies (Figures 2-4) suggest inadequately powered samples may have reduced precision and thus represent a limitation in generalizability of the findings.

Second, significant sources of bias were identified, further weakening the evidence (Table 2). Major sources of bias included: studies without randomisation or allocation concealment, a lack of participant/personnel blinding and incomplete outcome data. Further, no study reported the use of intention to treat analysis and reporting of outcome assessor blinding was inconsistent. Poor reporting of study conduct which limited assessment of risk of bias in some studies also precludes full evaluation of the evidence.

Third, the limited detail of reporting and lack of reproducibility potentially reduces the robustness of research findings if the study cannot be replicated and limits implementation. Reporting of dosage was unclear in some studies, format often missing, participant adherence reported in few studies and assessment of fidelity uniformly lacking. Comparison interventions were ambiguously reported. Known geographical and economical variability in clinical practice[16] reduces the utility of terms such as “routine physiotherapy,” (without further definition) and further limits reproducibility.

Fourth, limitations in outcome measurement selection may have influenced the generalisability and transferability of some findings. Outcome measure responsiveness is key to determining intervention effectiveness[38]. The GMFM and the PEDI are a well-established

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tools for evaluating change in children with CP[39], and were used in six[27 28 30 33 35 36] and four studies[25 28 33 35] respectively. However, concerns have been raised about the responsiveness of these measures in higher functioning children[39], the target population of this review. Whilst common use makes them appealing, the broad focus of these measures means the skills targeted by the TST may not have been adequately captured, especially given only seven of the 13 included studies measured the specific skill being trained[14 26 28-30 32 34]. Further, far fewer participation-related outcome measures were used compared to activity outcomes thus any evidence that improved activity through TST leads to improved participation is weak at best.

Limitations of this review

By narrowing the inclusion criteria in terms of; population to ambulant school aged children with CP, intervention - to gross motor skill TST, and outcomes - to those in the activity and participation domains, the authors intended to enable specific conclusions to be drawn. This was not possible for the questions posed by this review largely due to study design issues and heterogeneity across the included studies, which also precluded meta-analysis. However, the methodology of the review itself is not without limitations. The impact of publication bias is not known. Inclusion was limited to published academic articles in English meaning some studies, including any grey literature, may have been missed. Further, non-randomised studies were included in this review. Although the Cochrane Risk of Bias tool and the GRADE system are the most widely used systems for assessing risk of bias and the quality of the evidence across outcomes respectively[20 24], these tools do emphasise randomised studies.

Conclusions and recommendations

Despite the largely positive within-group effects of TST over time across each outcome of interest, between-group effects were conflicting for specific skills performance and functional skills, positive for participation-related outcomes while no difference or negative effects were found for gross motor function. Given the low-moderate quality of this evidence, there is currently limited evidence to support task-specific gross motor skills training for improving activity and participation-related outcomes in children with CP. Clear recommendations around whether TST is superior to other interventions cannot be made.

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Before conclusions can be made about any relationship of motor learning strategies to outcome, more consistent reporting and studies designed to test this are required.

The importance of tailoring motor interventions to individual goals and lifelong physical activity is increasingly being recognised[4 40]. Thus, to enable specific recommendations, strengthening the evidence is imperative. Adequately powered samples, rigorous study design, consistent reporting with attention to reporting interventions to allow for reproducibility is required. Future challenges for TST research also include considering issues with outcome measure responsiveness and intervention heterogeneity, and optimising TST through the use of motor learning strategies. Lastly, consensus in the terminology used to describe motor interventions for children with CP is required to ensure appropriate evidence synthesis and accurate guidance for clinicians and families.

Competing interests

The authors have no competing interests to declare.

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Data sharing statement

Data relating to the qualitative synthesis is available on request to the corresponding author.

Author contributions

All named authors contributed to the design of this review. RT collected data, carried out data analyses and led interpretation and reporting. CB collected data and contributed to interpretation and reporting. ARH, JLM and AJS contributed to data analysis, interpretation and reporting. All named authors approved the final manuscript as submitted.

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Figure legend

Figure 1: PRISMA flow chart of included and excluded studies

Figure 2a: Gross motor skill performance - Task specific vs alternative: walking performance at T1 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 2b: Gross motor skill performance - Task specific vs alternative: walking performance at T2 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 3a: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T1.

Figure 3b: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T2.

Figure 3c: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T1.

Figure 3d: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T2.

Figure 3e: Gross motor function - Task specific vs alternative: Overall GMFM score at T1.

Figure 3f: Gross motor function - Task specific vs alternative: Overall GMFM score at T2.

Figure 4a: Functional skills - Task specific vs alternative: Self-care at T1.

Figure 4b: Functional skills - Task specific vs alternative: Self-care at T2.

Figure 4c: Functional skills - Task specific vs alternative: Mobility skills at T1

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APPENDIX 1

Search strategy for Ovid databases

1. cerebral palsy.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
2. child*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. (Task specific or skill specific or task oriented or activity focussed or task practice or goal directed).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
4. (motor imagery or implicit or error reduced or neuromotor task or parent assisted or group or cognitive orientation or repetit*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. (perceptual motor or kinaesthetic explicit or action observation or dual task or trial and error or practice or imitation or experiential or discrimination or discovery or errorless or analogy or observational or exploratory).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
6. (Training or intervention or therapy or approach or learning or program).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. (Motor skill* or motor function* or movement skill* or Motor performance or task performance or occupational performance or motor competence or task competence or occupational competence or skill acquisition or skill attainment or skill proficiency or skill achievement or goal attainment).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
8. 3 or 4 or 5
9. 1 and 2 and 6 and 7 and 8

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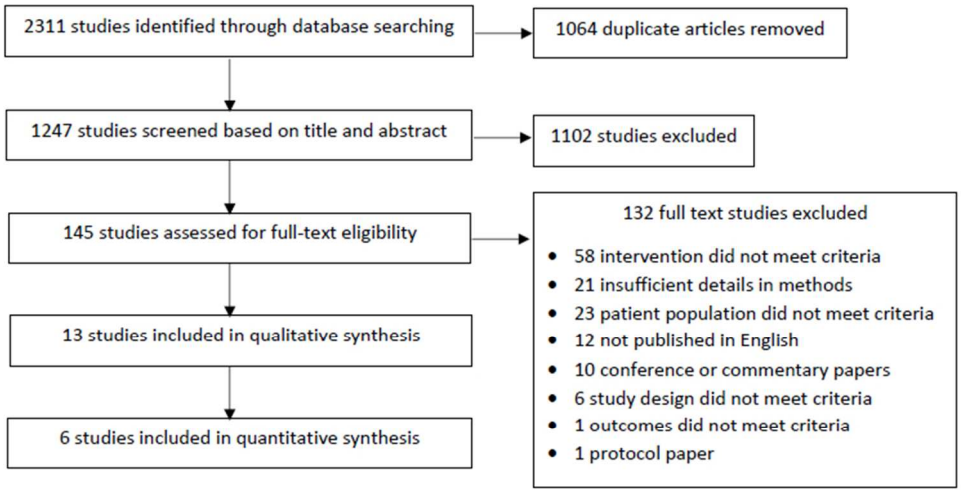

205x111mm (96 x 96 DPI)

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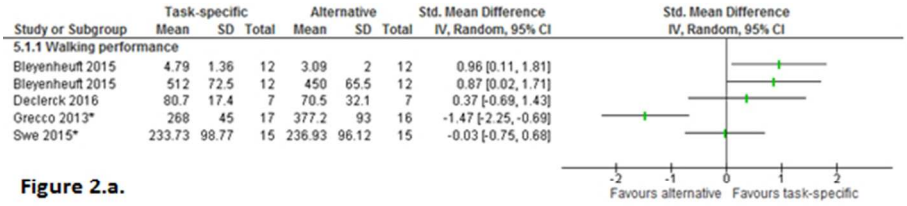

Figure 2.a.

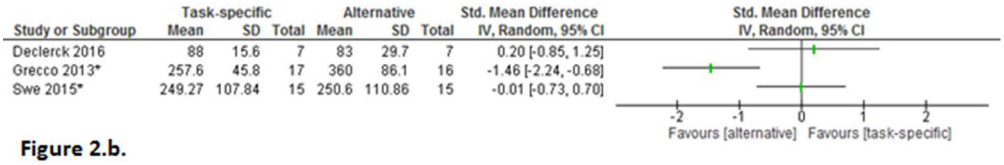

Figure 2.b.

177x78mm (96 x 96 DPI)

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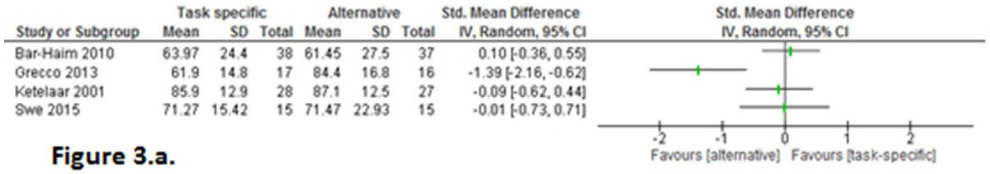

Figure 3.a.

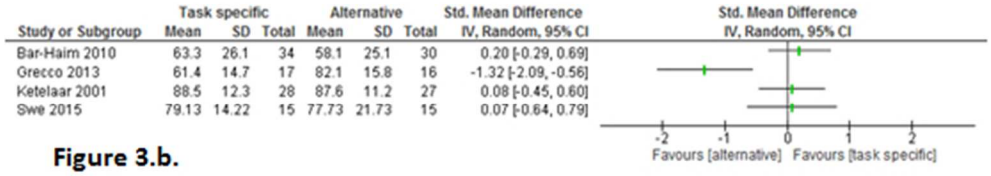

Figure 3.b.

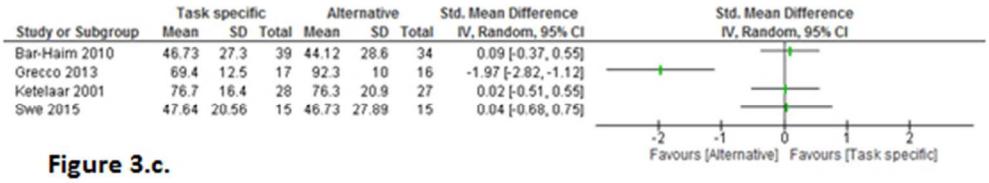

Figure 3.c.

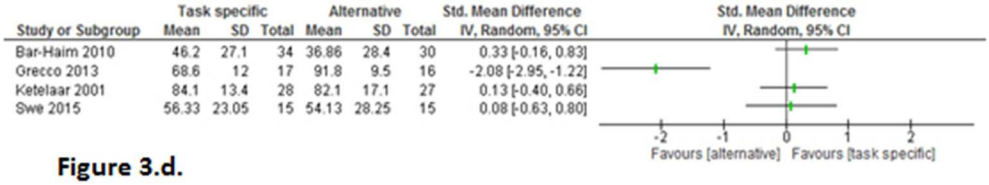

Figure 3.d.

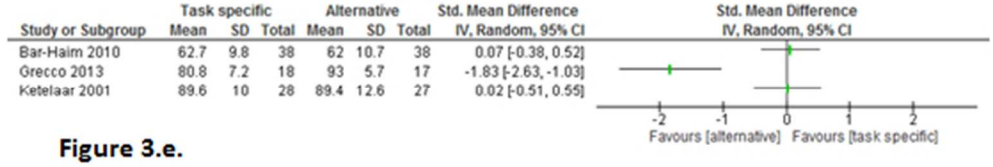

Figure 3.e.

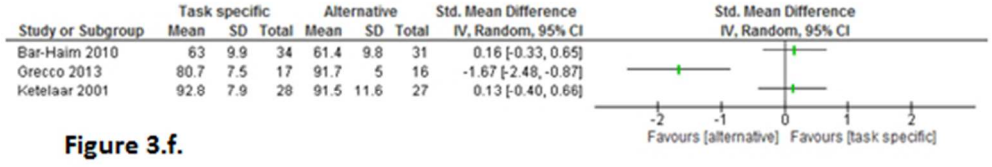

Figure 3.f.

149x178mm (96 x 96 DPI)

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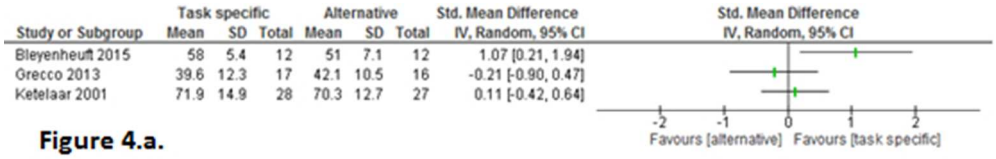

Figure 4.a.

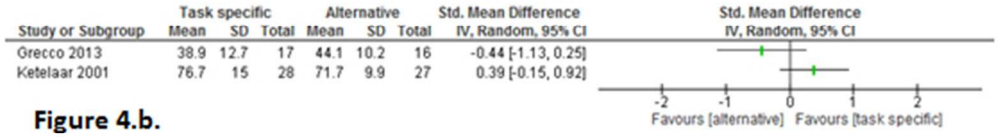

Figure 4.b.

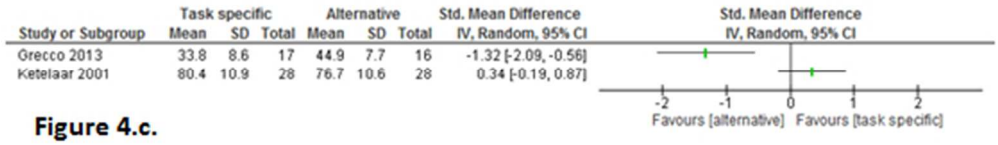

Figure 4.c.

148x79mm (96 x 96 DPI)

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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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Keywords:	Neurodisability, Physcial therapy, Rehabilitation

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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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ABSTRACT

OBJECTIVES: The primary objective is to systematically evaluate the evidence for the effectiveness of task-specific training (TST) of gross motor skills for improving activity and/or participation outcomes in ambulant school aged children with cerebral palsy (CP). The secondary objective is to identify motor learning strategies reported within TST and assess relationship to outcome.

DESIGN: Systematic review

METHOD: Relevant databases were searched for studies including; children with CP (mean age >4 years and >60% of the sample ambulant); TST targeting gross motor skills; and activity (skill performance, gross motor function and functional skills) and/or participation-related outcomes. Quality of included studies was assessed using standardised tools for risk of bias, study design and quality of evidence across outcomes. Continuous data were summarised for each study using standardised mean difference (SMD) and 95% confidence intervals (CI).

RESULTS: Thirteen studies met inclusion criteria: eight randomised controlled trials (RCT), three comparative studies, one repeated-measures study and one single-subject design study. Risk of bias was moderate across studies. Components of TST varied and were often poorly reported. Within-group effects of TST were positive across all outcomes of interest in 11 studies. In RCTs, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes (one study: Life-HABITS performance SMD = 1.19, 95% CI 0.3, 2.07, p< 0.001; Life-HABITS satisfaction SMD = 1.29, 95% CI 0.40, 2.18, p =0.001) while no difference or negative effects were found for gross motor function. The quality of evidence was low-moderate overall. Variability and poor reporting of motor learning strategies limited assessment of relationship to outcome.

CONCLUSIONS: Limited evidence for TST for gross motor skills in ambulant children with CP exists for improving activity and participation-related outcomes and recommendations for use over other interventions are limited by poor study methodology and heterogeneous interventions.

REGISTRATION: PROSPERO ID42016036727

KEY MESSAGES

What is known about the subject

- Strong evidence for motor interventions involving task specificity for functional mobility in adults post stroke and for upper limb function in children with cerebral palsy exists
- The effectiveness of task-specific gross motor skills training in ambulant school aged children with cerebral palsy has not been systematically evaluated or synthesised

What this study adds

- A low-moderate overall quality of evidence was found for task-specific gross motor skills training for ambulant school aged-children with cerebral palsy
- Limited evidence for task-specific training to improve specific skills performance, functional skills and participation-related outcomes exists
- While clear recommendations for use of task-specific training over other interventions are limited, ways to strengthen the evidence in future studies are identified

Short title: Task-specific gross motor skills training in CP

Key words: cerebral palsy, task-specific training, gross motor skills, activity, participation

ABBREVIATIONS

CP	Cerebral palsy
GMFCS	Gross motor Function Classification System
ICF	International Classification of Functioning, Disability and Health
TST	Task-specific training
AACPDM	American Academy of Cerebral Palsy and Developmental Medicine
SMD	Standardised mean difference
CI	Confidence interval
RCT	Randomised controlled trial
NDT	Neurodevelopmental therapy
GMFM	Gross Motor Function Measure

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PEDI Pediatric Evaluation of Disability Inventory

Cerebral palsy (CP) is an umbrella term used to describe a group of disorders of movement which cause varying degrees of activity limitations[1]. The most widely used means for classifying gross motor function in children with CP is the Gross Motor Function Classification System (GMFCS)[2]. Children classified GMFCS I are able to walk and run but have limitations with speed, balance and coordination whilst children classified GMFCS V are transported in a wheelchair in all settings. Although the focus of the GMFCS is on functional mobility, the realm of gross motor activities, that is skills involving movement of the large muscles of the limbs or whole body, undertaken by children is much broader[3]. Development of gross motor skills underpins functional, play and social activities across childhood and complex movement skills required for sports in older children[3]. In children with CP, limitations in gross motor function increase as GMFCS level increases, however, children at all GMFCS levels (I-V) participate, on average, less in physical activities than their typically developing peers[2]. This is an issue because of the known poor health outcomes in adulthood due to inactivity in childhood[4]. Effective interventions tailored to GMFCS levels and developmental stages are required to improve these outcomes in this population.

The International Classification of Functioning, Disability and Health (ICF) has become the common language for clinicians, researchers and families for understanding the effect of CP on the individual and for targeting interventions[5,6]. Where interventions previously focused on remediating limitations in body structures and functions, there has been a more recent acknowledgement of the importance of the effect of interventions within the activity and participation domains[7 8]. Clinicians working with children with CP need guidance from evidence synthesis to implement effective means of improving physical skills *and* improve the uptake of these skills in the child's daily life. The historical bias towards impairment-focused motor interventions yielded few effective treatments[6 9 10] thus more functional approaches have emerged.

Task-specific training (TST) involves practice of context-specific tasks where the intervention focuses on the skills needed for a task(s)[11] - there is similarity between the training task and the goal of the intervention. Although Level I evidence exists for TST to improve gross motor activities in adults after stroke[12], the majority of high level evidence for interventions involving task specificity in children with CP relates to training of upper limb or fine motor activities[6 13] with limited evidence for gross motor skills training. TST inherently involves principles of motor learning with components including context, practice and

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dosage[11]. Other motor learning strategies, such as feedback and task modification, have the potential to optimise TST, however, this has not been systematically studied[14]. TST should involve varied components depending on the requirements of the skill, the environment and the function of the child[15]. Moreover, training for a child of higher-level motor function (e.g. GMFCS I-III) should be targeted towards different skills compared to training with a child of lower-level motor function (e.g. GMFCS IV-V). Similarly, children of different ages and developmental stages have varying learning capabilities, and physical demands placed on them by their context[16].

Previous systematic reviews of motor interventions in children with CP have been broad in terms of ages (including infants and children) and motor function (all GMFCS levels), and included interventions have been heterogeneous[17 18]. TST may be a promising approach for ambulant children who have specific gross motor skills goals, however, there has been no systematic review to examine the effectiveness of this approach. The primary aim of this study is to evaluate and synthesise the evidence for the effectiveness of task-specific gross motor skills training in ambulant children aged 4 – 18 years with CP for activity and participation outcomes. The secondary aim of this study is to identify motor learning strategies reported within TST and assess relationship to outcome.

METHOD

Eligibility criteria

Published studies were included if they met all of the following criteria:

1. Level of evidence: All group design studies categorised as level II-IV using the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) levels of evidence[19] and studies classified as level I single-subject designs involving over 10 participants.
2. Population: The majority (i.e. > 60%) of the participants were ambulant children with CP (GMFCS I-III), and the mean sample age was four – 18 years.
3. Interventions: TST of gross motor skills where there was similarity between the training task and the goal of the intervention, including those interventions described as involving motor learning strategies/coaching, goal-directed training, activity focused training and/or functional skills training. Any duration or intensity of TST.

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4. Comparison: Studies comparing TST to another intervention, another type of TST or no intervention.
5. Outcomes: Activity outcomes including; gross motor skill performance (specific to the task being trained or other gross motor task to assess for transferability), gross motor function and functional skills; and participation-related outcomes. Only studies reporting outcomes separately for children with CP.

Exclusion criteria: TST was applied within a combined intervention approach and the influence of TST could not be isolated (e.g. Botulinum Toxin-A, virtual reality, treadmill training, orthoses or robotics), greater than ten percent of the intervention was passive or the article was not in English.

Search strategy

Relevant articles were identified from Medline, EMBASE, CINAHL, Psycinfo, SPORTDiscus and PubMed with all searches limited to articles published in peer-reviewed journals in English. A search was conducted in June 2016 and search terms were tailored for each database. Reference lists of included studies and related reviews were also searched. See Appendix 1 for full search strategy for Ovid databases (Medline, EMBASE and PubMed).

Study selection, data extraction and quality assessment

Study selection, data extraction and quality assessment was completed by two authors (RT and CB) independently, with a third author (AS, AH or JM) to resolve any disagreements. If inclusion was uncertain from abstract, the full text was retrieved.

For included studies, data were extracted using a customised form based on the Cochrane recommendations[20]. Data extracted included: study details (author, year, country, funding), study design, AACPDM level of evidence[19], study sample characteristics , detailed TST characteristics (including motor learning strategies), comparison intervention characteristics, outcomes measured and associated ICF domain, effects of the intervention and conclusions. Reported motor learning strategies in all TST, including when studies involved a comparison of two or more TST approaches, were identified using pre-defined motor learning strategy codes based on current literature[21 22]. Authors were contacted for complete data extraction when needed.

Risk of bias for individual studies was assessed using the Cochrane Risk of Bias Tool[20]. Overall risk of bias was determined by predominant level of bias across the domains

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of the Risk of Bias Tool. Intervention replicability was assessed using relevant components of the Template for Intervention Descriptions and Replication (TIDieR) Checklist[23]. The Grading of Recommendations Assessment Development and Evaluation (GRADE) system, was used to assess the quality of the evidence across outcomes[24].

Narrative synthesis and data analysis

Study design and AACPDM level of evidence were used for narrative synthesis. Higher-level group designs (AACPDM levels II-III) were reported together, whilst lower-level group designs (IV) and single-subject design studies were reported separately. Within the level II-III group designs, studies were considered as either 'TST versus a comparison' or 'TST versus TST'. Finally, within each grouping, activity outcomes were organised into 3 constructs; gross motor skill performance, functional skills and gross motor function whilst participation-related outcomes were grouped together.

Analyses were conducted using Cochrane Review Manager Software RevMan 5.3. Only data from level II studies comparing TST to comparison interventions were included in quantitative analysis to ensure comparison was between studies of similar design. Continuous data were summarized for each study within outcomes of interest using standardized mean difference (SMD) and 95% confidence intervals (CIs). A SMD of 0.2 was considered small, 0.4-0.6 moderate and 0.8 a large effect size[20]. Given the heterogeneity between studies in tasks and characteristics of the TST and comparison interventions, meta-analysis was not undertaken.

RESULTS

Following removal of duplicates, the abstracts of 1247 studies were screened and 145 full-text articles were retrieved for full appraisal (Figure 1). Thirteen studies involving 405 participants met inclusion criteria and underwent narrative synthesis with six of these studies involving 237 individuals included in data analysis. Characteristics of included studies are summarised in Table 1.

The 13 studies included 12 group designs and one single-subject design (level I). The group designs involved eight randomised controlled trials (RCTs) (level II), three comparative studies with concurrent controls (level III) and one repeated measures study (level IV). Eleven studies did not report adverse events as an outcome and two studies reported no adverse events[25 26].

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Risk of bias assessment

Risk of bias was low in four studies[14 27-30], unclear in seven studies[14 25 26 31-34] and high in two studies[34-36] leading to an overall moderate risk of bias across the studies (Table 2). Major sources of bias included: studies without randomisation or allocation concealment, a lack of participant/personnel blinding and incomplete outcome data. No study reported the use of intention to treat analysis and reporting of outcome assessor blinding was inconsistent. Poor reporting of study conduct limited assessment of risk of bias in the seven studies with unclear risk of bias.

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Table 1: Characteristics of included studies

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Study	Study design	AACPDM LoE	CP n	Age Range, Mean and SD in years	GMFCS levels / proportions	Task-specific training (T)	Comparison (C) or other task-specific training	Activity outcomes			*Participation related outcomes (measure)*	*Other outcomes (ICF domain)*
								Gross motor function	*Functional skills*	*Gross motor skill performance*		
								Outcome measured used				
1. Group research designs												
a. Level II – III studies (Task specific vs comparison)												
Bar-Haim 2010[27]	RCT	II	Total = 78 T = 39 C = 39	Range = 5.5 – 12.2 Total = 8.9 +/- 1.7 T = 8.8 +/- 1.7 C = 8.9 +/- 1.7	T II = 36%, III = 64% C II = 44% III = 56%	Motor learning coaching	Routine physiotherapy - NDT	GMFM	Parent survey			Mechanical efficiency (BSF)
Bleyenheuft 2015[25]	Crossover RCT	II	Total = 24* T = 12 C = 12	T = 8.9 +/- 1.7 C = 8.5 +/- 1.7	T I = 50% II = 50% C I = 42% II = 58%	Hand arm bimanual intensive training including lower extremity (HABITILE)	Routine physiotherapy (in general had NDT while waiting for delayed HABITILE)		PEDI (self-care domain only)	6MWT ABILOCO Kids	Social participation (Life-H Performan-ce and Satisfaction)	Body weight distribution (BSF) Mean step length (BSF) UL function (Act) Adverse events
Declerck 2016[26]	RCT	II	Total = 14 T = 7 C = 7	Range = 7 – 17 T = 8.7 +/- 3.4 C = 11.8 +/- 3.5	T I = 14% II = 86% C I = 29% II = 57% III = 14%	Swimming skills program	Routine physiotherapy			WOTA-2† 1MWT	Adherence/ enjoyment	Pain (BSF) Fatigue (BSF) Adverse events
Grecco 2013[28]	RCT	II	Total = 36* T = 17 C = 18	T = 6.8 +/- 2.6 C = 6.0 +/- 1.5	T I = 47% II = 41% III = 12% C I = 31% II = 50% GMFCS III = 19%	Overground walking	Treadmill training	GMFM	PEDI	6MWT† TUG		Balance (Act)
Ketelaar	RCT	II	Total = 55	Range (total) =	T	Functional	Routine	GMFM	PEDI			

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2001[33]			T = 28 C = 27	2.0 – 7.25 T = 4.5 +/- 1.7 C = 4.7 +/- 1.7	mild = 79%, mod = 21% C mild = 77%, mod = 23%	therapy	physiotherapy - generally NDT or Votja method.					
Kumban 2013[29]	RCT	II	Total = 21 T = 10 C = 11	T = 12.3 +/- 2.6 C = 12.4 +/- 2.4	T I-II = 60%, III = 40% C I-II = 55% III = 45%	Regular PT + sit-to-stand task training	Routine physiotherapy			FTSST†		Balance ability (Act) Functional balance and lower limb strength (Act)
Swe 2015[30]	RCT	II	Total = 30* T = 15 C = 15	Range (total)= NS Total = 13.2 +/- 3.4 T = 13.0 +/- 3.6 C = 13.4 +/- 3.3	T II = 53% III = 47% C II = 67%, III = 33%	Overground walking	Partial weight support treadmill training	GMFM (standing and walking domains only)		6MWT† 10MWT†		
Capio 2015[31]	Comparative study with concurrent controls	III	Total = 24 T = 12 C = 12	T = 6.92 +/- 3.04 C = 7.98 +/- 1.74	T I = 33% II = 58% III = 8% C I = 25% II = 58% III = 17%	Functional motor skills training	Routine physiotherapy or physical education	TGMD- 2			Physical activity (accelerome-ter)	
b. Level II – III (task specific vs task specific)												
Hemayatt - alab 2013[32]	RCT	II	Total = 20 T1 = 10 T2 = 10	Total = 11.6 +/- 1.5 T1 = 11.9 +/- 1.6 T2 = 11.3 +/- 1.4	I-III Proportions not stated	Throwing task + self-control feedback	Throwing task + yoked feedback			Throwing accuracy†		
Hemayatt - alab 2010[34]	Comparative study with concurrent controls	III	Total = 24 T1 = 8 T2 = 8 T3 = 8	Range (total) = 7-15	I = 100%	T1 = dart throwing task + 50% KR T2 = dart throwing task + 100% KR	T3 = Dart throwing task + 0% KR			Throwing accuracy†		
Lowing	Comparative	III	Total = 44	Total = 4.1 +/-	T	Activity	Goal directed	GMFM	PEDI		Goal	

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2009[35]	study with concurrent controls		T = 22 C = 22	1.4 T = 4.3 +/- 0.7 C = 3.8 +/- 1.3	I = 45% II = 23%, III = 14% IV = 18% C I = 41% II = 23%, III = 22% IV = 14%	focussed individual therapy	functional group therapy				attainment (GAS) in GDT group only	
c. Level IV												
Sorsdahl 2010[36]	Repeated measures design	IV	T = 22*	Range = 2.8 – 9.25 Average age = 5.5	T I = 36% II = 23%, III = 27% IV-V = 14%	Functional family-centred goal directed therapy		GMFM GMPM			Goal attainment (GAS)	Quality of fine motor movements (Act)
2. Single-subject research designs												
Thorpe 2002[14]	13 x n=1 studies with random sampling to protocols (A, B or C)	I	N(total) = 13	Range = 6.0 – 12.7 Age = 8.6 +/- 1.89	III = 100%	All protocols = task specific A = 2 x no AI 1 x KP and 5 x KP+CS	B = 3 x no AI, 2 x KP and 3 x KP+CS C = 4 x no AI, 3 x KP and 1 x KP+CS			Change in backward displacement†		

Table 1 notes and abbreviations: * = sample size justified with statistical consideration, † = skill performance outcome is specific to skill being trained LoE = level of evidence, SD = standard deviation, GMFCS = Gross Motor Function Classification System, ICF = International Classification of Functioning, Disability and Health, NDT = Neurodevelopmental therapy, GMFM = Gross motor function measure (-88 or -66), BSF = Body structures and functions, PEDI = Pediatric Evaluation of Disability Inventory, 6MWT = Six minute walk test, ABILOCO = measure of locomotion ability, Life-H = Assessment of Life Habits, Act = activity, WOTA-2 = Water Orientation Test Alyn – 2nd edition, 1MWT = one minute walk test, 10MWT = ten metre walk test, TGMD – 2 = Test of Gross Motor Development – 2nd edition, KR = Knowledge of results, GAS = Goal Attainment Scale, GMPM = Gross Motor Performance Measure, AI = Augmented Information, KP = Knowledge of Performance, CS = cognitive strategies

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Table 2: Risk of bias within included studies

	Study	Selection bias		Performance bias	Detection bias	Attrition bias	Reporting bias	Other bias	Overall bias
		Random sequence generation	Allocation concealment	Participant and personnel blinding	Outcome assessor blinding	Incomplete outcome data	Selective outcome reporting		
Group research designs									
Level II and III studies	**Bar-Haim 2010**[27]	Low	Low	High	Low	High	Low	High	**Low**
	Bleyenheuft 2016[25]	Low	Low	High	Unclear	High	Unclear	Unclear	**Unclear**
	Declerck 2016[26]	Low	Low	High	Unclear	Unclear	Low	High	**Unclear**
	Grecco 2013[28]	Low	Low	High	Low	Unclear	Low	Unclear	**Low**
	Ketelaar 2001[33]	Low	Unclear	High	Low	Unclear	Unclear	Unclear	**Unclear**
	Kumban 2013[29]	Low	Low	High	Low	High	Low	Unclear	**Low**
	Swe 2015[30]	Low	Low	High	Low	Low	Low	Unclear	**Low**
	Capio 2015[31]	High	High	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2013[32]	High	Unclear	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2010[34]	Unclear	High	High	Unclear	Low	Low	Unclear	**Unclear**
	Lowing 2009[35]	High	High	High	High	Low	Low	High	**High**
IV	**Sorsdahl 2010**[36]	High	High	High	Low	High	Low	Unclear	**High**
Single-subject design									
I	**Thorpe 2002**[14]	Low	Unclear	High	High	Low	Low	Unclear	**Unclear**

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Level II-III group designs

(i) Task-specific training versus comparison

Interventions

Seven RCTs and one comparative study[31] (Table 1) compared TST to another intervention. The target of TST varied; four studies involved training a variety of tasks or movement skills[21 25 31 33], one focused on swimming[26] and another trained sit to stand[29]. Six studies compared TST to 'routine physiotherapy'[25-27 29 31 33]. This was generally poorly defined; described as neurodevelopmental therapy (NDT)[25 27 33] in three studies, physical education in one study[31] or not reported [26 29]. The remaining two studies involved over-ground walking compared to treadmill training; with over-ground walking deemed as the TST, as treadmill training was considered not specific to the gross motor aim of improving over-ground walking.

Two of the eight studies were goal-directed while four studies reported the TST was driven by motor learning principles[25 27 31 33] (Table 3). Repetitive practice was the most commonly reported motor learning strategy with feedback and task modification in four[25 27 28 33] and five[25 27 29 31 33] studies respectively. Physiotherapists generally conducted interventions but characteristics varied widely. Overall intervention time ranged from three[31] to 90 hours[25] over periods from 10 days[25] to six months[33]. Reported settings were largely ecological while format was group-based in two studies[25 26], otherwise was not stated.

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Table 3: Interventions: Components of task-specific training for included studies

Study	Task-specific intervention/s	Key characteristics of task-specific training									Components of the TIDiER checklist					
		Overall approach				Motor learning strategies reported					Dosage (Total hours)	Setting	Format	Providers	Adherence Reported?	Fidelity Reported?
		Goal-directed	*Motor learning driven*	*Feed back*	*Cogn. strat.*	*Repet. practice*	*Task mod.*	*Environ. mod*	*Obs. learning*	*Dual task learning*						
Group designs																
Bar-Haim 2010[27]	Motor learning coaching	✓	✓	✓		✓	✓	✓		✓	1 hour, 3 days/ week for 3 months (36 hours)	School	NS	Trained PT		
Bleyenheuft 2015[25]	HABITILE		✓	✓		✓	✓	✓		✓	9 hours/day for 10 consecutive days (90 hours)	Recreation camp	Group	Trained PTs, OTs, and PT / OT students	✓	
Declerck 2016[26]	Swimming skills program					✓					40-50 mins, 2 days/week for 10 weeks (15 hours)	Swimming pool	Group	PT and PT students	✓	
Grecco 2013[28]	Overground walking			✓		✓	✓				30 mins, 2 days/week for 7 weeks (7 hours)	NS	NS	Therapist		
Ketelaar 2001[33]	Functional therapy	✓	✓	✓		✓	✓	✓			6 months – intensity or hours NS (NS)	Clinic and home	NS	Trained PTs and parents		
Kumban 2013[29]	Sit-to-stand task training					✓	✓				20 min, 3 days/ week for 6 weeks (6 hours)	School	NS	Child's own PT		
Swe 2015[30]	Overground walking					✓	✓				30 mins, 2 days/ week for 8	School	NS	PTs	✓	

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AACPDM Level III & IV task-specific sys Comparison

										weeks (8 hours)				
	Capio 2015[31]	Functional motor skills training		✓			✓	✓			45 mins, 1 day/week for 4 weeks (3 hours)	NS	NS	Each child's regular PT	✓
Level I & III RCTs/Pseudo-RCTs	Hemayattal-ab 2010[32]	Throwing task with different feedback conditions		✓	✓		✓			✓	2 sessions in 2 days (NS)	NS	NS	NS	
	Hemayattal-ab 2013[34]	Throwing task with different feedback conditions		✓	✓		✓			✓	8 sessions Intensity or hours NS (NS)	NS	NS	Specialist trainers	
	Lowing 2009[35]	Activity focussed individual therapy vs goal-directed therapy	✓ (goal group only)				✓ inc. home program				3 sessions / fortnight for 12 weeks (NS)	Clinic and home / preschool	Group and indiv	PT, OT, SP, teacher and parents	
Level IV	Sorsdahl 2010[36]	Functional family-centred goal directed therapy	✓		✓		✓ inc. home program	✓	✓		3 hours, 5 days/ week for 3 weeks (45 hours)	Clinic	Group	Trained PTs and parents	✓
Single-subject design															
Level I	Thorpe 2002[14]	Novel motor task learning with different conditions		✓	✓	✓	✓				3 sessions over 5 days (NS)	School	Indiv	PT (also the PI)	

Table 3 abbreviations: TIDiER = Template for Intervention Descriptions and Replication, Cogn. = cognitive, Strat. = strategies, Mod = modification, Environ = environmental, Obs = Observational, HABITILE = Hand Arm Bimanual Intensive Training Including Lower Extremity, PT = Physiotherapist, OT = Occupational therapist, SP = Speech therapist, NS = Not stated, Indiv = individual, Trained = trained in specific intervention protocol, inc = including, PI = Principal investigator

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Outcomes and effects

All eight studies reported outcomes at the activity level, three reported participation-related outcomes and three involved body structure and function outcomes (Table 1). Outcomes were measured at one to three time points. In general T1 was immediately following the intervention, whilst T2 (4 weeks – 6 months)[25-30 33] and T3 (2 – 12 months)[26 33] were varied.

Activity outcomes

Gross motor skill performance was measured in five of the eight studies[25 26 28-30] with four measuring skill performance specific to the training tasks[26 28-30] (Table 1). Overall, skills and measures were varied. Four studies measured walking performance:[25 26 28 30] using the six minute walk test (6MWT)[25 28 30]or the timed up and go, one minute walk test, ten metre walk test and the ABILOCO-Kids questionnaire [25 26 28 30]. Between-group effects of TST on walking performance were mixed (Figures 2a-b). Large effects favouring TST on walking performance at T1 were found (ABILOCO Kids SMD = 0.96, 95% CI = 0.11, 1.81; 6MWT SMD = 0.87, 95% CI 0.02, 1.71) by *Bleyenheuft 2015*[25]. However, large negative effects at both T1 (6WMT SMD = -1.47, 95%CI -2.25, -0.69) and at 4 weeks follow up (6WMT SMD = 1.46, 95%CI 0.69, 2.24)) were found by *Grecco 2013*[28]. Non-significant mixed effects were found at both time points in the remaining two studies[26 30]. Swimming performance was measured using the Water Orientation Test Alyn – 2nd edition in one study[26] and sit to stand performance was measured using the five-times sit to stand test in another[29]. There was no between-group effect found for TST on sit to stand[29] or swimming performance at T1[26]. Within-group effects of TST were positive for all skill performance outcomes in each study, except for the GMFCS III subgroup in one study[29].

Overall gross motor function was measured in five of the eight studies[27 28 30 31 33] (Table 1). Four studies[27 28 30 33] utilised the Gross Motor Function Measure (GMFM)[37]. Three studies measured all domains of the GMFM[27 28 33] (A: lying and rolling, B: sitting, C: crawling and kneeling, D: standing, E walking running and jumping) while one study measured domains D and E only[30]. Nil or negative effect of TST was found at T1 or at T2 (Figures 3a-d)[27 28 30 33] on domains D and E. Three studies demonstrated no effect of TST at T1 or T2 in either domain[27 30 33] while the fourth study *(Grecco 2013)* showed a large negative effect for both domains at T1 (GMFM-D SMD = -1.39, 95%CI -2.16, -0.62; GMFM-E SMD = -1.97, 95%CI -2.82, -1.12) and T2 (GMFM-D SMD = -1.32, 95%CI -2.09, -0.56; GMFM-E SMD = -2.08, 95%CI -2.95, -1.22)[28].

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No significant effects of TST were found (Figures 3e-f)[27 33] on total GMFM score. Again, a large effect favouring the comparison was found for total score at both time points (T1 SMD = -1.83, 95%CI -2.63, -1.03; T2 SMD = -1.67, 95%CI -2.48, -0.87) by *Grecco 2013*[28]. The remaining level III design study reported a significant positive effect of TST on gross motor function as measured by the Test of Gross Motor Development - 2nd edition[31]. Within-group effects of TST on gross motor function were positive in all five studies.

Functional skill outcomes were measured in four of the eight studies[25 27 28 33] (Table 1). Functional skills included; self-care, assessed by the self-care domain of the Pediatric Evaluation of Disability Inventory (PEDI), in three studies[25 28 33]; mobility, measured by the PEDI mobility domain in two studies[28 33] and a parent questionnaire[27]; and social function assessed by the PEDI social function domain in one study[28]. A large effect (SMD 1.07, 95%CI 0.21, 1.94) favouring TST was found for self-care skills at T1 in *Bleyenheuft 2015*[25]. No significant effects were found for the other two studies measuring self-care at T1 and T2[28 33] (Figures 4a-b). No effect of TST on mobility skills (PEDI mobility domain) was found at T1 (Figure 4c) but a moderate effect was found at six months (T2 SMD = 0.58, 95% CI 0.04, 1.12,) and a moderate-large effect found at 12 month follow up (T3 SMD 0.76 95%CI 0.21, 1.31) by *Ketelaar 2007*[33]. In contrast, a large negative effect (SMD -1.32, 95%CI -2.09, -0.56) was found for mobility skills at T1 by *Grecco 2013*[28] (Figure 4c). No effect on mobility on the parent questionnaire was found at T1 or 6 months post[27] or on social function T1 or at 4 weeks follow up[28]. Within-group effects of TST on all functional skills outcomes were positive in all four studies.

Participation-related

Participation-related outcomes were measured in only three of the eight studies[25 26 31] (Table 1). Measures included social participation using the Life-HABITS performance and satisfaction questionnaire[25], adherence and enjoyment[26] and participation in physical activity using an accelerometer[31]. Large effects favouring TST were found for both social participation performance (SMD = 1.19, 95%CI 0.31, 2.07) and satisfaction (SMD = 1.29, 95%CI 0.40, 2.18) by *Bleyenheuft 2015*[25]. Positive effects of TST on weekend physical activity were reported at T1[31]. High adherence rates and enjoyment levels for TST were also reported but without comparison group data [26].

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(ii) Task-specific versus task-specific

Interventions

Three studies compared two or more task-specific interventions including one RCT (level II)[32] and two comparative studies (level III)[34 35]. Two studies[32 34] compared the effect of TST programs with different feedback conditions on a throwing task (Table 1). The throwing task training was informed by motor learning principles but was not goal-directed (Table 3). The intensity of TST in both studies was approximately two hours over two days[32 34]. The other study examined the effects of goal-directed TST compared to activity-based TST[35], with 18 sessions over 12 weeks with parent involvement across various settings[35]. The goal-directed program involved group and individual sessions while the activity program was individual only.

Outcomes

Gross motor skill performance (specific to the trained task), assessed by throwing accuracy, was the sole outcome in both throwing studies[32 34]. This was assessed immediately post intervention (T1 - acquisition) in both studies, 24 hours later in one[32] (T2 - retention) and three days later (T2) in the other[34]. Outcomes were assessed at T1 only in the remaining study: including gross motor function (total GMFM score) and functional skills (all domains of the PEDI). The participation-related outcome, goal attainment, was assessed in the goal-directed group only by the goal attainment scale (GAS) [35].

Knowledge of results provided every trial was found to improve throwing accuracy significantly more than 50% or no feedback at T1. However, at T2, those receiving knowledge of results 50% of the time performed significantly better than those who received feedback every trial or no feedback at all[34]. Self-controlled feedback was found to improve throwing accuracy significantly more than yoked feedback at T2 but not at T1[32]. Goal-directed training improved overall gross motor function and the functional skills of mobility and self-care significantly more than activity-based training[35]. There was no difference in social function between the groups. Eighty-five percent of goals were attained with goal-directed TST[35].

Level IV group design

One repeated measures study involving goal-directed, group-based, intensive TST was included[36] (Table 1, Table 3). Gross motor function (overall GMFM score), functional skills (mobility, self-care and social function using the PEDI) and goal attainment (GAS) were measured at three baseline time points, immediately (T1) and 3 weeks (T2) after the intervention. Positive effects were reported for overall gross motor function at both T1 and T2, and for self-care skills at T2, while 66% of goals were attained at T2.

Single-subject design

One Level I single-subject design study involving randomisation to one of three feedback protocols for learning to move an exercise vehicle backward was included[14] (Table 1). Gross motor skill performance (specific to the trained task) was measured using backwards displacement of the vehicle two days following the training (T1). Eight of the 13 participants demonstrated significant improvement at T1. No specific feedback protocol was clearly superior.

Intervention replicability

Reporting of intervention characteristics varied widely (Table 3). Four studies reported intervention providers were trained in the intervention protocol[25 27 33 36], five studies reported the format of the intervention[14 25 26 35 36] while participant adherence was described in five studies[25 26 30 31 36] . No study reported provider fidelity while comparison interventions were generally ambiguously reported.

Quality of the evidence by outcome

The overall quality of evidence was moderate for gross motor skill performance and functional skills and low for gross motor function and participation-related outcomes.

DISCUSSION

To our knowledge, this is the first systematic review to evaluate and synthesise the evidence for task specific gross motor skills training in ambulant school aged children with CP. Given the focus of TST is on practice of tasks rather than remediating impairments, and the increasing recognition of importance of child and family-centred effects of interventions,

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this review explicitly focused on activity and participation outcomes. In general, positive within-group effects of TST were reported across outcomes of interest. However, in RCTs where TST was compared to comparison interventions, between-group effects were largely non-significant with the exception of two studies[25 28] reporting large but conflicting effects. Overall, there was moderate quality evidence for conflicting effects of TST to improve specific skills performance and functional skills but low quality evidence showing no difference or negative effects on gross motor function. For participation-related outcomes, low quality evidence for positive effects of TST was found. Positive effects across all outcomes were generally found immediately following TST, with evidence of longer-term retention lacking.

The secondary aim of this study was to identify motor learning strategies reported in TST and assess any relationship to outcome. The overall poor reporting of motor learning strategies and heterogeneity in interventions and outcomes limited definitive conclusions. However, this review provides some insight into potentially important characteristics of interventions, in particular; practice dosage, feedback and goals. Unsurprisingly, given its intrinsic relationship to TST, the most consistently reported motor learning strategy was repetitive practice. While reporting of dosage was variable, the largest positive effects of TST were found where dosage was highest[25]. Although no specific feedback condition emerged as clearly superior in the three studies comparing these[14 32 34], results suggest different feedback conditions may influence the phases of training differently. Replication of these studies for varying tasks is required to provide further clarity into the role of feedback. Although causal inferences are limited due to lack of randomisation, better activity outcomes were found when the TST was goal-directed[35]. These results are consistent with the growing evidence base for interventions targeted towards the goals of children with CP and their families[6 16].

Our review found some conflicting results between studies, with large positive and negative effects found in *Bleyenheuft 2015*[25] and *Grecco 2013*[28] respectively. The TST in *Bleyenheuft 2015*[25] was high in dose, explicitly driven by motor learning principles and involved of a wide range of gross motor tasks including ball skills, cycling and walking. By contrast, in *Grecco 2013*[28] dosage of TST (over-ground walking) was low in comparison and limited motor learning strategies were reported. The comparison treatment in this study was treadmill training without body weight support – training that may be regarded as task-related but was not deemed TST given the primary goal of the study was to improve over-ground walking. Whilst practice dose was equal between the groups in *Grecco 2013*[28], task

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progression was potentially greater with the use of treadmill functions. These results suggest task-related adjuncts, such as treadmill training, may be more beneficial than pure TST for developing (rather than acquiring) gross motor skills, such as walking in this already-ambulant population.

Limitations across the included studies

Low-moderate quality of evidence was found across the outcomes due to significant methodological limitations in the studies, in addition to risk of bias. First, sample size calculation was not reported in most studies (Table 1). Wide confidence intervals found for outcome data from Level II-III studies (Figures 2-4) suggest inadequately powered samples may have reduced precision and thus limit the generalizability of the findings. Second, the limited detail in reporting potentially reduces the robustness of research findings if the study cannot be replicated and limits implementation. Third, limitations in outcome measurement selection may have influenced the generalisability and transferability of some findings. The GMFM and the PEDI are a well-established tools for evaluating change in children with CP[38], and were used in six[27 28 30 33 35 36] and four studies[25 28 33 35] respectively. However, concerns have been raised about the responsiveness of these measures in higher functioning children[38], the target population of this review. Whilst common use makes them appealing, their broad focus means skills targeted by the TST may not have been adequately captured.. Finally, far fewer participation-related outcome measures were used compared to activity outcomes thus any evidence that improved activity through TST leads to improved participation is weak at best.

Limitations of this review

This review chose to narrow the inclusion criteria to ambulant school aged children with CP, interventions of gross motor skill TST, and activity and participation domains outcomes, in order to draw specific conclusions. This was not possible for all questions posed by this review due to study design issues and heterogeneity, which also precluded meta-analysis. Further, the methodology of the review itself has some limitations. The impact of publication bias was not evaluated. Inclusion was limited to published articles in English meaning some studies, including grey literature, may have been missed. Further, non-randomised studies were retained in this review. Although the Cochrane Risk of Bias tool and the GRADE system are the most widely used systems for assessing risk of bias and the

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quality of the evidence across outcomes respectively[20 24], these tools do emphasise randomised studies.

Conclusions and recommendations

Despite the largely positive within-group effects of TST over time across each outcome of interest, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes while no difference or negative effects were found for gross motor function. Given the low-moderate quality of this evidence, there is currently limited evidence to support task-specific gross motor skills training for improving these activity and participation-related outcomes in children with CP. Clear recommendations around whether TST is superior to other interventions cannot be made. Before conclusions can be made about any relationship of motor learning strategies to outcome, more consistent reporting and studies designed to test this are required.

The importance of tailoring motor interventions to individual goals and lifelong physical activity is increasingly being recognised[4 39]. Thus, to enable specific recommendations, strengthening the evidence base is imperative. Adequately powered samples, rigorous study design and consistent reporting with attention to reporting interventions to allow for reproducibility and appropriate evidence synthesis is required. Future challenges also include considering issues with outcome measure responsiveness and intervention heterogeneity, and optimising TST through the use of motor learning strategies.

Competing interests

The authors have no competing interests to declare.

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Data sharing statement

Data relating to the quantitative synthesis is available on request.

Author contributions

All named authors contributed to the design of this review. RT collected data, carried out data analyses and led interpretation and reporting. CB collected data and contributed to interpretation and reporting. ARH, JLM and AJS contributed to data analysis, interpretation and reporting. All named authors approved the final manuscript as submitted.

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Figure legend

Figure 1: PRISMA flow chart of included and excluded studies

Figure 2a: Gross motor skill performance - Task specific vs alternative: walking performance at T1 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 2b: Gross motor skill performance - Task specific vs alternative: walking performance at T2 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 3a: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T1.

Figure 3b: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T2.

Figure 3c: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T1.

Figure 3d: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T2.

Figure 3e: Gross motor function - Task specific vs alternative: Overall GMFM score at T1.

Figure 3f: Gross motor function - Task specific vs alternative: Overall GMFM score at T2.

Figure 4a: Functional skills - Task specific vs alternative: Self-care at T1.

Figure 4b: Functional skills - Task specific vs alternative: Self-care at T2.

Figure 4c: Functional skills - Task specific vs alternative: Mobility skills at T1

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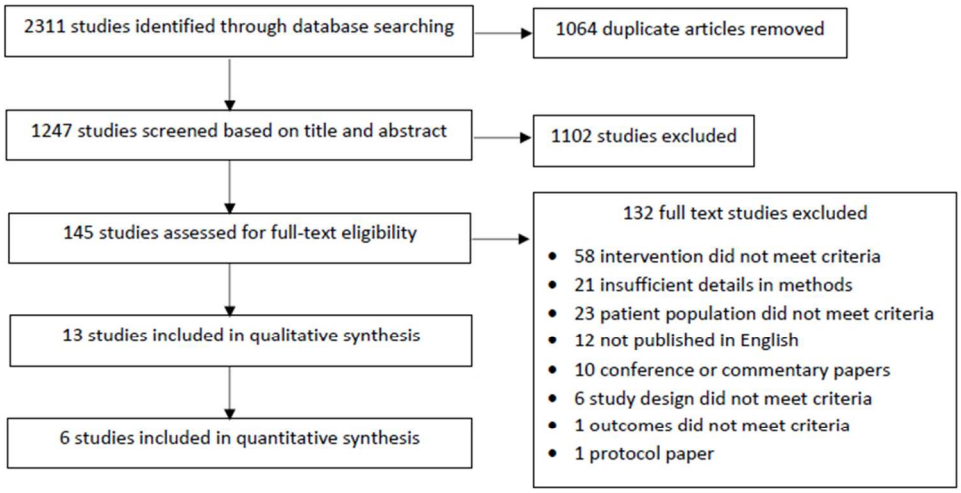

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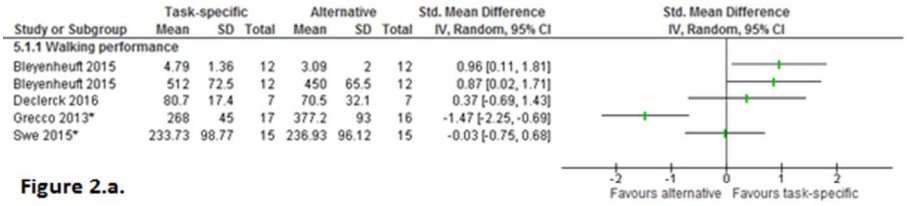

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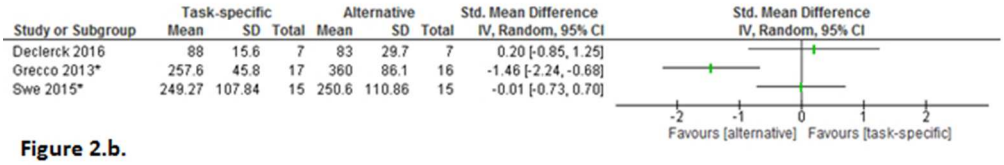

Figure 2.b.

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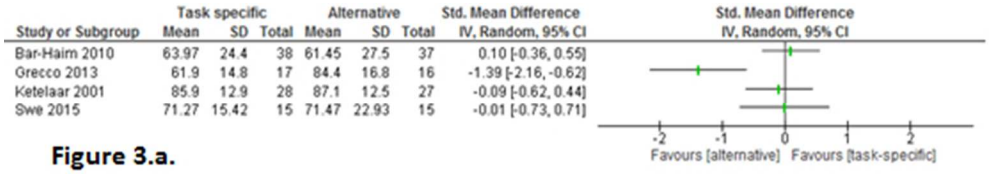

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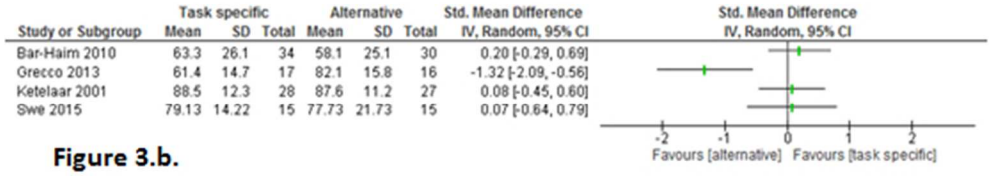

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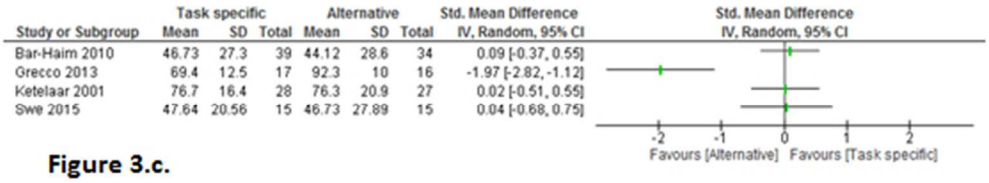

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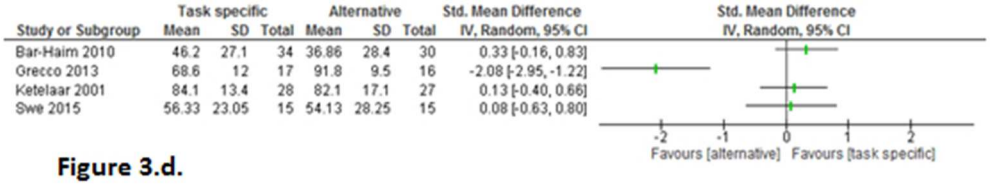

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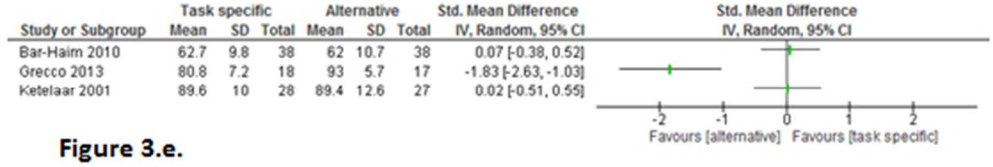

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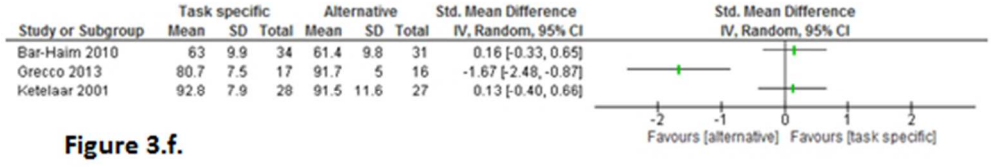

Figure 3.f.

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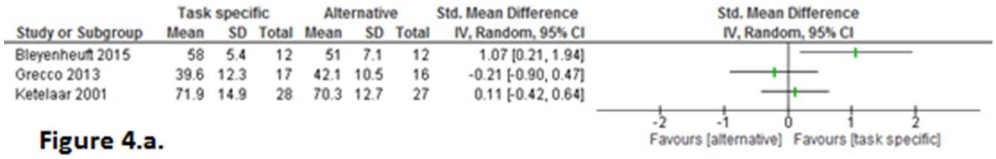

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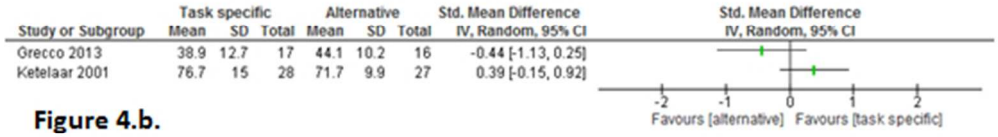

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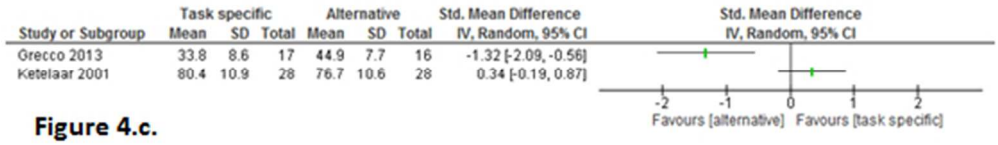

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APPENDIX 1

Search strategy for Ovid databases

1. cerebral palsy.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
2. child*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. (Task specific or skill specific or task oriented or activity focussed or task practice or goal directed).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
4. (motor imagery or implicit or error reduced or neuromotor task or parent assisted or group or cognitive orientation or repetit*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. (perceptual motor or kinaesthetic explicit or action observation or dual task or trial and error or practice or imitation or experiential or discrimination or discovery or errorless or analogy or observational or exploratory).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
6. (Training or intervention or therapy or approach or learning or program).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. (Motor skill* or motor function* or movement skill* or Motor performance or task performance or occupational performance or motor competence or task competence or occupational competence or skill acquisition or skill attainment or skill proficiency or skill achievement or goal attainment).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
8. 3 or 4 or 5
9. 1 and 2 and 6 and 7 and 8

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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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Keywords:	Neurodisability, Physcial therapy, Rehabilitation

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Task-specific gross motor skills training for ambulant school aged children with cerebral palsy: a systematic review

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ABSTRACT

OBJECTIVES: The primary objective is to systematically evaluate the evidence for the effectiveness of task-specific training (TST) of gross motor skills for improving activity and/or participation outcomes in ambulant school aged children with cerebral palsy (CP). The secondary objective is to identify motor learning strategies reported within TST and assess relationship to outcome.

DESIGN: Systematic review

METHOD: Relevant databases were searched for studies including: children with CP (mean age >4 years and >60% of the sample ambulant); TST targeting gross motor skills; and activity (skill performance, gross motor function and functional skills) and/or participation-related outcomes. Quality of included studies was assessed using standardised tools for risk of bias, study design and quality of evidence across outcomes. Continuous data were summarised for each study using standardised mean difference (SMD) and 95% confidence intervals (CI).

RESULTS: Thirteen studies met inclusion criteria: eight randomised controlled trials (RCT), three comparative studies, one repeated-measures study and one single-subject design study. Risk of bias was moderate across studies. Components of TST varied and were often poorly reported. Within-group effects of TST were positive across all outcomes of interest in 11 studies. In RCTs, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes (one study: Life-HABITS performance SMD = 1.19, 95% CI 0.3, 2.07, p< 0.001; Life-HABITS satisfaction SMD = 1.29, 95% CI 0.40, 2.18, p =0.001) while no difference or negative effects were found for gross motor function. The quality of evidence was low-moderate overall. Variability and poor reporting of motor learning strategies limited assessment of relationship to outcome.

CONCLUSIONS: Limited evidence for TST for gross motor skills in ambulant children with CP exists for improving activity and participation-related outcomes and recommendations for use over other interventions are limited by poor study methodology and heterogeneous interventions.

REGISTRATION: PROSPERO ID42016036727

KEY MESSAGES

What is known about the subject

- Strong evidence for motor interventions involving task specificity for functional mobility in adults post stroke and for upper limb function in children with cerebral palsy exists
- The effectiveness of task-specific gross motor skills training in ambulant school aged children with cerebral palsy has not been systematically evaluated or synthesised

What this study adds

- A low-moderate overall quality of evidence was found for task-specific gross motor skills training for ambulant school aged-children with cerebral palsy
- Limited evidence for task-specific training to improve specific skills performance, functional skills and participation-related outcomes exists
- While clear recommendations for use of task-specific training over other interventions are limited, ways to strengthen the evidence in future studies are identified

Short title: Task-specific gross motor skills training in CP

Key words: cerebral palsy, task-specific training, gross motor skills, activity, participation

ABBREVIATIONS

CP	Cerebral palsy
GMFCS	Gross motor Function Classification System
ICF	International Classification of Functioning, Disability and Health
TST	Task-specific training
AACPDM	American Academy of Cerebral Palsy and Developmental Medicine
SMD	Standardised mean difference
CI	Confidence interval
RCT	Randomised controlled trial
NDT	Neurodevelopmental therapy
GMFM	Gross Motor Function Measure

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PEDI Pediatric Evaluation of Disability Inventory

Cerebral palsy (CP) is an umbrella term used to describe a group of disorders of movement which cause varying degrees of activity limitations[1]. The most widely used means for classifying gross motor function in children with CP is the Gross Motor Function Classification System (GMFCS)[2]. Children classified GMFCS I are able to walk and run but have limitations with speed, balance and coordination whilst children classified GMFCS V are transported in a wheelchair in all settings. Although the focus of the GMFCS is on functional mobility, the realm of gross motor activities, that is skills involving movement of the large muscles of the limbs or whole body, undertaken by children is much broader[3]. Development of gross motor skills underpins functional, play and social activities across childhood and complex movement skills required for sports in older children[3]. In children with CP, limitations in gross motor function increase as GMFCS level increases, however, children at all GMFCS levels (I-V) participate, on average, less in physical activities than their typically developing peers[2]. This is an issue because of the known poor health outcomes in adulthood due to inactivity in childhood[4]. Effective interventions tailored to GMFCS levels and developmental stages are required to improve these outcomes in this population.

The International Classification of Functioning, Disability and Health (ICF) has become the common language for clinicians, researchers and families for understanding the effect of CP on the individual and for targeting interventions[5,6]. Where interventions previously focused on remediating limitations in body structures and functions, there has been a more recent acknowledgement of the importance of the effect of interventions within the activity and participation domains[7 8]. Clinicians working with children with CP need guidance from evidence synthesis to implement effective means of improving physical skills *and* improve the uptake of these skills in the child's daily life. The historical bias towards impairment-focused motor interventions yielded few effective treatments[6 9 10] thus more functional approaches have emerged.

Task-specific training (TST) involves practice of context-specific tasks where the intervention focuses on the skills needed for a task(s)[11] - there is similarity between the training task and the goal of the intervention. Although Level I evidence exists for TST to improve gross motor activities in adults after stroke[12], the majority of high level evidence for interventions involving task specificity in children with CP relates to training of upper limb or fine motor activities[6 13] with limited evidence for gross motor skills training. TST inherently involves principles of motor learning with components including context, practice and

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dosage[11]. Other motor learning strategies, such as feedback and task modification, have the potential to optimise TST, however, this has not been systematically studied[14]. TST should involve varied components depending on the requirements of the skill, the environment and the function of the child[15]. Moreover, training for a child of higher-level motor function (e.g. GMFCS I-III) should be targeted towards different skills compared to training with a child of lower-level motor function (e.g. GMFCS IV-V). Similarly, children of different ages and developmental stages have varying learning capabilities, and physical demands placed on them by their context[16].

Previous systematic reviews of motor interventions in children with CP have been broad in terms of ages (including infants and children) and motor function (all GMFCS levels), and included interventions have been heterogeneous[17 18]. TST may be a promising approach for ambulant children who have specific gross motor skills goals, however, there has been no systematic review to examine the effectiveness of this approach. The primary aim of this study is to evaluate and synthesise the evidence for the effectiveness of task-specific gross motor skills training in ambulant children aged 4 – 18 years with CP for activity and participation outcomes. The secondary aim of this study is to identify motor learning strategies reported within TST and assess relationship to outcome.

METHOD

Eligibility criteria

Published studies were included if they met all of the following criteria:

1. Level of evidence: All group design studies categorised as level II-IV using the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) levels of evidence[19] and studies classified as level I single-subject designs involving over 10 participants.
2. Population: The majority (i.e. > 60%) of the participants were ambulant children with CP (GMFCS I-III), and the mean sample age was four – 18 years.
3. Interventions: TST of gross motor skills where there was similarity between the training task and the goal of the intervention, including those interventions described as involving motor learning strategies/coaching, goal-directed training, activity focused training and/or functional skills training. Any duration or intensity of TST.

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4. Comparison: Studies comparing TST to another intervention, another type of TST or no intervention.
5. Outcomes: Activity outcomes including; gross motor skill performance (specific to the task being trained or other gross motor task to assess for transferability), gross motor function and functional skills; and participation-related outcomes. Only studies reporting outcomes separately for children with CP.

Exclusion criteria: TST was applied within a combined intervention approach and the influence of TST could not be isolated (e.g. Botulinum Toxin-A, virtual reality, treadmill training, orthoses or robotics), greater than ten percent of the intervention was passive or the article was not in English.

Search strategy

Relevant articles were identified from Medline, EMBASE, CINAHL, Psycinfo, SPORTDiscus and PubMed with all searches limited to articles published in peer-reviewed journals in English. A search was conducted in June 2016 and search terms were tailored for each database. Reference lists of included studies and related reviews were also searched. See Appendix 1 for full search strategy for Ovid databases (Medline, EMBASE and PubMed).

Study selection, data extraction and quality assessment

Study selection, data extraction and quality assessment was completed by two authors (RT and CB) independently, with a third author (AS, AH or JM) to resolve any disagreements. If inclusion was uncertain from abstract, the full text was retrieved.

For included studies, data were extracted using a customised form based on the Cochrane recommendations[20]. Data extracted included: study details (author, year, country, funding), study design, AACPDM level of evidence[19], study sample characteristics , detailed TST characteristics (including motor learning strategies), comparison intervention characteristics, outcomes measured and associated ICF domain, effects of the intervention and conclusions. Reported motor learning strategies in all TST, including when studies involved a comparison of two or more TST approaches, were identified using pre-defined motor learning strategy codes based on current literature[21 22]. Authors were contacted for complete data extraction when needed.

Risk of bias for individual studies was assessed using the Cochrane Risk of Bias Tool[20]. Overall risk of bias was determined by predominant level of bias (at least four of the

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seven domains) across the domains of the Risk of Bias Tool. Intervention replicability was assessed using relevant components of the Template for Intervention Descriptions and Replication (TIDieR) Checklist[23]. The Grading of Recommendations Assessment Development and Evaluation (GRADE) system, was used to assess the quality of the evidence across outcomes[24].

Narrative synthesis and data analysis

Study design and AACPDM level of evidence were used for narrative synthesis. Higher-level group designs (AACPDM levels II-III) were reported together, whilst lower-level group designs (IV) and single-subject design studies were reported separately. Within the level II-III group designs, studies were considered as either 'TST versus a comparison' or 'TST versus TST'. Finally, within each grouping, activity outcomes were organised into 3 constructs; gross motor skill performance, functional skills and gross motor function whilst participation-related outcomes were grouped together. Gross motor skill performance outcomes included those measuring a specific gross motor skill (e.g. walking or throwing), functional skills outcomes included those measuring a broader range of skills in domains related to function (e.g. self-care or social skills) while gross motor function outcomes included those measuring a range of gross motor related skills.

Analyses were conducted using Cochrane Review Manager Software RevMan 5.3. Only data from level II studies comparing TST to comparison interventions were included in quantitative analysis to ensure comparison was between studies of similar design. Continuous data were summarized for each study within outcomes of interest using standardized mean difference (SMD) and 95% confidence intervals (CIs). A SMD of 0.2 was considered small, 0.4-0.6 moderate and 0.8 a large effect size[20]. Given the heterogeneity between studies in tasks and characteristics of the TST and comparison interventions, meta-analysis was not undertaken.

RESULTS

Following removal of duplicates, the abstracts of 1247 studies were screened and 145 full-text articles were retrieved for full appraisal (Figure 1). Thirteen studies involving 405 participants met inclusion criteria and underwent narrative synthesis with six of these studies involving 237 individuals included in data analysis. Characteristics of included studies are summarised in Table 1.

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The 13 studies included 12 group designs and one single-subject design (level I). The group designs involved eight randomised controlled trials (RCTs) (level II), three comparative studies with concurrent controls (level III) and one repeated measures study (level IV). Eleven studies did not report adverse events as an outcome and two studies reported no adverse events[25 26].

Risk of bias assessment

Risk of bias was low in four studies[14 27-30], unclear in seven studies[14 25 26 31-34] and high in two studies[34-36] leading to an overall moderate risk of bias across the studies (Table 2). Major sources of bias included: studies without randomisation or allocation concealment, a lack of participant/personnel blinding and incomplete outcome data. No study reported the use of intention to treat analysis and reporting of outcome assessor blinding was inconsistent. Poor reporting of study conduct limited assessment of risk of bias in the seven studies with unclear risk of bias.

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Table 1: Characteristics of included studies

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Study	Study design	AACPDM LoE	CP n	Age Range, Mean and SD in years	GMFCS levels / proportions	Task-specific training (T)	Comparison (C) or other task-specific training	Activity outcomes			*Participation related outcomes (measure)*	*Other outcomes (ICF domain)*
								Gross motor function	*Functional skills*	*Gross motor skill performance*		
								Outcome measured used				
1. Group research designs												
a. Level II – III studies (Task specific vs comparison)												
Bar-Haim 2010[27]	RCT	II	Total = 78 T = 39 C = 39	Range = 5.5 – 12.2 Total = 8.9 +/- 1.7 T = 8.8 +/- 1.7 C = 8.9 +/- 1.7	T II = 36%, III = 64% C II = 44% III = 56%	Motor learning coaching	Routine physiotherapy - NDT	GMFM	Parent survey			Mechanical efficiency (BSF)
Bleyenheuft 2015[25]	Crossover RCT	II	Total = 24* T = 12 C = 12	T = 8.9 +/- 1.7 C = 8.5 +/- 1.7	T I = 50% II = 50% C I = 42% II = 58%	Hand arm bimanual intensive training including lower extremity (HABIT-ILE)	Routine physiotherapy (in general had NDT while waiting for delayed HABIT-ILE)		PEDI (self-care domain only)	6MWT ABILOCO Kids	Social participation (Life-H Performance and Satisfaction)	Body weight distribution (BSF) Mean step length (BSF) UL function (Act) Adverse events
Declerck 2016[26]	RCT	II	Total = 14 T = 7 C = 7	Range = 7 – 17 T = 8.7 +/- 3.4 C = 11.8 +/- 3.5	T I = 14% II = 86% C I = 29% II = 57% III = 14%	Swimming skills program	Routine physiotherapy			WOTA-2† 1MWT	Adherence/ enjoyment	Pain (BSF) Fatigue (BSF) Adverse events
Grecco 2013[28]	RCT	II	Total = 36* T = 17 C = 18	T = 6.8 +/- 2.6 C = 6.0 +/- 1.5	T I = 47% II = 41% III = 12% C I = 31% II = 50% GMFCS III = 19%	Overground walking	Treadmill training	GMFM	PEDI	6MWT† TUG		Balance (Act)
Ketelaar	RCT	II	Total = 55	Range (total) =	T	Functional	Routine	GMFM	PEDI			

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2001[33]			T = 28 C = 27	2.0 – 7.25 T = 4.5 +/- 1.7 C = 4.7 +/- 1.7	mild = 79%, mod = 21% C mild = 77%, mod = 23%	therapy	physiotherapy - generally NDT or Votja method.					
Kumban 2013[29]	RCT	II	Total = 21 T = 10 C = 11	T = 12.3 +/- 2.6 C = 12.4 +/- 2.4	T I-II = 60%, III = 40% C I-II = 55% III = 45%	Regular PT + sit-to-stand task training	Routine physiotherapy			FTSST†		Balance ability (Act) Functional balance and lower limb strength (Act)
Swe 2015[30]	RCT	II	Total = 30* T = 15 C = 15	Total = 13.2 +/- 3.4 T = 13.0 +/- 3.6 C = 13.4 +/- 3.3	T II = 53% III = 47% C II = 67%, III = 33%	Overground walking	Partial weight support treadmill training	GMFM (standing and walking domains only)		6MWT† 10MWT†		
Capio 2015[31]	Comparative study with concurrent controls	III	Total = 24 T = 12 C = 12	T = 6.92 +/- 3.04 C = 7.98 +/- 1.74	T I = 33% II = 58% III = 8% C I = 25% II = 58% III = 17%	Functional motor skills training	Routine physiotherapy or physical education	TGMD- 2			Physical activity (accelerome-ter)	
b. Level II – III (task specific vs task specific)												
Hemayatt - alab 2013[32]	RCT	II	Total = 20 T1 = 10 T2 = 10	Total = 11.6 +/- 1.5 T1 = 11.9 +/- 1.6 T2 = 11.3 +/- 1.4	I-III Proportions not stated	Throwing task + self-control feedback	Throwing task + yoked feedback			Throwing accuracy†		
Hemayatt - alab 2010[34]	Comparative study with concurrent controls	III	Total = 24 T1 = 8 T2 = 8 T3 = 8	Range (total) = 7-15	I = 100%	T1 = dart throwing task + 50% KR T2 = dart throwing task + 100% KR	T3 = Dart throwing task + 0% KR			Throwing accuracy†		
Lowing	Comparative	III	Total = 44	Total = 4.1 +/-	T	Activity	Goal directed	GMFM	PEDI		Goal	

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2009[35]	study with concurrent controls		T = 22 C = 22	1.4 T = 4.3 +/- 0.7 C = 3.8 +/- 1.3	I = 45% II = 23%, III = 14% IV = 18% C I = 41% II = 23%, III = 22% IV = 14%	focussed individual therapy	functional group therapy				attainment (GAS) in GDT group only	
c. Level IV												
Sorsdahl 2010[36]	Repeated measures design	IV	T = 22*	Range = 2.8 – 9.25 Average age = 5.5	T I = 36% II = 23%, III = 27% IV-V = 14%	Functional family-centred goal directed therapy		GMFM GMPM			Goal attainment (GAS)	Quality of fine motor movements (Act)
2. Single-subject research designs												
Thorpe 2002[14]	13 x n=1 studies with random sampling to protocols (A, B or C)	I	N(total) = 13	Range = 6.0 – 12.7 Age = 8.6 +/- 1.89	III = 100%	All protocols = task specific A = 2 x no AI 1 x KP and 5 x KP+CS	B = 3 x no AI, 2 x KP and 3 x KP+CS C = 4 x no AI, 3 x KP and 1 x KP+CS			Change in backward displacement†		

Table 1 notes and abbreviations: * = sample size justified with statistical consideration, † = skill performance outcome is specific to skill being trained LoE = level of evidence, SD = standard deviation, GMFCS = Gross Motor Function Classification System, T = task-specific intervention, C = comparison, ICF = International Classification of Functioning, Disability and Health, NDT = Neurodevelopmental therapy, GMFM = Gross motor function measure (-88 or -66), BSF = Body structures and functions, PEDI = Pediatric Evaluation of Disability Inventory, 6MWT = Six minute walk test, ABILOCO = measure of locomotion ability, Life-H = Assessment of Life Habits, Act = activity, WOTA-2 = Water Orientation Test Alyn – 2nd edition, 1MWT = one minute walk test, 10MWT = ten metre walk test, TGMD – 2 = Test of Gross Motor Development – 2nd edition, KR = Knowledge of results, GAS = Goal Attainment Scale, GMPM = Gross Motor Performance Measure, AI = Augmented Information, KP = Knowledge of Performance, CS = cognitive strategies

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Table 2: Risk of bias within included studies

	Study	Selection bias		Performance bias	Detection bias	Attrition bias	Reporting bias	Other bias	Overall bias
		Random sequence generation	Allocation concealment	Participant and personnel blinding	Outcome assessor blinding	Incomplete outcome data	Selective outcome reporting		
Group research designs									
Level II and III studies	**Bar-Haim 2010**[27]	Low	Low	High	Low	High	Low	High	**Low**
	Bleyenheuft 2016[25]	Low	Low	High	Unclear	High	Unclear	Unclear	**Unclear**
	Declerck 2016[26]	Low	Low	High	Unclear	Unclear	Low	High	**Unclear**
	Grecco 2013[28]	Low	Low	High	Low	Unclear	Low	Unclear	**Low**
	Ketelaar 2001[33]	Low	Unclear	High	Low	Unclear	Unclear	Unclear	**Unclear**
	Kumban 2013[29]	Low	Low	High	Low	High	Low	Unclear	**Low**
	Swe 2015[30]	Low	Low	High	Low	Low	Low	Unclear	**Low**
	Capio 2015[31]	High	High	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2013[32]	High	Unclear	High	Unclear	Unclear	Unclear	Unclear	**Unclear**
	Hemayattalab 2010[34]	Unclear	High	High	Unclear	Low	Low	Unclear	**Unclear**
	Lowing 2009[35]	High	High	High	High	Low	Low	High	**High**
IV	**Sorsdahl 2010**[36]	High	High	High	Low	High	Low	Unclear	**High**
Single-subject design									
I	**Thorpe 2002**[14]	Low	Unclear	High	High	Low	Low	Unclear	**Unclear**

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Level II-III group designs

(i) Task-specific training versus comparison

Interventions

Seven RCTs and one comparative study[31] (Table 1) compared TST to another intervention. The target of TST varied; four studies involved training a variety of tasks or movement skills[21 25 31 33], one focused on swimming[26] and another trained sit to stand[29]. Six studies compared TST to 'routine physiotherapy'[25-27 29 31 33]. This was generally poorly defined; described as neurodevelopmental therapy (NDT)[25 27 33] in three studies, physical education in one study[31] or not reported [26 29]. The remaining two studies involved over-ground walking compared to treadmill training; with over-ground walking deemed as the TST, as treadmill training was considered not specific to the gross motor aim of improving over-ground walking.

Two of the eight studies were goal-directed while four studies reported the TST was driven by motor learning principles[25 27 31 33] (Table 3). Repetitive practice was the most commonly reported motor learning strategy with feedback and task modification in four[25 27 28 33] and five[25 27 29 31 33] studies respectively. Physiotherapists generally conducted interventions but characteristics varied widely. Overall intervention time ranged from three[31] to 90 hours[25] over periods from 10 days[25] to six months[33]. Reported settings were largely ecological while format was group-based in two studies[25 26], otherwise was not stated.

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Table 3: Interventions: Components of task-specific training for included studies

Study	Task-specific intervention/s	Key characteristics of task-specific training									Components of the TIDiER checklist					
		Overall approach				Motor learning strategies reported					Dosage (Total hours)	Setting	Format	Providers	Adherence Reported?	Fidelity Reported?
		Goal-directed	*Motor learning driven*	*Feed back*	*Cogn. strat.*	*Repet. practice*	*Task mod.*	*Environ. mod*	*Obs. learning*	*Dual task learning*						
Group designs																
Bar-Haim 2010[27]	Motor learning coaching	✓	✓	✓		✓	✓	✓		✓	1 hour, 3 days/ week for 3 months (36 hours)	School	NS	Trained PT		
Bleyenheuft 2015[25]	HABIT-ILE		✓	✓		✓	✓	✓		✓	9 hours/day for 10 consecutive days (90 hours)	Recreation camp	Group	Trained PTs, OTs, and PT / OT students	✓	
Declerck 2016[26]	Swimming skills program					✓					40-50 mins, 2 days/week for 10 weeks (15 hours)	Swimming pool	Group	PT and PT students	✓	
Grecco 2013[28]	Overground walking			✓		✓	✓				30 mins, 2 days/week for 7 weeks (7 hours)	NS	NS	Therapist		
Ketelaar 2001[33]	Functional therapy	✓	✓	✓		✓	✓	✓			6 months – intensity or hours NS (NS)	Clinic and home	NS	Trained PTs and parents		
Kumban 2013[29]	Sit-to-stand task training					✓	✓				20 min, 3 days/ week for 6 weeks (6 hours)	School	NS	Child's own PT		
Swe 2015[30]	Overground walking					✓	✓				30 mins, 2 days/ week for 8	School	NS	PTs	✓	

AACPDM Level II & III task-specific sys Comparison

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										weeks (8 hours)				
	Capio 2015[31]	Functional motor skills training		✓			✓	✓			45 mins, 1 day/week for 4 weeks (3 hours)	NS	NS	Each child's regular PT	✓
Level I & III RCTs/Pseudo-RCTs	Hemayattal-ab 2010[32]	Throwing task with different feedback conditions		✓	✓		✓			✓	2 sessions in 2 days (NS)	NS	NS	NS	
	Hemayattal-ab 2013[34]	Throwing task with different feedback conditions		✓	✓		✓			✓	8 sessions Intensity or hours NS (NS)	NS	NS	Specialist trainers	
	Lowing 2009[35]	Activity focussed individual therapy vs goal-directed therapy	✓ (goal group only)				✓ inc. home program				3 sessions / fortnight for 12 weeks (NS)	Clinic and home / preschool	Group and indiv	PT, OT, SP, teacher and parents	
Level IV	Sorsdahl 2010[36]	Functional family-centred goal directed therapy	✓		✓		✓ inc. home program	✓	✓		3 hours, 5 days/ week for 3 weeks (45 hours)	Clinic	Group	Trained PTs and parents	✓
Single-subject design															
Level I	Thorpe 2002[14]	Novel motor task learning with different conditions		✓	✓	✓	✓				3 sessions over 5 days (NS)	School	Indiv	PT (also the PI)	

Table 3 abbreviations: TIDiER = Template for Intervention Descriptions and Replication, Cogn. = cognitive, Strat. = strategies, Mod = modification, Environ = environmental, Obs = Observational, HABITILE = Hand Arm Bimanual Intensive Training Including Lower Extremity, PT = Physiotherapist, OT = Occupational therapist, SP = Speech therapist, NS = Not stated, Indiv = individual, Trained = trained in specific intervention protocol, inc = including, PI = Principal investigator

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Outcomes and effects

All eight studies reported outcomes at the activity level, three reported participation-related outcomes and three involved body structure and function outcomes (Table 1). Outcomes were measured at one to three time points. In general T1 was immediately following the intervention, whilst T2 (4 weeks – 6 months)[25-30 33] and T3 (2 – 12 months)[26 33] were varied.

Activity outcomes

Gross motor skill performance was measured in five of the eight studies[25 26 28-30] with four measuring skill performance specific to the training tasks[26 28-30] (Table 1). Overall, skills and measures were varied. Four studies measured walking performance:[25 26 28 30] using the six minute walk test (6MWT)[25 28 30]or the timed up and go, one minute walk test, ten metre walk test and the ABILOCO-Kids questionnaire [25 26 28 30]. Between-group effects of TST on walking performance were mixed (Figures 2a-b). Large effects favouring TST on walking performance at T1 were found (ABILOCO Kids SMD = 0.96, 95% CI = 0.11, 1.81; 6MWT SMD = 0.87, 95% CI 0.02, 1.71) by *Bleyenheuft 2015* (HABIT-ILE vs routine physiotherapy)[25]. However, large negative effects at both T1 (6WMT SMD = -1.47, 95%CI -2.25, -0.69) and at 4 weeks follow up (6WMT SMD = -1.46, 95%CI -2.24, -0.68) were found by *Grecco 2013* (over ground walking vs treadmill training)[28]. Non-significant mixed effects were found at both time points in the remaining two studies[26 30]. Swimming performance was measured using the Water Orientation Test Alyn – 2nd edition in one study[26] and sit to stand performance was measured using the five-times sit to stand test in another[29]. There was no between-group effect found for TST on sit to stand[29] or swimming performance at T1[26]. Within-group effects of TST were positive for all skill performance outcomes in each study, except for the GMFCS III subgroup in one study[29].

Overall gross motor function was measured in five of the eight studies[27 28 30 31 33] (Table 1). Four studies[27 28 30 33] utilised the Gross Motor Function Measure (GMFM)[37]. Three studies measured all domains of the GMFM[27 28 33] (A: lying and rolling, B: sitting, C: crawling and kneeling, D: standing, E walking running and jumping) while one study measured domains D and E only[30]. Nil or negative effect of TST was found at T1 or at T2 (Figures 3a-d)[27 28 30 33] on domains D and E. Three studies demonstrated no effect of TST at T1 or T2 in either domain[27 30 33] while the fourth study *(Grecco 2013)* showed a large negative effect for both domains at T1 (GMFM-D SMD = -1.39, 95%CI -2.16, -0.62; GMFM-E SMD = -1.97, 95%CI -2.82, -1.12) and

T2 (GMFM-D SMD = -1.32, 95%CI -2.09, -0.56; GMFM-E SMD = -2.08, 95%CI -2.95, -1.22)[28]. No significant effects of TST were found (Figures 3e-f)[27 33] on total GMFM score. Again, a large effect favouring the comparison was found for total score at both time points (T1 SMD = -1.83, 95%CI -2.63, -1.03; T2 SMD = -1.67, 95%CI -2.48, -0.87) by *Grecco 2013*[28]. The remaining level III design study reported a significant positive effect of TST on gross motor function as measured by the Test of Gross Motor Development - 2nd edition[31]. Within-group effects of TST on gross motor function were positive in all five studies.

Functional skill outcomes were measured in four of the eight studies[25 27 28 33] (Table 1). Functional skills included; self-care, assessed by the self-care domain of the Pediatric Evaluation of Disability Inventory (PEDI), in three studies[25 28 33]; mobility, measured by the PEDI mobility domain in two studies[28 33] and a parent questionnaire[27]; and social function assessed by the PEDI social function domain in one study[28]. A large effect (SMD 1.07, 95%CI 0.21, 1.94) favouring TST was found for self-care skills at T1 in *Bleyenheuft 2015*[25]. No significant effects were found for the other two studies measuring self-care at T1 and T2[28 33] (Figures 4a-b). No effect of TST on mobility skills (PEDI mobility domain) was found at T1 (Figure 4c) but a moderate effect was found at six months (T2 SMD = 0.58, 95% CI 0.04, 1.12,) and a moderate-large effect found at 12 month follow up (T3 SMD 0.76 95%CI 0.21, 1.31) by *Ketelaar 2007* (Functional therapy vs routine physiotherapy)[33]. In contrast, a large negative effect (SMD -1.32, 95%CI -2.09, -0.56) was found for mobility skills at T1 by *Grecco 2013*[28] (Figure 4c). No effect on mobility on the parent questionnaire was found at T1 or 6 months post[27] or on social function T1 or at 4 weeks follow up[28]. Within-group effects of TST on all functional skills outcomes were positive in all four studies.

Participation-related

Participation-related outcomes were measured in only three of the eight studies[25 26 31] (Table 1). Measures included social participation using the Life-HABITS performance and satisfaction questionnaire[25], adherence and enjoyment[26] and participation in physical activity using an accelerometer[31]. Large effects favouring TST were found for both social participation performance (SMD = 1.19, 95%CI 0.31, 2.07) and satisfaction (SMD = 1.29, 95%CI 0.40, 2.18) by *Bleyenheuft 2015*[25]. Positive effects of TST on weekend physical activity were reported at T1[31]. High adherence rates and enjoyment levels for TST were also reported but without comparison group data [26].

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(ii) Task-specific versus task-specific

Interventions

Three studies compared two or more task-specific interventions including one RCT (level II)[32] and two comparative studies (level III)[34 35]. Two studies[32 34] compared the effect of TST programs with different feedback conditions on a throwing task (Table 1). The throwing task training was informed by motor learning principles but was not goal-directed (Table 3). The intensity of TST in both studies was approximately two hours over two days[32 34]. The other study examined the effects of goal-directed TST compared to activity-based TST[35], with 18 sessions over 12 weeks with parent involvement across various settings[35]. The goal-directed program involved group and individual sessions while the activity program was individual only.

Outcomes

Gross motor skill performance (specific to the trained task), assessed by throwing accuracy, was the sole outcome in both throwing studies[32 34]. This was assessed immediately post intervention (T1 - acquisition) in both studies, 24 hours later in one[32] (T2 - retention) and three days later (T2) in the other[34]. Outcomes were assessed at T1 only in the remaining study: including gross motor function (total GMFM score) and functional skills (all domains of the PEDI). The participation-related outcome, goal attainment, was assessed in the goal-directed group only by the goal attainment scale (GAS) [35].

Knowledge of results provided every trial was found to improve throwing accuracy significantly more than 50% or no feedback at T1. However, at T2, those receiving knowledge of results 50% of the time performed significantly better than those who received feedback every trial or no feedback at all[34]. Self-controlled feedback was found to improve throwing accuracy significantly more than yoked feedback at T2 but not at T1[32]. Goal-directed training improved overall gross motor function and the functional skills of mobility and self-care significantly more than activity-based training[35]. There was no difference in social function between the groups. Eighty-five percent of goals were attained with goal-directed TST[35].

Level IV group design

One repeated measures study involving goal-directed, group-based, intensive TST was included[36] (Table 1, Table 3). Gross motor function (overall GMFM score), functional skills (mobility, self-care and social function using the PEDI) and goal attainment (GAS) were measured at three baseline time points, immediately (T1) and 3 weeks (T2) after the intervention. Positive effects were reported for overall gross motor function at both T1 and T2, and for self-care skills at T2, while 66% of goals were attained at T2.

Single-subject design

One Level I single-subject design study involving randomisation to one of three feedback protocols for learning to move an exercise vehicle backward was included[14] (Table 1). Gross motor skill performance (specific to the trained task) was measured using backwards displacement of the vehicle two days following the training (T1). Eight of the 13 participants demonstrated significant improvement at T1. No specific feedback protocol was clearly superior.

Intervention replicability

Reporting of intervention characteristics varied widely (Table 3). Four studies reported intervention providers were trained in the intervention protocol[25 27 33 36], five studies reported the format of the intervention[14 25 26 35 36] while participant adherence was described in five studies[25 26 30 31 36] . No study reported provider fidelity while comparison interventions were generally ambiguously reported.

Quality of the evidence by outcome

The overall quality of evidence was moderate for gross motor skill performance and functional skills and low for gross motor function and participation-related outcomes.

DISCUSSION

To our knowledge, this is the first systematic review to evaluate and synthesise the evidence for task specific gross motor skills training in ambulant school aged children with CP. Given the focus of TST is on practice of tasks rather than remediating impairments, and the increasing recognition of importance of child and family-centred effects of interventions,

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this review explicitly focused on activity and participation outcomes. In general, positive within-group effects of TST were reported across outcomes of interest. However, in RCTs where TST was compared to comparison interventions, between-group effects were largely non-significant with the exception of two studies[25 28] reporting large but conflicting effects. Overall, there was moderate quality evidence for conflicting effects of TST to improve specific skills performance and functional skills but low quality evidence showing no difference or negative effects on gross motor function. For participation-related outcomes, low quality evidence for positive effects of TST was found. Positive effects across all outcomes were generally found immediately following TST, with evidence of longer-term retention lacking.

The secondary aim of this study was to identify motor learning strategies reported in TST and assess any relationship to outcome. The overall poor reporting of motor learning strategies and heterogeneity in interventions and outcomes limited definitive conclusions. However, this review provides some insight into potentially important characteristics of interventions, in particular; practice dosage, feedback and goals. Unsurprisingly, given its intrinsic relationship to TST, the most consistently reported motor learning strategy was repetitive practice. While reporting of dosage was variable, the largest positive effects of TST were found where dosage was highest[25]. Although no specific feedback condition emerged as clearly superior in the three studies comparing these[14 32 34], results suggest different feedback conditions may influence the phases of training differently. Replication of these studies for varying tasks is required to provide further clarity into the role of feedback. Although causal inferences are limited due to lack of randomisation, better activity outcomes were found when the TST was goal-directed[35]. These results are consistent with the growing evidence base for interventions targeted towards the goals of children with CP and their families[6 16].

Our review found some conflicting results between studies, with large positive and negative effects found in *Bleyenheuft 2015*[25] and *Grecco 2013*[28] respectively. The TST in *Bleyenheuft 2015*[25] was high in dose, explicitly driven by motor learning principles and involved of a wide range of gross motor tasks including ball skills, cycling and walking. By contrast, in *Grecco 2013*[28] dosage of TST (over-ground walking) was low in comparison and limited motor learning strategies were reported. The comparison treatment in this study was treadmill training without body weight support – training that may be regarded as task-related but was not deemed TST given the primary goal of the study was to improve over-ground walking. Whilst practice dose was equal between the groups in *Grecco 2013*[28], task

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progression was potentially greater with the use of treadmill functions. These results suggest task-related adjuncts, such as treadmill training, may be more beneficial than pure TST for developing (rather than acquiring) gross motor skills, such as walking in this already-ambulant population.

Limitations across the included studies

Low-moderate quality of evidence was found across the outcomes due to significant methodological limitations in the studies, in addition to risk of bias. First, sample size calculation was not reported in most studies (Table 1). Wide confidence intervals found for outcome data from Level II-III studies (Figures 2-4) suggest inadequately powered samples may have reduced precision and thus limit the generalizability of the findings. Second, the limited detail in reporting potentially reduces the robustness of research findings if the study cannot be replicated and limits implementation. Third, limitations in outcome measurement selection may have influenced the generalisability and transferability of some findings. The GMFM and the PEDI are a well-established tools for evaluating change in children with CP[38], and were used in six[27 28 30 33 35 36] and four studies[25 28 33 35] respectively. However, concerns have been raised about the responsiveness of these measures in higher functioning children[38], the target population of this review. Whilst common use makes them appealing, their broad focus means skills targeted by the TST may not have been adequately captured. Finally, far fewer participation-related outcome measures were used compared to activity outcomes thus any evidence that improved activity through TST leads to improved participation is weak at best.

Limitations of this review

This review chose to narrow the inclusion criteria to ambulant school aged children with CP, interventions of gross motor skill TST, and activity and participation domains outcomes, in order to draw specific conclusions. This was not possible for all questions posed by this review due to study design issues and heterogeneity, which also precluded meta-analysis. Further, the methodology of the review itself has some limitations. The impact of publication bias was not evaluated. Inclusion was limited to published articles in English meaning some studies, including grey literature, may have been missed. Further, non-randomised studies were retained in this review. Although the Cochrane Risk of Bias tool and the GRADE system are the most widely used systems for assessing risk of bias and the

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quality of the evidence across outcomes respectively[20 24], these tools do emphasise randomised studies.

Conclusions and recommendations

Despite the largely positive within-group effects of TST over time across each outcome of interest, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes while no difference or negative effects were found for gross motor function. Given the low-moderate quality of this evidence, there is currently limited evidence to support task-specific gross motor skills training for improving these activity and participation-related outcomes in children with CP. Clear recommendations around whether TST is superior to other interventions cannot be made. Before conclusions can be made about any relationship of motor learning strategies to outcome, more consistent reporting and studies designed to test this are required.

The importance of tailoring motor interventions to individual goals and lifelong physical activity is increasingly being recognised[4 39]. Thus, to enable specific recommendations, strengthening the evidence base is imperative. Adequately powered samples, rigorous study design and consistent reporting with attention to reporting interventions to allow for reproducibility and appropriate evidence synthesis is required. Future challenges also include considering issues with outcome measure responsiveness and intervention heterogeneity, and optimising TST through the use of motor learning strategies.

Competing interests

The authors have no competing interests to declare.

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Data sharing statement

Data relating to the quantitative synthesis is available on request.

Author contributions

All named authors contributed to the design of this review. RT collected data, carried out data analyses and led interpretation and reporting. CB collected data and contributed to interpretation and reporting. ARH, JLM and AJS contributed to data analysis, interpretation and reporting. All named authors approved the final manuscript as submitted.

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Figure legend

Figure 1: PRISMA flow chart of included and excluded studies

Figure 2a: Gross motor skill performance - Task specific vs alternative: walking performance at T1 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 2b: Gross motor skill performance - Task specific vs alternative: walking performance at T2 (NB Grecco 2013 TUG scores not included) *= outcome is specific to task being trained

Figure 3a: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T1.

Figure 3b: Gross motor function - Task specific vs alternative: Standing ability (GMFM domain D) at T2.

Figure 3c: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T1.

Figure 3d: Gross motor function - Task specific vs alternative: Walking, running and jumping ability (GMFM Domain E) at T2.

Figure 3e: Gross motor function - Task specific vs alternative: Overall GMFM score at T1.

Figure 3f: Gross motor function - Task specific vs alternative: Overall GMFM score at T2.

Figure 4a: Functional skills - Task specific vs alternative: Self-care at T1.

Figure 4b: Functional skills - Task specific vs alternative: Self-care at T2.

Figure 4c: Functional skills - Task specific vs alternative: Mobility skills at T1

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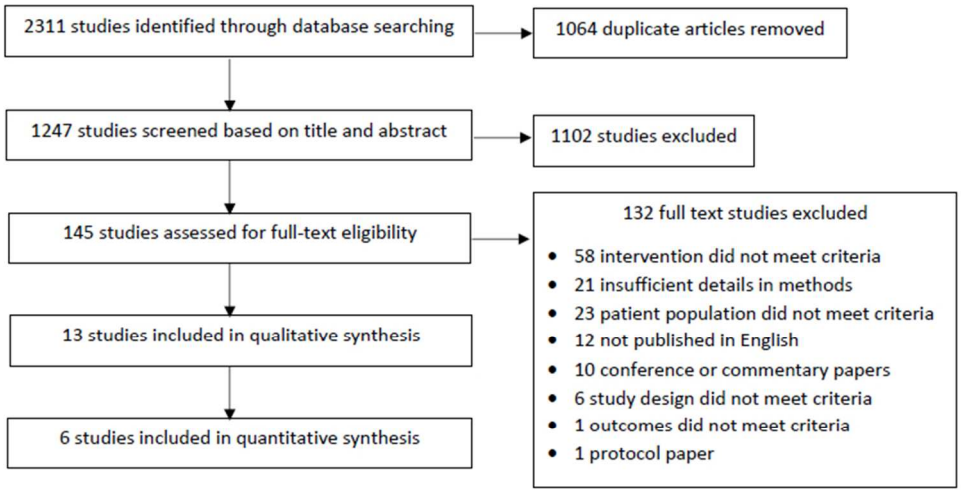

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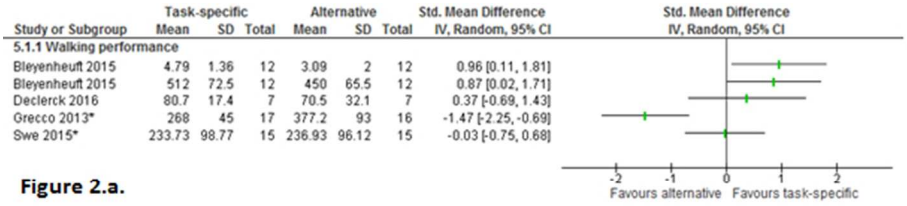

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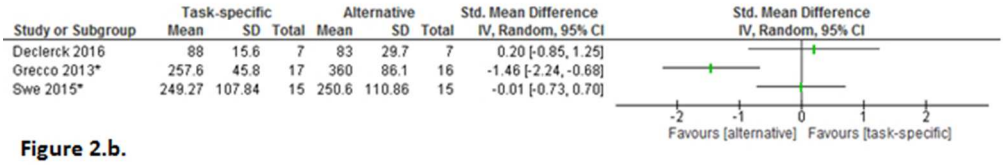

Figure 2.b.

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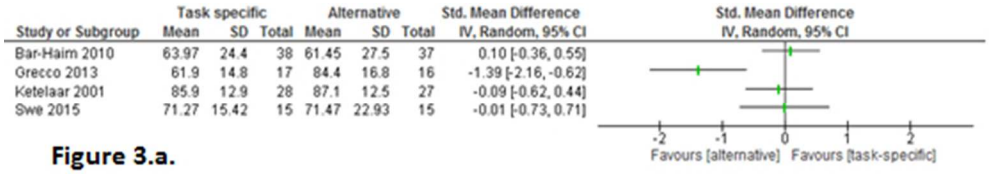

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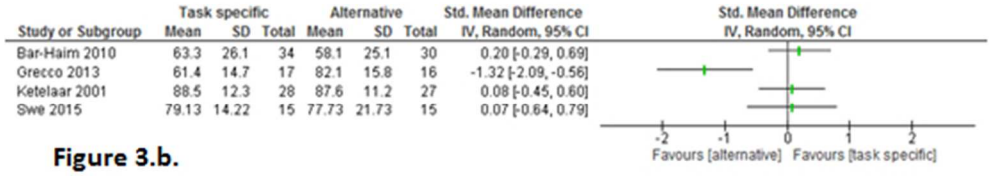

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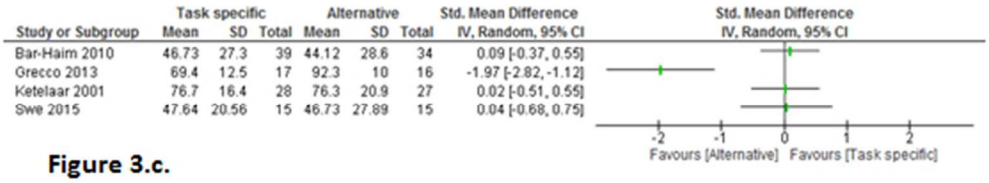

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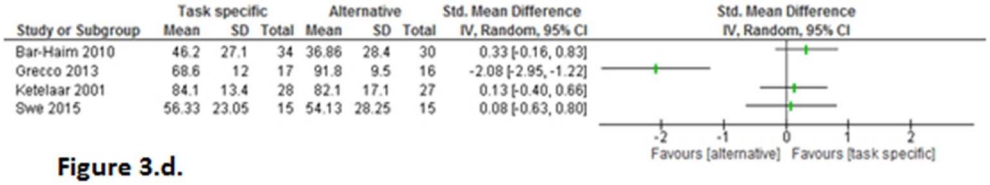

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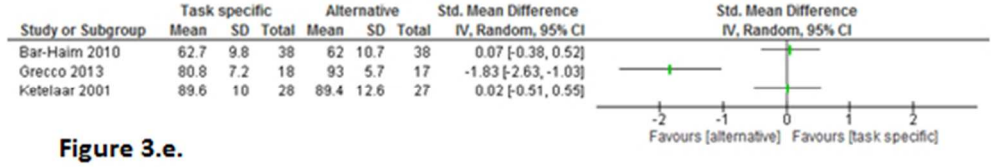

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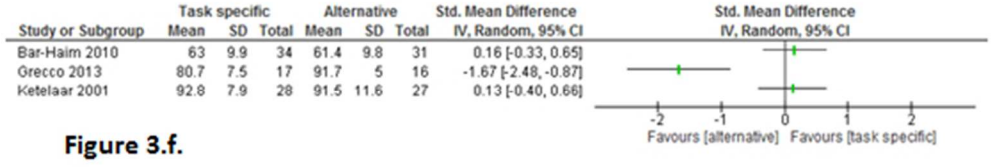

Figure 3.f.

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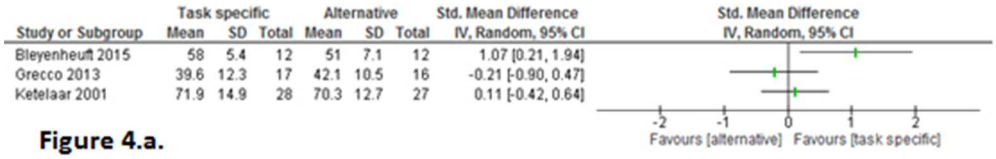

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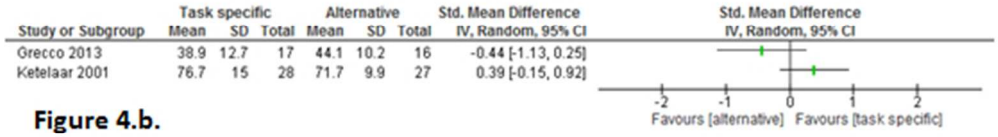

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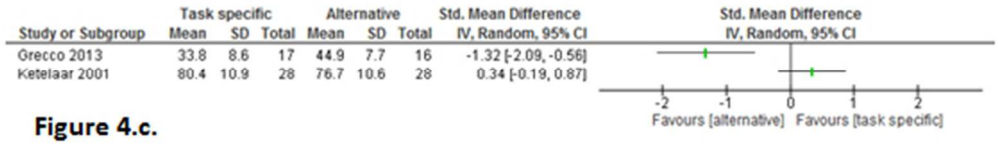

Figure 4.c.

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APPENDIX 1

Search strategy for Ovid databases

1. cerebral palsy.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
2. child*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. (Task specific or skill specific or task oriented or activity focussed or task practice or goal directed).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
4. (motor imagery or implicit or error reduced or neuromotor task or parent assisted or group or cognitive orientation or repetit*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. (perceptual motor or kinaesthetic explicit or action observation or dual task or trial and error or practice or imitation or experiential or discrimination or discovery or errorless or analogy or observational or exploratory).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
6. (Training or intervention or therapy or approach or learning or program).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. (Motor skill* or motor function* or movement skill* or Motor performance or task performance or occupational performance or motor competence or task competence or occupational competence or skill acquisition or skill attainment or skill proficiency or skill achievement or goal attainment).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
8. 3 or 4 or 5
9. 1 and 2 and 6 and 7 and 8

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