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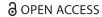
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Defining Functional Therapy in Research Involving Children with Cerebral Palsy: A Systematic Review

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ABSTRACT

Aim: To review definitions and elements of interventions in studies, which used the word "functional" to describe their intervention for children with cerebral palsy (CP), and to determine whether definitions and elements are similar to criteria of functional therapy described in the Dutch Guidelines.

Methods: Systematic review of intervention studies, which used the word "functional" to describe interventions for children with CP. We described criteria of functional therapy that were used to describe the intervention, and whether criteria were described similarly to the descriptions used in the Dutch Guidelines.

Results: Of the 27 included studies, criteria "based on the activities/participation level of the ICF-CY", "goal-directed" and "context-specific" were referred to the most (40–59.3%). Descriptions of these criteria were less comparable to the suggested definition (43.8–69.2%). The remaining three criteria ("active involvement", "task-specific", and "focused on functionality instead of normality") were referred to less frequently (18.5–33.3%). The descriptions reported for these criteria were, however, the most comparable with the suggested definitions (80–100%).

Conclusions: The included studies, in general have not used criteria of functional therapy. Future studies have to describe the elements of interventions in detail. Moreover, it is important to reach consensus on the definition and elements of functional therapy.

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KEYWORDS

Active involvement; activity; cerebral palsy; contextspecific; functional therapy; functionality; goal-directed; ICF-CY; participation; task-specific

Introduction

There is an increasing number of studies of children with cerebral palsy (CP) that refer to their intervention as functional therapy. However, there is no standard description of

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what constitutes functional therapy. Ketelaar, Vermeer, Hart, van Petegem-van Beek, and Helders (2001) described functional therapy as: "Functional physical therapy emphasizes the learning of motor abilities that are meaningful in the child's environment and perceived as problematic by either the child or parents. Children practice these motor abilities in functional situations with the child having an active role in finding solutions for motor problems rather than having the physical therapist's handling result in a solution. Functional goals, in terms of skills, are established with parents and children based on their priorities. Functional activities are assumed to be learned by repetitive practice of goal-related tasks in functional situations" (p. 1536) (Ketelaar et al., Ketelaar et al., 2001). Another study by Ahl, Johansson, Granat, and Carlberg, (2005) described functional therapy in a similar manner: "Physical therapy in this context emphasizes the interaction between the individual, the task, and the environment as a basis for the child to learn; children actively seek effective solutions to problems that arise in situations they desire to master. The approach is called 'functional' as primarily functional skills (e.g. dressing and climbing stairs) are practiced to achieve functional goals. Defining specific goals can enhance motivation and, possibly, the child's capacity to learn. In this context, the child can become an active problem solver instead of a passive recipient of treatment" (p. 613) (Ahl et al., 2005).

Over the last decade, there has been a substantial increase in therapy interventions addressing motor abilities of children with CP (Novak et al., 2013). Some of these studies refer to the intervention as functional therapy, but an elaborated description of the intervention is frequently missing (Sakzewski, Reedman, & Hoffmann, 2016). It can be questioned whether studies which refer to their intervention as functional therapy indeed examined interventions that align with the definition of functional therapy as described by Ketelaar et al. (2001) and Ahl et al. (2005). Vice versa, there may be other interventions that align with the definition of functional therapy, yet do not use this terminology to label their intervention.

Based on the lack of consensus on the definition and criteria for functional therapy, experts in the field of CP introduced six criteria to guide the implementation of functional therapy for children with CP. The group of experts consisted of members of the CP guideline steering group. Within this group of experts a total of five members were part of the consensus group, consisting of two experienced pediatric physical therapist, a pediatric occupational therapist, pediatric rehabilitation physician, and human movement scientist. The six criteria were selected during several expert meetings. First, all possible names and definitions of functional therapy were identified based on a comprehensive review of the literature and clinical experience, and discussed. The second step was to synthesize all names and definitions into a set of criteria defining functional therapy. After reaching consensus the criteria were then discussed with the other members of the steering group, which approved the following six criteria of functional therapy: (1) goal-directed, (2) based on the activities/participation level of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY), (3) task-specific, (4) active involvement of the child and parents to find solutions, learn motor skills and discover new possibilities, (5) focused on functionality rather than normality and (6) context specific (Revalidatieartsen, 2015). See Table 1 for a more elaborated description of the criteria.



Table 1. Criteria of functional therapy of the Dutch guidelines (Revalidatieartsen, 2015).

1. Goal-directed

The goals have been formulated in consultation with parents and/or child. The goals are specific, measurable, achievable, relevant and time-bound (SMART) and should preferably be determined using a valid and reliable measurement instrument, such as the Goal Attainment Scale (GAS)a or the Canadian Occupational Performance Measures (COPM)b

2. Based on the activities/participation level of the ICF-CYc

The goals of the therapy should always be formulated at the ICF-CY level of activities and/or participation 3. Task-specific

The intervention is based on a task analysis and focuses on practicing the activities (or partial activities), which are formulated in the goals

- 4. Active involvement of the child and parents to find solutions, learn motor skills and discover new possibilities During therapy, the child is actively trying to find solutions regarding motor activities. The approach is basically hands-off
- 5. Focused on functionality rather than normality

The focus of the treatment and the most important goal of the intervention is functionality and not normality. It is important that the child is successful in performing the motor tasks, and the quality of the performance is not the main focus

6. Context-specific

Therapy takes into account contextual factors that specifically concern the formulated goals, and if possible takes place in the context that is relevant to the child.

The aim of this systematic review was to review definitions and elements of interventions in studies, which used the word "functional" to describe their intervention for children with CP (aged <18 years), and to determine whether definitions and elements are similar to the criteria of functional therapy described in the Dutch Guidelines. The findings will provide evidence as to whether or not the construct functional therapy is generalizable across studies that specifically stated they used functional therapy and to provide recommendations for standardized criteria for clinical practice and research.

Methods

This systematic review was conducted in accordance with the PRISMA guidelines, A completed PRISMA checklist can be found in the Online Supplementary Appendix A.

Inclusion/Exclusion Criteria

This review included all intervention studies, which used the word "functional" to describe interventions for children with CP; only therapies that focused on oral function were excluded. Studies included randomized controlled trials (RCTs), clinical trials (CTs), two group comparison, non-randomized CTs, pre-post study design, CTs with only one data point and studies with a single subject design. Participants were children and/or adolescents with any type of CP. Studies which comprised both children with CP and children with other disorders were included if at least 50% of the study sample had a diagnosis of CP.

^aTurner-Stokes (2009).

^bLaw et al. (1990).

^cWHO (2007).

Literature Searches

Literature searches were performed using search strategies developed by one reviewer (MG). A total of five electronic databases were searched: EMBASE, MEDLINE, PubMed, CINAHL, and PEDro. The first search was performed on March 2016 with an update in January 2018. The search strategy was adapted according to the configuration and requirements of each database. The searches were developed using keywords (e.g. MeSH terms) and text words, combining population, and intervention related terms. The population related keyword was "cerebral palsy". Intervention related search terms were "functional" in combination with "exercise", "intervention", "practice", "program", "therapy", "training", "treatment", "rehabilitation", or their plurals. Additional intervention related search terms were "task-oriented", "task-specific", and "goal-directed". Literature searches were limited to articles published between 2000 and 2018, the English language and human subjects.

Identified references were downloaded in Endnote X7 software (Thomson Reuters, New York) for further assessment and handling, and duplicate records were removed.

Study Selection and Data Extraction

The first phase of the selection process was screening titles and abstracts on eligibility criteria to remove irrelevant reports. If there was any doubt regarding relevance, the report remained included at this stage. Subsequently, full texts were retrieved for all potentially relevant reports. Full-text reports were selected based on the inclusion and exclusion criteria. Multiple reports of the same study were identified and only the primary outcome paper of a study was included. Any available protocol papers of the included studies were retrieved for the data extraction.

The first phase of the selection process (i.e. screening of titles and abstracts) was performed by three reviewers (ER, MG, and MK), where each abstract was screened by two reviewers independently.

The second phase of the selection process (i.e. examination of the full-text reports) was performed independently by the same reviewers (ER, MG, and MK) in the same way as the first phase. In the case of discrepancies in any phase of the selection process, the third independent reviewer mediated to reach consensus.

After the selection process, data extraction was performed. Of the included studies, the following data were extracted: demographic information (age and type CP), methodology (study design, number of participants), whether the criteria of functional therapy ("goal-directed", "based on activities/participation level of the ICF-CY", "task-specific", "active involvement", "focused on functionality rather than normality", and "context-specific") were used to describe the intervention. We checked whether criteria were described similarly to the descriptions used in the Dutch Guidelines. In addition, we checked whether the criteria were mentioned in the method section. Mentioning the criteria in the method section is an indicator that it was part of the intervention.

Agreement

Agreement between the reviewers was determined by calculating Cohen's Kappa (κ). Two independent reviewers checked if the authors used "functional" to describe their

intervention. There was a high agreement ($\kappa = 0.91$) for "functional (therapy)" between the two reviewers. A third reviewer was consulted to reach consensus.

There were moderate to high levels of agreement between reviewers when determining whether criteria were mentioned in a study (goal-directed, $\kappa = 0.92$; based on activities/participation level of the ICF-CY $\kappa = 0.70$; task-specific $\kappa = 0.78$; active involvement $\kappa = 0.74$; focused on functionality rather than normality $\kappa = 0.63$; context specific $\kappa = 0.78$). When determining whether the definition of the criteria aligned with those used in this review, agreement between reviewers was moderate to good (goaldirected $\kappa = 0.64$; based on activities/participation level of the ICF-CY $\kappa = 0.68$; taskspecific $\kappa = 0.89$; active involvement $\kappa = 0.82$; focused on functionality rather than normality $\kappa = 1.00$; context specific $\kappa = 0.91$). Consensus was reached for all criteria and definitions by consulting a third independent reviewer.

Results

Study Selection

Literature searches of electronic databases retrieved 3158 titles/abstracts. After de-duplication, a total of 2652 titles/abstracts were screened, and 2592 articles were excluded because there was no therapeutic intervention. Full-text reports of 60 potentially relevant studies were selected for further examination. Of these, 33 studies were excluded after further examination for the following reasons: an adult population (1 article) or the authors did not use the word "functional" to describe their intervention (32 articles). A total of 27 studies met the criteria for inclusion in the review. A flow diagram of the identification and selection of studies for inclusion in this review is presented in Figure 1.

Study Characteristics

All characteristics of the included studies are shown in Table 2. Several different types of intervention approaches were referred to by authors as functional therapy. For example home-based or modified constraint-induced movement therapy (CIMT) involving functional training of the involved hand (Chen et al., 2013; de Brito Brandao, Mancini, Vaz, Pereira de Melo, & Fonseca, 2010; Gordon et al., 2011; Chen et al., 2014; 2016; Xu, He, Mai, Yan, & Chen, 2015), goal-directed functional therapy (GDT) (Ahl et al., 2005; Lowing, Bexelius, & Brogren Carlberg, 2009; Lowing, Bexelius, & Carlberg, 2010; Storvold & Jahnsen, 2010), and functional (physical) training program (Akbari, Javad Zadeh, Shahraki, & Jahanshahi Javaran, 2009; Blundell, Shepherd, Dean, Adams, & Cahill, 2003; Ferre et al., 2017; Gorter, Holty, Rameckers, Elvers, & Oostendorp, 2009; Ketelaar et al., 2001; Kusumoto, Nitta, & Takaki, 2016; Lee, Ko, Shin, & Lee, 2015; Liao, Liu, Liu, & Lin, 2007; Moura et al., 2017; Peungsuwan, Parasin, Siritaratiwat, Prasertnu, & Yamauchi, 2017; Rameckers, Duysens, Speth, Vles, & Smits-Engelsman, 2010; Rameckers, Speth, Duysens, Vles, & Smits-Engelsman, 2009; Scholtes et al., 2010; van Vulpen, de Groot, Rameckers, Becher, & Dallmeijer, 2017).

The age of participants included in the studies ranged from 1 to 18 years, the majority were school-aged children. According to the Surveillance of Cerebral Palsy in Europe

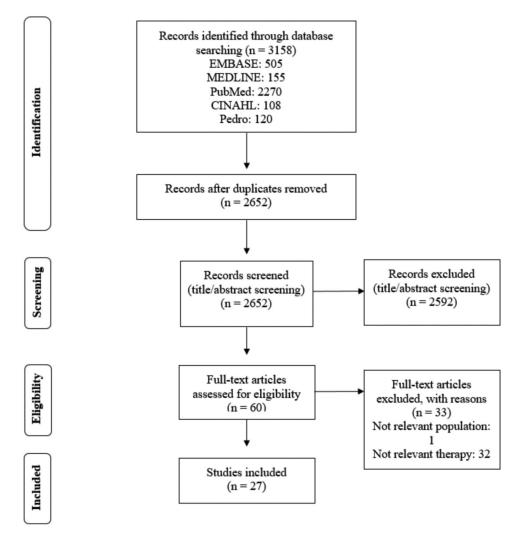


Figure 1. Flow diagram of the study identification and selection.

(SCPE) terminology, eleven studies included only children with unilateral CP (Chen et al., 2013; de Brito Brandao et al., 2010; Ferre et al., 2017; Gordon et al., 2011; Chen et al., 2014; 2016; Moura et al., 2017; Rameckers et al., 2009; 2010; Sung et al., 2005; Xu et al., 2015). Eight studies included both unilateral and bilateral CP (Akbari et al., 2009; Ketelaar et al., 2001; Lowing et al., 2009; 2010; Peungsuwan et al., 2017; Scholtes et al., 2010; Storvold & Jahnsen, 2010; van Vulpen et al., 2017). Five studies included children with bilateral CP (Ahl et al., 2005; Blundell et al., 2003; Kusumoto et al., 2016; Liao et al., 2007; Lin et al., 2015). Three studies did not specify the subtype of CP (Gorter et al., 2009; Lammi & Law, 2003; Lee et al., 2015).

Criteria and Definition of Criteria

All 27 studies referred to functional therapy, mostly in the methods section (23 studies; 85.2%). Of these 27 studies only one study referred to all six criteria, whereas four

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Study	Design	Diagnosis	Age	z	Intervention	Control
Xu et al. (2015)	Single-blinded RCT	Unilateral CP	2–14 years 6	0 89	CIMT (involved intensive functional training of the involved hand) $(n = 22)$ or CIMT plus electrical stimulation $(n = 23)$	Traditional occupational therapy (involved functional unimanual and bimanual training) ($n = 23$)
Chen et al. (2014)	RCT	Unilateral CP	6–12 years 4	45 F	Home-based constraint-induced therapy (involved intensive functional training of the more affected unner extremity) $(n=23)$	Traditional rehabilitation (involved functional unimanual and bimanual trainin) (n=22)
Chen et al. (2013)	Single-blinded RCT	Unilateral CP	6–12 years 4	0 44 0	Constraint-induced therapy (involved intensive functional training of the more affected arm) ($n = 24$)	Traditional rehabilitation (involved functional unilateral and bilateral arm training) (n = 23)
Storvold and Jahnsen (2010)	Multiple single-subject (ABA) design	Unilateral CP $(n = 4)$, bilateral CP $(n = 1)$ and quadrilateral CP $(n = 1)$	3–11 years	9	Intensive goal-directed functional training combining group and individual sessions	
de Brito Brandao et al. (2010)	Single-blinded RCT	Unilateral CP	4–8 years 8 months	J6 C	One week of bimanual functional training following constraint therapy $(n=8)$	Usual rehabilitation ($n=8$)
Rameckers et al. (2010)	Randomized design	Spastic unilateral CP	4–16 years 2	50 S	Standardized functional training program with vs. without the addition of chemodernervation of the muscles of the forearm and hand	
Lowing et al. (2009)	Prospective intervention study	Uni- $(n=17)$ or bilateral $(n=27)$ CP	Mean age 4 years 1 months	4	Goal-directed functional therapy (GDT) versus activity focused therapy (AT)	
Gorter et al. (2009)	Test-retest repeated measures design	C _D		13 F	Functional physical training program	
Rameckers et al. (2008)	RCT	Spastic unilateral CP	4–16 years 2	20 S	Standardized functional training program versus without the addition of chemodenervation of forearm and hand muscles	
Ahl et al. (2005)		Spastic bi- $(n=12)$ or trilateral $(n=2)$ CP	1 years 6 months- 6 years	14	Goal-directed functional therapy	
Lowing et al. (2010)	Prospective longitudinal intervention study	Uni- $(n=8)$ or bilateral $(n=14)$ CP		22 G	Goal-directed functional therapy	
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Study	Design	Diagnosis	Age	z	Intervention	Control
Akbari et al. (2009)	Pre- and post design study	Uni- $(n=7)$, bi- $(n=6)$ and quadrilateral (n=2) CP	Mean age 21.87 months	15	Functional physical therapy program	
Lammi and Law (2003)	Single subject, multiple baseline design	, do	3–3 years 6 months	m	Family-Centred Functional Therapy (FCFT)	
Ketelaar et al. (2001)	Randomized block design	Uni- $(n = 32)$, bi- $(n = 11)$ or quadrilateral $(n = 12)$ CP		55	Functional physical therapy	
Scholtes et al. (2010)	Single-blinded RCT	Spastic uni- $(n = 17)$ or bilateral $(n = 32)$ CP	mean age 10 years 5 months	49	Functional progressive resistance exercise $(n=24)$	Usual care $(n=25)$
Blundell et al. (2003)	Nonrandomized ABA trial	Spastic bilateral CP $(n=7)$ or spastic/ ataxic quadrilateral CP $(n=1)$	4-8 years	∞	Functional strength training	
Lee et al. (2015)	Single-blinded RCT	Spastic CP	5–10 years	56	General NDT and additional progressive functional training	General NDT
Gordon et al. (2011)	Randomized trial	Unilateral CP	3.5–10 years	45	90 h of CIMT ($n = 21$) versus an equivalent dosage of functional bimanual training (HABIT) ($n = 21$)	
Liao et al. (2007)	Single-blind, randomized block design	Spastic bilateral CP	5–12 years	20	Regular physical therapy plus functional strengthening program, the loaded sit-to-stand (STS) resistance exercise $(n=10)$	Regular physical therapy $(n=10)$
Sung et al. (2005)	Prospective case series	Unilateral CP	Mean age intervention group 33.2 months, mean age control group 43.2 months	31	Short-arm Scotchcast On the unaffected arm for 6 weeks plus Conventional rehabilitation program that included stretching Exercises and functional occupational therapy for the upper extremity (n = 18)	Conventional rehabilitation Program ($n=13$)
Chen et al. (2016)		Spastic unilateral CP	Younger group: $6-8$ years $(n = 11)$ and older group: $9-12$ years $(n = 12)$	23	Home-based constraint-induced therapy (CIT) (involved intensive functional training of the more affected upper-limb)	
Lin et al. (2015)		Spastic bilateral CP		12	Botulinum toxin type A (BoNT-A) combined with functional occupational therapy (OT)	

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		Functional training of the paretic upper limb combined with sham transcranial stimulation $(n=10)$	Physical therapy $(n=7)$	
24 Home-based HABIT ($n=12$) versus lower-limb functional intensive training (LIFT) ($n=12$)	Regular physical therapy (involved functional training) with slow loaded sit-to-stand exercise group (n = 8) or self-paced loaded sit-tostand exercise group (n = 8)	Functional training of the paretic I upper limb combined with anodic transcranial stimulation $(n=10)$	15 Functional exercise program and physical therapy $(n=8)$	22 Functional power training
24	16	50	15	22
2 years 6 months- 10 years 1 months	12–18 years	6–12 years	7–16 years	4–10 years
Spastic unilateral CP	Spastic bilateral CP	Spastic unilateral CP	Spastic uni- or bilateral CP	Spastic uni- or bilateral CP
RCT	Single-blind randomized clinical trial	Randomized sham- controlled study	RCT	Study protocol of double baseline trial
Ferre et al. (2016)	Kusumoto et al. (2016)	Moura et al. (2017)	Peungsuwan et al. (2017) RCT	van Vulpen et al. (2017) Study protocol of baseline trial

CIMT: constraint induced movement therapy; HABIT: hand-arm bimanual intensive therapy; NDT: neurodevelopmental treatment; RCT: randomized controlled trial.

Table 3. Criteria of functional therapy mentioned in the included papers, and in which section of the paper.

<u> </u>							
			Based on activities/ participation			Focused on functionality	
C4d	Functional	Goal-	level of the ICF-CY	Task-	Active	rather than	Context
Study	therapy	directed	ICF-CY	specific	involvement	normality	specific
Xu et al. (2015)	M	_	M	_		M	M
Chen et al. (2014)	M	_	M	_	M	_	M
Chen et al. (2013)	M	_	M	_	M	_	M
Storvold and Jahnsen (2010)	M	M	M	M	M	M	M
de Brito Brandao et al. (2010)	M	_	M	_	_	_	M
Rameckers et al. (2010)	A + D	M	M	M	_	_	_
Lowing et al. (2009)	M	M	M	_	M	M	ı
Gorter et al. (2009)	T + A + I	_	_	_	_	_	_
Rameckers et al. (2008)	M	M	_	M	_	_	_
Ahl et al. (2005)	M	M	M	M	M	_	M
Lowing et al. (2010)	T + A + I + M + D	M	M	M	M	_	M
Akbari et al. (2009)	M	M	M	M	M	M	_
Lammi and Law (2003)	M	M	1	1	I	_	1
Ketelaar et al. (2001)	M	M	_	M	M	M	M
Scholtes et al. (2010)	M	D	M	_		_	D
Blundell et al. (2003)	M	I + D	_	A + D		_	D
Lee et al. (2015)	M	_	_	_		_	_
Gordon et al. (2011)	Α	M	M	I + D	M	_	M
Liao et al. (2007)	M	_	_	I + D		_	_
Sung et al. (2005)	M	M	M	_		_	_
Chen et al. (2016)	M	_	_	_		_	M
Lin et al. (2015)	M	D	M	D		_	_
Ferre et al. (2016)	A + I + M	A + M + D	M	M		_	T + I + M
Kusumoto et al. (2016)	I + M	_	_	1		_	_
Moura et al. (2017)	A + I	_	_	_	_	_	_
Peungsuwan et al. (2017)	I + M	_	_	_	_	_	_
van Vulpen et al. (2017)	T + A + I + M	М	М		_		_

A: mentioned in the abstract; D: mentioned in the discussion; I: mentioned in the introduction; M: mentioned in the methods; T: mentioned in the title; —: not mentioned at all.

studies referred to five criteria. Three studies referred to four criteria, four studies referred to three criteria, and another four studies to two criteria. Four studies referred to only one criterion and seven studies did not refer to any criterion. Figure 2 lists the criteria and definitions mentioned in the method section of each study. A more detailed overview of the criteria of functional therapy and definitions mentioned in the included studies, and in which section of the paper the criteria were mentioned can be found in Tables 3 and 4.

Of the six proposed criteria, "based on the activities/participation level of the ICF-CY" was referred to the most in 16 (59.3%) of the studies followed by the criterion "goal-directed" in 13 (48.1%) and "context-specific" in 11 (40.7%). The descriptions of these criteria, however, were only comparable to the suggested definition of the Dutch guidelines in 7 (43.8%), 9 (69.2%), and 7 (63.6%) of the studies respectively. The remaining three criteria were referred to less frequently. The criterion "active involvement" was described in 9 (33.3%) studies, "task-specific" in 8 (29.6%) and "focused on functionality instead of normality" in 5 (18.5%). The descriptions reported for these three criteria were, however, the most comparable with the suggested definitions of the Dutch guidelines (80–100%). The descriptions for all criteria were reported in the methods section of each study. No other definitions for the criteria were found in the studies.

Table 4. Comparison of the proposed definitions with the definitions of the criteria that were mentioned in the methods section of the included papers.

	Goal-	Based on the activities/ participation level of the	Task-	Active	Focused on functionality rather than	Context
Study	directed	ICF-CY	specific	involvement	normality	specific
Xu et al. (2015)	_	No	_	_	No	No
Chen et al. (2014)	_	No	_	Yes	_	Yes
Chen et al. (2013)	_	No	_	Yes	_	Yes
Storvold and Jahnsen (2010)	Yes	Yes	Yes	Yes	Yes	Yes
de Brito Brandão et al. (2010)	_	No	_	_	_	No
Rameckers et al. (2010)	Yes	No	Yes	_	_	_
Lowing et al. (2009)	Yes	Yes	_	Yes	Yes	-
Gorter et al. (2009)	_	_	_	_	_	_
Rameckers et al. (2008)	No	_	Yes	_	_	_
Ahl et al. (2005)	Yes	Yes	Yes	Yes	_	Yes
Lowing et al. (2010)	Yes	Yes	Yes	Yes	_	Yes
Akbari et al. (2009)	Yes	Yes	Yes	Yes	Yes	_
Lammi and Law (2003)	Yes	_	_	_	_	_
Ketelaar et al. (2001)	No	_	Yes	Yes	Yes	Yes
Scholtes et al. (2010)	_	No	_	_	_	_
Blundell et al. (2003)	_	_	_	_	_	_
Lee et al. (2015)	_	_	_	_	_	_
Gordon et al. (2011)	Yes	Yes	_	Yes	_	Yes
Liao et al. (2007)	_	_	_	_	_	_
Sung et al. (2005)	No	No	_	_	_	_
Chen et al. (2016)	_	_	_	_	_	No
Lin et al. (2015)	_	No	_	_	_	_
Ferre et al. (2016)	No	No	No	_	_	No
Kusumoto et al. (2016)	_	_	_	_	_	_
Moura et al. (2017)	_	_	_	_	_	_
Peungsuwan et al. (2017)	_	_	_	_	_	_
van Vulpen et al. (2017)	Yes	Yes	_	_	_	_

Yes: description in line with suggested definition in this paper; No: description not in line with suggested definition in this paper; —: criteria were not mentioned in the method section.

Discussion

The aim of this systematic review was to review definitions and elements of interventions in studies which used the word "functional" to describe their intervention for children with CP, and to determine whether definitions and elements are similar to the criteria of functional therapy described in the Dutch Guidelines. A clear description of treatment methodology or taxonomy for rehabilitation is crucial to compare studies and to unravel the black box, called intervention (Craig et al., 2008; Dijkers, Hart, Tsaousides, Whyte, & Zanca, 2014). Our comprehensive literature search identified a range of different interventions targeting both upper and lower extremity function, all of which were described as functional therapy. Findings of this review demonstrate inconsistent descriptions and definitions of the multiple elements of functional therapy, with three or more of six criteria reported in less than 50% of the studies.

While all studies described their intervention as functional therapy, lack of a clear description of the essential elements made it difficult to match against our proposed six criteria. Firstly, it is shown that the total set of all six criteria to describe functional therapy was not or rarely used. Only one of the 27 studies referred to all six criteria in their article, whereas seven studies referred to none of the criteria. In most studies, there is a description of the intervention, but the intervention is not described in terms

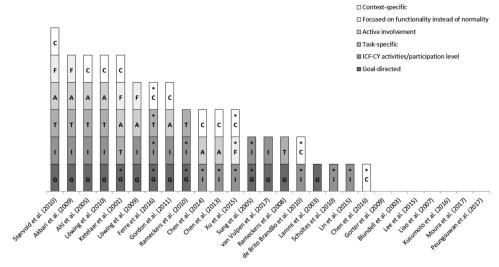


Figure 2. Criteria and definition mentioned in the method section of each study. When the definition was not in line with the proposed definition an * was placed in the box.

of the proposed criteria. Secondly, the referred criteria were used to a varying extent. The criteria "based on activities/participation level of the ICF-CY" and "goal-directed" were referred to most frequently while the criterion "focused on functionality rather than normality" was referred to only in five studies. The criteria "based on activities/participation level of the ICF-CY" and "goal-directed" are already well known and often used in reviews and papers to describe interventions for children with CP. The criterion "focused on functionality rather than normality" is a very specific element and therefore may not always be explicitly mentioned. The agreement between the reviewers for this criterion was moderate, so even if it is mentioned it can be interpreted differently. The criteria "taskspecific", "active involvement", and "context-specific" are based on motor learning principles and therefore not explicitly mentioned separately (Shumway-Cook & Woollacott, 2007). Thirdly, when the criteria were mentioned, a definition was not always provided. Eight studies referring to one or more criteria did provide definitions for all mentioned criteria, whereas seven studies naming one or more of the criteria did not provide any definition. When criteria were defined, these definitions were in extent in line with the definitions of the Dutch guidelines used in this review, only the definitions of the criterion "based on activities/participation level of the ICF-CY" showed minimal agreement with the definition used in this review. The definition used in this review links the ICF model to activity-based intervention goals, which is often not the case in the definitions mentioned in the studies. Also, the agreement between the reviewers for this criterion was moderate, so despite the fact that this criterion is mentioned most frequently, the definition may be too broad. Overall, it is obvious that the criteria of functional therapy and their definitions are not yet well incorporated in research about functional therapy.

The lack of reporting on the elements and definitions of functional therapy does not necessarily mean that the studies did not use all these components. Hoffmann, Erueti, and Glasziou (2013) investigated the completeness of descriptions of non-pharmacological interventions in randomized trials. The results showed that 39% of

the interventions were adequately described, but after contact with the authors to get more information, it increased to 59% (Hoffmann et al., 2013). We may have been able to get additional information if we had approached the authors directly. Possibly a percentage of the excluded studies might have included functional interventions, without describing the therapy as functional, for example the studies of Cameron et al. (2016), Kruijsen-Terpstra et al. (2016)and Law et al. (2011). Also, several excluded studies did indeed describe elements of functional therapy but were not included on the sole basis that they did not use "functional" to describe their interventions. However, this review was not intended to include all papers examining functional therapy but aimed to examine the use of terminology. We also saw that different terms have been used to describe a "functional" approach of the intervention, such as task-oriented, activity-focused, and goal-directed. It is essential to use the same terminology when describing interventions, so it is clear what has been done and to compare intervention approaches.

Rehabilitation approaches for children with CP are inherently complex, multi-faceted, and comprise a number of interconnected elements. A clear description of each of the elements of an intervention is required for standardization, usability and replication. Unclear descriptions will hamper evidence uptake into clinical practice (implementation), and interventions are unlikely to be delivered as intended with fidelity. Therefore, a clear description of the intervention is of great importance. A tool for intervention description and replication are the TIDieR guidelines (Hoffmann et al., 2014). When interventions are clearly described in clinical guidelines or in consensus statements, the protocols can be more readily embedded in clinical practice. The consensus statement in stroke research is an example, in which definitions and a shared vision for new standards in stroke recovery have been agreed upon internationally (Bernhardt et al., 2017). The Dutch Guideline for diagnosis and treatment of children with spastic CP might be a starting point to bring people together internationally to reach consensus on the definitions of these elements of functional therapy.

Our systematic review concluded that the set criteria and definitions of functional therapy were frequently not used in the included studies. It is of great importance that interventions are described in detail for both research and clinical practice. The criteria as described in the Dutch Guideline for diagnosis and treatment of children with spastic CP are a helpful tool to describe elements of functional interventions. The next step is to reach international consensus on the elements of functional therapy to promote standardization and widespread use.

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