

Clinical Rehabilitation

<http://cre.sagepub.com/>

Occupational therapy for children with cerebral palsy: a systematic review

Esther MJ Steultjens, Joost Dekker, Lex M Bouter, Jos CM van de Nes, Brigitte LM Lambregts and
Cornelia HM van den Ende

Clin Rehabil 2004 18: 1

DOI: 10.1191/0269215504cr697oa

The online version of this article can be found at:

<http://cre.sagepub.com/content/18/1/1>

Published by:



<http://www.sagepublications.com>

Additional services and information for *Clinical Rehabilitation* can be found at:

Email Alerts: <http://cre.sagepub.com/cgi/alerts>

Subscriptions: <http://cre.sagepub.com/subscriptions>

Reprints: <http://www.sagepub.com/journalsReprints.nav>

Permissions: <http://www.sagepub.com/journalsPermissions.nav>

Citations: <http://cre.sagepub.com/content/18/1/1.refs.html>

Occupational therapy for children with cerebral palsy: a systematic review

Esther MJ Steultjens Netherlands Institute for Health Services Research, Utrecht, **Joost Dekker** Department of Rehabilitation Medicine and Institute for Research in Extramural Medicine (EMGO-Institute), **Lex M Bouter** Institute for Research in Extramural Medicine (EMGO-Institute), **Jos CM van de Nes** University of Professional Education, **Brigitte LM Lambregts** SPD Amstel en Zaan, Amsterdam and **Cornelia HM van den Ende** Netherlands Institute for Health Services Research, Utrecht, The Netherlands

Received 6th March 2003; returned for revisions 12th May 2003; revised manuscript accepted 20th July 2003.

Objective: Occupational therapy (OT) for cerebral palsy focuses on the development of skills necessary for the performance of activities of daily living. The aim of this systematic review was to determine whether OT interventions improve outcome for children with cerebral palsy (CP).

Methods: An extensive search in MEDLINE, CINAHL, EMBASE, AMED and SCISEARCH was performed. Studies with controlled and uncontrolled designs were included. Six intervention categories were distinguished and individually analysed using a best-evidence synthesis. This synthesis is based on the type of design, the methodological quality, the type of outcome measures and the statistical significance of the findings.

Results: Seventeen studies were included in this review, seven of which were randomized controlled trials (RCTs). One RCT had a high methodological quality. The analyses resulted in insufficient evidence of the efficacy of occupational therapy in all intervention categories, due to the low methodological quality of studies presenting statistically nonsignificant results.

Conclusion: Despite the reasonable number of studies identified, the inconclusive findings regarding the efficacy of occupational therapy for children with cerebral palsy may be a reflection of the difficulties in efficacy research in OT for children with CP. Future research should critically reflect on methodological issues.

Introduction

Cerebral palsy (CP) is a static encephalopathy that can be defined as a nonprogressive disorder of posture and movement. It is often associated

with epilepsy and abnormalities of speech, vision and intellect, resulting from a defect or lesion in the developmental brain. CP is a common disorder with an estimated prevalence of 2/1000 in the general population.¹ A large variety of symptomatology is seen in children with CP.² It is also a condition that occurs early in life and is present throughout a person's lifetime. It can affect all aspects of a person's development throughout their life. The focus for treatment should be on

Address for correspondence: Esther MJ Steultjens, Netherlands Institute for Health Services Research, PO Box 1568, 3500 BN Utrecht, The Netherlands. e-mail: e.steultjens@nivel.nl

the facilitation of independence.³ The management of a child with cerebral palsy, with the objective of optimizing functional abilities, typically includes the input of many disciplines, including occupational therapy (OT).⁴ One study⁵ reported that 50% of children with CP receive OT. Occupational therapy focuses on the development of skills necessary for the performance of activities of daily living. These activities include play, self-care activities such as dressing, grooming and feeding, and fine motor tasks such as writing and drawing. OT also addresses cognitive and perceptual disabilities, especially in the visual-motor area. Another aspect of OT is the adaptation of equipment and seating to allow better upper extremity use and to promote functional independence.⁶ Furthermore, parent counselling is an important aspect of the OT treatment with regard to optimizing parental support for improving the functional abilities of the child with CP. Different approaches to treatment are taken within OT, such as neurodevelopmental treatment (NDT)⁷ and sensory integration (SI).⁸

Until now, no systematic summary has been produced of the evidence of the efficacy of various OT interventions in children with CP. Five reviews^{9–13} do address OT-related topics in CP but three of these were narrative in origin,^{11–13} while the two systematic reviews^{9,10} focused on the efficacy of very specific interventions applied within OT, viz. NDT in general and adaptive seating respectively. The objective of our systematic review, therefore, was to determine whether OT interventions improve functional ability and social participation for children with CP.

Materials and methods

An extensive search was conducted utilizing the following resources: MEDLINE (1966 until June 2003), CINAHL (1982 until December 2002), EMBASE (1982 until December 2002), SCISEARCH (1974 until December 2002), AMED (1985 until December 2002), Cochrane Controlled Trials Register, The Rehabilitation and Related Therapies (RRT) Field (Cochrane Collaboration), and two Dutch libraries of med-

ical and rehabilitation literature (Dutch National Institute Allied Health Professions (NPI), Netherlands Institute for Health Services Research (NIVEL)).

The computerized search strategy used in PubMed was: ‘Cerebral Palsy’[MESH] AND (‘occupational therapy’[MESH] OR ‘activities of daily living’[MESH] OR ‘exercise therapy’[MESH] OR ‘splints’[MESH] OR ‘self-help devices’[MESH] OR ‘Early Intervention (Education)’[MESH] OR (‘parents/education’[MESH] OR ‘parents/psychology’[MESH]) OR ‘professional family relations’[MESH] OR ‘play and playthings’[MESH]) Limits: Human. The search strategy was adapted by an experienced medical librarian to make it applicable to the other databases.

In addition, the reference lists of all studies identified were scanned and the corresponding authors of papers eligible for inclusion were contacted by mail to identify further studies.

The inclusion of articles was assessed by two independent reviewers (EMJS, CHME), first on the basis of the title and abstract. The article was read in the event of uncertainty. Disagreements were resolved by discussion.¹⁴ All four inclusion criteria had to be met: (1) efficacy studies with either a controlled design or a design other than controlled (ODs) such as pre–post tests or time-series; (2) evaluating OT interventions in children (<19 years) with clinically diagnosed cerebral palsy; (3) used outcome measures: ‘functional ability’ (including motor skills and dexterity) or ‘social participation’, or process measures, which are measures considered to be indicators of successful treatment (e.g., ‘upper-extremity function’, ‘muscle tone’ or ‘balance’); and (4) full-length publications or manuscripts.

Occupational therapy interventions were classified into five specific intervention categories, viz. (1) training of sensorimotor functions including play activities to facilitate motor performance; (2) training of skills including training of daily activities such as feeding, personal hygiene, writing etc.; (3) parental counselling in which parents are educated how to stimulate independence in their child; (4) advice and instruction regarding the use of assistive devices including the provision of mobility aids like wheelchairs and bathroom devices; and (5) provision of splints

such as hand orthoses to facilitate hand function. Furthermore, a sixth category was defined as 'comprehensive OT' (when all five specific intervention categories were part of the OT treatment evaluated). This classification is based on the International Classification of Functioning, Disability and Health (ICF)¹⁵ and enables the categorization of all interventions possible in OT. A group of four occupational therapists (including EMJS and BLML) and reviewer CHME reached consensus on this classification. This group assessed whether the interventions evaluated in each study could be regarded as OT and if so allocated them to one of the intervention categories. The criteria applied were that the intervention had most likely been part of an OT treatment plan and that the treatment was aimed at enhancing performance of daily activities. Disagreements were resolved by discussion.

The methodological quality of all studies was independently assessed by two reviewers (EMJS, BLML). Disagreements were resolved by discussion. If no consensus was reached, a third reviewer (CHME) made the final decision. A list of methodological criteria recommended by van Tulder *et al.*¹⁴ was used for randomized controlled trials (RCTs) and controlled clinical trials (CCTs). This list, containing all the criteria proposed by Jadad *et al.*¹⁶ and Verhagen *et al.*,¹⁷ consists of 11 criteria for internal validity, six descriptive criteria and two statistical criteria (Appendix 1). One modification was made regarding the specification of the 'eligibility' criterion, viz. the condition of interest (i.e., the impairment or disability that indicated referral to OT) was added as an eligibility criterion, as proposed by Wells.¹⁸ All criteria were scored as 'yes', 'no', or 'unclear'. Studies were considered to be of 'high quality' if at least six criteria for internal validity, three descriptive criteria and one statistical criterion were scored positively.

To rate the methodological quality of the other designs (ODs), van Tulder's list was modified with regard to some items (Appendix 1). The amended list consisted of seven criteria for internal validity, four descriptive criteria and two statistical criteria. Studies were considered to be of 'sufficient quality' if at least four criteria for internal validity, two descriptive criteria and one statistical criterion were met.

Analysis of the results was performed separately for each intervention category. A standardized mean difference (Hedges' g)¹⁹ was calculated for continuous variables, and odds ratios with corresponding 95% confidence intervals were computed for dichotomous variables. In crossover trials without a washout period between intervention phases, data after the first phase were not included in the review. The primary analysis was focused on comparisons of an OT intervention group with a 'no treatment' control group. If a study compared the effect of more than two intervention groups, however, two reviewers (EMJS, CHME) decided by consensus how these comparisons would be classified. In the particular case of the comparison of two interventions, the predominant contrast had to be the OT treatment provided.

We anticipated finding too much diversity among the studies, in terms of patients (classification of CP), interventions (duration, frequency and setting) and outcome measures (diversity, presentation of the results), to make a quantitative analysis (meta-analysis) appropriate, and we therefore formulated a best-evidence synthesis. Our best-evidence synthesis is based on the one proposed by van Tulder *et al.*²⁰ and was modified for the purposes of this review by attributing the appropriate level of evidence to the efficacy of OT, taking into account the design of the studies, the methodological quality, the type of outcome measures and the statistical significance of the findings (Appendix 2). A sensitivity analysis was performed by excluding low-quality studies.

Results

The search strategy resulted in a list of 1004 references of studies. The first selection based on title and abstract obtained 128 full articles. Forty-seven studies of these 128 publications concerned the efficacy of OT in children with cerebral palsy. Seventeen of these studies fulfilled all four inclusion criteria. Thirty OT studies²¹⁻⁵⁰ were excluded because a single subject design was used, children with diseases other than CP participated in the study, or the outcome measures were beyond the scope of our review (Appendix 3).

The methodological quality was assessed in

eight RCTs/CCTs and nine ODs (Table 1). One RCT had a high methodological quality. Two of the ODs had sufficient methodological quality. The raters disagreed on 25% of the items. Specifically the descriptive items and the 'compliance' and 'intention to treat analysis' items were scored differently. All disagreements were resolved after discussion. Results of studies that contribute to the conclusion of the best-evidence synthesis will be presented separately for each intervention category.

Comprehensive OT

Two RCTs^{51,52} (Table 2) of low methodological quality compared an intensive NDT and splinting programme with a regular NDT programme and a functional approach respectively. The dexterity outcome measure was evaluated in both studies. No significant differences between groups were reported (Table 3). Both RCTs mea-

sured upper extremity function. No significant differences between groups were reported on this process measure (data not presented). There is insufficient evidence, therefore, for the efficacy of comprehensive OT on dexterity and upper extremity function.

Training of sensorimotor functions

One RCT⁵³ and one CCT⁵⁴ were identified (Table 2). Both studies had low methodological quality. The RCT presented nonsignificant results on the dexterity outcome measures (Table 3). There is insufficient evidence, therefore, for the efficacy of the training of sensorimotor functions on dexterity.

Training of skills

One low-quality OD⁵⁵ (Table 2) evaluated training focused on dressing. No significant results were found on the functional ability out-

Table 1 Fulfilled items of methodological quality

First author	Design	Internal validity	Descriptive	Statistical	MQ
Comprehensive OT					
Law ⁵¹	RCT	b1, i, j, l, n	a, c, m1, m2	o, q	-
Law ⁵²	RCT	b1, i, j, l, n	a, m1, m2	o, q	-
Training of sensorimotor function					
Bumin ⁵⁴	CCT	j, n	a, m1	o, q	-
Talbot ⁵³	RCT	b1, j, l, n	d, m1	o	-
Training of skills					
Guidetti ⁵⁵	OD	j, n, p	a, d, m1	o	-
Training of sensorimotor function versus training of skills					
Carlsen ⁵⁶	RCT	b1, j, n,	d, m1	o, q	-
Parental counselling					
Hanzlik ⁵⁷	RCT	b1, f, g, j, n	a, c, d, m1	o, q	-
McConachie ⁵⁸	RCT	b1, i, j	a, c, k, m2	o, q	-
Advice/instruction regarding assistive devices					
Noronha ⁵⁹	OD	j, n, p	d, m1	o, q	-
Pope ⁶⁰	OD	j, l,	a, m2	o	-
Provision of splints					
Exner ⁶¹	RCT	b1, f, j, l, n, p	a, c, d, m1	o	+
Blair ⁶⁵	OD	i, j, l, n	d, k, m1	o, q	+
Edmondson ⁶⁶	OD	g, n, p	a, d, k, m2	o	-
Flegle ⁶²	OD	g, j, p	a	o	-
Nicholson ⁶⁷	OD	g, j, n, p	a, d, k, m1	o	+
Reid ⁶³	OD	j, n, p	a, d, m1	o	-
Steer ⁶⁴	OD	n, p	a, d, m1	o	-

RCT, randomized controlled trial; CCT, controlled clinical trial; OD, other than controlled design; MQ, methodological quality; + high methodological quality or for ODs sufficient methodological quality; - low methodological quality. See Appendix 1 for meanings of a-q.

come measure (Table 3). There is insufficient evidence, therefore, for the efficacy of the training of skills on functional ability and motor skills.

Training of sensorimotor function versus training of skills

One low-quality RCT⁵⁶ compared a training of sensorimotor function approach with a functional approach. Nonsignificant results were reported on the motor skills outcome measure (Table 3). Insufficient evidence exists, therefore, for a difference in efficacy between training of sensorimotor function and training of skills.

Parental counselling

Two low-quality RCTs^{57,58} (Table 2) evaluated an intervention focused on the parent-child dyad. Both studies measured the functional ability outcome measure and reported nonsignificant results (Table 3). There is insufficient evidence, therefore, for the efficacy of parental counselling on the functional ability of children with CP.

Advice and instruction regarding the use of assistive devices

Two ODs^{59,60} (Table 2) were identified. Both studies had a low methodological quality and both studies reported nonsignificant results on the functional ability outcome measure (Table 3). There is insufficient evidence, therefore, for the efficacy of the use of assistive devices on functional ability.

Provision of splints

One high-quality crossover RCT⁶¹ and three low-quality ODs⁶²⁻⁶⁴ evaluated the efficacy of (arm-) hand splints (Table 2). The RCT⁶¹ reported no significant differences in the motor skills outcome measure between the three types of splints evaluated (Table 3). Three ODs⁶⁵⁻⁶⁷ (Table 2) evaluated the use of Lycra garments. Two ODs^{65,67} were of sufficient methodological quality. One of the three ODs identified⁶⁷ presented a significant increase in the functional ability outcome measure (Table 3). There is insufficient evidence, therefore, for the efficacy of splinting and of wearing Lycra garments on upper extremity function and functional ability.

Discussion

This systematic review explored the efficacy of several occupational therapy interventions in children with cerebral palsy. Six intervention categories were individually analysed for their efficacy on the outcome measures of functional ability as well as on the process measures of upper extremity function. In all intervention categories, the analyses produced insufficient evidence for the efficacy of occupational therapy as a consequence of the low methodological quality of most studies. The analysis of results was hampered by the lack of a clear distinction between outcome measures at the activities and participation level and measures at the body function level (ICF). This was a particular problem where the classification of measurement instruments for motor development in children was concerned, since both levels are often incorporated in the same instrument. Each outcome instrument was classified either at skills level or at functions level, according to descriptions we found in the literature.⁶⁸ The decision made was based on the main items favouring one level. An alternative categorization would not have altered the findings of the review.

The outcome of our review corresponds with the conclusions of reviews of topics related to the efficacy of OT for children with CP,^{9,10,12,13} which all conclude that the evidence for efficacy is inconclusive on account of methodological flaws in original studies. As a consequence, it would still be premature to draw conclusions regarding the efficacy of OT from the evidence presently available.

Clinical messages

- The efficacy of occupational therapy (OT) practice for children with cerebral palsy is still inconclusive.
- Functional ability and social participation should be the main outcome measures in evaluating OT efficacy.
- Future efficacy research needs attention for methodological quality issues.

Table 2 Characteristics of included studies

First author	N	Methods	Inclusion criteria	Intervention	Outcome and process measures	Duration of intervention
Comprehensive OT						
Law ⁵¹	79	RCT	Spastic CP, wrist/hand involved, age 1.5–8 years	I1: intensive NDT + cast I2: regular NDT + cast I3: intensive NDT I4: regular NDT Outpatients	Peabody fine motor scale QUEST (quality of upper extremity skills test)	I1 + I3: 2 × 45 min, a week 6 months, 30 min a day (home) I2 + I4: 1 × 45 min a month 6 months, 3 × 15 min a week at home
Law ⁵²	52	RCT	CP, age 1.5–4 years, hand and wrist function not isolated possible	I: intensive NDT + cast I2: regular OT Outpatients	Peabody fine motor scale QUEST (quality of upper extremity skills test)	I1: 2 × 45 min a week + 30 min daily at home I2: 1 × 45, min a week/month
Training of sensorimotor function						
Burnin ⁵⁴	41	CCT	Spastic diplegic CP	I1: individually sensory perceptual Motor (SPM) training + home programme I2: group SPM training + at home Outpatients	Physical ability test SCMAT (Southern California motor accuracy test)	3 × 1.5 hours a week for 3 months
Talbot ⁵³	59	RCT	CP, attending school for handicapped children	I1: tracing + auditorially feedback I2: tracing without feedback At school	SCMAT (Southern California motor accuracy test)	2 × 10 min 5 days a week for 4 weeks
Training of skills						
Guidetti ⁵⁶	5	OD	CP, age 4–7 years, participating in dressing	I: dressing and undressing during play Outpatients	Klein-Bell scale	2 × 60 min a week for 10 weeks
Training of cognitive functions versus training of skills						
Carlsen ⁵⁶	20	RCT	CP, age 0–5 years, mother active in clinical sessions and with home management	I1: facilitation group, sensory organization, postural stability I2: functional approach, self-care Outpatients	BMS, (Bayley motor development scale) DDST (Denver developmental screening test)	2 × 1 hour a week for 6 weeks
Parental counselling						
Hanzlik ⁵⁷	20	RCT	CP, no major sensory handicap, not able to ambulate independently	I1: verbal instruction behaviour I2: NDT instruction At home	Independent play (observation)	1 × 1 hour

McConachie ⁵⁸	58	RCT	CP, age 1.5–5 years	I1: distance training, urban population R1: mother-child group, urban I2: distance training, rural population R2: health advice, rural Outpatients	IBAS (independent behaviour assessment scale)	I1: 9 × 1.5 hour in 9 months R1: each day I2: 9 × 1.5 hour in 9 months R2: 1x
Advice/instruction regarding assistive devices						
Noronha ⁵⁹	10	OD	Spastic diplegic CP, IQ not < 1 SD from mean	I: prone stander, At school	Jebsen Taylor hand function test, immediate effect	1 × difference in test, sitting versus prone stander
Pope ⁶⁰	9	OD	CP, major postural deficit, at risk for spinal deformities	I: SAM seating system At home and school	Mobility, 5 point scale	each day 0–12 hours for 3 years
Provision of splints						
Exner ⁶¹	12	RCT	Spastic hemiplegic CP, age 2–16 years, mental age ≥ 18 months, no visual deficit	I1: orthokinetic cuff I2: short opponens thumb splint I3: MacKinnon splint Outpatients	Bilateral hand use (observation)	8 hours each day for 6 weeks
Blair ⁶⁵	25	OD	CP, age 1.25–14 years, suboptimal proximal stability	I: Lycra garment (UP suit) At home and school	Gross and fine motor function, (observation)	8 hours each day for 3 weeks
Edmondson ⁶⁶	15	OD	CP	I: Lycra garment At school and home	Gross-motor skills, balance, fine motor function	6 ours each day for 12 months
Flegle ⁶²	3	OD	Spastic hemiplegic CP, grasp difficulties	I: MacKinnon splint At school	Grasp skill test	8 hours each day
Nicholson ⁶⁷	12	OD	CP, significant impairment of upper limb function	I: Lycra garment At home/outpatients	PEDI (Paediatric Evaluation of Disability/Inventory)	6 hours each day for 6 weeks
Reid ⁶³	10	OD	CP, full passive range of motion in both arms	I: Hand positioning device At school	Visual-motor accuracy test	1 × 1.5 hour, immediate effect
Steer ⁶⁴	6	OD	CP, developing contractures	I: Elbow/wrist cast Outpatients	Passive range of motion, goniometer	24 hours a day for 1 week, than 2 × new cast

RCT, randomized controlled trial; CCT, controlled clinical trial; OD, other than controlled design; CP, cerebral palsy; I, intervention group; R, reference group; NDT, neurodevelopmental treatment.

The poor methodological quality of OT studies is also a factor in the recent studies. In the last few decades a lot of attention has been paid to raising the methodological quality of randomized controlled trials, as is shown by the 'CONSORT statement' for instance.⁶⁹ It should be a matter of great concern that this development is not reflected in the recent studies included in this

review. We are not able to support or refute the efficacy of OT in children with CP on the basis of the outcome of our review. A reason for this inconclusiveness could be the possible masking of significant interactions between such variables as IQ, age, type of CP, degree of impairments, parental participation, emotional disturbance, intensity of treatment and type of intervention in

Table 3 Effects on motor skills, dexterity and functional ability

Reference (N)	Design	Methodological quality	Motor skills		Functional ability	
			Mean (SD) baseline	SMD [CI]	Mean (SD) baseline	SMD [CI]
Comprehensive OT						
Law ⁵¹ (79)	RCT	low	I: 25.0 (17.5)	0.14 ^a	–	–
NDT			R: 27.3 (20.3)	(–0.52;0.79)	–	–
Law ⁵¹ (79)	RCT	low	I: 30.6 (18.4)	0.14 ^a	–	–
cast			R: 27.3 (20.3)	(–0.52;0.80)	–	–
Law ⁵² (52)	RCT	low	I: 20.4 (9.0)	0.10 ^a	–	–
			R: 19.2 (8.6)	(–0.45;0.66)	–	–
Training of sensorimotor function						
Bumin ⁵⁴ (41)	CCT	low	NR	0.85 ^a	NR	0.50
				(0.00;1.70)		(–0.32;1.33)
Talbot ⁵³ (59)	RCT	low	NR	NE ^a	–	–
Training of skills						
Guidetti ⁵⁵ (5)	OD	low	–	–	I: 52.6 (7.5)	<i>p</i> ≥ 0.05
Training of sensorimotor functions vs training of skills						
Carlsen ⁵⁶ (20)	RCT	low	I: 17.5 (10.9)	0.12	–	–
			R: 16.1 (9.2)	(–0.68;0.92)		
Parental counselling						
Hanzlik ⁵⁷ (20)	RCT	low	–	–	I: 0.09 (0.1)	0.17
					R: 0.17 (0.1)	(–0.71;1.05)
McConachie ⁵⁸ (58)	RCT	low	–	–	I: –2.6 (1.1)	0.27
urban					R: –2.0 (1.5)	(–5.0;1.04)
McConachie ⁵⁸ (58)	RCT	low	–	–	I: –2.1 (1.8)	0.05
rural					R: –1.8 (2.0)	(–0.68;0.77)
Advice/instruction regarding assistive devices						
Noronha ⁵⁹ (10)	OD	low	NR	<i>p</i> = 0.87	–	–
Pope ⁶⁰ (9)	OD	low	–	–	I: 2.7 (0.5)	NR
Provision of splints						
Exner ⁶¹ (12)	RCT	high	NR	NE	–	–
Blair ⁶⁵ (25)	OD	sufficient	NR	NR	–	–
Edmondson ⁶⁶ (15)	OD	low	–	–	I:54.9 (22.1)	NR
Nicholson ⁶⁷ (12)	OD	sufficient	–	–	NR	<i>p</i> = significant

^aDexterity.

ADL, activities of daily living; SMD, standardized mean difference; CI, 95% confidence interval; I, intervention group; R, reference group; RCT, randomized controlled trial; CCT, controlled clinical trial; OD, other than controlled design; NR, not reported; NE, not estimable; – not assessed.

present research paradigms.¹³ Occupational therapists consider and explore this whole range of child characteristics, environmental characteristics and CP symptoms to formulate individually focused goals and treatment planning. The characteristics of an RCT in which large homogeneous groups are evaluated on the efficacy of a homogeneous intervention are in contradiction with this clinical practice, which might provide an explanation for the weakness of most studies included in this review. The inclusion of a sufficiently large homogeneous population should be an objective of future efficacy research. The criteria of sample size and homogeneity should both be fulfilled, and a pilot study might be required to establish whether these criteria can be fulfilled in CP research.

The large variability in OT treatment for children might explain the 15 single-case studies identified in this review. Single-case studies using a repeated time-series design deal with the variability problem, but the single-case design studies were excluded from our review, however, because of the difficulty in generalizing evidence from these studies to a larger population of children with CP.

Another reason for the inconclusive findings could be the outcome measures chosen. The instrumentation used may have been insensitive to the subtle but important motor progress exhibited in children with CP. Furthermore, relevant gains in nonmotor areas, such as emotional status, parent/child interaction, language development and cognitive development, might have been expected but were evaluated sparsely.¹³ The main goals of occupational therapy are to increase functional abilities and to enhance social participation and well-being. Instruments like the Pediatric Evaluation of Disability Inventory (PEDI)⁷⁰ have recently been developed to measure a wide range of functional abilities (not only motor skills or dexterity), but were used in only a few studies included in our review. Social participation and well-being were not measured at all, which demonstrates the need for the development and use of reliable and valid comprehensive assessment batteries, incorporating outcomes that reflect the aims of occupational therapy.

Although we were able to identify a reasonable

number of studies, the inconclusive findings regarding the efficacy of occupational therapy for children with CP reflect the difficulties in efficacy research into OT for children with CP. Future research should critically reflect on methodological issues such as homogeneity, sample size and outcome measures.

Acknowledgements

This study was funded by a grant (grant number 00-04) from the Dutch Healthcare Insurance Board (College voor Zorgverzekeringen (CVZ)).

The authors would like to thank Mrs M Breedijk and Mr J Wielders for discussing occupational therapy issues and Mrs E Breuning for making the search strategy applicable to the several databases.

References

- Behrman RE, Kliegman R, Jenson HB. Encephalopathies; cerebral palsy. In: Behrman RE, Kliegman R, Jenson HB, eds. *Nelson textbook of pediatrics*, sixteenth edition. Philadelphia: WB Saunders; 2000: 843–45.
- DeLisa JA, Gans BM, Bockenek WL. *Rehabilitation medicine: principles and practice*. Philadelphia: Lippincott-Raven, 1998.
- Rosenbaum P. Cerebral palsy: what parents and doctors want to know. *BMJ* 2003; **326**: 670–74.
- Rosen BA. Cerebral palsy. In Hultman CB, eds. *Gellis and Kagan's current pediatric therapy*, fifteenth edition. Philadelphia: WB Saunders, 1996: 79–82.
- Koman LA. *Impact of cerebral palsy*. Accessed 5 March 2001, from www.ucpa.org/ucp_generaldoc.cfm/123/4/24/24-6607/106.2002
- Wilsdon J. Cerebral palsy. In: Turner A, Foster M, Johnson SE eds. *Occupational therapy and physical dysfunction*. New York: Churchill Livingstone, 1996: 395–432.
- Bobath B. *Motor development in the different types of cerebral palsy*. New York: William Heinemann, 1975.
- Ayres AJ. *Sensory integration and learning disorders*. Los Angeles: Western Psychological Services, 1972.
- Brown GT, Burns SA. The efficacy of neurodevelopmental treatment in paediatrics: a systematic review. *Br J Occup Ther* 2001; **64**: 235–44.
- Roxborough L. Review of the efficacy and effectiveness of adaptive seating for children with cerebral palsy. *Assist Technol* 1995; **7**: 17–25.

- 11 Gormley ME. The treatment of cerebral origin spasticity in children. *Neurorehabilitation* 1999; **12**: 93–103.
- 12 Hur JJ. Review of research on therapeutic interventions for children with cerebral palsy. *Acta Neurol Scand* 1995; **91**: 423–32.
- 13 Parette HP, Hourcade Jr. A review of therapeutic intervention research on gross and fine motor progress in young children with cerebral palsy. *Am J Occup Ther* 1984; **38**: 462–68.
- 14 Tulder van MW, Assendelft WJJ, Koes BW, Bouter L. Method guidelines for systematic reviews in the Cochrane collaboration Back review group for spinal disorders. *Spine* 1997; **22**: 2323–30.
- 15 WHO. *International Classification of Functioning, Disability and Health*. Geneva: WHO, 2001.
- 16 Jadad AR, Moore RA, Carroll D *et al.* Assessing the quality of reports of randomized clinical trials: is blinding necessary? *Controlled Clin Trials* 1996; **17**: 1–12.
- 17 Verhagen AP, Vet de HCW, Bie de RA *et al.* The delphi list: A criteria list for quality assessment of randomized clinical trials for conducting systematic reviews developed by delphi consensus. *J Clin Epidemiol* 1998; **51**: 1235–41.
- 18 Wells GA, Shea B, O'Connell D, Peterson J, Welch V, Tugwell P. The Newcastle-Ottawa scale (NOS) for assessing the quality of nonrandomized studies in meta-analyses. *3rd symposium on systematic reviews*. Accessed 2000 from www.lri.ca
- 19 Hedges LV. Estimation of effect size from a series of independent experiments. *Psychol Bull* 1982; **92**: 490–99.
- 20 Tulder van MW, Cherkin DC, Berman B, Lao L, Koes BW. Acupuncture for low back pain. In: *The Cochrane Library*, Issue 1. Oxford: Update Software, 2003.
- 21 Barray V, Bourrelis C, D'Heilly N, Lacet P. Constructive dyspraxia in premature children born with cerebral palsy: Long-term therapeutic results in 13 children. *Ann Readapt Med Phys* 1999; **42**: 46–50.
- 22 Case-Smith J. Fine motor outcomes in pre-school children who receive occupational therapy services. *Am J Occup Ther* 1996; **50**: 52–61.
- 23 Colbert AP, Doyle KM, Webb WE. DESEMO seats for young children with cerebral palsy. *Arch Phys Med Rehabil* 1986; **67**: 484–86.
- 24 Crawford PJ, Burril C, Grogan J. The effect of modifying toothbrush handles on plaque control in handicapped children: preliminary report. *Proc Br Paedod Soc* 1977; **7**: 11–13.
- 25 Damle SG, Bhavsar JP. Plaque removing efficacy of individually modified toothbrushes in cerebral palsy children. *ASDC J Dent Child* 1995; **62**: 279–82.
- 26 Fetters L, Kluzik JA. The effects of neurodevelopmental treatment versus practice on the reaching of children with spastic cerebral palsy. *Phys Ther* 1996; **76**: 346–58.
- 27 Fisher WE, Seeger BR. A comparative evaluation of special seating for severely disabled children. *Australas Phys Eng Sci Med* 1987; **10**: 128–43.
- 28 Hankinson J, Morton RE. Use of a lying hip abduction system in children with bilateral cerebral palsy: a pilot study. *Dev Med Child Neurol* 2002; **44**: 177–80.
- 29 Hasdai A, Jessel AS, Weiss PL. Use of a computer simulator for training children with disabilities in the operation of a powered wheelchair. *Am J Occup Ther* 1998; **52**: 215–20.
- 30 Hulme JB, Shaver J, Acher S, Mullette L, Eggert C. Effects of adaptive seating devices on the eating and drinking of children with multiple handicaps. *Am J Occup Ther* 1987; **41**: 81–89.
- 31 Hulme JB, Gallagher T, Walsh J, Niesen S, Waldron D. Behavioral and postural changes observed with use of adaptive seating by clients with multiple handicaps. *Phys Ther* 1987; **67**: 1060–67.
- 32 Manley MT, Gurtowski J. The vertical wheeler: a device for ambulation in cerebral palsy. *Arch Phys Med Rehabil* 1985; **66**: 717–20.
- 33 Palmer FB, Shapiro BK, Allen MC *et al.* Infant stimulation curriculum for infants with cerebral palsy: effects on infant temperament, parent-infant interaction and home environment. *Pediatrics* 1990; **85**: 411–15.
- 34 Reid DT. An instrumentation approach for assessing the effects of a hand positioning device on reaching motion of children with cerebral palsy. *Occup Ther J Res* 1992; **12**: 278–95.
- 35 Rennie DJ, Attfield SF, Morton RE, Polak FJ, Nicholson J. An evaluation of lycra garments in the lower limb using 3-D gait analysis and functional assessment (PEDI). *Gait Posture* 2000; **12**: 1–6.
- 36 Barnes KJ. Improving prehension skills of children with cerebral palsy. *Occup Ther J Res* 1989; **6**: 227–39.
- 37 Barnes KJ. Direct replication: relationship of upper-extremity weight bearing to hand skills of boys with cerebral palsy. *Occup Ther J Res* 1989; **9**: 235–42.
- 38 Crocker MD, MacKay-Lyons M, McDonnell E. Forced use of the upper extremity in cerebral palsy: a single-case design. *Am J Occup Ther* 1997; **51**: 824–33.
- 39 Durfee JL, Billingsley FF. A comparison of two computer input devices for upper case letter matching. *Am J Occup Ther* 1998; **53**: 214–20.
- 40 Everson JM, Goodwyn R. A comparison of the use of adaptive microswitches by students with cerebral palsy. *Am J Occup Ther* 1987; **41**: 739–44.
- 41 Goodman G, Bazyk S. The effects of a short thumb opponens splint on hand function in cerebral palsy: a single-subject study. *Am J Occup Ther* 1991; **45**: 726–31.
- 42 Hsieh MC, Luo CH. Morse code text typing training of a teenager with cerebral palsy using a six-switch Morse keyboard. *Technol Disabil* 1999; **10**: 169–73.

- 43 Hulme JB, Bain B, Hardin M, McKinnon A, Waldron D. The influence of adaptive seating devices on vocalization. *J Commun Disord* 1989; **22**: 137–45.
- 44 Kinghorn J, Roberts G. The effect of an inhibitive weight-bearing splint on tone and function: a single-case study. *Am J Occup Ther* 1996; **10**: 807–15.
- 45 Lilly LA, Powell NJ. Measuring the effects of neurodevelopmental treatment on the daily living skills of 2 children with cerebral palsy. *Am J Occup Ther* 1990; **44**: 139–45.
- 46 McCormack DJ. The effects of keyguard use and pelvic positioning on typing speed and accuracy in a boy with cerebral palsy. *Am J Occup Ther* 1990; **44**: 312–15.
- 47 Reid D, Koheil R. EMG biofeedback training to promote hand function in a cerebral palsied child with hemiplegia. *Hand Rehabil Occup Ther* 1987; **4**: 97–107.
- 48 Sakemiller LM, Nelson DL. Eliciting functional extension in prone through the use of a game. *Am J Occup Ther* 1998; **52**: 150–57.
- 49 Smiths LH, Harris SR. Upper extremity inhibitive casting for a child with cerebral palsy. *Phys Occup Ther Pediatr* 2002; **5**: 71–79.
- 50 Tona JL, Schneck CM. The efficacy of upper extremity inhibitive casting – a single-subject pilot-study. *Am J Occup Ther* 1993; **47**: 901–10.
- 51 Law M, Cadman D, Rosenbaum P, Walter S, Russell D, DeMatteo C. Neurodevelopmental therapy and upper-extremity inhibitive casting for children with cerebral palsy. *Dev Med Child Neurol* 1991; **33**: 379–87.
- 52 Law M, Russell D, Pollock N, Rosenbaum P, Walter S, King G. A comparison of intensive neurodevelopmental therapy plus casting and a regular occupational therapy program for children with cerebral palsy. *Dev Med Child Neurol* 1997; **39**: 664–70.
- 53 Talbot ML, Junkala J. The effect of auditorially augmented feedback on the eye-hand coordination of students with cerebral palsy. *Am J Occup Ther* 1981; **35**: 525–28.
- 54 Bumin G, Kayihan H. Effectiveness of two different sensory-integration programmes for children with spastic diplegic cerebral palsy. *Disabil Rehabil* 2001; **23**: 394–99.
- 55 Guidetti S, Soderback I. Description of self-care training in occupational therapy: case studies of five Kenyan children with cerebral palsy. *Occup Ther Int* 2001; **8**: 34–48.
- 56 Carlsen PN. Comparison of two occupational therapy approaches for treating the young cerebral-palsied child. *Am J Occup Ther* 1975; **29**: 267–72.
- 57 Hanzlik JR. The effect of intervention on the free-play experience for mothers and their infants with developmental delay and cerebral palsy. *Phys Occup Ther Pediatr* 1989; **9**: 33–51.
- 58 McConachie H, Huq S, Munir S, Ferdous S, Zaman S, Khan NZ. A randomized controlled trial of alternative modes of service provision to young children with cerebral palsy in Bangladesh. *J Pediatr* 2000; **137**: 769–76.
- 59 Noronha J, Bundy A, Groll J. The effect of positioning on the hand function of boys with cerebral palsy. *Am J Occup Ther* 1989; **43**: 507–12.
- 60 Pope PM, Bowes CE, Booth E. Postural control in sitting the SAM system: evaluation of use over three years. *Dev Med Child Neurol* 1994; **36**: 241–52.
- 61 Exner CE, Bonder BR. Comparative effects of three hand splints on bilateral hand use, grasp, and arm-hand posture in hemiplegic children: a pilot study. *Occup Ther J Res* 1983; **3**: 75–92.
- 62 Flegle JH, Leibowitz JM. Improvement in grasp skill in children with hemiplegia with the MacKinnon splint. *Res Dev Disabil* 1988; **9**: 145–51.
- 63 Reid DT, Sochaniwskyj A. Influences of a hand positioning device on upper-extremity control of children with cerebral palsy. *Int J Rehabil Res* 1992; **15**: 15–29.
- 64 Steer V. Upper limb serial casting of individuals with cerebral palsy; a preliminary report. *Aust Occup Ther J* 1989; **36**: 69–77.
- 65 Blair E, Ballantyne J, Horsman S, Chauvel P. A study of a dynamic proximal stability splint in the management of children with cerebral palsy. *Dev Med Child Neurol* 1995; **37**: 544–54.
- 66 Edmondson J, Fisher K, Hanson C. How effective are lycra suits in the management of children with cerebral palsy. *J Assoc Pediatr Phys* 1999; **90**: 49–57.
- 67 Nicholson JH, Morton RE, Attfield S, Rennie D. Assessment of upper-limb function and movement in children with cerebral palsy wearing lycra garments. *Dev Med Child Neurol* 2001; **43**: 384–91.
- 68 Van Deusen J, Brunt D. *Assessment in occupational therapy and physical therapy*, first edition. Philadelphia: WB Saunders, 1997.
- 69 Begg C, Cho M, Eastwood S *et al*. Improving the quality of reporting of randomized clinical trials: the CONSORT statement. *JAMA* 1996; **276**: 637–39.
- 70 Haley SM, Coster WJ, Ludlow LH *et al*. *Pediatric evaluation of disability inventory: development, standardization and administration manual*, first edition. Boston: New England Medical Center Hospital, 1992.

Appendix 1 – Criteria of methodological quality

Randomized controlled trials (RCTs) and controlled clinical trials (CCTs)

Patient selection

- a) Were the eligibility criteria specified?
- b) Treatment allocation:
 - 1) Was a method of randomization performed?
 - 2) Was the treatment allocation concealed?
- c) Were the groups similar at baseline regarding the most important prognostic indicators?

Interventions

- d) Were the index and control interventions explicitly described?
- e) Was the care provider blinded for the intervention?
- f) Were co-interventions avoided or comparable?
- g) Was the compliance acceptable in all groups?
- h) Was the patient blinded to the intervention?

Outcome measurement

- i) Was the outcome assessor blinded to the interventions?
- j) Were the outcome measures relevant?
- k) Were adverse effects described?
- l) Was the withdrawal/drop out rate described and acceptable?
- m) Timing follow-up measurements:
 - 1) Was a short-term follow-up measurement performed?
 - 2) Was a long-term follow-up measurement performed?
- n) Was the timing of the outcome assessment in both groups comparable?

Statistics

- o) Was the sample size for each group described?
- p) Did the analysis include an intention-to-treat analysis?
- q) Were point estimates and measures of variability presented for the primary outcome measures?

Other than controlled designs (OD)

Patient selection

- a) Were the eligibility criteria specified?

Interventions

- d) Was the intervention explicitly described?
- f) Were co-interventions avoided?
- g) Was the compliance acceptable?

Outcome measurement

- i) Was the outcome assessor not involved in the treatment?
- j) Were the outcome measures relevant?
- k) Were adverse effects described?
- l) Was the withdrawal/drop out rate described and acceptable?
- m) Timing follow-up measurements:
 - 1) Was a short-term follow-up measurement performed?
 - 2) Was a long-term follow-up measurement performed?
- n) Was the timing of the outcome assessment in all patients comparable?

Statistics

- o) Was the sample size of the patient group described?
- p) Did the analysis include an intention-to-treat analysis?
- q) Were point estimates and measures of variability presented for the primary outcome measures?

Internal validity criteria: b, e, f, g, h, i, j, l, n, p; descriptive criteria: a, c, d, k, m; statistical criteria: o, q.

Appendix 2 – Best-evidence synthesis

Strong evidence	Provided by consistent, statistically significant findings in <i>outcome</i> measures in at least two high-quality RCTs ^a
Moderate evidence	Provided by consistent, statistically significant findings in <i>outcome</i> measures in at least one high-quality RCT and at least one low-quality RCT or high-quality CCT ^a
Limited evidence	Provided by statistically significant findings in <i>outcome</i> measures in at least one high-quality RCT ^a
or	Provided by consistent, statistically significant findings in <i>outcome</i> measures in at least two high-quality CCTs ^a (in the absence of high-quality RCTs)
Indicative findings	Provided by statistically significant findings in <i>outcome and/or process</i> measures in at least one high-quality CCT or low-quality RCT ^a (in the absence of high-quality RCTs)
or	Provided by consistent, statistically significant findings in <i>outcome and/or process</i> measures in at least two ODs with sufficient quality (in the absence of RCTs and CCTs) ^a
No or insufficient evidence	In the case that results of eligible studies do not meet the criteria for one of the above stated levels of evidence
or	In the case of conflicting (statistically significant positive and statistically significant negative) results among RCTs and CCTs
or	In the case of no eligible studies

^aIf the number of studies showing evidence is less than 50% of the total number of studies found within the same category of methodological quality and study design (RCTs, CCTs or ODs), we state no evidence. RCT, randomized controlled trial; CCT, controlled clinical trial; OD, design other than controlled.

Appendix 3 – Excluded OT studies with the reason for exclusion

Reference	Design and/or other reason for exclusion
Barray ²¹	Multidisciplinary, outcome measures not included in review
Case-Smith ²²	Participants with CP and other diseases
Colbert ²³	Outcome measures not included in review
Crawford ²⁴	Outcome measures not included in review
Damle ²⁵	Outcome measures not included in review
Fetters ²⁶	Outcome measures not included in review
Fisher ²⁷	Participants with CP and other diseases
Hankinson ²⁸	Outcome measures not included in review
Hasdai ²⁹	Participants with CP and other diseases
Hulme ³⁰	Participants with CP and other diseases
Hulme ³¹	Participants with CP and other diseases
Manley ³²	Outcome measures not included in review
Palmer ³³	Outcome measures not included in review
Reid ³⁴	Outcome measures not included in review
Rennie ³⁵	Participants with CP and other diseases
Barnes ³⁶	Single subject design
Barnes ³⁷	Single subject design
Crocker ³⁸	Single subject design
Durfee ³⁹	Single subject design
Everson ⁴⁰	Single subject design, outcome measures not included in review
Goodman ⁴¹	Single subject design
Hsieh ⁴²	Single subject design
Hulme ⁴³	Single subject design, participants with CP and other diseases
Kinghorn ⁴⁴	Single subject design
Lilly ⁴⁵	Single subject design
McCormack ⁴⁶	Single subject design
Reid ⁴⁷	Single subject design
Sakemiller ⁴⁸	Single subject design
Smiths ⁴⁹	Single subject design
Tona ⁵⁰	Single subject design