Adaptive seating and adaptive riding in children with cerebral palsy

Angsupaisal, Mattana

Part II

Chapter 4

Review:
on the effects of adaptive seating systems (AdSS) in children with severe CP

Chapter 4

Adaptive seating systems in children with severe cerebral palsy across International Classification of Functioning, Disability and Health for Children and Youth version domains: a systematic review

Mattana Angsupaisal
Carel G.B. Maathuis
Mijna Hadders-Algra

Developmental Medicine & Child Neurology 2015 April; 57: 919–931

ABBREVIATIONS

AdSS Adaptive seating system
COPM Canadian Occupational Performance Measure
GAS Goal Attainment Scaling
ICF-CY International Classification of Functioning, Disability and Health for Children and Youth version
SSRD Single-subject research design

ABSTRACT

Aim The aim of this study was to systematically review the effect of adaptive seating systems (AdSSs) in young people < 19 years of age with severe cerebral palsy (CP), with particular focus on child-related outcomes across all components of the functioning and disability domains of the International Classification of Functioning, Disability, and Health for Children and Youth version (ICF-CY).

Method Literature searches of studies published from 1975 to October 2014 were performed. Methodological quality and the risk of bias were analysed using Sackett's level of evidence, the American Academy for Cerebral Palsy and Developmental Medicine guidelines, and Mallen criteria for observational studies.

Results Nine studies fulfilled the selection criteria. All studies had level IV evidence and were of moderate methodological quality. The results focused on the effects of AdSSs on postural control and on upper extremity function and on additional child-related outcomes. The results suggested that AdSSs that include trunk and hip support devices may improve postural control outcomes, and that special-purpose AdSSs may improve self-care and play behaviour at home.

Interpretation Because of a low level of evidence and the moderate methodological quality of the studies available, no robust conclusions can be drawn. Nevertheless, the data suggest that AdSSs may be able to improve activity and participation at home among children with severe CP. More studies of high methodological quality addressing the effect of AdSSs on activity and participation are urgently needed. Suggestions for future research are provided.

WHAT THIS PAPER ADDS

— Low levels of evidence from the available studies precludes pertinent conclusions on the effectiveness of AdSSs in children with severe CP.
— Limited evidence suggests that special-purpose AdSSs may improve self-care and play behaviour.

Introduction

Adaptive seating systems (AdSSs) are part of the postural management programme recommended in multifaceted guidance for children with severe cerebral palsy (CP).[1,2] The survey of the Surveillance of Cerebral Palsy in Europe (SCPE)[3] showed that about 40% of children with CP have a severe form of CP, which means that their gross motor function is classified in level IV or V according to the Gross Motor Function Classification System (GMFCS).[4] Children with severe forms of CP face serious limitations of functioning, activity, and participation. The functional limitations are apparent from the children's mobility. Children classified in GMFCS level IV usually function in supported sitting with limited self-mobility. They require adaptive seating, which provides support at the level of the pelvis and trunk, and they need physical assistance for most transfers.[4,5] Children functioning in GMFCS level V have severe limitations in pelvis, trunk, and head control.[5] They typically require a seating system which provides support for head, trunk, pelvis, and legs, and they depend on adult assistance for mobility and self-care throughout the day.[4,5] Their severe postural dysfunction[5] interferes significantly with activities of daily living, such as eating, playing, dressing, and undressing; it also restricts their participation, such as their interaction with friends or family members.[4,6,7]

Adaptive seating for children with severe CP has traditionally been considered a therapeutic intervention that focuses on the improvement of positioning and functioning, and the prevention of long-term complications, such as spinal deformity, hip displacement, and respiratory problems.[6,8–13] Recently, however, a paradigm shift occurred in paediatric rehabilitation. The primary goal of intervention is no longer to improve the child's body function and structure, but to improve the child's ability to undertake daily activities and enhance participation.[2,6–12] This also holds true for the goal of AdSSs in children with severe CP.[1,2,6–12] In general, AdSSs in children with severe CP are highly individualized.[4,13] They may be attached to a mobility base, thereby creating a wheelchair seating system.[4,13] Other specific variants consist of AdSSs designed for activities at school, or AdSSs used for eating or using the toilet.[2,12,13] Whether or not AdSSs can effectively maximize comfort and ease of care, promote activities and participation, and assist in delaying the development of spinal and/or hip deformity in children with severe CP is not clear.[6,12]

The International Classification of Functioning, Disability and Health for Children and Youth version (ICF-CY)[9] offers an ideal framework for the evaluation of AdSSs in children with severe CP.[6,8,9,12] The ICF-CY framework views 'health condition' and 'functioning and disability' as a dynamic interaction of three components: (1) body functions and structures/impairments, (2) activities/activity limitations, and (3) participation/restrictions. In addition, these components (or 'domains') are continuously affected by contextual factors, such as personal and environmental factors.[6,7,9–11] Interventions using AdSSs can be viewed as influencing 'the immediate environment' of the child.[2,9,11] The AdSS may – together with other contextual elements (e.g. caregiver and societal attitudes and caregiver needs) – act as a facilitator of change in function.[2,6,8,9,11,12] For instance, it is conceivable that sitting more upright with more comfort is associated with better eye contact, which may, in turn, affect activity (e.g. meaningful play) and participation (e.g. interaction

with friends and family members at home, at school, and in the community). The application of the ICF-CY framework in the systematic evaluation of the evidence of the effectiveness of AdSSs results in an overview of outcomes across the three domains of body functions and structures, activities, and participation.[6,9,12]

There are five published papers reviewing the literature available on AdSS.[14–18] Only one of them, that by Chung et al.,[16] focused on the effect of AdSSs in children with non-ambulatory CP (GMFCS levels IV and V). That review focused on the postural effects of AdSSs. It may be regarded as a pioneer paper in this area of research as it assessed the methodological quality of the studies included, and classified outcomes according to the ICF.[19] Last year, as 7 years had passed since the review of Chung et al.[16] and the adjustment of the ICF framework[19] to the paediatric field (ICF-CY),[9] we considered it time for a new review.

The aim of this systematic review is to evaluate and provide an updated critical review and synthesis of literature available on the effects of AdSSs in children with severe CP (GMFCS levels IV and V), across all components of functioning and disability (body functions and structures, activity, participation),[9] especially taking into account the methodological quality of the studies reviewed. We aimed to answer three questions. First, what are the functional effects of specific components of AdSSs, i.e. (a) hip- and pelvis-stabilizing devices and (b) trunk support devices? Second, does the functional effect of AdSSs depend on the severity of CP (GMFCS level IV or V) or the type of CP? In other words, is the effect different for children with bilateral spastic CP, unilateral spastic CP, and dyskinetic CP? In the evaluation of the above-mentioned functional effects, the three domains of the ICF-CY framework are addressed separately. Up until now, reviews pooled the effects on the level of activity and participation. Therefore, the third question is 'What are the effects of the factors addressed in questions 1 and 2 on the level of (a) body functions and structures, (b) activities, and (c) participation?'

To this end we reviewed the literature published between 1975 and October 2014, and systematically evaluated the effect of specific components of AdSSs and specific characteristics of the children with severe CP. The review focuses on children with CP in GMFCS levels IV and V, as children with these most severe forms of CP may profit most from AdSSs,[1,2,4–6,8] as their postural control is most impaired.[5] In addition, we assessed methodological quality using three approaches: (1) the system of Sackett et al.,[20] (2) the American Academy for Cerebral Palsy and Developmental Medicine's (AACPDM) methodology for conducting systematic reviews,[21,22] and (3) the appraisal of Mallen et al.[23] The conclusion about the effect of AdSSs was based only on studies meeting the criteria for sufficient methodological quality.

Method

Search strategy

A literature search was performed to identify studies published from 1975 to October 2014. Electronic databases searched were PubMed, EMBASE, Scopus, CINAHL, Ovid MEDLINE, and Allied Health Evidence (searched across four databases: PEDro, OTseeker, SpeechBITE, and PsycBITE). Reference lists in original studies and reviews were examined and *Developmental Medicine and*

Child Neurology was also hand searched for appropriate articles. We combined the search terms 'cerebral palsy', 'adaptive seating', 'adaptive seating system', 'support sitting', 'seated posture', and 'sitting posture'. Medical subject headings (MeSH) and a Thesaurus were used to customize the search terms for each database.

Publications were included if (1) they evaluated groups which consisted of individuals with severe forms of CP, that is individuals who were described as functioning in GMFCS level IV or V or as having severe CP or being non-ambulatory; (2) the study population age was less than or equal to 18 years; (3) interventions focused on the effect of the entire seat (the adaptive seating system, AdSS) or the effect of specific elements of the AdSS; and (4) outcomes included sitting posture and/or postural outcomes, upper extremity function, activity, participation, and quality of life. Articles were excluded if (1) the children had comorbidities unrelated to CP; (2) participants classified in GMFCS levels IV or V and aged less than or equal to 18 years constituted less than 50% of the whole group; and (3) the article was a review, survey, anecdote, letter, or comment, or was unpublished, or came from a non-peer-reviewed source. Papers were not excluded on the basis of the language in which the paper was published.

Evaluation procedure

Two reviewers (MA and MH-A) independently read the papers and summarized their findings on a data extraction summary form, including the assessment of methodological quality and risk of bias. In case of disagreement or any discrepancies in scores, details were discussed until consensus was reached. The consensus is reported in the tables. The evaluation started with an assessment of the level of quantitative evidence according to the Sackett methodology,[20] supplemented by the criteria of the AACPDM methodology (revision 1.2, 2008 version).[21,22] The AACPDM methodology for group design studies[21] and single-subject design studies[24] consists of 7-item scales and 14-item scales respectively (for descriptor details see Tables I and Table SI [online supporting information]). While applying the AACPDM methodology,[21] we discovered that some items could be interpreted in multiple ways in our study. We therefore developed a more detailed description of the item criteria (Table SI, online supporting information). Each item has a dichotomous score ('yes', for criterion present; 'no', for criterion not present). The number of items for which the criterion was present defines the methodological quality score. The scores of group studies (Table I) were used to classify the methodological quality as strong (6 or 7), moderate (3–5), or weak (≤2). This means that we slightly modified the interpretation of the AACPDM, which considers a score of 3 as weak, as none of the studies fulfilled criterion 4 (whether or not assessors were masked for intervention). In the case of single-subject studies (Table SII, online supporting information), the quality of the methodology was assessed as strong (scores 11–14), moderate (7–10), or weak (≤7).[24]

In addition to the AACPDM methodology, we used the criteria of Mallen et al.[23] to assess methodological quality and risk of bias. The Mallen score has been designed for the assessment of observational studies.[23] Of the 24 Mallen criteria, 17 were selected as they met the specific needs of this review. Again, the items were scored as 'yes', for criterion present, or 'no', for criterion not present. The number of items fulfilling the criterion resulted in the Mallen scores (Table SIII, online supporting information).

Table I Sixteen included group design studies: methodology assessment according to the AACPDM (revision 1.2)[21]

Study	Research design	Level of evidence[a]	AACPDM conduct questions[b]							Quality score	Quality summary
			1[c]	2[c]	3	4[c]	5	6[c]	7[c]		
Ryan et al.[25]	A within-subject ABA design	IV	y	n	y	n	y	y	n	4	Moderate
McDonald and Surtees[26]	Cohort study of case series, experimental design, 2´	IV	y	y	y	n	n	y	n	4	Moderate
McDonald and Surtees[27]	Case series, experimental design, 2´	IV	y	y	y	n	n	y	n	4	Moderate
Ekblom and Myhr[28]	Experimental design, 2x4 factorial	IV	y	y	y	n	n	y	n	4	Moderate
Rigby et al.[29]	A within-subject ABA design	IV	n	n	y	n	y	y	n	3	Moderate
Holmes et al.[30]	Case series with three conditions	IV	y	y	y	n	n	n	n	3	Moderate
Cimolin et al.[31]	Case series, experimental design, two-sessions	IV	n	y	y	n	n	y	n	3	Moderate
Pope et al.[32]	Cohort study of case series	IV	y	y	n	n	n	y	n	3	Moderate
Vekerdy[33]	Cohort study without concurrent control group	IV	y	y	y	n	n	n	n	3	Moderate
Hulme et al.[34]	Cohort study, pre–post tests	IV	y	n	y	n	n	n	n	2	Weak
Hulme et al.[35]	Cohort study of case series	IV	n	n	y	n	n	y	n	2	Weak
Ryan et al.[36]	One group, case series, pre–post tests	IV	n	y	n	n	n	y	n	2	Weak
Seeger et al.[37]	Experimental design, four sessions	IV	n	y	n	n	n	y	n	2	Weak
Myhr and von Wendt[38]	Quasi-experimental, single group, pre–post tests	IV	n	y	n	n	n	n	n	1	Weak
Lacoste et al.[39]	Expert opinion	V	n	n	n	n	n	y	n	1	Weak
Stewart and McQuilton[40]	Expert opinion	V	n	y	n	n	n	n	n	1	Weak

[a]Sackett's level of evidence. [b,c]See Table SI (online supporting information) for the full description of the AACPDM criteria including their contribution to the evaluation of the risk of bias. Methodological quality is judged as strong ('yes' score on six or more questions), moderate (score 3–5) or weak (score ≤2). AACPDM, American Academy for Cerebral Palsy and Developmental Medicine; ABA (research design), baseline–intervention–baseline study design; y, yes; n, no.

The contents of the studies were summarized on the data extraction form, focusing on descriptors of study design, population, participant characteristics, specific elements of the AdSS, outcome measures, results, and conclusion (MA and MH-A). Attention was paid to age, type and severity of CP (GMFCS levels IV and V), the specific components of the AdSS, and outcomes of interest. For the description of the type of CP we used the SCPE classification,[42] and for severity either the GMFCS[4] or the classification 'severe or non-ambulatory', which were the descriptions used in studies dating before the introduction of the GMFCS.[43]

We focused on the effect of (i) hip- and pelvis-stabilizing devices and (ii) trunk support devices, as theoretically these elements are regarded as essential in the achievement of a 'functional sitting posture'.[5,12,13] The details of the postural support devices were specified: support for neck, trunk, and hip and pelvis; hip abduction (use of abductor wedge); support for knees and feet; and use of a table in front.

Results

Study selection

Of the 92 studies yielded in the search strategy, 36 met the eligibility criteria. A flow chart of selection process is shown in Figure 1. The full texts of these 36 articles were retrieved and examined in more detail. We did not exclude studies using a single-subject research design (SSRD) as recommended by the AACPDM.[21,24] Seventeen original studies[25–41] met all inclusion criteria. These 17 articles were reviewed in detail with two aims: (1) to assess methodological quality and (2) to evaluate the specifics of the AdSSs and their effects on the functions of children at all levels of the ICF-CY. The evaluative procedures in following sections reflect the PICOS approach (Participants, Interventions, Comparators, Outcomes, Study design), as emphasized in the PRISMA (Preferred Reporting Items for Systemic Reviews and Meta-analyses) statement[44] for reporting systematic reviews.

Methodological assessment

Level of evidence and methodological quality

The research designs, Sackett's levels of evidence,[20] and the AACPDM[21] methodological quality scores are shown in Table I and Table SII (online supporting information). Of the 17 studies included in the methodological quality assessment, 16 used a group design[25–40] and one used a SSRD.[41] Of the 16 group design studies, 14[25–38] were classified as level IV and two[39,40] as level V evidence (Table I). The only SSRD study[41] was rated as evidence level III (Table SII, online supporting information). Even though the AACPDM Treatment Outcomes Committee[21] recommends applying the AACPDM quality assessment only for studies with at least level III evidence, we decided that an analysis of the methodological quality of all included studies was warranted in order to provide the reader with a critical synthesis of the limited evidence available (see Tables I, SI and SII [online supporting information]). The evaluation using the AACPDM criteria (Table SI) showed

that 9 out of the 16[25–33] group design studies were of moderate methodological quality, whereas the remaining seven studies[34–40] were of weak methodological quality (Table I). The methodology of the SSRD study[41] was interpreted as weak (Table SII). We excluded the eight studies[34–40,41] with weak methodological quality, leaving nine studies[25–33] to be included in the in-depth evaluation of the systematic review (Fig. 1).

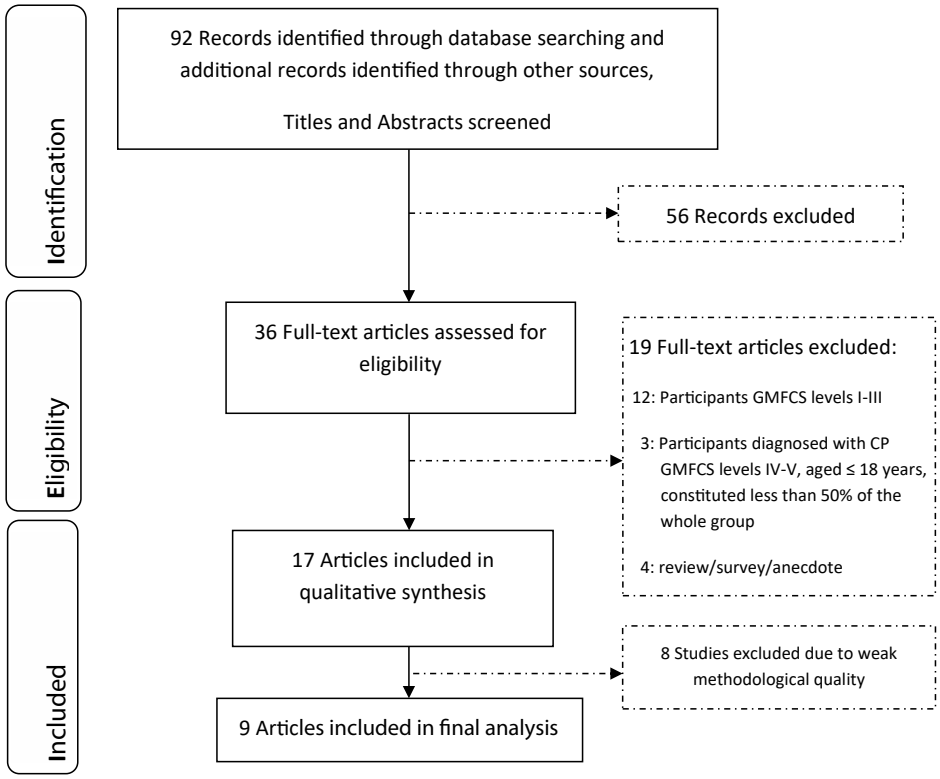

Figure 1 Inclusion and exclusion of studies found in the search strategy.

Extended methodological quality and risk of bias

In order to get more insight into the methodological problems in the field, we also applied the adapted Mallen methodological quality evaluation.[23] The Mallen score of the nine selected studies varied from 7 to 11 points. Criteria which all or most studies fulfilled (≥7/9 studies) were description of losses and completers (all studies), description of clear inclusion and exclusion criteria, description of the intervention, description of the outcomes, the numerical description of important outcomes, description of the main findings, and a conclusion supported by findings.

Criteria which were never (0/9 studies) or infrequently (< 3/9 studies) fulfilled were: reliable assessment of disease state (i.e. it was not reported how CP was diagnosed) (never), grading level of evidence (never), blinding (never), and a description of the type of study (infrequent) (Table SIII, online supporting information).

The evaluation of methodological quality according to the AACPDM[21] and Mallen[23] also well represents and reflects the evaluation of 'risk of bias' as recommended by PRISMA[44] (see Tables I, SI–III [online supporting information]). The most common markers for risk of bias in the nine studies were the absence of a reliable and standardized assessment of the disease state (all studies), the absence of masked assessors (all studies), and no controlling for confounding variables (all studies; for example, studies did not control for the presence of severe cognitive and visual impairments or dyskinesia). Most studies did take into account the potential risk of bias due to unclear presentation of inclusion and exclusion criteria (in general: clear inclusion and exclusion criteria), unclear presentation of losses and completers (in general: clear presentation of losses and completers), selective attrition (in general: little attrition), type of outcome measures (in general: valid measures), unclear presentation of main findings (in general: clear presentation), and funding resources (in general: disclosed).

Contents of the studies

Study characteristics and main outcomes of interest

Table SIV (online supporting information) summarizes the design, level of evidence, methodological quality, and demographics of the nine selected studies. The selected studies involved 134 children with CP and five typically developing children (a control group) with an age ranging from 2 to 21 years. Most children were studied at school age, that is they were aged 6 to 12 years[25–27,29–33] (GMFCS age band IV). In five studies,[25,28,29,32,33] children below the age of 6 years were also studied. Fifty-three children were included in two studies: 30 children were studied by Ryan et al.[25] and Rigby et al.[29] and 23 were assessed by McDonald and Surtees.[26,27] All but one study[28] applied single-group designs in which participants served as their own control.

The severity of CP was well specified in seven studies,[26–28,30–33] and moderately well specified in two studies,[25,29] implying that a group of children with moderate to severe CP (GMFCS levels III and IV) was studied.[25,29] The type of CP was clearly specified in five studies;[26,28,30–32] most children included had bilateral spastic CP. The type of CP was not clearly specified in the remaining four studies.[25,27,29,33] McDonald and Surtees[27] included in their study group children with bilateral CP and/or dyskinetic CP, whereas Verkerdy[33] studied a group of children with either unilateral or bilateral spastic CP. In the two papers of Ryan, Rigby and colleagues,[25,29] the type of CP was not clearly stated. Four studies[26,27,31,33] included children with dyskinetic CP, of which one[31] dealt with children with dyskinetic CP only. No specific information was present on children with ataxic CP.

Table II, III, IV, and Table SIV (online supporting information) show that the studies varied substantially in the intervention evaluated and the methods used to assess the effects. The extraction suggested that outcomes could be organized best into three main categories: postural control, upper extremity function, and additional outcomes (such as quality of life, child comfort,

parents'/caregivers' experience, and daily life performance). All outcomes were specified according to the ICF-CY framework (body functions and structures, activity, participation).[9] There follows, below, a qualitative synthesis of the main results guided by the questions addressed in this study.

Effect of specific components of adaptive seating devices

The seating systems often included supports at the level of the foot, pelvis, and trunk, but infrequently at the level of the neck. Three studies[26,27,30] applied 'knee blocks' as part of the so-called CAP-II modular seating system. Only two studies[28,32] assessed the effect of AdSSs when a table was placed in front of the child.

The duration of the AdSS intervention and that of the measurement of the effect varied across the studies from instantaneous effects measured over a few seconds or minutes to studies that followed the children for months or years.

Effect of specific components of adaptive seating devices on postural function

Seven studies[26–28,30–33] addressed the effect of a specific postural device in combination with an AdSS on postural control outcomes. All but one study[33] evaluated outcomes at the level of body functions and structures only (Table II).

Effect of a hip- and pelvis-stabilizing device

Three studies[26–28] evaluated the effect of hip- and pelvis-stabilizing devices; two[26,27] of these examined knee blocks and a sacral pad, while the other[28] evaluated a hip abduction orthosis without knee blocks. No study reported a significant effect of the device on postural function.

Effect of a trunk and hip support device

Three studies[30,31,33] evaluated the effect of a trunk and hip support device. Holmes et al.[30] and Cimolin et al.[31] reported a positive effect of trunk and hip support. Holmes et al.[30] suggested that asymmetrical lateral supports, arranged in a '3-point force system', were associated with a more symmetrical trunk posture, but without affecting pelvic obliquity in children with non-ambulatory CP who had scoliosis. The study of Cimolin et al.[31] in children with severe dyskinetic CP indicated that the presence of a dynamic back rest (including neck support) was associated with less extensor thrust. Two other studies[32,33] did not report a significant effect of a trunk and hip support device – those were the two studies of the lowest methodological quality as reflected by the Mallen scores. The study of Pope et al.[32] did not report a significant effect of an entire seating system (the Seating and Mobility system [SAM] including a forward tilted saddle seat). The study of Vekerdy[33] was not able to demonstrate an effect of the TLSO-SIDO® device in correcting scoliotic curves in children with severe CP.

Table II Effect of specific components of adaptive seating systems on postural control outcomes

	Adaptation tested							Duration				
		Support during sitting										
Study	Adaptive seating conditions	Ta	N	Tr	H	Aw	Kn	F	Measurement	Intervention	Postural function tests	Result	ICF-CY
McDonald and Surtees[26]	CAP-II seating system, without [E]/ with [C] knee blocks and sacral pads	–	+	+	+	+ S	+ S[a]	+	±3 measures × 80s	4wk	1. PC, postural alignment (direct observation, modified SPCM) 2. Force on knee blocks 3. Pressure on sacral pads	E = C E = C E = C	BS/F
McDonald and Surtees[27]	CAP-II seating system, without [E]/ with [C] knee blocks and sacral pads	–	+	+	+	+ S	+ S[a]	+	±3 measures × 80s	4wk	PC, postural alignment (direct observation, modified SPCM)	E = C	BS/F
Ekblom and Myhr[28]	Standard chair with hip AO at three spring lengths [E] and without hip AO [C]	+	–	–	+	+ S	–	+	4 conditions × 3 min	4 conditions × 3min	1. PC head, trunk, foot (video observation, SAS) 2. PC EMG on AL, RF, GA, ES	E = C E = C	BS/F
Holmes et al.[30]	CAP-II modular seating system; with symmetric axillar support [E1], with asymmetric lateral support [E2], or with bilateral pelvic support [C]	–	–	+ S	+ S	+	+	+	3 measures × ±30s	3 measures × ±30s	PC Kine: (1) spinous process angle correction, (2) pelvic obliquity, (3) symmetry of forces applied on supports	E1 = C, E2 > C E = C E2 > E1	BS/F
Cimolin et al.[31]	X-PANDA with dynamic back rest [E] and rigid back rest [C]	–	+	+ S	+ S	+	–	+	2 conditions × 6min	2 conditions × >6min (after the subject familiarized with AdSS, the test began)	PC Kine, extensor thrust: (1) head anterior–posterior, (2) head vertical, (3) trunk anterior–posterior, (4) trunk vertical Kinetics: (1) head; (2) trunk	 E > C E = C E > C E < C E = C E < C	BS/F
Pope et al.[32]	SAM systems with saddle seat including forward tilting of a seat surface	+	–	+	+	+	–	+	?	3y follow-up	PC, postural ability (photograph)	E = C?	BS/F
Vekerdy[33]	AdSS with TLSO-SIDO®	–	–	+ S	+ S	–	–	+	<1s	4–18mo (mean 12.7mo SD 30.1mo)	Cobb angle of (1) trunk kyphosis, (2) lumbar lordosis Q; Changes in posture and changes in ADL	 E = C E = C sam	BS/F A

±, approximately; AdSS, adaptive seating system with or without mobility base. Although most studies addressed the effects of specific components (indicated by 'S' in all column 'Support during sitting'), some studies evaluated a whole complex system without a specific focus on seating components (no 'S' indicated).
Adaptation tested: C, control group; E, experimental group; asymmetrical lateral support of trunk using the three-point force system at axilla, apex of the scoliosis, and pelvis; symmetrical axillary supports at two sides of both axillae and pelvis; hip AO, hip abduction orthosis; TLSO-SIDO®, thoracic–lumbar–sacral orthosis with non-rigid SIDO® frame (this frame served as postural support for the child to be able to move his/her trunk in the TLSO).
Supports during sitting: Ta, table; N, neck; Tr, trunk; H, hip; Aw, abduction wedge; Kn, knee blocks; F, foot; –, without support; +, with support; ±, with and without support (a support can be detached); ?, no detail/unclear information; S, the level of support being evaluated in the study. [a]Study focused on the effect of the *removal* of the knee blocks.
Outcome measurements: ADL, activity daily living. Muscles for EMG test: AL, adductor longus; ES, erector spinae; GA, gastrocnemius; RF, rectus femoris; PC, postural control; PC EMG, postural control measured by means of surface electromyography; PC Kine, postural control measured by means of kinematics; Q, questionnaire; SAS, sitting assessment scale; SCPM, seated postural control measure.
Result: A, activity domain; BS/F, body structure and function domain; P, participation domain; ICF-CY, International Classification of Functioning, Disability and Health, Child and Youth version; C, control group/ conditions; E, experimental group/conditions; E1, experimental condition 1; E2, experimental condition 2; E > C, experiment group is better than control group; E < C, experiment group is worse than control group; E = C, no significant difference between experiment and control group;?, no detail or result was not clear; sam, statistical analysis missing.

Effect of specific components of adaptive seating devices on upper extremity function

Four studies[28,29,31,32] evaluated the effect of various postural support devices in combination with an AdSS on upper extremity function (Table III).

Effect of a hip- and pelvis-stabilizing device

Ekblom and Myhr[28] evaluated the effect of a hip abduction orthosis. The study could not demonstrate an effect of the orthosis on upper extremity function, either in the domain of body functions or in the activities domain.

Effect of a trunk support device

Cimolin et al.[31] evaluated the effect of a trunk support device on upper extremity function. The results suggested that, in children with severe dyskinetic CP, the X-PANDA seating system with a dynamic back rest was associated with more involuntary arm movements than the X-PANDA seating with a rigid back rest. Apparently the dynamic back rest was associated with better head and trunk stability at the expense of increased involuntary arm movements.

Finally, two studies[29,32] assessed the effect of an AdSS that provided postural support at the trunk and hip level without focusing on specific AdSS components. Rigby et al.[29] demonstrated that specific-purpose AdSSs, the Flip2sit and the Aquanaut toilet system, were associated with a positive effect on self-care and play behaviour as measured with the Canadian Occupational Performance Measure (COPM).[45] The other study, by Pope et al.,[32] did not allow for pertinent conclusions as it lacked statistical analysis.

Effect of adaptive seating devices on additional outcomes

Three studies[25,29,32] paid attention to outcomes outside the domains of postural control and upper extremity function (Table IV).

Effect of a hip- and pelvis-stabilizing device.

None of the studies evaluated the effect of a hip- and pelvis-stabilizing device on additional outcomes.

Table III Effect of specific components of AdSS on upper extremity function outcomes

	Adaptation tested								Duration			
		Supports during sitting										
Study	Adaptive seating conditions	Ta	N	Tr	H	Aw	Kn	F	Measurement	Intervention	Upper extremity tests	Result	ICF-CY
Ekblom and Myhr[28]	Standard chair with hip AO at three spring lengths [E], and without hip AO [C]	+	−	−	+	±S	−	+	4 conditions ´	4 conditions ´	Video observation; SAS (arm–hand movement control)	E = C	BS/F, A
Rigby et al.[29]	1. Flip2sit activity seat, 2. Aquanaut toilet system for postural sitting supports; home activities	±?	±?	±?	±?	±?	±?	±?	6wk	6wk	COPM: (1) self-care, (2) play, (3) socialize/recreation	E > C E > C E = C	A, P
Cimolin et al.[31]	X-PANDA with dynamic back rest [E] and rigid back rest [C]	−	+ S	+ S	+	+	−	+	2 conditions × ±6min	2 conditions × >6min (after the subject familiarized with AdSS, the test began)	Kinematic measurement of involuntary movements of upper extremities: (1) excursion, (2) jerk	E < C E < C	BS/F
Pope et al.[32]	SAM systems with saddle seat including forward tilting of a seat surface	+	−	+	+	+	−	+	?	3y follow-up	Video observation; quality of arm–hand function (manipulating blocks)	sam	A

Supports during sitting: Ta, table; N, neck; Tr, trunk; H, hip; Aw, abduction wedge; Kn, knee blocks; F, foot; –, without support; +, with support; ±, with and without support (a support can be detached); ?, no detail/unclear information; S, the level of support being evaluated in the study.
Outcome measurements: COPM, Canadian Occupational Performance Measure; Q, questionnaire; SAS, sitting assessment scale.
Result: A, activity domain; BS/F, body structure and function domain; P, participation domain; ICF-CY, International Classification of Functioning, Disability and Health, Child and Youth version; E, Experimental group/conditions; C, control group/conditions; E > C, experiment group is better than control group; E < C, experiment group is worse than control group; E = C, no significant difference between experiment and control group;?, no detail or result was not clear; sam, statistical analysis missing.

Effect of a postural support device

Three studies[25,29,32] addressed the effect of postural sitting supports that included trunk and hip support but did not address the effect of AdSS components. Pope et al.[32] did not find a significant effect of the SAM seating system applied for 3 years on the range of motion of hip and knees and on daily activities. The other two studies[25,29] had a better methodological quality than the Pope et al. study.[32] Both studies reported a beneficial effect of specific-purpose AdSSs. Ryan et al.[25] reported a positive effect of the Flip2sit and the Aquanaut toilet system on parental views about the impact of the AdSS in daily life. The qualitative study of Rigby et al.[29] underlined the positive effect of the specific seating systems of the Ryan et al. study.[25]

Effect of severity and type of CP

The effect of the severity of CP (GMFCS level IV vs V) on postural function, upper extremity function, and additional outcomes could not be assessed as the studies did not supply sufficient information to allow for this evaluation. The same held true for the effect of the type of CP. The studies either did not include sufficient information on the type of CP[25,27,29,33] or included only children with one type of CP.[28,30–32] Only the study of McDonald and Surtees[26] specified the presence of two types of CP in the study group. However, these authors did not address whether the type of CP modified the effect of the intervention.

Effect of adaptive seating devices across three domains of the ICF-CY

Seven studies[26–28,30–33] addressing the effect of intervention in terms of postural function (Table II) primarily focused on outcome in the domain of body functions and structure. An exception was the study of Vekerdy,[33] which included outcome in terms of activities. Only two[30,31] of the seven 'postural' studies found a positive effect of an AdSS component on postural function in the domain of body function and structure. These studies were two out of the three studies[30,31,33] that evaluated the effect of a trunk and hip support device. The third study[33] that did not find a significant effect of a trunk and hip support device was the study with the lowest methodological quality score.

Three of the four studies[28,29,32] that addressed outcome in terms of upper extremity function (Table III) evaluated outcome in the activity domain. Only the study by Rigby et al.[29] demonstrated a positive effect of a seating device on upper extremity function. This study evaluated the effect of an entire seating system with a well-validated outcome measure, the COPM. The study by Cimolin et al.[31] was the only one that evaluated outcomes at the body function and structure level. It found that use of a dynamic back rest in children with dyskinetic CP was associated with an increase in involuntary arm movements.

Table IV Additional outcomes

	Adaptation tested								Duration				
		Supports during sitting											
Study	**Adaptive seating conditions**	**Ta**	**N**	**Tr**	**H**	**Aw**	**Kn**	**F**	**Measurement**	**Intervention**	**Upper extremity tests**	**Result**	**ICF-CY**
Ryan et al.[25]	AdSSs (Flip2sit activity seat and Aquanaut toilet system) for sitting support and home activities	±?	±?	±?	±?	±?	±?	±?	6wk	6wk	FIAT (parents' views on impact of ASS in daily life)	E>C	A, P
Rigby et al.[29]	AdSSs (Flip2sit activity seat and Aquanaut toilet system) for sitting support and home activities	±?	±?	±?	±?	±?	±?	±?	6wk	6wk	Home activity log; parents' views on impact of AdSS	sam	A, P
Pope et al.[32]	SAM systems with saddle seat including forward tilting of a seat surface	+	-	+	+	+	–	+	?	3y follow-up	ROM hip and knee ADL (feeding, computer activities)	E=C? sam	BS/F A

Supports during sitting: –, without support; +, with support; ±, with and without support (a support can be detached); ?, no detail/unclear information; S, the level of support being evaluated in the study.
Outcome measurements: ADL, activity of daily living; FIAT, Family Impact of Assistive Technology Scale; ROM; range of motion.
Result: A, activity domain; BS/F, body structure and function domain; P, participation domain; ICF-CY, the international classification of functioning, disability and health-Child and Youth version; CP type: BS, bilateral spastic; US, unilateral spastic; E, Experimental group/conditions; C, Control group/conditions; E>C, experiment group is better than control group; E<C, experiment group is worse than control group; E=C, no significant difference between experiment and control group;?, no detail or result was not clear; sam, statistical analysis missing.

The three studies[25,29,32] that assessed the effect of AdSS on additional outcomes focused primarily on outcome in the activity domain (Table IV). Two[25,29] of them also evaluated outcome in terms of participation, and one[32] also addressed outcome in terms of body function and structure. Again, the study[25] that evaluated the effect of an entire specific-purpose AdSS with a well-validated outcome measure, the Family Impact of Assistive Technology (FIAT),[46] was the study that reported a positive effect of the seating intervention. Four of the five studies[25,28,29,33] that evaluated outcome in the activity or participation domain had been published after the introduction of the ICF in 2001, of which two[25,29] were published after the introduction of the ICF-CY in 2007.

Discussion

This systematic review aims to critically evaluate and update the evidence on whether or not AdSSs positively affect postural control, upper extremity function, and additional outcomes in children with severe CP across the three ICF-CY domains (body functions and structure, activity, and participation). The review revealed that the nine methodologically best studies still had a low level of evidence with a moderate and heterogeneous methodological quality. This means that it is currently impossible to draw robust conclusions about the functional effect of seating devices in children with severe CP.

None of the nine studies reviewed in detail reported significant adverse side effects of the AdSSs studied. Nevertheless, caution is warranted. For example, children with scoliosis in GMFCS levels IV or V might function better with a postural support device counteracting the asymmetry of scoliosis, but this type of specific AdSS may increase the uneven pressure distribution on the skin or may cause stress and pain as a result of the forces induced by the AdSS.[6]

The studies did not provide sufficient information to evaluate a potential effect modification depending on the severity and the type of CP. In addition, most studies were of children of school age, with only a minority addressing the effect of AdSSs before school age. It is, however, conceivable that the effect of AdSSs at an early age is greater than that at later ages,[1,8,47] as the nervous system has a higher plasticity at an early age.[48] Therefore, we recommend that future studies address the effect of AdSSs on activity and participation in young children with severe CP.

In the following paragraphs we address methodological considerations, the outcomes and the effects of specific components of the AdSS in view of the framework of the ICF-CY, and the limitations of our review. We end with concluding remarks and perspectives for future research.

Methodological considerations

The Mallen assessment revealed that none of the nine studies evaluated had 'participants representative of the population' and, therefore, the findings of the studies cannot be generalized to the entire (sub)group of children with CP who are classified in GMFCS level IV or V. The analyses of methodological quality according to the AACPDM and Mallen also allowed us to evaluate the risk of bias in the individual studies. Some types of bias reflect the difficulties encountered in this field.

First, it is not easy for masked assessors to assess children: often the assessor is aware of the seating condition. One option available to prevent this assessor bias would be the use of video assessment in which – during the analysis – critical parts of the seating system are covered. Another option – applicable only for the analysis of outcome at the body function and structure level – would be the masked analysis of physiological parameters, such as kinematics or electromyography.

A second form of bias is the confounding by comorbidity, such as visual and cognitive impairment. Although comorbidities are inevitable in children with severe CP, this form of bias might be overcome by multicentre trials allowing for adjustment for specific sensory limitations, degree of cognitive impairment, and presence and severity of epilepsy. This requires that this information is documented in a standardized way.[1,2,7]

A third problem encountered is the limited number of standardized, validated, and reliable assessment procedures available for the evaluation of function in children with severe CP. Although most studies included in the review used valid measures, the sensitivity and responsiveness to change of many measurements were problematic. For example, the relatively insensitive direct observation methods applied in some studies[26,27,32] may have missed subtle changes in child function. Ryan[12] suggested that 'measurement scales must have adequate levels of reliability, validity and be responsive to meaningful changes in dimensions of the ICF-CY'.[12] Quantitative, computer-based video analysis including a standardized assessment protocol observation[49] may pave the way for dedicated analysis of outcome in all domains of the ICF-CY.[6,9] Another available tool is Goal Attainment Scaling (GAS).[50] This tool has been successfully applied to document effect of intervention in children with milder forms of CP.[51,52] GAS can potential document changes in activities and participation of children with severe CP.[50–52]

In this difficult area of research, the prospective cohort, the mixed-methods design involving quantitative and qualitative methods, single-subject research methodologies and qualitative inquiries with a rigorous methodology presumably are the most appropriate approaches.[7,9,12,24,53] We urge that efforts are made to improve the methodological quality in this area: may the severity of CP not be an excuse for poor methodological quality.

Outcomes and the framework of the ICF-CY

Body functions and structures

The studies[26–28,30–33] mostly addressed outcome in terms of postural control (correction of the alignment of the spinal deformities and correction of the alignment of the pelvis, hips, and knees, including a windswept posture) or in terms of upper extremity control outcomes (the control of arm–hand movements and involuntary movements). Most studies were not able to provide evidence of a beneficial effect of the seating system on postural control outcomes or upper extremity function. Nevertheless, two findings on the effect of trunk and hip support devices[30,31] are noteworthy: (1) the use of the CAP-II seating system with three-point lateral trunk supports[30] significantly improved symmetrical trunk posture in children with severe CP with scoliosis; and (2) the use of the X-PANDA seating system combined with a dynamic back rest[31] in children with

dyskinetic CP was associated with improved head and trunk stability in association with reduced extensor trust, but at the expense of an increase in involuntary arm movements. However, caution in the interpretation of these findings is needed. The findings cannot be generalized, as the AdSS interventions[30,31] were based on short-lasting evaluations of the child's capacity in experimental conditions.[11,12,56] Short-lasting effects on capacity may differ from long-lasting effects on performance in daily life.[11,12,56]

Another important issue, which none of the studies addressed, is the evaluation of comfort in a specific AdSS in children with CP who have severe communicative and cognitive impairments. Various research strategies may be applied here. One solution might be the use of long-lasting (e.g. an entire day) pressure distribution measurements at the seating interface. A second option is the use of the Caregiver Priorities & Child Health Index of life with Disabilities (CPCHILD) questionnaire,[54] a novel instrument to use with children with severe CP to evaluate the child's comfort, the ease of care, health, and quality of life.[54] Third, the previously mentioned method of quantitative video analysis[49] may be used to evaluate the child's non-verbal behaviour, using scoring systems similar to the ones used in the evaluation of pain in newborn infants.[55]

Activity

The studies addressing outcome in terms of activities dealt with prehension, self-care, and play. The more recent ones, which evaluated specific-purpose AdSS, indicated that the AdSSs were associated with improvements in these activities in the daily life situation.[25,29] Activities and participation are – as the ICF-CY framework highlights – strongly dependent on the child's context, such as the family and school.[6,9–11] We therefore suggest that future studies on AdSSs use a battery of tools to assess the effect of AdSS on daily life activities, including the assessment of family wishes and burden.[6,9,11]

Participation

Currently, optimization of home and community participation is the major goal of paediatric rehabilitation services, including AdSS intervention.[7–11] Only two studies[25,29] supplied valid information on outcomes in both activity and participation domains. Of these, the study[25] with the higher methodological quality reported a beneficial effect of the AdSS on the children's play, but not on their socialization and recreation. This study suggests that it may be possible to improve participation in children with severe CP at home, at school, and in the community by means of the specific 'immediate environment'[2,9,47] formed by the AdSS. Future studies on the effect of AdSS on participation in children with severe CP are urgently needed.

Evaluation of participation is not possible in a laboratory setting.[8,11,56] The best options available are questionnaires and interviews. In young children and in children with limited communicative ability, a proxy respondent, e.g. the primary caregiver, is an acceptable alternative to the child.[7,9,53] The tools available are the COPM[45], GAS,[50–52] and two novel tools developed for children with severe disability, the CPCHILD[54] and the novel version of the FIAT, the Family Impact of Assistive Technology Scale for Adaptive Seating (FIAT-AS).[53]

Effect of severity and type of cerebral palsy on the effect of adaptive seating devices

Presumably the putative effect of adaptive seating depends, in particular, on the severity of CP. The limited evidence available suggests that children with CP in GMFCS level V lack the basic level of postural control, and that in children in GMFCS level IV this basic level of control is present with impairments.[5] The benefit of hip, pelvis, and trunk support devices in children classified in GMFCS level IV or V presumably is primarily mediated by the provision of support, which reduces the number of degrees of freedom which have to be controlled, thereby reducing the demand of the postural control task.[5] In the current review we were not able to assess the effect of severity of CP (GMFCS level IV vs V) on the effect of AdSSs owing to a lack of detail in the studies. Similarly, we were unable to assess the effect of the type of CP on the effect of AdSSs on outcome, as insufficient information was available. Theoretically, the AdSSs that may be beneficial for children with dyskinetic CP might differ from those that may be most appropriate for children with spastic CP, as the postural problems associated with the two types of CP are very different.[3,5]

Strengths and limitations of this review

It is generally recognized that research in this area is notoriously difficult.[5,6] This is reflected by the low grades of evidence of the studies included in the review. This low grade of evidence is a serious limitation of this study. However, rather than casting away all low-level evidence, we systematically reviewed the material available. To this end we adapted the standard systematic review protocol, as we considered it to be too restricted for our purpose; we applied a triple methodological quality assessment. The detailed methodological analysis may be regarded as the strength of the study. It allowed us to highlight the difficulties and risks of bias in the field. In addition, it offered opportunities to suggest solutions and strategies for future research. The second strength of the review is its systematic and structured organization. Not only did we apply a rigorous methodological evaluation, we also organized outcomes systematically, that is according to functional domain (postural function, upper extremity function, and additional outcomes) with special attention to the components of the ICF-CY. Nevertheless, it is debatable whether our selection was the optimal one for synthesizing the best evidence on this topic.

The most serious limitation of our review is that it is based on studies with a low level of evidence. The second limitation is related to this first limitation. We slightly adapted the criteria of the AACPDM[21] score for moderate methodological quality in order to preserve a sufficient number of studies in the review. This is a choice that may be debated. Finally, the heterogeneity of the studies precluded the performance of a meta-analysis to balance overall outcomes across studies.

Conclusion

Adaptive seating systems constitute important assistive devices in the daily life of children with severe CP who are classified in GMFCS level IV or V. The current systematic review showed that the studies available on the effect of AdSSs have a low level of evidence and are of moderate method-

ological quality. The low level of evidence precludes pertinent conclusions on the effectiveness of AdSSs in children with severe CP. This reflects how little we know in this field. However, the limited evidence available suggests that special-purpose AdSSs may improve activities and participation.

The limited evidence emphasizes the urgent need for studies of high methodological quality. Our review has discussed the many difficulties that studies in this area may encounter. Nevertheless, we think that time and tools are ready for new studies that may unravel which components of AdSS are effective promoters of participation in children with severe CP. We have summarized our suggestions for future research in Table SV (online supporting information).

Acknowledgements

MA was supported financially by the Naresuan University, Phitsanulok, Thailand, and the Graduate School for Behavioural and Cognitive Neurosciences, University of Groningen.

The authors have stated that they had no interests that might be perceived as posing a conflict or bias.

Supporting information

Additional supporting information may be found in the online version of this article:

Table SI: Conduct questions with item criteria for quality assessment; adapted from AACPDM Quality Assessment Scale for group study.
Table SII: One included single subject design study: methodology assessment according to the AACPDM (revision 1.2).
Table SIII: Extended methodological quality criteria adapted from Mallen et al.[23] in nine studies included in the final analyses.
Table SIV: Summary of nine selected studies for review.
Table SV: Suggestions for future directions of studies on the effect of AdSS in children with severe CP across the domains of the ICF-CY.

Supporting information (online)

Table SI Conduct questions with item criteria for quality assessment; adapted from AACPDM Quality Assessment Scale[21] for group study

AACPDM conduct question	Specification of criteria
1. Were inclusion and exclusion criteria of the study population well described and followed? Both inclusion and exclusion criteria need to be met in order to score 'yes'*	1. Inclusion should at least be specified with (a) diagnosis of CP, (b) type of CP, (c) severity of CP, and (d) age of participants 2. Exclusion criteria should be mentioned
2. Was the intervention well described and was there adherence to the intervention assignment? (for two-group designs, was the control exposure also well described?) Both parts of the question need to be met in order to score 'yes'*	'Well described' implies that the intervention can be replicated on the basis of the description in the study
3. Were the measures used clearly described, valid, and reliable for measuring the outcomes of interest?	1. The primary outcome measure should be clearly described, valid. *and* reliable 2. If additional data on postural control or upper extremity function are provided, these outcome measures are also rated in terms of clarity of description, validity, and reliability
4. Was the outcome assessor unaware of the intervention status of the participants (i.e. were the assessors masked)?*	Clear description
5. Did the authors conduct and report appropriate statistical evaluation including power calculations? Both parts of the question need to be met in order to score 'yes'	Clear description
6. Were dropout/loss to follow-up reported and less than 20%? For two-group designs, was dropout balanced?*	Clear description
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?*	The presence of following confounders should have been taken into account: (a) severe visual impairment, (b) severe cognitive impairment, and (c) dyskinesia

*Conduct questions that represent an evaluation of risk of bias within studies. AACPDM, American Academy for Cerebral Palsy and Developmental Medicine; CP, cerebral palsy.

Table SII One included single subject design study: methodology assessment according to the AACPDM (revision 1.2)[24]

Study	Research design	Level of evidence	AACPDM conduct questions														Conclusion
			1	2	3[a]	4	5	6[a]	7	8	9	10	11	12	13	14	
Hulme et al.[41]	AB design, SSRD	SSRD III	n	n	n	y	y	y	y	y	n	y	n	n	n	n	6, Weak

Methodological quality is judged as strong (score 11–14), moderate (7–10), or weak (≤7). [a]An evaluation of risk of bias. SSRD, single subject research design. AACPDM, American Academy for Cerebral Palsy and Developmental Medicine.

Conduct questions for single-subject design studies (y = yes; n = no)

1. Was/were the participant(s) sufficiently well described to allow comparison with other studies or with the reader's own patient population?
2. Were the independent variables operationally defined to allow replication?
3. Were intervention conditions operationally defined to allow replication?
4. Were the dependent variables operationally defined as dependent measures?
5. Was inter-rater or intra-rater reliability of the dependent measures assessed before and during each phase of the study?
6. Was the outcome assessor unaware of the phase of the study (intervention vs control) in which the participant was involved?
7. Was stability of the data demonstrated in baseline, namely lack of variability or a trend opposite to the direction one would expect after application of the intervention?
8. Was the type of SSRD clearly and correctly stated, for example A-B (baseline – intervention), multiple baselines across subjects?
9. Were there an adequate number of data points in each phase (minimum of five) for each participant?
10. Were the effects of the intervention replicated across three or more subjects?
11. Did the authors conduct and report appropriate visual analysis, for example level, trend, and variability?
12. Did the graphs used for visual analysis follow standard conventions, for example x- and y- axes labelled clearly and logically, phases clearly labelled (A, B, etc.) and delineated with vertical lines, data paths separated between phases, consistency of scales?
13. Did the authors report tests of statistical analysis, for example celeration line approach, two-standard deviation band method, C-statistic, or other?
14. Were all criteria met for the statistical analyses used?

Table SIII Extended methodological quality criteria adapted from Mallen et al.[23] in nine studies included in the final analyses

Mallen quality criteria	Ryan et al.[25]	McDonald and Surtees[26]	McDonald and Surtees[27]	Ekblom and Myhr[28]	Rigby et al.[29]	Holmes et al.[30]	Cimolin et al.[31]	Pope et al.[32]	Vekerdy[33]
Accurate and appropriate outcome/ intervention measures in all participants	n	y	y	y	n	y	y	n	y
Appropriate statistical tests used	y	y	y	n	y	y	y	n	n
Blinding[a] (participants/subjects)	n	n	n	n	n	n	n	n	n
Participants characteristics described	n	y	n	y	n	n	n	y	n
Numerical description of important outcomes given	y	n	y	y	y	y	y	y	y
Outcomes clearly described[a]	y	y	y	y	y	y	y	n	y
Power calculation used	y	y	n	n	y	n	n	n	n
Grading of level of evidence	n	n	n	n	n	n	n	n	n
Losses and completers described[a]	y	y	y	y	y	y	y	y	y
Reliable assessment of disease state[a]	n	n	n	n	n	n	n	n	n
Clear inclusion/exclusion criteria[a]	y	y	y	y	n	y	n	y	y
Clear hypothesis	y	y	y	n	y	n	n	n	n
Intervention described[a]	n	y	y	y	n	y	y	y	y
Type of study stated	y	n	n	n	y	n	n	n	n
Main findings described[a]	y	n	y	y	y	y	y	y	y
Disclosure of funding source[a]	y	y	y	y	y	n	n	y	n
Conclusion supported by findings	y	y	y	y	y	y	y	n	n
Total score	**11**	**11**	**11**	**10**	**10**	**9**	**8**	**7**	**7**

Adapted Mallen score: the highest possible score is 17. [a]Criteria which contributed to the evaluation of risk of bias. y, yes; n, no.

Table SIV Summary of nine selected studies for review

Study	Research design	Sackett's level	Quality of study (score)		Nr of children with CP (age range; mean age)	Younger than 6y	Nr of TD children (age range; mean age)	CP clinical characteristics						
								GMFCS level/severity			Type			
			AACPDM	Mallen criteria[23]				I–II (mild)	III (moderate)	IV–V (severe)	US	BS	Dyskinetic	Mixed
Ryan et al.[25]	Within-subject ABA design	IV	4	11	30 (2.6–7.6y)	±	0		30 (levels III and IV)[a]		?	?	?	?
McDonald and Surtees[26]	Cohort study of case series, experimental design, 2 ´	IV	4	11	23 (7.4–14.3y; 10.8y)	–	0	–	–	23	–	18	5	–
McDonald and Surtees[27]	Case series, experimental design, 2 x 2 factorial	IV	3	11	23 (7.4–14.3y; 10.8y)	–	0	–	–	23	–	23 BS or dyskinetic		–
Ekblom and Myhr[28]	Experimental design, 2 ´	IV	4	10	4 (3–5y; 4y)	+	5 (5–9y; 7y)	1	1	2	–	4	–	–
Rigby et al.[29]	A within-subject ABA design	IV	3	10	30 (2.6–7.6y)	±	0		30 (levels III and IV)[a]		?	?	?	?
Holmes et al.[30]	Case series with three session	IV	3	9	16 (6.5–20.8y; 14.7y)	–	0	?	?	16	–	16	–	–
Cimolin et al.[31]	Case series, experimental two sessions	IV	3	8	10 (6 to 10y)	–	0	–	–	10	–	-	10	-
Pope et al.[32]	Cohort study of case series	IV	3	7	9 (2.5–9y)	±	0	–	–	9	–	9	-	-
Vekerdy[33]	Cohort without concurrent control group	IV	3	7	42 (1.7–11.2y; 4.5y)	+	0	–	7	35	35 (US and BS)		7	-

[a]Studies with presumably at least 50% of children functioning at GMFCS level IV. +, majority of children studied younger than 6y; ±, inclusion of children under and over 6y; –, children included older than 6y; AACPDM, American Academy for Cerebral Palsy and Developmental Medicine methodology; TD, typically developing; GMFCS, Gross Motor Function Classification System; US, unilateral spastic; BS, bilateral spastic; ABA (research design), baseline–intervention–baseline study design.

Table SV Suggestions for future directions of studies on the effect of AdSS in children with severe CP across the domains of the ICF-CY

Study design and groups

Prospective cohort design
Mixed methods design of the quantitative and qualitative study
High-quality single-subject research methodology
Multicentre trials to increase group size, thereby allowing for adjustment for confounding by co-morbidities
Document type and severity of CP with standardized instruments, such as the SCPE and GMFCS
Mask assessors for type of intervention
Study the effect of AdSS at pre-school age

Evaluation Tools, novel directions

ICF-CY	Outcome	Assessment
Body functions and structure/ impairments	Comfort/pain levels	Pressure distribution measurements Paediatric Pain Profile CPCHILD Video-analysis of facial expressions (cf. neonatal pain assessments)
	Posture	Computer-based video analysis Kinematics EMG
Activities/activity limitations	Achievement of functional activities, mobility, self-care, and social function	GAS FIAT-AS CPCHILD Computer-based video analysis of daily activities
Participation/restriction in participation	Caregiver assistance in activities, mobility, self-care, and social function Performance of children in different settings (home, school, community)	GAS FIAT-AS CPCHILD
Environmental and personal factors	Evaluation of presence of modifications of environment	PEDI: modification scales
	Evaluation of caregiver involvement	PEDI: caregiver assistance scales
	Evaluation of child characteristics	Assessment of, for example, cognition, vision, self-perception

AdSS, adaptive seating systems with/or without its mobility base; ICF-CY, International Classification of Functioning, Disability and Health, Child and Youth version; SCPE, Surveillance of Cerebral Palsy in Europe Guideline; GMFCS, Gross Motor Function Classification System; CPCHILD, Caregiver Priorities & Child Health Index of life with Disabilities questionnaire; EMG, surface electromyography; GAS, Goal Attainment Scaling; FIAT-AS, Family Impact of Assistive Technology Scale for Adaptive Seating; PEDI, Paediatric Evaluation of Disability Inventor.

References

1. Gericke T. Postural management for children with cerebral palsy: consensus statement. *Dev Med Child Neurol* 2006; **48:** 244.
2. Østenjø S, Carlberg EB, Vøllestad NK. The use and impact of assistive devices and other environmental modifications on everyday activities and care in young children with cerebral palsy. *Disabil Rehabil* 2005; **27:** 849–61.
3. Himmelmann K, Beckung E, Hagberg G, Uvebrant P. Bilateral spastic cerebral palsy: prevalence through four decades, motor function and growth. *Eur J Paediatr Neurol* 2007; **11:** 215–22.
4. Palisano R, Rosenbaum P, Bartlett D, Livingston M. The Gross Motor Function Classification System – Expanded and Revised. CanChild Centre for Childhood Disability Research, 2007. Available from: http://www.canchild.ca/en/measures/gmfcs_expanded_revised.asp (accessed 9 April 2013).
5. Hadders-Algra M, Brogren Carlberg E. Postural Control: a Key Issue in Developmental Disorders. London: Mac Keith Press, 2008.
6. McDonald R, Surtees R, Wirz S. The international classification of functioning, disability and health provides a model for adaptive seating interventions for children with cerebral palsy. *Br J Occup Ther* 2004; **67:** 293–302.
7. Palisano R, Kang LJ, Chiarello L, Orlin M, Oeffinger D, Maggs J. Social and community participation of children and youth with cerebral palsy is associated with age and gross motor function classification. *Phys Ther* 2009; **89:** 1304–14.
8. Wiart L, Darrah J. Changing philosophical perspectives on the management of children with physical disabilities – their effect on the use of powered mobility. *Disabil Rehabil* 2002; **24:** 492–8.
9. World Health Organization. International Classification of Functioning, Disability, and Health: Children and Youth version (ICF-CY). Geneva: World Health Organization, 2007.
10. Darrah J. Using ICF as a framework for clinical decision making in pediatric physical therapy. *Adv Physiother* 2008; **10:** 146–51.
11. Rosenbaum P, Stewart D. The World Health Organization International Classification of Functioning Disability and Health: a model to guide clinical thinking, practice and research in the field of cerebral palsy. *Semin Pediatr Neurol* 2004; **11:** 5–10.
12. Ryan S. An overview of systematic reviews of adaptive seating interventions for children with cerebral palsy: where do we go from here? *Disabil Rehabil Assist Technol* 2012; **7:** 104–11.
13. Healy A, Ramsey C, Sexsmith E. Postural support systems: their fabrication and functional use. *Dev Med Child Neurol* 1997; **39:** 706–10.
14. Roxborough L. Review of the efficacy and effectiveness of adaptive seating for children with cerebral palsy. *Assist Technol* 1995; **7:** 17–25.
15. Harris S, Roxborough L. Efficacy and effectiveness of physical therapy in enhancing postural control in children with cerebral palsy. *Neural Plast* 2005; **12:** 229–43.
16. Chung J, Evans J, Lee C, et al. Effectiveness of adaptive seating on sitting posture and postural control in children with cerebral palsy. *Pediatr Phys Ther* 2008; **20:** 303–17.
17. Stavness C. The effect of positioning for children with cerebral palsy on upper-extremity function: a review of the evidence. *Phys Occup Ther Pediatr* 2006; **26:** 39–48.

18. McNamara L, Casey J. Seat inclinations affect the function of children with cerebral palsy: a review of the effect of different seat inclines. *Disabil Rehabil Assist Technol* 2007; **2:** 309–18.
19. World Health Organization. International Classification of Functioning, Disability, and Health. Geneva: World Health Organization, 2001.
20. Sackett D, Straus S, Richardson S, Rosenberg W, Haynes R. Evidence-based Medicine: How to Practice and Teach EBM, 2nd edn. Edinburgh: Churchill Livingstone, 2000.
21. Darrah J, Hickman R, O'Donnell M, Vogtle L, Wiart L. AACPDM methodology to develop systematic reviews of treatment interventions (revision 1.2) [updated 2008 Dec 17]. Available from: http://www.aacpdm.org/UserFiles/file/systematic-review-methodology.pdf; accessed 29 May2012.
22. Wiart L, Kolaski K, Butler C, et al. Interrater reliability and convergent validity of the American Academy for Cerebral Palsy and Developmental Medicine methodology for conducting systematic reviews. *Dev Med Child Neurol* 2012; **54:** 606–11.
23. Mallen C, Peat G, Croft P. Quality assessment of observational studies is not commonplace in systematic reviews: review articles. *J Clin Epidemiol* 2006; **59:** 765–9.
24. Romeiser Logan L, Hickman R, Harris S, Heriza C. Single-subject research design: recommendations for levels of evidence and quality rating. *Dev Med Child Neurol* 2008; **50:** 99–103.
25. Ryan S, Campbell K, Rigby P, Fishbein-Germon B, Hubley D, Chan B. The impact of adaptive seating devices on the lives of young children with cerebral palsy and their families. *Arch Phys Med Rehabil* 2009; **90:** 27–33.
26. McDonald R, Surtees R. Longitudinal study evaluating a seating system using a sacral pad and kneeblock with children with cerebral palsy. *Disabil Rehabil* 2007; **29:** 1041–7.
27. McDonald R, Surtees R. Changes in postural alignment when using kneeblocks for children with severe motor disorders. *Disabil Rehabil Assist Technol* 2007; **2:** 287–91.
28. Ekblom B, Myhr U. Effects of hip abduction orthosis on muscle activity in children with cerebral palsy. *Physiother Theory Pract* 2002; **18:** 55–63.
29. Rigby P, Ryan S, Campbell K. Effect of adaptive seating devices on the activity performance of children with cerebral palsy. *Arch Phys Med Rehabil* 2009; **90:** 1389–95.
30. Holmes KJ, Michael SM, Thorpe SL, Solomonidis SE. Management of scoliosis with special seating for the non-ambulant spastic cerebral palsy population-a biomechanical study. *Clin Biomech* 2003; **18:** 480–7.
31. Cimolin V, Piccinini L, Avellis M, et al. 3D-Quantitative evaluation of a rigid seating system and dynamic seating system using 3D movement analysis in individuals with dystonic tetraparesis. *Disabil Rehabil Assist Technol* 2009; **4:** 422–8.
32. Pope P, Bowes C, Booth E. Postural control in sitting the SAM system: evaluation of use over three years. *Dev Med Child Neurol* 1994; **36:** 241–52.
33. Vekerdy Z. Management of seating posture of children with cerebral palsy by using thoracic–lumbar–sacral orthosis with non-rigid SIDO frame. *Disabil Rehabil* 2007; **29:** 1434–41.
34. Hulme JB, Gallacher K, 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:** 1060–7.
35. Hulme J, 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:** 81–9.
36. Ryan S, Snider-Riczker P, Rigby P. Community-based performance of a pelvic stabilization device for children with spasticity. *Assist Technol* 2005; **17:** 37–46.

37. Seeger B, Caudrey D, O'Mara N. Hand function in cerebral palsy: the effect of hip-flexion angle. *Dev Med Child Neurol* 1984; **26:** 601–6.
38. Myhr U, von Wendt L. Improvement of functional sitting positions for children with cerebral palsy. *Dev Med Child Neurol* 1991; **33:** 246–56.
39. Lacoste M, Therrien M, Prince F. Stability of children with cerebral palsy in their wheelchair seating: perceptions of parents and therapists. *Disabil Rehabil Assist Technol* 2009; **4:** 143–50.
40. Stewart P, McQuilton G. Straddle seating for the cerebral palsied child. *Brit J Occup Ther* 1987; **50:** 136–8.
41. Hulme J, Bain B, Hardin M, McKinnon A, Waldron D. The influence of adaptive seating devices on vocalization. *J Commun Disord* 1989; **22:** 137–45.
42. Surveillance of Cerebral Palsy in Europe Collaborative Group (SCPE), 2009. Available from: http://www-rheop.ujf-grenoble.fr/scpe2/site_scpe/index.php (accessed 9 April 2012).
43. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol* 2007; **49**(Suppl. 109): 8–14.
44. Liberati A, Altman D, Tetzlaff J, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration. *Br Med J* 2009; **339:** b2700.
45. Law M, Baptiste S, Carswell A, McColl M, Polatajko H, Pollock N. Canadian Occupational Performance Measure, 4th edn. Ottawa: CAOT Publications ACE, 2005.
46. Ryan S, Campbell K, Rigby P. Reliability of the family impact of assistive technology scale. *Arch Phys Med Rehabil* 2007; **88:** 1436–40.
47. Livingstone R, Paleg G. Practice considerations for the introduction and use of power mobility for children. *Dev Med Child Neurol* 2014; **56:** 210–22.
48. Hadders-Algra M. Early diagnosis and early intervention in cerebral palsy. *Front Neurol* 2014; **5:** 185.
49. Dirks T, Blauw-Hospers CH, Hulsho LJ, Hadders-Algra M. Differences between the family-centered 'COPCA' program and traditional infant physical therapy based on neurodevelopmental treatment principles. *Phys Ther* 2011; **91:** 1303–22.
50. Kiresuk T, Smith A, Cardillo J. Goal Attainment Scaling: Applications, Theory and Measurement. Hillsdale, NJ: Lawrence Erlbaum Associates, 1994.
51. McDougall J, Wright V. The ICF-CY and Goal Attainment Scaling: benefits of their combined use for pediatric practice. *Disabil Rehabil* 2009; **31:** 1362–72.
52. Ostensjo S, Oien I, Fallang B. Goal-oriented rehabilitation of preschoolers with cerebral palsy – a multicase study of combined use of the Canadian Occupational Performance Measure (COPM) and the Goal Attainment Scaling (GAS). *Dev Neurorehabil* 2008; **11:** 252–9.
53. Ryan E, Sawatzky B, Campbell KA, et al. Functional outcomes associated with adaptive seating interventions in children and youth with wheeled mobility needs. *Arch Phys Med Rehabil* 2014; **95:** 825–31.
54. Narayanan UG, Fehlings D, Weir S, et al. Initial development and validation of the Caregiver Priorities & Child Health Index of life with Disabilities (CPCHILD). *Dev Med Child Neurol* 2006; **48:** 804–12.
55. Cong X, McGrath JM, Cusson RM, Zhang D. Pain assessment and measurement in neonates: an updated review. *Adv Neonatal Care* 2013; **13:** 379–95.
56. Badley EM. Enhancing the conceptual clarity of the activity and participation components of the International Classification of Functioning, Disability, and Health. *Soc Sci Med* 2008; **66:** 2335–45.

Part III

Therapist Designed Adaptive Riding (TDAR) Intervention