Rannei Sæther

Assessment of Trunk Control in Children and Adolescents with Cerebral Palsy

A neglected perspective?

Thesis for the degree of Philosophiae Doctor

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Norwegian University of Science and Technology
Faculty of Medicine
Department of Laboratory Medicine,
Children's and Women's Health

NTNU – Trondheim
Norwegian University of
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Sammendrag

Et av hovedproblemene for mange barn, unge og voksne med cerebral parese (CP) er at de har redusert trunkus kontroll. Disse vanskene påvirker deres evne til å sitte og gå. Under gange er trunkus kontroll spesielt viktig, ettersom to tredjedeler av kroppsmassen er lokalisert i de øvre to tredjedeler av kroppshøyden, noe som gjør at kroppen har et høyt tyngdepunkt, og dermed blir ustabil. Likevel neglisjeres disse vanskene ofte når ressurskrevende behandlinger for å bedre gangfunksjonen vurderes. Slike behandlinger omfatter blant annet intramuskulære injeksjoner med botulinum toxin (BoNT-A), pumpebehandling med baklofen, ortopedisk-kirurgiske inngrep, og/ eller ortoser.

Dette kan delvis skyldes at det er få etablerte metoder for å undersøke trunkus kontroll i daglig klinisk arbeid, og at det er få studier som har undersøkt trunkus kontroll under gange i CP populasjonen. Hovedmålet med denne avhandlingen har derfor vært å bidra til bedre undersøkelse og forståelse av trunkus kontroll hos barn og ungdom med CP, både i sittende og under gange. Mer spesifikt var målet å identifisere og evaluere kliniske verktøy som undersøker trunkus kontroll samt å evaluere påliteligheten (intra- og inter observatør reliabilitet) og gyldigheten (validitet) av et slikt verktøy; Trunk Impairment Scale (TIS). Videre var det et mål å undersøke trunkus kontroll under gange samt å undersøke sammenhengen mellom trunkus kontroll i sittende og under gange.

I den første studien, en systematisk litteratur oversikt, identifiserte vi 22 kliniske verktøy som undersøker trunkus kontroll hos barn, ungdom og voksne med CP. Vi fant begrenset dokumentasjon av måleegenskapene til disse verktøyene, og informasjonen om egenskapen til å måle endring (responsiveness) var spesielt begrenset. Mangelen på verktøy som kan måle endring er en begrensning for gjennomføring av behandlingsstudier. *I den andre studien* fant vi at TIS, en av testene inkludert i den systematiske litteratur oversikten, viste høy inter- og intra observatør reliabilitet samt god validitet hos barn og ungdom med CP. *I studie tre* fant vi at barn og ungdom med CP hadde signifikante vansker med trunkus kontroll under gange, undersøkt med et 3-dimensjonalt askelerometer festet på nedre del av trunkus. Vanskene med trunkus kontroll ble reflektert gjennom økte trunkus akselerasjoner og mindre regularitet mellom steg, og de tenderte til å øke med økende alvorlighetsgrad av CP samt med økende gang hastighet. Til slutt, *i studie fire*, fant vi en moderat sammenheng mellom trunkus kontroll i sittende, undersøkt med en «del-test» av Trunk Control Measurement Scale og TIS, og trunkus kontroll under gange, undersøkt med et askelerometer festet på nedre del av trunkus.

Av relevans for klinisk praksis er at funnene i denne studien tyder på at de mindre tidkrevende «del-skalaene» kan benyttes for å innhente informasjon om trunkus kontroll under gange.

Det er et stort antall undersøkelsesverktøy beregnet på å undersøke trukus kontroll i ulike situasjoner tilgjengelig. Denne avhandlingen understreker at det er begrenset dokumentasjon av hvor godt disse verktøyene faktisk måler trunkus kontroll, og spesielt hvor gode de er til å måle effekt av behandling («responsiveness»). Våre resultater bekrefter at barn og ungdom med CP har vansker med trunkus kontroll både i sittende og under gange, og avhandlingen gir ny kunnskap om sammenhengen mellom disse to ulike oppgavene. Denne informasjonen bør føre til at fokuset i forbindelse med planlegging av intervensjoner som har til hensikt å bedre gangfunksjonen, utvides til også å omhandle en vurdering av pasientens trunkus kontroll.

Navn kandidat: Rannei Sæther

Institutt: Department of Laboratory Medicine, Children's and Women's Health
Veiledere: Torstein Vik, Jorunn Helbostad og Lars Adde
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“I am glad to say:

There is work enough left for you to do.”

R. Magnus 1926
L. Oddsson 1990

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Trondheim, 5 May 2014

Rannei Sæther

List of papers

Paper I

Clinical tools to assess balance in children and adults with cerebral palsy: A systematic review

Saether, R., Helbostad, J. L., Riphagen, II., & Vik, T. (2013). Clinical tools to assess balance in children and adults with cerebral palsy: A systematic review. *Developmental Medicine and Child Neurology*, 55(11): 988-999.

Paper II

Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy

Saether, R., Helbostad, J. L., Adde, L., Jorgensen, L., & Vik, T. (2013). Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy. *Research in Developmental Disabilities*, *34*(7): 2075–2084.

Paper III

Gait characteristics in children and adolescents with cerebral palsy assessed with a trunk-worn accelerometer

Saether R, Helbostad J. L., Adde L., Brændvik, S., Jorgensen L., Lydersen, S., & Vik T. (2014). Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy. *Research in Developmental Disabilities*, 35(7): 1733-81.

Paper IV

The relationship between trunk control in sitting and during gait in children and adolescents with cerebral palsy

Saether R, Helbostad J. L., Adde L., Brændvik, S., Jorgensen L., Lydersen, S., & Vik T. (2014). Submitted to: *Developmental Medicine and Child Neurology 04.04.14.* Accepted for publication 22.08.14.

Abbreviations

AP = anterioposterior

APA = anticipatory postural adjustment

BoNT = botulinum toxin

COP= center of pressure

COM = center of mass

COSMIN = consensus-based standards for selection of health measurement instruments

CP = cerebral palsy

CPG = central pattern generators

EBP= evidence based practice

EMG = electromyography

GMFCS = Gross Motor Function Classification System

GMFM = Gross Motor Function Measure

GRRAS= Guidelines for Reporting Reliability and Agreement Studies

HAT = head, arm, trunk segment

ICC = Intraclass Correlation Coefficient

ICF = International Classification of Functioning, Disability and Health

ICF CY= International Classification of Functioning, Disability and Health, Children and Youth

LSS = Level of Sitting Scale

ML = mediolateral

NGST = neural group selection theory

RPA = reactive postural adjustment

SATCo= Segmental Assessment of Trunk Control

SELMS = single-event multilevel surgery

SEM= standard error of measurement

TD = typical developing children

TCMS = Trunk Control Measurement Scale (SSB= static sitting balance, DSB-S= dynamic sitting balance-selective control, DSB-R= dynamic sitting balance-reaching)

TIS = Trunk Impairment Scale (SSB = static sitting balance, DSB = dynamic sitting balance, C = coordination)

TPS= Trunk Profile Score

TUG = Timed Up and Go

V = vertical

Summary

Poor trunk control is a primary impairment in children, adolescents, and adults with cerebral palsy (CP) and influences their activities in daily life such as sitting and walking. In the latter case, trunk control is especially important since two-thirds of the body mass (head, arms, and trunk) is located in the upper two-thirds of the body height, thus making the body unstable. Nonetheless, in the decision process leading to "gait interventions," such as orthopedic surgery, botulinum toxin injections, intrathecal baclofen, and/or the application of orthoses, the focus is mainly on the lower extremities.

This may partly be due to few established methods for assessment of trunk control in daily clinical work, and that few studies have examined trunk control during gait in children and adolescents with CP. The main aim of this thesis is therefore to contribute to better assessment and understanding of trunk control in children and adolescents with CP, both in the sitting position and during gait. The specific aims during the research were to identify and evaluate clinical tools to assess trunk control and to evaluate the intra- and inter-observer reliability and construct validity of one such tool: the Trunk Impairment Scale (TIS). Further aims were to investigate trunk control during gait, and finally to investigate the relationship between trunk control during sitting and during gait.

In the first study, a systematic literature review, 22 clinical tools for assessment of trunk control in children, adolescents and adults with CP were identified. However, there was moderate or limited evidence for the measurement properties of the tools, and scarce information on the measurement property responsiveness. The latter is a limitation for intervention research which is dependent of the ability to evaluate change. *In the second study*, it was found that the TIS, one of the tools included in the review, showed high intra- and inter- observer reliability, and the construct validity of the test involving children and adolescents with CP was considered as good. *In the third study*, children and adolescents with CP were found to have significant difficulties with trunk control during gait, which was assessed with a trunk-worn accelerometer. The difficulties were reflected in higher trunk acceleration and less regularity between strides than in children with typical development. These problems seemed to rise with increasing gross motor impairment and increasing speed. Finally, *in the fourth study*, a moderate relationship was found between trunk control during sitting assessed with a subscale score of the Trunk Control Measurement Scale and the TIS and trunk control during gait assessed with a trunk-worn accelerometer. With regard to

relevance for clinical practice, the findings of the latter study suggest that the some of the less time-consuming subscales may be used to gain information on trunk control during gait.

A large number of assessment tools of trunk control are available. This thesis reveals that there is limited evidence for the measurement properties of the tools, especially for responsiveness. The results confirm that children and adolescents with CP have impaired trunk control both during sitting and gait, and provide new knowledge of the relationship between the two tasks. This information may expand the focus on the lower limbs in gait assessment in children and adolescents with CP to include assessment of the trunk.

1. Introduction

Children with cerebral palsy (CP) are often referred for gait analysis prior to gait interventions. As a physiotherapist and member of St Olavs Hospital's neuro-orthopedic team, I was invited to some of the meetings where the results of gait analyses were interpreted. At these meetings: *We saw the child's lower limbs walk over the computer screen* (**Figure 1**). *From the movements of this half body, we were asked to interpret the walking pattern of the child with CP in order to make plans for intervention.* This experience summarizes much of my motivation for doing this work.

Figure1. Three-dimensional gait analysis

The topic of this thesis is the assessment of trunk control, with special focus on trunk control during gait in children and adolescents with CP. Gait is usually assessed in the decision process leading to treatments such as orthopedic surgery, botulinum toxin (BoNT) injections, and/or the application of orthoses. This thesis highlights concerns related to decision making, with the main focus on the assessment of the lower limbs.[1, 2] However, as a physiotherapist working with preterm children and young children with CP, I experienced trunk control as a rate-limiting factor, and thus probably a determinant (i.e., a factor that influences the outcome of interest)[3] in the development of activities of daily life, such as sitting and walking. I found support for my experiences in the literature.[4, 5] Consequently, I found it difficult to understand why assessments of trunk control seemed to be omitted from the decision-making process in gait interventions. However, in order to communicate my concerns to the neuro-orthopedic team, a multidisciplinary team making decisions on treatments such as surgery and BoNT injections, I was looking for a standardized clinical tool to assess trunk control in children with CP. I was only aware of one such tool, the Trunk Impairment Scale (TIS), which was developed for the assessment of trunk control in adults after stroke.[6] During my research for my master's thesis I assessed the measurement properties of the TIS in children with CP.[7] During this work, the following question emerged: "To what extent can we measure a

construct such as trunk control?" Measurement is fundamental in research and clinical practice, and it is important that researchers and practitioners can have "trust" in the tools that are used. The trust relates to the measurement properties of the tools, such as reliability and validity. It should be highlighted that consistency of both the quality and the results (i.e. have an Intraclass Correlation Coefficient > 0.75) of the studies of measurement properties have to be good. This is fundamental to ensure the validity of the reported results of studies in which assessment tools have been used.[8]

I consider a systematic review of trunk control tools including assessment of the measurement properties of the tools to be my contribution to clinicians and researchers working with children and adolescents (hereafter referred to as children) with CP, as well as exploring trunk control during sitting and walking in such children.

2. Background

In this section I first describe the definition and classification of CP (section 2.1), followed by definitions and some theoretical perspectives on trunk control (2.2). Thereafter, I review some recent literature on trunk control during sitting and gait (2.3) and interventions related to gait (2.4). This is followed by reflections on assessments in general, on some historical perspectives on assessment, and on measurement properties related to assessment. The section ends with a brief summary on assessment of trunk control during sitting and gait (2.5).

2.1 Terms and constructs

2.1.1 Cerebral palsy

CP is the most common motor disorder in children, with an incidence of approximately 2.2 per 1000 live births in developed countries.[9, 10] beginning in early childhood and persisting throughout the lifespan.[11]

CP is described as "a group of disorders of the development of movement and posture, causing activity limitation that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception and/or behavior, and/or by a seizure disorder."[1](1)12 In this new definition of CP the inclusion of postural abnormalities, as observed in the clinical picture, is clearly emphasized.

Children with CP have a wide variability in their presentation and severity of the disorder and they can be classified by the predominate type of motor disorder and by topographical distribution. The groups are unilateral (30%) and bilateral (58%), dyskinetic (7%), ataxic (4%), and mixed or unclassifiable (1%).[13] The severity of their gross motor impairment can be described by the Gross Motor Function Classification System (GMFCS), which is a reliable and valid classification containing five levels of severity, ranging from Level 1 (least affected) to Level 5 (most affected).[14] The GMFCS is based on self-initiated movement, with particular emphasis on trunk control during sitting and walking. Children classed as GMFCS Level I can perform all activities performed by their age-matched peers, albeit with some difficulties in their speed, balance, and coordination. Children classed as Level II have similar

[1] Bax M. et al., 2005. Page 572.

functional abilities when walking on flat and familiar surfaces but require support when negotiating uneven surfaces or stairs. Children classed as Level III are also independent walkers but require walking aids such as cane and crutches or a walker, and may use a wheelchair for longer distances. Children classed as GMFCS Levels IV and V are non-ambulatory.[15, 16]

Children with CP constitute a heterogeneous group, in which their disability results in limitations to their activity.

2.1.2 Trunk control

Despite that few clinicians would argue the importance of posture or balance to independence in activities such as sitting, standing, and walking, there is no universal definition of these constructs or agreement on how to assess them.[17] Constructs are described as abstract behaviors or events that cannot be directly observed, but can be inferred from other relevant observable variables.[8] Thus, in research, constructs have to be operationally defined (i.e., by the way they are measured).[18]

Impaired control of *posture* is an essential part of the definition of CP.[19] Shumway-Cook and Woollacott[17] include posture in the description of *postural control*, which involves controlling the body's position in space for the dual purposes of *orientation* and *stability.*

Postural *orientation* is defined as the ability to maintain an appropriate relationship between body segments, and between the body and the environment during a task.[20] Posture is often used to describe both the biomechanical alignment of the body and orientation of the body in the environment, and Shumway-Cook and Woollacott[17] include both of these concepts in the term postural orientation.

Postural stability, also referred to as ***balance***, is the ability to control the center of mass (COM) in relation to the base of support.[17] COM is defined as a point at the center of the total body mass (determined as the weighted average of the COM of each body segment) and is *located in the trunk.* Analysis of trunk movements may therefore yield information on balance in activities such as sitting and walking.[21] The main focus in this thesis is on ***trunk control***, and hereafter this term is used in the text. Trunk control involves stabilization and selective movements of the trunk.[22] Stabilization is essential for free and selective movements of the head and the extremities,[22] for example during gait.[21] Trunk movements are controlled in an

interaction with feet placement, and the "*trunk may be oriented secondary to foot position or vice versa.* "[(2)23]

Due to different aims and assessment methods, trunk control was operationalized differently in the studies reported in Papers I–IV.

In ***Paper 1***, a systematic literature review, we used the term '*balance*', and it was defined as the act of maintaining, achieving, and restoring the COM relative to the base of support. A broad search strategy was used, including many terms reflecting balance (see ***paper I***), to identify clinical assessment tools of balance.

In ***Paper II*** the term "*trunk control*" was used in an assessment of sitting. Trunk control was evaluated according to items describing 'qualities of trunk control' such as static balance, dynamic balance, and coordination.

In ***Paper III*** we used the terms '*progression'* and '*balance*' in the assessment of gait characteristics. However, the term trunk control may be more accurate, as the study focused on the movements of the trunk with the application of a trunk-worn accelerometer. The gait characteristics representing *progression* were gait speed, cadence, step time, and step length, and the gait characteristics representing *balance* were trunk accelerations, regularity between strides (where decreased regularity means increased variability), and asymmetry. The interpretation of the results was done according to earlier studies applying the same assessment method. In these studies a higher average dispersion of accelerations was found in subjects with impaired balance during gait,[24-27] while both increased and reduced variability between strides has been regarded as a marker of balance during gait.[28, 29] However, the above-mentioned gait characteristics are not separate entities, as reduced balance may lead to reduced gait speed and vice versa.[30]

In ***Paper IV*** we used the term *'trunk control'*. In the study on which the paper is based, trunk control was represented by trunk accelerations and regularity between strides, based on the findings in ***Paper III***.

[2] Moe-Nilssen R. et al., 2005. Page 165.

2.2. Theoretical perspectives

In this section I describe some theoretical perspectives on motor control and motor development related to trunk control.

2.2.1 Motor control

Motor control is defined as the ability to regulate or direct the mechanisms essential to movement.[31] Studies of motor control address questions such as how does the central nervous system organize the many individual muscles and joints into coordinated functional movements? Such questions are of interest in order to understand the underlying factors of trunk control. Different theories of motor control reflect philosophically varied views about how the brain controls movement. Such theories often reflect differences in the option about the importance of various neural components. There are several theories of motor control: reflex theory, hierarchical theory, motor program theory, system theory, dynamic action theory, and ecological theory.[31] In this thesis, elements from both *program theory* and *system theory* are described to illustrate aspects of trunk control in children with CP.

Based on *motor program theory*, Forsberg and Hirchfeldt[32] developed a *functional model of the organization of postural control* (thus including trunk control) during externally triggered perturbations studies of sitting adults. This model is also called the *central pattern generator (CPG) model.* The CPGs are specific neural circuits that generate rhythmical movements, such as locomotor rhythm.[31] Essential to the CPG model is its organization of *two functional levels of control.*[32-34] The *first level* consists of a network that coordinates the basic structure of postural synergies. At this level, *direction-specific* synergies are performed. This means that a forward sway induces activity in the muscles on the dorsal side of the body, while a backward sway induces activity in the muscles in the ventral muscles, and a similar synergy is present in the frontal plane. It has been hypothesized that the basic structure of postural synergies is generated by the above-mentioned spinal networks. The *second level* of control is involved in the *fine tuning* of the basic pattern of adjustment on the basis of multisensory afferent input from somatosensory, visual, and vestibular systems. Modulation can occur by means of: (1) the selection of the best-fitting muscle activation pattern from the repertoire of direction-specific patterns; (2) the recruitment of antagonist muscles; (3) the recruitment order of the direction-specific muscles; and (4) the degree of contraction of the direction-specific muscles.[35]

Nicolai Bernstein, a Russian scientist, who also participated in the development of motor program theories, looked at the nervous system and the body in a new way and contributed to the development of *system theory*.[36] He recognized that one cannot understand the neural control of movement without an understanding of characteristics of the system within which one is moving and the external and internal forces acting on the body. System theory takes into account not only the nervous system's contribution to action, but also the contribution of the muscular and skeletal systems, as well as forces of gravity. Movement emerges from the interaction of three factors: the individual, the task, and the environment. Requirements of trunk control thus vary with the task and environment (**Figure 2**).[36] Bernstein was the first to realize that the central problem in motor control, including trunk control, was organizing the redundant sets of elements, muscles, and joints in task-specific ways. He suggested that the motor problem posed by excessive degrees of freedom might be solved by organizing the elements into synergies.[36] Synergies have been defined as neural organizations of sets of elements with the purpose of stabilizing a particular feature of performance.[37] The description of muscle synergies is essential for the organization of the above-described functional model of the organization of postural control.

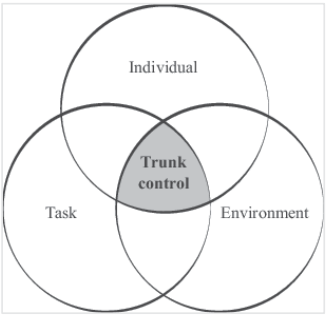

Figure 2. Trunk control emerges from an interaction of the individual, the task and the environment (modified from Shumway-Cook and Woollacott, 2012)[31]

2.2.2 Motor development

Concurrent with changes in insights into the neural mechanisms involved in motor control, knowledge of motor development also increased.[35] Motor development was initially regarded as an innate maturational process, described in neural-maturation theories, but gradually it became clear that motor development is also affected by experience. The extent to which experience affects motor development is still a matter of debate.[38] This is reflected in two theoretical frameworks that are most frequently used today:[38, 39] the dynamic systems theory, which

assigns a dominant role to experience, and the *neural group selection theory* (NGST), in which genetic endowment, epigenic cascades, and experience play equally prominent roles.[38] In this thesis the NGST is emphasized to facilitate the understanding of the development of trunk control and contributes to understanding of the effects of brain damage in early age.

Forsberg and Hirchfeldt[32] found support for their functional model of the organization of postural control (describing two levels of control) in NGST introduced by Edelman.[33, 40] This theory explains the variation in motor development on the basis of experience and selection. Healthy infants show great variation in spontaneous movements. During the phase of development called *primary variability*, the neural system explores all motor possibilities available for a function. This phase is consequently characterized by variability. At a certain point in time the nervous system starts to use the afferent information produced by behavior and experience for the selection of motor behavior that best fits a given situation. This is followed by the phase called *secondary variability*, in which the selection process is based on active trial-and-error experiences that are unique to the individual.

2.3 Previous research

The focus of this thesis is on *sitting* and *walking*. In this section I first describe reasons for focusing on these tasks. Thereafter, I present research on trunk control during sitting in typical developing (TD) children (2.3.1) and children with CP (2.3.2). In the following, I address aspects of trunk control (characteristics, control, and development) during gait in TD children (2.3.3) and in children with CP (2.3.4). Research related to TD children has been included, as knowledge of movement and development in such children may facilitate the understanding of impairments in children with CP. Lastly, I present an overview of the few published studies of trunk control during gait in children with CP (2.3.5). That there are few studies of trunk control may reflect that the focus of the gait assessment has been on the lower limbs. Thus, the main objective of including this section has been to facilitate an understanding of the important role of trunk control during gait.

Poor trunk control is a primary impairment in children, adolescents, and adults with CP and may affect their activities in daily life, such as sitting and walking.[21, 41] Assessment of trunk control during sitting may provide some information about primary impairment in trunk control. In assessments of walking, deviations in the trunk are most often interpreted as

compensation for impairments in the lower limbs, such as muscle weakness and impaired control. However, the deviations may also be due to a primary impairment in the trunk.[41] Moreover, primary impairments in the trunk may cause compensatory movements in the lower limbs, for example by allowing the pelvis to rotate anteriorly and thereby causing increased hip flexion.[42] It may therefore be important to reveal primary trunk deficits in order to plan the most appropriate "gait treatment," such as orthopedic surgery, botulinum toxin injections, and/or application of orthoses, in children with CP.

However, it may be difficult to separate deviations in trunk control during gait due to "primary" impairments of trunk control from those due to "secondary" impairments caused by impairments in the lower extremities,[42] even when using advanced laboratory equipment. Since trunk control during sitting is less influenced by impairments in the lower extremities, assessment of trunk control during sitting may be used as a first step to identify primary impairments. Even though the two tasks clearly differ, trunk control during sitting may be assessed as an indicator of impaired (primary) trunk control during gait if there is a relationship between trunk control during sitting and during gait. If such a relationship can be documented, the implementation of a short test of trunk control during sitting may provide information about primary impairments in trunk control and may thus lead to improved decision processes regarding the choice of gait interventions. Moreover, the results of such a sitting assessment might be used to select children who need a thorough assessment of the relationship between their trunk and lower limbs, including full-body three-dimensional (3D) gait analysis, before a treatment option is chosen.

2.3.1 Trunk control during sitting in TD children

Hedberg and colleagues[43] were the first to systematically study trunk control (specifically the adjustment of muscle activity) in very young infants. The results of their study indicated that at the age of one month infants can generate direction-specific adjustments of the trunk, meaning that the first level of control (2.2.1) is functionally active at this age and that it possibly has an innate origin. Other studies have shown that during the first six months, the phase of primary variability, an infant explores its movement repertoire, and a high degree of variability in movements may be observed. From six months onwards, the phase of secondary variability starts, in which an infant will select the most appropriate movement and less variability, may be observed. In this period the child develops the ability to adapt their trunk control to a specific situation. Around the age of 13–14 months, anticipatory control emerges,

suggesting that at this age infants develop the ability to integrate feed- forward control into trunk control.[44] Initially, infants develop a top-down recruitment order of postural muscles, whereas children who sit independently have a preference for bottom-up recruitment. It takes at least to adolescents before an adult type of trunk control has been achieved.[5]

2.3.2 Trunk control during sitting in children with CP

The complex nature of the trunk control (described in 2.2) induces vulnerability for dysfunctions in cases of adverse conditions during early life,[44] including children with CP. In general, children with CP can produce direction-specific postural muscular activity, and the *first level* of Hirschfeldt and Forsberg's[43] *functional model of organization of postural control* (referred to in 2.2.1) is intact. Only children with severe CP (GMFCS Level V), who cannot sit independently, entirely lack these adjustments. In children classed as GMFCS Level IV and in young children at Level III, a parietal loss of direction-specific adjustments has been found.[45] However, the most frequent dysfunctions in children with CP are related to the *second level*, which means that they have problems with adaption of postural muscular activity (fine-tuning of the basic direction-specific adjustments to environmental conditions, which is based on experience and sensory information from somatosensory, visual, and vestibular systems).[46] One study assessed the influence of two different sitting positions on postural adaption in children with CP compared to TD children and found that in children with CP the deficient adaptive capacity was more pronounced in the erect position than in the crouched position.[47]

Differences in trunk control due to topography and the severity of motor impairments rated according to the GMFCS have been assessed in a large study of 100 children with CP, in the age group 8–15 years.[48] The highest scores were obtained for children with hemiplegia (where a high score represented a high level of performance), followed by children with diplegia, while the lowest scores were obtained for children with quadriplegia. The trunk control scores significantly decreased with increasing GMFCS level. The finding concerning the trunk control score in relation to GMFCS level was in accordance with findings in our earlier study, a reliability study of a clinical test to assess trunk control (using the TIS).[7]

Moreover, several studies have described typical characteristics of trunk control in children with CP, and of these the most typical is a top-down recruitment of postural muscles, excessive degree of antagonist co-activation, and a lack of modulation to task-specific

constraints.[5, 45, 49, 50] Studies of anticipatory and compensatory trunk control during sitting in CP and TD children demonstrated that children with CP make fewer anticipatory compensatory adjustments and more compensatory adjustments (with stronger muscle activation after the initiation of movement) than TD children.[51]

Several studies have assessed trunk control and reaching during sitting. The children with CP invariably showed deficits in their ability to adapt (i.e., fine-tune) direction-specific activity, and the degree of their dysfunction was related to the severity of their CP.[52] The typical characteristics of movements with a top-down recruitment of postural muscles and excessive degree of antagonist co-activation were as described above. However, there was an absence of antagonistic co-contraction, indicating that the co-contraction in children with CP is situation-specific.[52] A study by van der Heide and colleagues[52] of preterm children with CP demonstrated that their ability to adapt or fine-tune their postural activity was based especially on information on trunk position. The authors argue that this finding fits with the emerging idea that afferent information from the trunk might be a major source of input for postural regulation.[53] In a separate study, van der Heide and colleagues[54] assessed the kinematic characteristics of trunk control during reaching in preterm children with CP and found that the sitting position before the onset of reaching differed between CP and TD children. The children with CP sat with a more reclined pelvis and a more "collapsed" trunk. The more reclined pelvic position was associated with a better quality of reaching movement in children with CP.

2.3.3 Trunk control during gait in typical development

Characteristics of gait

Gait is characterized by three essential requirements: progression, trunk control, and adaption. Progression is ensured through a basic locomotor pattern that produces and coordinates rhythmic patterns of muscle activation in the lower extremities, which in turn successfully move the body in the desired direction. The requirements of trunk control reflect the need to establish and maintain an appropriate posture for locomotion, and the demand for dynamic stability of the moving body. The third essential requirement of gait is the ability to adapt gait to meet the goals of an individual and the demands of the environment.[55] Although the focus of this thesis is mainly on trunk control, the three characteristics are interrelated and may influence each other.

In descriptions of gait, the body has been divided into two functionally different units: the passenger unit (head, arm, and trunk segment (HAT)) and the locomotor (lower limbs) unit (**Figure 3**).[56] Perry and colleagues[56] described the normal gait mechanisms to be so efficient that the demands of the passenger unit were reduced to a minimum, "making it a passive entity that rides on the locomotor system."[(3)] However, they emphasized that alignment of the passenger unit above the lower limbs was a major determinant of muscle action within the locomotor system. Other researchers, such as Winter et al.,[21] have highlighted the importance of trunk control (the passenger unit) during gait, as two-thirds of the body mass (HAT) is located at the upper two-thirds of the body height, thus making the body unstable. The body is therefore in a continuous state of imbalance during gait, as the COM remains outside the support surface for 80% of the time. The only way to prevent falling is to place the swing leg ahead of and lateral to the COM as it moves forward; in addition, the mass of the HAT would have to be regulated with respect to the hip.[57]

Figure 3. Functional division of the body (modified from Perry & Burnfield 2010,[56] with permission from SLACK Incorporated)

During gait the motion of the individual joints is large, yet the coordinated action of motion across all joints results in smooth forward progression of the COM through a fluctuating sinusoidal path (**Figure 4**).[56] However, the displacement of the COM has been related to some proposed overall goals of gait, namely moving the body with the least energy and the greatest stability.[58] Saunders and coworkers[59] identified "determinants" of normal gait that they proposed were responsible for saving body energy by minimizing the displacement of the COM. However, this hypothesis has been reconsidered and the smooth mechanical transfer of kinetic and gravitational energies has been suggested as more important for an energy

[3] Perry J et al., 2010. Page 20.

efficient gait.[60] Although the aim of gait-interventions often is to reduce energy consumption, this is not discussed further in this thesis.

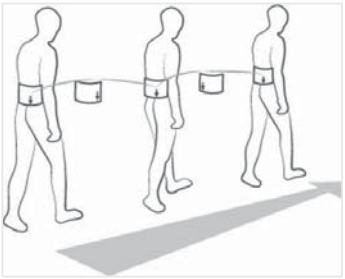

Figure 4. The normal path of the center of mass (COM) (modified from Perry & Burnfield 2010,[56] with permission from SLACK Incorporated)

Control

All parts of the central nervous system, from the spinal cord to the cortex and the major sensory systems, are involved in control of locomotion. There are indications that the central pattern generators within the spinal cord play an important role in generation of rhythmic movements underlying gait (2.2.1).[60] However, the goals for human walking are sophisticated and require control that is more complex, with input from supraspinal centers and collaboration between them. Especially important are the basal ganglia, cerebellum, and brainstem: the basal ganglia are important for planning, initiating, and generating smooth movements, the cerebellum for smooth execution and completion of movements and providing trunk control, and the brainstem for providing background posture and muscle tone. The cortical centers act as commanders of the voluntary movement.[61]

Several strategies are used to maintain trunk control when voluntary movements are performed, including "postural preparation," anticipatory postural adjustments (APAs), and reactive postural adjustments (RPAs) (**Figure 5**).[62, 63] During gait, trunk control is achieved through a combination of APAs and RPAs. Unlike reactive responses, which use sensory feedback to counteract disturbances to the trunk, APAs actively initiate movements to anticipate and counteract disturbances to the trunk in a feed-forward manner.[62] All sensory systems are involved in the proactive and reactive control of gait. The somatosensory systems contribute to keep an appropriate cadence and rhythm, vision helps to determine walking speed, and the vestibular system provides important information about position in space.[55]

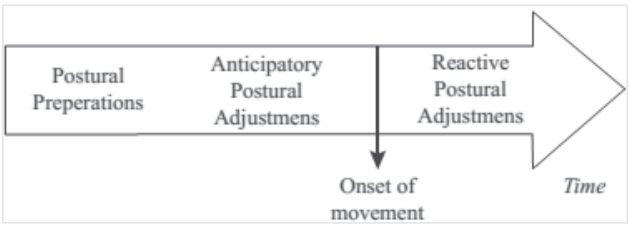

Figure 5. Movement strategies (modified from Frank and Earl, 1990[63] with permission from the American Physical Therapy Association)

All movements, including even normal locomotor movements, perturb the body by virtue of displacement of the COM and thus require reactive movements.[60] Winter[21] has documented the joint moments that counteract the perturbations produced by the normal locomotor movements. He found that anterioposterior (AP) motion of the trunk, with acceleration and deceleration in each step cycle, was primarily controlled by the hip extensors. The mediolateral (ML) movement of the body was primarily regulated by the hip abductors (the magnitude of destabilization controlled by the foot placement with respect to body COM). Collapse in the vertical (V) direction was prevented by controlling the movement about the knee joint.

Moreover, the trunk serves a number of control functions during gait, as it especially plays an important role in anticipatory trunk control,[64] in steering (i.e., moving the COM in a new direction),[65] in ensuring an upright posture, and in attenuation of gait-related oscillations to promote stability of the head (i.e., to stabilize optic flow and vestibular signals).[66] Furthermore, the trunk interacts with the lower limb movements to achieve efficient locomotion.[67]

In addition, there are important *non-neural* contributions from the musculoskeletal system and the environment to the control of gait. In particular, gravity plays an important role by contributing to the pendular movement of the COM (the COM of the body vaults over the stance leg in an arc), called the "inverted pendulum mechanism."[68] The pendulum mechanism conserves mechanical energy, and thus little mechanical work is needed.[58] The main work is done in the transitional gait phases: gait initiation (acceleration) and termination (deceleration). In addition, there are acceleration and deceleration phases within each step, caused by the breaking effect of foot strike in the AP direction, opposing gravity in the V direction, and opposing the effect of weight shift in the ML direction.[21]

Development

When looking at the three requirements for successful locomotion: a rhythmic stepping pattern (progression), control of the trunk, and the ability to modify gate (adaption), clearly a rhythmic stepping pattern is developed first.[68] Brenière and Bril[69] studied the emergence of locomotion and hypothesized that learning to walk is a two-stage process. In the initial phase infants learn to control their trunk, while in the second phase the locomotor pattern is progressively refined. They proposed that a high level of strength is required to control gravitational forces that tend to destabilize the upper body.

An important part of controlling gait is stabilizing the head. Assaiante et al.[70] found that from achieving stance until about six years of age, children organize gait in a bottom-up manner, using the support surface as a reference and controlling their head movements in an en bloc mode, which serves to reduce the degrees of freedom to be controlled. During this period, the children gradually learn to stabilize their hips, then their shoulders and finally their head. At about seven years of age, with mastery of the control of the head, there is a transition and the head is changed to an articulated mode, and top-down organization of trunk control during gait becomes dominant.

However, although gait patterns (kinematics) normally are developed to an adult pattern already by the age of five years,[71] trunk control during gait continues to develop into adolescence.[72]

2.3.4 Trunk control during gait in children with CP

Characteristics

The Surveillance of Cerebral Palsy in Europe revealed that 54% of the studied children walked without support by five years, whereas 16% walked with assistive devices, and 30% were unable to walk. Gait ability related significantly to CP type, IQ level, epilepsy, and severe visual and hearing impairment.[73] There is a wide range of "locomotor phenotypes" in children with CP.[74] In a large retrospective study as many as 14 specific gait abnormalities were described (Figure 6),[75] and in a systematic review 18 gait classifications were identified, whereby only the lower limbs were included in the classifications.2 Several gait indexes exist, which quantify deviations in kinetics and kinematics from "normal gait."[76, 77] A recent study presented a similar index, the Trunk Profile Score (TPS), which reflects the overall severity of

trunk movement pathology during gait.[41] Moreover a longitudinal assessment of sagittal trunk movements in children with CP has shown two main patterns. The most common pattern type occurred in 75% of the children and was characterized by an erect posture, with possible enhancement of lumbar lordosis and anteversion of the pelvis. By contrast, the second main type, which was less common and which occurred in 25% of the children, was characterized by "forward-bent trunk" walking.[78] Some researchers have argued that the abnormal pattern found in many patients with neurologic pathology is like the stepping pattern observed in young children. However, some children with CP persist in using this pattern.[79]

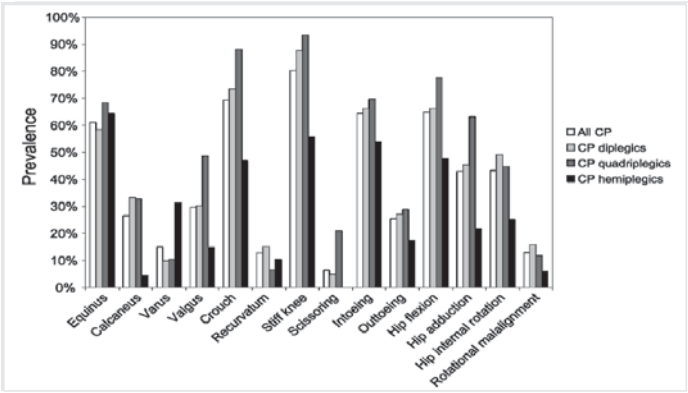

Figure 6. Different gait patterns described by Wren et al. (2005) (reproduced with permission from Wren[75])

Development

In children with CP gait, skills emerge and plateau later than in individuals with typical development. Moreover, deterioration in gait ability may occur, especially in the most impaired children (GMFCS III), and studies have shown that as many as 10–20% became non-ambulatory by the age of 40.[71] In the development of independent gait, both trunk control and muscle strength have been considered as rate-limiting factors,[69, 80] as described above. However, a decline in these factors may also contribute to a decline in gait ability. In a study conducted by Opheim and colleagues,[81] self-reported deterioration of gait in adults (> 40 years) with bilateral CP was regarded as a result of impaired trunk control by 65% of participants and reduced strength by 33%. In two follow-up studies, involving self-reported deterioration in gait in adults with bilateral CP, by the same research group, the differences could not be explained by differences in balance confidence, fear of falling, or balance,[82] work (COM and joint work), or gait profile score.[83] For future research, they suggested longitudinal studies, studies of measurement properties of the assessment tools, and further exploration of individual interpretations of gait deterioration with a qualitative design.[83]

Pathophysiological mechanisms

The primary disorder of the brain in children with CP may be associated with abnormal muscle tone, most often hypertonia, accompanied by loss of selective motor control, muscle weakness, and impaired trunk control. Moreover, the motor disorders may contribute to secondary musculoskeletal problems, including muscle contractures, bony deformities, and joint instability.[84] Impaired trunk control does not necessarily result from primary impairments, but might be a consequence of secondary factors or non-neural components.[74] However, the motor disorders of CP are often accompanied by disturbances in sensation and perception, a point that is emphasized in a new definition of CP.[19]

Woollacott and Crenna[74] have presented a "look-up chart" (**Figure 7**) of relevant pathophysiological mechanisms potentially contributing to abnormal gait in children with CP, The mechanisms include: (1) peripheral, non-neural components related to bone and passive muscle tendon properties; (2) central executive factors, including impaired muscle activation (paretic component), loss of selectivity in neuromuscular output (co-contraction component), and abnormal electromyography (EMG) recruitment upon stretching (spastic component); (3) central disturbance of sensory processing and sensorimotor integration; and (4) impaired higher level function.

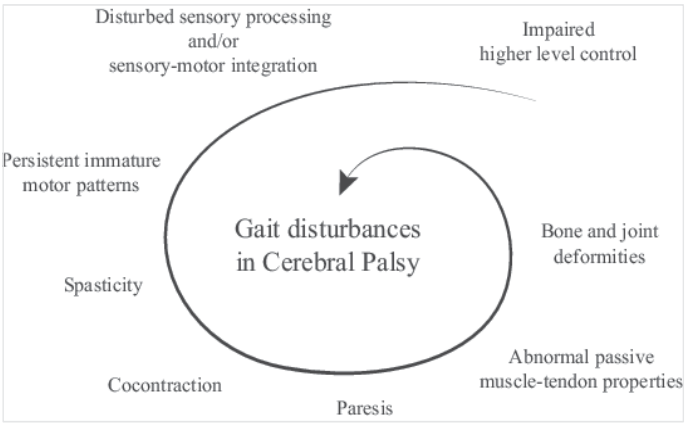

Figure 7. Look-up chart of relevant pathophysiological mechanisms potentially contributing to abnormal gait in children with cerebral palsy, arranged clockwise according to ranking from lower level peripheral mechanisms to the higher level central components (modified from Woollacott and Crenna, 2008,[74] with permission from Mac Keith Press)

Deviations observed in the trunk during gait are most often described as compensating for impairments such as weakness and impaired control in the lower limbs. However, these

deviations might equally be due to primary trunk impairments,[42] and might equally influence the movement of the lower limbs, consistent with the expression "*the trunk may be oriented secondary to foot position or vice versa.* "[4](4)23 The importance of "proximal stability" (trunk and hips) for "distal mobility" has been shown in studies of gait in children with CP[85, 86] and in gait development in TD children,[87] where increased hip muscle strength[85] and trunk control[86] has been shown to improve plantar flexor generating power at push-off and gait distance and spatiotemporal gait parameters, respectively. Trunk control (often referred to as "core stability in sports medicine") has also been found to be important for improving performance and preventing injuries during athletics.[88] Moreover, this relationship between trunk control and lower limb function is also supported by studies showing trunk control as a predictor of gait ability in children with CP and in adults after stroke.[89, 90]

2.3.5 Studies emphasizing trunk control during gait in children with CP

In the early 1990s, studies of gait analysis focused on hip, knee, and ankle kinematics, and the mainstream idea was that the upper body is a static "passenger unit" of a locomotor apparatus that is located in the lower limbs.[91] This idea has since been challenged by empirical evidence, as many studies have confirmed that the trunk plays a fundamental dynamic role during gait (2.3.3). According to these studies, which describe the function of the upper body during gait and the development of wearable wireless accelerometers for quantifying gait stability, the interest in trunk control during gait has increased during the past decade.

However, to my knowledge, there are a limited number of studies conducted with the main aim of assessing trunk control during gait of children with CP.[92-95] The few studies available have reported increased trunk displacement in the sagittal and/or frontal plane,[41, 92, 95] increased mechanical work in the head, arm, and trunk segment,[94] and altered trunk- and hip muscles' activation patterns (assessed with EMG)[93] in children with CP compared to children with TD. Moreover, a few studies have explored the effect of "trunk-targeted" interventions on different gait parameter in children with CP. Vibration of abdominal muscles has been found to increase both posture and the distance walked,[86] and another intervention, hippo therapy, has been reported to have an effect on gait speed and step length.[96, 97] Recently, some authors have studied trunk control during gait using optoelectronic and force plate motion analysis. A variety of parameters, including kinematics of the COM, the trunk, the upper

[4] Moe-Nilssen R. et al., 2005. Page 165.

limbs and head, center of pressure (COP), Floquet analysis, and a foot placement estimator, have been used to quantify trunk control. Several studies have reported displacement, velocity, accelerations, and variability in the AP, ML, and V directions of COM and COP in children with CP compared to TD children. More specifically, in children with CP, COM (displacement, velocity, and accelerations) was found to increase in the AP, ML, and V directions,[25, 98-101] and COP (displacement, velocity, and accelerations) in the ML direction.[25, 98] Children with CP also showed greater difference in COM and COP trajectories than TD children.[102] Moreover, kinematic analysis of the trunk showed increased forward tilt and increased range of motion in children with CP.[41, 92] The kinematic analysis of the upper limbs showed decreased arm swing on the least-affected side and the opposite on the most-affected side,[103] and a "guard position" with increased shoulder abduction and elbow flexion in children with CP.[92, 104] Kinematic analysis of the head showed greater variability of the head angle in the ML direction in children with CP.[105] Floquet analysis revealed that children with CP took wider steps and modulated their step length, compared to TD children in order to stabilize during gait.[106] Moreover, an assessment using a foot placement estimator showed marked instability in the AP and ML directions.[107] In addition, balance characteristics during gait has been assessed in CP and TD children using plantar pressure sensors in the shoes.[108] Symmetry and repeatability of the plantar pressure in children with CP were found to differ compared with the TD children.

Trunk-worn accelerometers may provide an alternative approach to assessing the gait characteristics of both progression and trunk control.[109] The method is less time-consuming and less expensive than 3D gait analysis and is not restricted to assessments conducted in a laboratory environment. The method is well established in assessments of adults,[109] but only two studies of children with CP have been identified in which a trunk-worn accelerometer was used.[24, 110] The latter two studies, which were conducted by the same research group, found that children with CP had higher accelerations of the COM, indicating impaired trunk control during gait, compared with TD children.[24, 110] The authors reported that the children with CP were able to walk at speeds comparable to those of the TD children, but had higher levels of trunk instability.

2.4 Gait interventions

In children with CP gait is usually assessed as part of the decision processes leading to interventions, and much effort is directed towards improving and maintaining their gait. In the following there will be some reflections on 'gait improvement' and interventions, followed by a brief description of the effect of some of these interventions.

What does "improving gait" actually mean? In a recent review covering the management of ambulatory children with CP, Narajanan, a pediatric orthopedic surgeon, answers: "*it may include improving speed and endurance, optimizing balance, preventing tripping and falls, reducing reliance on walking aids, reducing fatigue, and eliminating or preventing pain, with an expectation that this will preserve or improve their physical function and provide them with the ability to increase their participation in physical activities, recreations and sports.*"(5)84

To achieve the aforementioned goals, treatments may include physical therapy, orthoses, or more invasive methods. Physical therapy may include electrical stimulation and fitness, strength, and trunk control treatments, while invasive methods may include treatment with BoNT, intrathecal baclofen, and orthopedic surgery. These treatments are commonly used; for example, in Norway it has been registered that 62% of children with CP receive physiotherapy one to two times per week and 60% use orthoses.[111] The use of invasive methods are also common; 63-68% of the children with spastic CP have received BoNT treatment,[112] and 15% have received orthopedic surgery.[111] In a large study of gait abnormalities, Wren et al.[75] reported that 42% of the children with CP had undergone orthopedic surgery (including selective dorsal rhizotomy).

Despite the broad use of the above-mentioned interventions in the treatment of ambulatory children with CP, the evidence of their benefit has still not been adequately explored.[84] In a recent systematic review, Novak and coworkers[113] attempt to systematically describe the best available evidence for CP interventions, using the GRADE (Grading of Recommendations Assessment, Development and Evaluation) system, complemented by the Evidence Alert Traffic Light system, to provide knowledge translation guidance to clinicians about what to do. In the Traffic Light system, "green" represents strong level of evidence ("Apply the treatment"), "yellow" represents weak level of evidence ("Probably worth applying the

[5] Narayanan UG. et al., 2012. Page 173.

treatment") and "red" represents strong evidence for lack of effect ("Do not apply this treatment"). Only one of the interventions aimed to improve 'gait function', namely BoNT treatment, is given the green light. By contrast, orthopedic surgery and the use of orthoses are given the yellow light, thus indicating that more research is needed.

Nonetheless, Narayanan[84] claims that: "*The lack of evidence should not be interpreted as evidence of ineffectiveness, but should provide for larger, prospective multicenter, longitudinal controlled studies to generate higher quality evidence of effectiveness.* "[6] Another research group, comprising Rutz and colleagues,[114] has emphasized that there is considerable variation in the results regarding gait after surgery (single-event multilevel surgery (SELMS)), and that the factors responsible for this variation are poorly understood. However, some of the variation in the results of these studies may be related to impaired trunk control, one of several possible determinants of gait, which is seldom assessed in conjunction with gait assessment.[2] Gage and colleagues[115] are aware of this challenge and agree with Bleck's[116] statement: "*Of all the motor problems in cerebral palsy, deficient equilibrium reactions interfere the most with functional walking.* "[7] However, Gage and colleges[115] confess: "*To our shame, we have not instituted a formal balance-testing program at our hospital. It needs to be done*."[8]

Further detailed descriptions of potential interventions aimed at improving gait are considered beyond the scope of this thesis.

2.5 Assessment

In this section I address the concept of assessment from a clinical perspective today (2.5.1) and from a historical perspective (2.5.2). Thereafter I continue by addressing the importance of sound measurement properties of the assessments (2.5.3), and I introduce the International Classification of Functioning Disability and Health (ICF) as a model to analyze and categorize treatment outcomes (2.5.4). I complete the section with a description of assessment tools of trunk control (2.5.5).

[6] Narayanan UG. et al., 2012. Page 177.
[7] Bleck EE.et al., 1987. Page 32.
[8] Gage JR. et al., 2004. Page 184.

2.5.1 Assessment in a clinical perspective

An *outcome* is the end result of a process such as an intervention, and *outcome measures* are tools that may be used to assess change in particular attributes that are deemed meaningful to a person's life over time (called *assessment tools* in this thesis).[117] Assessment tools have been found to have important benefits to consumers of services (patients and their families), service providers, clinical managers, policymakers, and researchers.[118] The use of standardized tools serves to increase the accuracy of the assessment results, providing outcomes that are more reliable and reproducible.[119] However, the importance of objective documentation with the use of standardized tools has been stressed with the growing expectation of evidence-based practice.[120] Evidence-based practice (EBP) is defined by Sackett and colleagues[121] as the "*conscientious, explicit and judicious use of current best evidence in making decisions about care of individual patients*." However, EBP has also been described as the "*integration of best research evidence with our clinical expertise and our patient's unique values and circumstances*."[122] The latter definition emphasizes that there are different sources of information to apply in the process of clinical decision making.[120] In a historical perspective there have been different views of how to obtain knowledge.

2.5.2 Assessment in a historical perspective

Historically, it was essential to obtain "truth knowledge." For example, in the empirical tradition, in the 1700s, "truth knowledge" was obtained by observations through measuring and weighing under controlled conditions. Observations were considered to be independent of experience and theory, and neutrality and objectivity were considered as cardinal characteristics. Objective assessment, with no personal judgment involved, is still considered important, and in measurement theory, reliability, for example, has been defined as: "the proportion of the total variance in the measurements, which is due to 'true' differences between patients."[123] However, phenomenology, founded by Hussel (1859–1938) represented a different direction to empiricism. In this tradition, neutral and objective assessment has been considered impossible, since our earlier knowledge and experience is always included in an assessment.[124]

This thesis deals with assessment and assessment tools related to the empirical tradition, and the focus is on standardized assessment tools. Research has shown that the use of standardized measurement tools may lead to a shared understanding that facilitates communication.[125] However, it has been emphasized that quantification of outcomes and their determinants using

objective, standardized assessment tools is only one way to describe and characterize important information. Qualitative methods or combined methods can also be used to gather a more in-depth understanding of the construct of interest.[3] Moreover, when many measurement tools are available, it may be challenging to choose the most appropriate tool for a given situation. Evaluation of the quality of the assessment tools is then a core element when choosing the most appropriate tool.[8]

2.5.3 Measurement properties

The quality of an assessment tool is related to its clinical utility, generalizability, and measurement properties. *Clinical utility* has been related to clarity of instructions, format, time to complete assessment, qualifications, and cost,[126] whereas *generalizability* deals with the study population in terms of, for example, disease characteristics, age, and sex. However, if an assessment tool is applied in a study of a new population (i.e., another age group or another patient group) or situation or for another purpose, the *measurement properties* should be reassessed. The term measurement property (also called psychometric properties and clinimetrics) includes reliability, validity and responsiveness (**Figure 8**).[123] Both the methodological quality of the studies in which the measurement properties are assessed and the result of the studies are important, whereas the reported measurement properties of a tool can hardly be trusted if the methodological quality of the study from which the properties were obtained is poor. Moreover, the selection of instruments with good measurement properties will lead to the detection of smaller treatment effects or more power to draw stronger conclusions.[127]

Literature on measurement may be confusing due to wide variation in the names given to specific measurement properties and how they are defined.[8] The variation in terminology and definitions was one of the reasons that a research group recently initiated a study of how to achieve consensus-based standards for the selection of health measurement instruments (COSMIN). The COSMIN study aimed to reach consensus among approximately 50 experts with a background in psychometrics, epidemiology, statistics, and clinical medicine, regarding which measurement properties are considered important, their most appropriate terms and definitions, and how they should be assessed in terms of study design and statistical methods.[127] Several methodological guidelines for studies on measurement properties have

been proposed in the past, although none of them are widely used. It has been suggested that one reason may be that they are rather brief and not very user-friendly.[127]

The COSMIN group has proposed some criteria to assess the quality of studies of the measurement properties of health measurement instruments,[127, 128] and Terwee et al. have proposed some criteria to assess the results of studies of the measurement properties.[129] In this thesis, I used both the definitions used by the COSMIN group[123] and the "Terwee-criteria",[129] and further details are presented in Appendixes A and B.

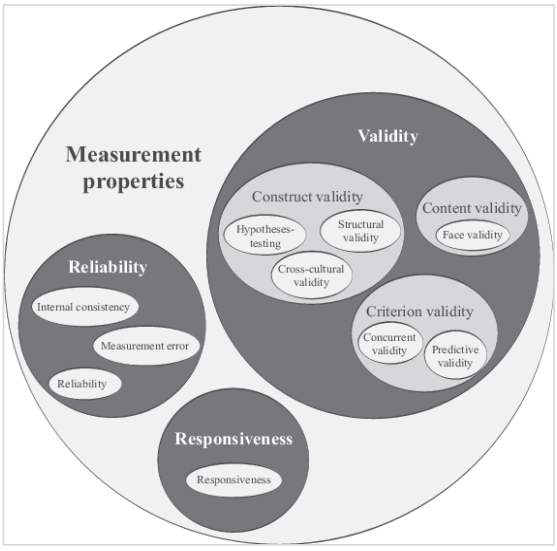

Figure 8. COSMIN's taxonomy of relationships between measurement properties (modified from Mokkink et al., 2010,[123] with permission from Elsevier)

2.5.4 International classification of functioning, disability, and health (ICF)

The ICF,[130] including the International Classification of Functioning Disability and Health for Children and Youth (ICF-CY),[131] provides a framework in which human functioning and disability are described as a dynamic interaction between various health conditions and environmental and personal factors (**Figure 9**). It provides a unified standard language that describes how people with a health condition function in their daily lives and is recommended as a model for analyzing and categorizing treatment outcomes. Moreover, this model may serve as a guide in the selection of assessment tools, treatment goals and outcomes of interests,[3] and is thus a useful framework in this thesis (***Paper I***).

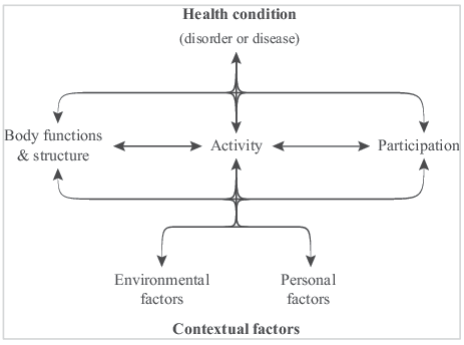

Figure 9.International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001.[130])

2.5.5 Assessment of trunk control

There is no universally established way of assessing trunk control, as this requires agreement on a definition of what to assess and/or measure: "What does the construct include?" Whereas weight and leg length can be 'directly assessed,' trunk control may probably be assessed both 'directly' and 'indirectly,' again depending on the definition (i.e., if defining trunk control during gait as "not falling," trunk control could easily be observed). However, other aspects of control may require more 'indirect measurements.'

Factors that cause or influence the outcome of interest (i.e., "gait outcomes") may be called *determinants* and the quality of trunk control may be one such determinant. The measurement of the outcomes and their determinants is said to be essential for informing decisions about treatment and targeting those who may benefit most.[3]

Sitting

In the absence of any overviews of clinical assessment tools for balance in children, adolescents, and adults with CP, it was deemed appropriate to conduct a review of the existing clinical tools and assessments that can be made without the need for laboratory equipment. The results of the review, on which this thesis is based, are presented in ***Paper I***[132]

Gait

A recent review identified 92 different quantitative measures of "gait stability" in the elderly[133] and in a more recent paper Bruijn and colleagues[134] have reviewed the current methods for assessing stability in human locomotion. Several of these methods were used in the relatively few studies aiming to assess trunk control during gait in children with CP (2.3.4)

(assessment of the COM, Floquet multiplier analysis, foot placement estimator, and variability measures). Most of the studies assessed the movements of the COM by optoelectronic and force plate motion analysis, although two studies assessed the movements of the COM with a trunk-worn accelerometer in children with CP.[24, 110] However, trunk-worn accelerometers have been used to assess trunk control during gait in other populations.[23, 27-29, 109, 135, 136] Despite the same assessment instrument being used, there was great variability in the outcome variables, such as accelerations (represented by root mean square),[24, 27, 29, 110, 135, 136] and variability measures (represented by the Lyapunov exponent, approximate entropy, the Harmony Index, and autocorrelations).[23, 24, 27-29, 110]

To summarize thus far, there is a need for assessment tools for trunk control and further assessment of trunk control during sitting and gait, as well as assessments of the relationship between the trunk control during sitting and gait.

Assessment tools are fundamental in research and clinical practice as they form the basis for diagnosis, prognosis, and evaluation of the results of interventions. In order to be able to trust the results of research it is essential that both the studies and the results of the studies of the measurement properties of the assessment tool are of good quality.[8] There are several methods for assessing balance during gait, but to date few studies have applied them in children with CP. The reason for this may be that, when planning "gait treatments, the main focus has been on the movements in the lower limbs in gait analysis.[2] The trunk has been considered as a "passenger unit,"[56] and movements of the trunk as compensations for impairments in the lower limbs. However, research has shown that controlling the trunk is a primary aim during gait,[21] and that trunk movement influences the moments in the lower extremities.[42] Thus, information on trunk control may be important for planning gait treatments. Research has shown that trunk control in children with CP is impaired both during sitting[48] and gait,[41] but the relationship between trunk control in these activities has not been not shown. If a relationship exists, there may be a possibility to use a short and easily applicable trunk control test during sitting in clinical practice when planning gait treatments.

3. Aims of the thesis

The main aim of this thesis is to contribute to better assessment and understanding of trunk control in children and adolescents with CP. The specific aims of the studies on which this thesis is based were as follows:

Paper I: To identify tools used in clinical practice with the aim of assessing trunk control in children, adolescents, and adults with CP, to describe the content of the tools, and to evaluate the quality of both the studies and the results of the measurement properties.

Paper II: To evaluate inter- and intra-observer reliability and construct validity of the TIS (assessed during sitting) in children and adolescents with cerebral palsy.

Paper III*:* To investigate characteristics of gait in children and adolescents with CP compared with TD children, with the main focus on trunk control, using data generated from a trunk-worn accelerometer.

Paper IV: To investigate the relationship between trunk control during sitting and gait in children and adolescents with cerebral palsy.

4. Material and methods

4.1 Study design

The four papers which form part of this thesis focus on assessment tools and assessment of trunk control in children with CP. ***Paper I*** is a systematic literature review of clinical tools used to assess trunk control in children and adults with CP. ***Paper II*** is a method study with a cross-sectional design, and ***Papers III and IV*** are observational studies, also with a cross-sectional design.

4.2 Study population

All children with CP included in the studies in this thesis were recruited from the neuro-orthopedic outpatient clinic at St. Olavs University Hospital (Trondheim, Norway), and children with no motor impairment were recruited from several mainstream schools. Exclusion criteria for all studies were treatment with BoNT in the lower extremities during the preceding 4–6 months and/or surgery during the preceding 12 months.

Paper I[132] is a systematic review of a total of 216 papers, of which 35 of the papers and 22 assessment tools were included for quality assessment. Papers were included in the review if they described a tool that met the following criteria: (1) the tool was designed for use in a clinical setting, assessment by a clinician in hospital or community, without the need for laboratory equipment; (2) the measurement properties of the tool had been evaluated in subjects with CP who were older than four years of age (since most children with CP are diagnosed by this age)[137] and (3) the paper was published in an English peer-reviewed journal or thesis. Papers were excluded if (1) the primary intention of the tool was not to assess trunk control, the trunk control assessment was part of a wider assessment of motor function (i.e., if a specific "trunk control score" could not be extracted from the assessment); (2) subjects with cerebral palsy comprised less than 30% of the total population; (3) they were reviews; and (4) they were case studies.

Paper II[138] reports a study that included a consecutive sample of 46 children in the age group 5–19 years: 37 children with CP (all subtypes within GMFCS Levels I–IV) and 9 children with typical development (controls). Among the 46 children, 25 had participated in our previous reliability study,[7] while for the study reported in Paper II the reliability of the TIS in

adolescents was assessed in 17 additionally recruited children with CP in the age group 13–19 years. However, construct validity was not addressed in our earlier study, and all 46 children (5–19 years) were therefore included in the validity part of this study. To qualify for inclusion, the children had to be able to able to sit on a bench without support and to understand instructions (**Figure 10**).

Paper III[139] reports a study with a consecutive sample of 70 children in the age group 5–18 years: 41 children (24 males) with spastic CP and 29 (13 males) children with no motor impairment. To qualify for inclusion, participants had to be able to understand certain instructions and to walk at least 10 m without support, shoes, or orthoses (**Figure 10**).

Paper IV[140] reports a study that included a consecutive sample of 26 children with spastic CP (17 males) in the age group 8–18 years (**Figure 10**).

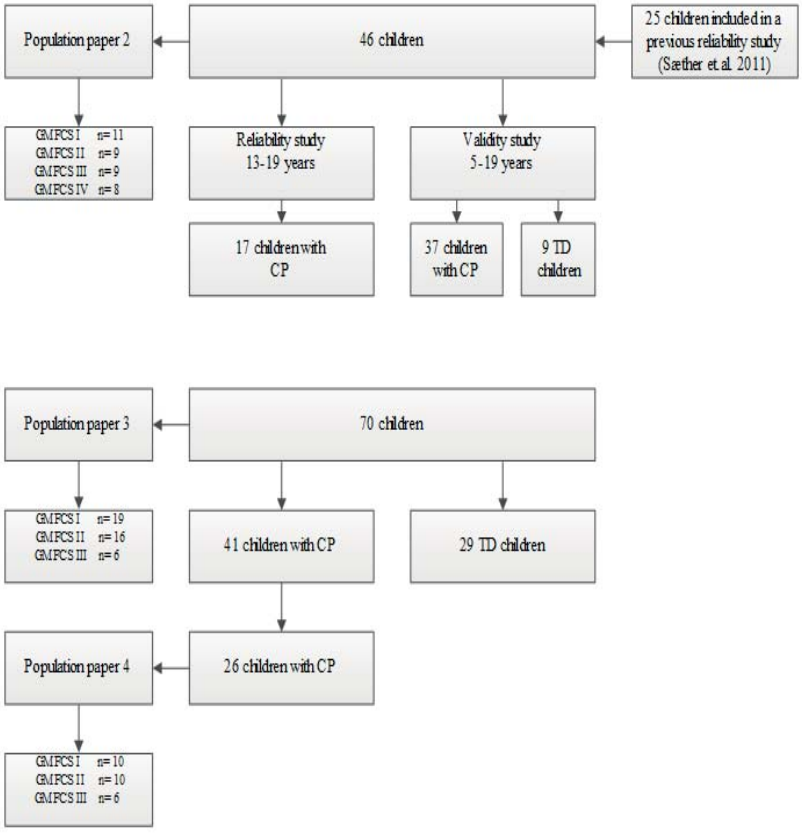

Figure 10. Study populations in Papers II–IV

4.3 Methods

Trunk control during sitting was assessed by the TIS in the study reported in *Paper II* and in addition the Trunk Control Measurement Scale (TCMS) was applied in the study reported in *Paper IV*. Trunk control during gait was assessed by a trunk-worn accelerometer in the studies reported in *Papers III and IV*.

4.3.1 Assessment of trunk control during sitting

Trunk control during sitting was tested with the Trunk Impairment Scale (TIS) and the Trunk Control Measurement Scale (TCMS). The TIS was originally developed to evaluate trunk control in adults after stroke, through assessment of static- (SSB) and dynamic balance (DSB) and trunk coordination (C) in the sitting position.[6] The total score—the sum of the three subscale scores—ranges from 0 (lowest performance) to 23 (best performance).[141] The TIS has been tested for reliability and for validity in children and adolescents with CP in the age group 5–19 years.[7, 138]

The TCMS is a further development of the TIS and was expanded to include assessment of selective trunk movements and dynamic reaching. The total score ranges from 0 (lowest performance) to 58 (best performance), where the total score is the sum of the three subscale scores: static sitting balance (SSB), dynamic sitting balance-selective movement control (DSB-S), and dynamic sitting balance-reaching (DSB-R). The TCMS has been tested for reliability and validity in children with CP, 8-15 years.[142]

4.3.2 Assessment of trunk control during gait

Trunk control during gait was tested with an accelerometer attached over the L3 region of the lower back with double-sided tape to acquire accelerometer and orientation data during gait. The sensor, a six degrees-of-freedom inertial sensor (MTx. XSens, Enschede, NL) (weight: 15 grams), contains tri-axial units of accelerometers, gyroscopes, and magnetometers and is connected to a battery-operated communication unit (weight: 300 grams), which was also worn by the children. Data were acquired at a sampling frequency of 100 Hz and transmitted in real time to a laptop by Bluetooth technology.[143] Gait time was registered by photoelectric cells synchronized with the accelerometer device. The trunk-worn accelerometer has

previously been tested for precision and accuracy,[144] test-retest reliability of accelerations,[145, 146] spatiotemporal parameters,[145] and variability measures[28] during gait in healthy adults.

4.3.3 Data acquisition and analysis of gait variables

A customized software application TRASK, run under Matlab R2011b (Math Works Inc., Natrick, MA), was used for signal processing and calculation of gait variables. By applying the accelerometer as an inclinometer, the average tilt of the measuring axes could be calculated for each sample, thus eliminating gravity bias.[147] The raw acceleration data were transformed into a horizontal-vertical coordinate system by a trigonometric algorithm and reported along the AP, ML, and V axes.[148] Trunk acceleration amplitudes for each gait interval were expressed by root mean square (RMS) values (hereafter referred to as trunk accelerations). The periodicity of the acceleration curve enabled steps and strides to be registered. Interstep and interstride trunk acceleration regularity were calculated using an unbiased autocorrelation procedure, in which an acceleration time series was correlated to the same series at a phase shift equivalent to one step and one stride. Perfect replication of the signals between consecutive steps or strides gives an autocorrelation coefficient of 1.[149] Trunk asymmetry was calculated for each of the AP, ML, and V axes by subtracting the interstep regularity from interstride regularity, thus reflecting differences in regularity between left and right steps beyond the regularity between strides. A perfect symmetry has the value of zero, whereas a positive value indicates an asymmetric gait pattern.[150] Step time and cadence for each walk was estimated from the autocorrelation curve of the vertical axis. Mean step length for each walk was calculated as gait distance and number of steps. The mean value of two walks (back and forth along the pathway) at preferred speed was used to compare gait variables between children with CP and children with TD. A mean of 9.6 (SD1.9) (***Paper III***) and 9.4 steps (SD 1.7) (***Paper IV***) was used in the calculations for the study populations.

4.3.4 Other variables

Information on gross motor function, described by the GMFCS,[16] and the classification of CP subtypes was obtained from hospital medical records for each child.

Each participant's height was measured to the nearest cm with a stadiometer, and their weight was measured to the nearest 0.5 kg using an electronic weight (Seca digital 770) while they

were wearing light clothing. The child's body mass index was then calculated using the formula: weight (kg)/height^2 (m^2).

4.4 Ethics

The studies in this thesis were performed in accordance with the Helsinki Declaration and approved by the Regional Committee for Medical Health Research Ethics: ***Paper II*** (Ref. nr. 2009/811-22, 2010/ 1889-8 and 2010/1889-11. ***Papers III*** **and** ***IV*** (Ref. nr. 2010/1991).Written informed consent was obtained from the participants and their parents prior to participation in the study.

4.5 Data analysis and statistical methods

Paper I is a systematic literature review and the searches were performed in CINAHL, Embase, PubMed, and MEDLINE up to 30 November 2012. The first search aimed to identify clinical assessment tools for assessing trunk control in cerebral palsy (details of the database search are presented in online supporting information, S1). Subsequently, the names of the tools identified during the first search were used in a complementary search, which aimed to identify additional studies of the *measurement properties* of assessment tools of trunk control.

The *characteristics* of the clinical trunk control tools, namely the focus of assessment, clinical utility, scale construction, and characteristics of the study population, of the included clinical trunk control tools were extracted into a pre-established review table, adapted from the CanChild Outcome Measures Rating Form.[126]

The *methodological quality of the studies* was rated according to the COSMIN.[151] The COSMIN checklist consists of nine measurement properties, with 5–18 items per measurement property, which address aspects of design and statistical methods. The measurements properties are internal consistency, reliability, measurement error, content validity, construct validity (structural validity, hypothesis testing, and cross-cultural validity), criterion validity, and responsiveness.[151] A study's quality is rated on a 4-point rating scale ("poor," "fair," "good," or "excellent") for each of the nine properties.[128] In accordance with the COSMIN, the overall quality score per measurement property was determined by the lowest rating of any of the items ("worst score counts"). A high level of inter-rater agreement for the COSMIN checklist has been documented.[152]

The *results obtained in each study* regarding the value of the measurement properties were assessed using Terwee's criteria.[129] The results of the studies were rated as "positive," "indeterminate," or "negative."

In order to determine an "overall score" for each measurement property per clinical trunk control tool, the *level of evidence* was estimated in a similar manner to that recommended by the Cochrane Back Review Group[153] and used in a number of reviews of measurement properties (Appendix C).[154-156] The *level of evidence* combines *number of studies*, *consistency of rating of results* according to Terwee,[129] and *quality of studies* according to the COSMIN [128] in a rating of the documented measurement properties per clinical trunk control tool as "strong," "moderate," "limited," "unknown," or "conflicting."

Paper II reports a method study that evaluated the reliability and validity of the TIS. *Relative reliability* was assessed by calculating an Intraclass Correlation Coefficient (ICC). The intra- and inter-observer reliability of total TIS score was assessed using ICC (1,1) and (3,1). For the items in the "static sitting balance subscale," "dynamic sitting balance subscale," and "coordination subscale" score, kappa and weighted kappa statistic (κ) were used to test agreement, or agreement was expressed as percent agreement if the κ value could not be calculated. To describe *absolute reliability*, the standard error of measurement (SEM) was calculated as the square root of the mean within-subject variance. The consistency of the measurements was also verified graphically using Bland and Altman plots[157] for the total TIS score.

Paper III reports an observational study that assessed gait characteristics in children with CP compared to TD children. Gait parameters for the CP and the TD group were compared using a three-step procedure: first, unadjusted, using Student's t-test; second, adjusted for the potential confounders gender, age, height, and BMI, one at a time, in linear regression, and with only confounders changing the coefficient for group more than 10% included in the model from this step; and third, since many gait variables have been shown to be associated with gait speed,[158] the role of gait speed was assessed as a possible mediator by adding it as a covariate. The results of the unadjusted analysis were also calculated in terms of standard deviation scores ("z-scores") for each gait variable, using the mean value and SD in the control group as reference.

Differences between the CP and the TD group in gait strategy with increasing gait speed were assessed using a linear mixed model with acceleration at each of three velocities (slow,

preferred, and fast gait) as a dependent variable, group, and measured velocity, their interaction as fixed effects, and subject as a random effect.

The TD children were also compared with children with different GMFCS levels with respect to differences in gait parameters in two steps: first, by One-Way Analysis of Variance (ANOVA); and second, if the ANOVA indicated a statistically significant difference between the groups, a Scheffe's post-hoc test was applied to localize the difference. The same procedure was used to compare TD children with the two CP subtypes (unilateral and bilateral). Finally, the two CP subtypes were compared with respect to the differences in gait parameters using the Student's t-test.

Paper IV reports an observational study that assessed the relationship between trunk control during sitting and gait. The relationship was first assessed by calculating the Pearson's (R) correlation coefficient. To control for the potential confounders age, height and gait speed, the partial correlation coefficient (R_p) was calculated.

5. Main results

Paper I: Clinical tools to assess balance in children and adults with cerebral palsy: A systematic review

Paper I presents a review of tools used to assess trunk control in clinical practice in children and adults with CP, a description of their content and measurement properties, and an evaluation of the quality of the studies that have examined these properties. The COSMIN was used to assess the quality of studies and the Terwee criteria were used to assess the result of studies, and a Modified CanChild Outcome Rating Form was used to describe the content.

Abstracts from 1529 papers were evaluated and 216 papers were included for a full text check. Finally, a total of 35 papers and 22 clinical trunk control tools were included for quality assessment. The content and focus of the tools varied significantly. Moderate or limited evidence was found for most of the measurement properties of the tools. The strongest evidence was found for the TCMS and the Level of Sitting Scale (LSS) in the category "maintain balance," and the Timed Up and Go (TUG) and the Segmental Assessment of Trunk Control (SATCO) in the respective categories "achieve balance" and "restore balance." Information on the measurement property responsiveness was scarce.

Paper II: Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy

Paper II presents findings relating to the intra- and inter-observer reliability in adolescents (13–19 years) and construct validity of the TIS for children and adolescents (5–19 years). Video recordings of 17 children with CP classed as GMFCS Levels I–IV were analyzed by three observers on two occasions. For construct validity, the TIS was compared with Gross Motor Function Measure (GMFM) in 37 children classed as GMFCS Levels I–IV. High intra- and inter-observer reliability was found, with high ICC and kappa values. However, the smallest detectable difference was somewhat high, partly due to consistently lower TIS scores made by one observer. The construct validity was considered good, as there was a high correlation between the TIS total score and the dimension scores of the GMFM. The findings, reported in Paper II, suggest the TIS is a reliable and valid measure of trunk control for both children and adolescents with cerebral palsy.

Paper III: Gait characteristics in children and adolescents with cerebral palsy assessed with a trunk-worn accelerometer

Paper III presents gait characteristics reflecting trunk control and progression in children and adolescents with CP compared with TD children. In the study, a total of 70 children—41 children (24 males) with CP and a gross motor function corresponding to GMFCS Levels I–III, and 29 TD children (13 males) in the age group 5–18 years (mean 11.1 years)—walked back and forth along a 5 meter walkway. A tri-axial accelerometer worn on the lower back was used to assess their gait characteristics. Data were recorded along the AP, ML, and V axes. To assess the magnitude of potential differences in gait characteristics, standard deviation scores (z-scores) were calculated using TD children as reference. Gait parameters related to trunk control, such as accelerations, were higher in all three directions in children with CP (z-scores between 0.4 and 0.7) than TD children and increased with increasing GMFCS levels. The differences in accelerations in the AP and V directions increased between children with CP and TD children with increasing speed. Also asymmetry in trunk accelerations differed significantly between the two groups in all three directions (z-scores were between 0.8 and 1.8 higher in the CP group), whereas interstride regularity differed only slightly between the groups and only in the AP direction. Gait characteristics also differed between children with the spastic unilateral and bilateral CP subtypes for accelerations and asymmetry in the AP and ML directions. In contrast to the findings related to trunk control during gait, there were no differences between the groups in parameters related to progression. The results show that the children with CP had significant difficulties with trunk control, but not with progression during gait, and that these problems increased with both increased gross motor impairment and speed.

Paper IV: The relationship between trunk control during sitting and gait in children and adolescents with cerebral palsy

Paper IV reports a study that assessed the relationship between trunk control during sitting and gait in 26 children with CP (17 males) with a mean age of 11.7 years (range: 8–18 years) and gross motor function corresponding to GMFCS Levels I–III. Trunk control during sitting was assessed with the TIS and the TCMS, and trunk control during gait by a tri-axial accelerometer worn on the lower back when the children walked back and forth along a 5 meter walkway at preferred speed. Gait variables representing trunk control were trunk accelerations and interstride regularity in the AP, ML, and V directions. It was found that

trunk control during sitting, assessed with the TCMS total score and TIS DSB, correlated "moderately to good" (R_p = 0.67 and 0.66, respectively) with trunk control during gait. Moreover, some subscale scores correlated nearly equivalent (TCMS DSB-R) subscale score) or even higher (TIS DSB) with trunk control during gait. The results suggest that two subscales of these tools, which are less time-consuming, may be applied in clinical assessment of trunk control. Future studies are needed to explore how this information may be applied in the planning of interventions aimed to improve gait performance in children and adolescents with CP.

6. Discussion

6.1 Main findings

In the first part of the study (***Papers I and II***) 22 clinical tools for the assessment of trunk control in children and adults with CP were identified through a systematic literature review. However, there was moderate or limited evidence for the measurement properties of these tools, and scarce information on the measurement property responsiveness. Results from the method study of the TIS (one of the tools identified in the review), showed high intra- and inter-observer reliability, and the construct validity was considered good.

In the second part of the study (***Papers III and IV***) it was found that children with CP had significant difficulties in trunk control during gait, as reflected in higher trunk accelerations in the AP, ML, and V directions and lower regularity in the AP direction when compared with TD children. Moreover, these problems seemed to increase with increasing gross motor impairment and increasing speed. A final finding was a moderate relationship between trunk during sitting assessed with both the TCMS total score and TIS DSB, and trunk control during gait assessed with an accelerometer. Moreover, some subscale scores of the sitting trunk control tests correlated almost equivalent with trunk control during gait. Before I discuss the results, I will address some aspects related to the validity of the results.

6.2 Validity

In this section I discuss the construct, chance, bias, confounding, methodical considerations and generalizability.

6.2.1 The construct

In research, constructs (such as trunk control) have to be operationally defined, and the manner in which this is done may affect the validity of a study. In the research related to trunk control there is a lack of a universal definition and how the construct should be operationalized, which has been stressed by some researchers in the field.[31]

In ***Papers I and III*** the term "balance" is used. The rationale behind the usage was that the term is to a large extent universally known. However, in these papers trunk control is treated

as synonymous with balance, the rationale being that the COM is located in the trunk, and that analysis of trunk motions gives information on balance.[21] In the systematic review, ***Paper I,*** balance is described as the act of maintaining, achieving, or restoring the COM relative to the base of support. The lack of a universally accepted definition has resulted in variety of terms being used by researchers in the field, which in turn made the identification and selection of relevant tools challenging. To avoid selection bias, a comprehensive search strategy was adopted and two searches conducted (described in section 4.5).

For the studies reported in ***Papers III and IV*** trunk control during gait was operationalized as asymmetry, accelerations (RMS), and regularity (autocorrelation). However, asymmetry was found to be more related to the CP subtype (unilateral) than to trunk control. Both the choice of parameters representing trunk control and the interpretation of the results were related to earlier studies of subjects with impaired trunk control. Higher average dispersion of accelerations have been found in subjects with impaired trunk control,[24, 25] while regularity (increased variability) has been regarded as a indicator of both impaired and improved control (section 2.5.5).[28] Thus, researchers may interpret the results of assessments of trunk control during gait differently.

6.2.2 Chance

The low p-values indicate that it is unlikely that the main findings in this thesis are due to chance. However, due to the relatively small sample size, the results should be interpreted with caution. No a priori power calculation were made for the studies in ***Paper III*** (n-CP = 29, n-TD = 41) and ***Paper IV*** (n = 26) as there were no earlier studies of children in the age group 5–18 years with both unilateral and bilateral CP and with outcomes based on assessment with a trunk-worn accelerometer.

Sample size estimation for reliability (***Paper II***) is not issue of statistical significance because it concerns whether the reliability parameter approaches 1, not its statistical difference. However, an adequate sample size is important to obtain an acceptable confidence interval around the estimated reliability parameter.[8] Guidelines for the calculation of sample size are scarce, although de Vet et al.[8] and Bonett[159] have described how many measurements (or observers) per patient are necessary to reach a specific confidence interval for the estimated parameter. Bonett[159] suggests that, for example, for a sample of 21 children a confidence interval of 0.2 for a reliability parameter of 0.9 should be obtained. The COSMIN group

suggest that a sample size of more than 30 is required to achieve fair ratings and more than 50 to reach good ratings.[128]

The sample size in ***Paper II*** (n = 17) should ideally have been larger, but in accordance with Bonett,[159] the high correlation coefficients (ICC 3,1 from 0.94 to 0.98) and narrow confidence intervals (from 0.85 to 1.00) indicate that the sample size was sufficient to justify the conclusion that the TIS has high intra- and inter-rater reliability.

6.2.3 Bias

Bias can be defined as any systematic error that results in an incorrect estimate of the association between exposure and outcome.[160]

Selection bias

Selection bias is a systematic error that stems from the procedure used to select the subject of study.[160] In the research for this thesis there may have been three elements of selection bias: one related to the selection of children for inclusion in the studies in ***Papers II–IV*** and two related to language and focus in the systematic review (***Paper I***).

A consecutive sample of children with CP was recruited from the neuro-orthopedic outpatient clinic at St. Olavs Hospital (Trondheim, Norway) (***Papers II–IV***). One could speculate that children visiting this clinic are a representative sample of children in need of special orthopedic treatment. However, regular neuro-orthopedic follow-up is recommended for children with CP in order to prevent contractures and deformities.[161] Thus, it is unlikely that results reported in ***Papers II–IV*** were affected by selection bias.

A limitation of the systematic review (***Paper I***) may be the inclusion of studies published in English only, and tools developed by researchers publishing in other languages may have been missed. A second limitation may be the exclusion of tools that assess trunk control in laboratory settings, such as EMG and kinetic and kinematic analyses.[162] However, it was considered that a separate systematic review of such tools would have been more appropriate, since it has been recommended that informative reviews should be restricted to a limited number of tools and with more details on each tool, rather than larger reviews with limited information.[8] Further, clinical trunk control tools for children less than four years of age were excluded from the review because was deemed more appropriate to review them in a separate study.

Information bias

Systematic error in a study can arise if the information collected about or from the study subjects is erroneous.[160]

In the research for this thesis there may have been *misclassification* of the GMFCS level in the children with CP. However, the reliability of the GMFCS classification has been found to be high,[16] and the classification was done by experienced physiotherapists with a thorough knowledge of the child in each case. Misclassification is unlikely in ***Paper II*** as a good relationship was observed between the GMFCS classifications and the results of the test of gross motor function conducted using the gross motor function measure (GMFM). In the study reported in ***Paper III*** a potential misclassification may have diluted the differences in gait parameters between children at different GMFCS levels. Thus, the main findings of this thesis are unlikely to be due to misclassification.

Prevention of potential bias may be accomplished through careful study design. In the study reported in ***Paper I*** two of the authors extracted data and conducted the quality assessment independently in order to avoid information bias. In the study in ***Paper IV*** the examiner conducted the trunk control test blind from the results of the gait parameters. By contrast, the study design reported in ***Paper II*** was not optimal because the physiotherapist who obtained the video recording and instructed the children also participated in the video-based scoring of the TIS. She scored consistently lower than the observers who only watched the video recording of the children. The differences in scores obtained by the physiotherapist may thus be at least partly due to the fact that this observer had "more information" on the child, as she both instructed and observed the child on video. For the study in ***Paper III*** the relatively short walkway (5 meters), with also a relatively short acceleration pathway (2 meters) before and after the measurements were obtained, may have influenced the results of the study. The findings indicate that the main problem of gait control in children with CP is related to impaired trunk control, and may to a lesser extent be related to progression. It should not be overlooked that children with CP and TD children need different times to reach their preferred speed, and that they may start to decelerate at different times. Such differences may have contributed to the similarity in the progression parameters.

6.2.4 Confounding

If the results of a study cannot be attributed to chance or bias, the possibility of confounding should be considered. In order to control for possible confounding factors (i.e., factors associated with both independent (CP) and dependent (trunk-control parameters) variables; multivariable analysis, linear regression, and partial correlation were applied in the studies on which this thesis is based.

The multivariable analyses (***Paper III***) indicated that the results could not be explained by differences in age, sex, height, BMI, or gait speed. In the partial correlation analysis (***Paper IV***), the relationship between trunk control during sitting and during gait increased when gait speed was included as a covariate.

Including children who had been treated with BoNT or had undergone surgery may have been a potential confounder of the findings in ***Papers III*** **and** ***IV*** (not mentioned in either of the papers). The effect of such interventions on the movements of the COM has, to my knowledge, only been assessed in one study[163] In the study Massad et al.[163] found that surgery (on equines) reduced the vertical COM displacement during gait in children with CP (the treatment with BoNT did not show such an effect). In the study referred in ***Paper III*** 83% of the children at GMFCS Level III had undergone surgery at an earlier date, whereas 52% and 62% of the children at GMFCS Levels I and II had undergone surgery, respectively (unpublished data). If the surgery had resulted in greater improvements to the COM accelerations in children classed as GMFCS Level III compared to the children classed as GMFCS Level II (who had had undergone less surgery), it is possible that including children with orthopedic surgery may have diluted the differences in trunk control between the groups, thus diminishing the strength of the association between CP and trunk control found in the study. According to the findings reported in ***Paper IV***, the effect of surgery may potentially have weakened the relationship between trunk control in sitting and during gait.

6.2.5 Methodical considerations

In the following subsections I discuss the methodical issues related to Papers I–IV.

Paper I

The COSMIN[128] and the Terwee criteria[129] were used to rate the quality and the results of studies of measurement properties of trunk control tools, respectively. The ICF[130] was used to describe the "focus" of the tools and level of evidence to determine the overall score of each measurement property of the tools.

The methodological quality of the studies included in the review was rated according to the COSMIN four-scale rating system.[128] Some other methods have been developed to assess the quality of studies of measurement properties, but these are seldom used which, possibly due to the lack of user-friendly checklists and lack of evaluation of reliability and validity.[127] However, the reliability of the COSMIN four-scale rating system has not been assessed to date, and this is a limitation of the study reported in Paper I, although the four-scale rating system has been developed from the COSMIN checklist, which has a high level of inter-rater agreement.[152]

The COSMIN checklist is based on the "worst score counts" principal. This may be considered a too strict evaluation, and the COSMIN Delphi panel (which developed the checklist) considered that the methodological quality of a study should be rated as good when most (but not all) items are adequate, and poor when a defined number of items are inadequate.[128] However, a study may not be regarded as better than its "weakest link," and in future studies the COSMIN checklist may be used as a guide when designing studies of measurement properties, in order to ensure better quality.

However, the COSMIN checklist may not be optimal for all measurement properties, as content validity was rated as excellent for most of the tools in the reported study in Paper I. An example of one item from the COSMIN checklist regarding content validity is: "Was there an assessment of whether all items refer to relevant aspects of the construct to be measured?" In the studies reported in Paper I, we found it difficult to apply the four-point scale when scoring this item, as the "excellent" and "fair" alternatives were the most relevant and there was no alternative to rate it as somewhere between the two alternatives (i.e., as "good").

In Paper I the focus of each tool corresponding to the function, activity, or participation domains according to the ICF was described. To be more precise in the determination of which domain or combination of domains the tool actually measures, the most relevant components of an assessment could have been identified by applying the Linking Assessment Items to the ICF,[164] whereby each item of the assessment may be assigned an ICF code.

To my knowledge, there are no consensus-based criteria available for assessing the adequacy of a measurement property. Quality criteria have been suggested by Terwee et al.,[129] and were applied in the study in Paper I. These criteria combine the standards for methodological quality (such as study design) with the adequacy of the results (i.e., ICC > 0.70), and thus there is some overlap with the COSMIN rating system regarding study design. Moreover, there may not be one relevant criterion (for a specific measurement property) for all research. One solution may be to reach an agreement on certain criteria and if either stricter or more lenient criteria are applied researchers would need to provide and explanation for the usage.

In order to determine an overall score for *each* measurement property per clinical trunk control tool, the *level of evidence* was estimated (Appendix C). The last step to be carried out when conducting a systematic review is the synthesis of data, either to draw an overall conclusion about the quality of an assessment tool to measure a specific construct or to select the "best" assessment tool.[8] However, the inherent heterogeneity of CP populations poses a limitation for the synthesis of data, since ideally only results obtained in homogeneous populations should be combined.[8]

In the synthesis of data, all measurement properties should be considered together, and the number of studies in which the measurement properties of the tool is investigated, the methodological quality of those studies, and the results of the studies should be taken into account.[8] For example, when low scores for a reliability parameter (e.g., ICC < 0.4) are found in a number of studies of good methodological quality, there is strong evidence that the assessment tool has low reliability, but when high internal consistency is found in a number of studies of fair quality, there is only moderate evidence of high internal consistency.[8] However, to date, no well-designed methods have been established to combine the evidence of measurement properties from different studies.

The strengths of the systematic review in Paper I are that both the methodical quality of the studies of the measurement properties and the results of the studies (i.e., the reported measurements properties) were assessed by utilizing existing methods.[128] This point is important, as described in the example above, because the reported measurement properties of a tool can hardly be trusted if the methodological quality of the study from which the properties were obtained is poor.

Lack of good reporting of primary studies is a problem when conducting a systematic review. Poorly reported studies will limit a reader's ability to assess the methodological quality of a

study. However, relevant information can be deduced from the COSMIN checklist,[128] and the recently published guidelines for reporting reliability and agreement studies (GRRAS).[165]

Paper II

For the study reported in Paper II, video assessment was used in examinations of intra- and inter-observer reliability of the TIS. The method was considered a strength of the study because video assessment ensured that the variability of the scoring was unrelated to the child's performance or the instructions. However, there may be a need for future studies to assess the reliability of a "real-time" scoring of the TIS.

Paper III and IV

For the studies in Paper III and IV, trunk control during gait was assessed with the use of a trunk-worn accelerometer. A limitation of both studies was the lack of information on the validity and reliability of the use of trunk-worn accelerometer in children with CP, although high reliability has been reported in studies with healthy adults.[144-146] Moreover, in the case of the study reported in Paper III, the documented differences between subgroups in children with CP may indicate face validity in children with CP. An advantage of applying a trunk-worn accelerometer is the 'direct' measurement of 3D accelerations, which eliminates errors associated with differentiating displacement and velocity data.[109] Data measured from small inertial sensors attached directly to the body have the advantage of identifying human motion in a wide variety of environments and such sensors do not interfere with a child's movements.

6.3 Trunk control in children with CP

In the preceding section I have addressed some limitations of the validity of the studies on which this thesis is based, yet it is unlikely that the main findings were due to chance, bias, or confounding. Thus, the internal validity of the studies included in this thesis may be acceptable, and in the following subsections I therefore discuss some implications of the main findings.

6.3.1 Strength of the associations

The findings of the study reported in ***Paper III*** indicate that there are considerable differences in trunk control between children with CP and TD children during gait, as suggested by

differences in the asymmetry variables between 0.8 and 1.8 standard deviations, while deviations in the acceleration variables were between 0.4 and 0.7 standard deviations and for AP stride regulatory around 0.3 standard deviations. The accelerations during gait increased among children with CP with increasing GMFCS levels, which is consistent with increasing difficulties in gait with increasing impairments in gross motor function. By contrast, the largest deviations in the asymmetry variables were observed among children classed as GMFCS Level I, with no further deviations for children classed as GMFCS Levels II and III. This finding may be explained by the fact that all children classed as GMFCS Level I had spastic unilateral CP. The results regarding asymmetry were most similar for TD children and children classed as GMFCS Level III, which may be due to the fact that the latter had bilateral CP. Thus, although gait asymmetry is considered to be a good indicator of most gait abnormalities,[166] the results suggest that it may reflect other aspects than trunk control during gait.

In contrast to the marked differences in trunk control during gait between children with CP and TD children, all variables suggesting progression showed less than 0.5 standard deviations between the groups (***Paper III***). Thus, the findings indicate that the main problem of gait control in children with CP is related to impaired trunk control, and may to a lesser extent be related to progression.

The association between trunk control during sitting and gait was only moderate. However, it is not reasonable to expect a good to excellent correlation between the two, since they are two different tasks (***Paper IV***). Thus, the moderate to good correlation found in our study suggests that trunk control during sitting, assessed with the TCMS total score and the TIS DSB may provide valuable information regarding the primary impairments of trunk control.

6.3.2 Consistency with other studies

Systematic reviews of trunk control

In 1997, Westcott et al.[167] published a paper evaluating theories and assessment tools for "postural stability" in children in general. Later, a review published by Harris et al.,[168] addressed the efficacy and effectiveness of physical therapy in enhancing "postural control," and Chung and colleagues[169] reported the effectiveness of adaptive seating in sitting posture and postural control in children with CP. Several assessment tools were applied in the above-

mentioned papers. However, the review in Paper I[132] is the first to identify and evaluate clinical trunk control tools in children and adults with CP.

Recently, two published reviews, one by Field et al.[170] and one by Banas et al.,[171] identified and evaluated clinical tools to assess "sitting balance" in children with motor impairments and children with CP, respectively. The inclusion criteria in the two reviews and in the review in ***Paper I*** were dissimilar, and thus there were some variations in the included sitting assessments. However, the literature searches seem to have identified mainly the same tools, despite the lack of a universally accepted definition of the construct (six of the tools were included in all the three reviews). There was some similarity in the methods used for evaluating the tools and the studies of the measurement properties. The COSMIN checklist or the 4 point scale was used in all three studies. The main conclusion of the studies of Field et al.[170] and Banas et al.[171] was consistent with the findings reported in ***Paper I***, namely that studies of responsiveness are scarce, which means that the tools may not yet be applicable in evaluations of the results of trunk control interventions.

A recent study, conducted by Pavo and colleagues,[172] had the ambiguous aim of reviewing papers assessing postural control in children with CP, with a focus on both describing methods to assess it (identical to the aim of in ***Paper I***) and characterizing the children's motor responses. Pavo et al.'s [172] paper reports mainly laboratory methods, such as EMG and kinetic and kinematic measures, and mentions only two clinical assessments. Thus, the authors highlighted the lack of studies using clinical tests that are readily applicable. One reason for this may be that there is a lack of responsive clinical assessment tools, which is a prerequisite for measuring change, as reported in ***Paper I*** and in the above-mentioned reviews by Banas et al.[171] and Field et al.[170]

However, a disadvantage of the clinical tools, identified from the above reviews, is that they make measures at an ordinal level, which means that the items are ranked but the distance between the items is not known. Thus, when measuring change, one cannot know how much a child has changed, yet such information becomes available when using scales at an interval level. However, there are now available models, Rasch models, allowing conversation of raw data into interval scores.[8]

Trunk control during sitting and gait

The results of the assessment of trunk control during sitting, reported in ***Paper II***, showing decreasing scores on the trunk control test with increasing GMFCS level, are consistent with earlier findings.[5; 48] However, to my knowledge, the study reported in ***Paper IV*** is the first to have assessed the relationship between trunk control during sitting and during gait in children with CP. The relation between trunk control in standing and during gait has been assessed in children with spastic diplegia. In such studies, the correlation between dynamic trunk control tasks and spatiotemporal gait parameters (gait speed, step length, cadence) have been found to higher than the correlation between the static trunk control tasks and the spatiotemporal gait parameters.[173, 174] This finding is in agreement with the results in ***Paper IV***, where the dynamic subscales for both the TIS and the TCMS showed the highest correlations with the gait parameters. This may be explained by "task specificity," as gait is a task that requires more dynamic trunk control than static trunk control.

I am aware of only two very recent studies that assess trunk control during gait in children with CP and that applied the same method as used in the studies referred in ***Paper III*** and **IV** namely involving a trunk-worn accelerometer. However, the two studies, which were conducted by members of the same research group,[24, 110] included only children with unilateral CP, with a mean age of 5 years. Thus, the study presented in ***Paper III*** was the first to use a trunk-worn accelerometer to investigate gait characteristics in older children with CP, including children with spastic bilateral CP. The higher trunk accelerations found in all three directions in the CP group are consistent with the findings in two studies of younger children with unilateral CP assessed with the using a trunk-worn accelerometer,[24, 110] and with the findings of Hsue et al.,[25] who assessed trunk control during gait by optoelectronic motion analysis. In the study reported in ***Paper III*** children with CP presented less regularity between strides (in the AP direction) than TD children. This finding is in agreement with earlier studies reporting increased variability in gait characteristics in children with CP.[25, 106, 175, 176]

6.4 Interpretation of the results of trunk control during gait

The studies included in this thesis did not focus on the cause of the observed differences in trunk control during gait in children with CP and TD children, as information on underlying mechanisms—which may influence trunk control, such as footfall, range of motion, abnormal

muscle tone, loss of selective motor control, and muscle weakness—was not collected. Thus, the interpretations were based on knowledge of the patient group and theoretical perspectives.

The increased accelerations observed in all three directions in children with CP compared with TD children in the study reported in ***Paper III*** and other studies of children with CP,[24, 25, 110] as well as in studies of other groups, such as stroke patients,[26, 177] fatigued elderly people (ML direction),[27, 178] and children with dyslexia,[179] have been interpreted as a sign of impaired control. The increased trunk accelerations observed in the study reported in ***Paper III*** may have been a consequence of a primary trunk deficit or a compensatory mechanism for impairments in the lower limbs, or both, consistent with the notion that the "*trunk may be oriented secondary to foot position or vice versa."*[9][23] A possible explanation for the higher accelerations may be the inability to attenuate the oscillations of the COM arising from abnormal muscle tone, muscle weakness, and joints stiffness in the lower limbs, and in the trunk in the case of the children with CP.[25, 66] Increased accelerations of the COM may also be a result of excessive reactive adjustments, which may be explained by inadequate anticipatory trunk control.[148] Earlier studies have shown that children with CP have impaired anticipatory trunk control when standing.[180, 181] Moreover, studies have shown that in healthy adults the proximal (hip/trunk) muscles are the primary contributors to anticipatory trunk control during gait.[64, 182] Impaired trunk control has been reported in children classed at all GMFCS levels, and may thus explain impaired anticipatory control.[7, 48, 138, 176]

The steeper increase in accelerations with higher gait speed in children with CP compared with TD children may also be seen as compensatory mechanisms. Hence, children with CP have to "pay a price" in terms of a higher "cost" in order to be able to walk faster.[183] The compensatory mechanisms may be due to insufficient muscle strength, such as in the plantar flexors, and that children with CP therefore may use their hips, pelvis, and trunk (proximal joints) to produce a momentum to propel their extremities forward, as suggested by some authors.[25, 101, 184, 185] Gage [185] describes it like walking in deep mud or snow: "*one is forced to derive the power for mobility from 'pull up' from hips and knees, as opposed to 'push-off' from ankle and foot.*"[10] It is likely that such proximal compensations may have contributed to the increased trunk accelerations in the studied children with CP, and hence challenged their trunk control.

[9] Moe-Nilssen R. et al., 2005. Page 165.
[10] Gage JR. et al., 2004. Page 213.

Lateral trunk variables, especially ML regularity between strides, have been found to be important for trunk control during gait.[28] However, in the study reported in ***Paper III*** differences in AP stride regularity were found between children with CP and TD children: the children with CP had less regular (more variable) strides than the TD children. This finding is in agreement with studies of fit and frail older adults,[23] and in older adults in a fatigue group compared with a non-fatigue group.[27] However, in contrast to the study in ***Paper III***, the two studies of older adults found decreased ML variability between strides, and the authors suggested this was a strategy to control the multiple degrees of freedom. However, the interpretation of variability is unclear, as variability may indicate two opposites: adaptability or impairment.[28, 186] Traditionally, variability in motor functions has been regarded as an indicator of impaired control.[28] However, with regard to dynamic system theory, variability has also been interpreted as a positive factor for control. Latash et al.[182] proposed that motor variability should be classified as either 'good' or 'bad', where 'good' variability is believed to assist in achieving a successful outcome, whereas 'bad' variability is described to cause problems in performance.

However, the increased variability observed in children with CP in ***Paper III*** may also be explained according to a theory of motor development, the neural group selection theory (briefly described in section 2.2.2). This may indicate that children with CP are still in the phase of development called primary variability, in which the motor system explores motor possibilities, and that they not have been able to select the motor behavior that best fits a given situation, which occur in the phase known as secondary variability.

6.5 Generalizability

In the preceding subsections I have discussed the internal validity of the studies included in this thesis. However, even if the results were internally valid, they may not be externally valid. External validity refers to whether the results may be applicable to other similar populations.[160] The included subjects with CP comprised a heterogeneous population, including children classed as GMFCS Levels I–IV (***Paper II***) and GMFCS Levels I–III (***Papers III and IV***) in the age range 5–18 years. It is not likely that this population differed greatly from populations in other high-income countries, in which diagnostic criteria, treatment traditions, and follow-up programs are similar. Nonetheless, the findings need to be replicated in other populations in order to document their external validity.

7. General conclusions

Identifying determinants to target “gait treatments” is difficult, since children with CP represent a heterogeneous group with a large variety of gait patterns and underlying impairments. The aim was to identify assessment tools developed to assess trunk control in children with CP, which is one possible determinant of “gait outcomes.” Through a systematic literature review, 22 tools developed to assess trunk control in a clinical setting were identified, and a moderate or limited level of evidence was found for most of the measurement properties. Notably, responsiveness was scarcely documented. Moreover, the measurement properties of the TIS were examined and found to be a reliable and valid assessment tool of trunk control in children and adolescents with CP in the age group 5–19 years.

To provide more knowledge of trunk control during gait, children with CP were compared with TD children, and significant differences were found between the two groups. The problems relating to trunk control seemed to increase with increasing gross motor impairment and with increasing speed. Moreover, a moderate relationship was found between trunk control during sitting assessed with the TIS and the TCMS, and trunk control during gait assessed with a trunk-worn accelerometer. The results indicate that the sitting assessment may provide some information on the ability to control the trunk during gait.

8. Clinical implications

The systematic review revealed that the responsiveness of the assessment tools had scarcely been documented, thus indicating that results of existing studies evaluating the effects of treatment of trunk control in subjects with CP should be interpreted with caution.

The moderate relationship between trunk control during sitting assessed with two trunk control tests and trunk control during gait assessed with a trunk-worn accelerometer indicates that the sitting assessment may provide some information on the ability to control the trunk during gait. The results thus suggest that two subscales of these tools, which are less time-consuming, may be applied in clinical assessments of trunk control. This finding may be valuable in clinical practice, when planning treatment of the lower limbs, as impaired control of the trunk may influence movement in the lower limbs,[23] thus making it more difficult to predict the results of the intervention. In children with poor trunk control during sitting, augmented examination of the relationship between the trunk and the lower limbs may be required, whereas if trunk control during sitting is good, more advanced examinations may be omitted.

9. Future research

There is a need for further studies in order to identify determinants that may act as barriers or facilitators to gait outcomes. Advances in statistical methods, such as cluster analysis and principal component analysis[187] and linear discriminant analysis,[188] have been applied to predict outcomes (crouch gait and outcomes of rectus femoris transfer surgery, respectively) Moreover, development and testing of multivariable models have been suggested in the management of complex conditions such as CP.[189, 190] However, appropriate assessment tools are a prerequisite for the quality of the implemented data in these models as well as for all other research. The measurement properties of the tools need to be assessed by high quality studies, and in particular there is a lack of studies of responsiveness.[132] This is a limitation for intervention research, which is dependent on the ability to evaluate change.[8]

To obtain a higher level of evidence for the measurement properties of the tools, higher quality method studies may be warranted. The COSMIN methodology may be applied when planning and evaluating method studies, to ensure proper study design and statistical methods. However, methods to combine evidence of measurement properties from different studies are have not yet been well established. Thus, more work needs to be done on the methodology for data synthesis of measurement properties, such as statistical pooling and best evidence synthesis.[8] Moreover, in research it may be desirable also to quantify trunk control using new technology that has the advantage of measuring changes in trunk control that are too small to be observed. A separate systematic review of available "laboratory methods" in children with CP may be desirable.

Moreover, further studies are needed to explore the relation between progression and trunk control during gait, and to unravel how (primary) impairments in trunk control and compensatory mechanisms due to impairments of the lower limbs affect balance during gait in children with CP. The results suggest that assessments of trunk control during sitting through a readily accomplished clinical test will provide insights into trunk control during gait. However, future studies are needed to explore how such information could be applied in the planning of interventions to improve gait performance in children and adolescents with CP.

Computerized 3-D gait analyses have increased our understanding of gait in general and of pathologic gait in children with CP in particular. Recognizable gait patterns can be classified and used when making decisions and evaluating outcomes. However, there are still many

sources of variability related to this method, including the patients themselves, the gait laboratories, testing, processing, and interpretation of data, and surgical recommendations.[191] A review of gait classifications has indicated that currently laboratories mainly utilize variables related to the lower limbs.[2] However, in a recent study a high level of reliability was found for a model assessing trunk kinematics in children with CP.[192] Future research would probably benefit from agreement on a model that includes the total body as a basis for interpretation, as well as the newly developed gait index, the TPS,[41] which reflects the overall severity of trunk movement pathology in addition to the gait indexes addressing deviations in the lower limbs. However, one limitation of 3-D gait analysis is its restriction to a laboratory environment. Trunk-worn accelerometers may provide an alternative approach to assessing the gait characteristics as spatiotemporal parameters and trunk control.[109] This method is comparatively less time-consuming and less expensive, and is not restricted to assessments conducted in a laboratory environment. However, new methods are emerging in motion analyses that will facilitate our understanding of the complexity of gait pathology, such as "modeling" and "simulation" of the musculoskeletal system.[193]

There is an ongoing debate in the field[194] regarding the next steps, and prioritizing the development of in-depth, subgroups-specific, valid, and patient-orientated studies seems to be the most desirable approach.

10. References

1. McGinley JL, Dobson F, Ganeshalingam R, et al. Single-event multilevel surgery for children with cerebral palsy: a systematic review. Dev Med Child Neurol. 2012;54(2):117-28.
2. Dobson F, Morris ME, Baker R, Graham HK. Gait classification in children with cerebral palsy: a systematic review. Gait Posture. 2007;25(1):140-52.
3. Majnemer A. Selection and use og outcome measures. In: Majnemer A, editor. Measures for children with developmental disabilities An ICF-CY approach. London: Mac Keith Press; 2012. p. 3-9.
4. Mayston MJ. People with cerebral palsy: effects of and perspectives for therapy. Neural Plast. 2001;8(1-2):51-69.
5. de Graaf-Peters VB, Blauw-Hospers CH, Dirks T, et al. Development of postural control in typically developing children and children with cerebral palsy: Possibilities for intervention? Neurosci Biobehav Rev. 2007;31(8):1191-200.
6. Verheyden G, Nieuwboer A, Mertin J, et al. The Trunk Impairment Scale: a new tool to measure motor impairment of the trunk after stroke. Clin Rehabil. 2004;18(3):326-34.
7. Saether R, Jorgensen L. Intra- and inter-observer reliability of the Trunk Impairment Scale for children with cerebral palsy. Res Dev Disabil. 2011;32(2):727-39.
8. de Vet H, Terwee C, Mokkink LB, Knol DL. Measurement in Medicine. 1 ed. New York: Camebridge University Press; 2011.
9. Andersen GL, Irgens LM, Haagaas I, et al. Cerebral palsy in Norway: prevalence, subtypes and severity. Eur J Paediatr Neurol. 2008 Jan;12(1):4-13.
10. Europe. SoCPi. Surveillance of Cerbral Palsy in Europe (SCPE): a collaboration of cerebral palsy surveys and registers. Dev Med Child Neurol. 2000;42:816-24.
11. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl. 2007;109:8-14.
12. Bax M, Goldstein M, Rosenbaum P, Leviton A, Paneth N. Proposed definition and classification of cerebral palsy. Dev. Med. Child Neurol. 2005;47:571-76.
13. Krageloh-Mann I, Cans C. Cerebral palsy update. Brain Dev. 2009;31(7):537-44.
14. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. Dev Med Child Neurol. 2008;50(10):744-50.
15. Rosenbaum PL, Palisano RJ, Bartlett DJ, Galuppi BE, Russell DJ. Development of the Gross Motor Function Classification System for cerebral palsy. Dev Med Child Neurol. 2008;50(4):249-53.
16. Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39:214-23.
17. Shumway-Cook A, Wollacott MH. Motor Control. In: Lupash E, editor. Motor control Translating research into clinical practice. Baltimore: Wolters Kluwer/Lippincott Williams & Wilkins; 2012. p. 161-93.
18. Streiner DL, Norman GR. Health Measurement Scales. 4 ed. New York: Oxford University Press; 2008.
19. Bax M, Goldstein M, Rosenbaum P, et al. Proposed definition and classification of cerebral palsy, April 2005. Dev Med Child Neurol. 2005 Aug;47(8):571-76.
20. Horak F, Macpherson J. Postural orientation and equilibrium. Handbook of physiology. New York: Oxford University Press; 1996. p. 256-92.
21. Winter DA. Human balance and posture control during standing and walking. Gait Posture. 1995;3(4):193-214.
22. Verheyden G, Vereeck L, Truijen S, et al. Trunk performance after stroke and the relationship with balance, gait and functional ability. Clin Rehabil. 2006 May;20(5):451-58.
23. Moe-Nilssen R, Helbostad JL. Interstride trunk acceleration variability but not step width variability can differentiate between fit and frail older adults. Gait Posture. 2005;21(2):164-70.
24. Iosa M, Marro T, Paolucci S, Morelli D. Stability and harmony of gait in children with cerebral palsy. Res Dev Disabil. 2012;33(1):129-35.

25. Hsue BJ, Miller F, Su FC. The dynamic balance of the children with cerebral palsy and typical developing during gait Part II: Instantaneous velocity and acceleration of COM and COP and their relationship. Gait Posture. 2009;29(3):471-76.
26. Iosa M, Fusco A, Morone G, et al. Assessment of upper-body dynamic stability during walking in patients with subacute stroke. J Rehabil Res Dev. 2012;49(3):439-50.
27. Helbostad JL, Leirfall S, Moe-Nilssen R, Sletvold O. Physical fatigue affects gait characteristics in older persons. J Gerontol A Biol Sci Med Sci. 2007;62(9):1010-15.
28. Moe-Nilssen R, Aaslund MK, Hodt-Billington C, Helbostad JL. Gait variability measures may represent different constructs. Gait Posture. 2010 May;32(1):98-101.
29. Huisinga JM, Mancini M, St George RJ, Horak FB. Accelerometry reveals differences in gait variability between patients with multiple sclerosis and healthy controls. Ann Biomed Eng. 2013;41(8):1670-79.
30. England SA, Granata KP. The influence of gait speed on local dynamic stability of walking. Gait Posture. 2007;25(2):172-8.
31. Shumway-Cook A, Woollacott MH. Motor control: Issues and Theories. In: Lupash E, editor. Motor control: Translating research into clinical practice. Baltimore: Lippincott Williams & Wilkins; 2012. p. 3-20.
32. Forssberg H, Hirschfeld H. Postural adjustments in sitting humans following external perturbations: muscle activity and kinematics. Exp Brain Res. 1994;97(3):515-27.
33. Hadders-Algra M. The neuronal group selection theory: promising principles for understanding and treating developmental motor disorders. Dev Med Child Neurol. 2000 Oct;42(10):707-15.
34. Hadders-Algra M. The neural group selection theory: a framework to explain variation in normal motor development. Dev Med Child Neurol. 2000;42:566-72.
35. Hadders-Algra M. Development of postural control. In: Hadders-Algra M, Carlberg EB, editors. Postural control: A key issue in developmental disabilities. London: Mac Keith Press; 2008. p. 22-73.
36. Shumway-Cook A, Woollacott MH. Motor control: Issues and theories. In: Shumway-Cook A, Woollacott MH, editors. Motor Control Translating research into clinical practice. 3 ed. Baltimore: Lippincott Williams & Wilkins; 2007. p. 3-20.
37. Latash M, Hadders-Algra M. What is posture and how is it controlled ? In: Algra MH, Carlberg EB, editors. Postural Control: A key issue in developmental disorders. London: Mac Keith Press; 2008. p. 3-21.
38. Hadders-Algra M. Development of postural control. In: Carlberg EB, Hadders-Algra M, editors. Postural control: A key issue in developmental disorders. 1 ed. London: Mach Keith Press2008.
39. Campbell SK, Linden DWV, Palisano RJ. Physical therapy for children. 3 ed. St. Louis Missouri: Saunders Elsevier; 2007.
40. Sporns O, Edelman G. Solving Bernstein's problem: a proposal for the development of coordinated movement by selection. Child Dev. 1993;64:960-81.
41. Heyrman L, Feys H, Molenaers G, et al. Three-dimensional head and trunk movement characteristics during gait in children with spastic diplegia. Gait Posture. 2013;38(4):770-6.
42. Trost J. Physical assessment and observational gait analysis. In: Hart H, editor. The treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2004. p. 71-98.
43. Hedberg A, Forssberg H, Hadders-Algra M. Postural adjustments due to external perturbations during sitting in 1-month-old infants: evidence for the innate origin of direction specificity. Exp Brain Res. 2004 Jul;157(1):101-7.
44. Hadders-Algra M. Development of postural control during the first 18 months of life. Neural Plast. 2005;12(2-3):99-108; discussion 263-72.
45. Carlberg EB, Hadders-Algra M. Postural dysfunction in children with cerebral palsy: some implications for therapeutic guidance. Neural Plas. 2005;12(2-3):221-8.
46. Hadders-Algra M, Carlberg EB. Postural Control: A key issue in developmental disorders. Hadders-Algra M, Carlberg EB, editors. London: Mac Keith Press; 2008.

47. Brogren E, Forssberg H, Hadders-Algra M. Influence of two different sitting positions on postural adjustments in children with spastic diplegia. Dev Med Child Neurol. 2001 Aug;43(8):534-46.
48. Heyrman L, Desloovere K, Molenaers G, et al. Clinical characteristics of impaired trunk control in children with spastic cerebral palsy. Res Dev Disabil. 2013;34(1):327-34.
49. Brogren E, Hadders-Algra M, Forssberg H. Postural control in children with spastic diplegia: muscle activity during perturbations in sitting. Dev Med Child Neurol. 1996 May;38(5):379-88.
50. Brogren E, Hadders-Algra M, Forssberg H. Postural control in sitting children with cerebral palsy. Neurosci Biobehav Rev. 1998 Jul;22(4):591-6.
51. Bigongiari A, de Andrade e Souza F, Franciulli PM, et al. Anticipatory and compensatory postural adjustments in sitting in children with cerebral palsy. Human Movement Science. 2011;30(3):648-57.
52. van der Heide JC, Begeer C, Fock JM, et al. Postural control during reaching in preterm children with cerebral palsy. Dev Med Child Neurol. 2004 Apr;46(4):253-66.
53. Gurfinkel VS, Lipshits MI, Mori S, Popov KE. Stabilization of body position as the main task of postural regulation. Hum Physiol. 1981;7(3):155-65.
54. van der Heide JC, Fock JM, Otten B, Stremmelaar E, Hadders-Algra M. Kinematic characteristics of postural control during reaching in preterm children with cerebral palsy. Pediatr Res. 2005 Sep;58(3):586-93.
55. Shumway-Cook A, Woollacott MH. Mobility functions. In: Lupash E, editor. Motor control: Translating research into clinical practice. Baltimore: Lippincott Williams & Wilkins; 2012. p. 315-47.
56. Perry J, Burnfield J. Basic functions. Gait Analysis Normal and pathological fuction. Thorofare: Slack Incorporated; 2010. p. 19-50.
57. Massion J, Woollacott MH. Posture and Equilibrium. In: Bronstein A, Brandt T, Wollacott MH, Nutt JG, editors. Clinical disorders of balance, posture and gait. London: Arnold; 2004. p. 1-19.
58. Kuo AD, Donelan JM. Dynamic principles of gait and their clinical implications. Phys Ther. 2010;90(2):157-74.
59. Saunders JB, Inman VT, Eberhart HD. The major determinants in normal and pathological gait. J Bone Joint Surg Am. 1953:543-58.
60. Shumway-Cook A, Woollacott MH. Control of normal mobility. In: Lupash E, editor. Motor control: Translating research into clinical practice. Baltimore: Lippincott Williams & Wilkins; 2012. p. 315-48.
61. Peacock WJ. The neural control of movement. In: Gage JR, editor. The treatment of gaitproblems in cerebral palsy. London: Mac Keith Press; 2009. p. 3-20.
62. Liu WY, Zaino CA, McCoy SW. Anticipatory postural adjustments in children with cerebral palsy and children with typical development. Pediatr Phys Ther. 2007;19(3):188-95.
63. Frank JS, Earl M. Coordination of posture and movement. Phys Ther. 1990;70(12):855-63.
64. Tang PF, Woollacott MH, Chong RK. Control of reactive balance adjustments in perturbed human walking: roles of proximal and distal postural muscle activity. Exp Brain Res. 1998;119(2):141-52.
65. Patla AE, Adkin A, Ballard T. Online steering: coordination and control of body center of mass, head and body reorientation. Exp Brain Res. 1999;129(4):629-34.
66. Kavanagh JJ, Morrison S, Barrett RS. Coordination of head and trunk accelerations during walking. Eur J Appl Physiol. 2005;94(4):468-75.
67. Thorstensson A, Nilsson J, Carlson H, Zomlefer MR. Trunk movements in human locomotion. Acta Physiol Scand. 1984;121(1):9-22.
68. Shumway-Cook A, Woollacott MH. A life span perspective of mobility. In: Lupash E, editor. Motor control: Translating research into clinical practice. Baltimore: Lippincott Williams & Wilkins; 2012. p. 348-80.
69. Breniere Y, Bril B. Development of postural control of gravity forces in children during the first 5 years of walking. Exp Brain Res. 1998;121(3):255-62.
70. Assaiante C, Mallau S, Viel S, Jover M, Schmitz C. Development of postural control in healthy children: a functional approach. Neural Plast. 2005;12(2-3):109-18; 263-72.
71. Koop S. The natural history of ambulation in cerebral palsy. In: Gage JR, editor. The treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009. p. 167-78.

72. Assaiante C. Development of locomotor balance control in healthy children. Neurosci Biobehav Rev. 1998 Jul;22(4):527-32.
73. Beckung E, Carlsson G, Carlsdotter S, Uvebrant P. The natural history of gross motor development in children with cerebral palsy aged 1 to 15 years. Dev Med Child Neurol. 2007;49(10):751-6.
74. Wollacott MH, Crenna P. Postural control in standing and walking in children with cerebral palsy. In: Hadders-Algra M, Carlberg EB, editors. Postural control: A key issue in developmental disorders. London: Mac Keith Press; 2008. p. 97-130.
75. Wren TA, Rethlefsen S, Kay RM. Prevalence of specific gait abnormalities in children with cerebral palsy: influence of cerebral palsy subtype, age, and previous surgery. J Pediatr Orthop. 2005;25(1):79-83.
76. Baker R, McGinley JL, Schwartz MH, et al. The gait profile score and movement analysis profile. Gait Posture. 2009;30(3):265-9.
77. Schwartz MH, Rozumalski A. The Gait Deviation Index: a new comprehensive index of gait pathology. Gait Posture. 2008;28(3):351-7.
78. Yokochi K. Gait patterns in children with spastic diplegia and periventricular leukomalacia. Brain Dev. 2001;23(1):34-7.
79. Leonard CT, Hirschfeld H, Forssberg H. The development of independent walking in children with cerebral palsy. Dev Med Child Neurol. 1991;33(7):567-77.
80. Prosser LA, Ohlrich LB, Curatalo LA, Alter KE, Damiano DL. Feasibility and preliminary effectiveness of a novel mobility training intervention in infants and toddlers with cerebral palsy. Dev Neurorehabil. 2012;15(4):259-66.
81. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study. Dev Med Child Neurol. 2009;51(5):381-8.
82. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Balance in relation to walking deterioration in adults with spastic bilateral cerebral palsy. Phys Ther. 2012;92(2):279-88.
83. Opheim A, McGinley JL, Olsson E, Stanghelle JK, Jahnsen R. Walking deterioration and gait analysis in adults with spastic bilateral cerebral palsy. Gait Posture. 2013;37(2):165-71.
84. Narayanan UG. Management of children with ambulatory cerebral palsy: an evidence-based review. J Pediatr Orthop. 2012;32(2):172-81
85. Eek MN, Tranberg R, Zugner R, Alkema K, Beckung E. Muscle strength training to improve gait function in children with cerebral palsy. Dev Med Child Neurol. 2008;50(10):759-64.
86. Unger M, Jelsma J, Stark C. Effect of a trunk-targeted intervention using vibration on posture and gait in children with spastic type cerebral palsy: a randomized control trial. Dev Neurorehabil. 2013;16(2):79-88.
87. Ledebt A, Bril B. Acquisition of upper body stability during walking in toddlers. Dev Psychobiol. 2000;36(4):311-24.
88. Kibler WB, Press J, Sciascia A. The role of core stability in athletic function. Sports Med. 2006;36(3):189-98.
89. Wu YW, Day SM, Strauss DJ, Shavelle RM. Prognosis for ambulation in cerebral palsy: a population-based study. Pediatrics. 2004;114(5):1264-71.
90. Verheyden G, Nieuwboer A, De Wit L, et al. Trunk performance after stroke: an eye catching predictor of functional outcome. J Neurol Neurosurg Psychiatry. 2007 Jul;78(7):694-8.
91. Iosa M, Fusco A, Morone G, Paolucci S. Development and decline of upright gait stability. Front Aging Neurosci. 2014;6(14).
92. Romkes J, Peeters W, Oosterom AM, et al. Evaluating upper body movements during gait in healthy children and children with diplegic cerebral palsy. J Pediatr Orthop B. 2007;16(3):175-80.
93. Prosser LA, Lee SC, VanSant AF, Barbe MF, Lauer RT. Trunk and hip muscle activation patterns are different during walking in young children with and without cerebral palsy. Phys Ther. 2010;90(7):986-97.
94. Van de Walle P, Hallemans A, Truijen S, et al. Increased mechanical cost of walking in children with diplegia: The role of the passenger unit cannot be neglected. Res Dev Disabil. 2012;33(6):1996-2003.

95. Krautwurst BK, Wolf SI, Heitzmann DW, et al. The influence of hip abductor weakness on frontal plane motion of the trunk and pelvis in patients with cerebral palsy. Res Dev Disabil. 2013;34(4):1198-203.
96. Kwon JY, Chang HJ, Lee JY, et al. Effects of hippotherapy on gait parameters in children with bilateral spastic cerebral palsy. Arch Phys Med Rehabil. 2011 May;92(5):774-9.
97. Manikowska F, Jozwiak M, Idzior M, Chen PJ, Tarnowski D. The effect of a hippotherapy session on spatiotemporal parameters of gait in children with cerebral palsy - pilot study. Ortop Traumatol Rehabil. 2013;15(3):253-7.
98. Hsue BJ, Miller F, Su FC. The dynamic balance of the children with cerebral palsy and typical developing during gait. Part I: Spatial relationship between COM and COP trajectories. Gait Posture. 2009;29(3):465-70.
99. Massaad F, Dierick F, van den Hecke A, Detrembleur C. Influence of gait pattern on the body's centre of mass displacement in children with cerebral palsy. Dev Med Child Neurol. 2004;46(10):674-80.
100. Cherng R, Chou L, Su F, Shaughnessy WJ, Kaufman KR. Using Motion of the Whole-Body Center of Mass to Assess the Balance during Gait of Children with Spastic Cerebral Palsy. Journal of Medical and Biological Engineering. 2007;27(3):150-5.
101. Feng J, Pierce R, Do KP, Aiona M. Motion of the center of mass in children with spastic hemiplegia: Balance, energy transfer, and work performed by the affected leg vs. the unaffected leg. Gait Posture. 2013;25(13):Article in press.
102. Wallard L, Dietrich G, Kerlirzin Y, Bredin J. Balance control in gait children with cerebral palsy. Gait Posture. 2014;26(14):00073-3.
103. Bruijn SM, Meyns P, Jonkers I, Kaat D, Duysens J. Control of angular momentum during walking in children with cerebral palsy. Res Dev Disabil. 2011;4:2860-6
104. Meyns P, Desloovere K, Van Gestel L, et al. Altered arm posture in children with cerebral palsy is related to instability during walking. Eur J Paediatr Neurol. 2012;16(5):528-35.
105. Wallard L, Bril B, Dietrich G, Kerlirzin Y, Bredin J. The role of head stabilization in locomotion in children with cerebral palsy. Ann Phys Rehabil Med. 2012;2(12):1286-9.
106. Kurz MJ, Arpin DJ, Corr B. Differences in the dynamic gait stability of children with cerebral palsy and typically developing children. Gait Posture. 2012;36(3):600-4.
107. Bruijn SM, Millard M, van Gestel L, et al. Gait stability in children with Cerebral Palsy. Res Dev Disabil. 2013;34(5):1689-99.
108. Li H. Balancing charachteristics of children with spastic cerebral palsy during gait measurement using plantar pressure gait analysis system Journal of Clinical Rehabilitative Tissue Engineering Research. 2009;13(17):3387-91.
109. Kavanagh JJ, Menz HB. Accelerometry: a technique for quantifying movement patterns during walking. Gait Posture. 2008;28(1):1-15.
110. Iosa M, Morelli D, Marro T, Paolucci S, Fusco A. Ability and stability of running and walking in children with cerebral palsy. Neuropediatrics. 2013;44(3):147-54.
111. Jahnsen R, Elkjær S, Myklebust G. Årsrapport: Cerebral parese oppfølgingsprogram (CPOP). Oslo: Oslo Universitetssykehus. 2012.
112. Elkamil AI, Andersen GL, Skranes J, Lamvik T, Vik T. Botulinum neurotoxin treatment in children with cerebral palsy: a population-based study in Norway. Eur J Paediatr Neurol. 2012;16(5):522-7.
113. Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. Dev Med Child Neurol. 2013;55(10):885-910.
114. Rutz E, Tirosh O, Thomason P, Barg A, Graham HK. Stability of the Gross Motor Function Classification System after single-event multilevel surgery in children with cerebral palsy. Dev Med Child Neurol. 2012;54(12):1109-13.
115. Gage JR, Schwartz M. Pathological gait and lever-arm dysfunction. In: Gage JR, editor. The treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2004. p. 180-204.
116. Bleck EE. Orthopaedic management in cerebral palsy. London: Mac Keith Press; 1987.
117. Majnemer A, Limperopoulos C. Importance of outcome determination in pediatric rehabilitation. Dev Med Child Neurol. 2002;44(11):773-7.

118. Majnemer A. Benefits of using outcome measures in pediatric rehabilitation: Phys Occup Ther Pediatr. 2010 Aug;30(3):165-7.
119. Majnemer A, Mazer B. New directions in the outcome evaluation of children with cerebral palsy. Semin Pediatr Neurol. 2004;11(1):11-7.
120. Portney LG, Watkins MP. Foundations of Clinical Research. 3 ed. New Jersey: Pearson Education; 2009.
121. Sackett DL, Rosenberg WM, Gray JA, Haynes RB, Richardson WS. Evidence based medicine: what it is and what it isn't. 1996. Clin Orthop Relat Res. 2007;455:3-5.
122. Straus S. Evidence-based medicine : how to practice and teach it. Edinburgh: Churchill Livingstone/Elsevier; 2011.
123. Mokkink LB, Terwee CB, Patrick DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. J Clin Epidemiol. 2010;63(7):737-45.
124. Thornquist E. Vitenskapsfilosofi og vitenskapsteori. 2 ed. Bergen: Fagbokforlaget; 2006.
125. Tyson S, Greenhalgh J, Long AF, Flynn R. The use of measurement tools in clinical practice: an observational study of neurorehabilitation. Clin Rehabil. 2010;24(1):74-81.
126. Law M, MacDermid J. Evidence- Based Rehabilitation. 2 ed. Thorofare: SLACK Incorporated; 2008.
127. Mokkink LB. COSMIN:Development and evaluation of a checklist to assess the methodological quality of studies on measurement properties [Thesis]. Amsterdam: VU University Medical Center; 2010.
128. Terwee CB, Mokkink LB, Knol DL, et al. Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. Qual Life Res. 2012;21(4):651-7.
129. Terwee CB, Bot SD, de Boer MR, et al. Quality criteria were proposed for measurement properties of health status questionnaires. J Clin Epidemiol. 2007;60(1):34-42.
130. World Health Organization. International Classification of Functioning, Disability and Health. Geneva: World Health Organization; 2001.
131. World Health Organization. International Classification of Functioning, Disability and Health-Children and Youth version: ICF-CY. Geneva: World Health Organization; 2007.
132. Saether R, Helbostad JL, Riphagen, II, Vik T. Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review. Dev Med Child Neurol. 2013;11:988-99
133. Hamacher D, Singh NB, Van Dieen JH, Heller MO, Taylor WR. Kinematic measures for assessing gait stability in elderly individuals: a systematic review. J R Soc Interface. 2011;8(65):1682-98.
134. Bruijn SM, Meijer OG, Beek PJ, van Dieen JH. Assessing the stability of human locomotion: a review of current measures. J R Soc Interface. 2013;10(83):6.
135. Kavanagh J, Barrett R, Morrison S. The role of the neck and trunk in facilitating head stability during walking. Exp Brain Res. 2006;172(4):454-63.
136. Mazza C, Zok M, Cappozzo A. Head stabilization in children of both genders during level walking. Gait Posture. 2010;31(4):429-32.
137. Surveillance of Cerebral Palsy in Europe. A collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). Dev Med Child Neurol. 2000 Dec;42(12):816-24.
138. Saether R, Helbostad JL, Adde L, Jorgensen L, Vik T. Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy. Res Dev Disabil. 2013;34(7):2075-84.
139. Saether R, Helbostad JL, Adde L, et al. Gait characteristics in children and adolescents with cerebral palsy assessed with a trunk-worn accelerometer. Res Dev Disabil. 2014;25(14):00079-1.
140. Saether R, Helbostad JL, Adde L, et al. The relationship between trunk control in sitting and during gait in children and adolescents with cerebral palsy. Dev Med Child Neurol. 2014;april:submitted.
141. Verheyden G, Nuyens G, Nieuwboer A, et al. Reliability and validity of trunk assessment for people with multiple sclerosis. Phys Ther. 2006 Jan;86(1):66-76.

142. Heyrman L, Molenaers G, Desloovere K, et al. A clinical tool to measure trunk control in children with cerebral palsy: the Trunk Control Measurement Scale. Res Dev Disabil. 2011;32(6):2624-35.
143. Aaslund MK, Helbostad JL, Moe-Nilssen R. Familiarisation to body weight supported treadmill training for patients post-stroke. Gait Posture. 2011;34(4):467-72.
144. Moe-Nilssen R. Test-retest reliability of trunk accelerometry during standing and walking. Arch Phys Med Rehabil. 1998;79(11):1377-85.
145. Henriksen M, Lund H, Moe-Nilssen R, Bliddal H, Danneskiod-Samsoe B. Test-retest reliability of trunk accelerometric gait analysis. Gait Posture. 2004 Jun;19(3):288-97.
146. Kavanagh JJ, Morrison S, James DA, Barrett R. Reliability of segmental accelerations measured using a new wireless gait analysis system. J Biomech. 2006;39(15):2863-72.
147. Moe-Nilssen R. A new method for evaluating motor control in gait under real-life environmental conditions. Part 1: The instrument. Clin Biomech. 1998;13(4-5):320-7.
148. Moe-Nilssen R. A new method for evaluating motor control in gait under real-life environmental conditions. Part 2: Gait analysis. Clin Biomech. 1998;13(4-5):328-35.
149. Moe-Nilssen R, Helbostad JL. Estimation of gait cycle characteristics by trunk accelerometry. J Biomech. 2004 Jan;37(1):121-6.
150. Hodt-Billington C, Helbostad JL, Vervaat W, Rognsvag T, Moe-Nilssen R. Changes in gait symmetry, gait velocity and self-reported function following total hip replacement. J Rehabil Med. 2011;43(9):787-93.
151. Mokkink LB, Terwee CB, Knol DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: a clarification of its content. BMC Medical Research Methodology. 2010;10:22.
152. Mokkink LB, Terwee CB, Gibbons E, et al. Inter-rater agreement and reliability of the COSMIN (COnsensus-based Standards for the selection of health status Measurement Instruments) checklist. BMC Med Res Methodol. 2010;10:82.
153. van Tulder M, Furlan A, Bombardier C, Bouter L. Updated method guidelines for systematic reviews in the cochrane collaboration back review group. Spine. 1976;28(12):1290-9.
154. Benfer KA, Weir KA, Boyd RN. Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: a systematic review. Dev Med Child Neurol. 2012;54(9):784-95.
155. Schellingerhout JM, Verhagen AP, Heymans MW, et al. Measurement properties of disease-specific questionnaires in patients with neck pain: a systematic review. Qual Life Res. 2012;21(4):659-70.
156. Uijen AA, Heinst CW, Schellevis FG, et al. Measurement properties of questionnaires measuring continuity of care: a systematic review. PLoS One. 2012;7(7):31.
157. Bland JM, Altman DG. Statistical methods for assessing agreement between two methods of clinical measurement. Lancet. 1986(1):307-10.
158. Schwartz MH, Rozumalski A, Trost JP. The effect of walking speed on the gait of typically developing children. J Biomech. 2008;41(8):1639-50.
159. Bonett DG. Sample size requirements for estimating intraclass correlations with desired precision. Stat Med. 2002;21(9):1331-5.
160. Hennekens CH, Buring JE. In: Mayrent SL, editor. Epidemiology in medicine. Boston: Little, Brown; 1987.
161. Robb JE, Hagglund G. Hip surveillance and management of the displaced hip in cerebral palsy: J Child Orthop. 2013 Nov;7(5):407-413. Epub 2013 Aug 18.
162. Yelnik A, Bonan I. Clinical tools for assessing balance disorders. Neurophysiol Clin. 2008;38(6):439-45.
163. Massaad F, van den Hecke A, Renders A, Detrembleur C. Influence of equinus treatments on the vertical displacement of the body's centre of mass in children with cerebral palsy. Dev Med Child Neurol. 2006;48(10):813-8.
164. Cieza A, Geyh S, Chatterji S, et al. ICF linking rules: an update based on lessons learned. J Rehabil Med. 2005;37(4):212-8.
165. Kottner J, Audige L, Brorson S, et al. Guidelines for Reporting Reliability and Agreement Studies (GRRAS) were proposed. J Clin Epidemiol. 2011;64(1):96-106.

166. Auvinet B, Berrut G, Touzard C, et al. Reference data for normal subjects obtained with an accelerometric device. Gait Posture. 2002 Oct;16(2):124-34.
167. Westcott SL, Lowes LP, Richardson PK. Evaluation of postural stability in children: current theories and assessment tools. Phys Ther. 1997;77(6):629-45.
168. Harris SR, Roxborough L. Efficacy and effectiveness of physical therapy in enhancing postural control in children with cerebral palsy. Neural Plast. 2005;12(2-3):229-43.
169. 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 Winter;20(4):303-17.
170. Field D, Livingstone R. Clinical tools that measure sitting posture, seated postural control or functional abilities in children with motor impairments: a systematic review. Clin Rehabil. 2013;27(11):994-1004.
171. Banas BB, Gorgon EJ. Clinimetric Properties of Sitting Balance Measures for Children with Cerebral Palsy: A Systematic Review. Phys Occup Ther Pediatr. 2014;3:3.
172. Pavao SL, dos Santos AN, Woollacott MH, Rocha NA. Assessment of postural control in children with cerebral palsy: a review. Res Dev Disabil. 2013;34(5):1367-75.
173. Liao HF, Jeng SF, Lai JS, Cheng CK, Hu MH. The relation between standing balance and walking function in children with spastic diplegic cerebral palsy. Dev Med Child Neurol. 1997;39(2):106-12.
174. Moore JG. The relationship between dynamic balance and walking in children. Coral Gables, Florida: University of Miami; 2006.
175. Katz-Leurer M, Rotem H, Keren O, Meyer S. Balance abilities and gait characteristics in post-traumatic brain injury, cerebral palsy and typically developed children. Dev Neurorehabil. 2009 Apr;12(2):100-5.
176. Prosser LA, Lauer RT, VanSant AF, Barbe MF, Lee SC. Variability and symmetry of gait in early walkers with and without bilateral cerebral palsy. Gait Posture. 2010;31(4):522-6.
177. Mizuike C, Ohgi S, Morita S. Analysis of stroke patient walking dynamics using a tri-axial accelerometer. Gait Posture. 2009;30(1):60-4.
178. Kavanagh JJ, Morrison S, Barrett RS. Lumbar and cervical erector spinae fatigue elicit compensatory postural responses to assist in maintaining head stability during walking. J Appl Physiol. 1985;101(4):1118-26.
179. Moe-Nilssen R, Helbostad JL, Talcott JB, Toennessen FE. Balance and gait in children with dyslexia. Exp Brain Res. 2003;150(2):237-44.
180. Girolami GL, Shiratori T, Aruin AS. Anticipatory postural adjustments in children with hemiplegia and diplegia. J Electromyogr Kinesiol. 2011;21(6):988-97.
181. Tomita H, Fukaya Y, Ueda T, et al. Deficits in task-specific modulation of anticipatory postural adjustments in individuals with spastic diplegic cerebral palsy. J Neurophysiol. 2011;105(5):2157-68.
182. Latash M, Hadders-Algra M. What is posture and how is it controlled. In: Hadders-Algra M, Carlberg EB, editors. Postural Control: A key issue in developmental disorders. London: Mac Keith Press; 2008. p. 3-21.
183. Mulder T, Zijlstra W, Geurts A. Assessment of motor recovery and decline. Gait Posture. 2002;16(2):198-210.
184. Abel MF, Damiano DL. Strategies for increasing walking speed in diplegic cerebral palsy. J Pediatr Orthop. 1996;16(6):753-8.
185. Gage JR. Spesific problems of the hips, knees and ankles. In: Gage JR, editor. The treatment of gaitproblems in cerebral palsy. London: Mac Keith Press; 2004. p. 205-16.
186. Harbourne RT, Stergiou N. Movement variability and the use of nonlinear tools: principles to guide physical therapist practice. Phys Ther. 2009;89(3):267-82.
187. Rozumalski A, Schwartz MH. Crouch gait patterns defined using k-means cluster analysis are related to underlying clinical pathology. Gait Posture. 2009;30(2):155-60.
188. Reinbolt JA, Fox MD, Schwartz MH, Delp SL. Predicting outcomes of rectus femoris transfer surgery. Gait Posture. 2009;30(1):100-5.
189. Bartlett DJ, Palisano RJ. A multivariate model of determinants of motor change for children with cerebral palsy. Phys Ther. 2000;80(6):598-614.

190. Hicks JL, Delp SL, Schwartz MH. Can biomechanical variables predict improvement in crouch gait? Gait Posture. 2011;34(2):197-201.
191. Narayanan UG. The role of gaitanalysis in the orthopaedic management of ambulatory cerebral pasly. Curr Opin Pediatr. 2007;19(1):38-43.
192. Heyrman L, Feys H, Molenaers G, et al. Reliability of head and trunk kinematics during gait in children with spastic diplegia. Gait Posture. 2012;37(3):429-9.
193. Hicks JL, Schwartz M, Scott LD. Modeling and simulation of normal and pathological gait. In: Gage JR, Schwartz M, Koop S, Novacheck TF, editors. The treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009. p. 285-305.
194. Novak I, McIntyre S, Morgan C, et al. Novak et al. reply: Dev Med Child Neurol. 2014 Apr;56(4):403-6.

Appendix A

COSMIN definitions of domains, measurement properties, and aspects of measurement properties[123]

Term			Definition
Domain	Measurement property	Aspect of a measurement property	
Reliability			The degree to which the measurement is free from measurement error
Reliability (extended definition)			The extent to which scores for patients who have not changed are the same for repeated measurement under several conditions: e.g. using different sets of items from the same health related-patient reported outcomes (HR-PRO) (internal consistency); over time (test-retest); by different persons on the same occasion (inter-rater); or by the same persons (i.e. raters or responders) on different occasions (intra-rater)
	Internal consistency		The degree of the interrelatedness among the items
	Reliability		The proportion of the total variance in the measurements which is due to 'true'† differences between patients
	Measurement error		The systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured
Validity			The degree to which an HR-PRO instrument measures the construct(s) it purports to measure
	Content validity		The degree to which the content of an HR-PRO instrument is an adequate reflection of the construct to be measured
		Face validity	The degree to which (the items of) an HR-PRO instrument indeed looks as though they are an adequate reflection of the construct to be measured
	Construct validity		The degree to which the scores of an HR-PRO instrument are consistent with hypotheses *(for instance with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups)* based on the assumption that the HR-PRO instrument validly measures the construct to be measured
		Structural validity	The degree to which the scores of an HR-PRO instrument are an adequate reflection of the dimensionality of the construct to be measured
		Hypotheses testing	Idem construct validity
		Cross-cultural validity	The degree to which the performance of the items on a translated or culturally adapted HR-PRO instrument are an adequate reflection of the performance of the items of the original version of the HR-PRO instrument
	Criterion validity		The degree to which the scores of an HR-PRO instrument are an adequate reflection of a 'gold standard'
Responsiveness			The ability of an HR-PRO instrument to detect change over time in the construct to be measured
	Responsiveness		Idem responsiveness
Interpretability*			Interpretability is the degree to which one can assign qualitative meaning - that is, clinical or commonly understood connotations – to an instrument's quantitative scores or change in scores.

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Appendix B

Quality criteria for measurement properties of health status questionnaire[129]

	Property	Definition		Quality criteria
1	Content validity	The extent to which a domain of interest is comprehensively sampled by the items in the questionnaire	+	A clear description is provided of the measurement aim, the target population, the concept that are being measured, and the item selection AND target population and (investigators OR experts) were involved in the item selection
			?	A clear description of above mentioned aspects is lacking OR only target population involved OR doubtful design or method
			-	No target population involvement
			0	No information found on target population involvement
2	Internal consistency	The extent to which items in a (sub)scale are intercorrelated , thus measuring the same construct	+	Factor analysis performed adequate sample size (7*#items and≥ 100) AND Cronbach's alpha (s) calculated per dimension AND Cronbach's alpha (s) between 0.70 and 0.95
			?	No factor analysis OR doubtful design or method
			-	Cronbach's alpha(s) <0.70 or >0.95, despite adequate design and method
			0	No information found on internal consistency
3	Criterion validity	The extent to which scores on a particular questionnaire relate to a gold standard	+	Convincing arguments that gold standard is "gold" AND correlation with gold standard ≥ 0.70
			?	No convincing arguments that gold standard is "gold" OR doubtful design or method
			-	Correlation with gold standard < 0.70, despite adequate design and method
			0	No information found on criterion validity
4	Construct validity	The extent to which scores on a particular questionnaire relate to other measures in a manner that is consistent with theoretically derived hypothesis concerning the concepts that are being measured	+	Specific hypothesis were formulated AND at least 75% of the results are in accordance with these hypothesis
			?	Doubtful design or method (e.g., no hypotheses)
			-	Less than 75% of hypotheses were confirmed, despite adequate design and methods
			0	No information found on construct validity
5	Reproducibility			
5.1	Agreement	The extent to which scores on repeated measures are close to each other (absolute measurement error)	+	+ MIC < SDC or MIC outside LOA OR convincing arguments that agreement is acceptable
			?	Doubtful design or method OR (MIC not

				defined AND no convincing arguments that agreement is acceptable)
			-	MIC ≥ SDC OR MIC equals or inside LOA, despite adequate design and methods
			0	No information found on agreement
5.2	Reliability	The extent to which patients can be distinguished from each other, despite measurement errors (relative measurement error)	+	ICC or weighted Kappa ≥ 0.70
			?	Doubtful design or method (e.g., time interval not mentioned)
			-	ICC or weighted Kappa < 0.70, despite adequate design and methods
			0	No information found on reliability
6	Responsiveness	The ability of a questionnaire to detect clinically important changes over time	+	SDC or SDC < MIC OR MIC outside LOA OR RR > 1.96 OR AUC ≥ 0.70
			?	Doubtful design or method
			-	SDC or SDC ≥MIC OR MIC equals or inside LOA OR RR ≤ 1.96 OR AUC <.70. despite adequate design and methods
			0	No information found on responsiveness
7	Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible score	+	≤15% of the respondents achieved the highest or lowest possible scores
			?	Doubtful design or method
				>15% of the respondents achieved the highest or lowest possible scores, despite adequate design and methods
			0	No information found on interpretation
8	Interpretability	The degree to which one can assign qualitative meaning to quantitative scores	+	Mean and SD scores presented of at least four relevant subgroups of patients and MIC defined
			?	Doubtful design or method OR less than four subgroups OR no MIC defined
			0	No information found on interpretability

MIC = minimal important change; SDC = smallest detectable change; LOA = limits of agreement; ICC = Intraclass correlation; SD, standard deviation.

[a] + = positive rating; ? = indeterminate rating; – = negative rating; 0 = no information available.

[b] Doubtful design or method = lacking of a clear description of the design or methods of the study, sample size smaller than 50 subjects (should be at least 50 in every (subgroup) analysis), or any important methodological weakness in the design or execution of the study.

Appendix C

Description of scoring criteria used to assess quality of studies, results of studies and level of evidence for measurement properties per balance tool

	Scoring	Level	Description
Cosmin[128]		excellent	
		good	
		fair	
		poor	
Terwee criteria[129]	+	positive rating	
	?	indeterminate rating	
	-	negative rating	
Level of evidence[153]	4	strong	Consistent findings in multiple studies of **good** methodological quality or in one study of **excellent** methodological quality
	3	moderate	Consistent findings in multiple studies of **fair** methodological quality or in one study of **good** methodological quality
	2	limited	One study of fair methodological quality
	1	unknown	Only studies of **poor** methodological quality
	0	conflicting	According to Terwee criteria (Appendix B)

Paper I

DEVELOPMENTAL MEDICINE & CHILD NEUROLOGY REVIEW

Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review

RANNEI SAETHER[1,2] | JORUNN L HELBOSTAD[3,4] | INGRID I RIPHAGEN[5] | TORSTEIN VIK[1,2]

1 Department of Laboratory Medicine, Children's and Women's Health, Faculty of Medicine, Norwegian University of Science and Technology; **2** Department of Paediatrics, St Olavs Hospital, Trondheim University Hospital; **3** Department of Neuroscience Norwegian University of Science and Technology, Trondheim; **4** Clinic for Clinical Services, St Olavs Hospital, Trondheim University Hospital; **5** Department of Cancer Research and Molecular Medicine Norwegian University of Science and Technology, Trondheim, Norway.

Correspondence to Rannei Saether, Department of Laboratory Medicine, Children's and Women's Health, Faculty of Medicine, Norwegian University of Science and Technology, Post-box 8905, NO-7491 Trondheim, Norway. E-mail: rannei.sather@ntnu.no

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We aimed to review tools used to assess balance in clinical practice in children and adults with cerebral palsy (CP), to describe their content and measurement properties and to evaluate the quality of the studies that have examined these properties. CINAHL, Embase, and PubMed/MEDLINE were searched. The COnsensus-based Standards for selection of health Measurement INstruments (COSMIN) was used to assess the 'quality of studies' and the Terwee criteria were used to assess the 'result of studies'. Twenty-two clinical balance tools were identified from 35 papers. The content and focus of the tools varied significantly. There was moderate or limited levels of evidence for most of the measurement properties of the tools; the strongest level of evidence was found for the Trunk Control Measurement Scale and the Level of Sitting Scale, in the category 'maintain balance', the Timed Up and Go and the Segmental Assessment of Trunk Control in the categories 'achieve balance' and 'restore balance' respectively. Information on responsiveness was scarce. Further studies providing better evidence for reliability and responsiveness for clinical balance tools are needed. In the meantime, results of studies evaluating effects of treatment of balance in individuals with CP should be interpreted with caution.

Impaired control of posture is the main component of the definition of cerebral palsy (CP).[1] The development of movement and posture may be altered by non-progressive damage to the brain and subsequent neurological impairments (spasticity, muscle weakness, co-contractions and visual impairment).[2,3] Studies indicate that children and adults with both mild and severe forms of CP have postural impairments.[4–9] Dysfunctional posture control interferes with the activities of daily life.[6]

Posture refers to the relationship between the different parts of the body, and between the body and a reference frame.[10] Control of posture is required in order to obtain balance, which may be defined as the act of maintaining, achieving, or restoring the centre of mass relative to the base of support. Balance is achieved by the complex integration of multiple body systems, including vestibular, visual, auditive, proprioceptive and higher level premotor systems. The functional goal of the balance system includes: (1) maintenance of a specific postural alignment, such as sitting or standing; (2) facilitation of voluntary movement, such as the movement transitions between postures; and (3) restoring equilibrium after external disturbances, such as a trip, slip, or push.[11]

There is no consensus on how, in a regular clinical setting, balance should be systematically assessed in individuals with CP. As balance is such a complex, task-dependent construct, it cannot be reflected by a single clinical balance tool.[12] Appropriate assessment tools are crucial in research as well as in the clinical management of CP. A good assessment tool should address the domain of concern, be reliable in the population of current interest, have good internal validity, be easily administered, and be responsive to change. Some of these criteria are not fulfilled for many assessment tools used to assess persons with CP.[13] Furthermore, many of the available tools were developed using children who did not necessarily have CP.[14] In a systematic review of assessment tools for children with CP, Ketelaar et al.[15] highlight the urgent need for outcomes that can be used to evaluate change. Several interventions have been proposed to improve balance in children and adults with CP, including hippotherapy and horseback riding,[16] constraint-induced therapy,[17] electrical stimulation,[18] virtual reality training,[19] adaptive seating,[20] and training on a moving platform.[21–23] These interventions are widely used in clinical practice, even though the evidence for their effectiveness is variable.[24,25] Describing and quantifying change in balancing abilities are, therefore, two central challenges to the research and clinical management of CP.[14]

A large number of systematic reviews of outcome measures of functional motor abilities[15,26] or activity limita-

tions[27,28] in CP have been published; however, these studies only address posture and balance as an integrated part of motor functions. A separate review aimed to identify and evaluate measures of balance activities in general for all neurological conditions.[29] Thus, as far as we know, no systematic review of assessment tools to evaluate balance in CP has been published to date.

The primary aim of our systematic review was to identify balance tools used in clinical practice aimed at assessing balance in children, adolescents, and adults with CP. The second aim was to describe the content of the tools. The third aim was to evaluate both the quality of the studies of the measurement properties and the results of the studies of the measurement properties.

What this paper adds

- This is first systematic review of clinical tools designed to assess balance in children and adults with CP.
- The study summarizes the content and measurement properties of the clinical balance tools.
- Information on the responsiveness of the clinical balance tools was found to be scarce.

METHOD

Search strategy

Searches were performed in CINAHL (through EbscoHost), Embase (through OvidSP, edition 1980 onwards), PubMed, and MEDLINE (through OvidSP, edition 1946 onwards) until 30 November 2012 (see Fig. 1). The first search aimed to identify clinical assessment tools for balance in CP. Both free-text terms and controlled terminology (MeSH, EMtree) were applied. Queries consisted of Boolean combinations of search term groups that represent the concepts 'cerebral palsy', 'balance', 'instrument', and 'properties'. In PubMed, additional search terms were used in an attempt to exclude reviews, animal studies, stroke, Parkinson's disease, and studies of children under the age of 4 years.

Subsequently, the names of the tools identified during the first search were used in a complementary search, which aimed to identify additional studies of the measurement properties of balance assessment tools. We applied Boolean combinations of search terms groups representing 'tool name in title' and 'validation'. Details of the database search are presented in Appendix S1 (online supporting information).

Finally, we conducted manual searches of the references in the included articles, reviews and published abstracts from conferences (The American Academy for Cerebral Palsy and Developmental Medicine and the European Academy of Childhood Disability, in the period 2008–2012). Studies assessing the measurement properties of the clinical balance tools were included for quality analysis.

Inclusion/exclusion criteria

Papers were included in our review if they described a tool that met the following criteria: (1) the tool was designed

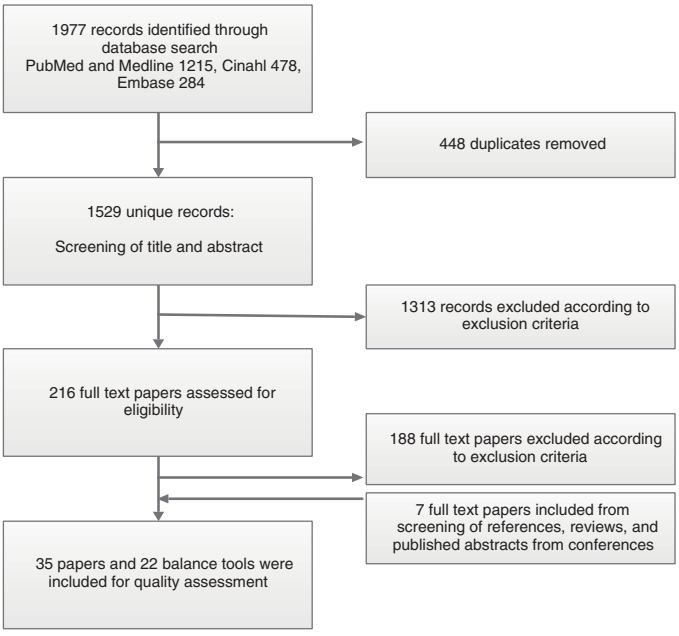

Figure 1: Processes performed to identify balance tools and studies of measurement properties.

for use in a clinical setting and assessment was by a clinician in a hospital or in the community, without the need for laboratory equipment; (2) the measurement properties of the tool had been evaluated in individuals with CP older than 4 years of age (since most children with CP are diagnosed by this age);[30] and (3) the paper was published in English in peer-reviewed journals or as theses. Papers were excluded if: (1) the primary intention of the tool was not to assess balance, the balance assessment was part of a wider assessment of motor function (i.e. if a specific 'balance score' could not be extracted from the assessment), (2) individuals with CP comprised less than 30% of the total population, (3) the papers were reviews, and (4) the papers were case studies.

Article selection

Two of the authors (RS and IR) independently reviewed titles and abstracts. To ensure consistent interpretation of the inclusion and exclusion criteria, the authors assessed a sample of 35 references from the list of identified papers before extraction of data. Disagreements in the interpretation of whether the criteria were met were discussed with a third author (TV). In cases of uncertainty, the paper in question was included for full text review.

Data extraction and quality assessment

Two of the authors (RS and TV) independently extracted data and performed the quality assessment of the data. The results were discussed until agreement was reached. The characteristics of the clinical balance tools of the included clinical balance tools (i.e. focus of assessment, clinical utility, scale construction, and characteristics of the study population), were extracted into a pre-established review table, adapted from the *CanChild* Outcome Measures Rating Form.[31] The focus of each tool was described according to the function, activity or participation domains of the International Classification of Functioning, Disability and Health (ICF),[32] primary purpose (discriminate, evaluate, and predict), functional goal of balance ('maintain', 'achieve', and 'restore'), whether it assessed capacity or performance and assessment position (sitting, standing, or walking). The clinical utility deals with the time to complete the assessment and the qualifications of the examiner, the scale construction deals with the tool's items and the characteristics of the study population cover age, sex, and the severity of CP.

The methodological quality of the studies was rated according to the COnsensus-based Standards for selection of health Measurement INstruments (COSMIN).[33] The COSMIN checklist consists of nine measurement properties, with 5 to 18 items per measurement property that address aspects of design and statistical methods. The measurements' properties are internal consistency, reliability, measurement error, content validity, construct validity (structural validity, hypothesis testing, cross-cultural validity), criterion validity, and responsiveness.[33] A study's quality is rated on a four-point rating scale ('poor', 'fair', 'good' or 'excellent') for each of the seven properties.[34] In accordance with the COSMIN, the overall quality score per measurement property was determined by the lowest rating of any of the items ('worst score counts'). However, according to the recommendations, not all criteria need to be used exactly as defined in the manual (i.e. they may be treated as a rule of thumb).[34] Hence, adaptations can be made in individual reviews and for our review we made two adaptations to the criteria. The first adaption was related to the description of missing items. Since none of the studies had indicated the number of missing cases, it seemed illogical to score how missing items were handled. Thus, the scoring on how missing items were handled was excluded. The second amendment was related to the scoring of the number of participants included, where according to the COSMIN guidelines, studies with samples of less than 30 participants were rated as 'poor'. Instead we rated sample sizes between 21 and 30 as 'fair', as has been done in other studies, since the number of participants mainly affects the precision of the estimates.[35]

Moreover, the terms used in the papers to describe measurement properties were not always identical to those used in the COSMIN and, in some cases, the definitions of the measurement properties could also differ somewhat from the definitions proposed by the COSMIN group. In such cases we applied the COSMIN taxonomy instead of the description in the paper.[36] A high level of interrater agreement for the COSMIN checklist has been documented.[37]

The results obtained in each study, regarding the value of the measurement properties, were assessed using Terwee's criteria.[38] The results of the studies were rated as 'positive', 'indeterminate' or 'negative' (see Table I). However, we did not apply this rating to content validity because the criteria proposed by Terwee are quite similar to the rating proposed by the COSMIN.

Data synthesis

In order to determine an 'overall score' for each measurement property per clinical balance tool, we estimated the level of evidence (Table I) in a similar manner to that recommended by the Cochrane Back Review Group[39] and used in a number of reviews of measurement properties.[35,40,41] The level of evidence combines the number of studies, consistency of rating of results according to Terwee[38] and the quality of studies according to the COSMIN[34] in a rating of the documented measurement properties per clinical balance tool as 'strong', 'moderate', 'limited', 'unknown', or 'conflicting'.

Tool categories

The control of balance has been identified to be associated with three broad categories of human activity[11,42] and we therefore divided the clinical balance tools into the same three categories: the ability to: (1) 'maintain balance', which refers to stability limits, i.e. how far can the body move over its base of support before changing support or

Table I: Description of scoring criteria used to assess the quality of studies, results of studies and level of evidence for measurement properties per balance tool

	Scoring	Level	Description
COSMIN[34]		Excellent	
		Good	
		Fair	
		Poor	
Terwee criteria[38]	+	Positive rating	
	?	Indeterminate rating	
	−	Negative rating	
Level of evidence[39]	4	Strong	Consistent findings in multiple studies of *good* methodological quality *or* in one study of *excellent* methodological quality
	3	Moderate	Consistent findings in multiple studies of *fair* methodological quality *or* in one study of *good* methodological quality
	2	Limited	One study of *fair* methodological quality
	1	Unknown	Only studies of *poor* methodological quality
	0	Conflicting	According to Tertwee criteria

losing balance and postural alignment; (2) 'achieve balance', which refers to the anticipation of a postural transition from one body position to another (in this study: mainly transition from one base of support to another); and (3) 'restore balance', which refers to postural responses to external perturbation. We added a further category, called 'other', for tools that did not fit the other three categories. These categories are broad representations of human activity, and a clinical balance tool could therefore fit with more than one category. However, we chose to select only one category, namely the one that fitted the content of the tool best. In addition, we also classified the balance tools into focus categories, based on the main focus of the tools, as it was described by the authors of the included papers.

RESULTS

Study selection

The database searches revealed 1529 records of possible interest. Figure 1 shows the selection process used to identify relevant papers. After screening the titles and abstracts of all identified papers, 1313 records were excluded and 216 records included for a full text check. After a full text check, a further 188 papers were excluded. Finally, seven papers were added after screening the references, reviews and published abstracts from conferences, and a total of 35 papers and 22 clinical balance tools were included for quality assessment. The 22 tools were the Berg Balance Scale (BBS), the Functional Reach Test (FRT), the Functional Walking Test (FWT), the Heel-to-Toe Stand (HTS), the Level of Sitting Ability (LSA), the Level of Sitting Scale (LSS), the Modified Posture Assessment Scale (MPAS), the Pediatric Balance Scale (PBS), the Pediatric Reach Test (PRT), the Pediatric Clinical Test of Sensory Interaction for Balance (P-CTSIB), the Posture Assessment Scale (PAS), the Posture and Posture Ability Scale (PPAS), the Seated Posture Control Measurement (SPCM), the Segmental Assessment of Trunk Control (SATCO), the Sitting Assessment for Children with Neuromotor Dysfunction (SACND), the Sitting Assessment Scale (SAS), the Spinal Alignment for Range of Motion Measure (SAROMM), the Timed One-Leg Stance (TOLS), the Timed Up and Down Stairs (TUDS), the Timed Up and Go (TUG), the Trunk Control Measurement Scale (TCMS), and Trunk Impairment Scale (TIS). Two of the clinical balance tools, PPAS and SPCM, were evaluated for adults (see Table II).

In accordance with the inclusion criteria, some commonly used clinical assessment tools were excluded from our review. Among the excluded tools were the following for which the measurement properties have not been tested in individuals with CP: the Ayres Southern California Sensory Integration Test, the Balance Evaluation System Test, the Bruininks–Oseretsky test of Motor Proficiency (only running speed has been tested in cases of CP), the Community Balance and Mobility Scale, the Movement Assessment Battery for Children (only the checklist has been tested in cases of CP), the Peabody Developmental Gross Motor Scale (tested in children with CP aged < 4y), and the Unified Balance Scale. Other tools were excluded because their primary purpose was not to assess balance; rather, the balance assessment was part of a wider assessment of motor function. These tools were the Ability for Basic Movement Scale for Children Type T, the ABILOCO-Kids Questionnaire, the Assessment of Behavioral Components Functional Ambulatory Categories, the Functional Mobility Scale, the Gillette Functional Assessment Questionnaire, the Gross Motor Function Measure, the Gross Motor Performance Measure, the Lateral Step Up Test, the Mobility Questionnaire, the Pediatric Evaluation of Disability Inventory, the Pediatric Outcomes Data Collection Instrument, the Pediatric Functional Independence Measure, the Activities Scale for Kids Performance, and the Quality of Upper Extremity Skills Test. The assessment of the Slump Test and the Standardized Walking Obstacle Course was based either on a case study or on a study in which the individuals with CP comprised less than 30% of the total study population.

Table II: Focus (according to ICF domain), clinical utility (time to complete and examiner qualifications), and scale (including number of items) of included balance tools as well as characteristics of the populations in which the tools were applied

	Focus					Clinical utility		Scale		Characteristics of study population				
Balance tool	ICF domain[a]	Primary purpose[b]	Type of balance[c]	Capacity/ performance[d]	Position[e]	Time to complete (min)	Examiner qualifications[f]	Level[g]	Number of items	Age (year)	Sex[h]	Total included[i]	GMFCS[j] (1–5)	Classification[k]
BBS[44,67]	A	D+E	M+A	C	Si+St	10–20	Na	O	14	5–12	53M+26F	66CP+14TD	1–4	21UI+37BI+3Q +3A+2Dy
FRT[44,45,65]	A	D+E	A	C	St	3	Nr	R	1	5–15	34M+22F	77CP+ 27TD+5O	1–4	15UI+15BI+3Q +3A+2Dy
FWT[53]	A	D+E	M+A	C	St+W	10	Nr	O	11	4.2–17	35M+21F	56CP	1–3	33UI+23BI
HTS[76]	A		M+A	C	St	Few	Na	R	2	5–12	6M+9F	15CP		5UI+7BI+ 1Dy+2A
LSA[43,66]	A	D+E+P	M+A	P	Si		Na	O	7 levels	1.5–19		148CP+68TD		4UI+12Q+ 15Dy+3 mixed
LSS[46–48,68]	A	D+E	M+A	P	Si	5–10	Na	O	8 levels	1–18	111M+94F	131CP+104O		1BI+2UI+ 19Q+1T
MPAS[77]	B	E	M	P	St	2–4	Na	O	3			3CP		
PAS[71]	B	E	M	P	St	2–4	Na	O	5	2.4–9.6	7M+4F	11CP		8BI+3Q
PBS[49,60,61,63]	A	D+E	M+A	C	Si+St	<15	Na	O	14	5–15	49M+45F	109CP		10M+5Mo+ 5Mo/S
P-CTCIB[69,70]	B+Ef	D	M	C	St	5	Na	O	6 conditions	6–14	18M+17F	35CP		8UI+24BI+3Q
PPAS[62]	B+A	D+E	M+A	P	Si	5	Na	O	7 levels	19–22		30 CP	1–5	
PRT[50]	A	D+E	A	C	Si+St+Ly	<15	Rec	R	6	2.6–14.1	15M+14F	10CP+19TD	1–4	5UI+2BI+3Q
SACND[54,75]	B+A	E	M+A	C/P	Si	2×5	Na	O	2 rest/reach	2.2–10.8	12M+10F	46CP+ 10TD+1O		24BI
SAROMM[59]	B	D	M+A	P	Si	15–30	Rec	O	26 4 alignment	2–18	17M+8F	25CP	1–5	3UI+12BI+10Q
SAS[52,73,74]	B+A+Ef	D+E	M+A	C/P	Si	5each position	Na	O	5 6 positions	2.6–16.2	8M+15F	33CP		25BI+3T+5Dy
SATCO[51,72]	B+A+Ef	D+E	M+A+R	C	Si		Nr	N	7	1.6–17		33CP+ 8TD+3O	1–5	6UI+10Q+1T +2Dy+2H
SPCM[46,47,68]	B+A	E	M+A	C/P	Si	<20–40	Rec	O	22 alignment 10 function	1.3–27.9	111M+94F	133CP–92O		1UI+1BI+ 15Q+1T
TCMS[55]	B+A	D+E	M+A	C	Si		Na	O	15	8–15	30M+26F	26CP+30TD	1–3	7UI+17BI+2Q
TIS[58]	B+A	D+E	M+A	C	Si	10	Nr	O	17	5–12	10M+15F	20CP+5TD	1–4	5UI+15BI
TOLS[56,65]	A		M+A	C	St	Few	Na	R	1	6.9–14	34M+36F	29CP+41TD	1–3	11UI+15BI +2Q+1H
TUDS[65]	A	D+E	A	C	W	<5	Na	R	1	8–14	14M+13F	27CP	1–3	
TUG[44,57,64,65]	A	D+E	A	C	Si+ St+W	<5	Na	R	1	3–17.5	50M+39F	113CP+ 27TD+8O	1–4	15UI+2BI+3Q+ 2Dys+3A+1H

Balance tools: BBS, Berg Balance Scale; FRT, Functional Reach Test; FWT, Functional Walking Test; HTS, Heel-to-Toe Stand; LSA, Level of Sitting Ability; LSS, Level of Sitting Scale; MPAS, Modified Posture Assessment Scale; PAS, Posture Assessment Scale; PBS, Pediatric Balance Scale; P-CTSIB, Pediatric Clinical Test of Sensory Interaction for Balance; PPAS, Posture and Postural Ability Scale; PRT, Pediatric Reach Test; SACND, Sitting Assessment for Children with Neuromotor Dysfunction; SAROMM, Spinal Alignment for Range of Motion Measure; SAS, Sitting Assessment Scale; SATCO, Segmental Assessment of Trunk Control; SPCM, Seated Posture Control Measurement; TCMS, Trunk Control Measurement Scale; TIS, Trunk Impairment Scale; TOLS, Timed One-Leg Stance; TUDS, Timed Up and Down Stairs; TUG, Timed Up and Go. [a]B, Body functions/-structures; A, Activities; P, Participation; Ef, Environmental factors. [b]E, evaluate; D, discriminate; P, predict. [c]M, maintain; A, achieve; R, restore. [d]C, capacity; P, performance (suggested by us, not described in papers). [e]Ly, lying; Si, sitting; St, standing; W, walking. [f]Rec, recommended; Nr, not required; Na, not addressed. [g]N, nominal; O, ordinal; I, interval; R, ratio. [h]M, male; F, female; (sex was not reported in all papers, and the number will differ from total included in [i]). [i]The total number included was the total number of participants in all studies per balance tool. CP, cerebral palsy; TD, typical development; O, Other. [j]GMFCS, Gross Motor Function Classification System. [k]BI, bilateral; Dy, dyskinetic; UI, unilateral; A, ataxia; Q, quadriplegia; T, triplegia; M, mild; Mo, moderate; S, severe; H, hypotonia.

Table III: The reported measurement properties of the included balance tools and their level of evidence

Category				Reliability												
				Internal consistency			Measurement error			Intrarater			Interrater			
Balance control		Focus of tool	Tool	Ref	Level of evidence	Result	Ref	Level of evidence	Result	Ref	Level of evidence	Result	Ref	Level of evidence	Result	
'Maintain'/ Stability (s) Posture (p)	p	'Posture'	MPAS										77	Unknown	+	
	p/s	'Posture'	PAS										71	Unknown	?	
	p	'Posture'	PPAS	62	Unknown	+							62	Limited	+	
	s	'Posture'	SAROMM				59	Limited[d]	?				59	Limited[d]	+	
	s	'Reaching'	FRT				45	Limited	?	44,65	Moderate	++	44,65	Moderate	+ +	
	p/s	'Reaching'	PRT							50	Limited	+	50	Limited	+	
	p/s	'Seating'	LSA													
	p/s	'Seating'	LSS							47	Moderate	+	46,68	Moderate	+ – [nc]	
	p/s	'Seating'	SAS							52,74	Unknown	??	52,73,74	Unknown	? ? ?	
	p/s	'Seating'	SPCM										46,47,68	Moderate	+ + +	
	p/s	'Trunk control'	SACND	75	Unknown	–							54,75	Limited	+ +	
	p/s	'Trunk control'	TCMS	55	Unknown	?	55	Moderate	+				55	Moderate	+	
		'Trunk control'	TIS				58	Limited	?	58	Limited	+	58	Limited	+	
'Achieve'		'Every-day task'	BBS										44	Limited	+	
		'Every-day task'	PBS							60,63	Limited	++	49,60,63	Moderate	+++	
		'Stability in gait'	FWT	53	Limited	+				53	Moderate	+	53	Moderate	+	
		'Stability in gait'	TUDS							65	Limited	+	65	Unknown	+	
		'Stability in gait'	TUG							65	Limited	+	57	Limited	+	
		'Standing'	HTS										76	Unknown	+	
		'Standing'	TOLS							65	Limited	+	56,65	Unknown	++	
'Restore'		'Trunk control'	SATCO+							51,72	Limited	+ ?[nc]	51,72	Limited	+ ?[nc]	
		'Sensory'	P-CTSIB										69	Limited[b]	+	
'Other'													69	Limited[c]	–	
													70	Unknown[a]	+	

Balance tools: BBS, Berg Balance Scale; FRT, Functional Reach Test; FWT, Functional Walking Test; HTS, Heel-to-Toe Stand; LSA, Level of Sitting Ability; LSS, Level of Sitting Scale; MPAS, Modified Posture Assessment Scale; PAS, Posture Assessment Scale; PBS, Pediatric Balance Scale; P-CTSIB, Pediatric Clinical Test of Sensory Interaction for Balance; PPAS, Posture and Postural Ability Scale; PRT, Pediatric Reach Test; SACND; Sitting Assessment for Children with Neuromotor Dysfunction; SAROMM, Spinal Alignment for Range of Motion Measure; SAS, Sitting Assessment Scale; SATCO, Segmental Assessment of Trunk Control (+could also belong into 'Maintain'); SPCM, Seated Posture Control Measurement; TCMS, Trunk Control Measurement Scale; TIS, Trunk Impairment Scale; TOLS, Timed One-Leg Stance; TUDS, Timed Up and Down Stairs; TUG,Timed Up and Go. *Level of evidence:* see Table I. *Result of study* (Terwee criteria): +, positive rating; ?, indeterminate rating; –, negative rating; +/–, conflicting; na, not assessed (see Table I); nc, not rated as conflicting (methods not comparable); ref, reference. [a]Time–sway. [b]Stance duration. [c]Motor Strategy. [d]Spine subscale. [e]Cross-cultural validity is not described in the Table (rating=unknown). *Balance control category:* The control of balance has been identified as associated with three broad categories of human activity: the ability to (1) *Maintain balance*, which refers to *stability limits(s)*, i.e. how far can the body move over its base of support before changing support or losing balance, as well as postural alignment (p), (2) *Achieve balance*, which refers to the anticipation of a postural transition from one body position to another (in this study; mainly transition from one base of support to another), and (3) *Restore balance*, which refers to postural responses to external perturbation. A further category called 'other' was added for tools that did not fit into the other three categories. The tools could fit in *more than one* category, but only one category, i.e. the one that best fitted the content of the tool was selected.

Characteristics of the clinical balance tools

The 22 clinical balance tools varied in focus, clinical utility, scale construction and characteristics of the study population (see Table II). The focus of 11 tools was within the body functions domain (ICF) while the focus of 18 tools was on the activity domain (ICF). Even if not clearly stated, the primary purpose could be assumed for the majority of the tools. The primary purpose of 16 tools was to discriminate between different levels of gross motor function in CP, and between individuals with CP and individuals with typical development, and/or evaluate treatments, and one tool also aimed to predict deformities. The ability to discriminate was assessed in 13 tools, while evaluative ability was assessed for only two tools. The ability to 'maintain balance' (n=18) and 'achieve balance' (n=19) was most commonly assessed, while only one tool assessed the ability to 'restore balance'. Balance was mainly assessed in sitting (n=14) or in standing position (n=11), while three tools assessed balance during walking.

Validity																
Test-retest			Content			Construct			Criterion			Responsiveness				
Ref	Level of evidence	Result	Ref	Level of evidence	Result	Ref	Level of evidence	Result	Ref	Level of evidence	Result	Ref	Level of evidence	Result	Total
															1
															1
						62	Limited	+							5
59	Limited[d]	+	59	Strong[d]	na	59	Limited[d]	+							12
44	Limited	+	45	Strong	na	44,45	Limited	+ ?							16
			50	Strong	na	50	Limited	+	50	Limited	–				12
			66	Strong	na	43	Unknown	?							5
46,68	Moderate	+ – [nc]	48,68	Strong	na	48	Moderate	?				47	Limited	?	18
			52,73,74	Strong	na										6
46,68	Limited	+ –[nc]	68	Strong	na							47	Limited	?	11
54	Limited	+	75	Strong	na	75	Unknown	–							10
55	Moderate	+	55	Strong	na	55	Moderate	+							17
			58	Strong	na	58	Limited	?							12
44	Limited	+	67	Strong	na	44,67	Limited	+ ?							10
49	Unknown	+	49	Moderate	na	63	Limited	?							11[e]
						53	Unknown	?							9
65	Unknown	+				65	Limited	?							7
44,64	Limited	+	44,57	Strong	na	44,57,64	Limited	+??							12
															1
			56,76	Strong	na										7
			51	Strong	na	51	Limited	+							10
															2
			69,70	Strong[c]	na	69	Limited[c]	+							8
															1

Clinical utility deals with issues such as examiner qualifications and time needed to complete the assessment. The qualifications needed to perform the assessment were seldom addressed specifically and were difficult to assume for 15 of the tools. For four tools we assumed that specific qualifications were not required, due to the design of the tool, whereas for three tools we assumed that specific qualifications were required. With the exception of four tools, the majority of the tools could be completed in less than 15 minutes. The *s*cale construction varied from seven tools with less than five items to the majority with between five and 15 items. Some of the tools were adaptations or modifications of other tools. This was the case for the PPAS and the LSS, which were adapted from the LSA. Further, the MPAS was modified from the PAS, the PBS was modified from the BBS, the PRT was adapted from the FRT and the TCMS was expanded from the TIS. Only a few tools (the PAS, MPAS, SAROMM, and SAS) were originally developed to be applied in studies of individuals with CP. The other tools were originally developed to assess balance in other populations, such as 'older people' (the BBS, FRT, P-CTSIB, TIS, and TUG), 'handicapped' children (the LSA, SPCM, SATCO, and SACND) and one tool was developed to study balance in typical developing children without disabilities (the FWT). The descriptions of the population with CP in which the tools had been applied varied. Half of the studies reported the Gross Motor Function Classification System (GMFCS) levels, while the other half reported clinical descriptions such as the topographical distribution and severity of CP. Some of the latter studies were published before GMFCS levels were available, but we assumed that seven tools would be appropriate for children in GMFCS levels I–V (according to the content of the tool), nine tools for children in GMFCS levels I–IV, four tools for children GMFCS I–III, and two tools for children with GMFCS levels I–II.

In 23 of the 35 papers the main aim was to study the measurement properties of a given tool.[43–65] In the other 12 papers, some measurement properties of a tool were assessed even though that was not the main aim of the paper.[66–77]

Quality of the studies

Details of how the quality of the measurement properties were addressed in accordance with the COSMIN[34] are

summarized in Tables SI and SII. In some studies more than one measurement property of reliability (intra-/interrater and test–retest, internal consistency, and measurement error) and validity (content, construct, and criterion) was assessed. Thus, in the 35 papers, a total of 70 reliability properties and 49 validity properties were assessed. The quality of how reliability properties had been studied was rated as 'poor' (n=23), 'fair' (n=40), and 'good' (n=7); none was rated as 'excellent'. Furthermore, the quality of how validity properties had been studied was rated as 'poor' (n=7), 'fair' (n=16), 'good' (n=6), and 'excellent' (n=20). The main reasons for the low quality scores were lack of a priori formulated hypotheses and the limited numbers of participants included in the studies, while content validity, including face validity, was the only validity property rated as excellent. Only five of the included studies had included more than 50 participants, the number of participants required to obtain a good quality score according to the COSMIN.[34]

Results of studies

The ratings of the reported results of the studies, according to Terwee criteria,[38] are summarized in Tables SI and SII, online supporting information. The results of the studies for reliability (n=70) were rated as 'negative' (n=5), 'indeterminate' (n=13), and 'positive' (n=52) (Table SI). The results of 21 studies of validity properties, excluding content validity (see section headed 'Data extraction and quality assessment') were 'negative' (n=2), 'indeterminate' (n=10), and 'positive' (n=9) (Table SII).

Data synthesis

In an attempt to help clinicians to select the appropriate tool for a specific clinical purpose, we divided the tools into four balance categories: 'Maintain balance', 'Achieve balance', 'Restore balance', and 'Other'. In addition, we identified eight different focus categories, based upon the authors' descriptions of the tools. We named these focus categories 'posture', 'reaching', 'seating' (adaption of seating position), 'trunk control' (quality of sitting), 'every-day tasks', 'stability in gait,' 'standing', and 'sensory' (influence of different sensory conditions). Table III shows an overall summary of measurement properties of the included tools that were studied and categorized, as well as the level of evidence of the properties in patients with CP. Table III shows that interrater reliability and content validity have been well studied and that there are strong levels of evidence for content validity for the majority of the tools, whereas the level of evidence for interrater reliability was assessed as moderate for only six tools (none was rated as strong). Regarding construct validity, as well as intrarater and test–retest reliability, the evidence of most tools was found to be limited or unknown. Table III also shows that internal consistency, measurement error, criterion validity, and responsiveness were reported and the level of evidence was mostly limited.

Overall, some tools within each category had a stronger level of evidence of the measurement properties than other tools (see description of level of evidence in Table I); however, most of the measurement properties of the tools were assessed as having moderate and limited levels of evidence. Within the category 'Maintain balance', two tools, the TCMS and the LSS, had the strongest level of evidence; within the category 'Achieve balance', the TUG had the strongest level of evidence; and within the categories 'Restore balance' and 'Other', the SATCO (could also have been in the 'Maintain balance' category) and the P-CTSIB were the only tools. We found the strongest level of evidence for the following tools within the categories related to the focus of the tool: the SAROMM ('posture'), FRT ('reaching'), LSS ('seating'), TCMS ('trunk control'), PBS ('every-day task'), TUG ('stability in gait'), TOLS ('standing'), and P-CTSIB ('sensory').

DISCUSSION

Our systematic review identified 22 tools that can be used in a regular clinical setting to assess balance in children with CP. Two tools were also validated in adults with CP. We found a strong level of evidence for content validity for the majority of the tools, while evidence was more limited for their intrarater, interrater, and test–retest reliability. In addition, there was little or no evidence for internal consistency, measurement error, criterion validity, and responsiveness for the vast majority of the tools.

The strengths of our study are that both the methodological quality of how the studies assessed the measurement properties and the results of the studies (i.e. the reported measurements properties) were taken into account.[34] This point is important because the reported measurement properties of a tool can hardly be trusted if the methodological quality of the study from which the properties were obtained is poor. In this study, the methodological quality of the reported studies was rated according to the COSMIN four-point rating scale (poor, fair, good, and excellent), a scoring system that has been applied in several systematic reviews.[34] Other standards for assessment of measurement properties have been published, but they were not developed to be used in systematic reviews.[34] However, we are aware of the limitation that the reliability of the COSMIN four-point rating scale has not yet been assessed, although it has been developed from the COSMIN checklist, which has a high level of interrater agreement.[37]

A further strength of our study is the description of how the methodological quality and the results of various studies were combined. de Vet et al.[78] argued that in order to grade each measurement property, the quality of the reviewed studies should be integrated with the results of those studies, although the methodology to do this is not yet well developed. The criteria for the level of evidence that we used were originally designed for systematic reviews of clinical trials,[39] but we believe that they also are applicable for reviews of measurement properties.

In order to help clinicians and researchers to determine the underlying cause of balance problems we organized the tools into different balance categories; although we are aware that these categories were broad and that one tool

may fit into more than one category. Moreover, the different modes of balance control, 'maintain', 'achieve', and 'restore balance', are not separate entities but also occur in combination. In addition, to help choosing the appropriate outcome measures for a specific intervention we also classified the focus of the tools into eight focus categories.

A limitation of the review is the inclusion of studies published in English only, as we may have missed tools developed by researchers publishing in other languages. A further limitation may be the exclusion of tools to assess balance in laboratory settings, such as electromyography (EMG), kinetic, and kinematic analyses.[79] However, we consider that a separate systematic review of such tools would be more appropriate, since it has been recommended that informative reviews should be written on a limited number of tools, including more details on each tool, than to write larger reviews that may lack some relevant information.[78] We also excluded clinical balance tools for children less than 4 years of age in this review, since in this case we consider a separate review of such tools to be more appropriate. Moreover, the inherent heterogeneity of the population with CP poses a limitation for the synthesis of data, since ideally only results obtained in homogeneous populations should be combined.[78] On the other hand, a heterogeneous sample, representing the entire population in which a tool should be used, is important in studies of measurement properties. Finally, it may be a limitation that we included studies where the main aim was not to assess the measurement properties of the tool, because it may be difficult to find these articles in a structured way, and it is often more difficult to interpret the evidence for validity, because no hypothesis has been formulated or tested.[78]

We are not aware of any previous systematic review of clinical balance tools in the population with CP. Tyson and Connell[29] published a review of psychometric properties and clinical utility of all available balance tools in all neurological conditions. They identified 30 tools, but the measurement properties of the tools have not been tested in the population with CP. Westcott et al.[12] presented a review of evaluations of postural stability in children in general and issues affecting the testing of this construct. Three of the tools, the FRT, TUG, and P-CTSIB, in the Westcott et al.[12] review were identified and included in our review.

The large number of tools identified through our search may reflect the many different focuses of the tools and the broad variety of impairments in control of balance inherent in the population with CP, as well as the lack of a uniform definition of balance. This means that the selection of an appropriate tool depends on the specific purpose of the assessment, the situation, and the patient's level of impairment. We attempted to assist clinicians and researchers in selecting appropriate clinical balance tools by dividing the tools into categories. Our assessment of the level of evidence may provide further help in choosing the most appropriate tool in the different categories. However, we are aware that a tool may fit into more than one of the categories. Despite the significant heterogeneity of the clinical picture of CP, it is an intriguing idea whether it is possible to assess balance in this population by using a more 'general' balance tool. Two such potential tools could be the Balance Evaluation System Test (BESTest)[80] or the Mini-BESTest,[81] which aim to identify the underlying postural control systems responsible for poor functional balance. However, the measurement properties of these tools have only been assessed in individuals with diagnoses other than CP. However, it may be argued that in this heterogenetic population one has to accept a number of different tools, depending on the purpose of the assessment.

We found a strong level of evidence for content validity for the majority of the tools. This may in part be due to the more interpretative way in which content validity is assessed compared with the other measurement properties, whereby the raters (RS and TV) had to judge to what extent the described items included in a tool reflected relevant aspects of the construct to be measured. It is also possible that the rating of the content validity, in accordance with the COSMIN, was not sensitive enough to differentiate between studies. In contrast to the high level of evidence for content validity, the measurement property reliability demonstrated a low level of evidence. The main reason for this result is that the studies were rated as 'fair' or 'poor' because of the low number of participants included in the studies. Users of the COSMIN are encouraged to judge what sample size is adequate for their purpose.[34] Consistent with this recommendation, we adjusted the number of individuals required to obtain an acceptable score, since the results of the reliability studies showed very high intraclass correlation coefficients values.[78] Nonetheless, only seven of the 70 properties of reliability were rated as good, and evidence was consistently rated as low.

Our review revealed that internal consistency, measurement error, criterion validity, and responsiveness have seldom been reported and the level of evidence is mostly limited (see Table III). The reason for little or no evidence for internal consistency may be that this measurement property is mainly assessed in questionnaires. In the case of criterion validity and responsiveness, the reason for little or no evidence may be due to our use of the COSMIN taxonomy,[36] as we interpreted the description given in the paper to fit with this taxonomy. Accordingly, we assigned studies of criterion validity as studies of construct validity. Moreover, according to the COSMIN taxonomy, responsiveness refers to the ability of a tool to detect change over time in the construct to be measured.[36] de Vet et al.[78] state that the treatment effects measured by a specific tool and responsiveness of the same tool should not be based on the effect size from the same study (i.e. the effect size is zero, either the intervention has no effect or the tool is not responsive). Effect size only has meaning as a measure of responsiveness if the magnitude of the effect of the intervention is known or assumed in advance.[78] Consequently, we did not include studies primarily aimed at assessing the effect of interventions in the systematic review of measurement properties. However, we are aware that there is little agreement on how this measurement property should be assessed.[82]

The limited reporting and level of evidence for responsiveness discussed above have implications for practice and research, since for most of the tools the intention was to evaluate the development and treatment of balance in the population with CP. Our findings strongly suggest that more studies of good quality are needed to assess the responsiveness, with a focus on meaningful change, of almost all the clinical balance tools included in this review. This means that the results of reviews of effect of treatments, such as hippotherapy and horseback riding,[16] and physical therapy techniques[24] on balance performance, should be interpreted with caution. The results of the studies of reliability in this review should also be interpreted with caution. Two out of three studies were rated as 'positive', but the quality of the studies assessing reliability was in most cases rated as poor to fair. Thus, increasing awareness and the use of checklists such as the COSMIN rating system may help in designing and reporting future studies of high quality.

CONCLUSION

Twenty-two tools developed to assess balance in a clinical setting were identified from 35 papers assessed in this review. The measurement properties of the tools were mainly studied in children; we found only two tools for adults. Within the categories of balance control, we found the strongest level of evidence for the Trunk Control Measurement Scale and the Level of Sitting Scale ('maintain balance' category), Timed Up and Go ('achieve balance' category), and the Segmental Assessment of Trunk Control ('restore balance' category). There is a need for further studies in order to provide more levels of evidence for the reliability and responsiveness of clinical balance tools. In the meantime, the results of studies evaluating the effects of treatment of balance in individuals with CP should be interpreted with caution.

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SUPPORTING INFORMATION

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

Appendix S1: Database search.

Table SI: The quality of studies assessing reliability of included balance tools and the rating of the reported results of the studies.

Table SII: The quality of studies assessing validity of included balance tools, and the rating of the reported results of the studies.

REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl* 2007; **49**(Suppl. 109): 8–14.
2. Gormley ME Jr. Treatment of neuromuscular and musculoskeletal problems in cerebral palsy. *Pediatr Rehabil* 2001; **4**: 5–16.
3. Carlberg EB, Hadders-Algra M. Postural control in sitting children with cerebral palsy. In: Hadders-Algra M, Carlberg EB, editors. Postural Control: A Key Issue in Developmental Disorders. Clinics in Developmental Medicine No. 179. London: Mac Keith Press, 2008: 74–96.
4. Heyrman L, Desloovere K, Molenaers G, et al. Clinical characteristics of impaired trunk control in children with spastic cerebral palsy. *Res Dev Disabil* 2012; **3**: 327–34.
5. Prosser LA, Lee SC, VanSant AF, Barbe MF, Lauer RT. Trunk and hip muscle activation patterns are different during walking in young children with and without cerebral palsy. *Phys Ther* 2010; **90**: 986–97.
6. de Graaf-Peters VB, Blauw-Hospers CH, Dirks T, et al. Development of postural control in typically developing children and children with cerebral palsy: possibilities for intervention? *Neurosci Biobehav Rev* 2007; **31**: 1191–200.
7. Burtner PA, Woollacott MH, Craft GL, Roncesvalles MN. The capacity to adapt to changing balance threats: a comparison of children with cerebral palsy and typically developing children. *Dev Neurorehabil* 2007; **10**: 249–60.
8. Carlberg EB, Hadders-Algra M. Postural dysfunction in children with cerebral palsy: some implications for therapeutic guidance. *Neural Plast* 2005; **12**: 221–8.
9. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Balance in relation to walking deterioration in adults with spastic bilateral cerebral palsy. *Phys Ther* 2012; **92**: 279–88.
10. Hadders-Algra M, Carlberg EB. Why bother about postural control? In: Hadders-Algra M, Carlberg EB, editors. Postural Control: A Key Issue in Developmental Disorders. Clinics in Developmental Medicine No. 179. London: Mac Keith Press, 2008: 1–2.
11. Mancini M, Horak FB. The relevance of clinical balance assessment tools to differentiate balance deficits. *Eur J Phys Rehabil Med* 2010; **46**: 239–48.
12. Westcott SL, Lowes LP, Richardson PK. Evaluation of postural stability in children: current theories and assessment tools. *Phys Ther* 1997; **77**: 629–45.
13. Vargus-Adams J. Understanding function and other outcomes in cerebral palsy. *Phys Med Rehabil Clin N Am* 2009; **20**: 567–75.
14. Rosenbaum PL, Rosenbloom L. Outcomes. In: Rosenbaum PL, Rosenbloom L, editors. Cerebral Palsy: From Diagnosis to Adult Life. London: Mac Keith Press, 2012: 151–60.
15. Ketelaar M, Vermeer A, Helders PJ. Functional motor abilities of children with cerebral palsy: a systematic literature review of assessment measures. *Clin Rehabil* 1998; **12**: 369–80.
16. Zadnikar M, Kastrin A. Effects of hippotherapy and therapeutic horseback riding on postural control or balance in children with cerebral palsy: a meta-analysis. *Dev Med Child Neurol* 2011; **53**: 684–91.
17. Zipp GP, Winning S. Effects of constraint-induced movement therapy on gait, balance, and functional locomotor mobility. *Pediatr Phys Ther* 2012; **24**: 64–8.
18. Karabay I, Dogan A, Arslan MD, Dost G, Ozgirgin N. Effects of functional electrical stimulation on trunk control in children with diplegic cerebral palsy. *Disabil Rehabil* 2012; **34**: 965–70.
19. Jelsma J, Pronk M, Ferguson G, Jelsma-Smit D. The effect of the Nintendo Wii Fit on balance control and gross motor function of children with spastic hemiplegic cerebral palsy. *Dev Neurorehabil* 2012; **3**: 3.
20. Ryan SE. 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.
21. Ledebt A, Becher J, Kapper J, et al. Balance training with visual feedback in children with hemiplegic cerebral palsy: effect on stance and gait. *Mot Control* 2005; **9**: 459–68.
22. Woollacott M, Shumway-Cook A, Hutchinson S, et al. Effect of balance training on muscle activity used in recovery of stability in children with cerebral palsy: a pilot study. *Dev Med Child Neurol* 2005; **47**: 455–61.
23. Shumway-Cook A, Hutchinson S, Kartin D, Price R, Woollacott M. Effect of balance training on recovery of stability in children with cerebral palsy. *Dev Med Child Neurol* 2003; **45**: 591–602.
24. Franki I, Desloovere K, De Cat J, et al. The evidence-base for basic physical therapy techniques targeting lower limb function in children with cerebral palsy: a systematic review using the International Classification

of Functioning, Disability and Health as a conceptual framework. *J Rehabil Med* 2012; **44**: 385–95.

25. Harris SR, Roxborough L. Efficacy and effectiveness of physical therapy in enhancing postural control in children with cerebral palsy. *Neural Plast* 2005; **12**: 229–43.

26. O'Neil ME, Fragala-Pinkham MA, Westcott SL, et al. Physical therapy clinical management recommendations for children with cerebral palsy – spastic diplegia: achieving functional mobility outcomes. *Pediatr Phys Ther* 2006; **18**: 49–72.

27. Debuse D, Brace H. Outcome measures of activity for children with cerebral palsy: a systematic review. *Pediatr Phys Ther* 2011; **23**: 221–31.

28. Harvey A, Robin J, Morris ME, Graham HK, Baker R. A systematic review of measures of activity limitation for children with cerebral palsy. *Dev Med Child Neurol* 2008; **50**: 190–8.

29. Tyson SF, Connell LA. How to measure balance in clinical practice. A systematic review of the psychometrics and clinical utility of measures of balance activity for neurological conditions. *Clin Rehabil* 2009; **23**: 824–40.

30. Socpi Europe. A collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). *Dev Med Child Neurol* 2000; **42**: 816–24.

31. Law M, MacDermid J. Evidence-Based Rehabilitation (2nd edition). Thorofare: SLACK Incorporated, 2008.

32. 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.

33. Mokkink LB, Terwee CB, Knol DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: a clarification of its content. *BMC Med Res Methodol* 2010; **10**: 22.

34. Terwee CB, Mokkink LB, Knol DL, et al. Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. *Qual Life Res* 2012; **21**: 651–7.

35. Benfer KA, Weir KA, Boyd RN. Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: a systematic review. *Dev Med Child Neurol* 2012; **54**: 784–95.

36. Mokkink LB, Terwee CB, Patrick DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol* 2010; **63**: 737–45.

37. Mokkink LB, Terwee CB, Gibbons E, et al. Inter-rater agreement and reliability of the COSMIN (COnsensus-based Standards for the selection of health status Measurement Instruments) checklist. *BMC Med Res Methodol* 2010; **10**: 82.

38. Terwee CB, Bot SD, de Boer MR, et al. Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007; **60**: 34–42.

39. van Tulder M, Furlan A, Bombardier C, Bouter L. Updated method guidelines for systematic reviews in the Cochrane Collaboration Back Review Group. *Spine* 1976; **28**: 1290–9.

40. Schellingerhout JM, Verhagen AP, Heymans MW, et al. Measurement properties of disease-specific questionnaires in patients with neck pain: a systematic review. *Qual Life Res* 2012; **21**: 659–70.

41. Uijen AA, Heinst CW, Schellevis FG, et al. Measurement properties of questionnaires measuring continuity of care: a systematic review. *PLoS ONE* 2012; **7**: 31.

42. Pollock AS, Durward BR, Rowe PJ, Paul JP. What is balance? *Clin Rehabil* 2000; **14**: 402–6.

43. Pountney TE, Cheek L, Green E, Mulcahy C, Nelham R. Content and criterion validation of the Chailey Levels of Ability. *Physiotherapy* 1999; **85**: 410–16.

44. Gan SM, Tung LC, Tang YH, Wang CH. Psychometric properties of functional balance assessment in children with cerebral palsy. *Neurorehabil Neural Repair* 2008; **22**: 745–53.

45. Niznik TM, Turner D, Worrell TW. Functional Reach as a Measurement of Balance for Children with Lower Extremity Spasticity. *Phys Occup Ther Pediatr* 1996; **15**: 1–16.

46. Fife SE. Reliability of a measure to assess outcomes of adaptive seating in children with neuromotor disabilities. *Can J Rehabil* 1993; **7**: 11–3.

47. Field DA, Roxborough LA. Responsiveness of the Seated Postural Control Measure and the Level of Sitting Scale in children with neuromotor disorders. *Disabil Rehabil Assist Technol* 2011; **6**: 473–82.

48. Field DA, Roxborough LA. Validation of the relation between the type and amount of seating support provided and Level of Sitting Scale (LSS) scores for children with neuromotor disorders. *Dev Neurorehabil* 2012; **15**: 202–8.

49. Franjoine MR, Gunther JS, Taylor MJ. Pediatric balance scale: a modified version of the Berg Balance Scale for the school-age child with mild to moderate motor impairment. *Pediatr Phys Ther* 2003; **15**: 114–28.

50. Bartlett D, Birmingham T. Validity and reliability of a Pediatric Reach Test. *Pediatr Phys Ther* 2003; **15**: 84–92.

51. Butler PB, Saavedra S, Sofranac M, Jarvis SE, Woollacott MH. Refinement, reliability, and validity of the Segmental Assessment of Trunk Control. *Pediatr Phys Ther* 2010; **22**: 246–57.

52. Myhr U, Von Wendt L, Sandberg KW. Assessment of sitting in children with cerebral palsy from videofilm: a reliability study. *Phys Occup Ther Pediatr* 1993; **12**: 21–35.

53. Quinn A, O'Regan M, Horgan F. Psychometric evaluation of the Functional Walking Test for children with cerebral palsy. *Disabil Rehabil* 2011; **33**: 2397–403.

54. Reid DT, Schuller R, Billson N. Reliability of the Sitting Assessment for Children with Neuromotor Dysfunction (SACND). *Phys Occup Ther Pediatr* 1996; **16**: 23–32.

55. Heyrman L, Molenaers G, Desloovere K, et al. A clinical tool to measure trunk control in children with cerebral palsy: the Trunk Control Measurement Scale. *Res Dev Disabil* 2011; **32**: 2624–35.

56. Liao HF, Mao PJ, Hwang AW. Test–retest reliability of balance tests in children with cerebral palsy. *Dev Med Child Neurol* 2001; **43**: 180–6.

57. Williams EN, Carroll SG, Reddihough DS, Phillips BA, Galea MP. Investigation of the Timed 'Up & Go' Test in children. *Dev Med Child Neurol* 2005; **47**: 518–24.

58. Saether R, Jorgensen L. Intra- and inter-observer reliability of the Trunk Impairment Scale for children with cerebral palsy. *Res Dev Disabil* 2011; **32**: 727–39.

59. Bartlett D, Purdie B. Testing of the Spinal Alignment and Range of Motion Measure: a discriminative measure of posture and flexibility for children with cerebral palsy. *Dev Med Child Neurol* 2005; **47**: 739–43.

60. Ries LG, Michaelsen SM, Soares PS, Monteiro VC, Allegretti KM. Cross-cultural adaptation and reliability analysis of the Brazilian version of Pediatric Balance Scale (PBS). *Rev Bras Fisioter* 2012; **16**: 205–15.

61. Yi SH, Hwang JH, Kim SJ, Kwon JY. Validity of Pediatric Balance Scales in Children with Spastic Cerebral Palsy. *Neuropediatrics* 2012; **25**: 25.

62. Rodby-Bousquet E, Agustsson A, Jonsdottir G, et al. Interrater reliability and construct validity of the Posture and Postural Ability Scale in adults with cerebral palsy in supine, prone, sitting and standing positions. *Clin Rehabil* 2012; **28**: 28.

63. Jin-Gang H, Ji-Hea W, Jooyeon K. Reliability of the Pediatric Balance Scale in the Assessment of the Children with Cerebral Palsy. *J Phys Ther Sci* 2012; **24**: 301–5.

64. Sanjivani D, Prema K. Intra-rater reliability of Timed 'Up and Go' test for children diagnosed with cerebral palsy. *Int J Ther Rehabil* 2012; **19**: 575–80.

65. Zaino CA, Marchese VG, Westcott SL. Timed Up and Down Stairs Test: preliminary reliability and validity of a new measure of functional mobility. *Pediatr Phys Ther* 2004; **16**: 90–8.

66. Green EM, Mulcahy CM, Pountney TE. An investigation into the development of early postural control. *Dev Med Child Neurol* 1995; **37**: 437–48.

67. Kembhavi G, Darrah J, Magill-Evans J, Loomis J. Using the Berg Balance Scale to distinguish balance abilities in children with cerebral palsy. *Pediatr Phys Ther* 2002; **14**: 92–9.

68. Fife SE, Roxborough LA, Armstrong RW, et al. Development of a clinical measure of postural control for assessment of adaptive seating in children with neuromotor disabilities. *Phys Ther* 1991; **71**: 981–93.

69. Lowes LP. Evaluation of standing balance of children with cerebral palsy and the tools for assessment. Pennsylvania: Allegheny University of the Health Sciences, 1997.

70. Lowes LP, Westcott SL, Palisano RJ, Effgen SK, Orlin MN. Muscle force and range of motion as predictors of standing balance in children with cerebral palsy. *Phys Occup Ther Pediatr* 2004; **24**: 57–77.

71. Bertoti DB. Effect of therapeutic horseback riding on posture in children with cerebral palsy. *Phys Ther* 1988; **68**: 1505–12.

72. Butler PB. A preliminary report on the effectiveness of trunk targeting in achieving independent sitting balance in children with cerebral palsy. *Clin Rehabil* 1998; **12**: 281–93.

73. Myhr U, von Wendt L. Improvement of functional sitting position for children with cerebral palsy. *Dev Med Child Neurol* 1991; **33**: 246–56.

74. Myhr U, von Wendt L, Norrlin S, Radell U. Five-year follow-up of functional sitting position in children with cerebral palsy. *Dev Med Child Neurol* 1995; **37**: 587–96.

75. Reid DT. Development and preliminary validation of an instrument to assess quality of sitting of children with

neuromotor dysfunction. *Phys Occup Ther Pediatr* 1995; **15**: 53–82.

76. Liao HF, Hwang AW. Relations of balance function and gross motor ability for children with cerebral palsy. *Percept Mot Skills* 2003; **96**: 1173–84.

77. Shelley M. The Effects of Neurodevelopmental Treatment on Postural Control During Reaching in Children with Spastic Cerebral Palsy. Boston: Boston University, 1991.

78. de Vet H, Terwee C, Mokkink LB, Knol DL. Measurement in Medicine (1st edn). New York: Cambridge University Press, 2011.

79. Yelnik A, Bonan I. Clinical tools for assessing balance disorders. *Neurophysiol Clin* 2008; **38**: 439–45.

80. Horak FB, Wrisley DM, Frank J. The Balance Evaluation Systems Test (BESTest) to differentiate balance deficits. *Phys Ther* 2009; **89**: 484–98.

81. Franchignoni F, Horak F, Godi M, Nardone A, Giordano A. Using psychometric techniques to improve the Balance Evaluation Systems Test: the mini-BESTest. *J Rehabil Med* 2010; **42**: 323–31.

82. Portney LG, Watkins MP. Foundations of Clinical Research (3rd edn). New Jersey: Pearson Education, 2009.

Paper II

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Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy

Rannei Saether [a,b,*], Jorunn L. Helbostad [c,d], Lars Adde [a,d], Lone Jørgensen [e,f], Torstein Vik [a,b]

[a] Department of Laboratory Medicine, Children's and Women's Health, Faculty of Medicine, Norwegian University of Science and Technology, Trondheim, Norway
[b] Department of Pediatrics, St. Olavs Hospital, University Hospital of Trondheim, Trondheim, Norway
[c] Department of Neuroscience, Norwegian University of Science and Technology, Trondheim, Norway
[d] Clinic for Clinical Services, St. Olavs University Hospital, Trondheim, Norway
[e] Department of Health and Care Sciences and the "Tromsø Endocrine Research Group", University of Tromsø, Tromsø, Norway
[f] Department of Clinical Therapeutic Services, University hospital of Northern Norway, Tromsø, Norway

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ABSTRACT

Standardized clinical tools are useful for treatment planning and evaluation, however clinical tools to assess quality in trunk movements in children with cerebral palsy (CP) are sparse. We have recently reported good intra- and inter-observer reliability of the Trunk Impairment Scale (TIS) in 5–12 year old children with CP. The aim of this study was to assess reliability in adolescents (13–19 years old), and to assess the construct validity in children and adolescents in the whole age spectrum from 5 to 19 years. Video recordings of 17 children with CP with Gross Motor Function Classification (GMFCS) level I–IV were analyzed by three observers on two occasions. For construct validity the TIS was compared with Gross Motor Function Measure (GMFM), in 37 children with GMFCS levels I–IV. Intraclass correlation coefficients varied between 0.82 and 0.98, and 86% of the kappa values varied between 0.61 and 1.00, suggesting high inter- and intra-observer reliability. The smallest detectable difference (SDD) of the TIS (scale range 0–23) varied between 2.55 and 3.82 for intra- and 4.07–8.23 for inter-observer observations. The high inter-observer SDD was partly due to consistently lower TIS scores by one observer. The correlation between the TIS total score and the dimension scores of the GMFM was high (Spearman's rho: 0.80–0.87), while decreasing GMFCS levels were associated with increasing total TIS score; both findings indicating good construct validity of the TIS. This study suggests that the TIS is a reliable and valid measure of trunk control for both children and adolescents with cerebral palsy.

1. Introduction

One of the key features in the definition of cerebral palsy (CP) is impaired control of posture (Rosenbaum et al., 2007). Control of posture is required in order to obtain balance, which may be defined as the ability to maintain, achieve or restore the center of mass relative to the base of support (Mancini & Horak, 2010). During development, the postural control system

* Corresponding author at: Norwegian University of Science and Technology, Faculty of Medicine, Department of Laboratory Medicine, Children's and Women's Health, Postboks 8905, 7491 Trondheim, Norway. Tel.: +47 99248133.
E-mail addresses: rannei.sather@ntnu.no (R. Saether), jorunn.helbostad@ntnu.no (J.L. Helbostad), lars.adde@ntnu.no (L. Adde), lone.jorgensen@uit.no (L. Jørgensen), torstein.vik@ntnu.no (T. Vik).

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tries to achieve a stable vertical posture of the head and trunk against the force of gravity, to make a base for adequate sitting, reaching, standing, walking and feeding (Forssberg, 1999; Redstone & West, 2004). The control of the trunk movements plays a crucial role in these activities of daily life (Bertenthal & Von Hofsten, 1998; Kavanagh, Barrett, & Morrison, 2006; Ledebt & Bril, 2000; Patla, Adkin, & Ballard, 1999; Saavedra, Joshi, Woollacott, & van Donkelaar, 2009; Schmid, De Nunzio, & Schieppati, 2005; Van de Walle et al., 2012). Studies indicate that children with both mild and severe forms of CP have postural impairments (de Graaf-Peters et al., 2007; Heyrman et al., 2012), and many of the children with CP use sitting instead of standing position when performing tasks of daily life. Consequently, they spend more time in sitting than healthy children (Carlberg & Hadders-Algra, 2005). More knowledge about trunk control is of particularly importance when clinicians and researchers are planning and evaluating treatment in children with CP (Heyrman et al., 2012).

Despite its clinical importance, research on the specific characteristics of impaired trunk control in children with CP is rare. Trunk control in children with CP has been explored in studies of postural control (Heyrman et al., 2012). Such studies include assessment of the trunk as a single unit either in force platform studies (Ju, Hwang, & Cherng, 2012; Kyvelidou, Harbourne, Willett, & Stergiou, 2013; Liao, Yang, Hsu, Chan, & Wei, 2003), or in kinematic analysis (Coluccini, Maini, Martelloni, Sgandurra, & Cioni, 2007). In addition, muscle activation patterns of the trunk have been analyzed in electromyography studies (Bigongiari et al., 2011; Brogren, Forssberg, & Hadders-Algra, 2001; Hadders-Algra, van der Fits, Stremmelaar, & Touwen, 1999; Prosser, Lee, VanSant, Barbe, & Lauer, 2010; Roncesvalles, Woollacott, & Burtner, 2002). However, we are aware of only one study that have assessed trunk control in children with CP related to CP-subtype and to severity of gross motor impairment (Heyrman et al., 2012). In that study, Heyrman et al. speculated that the reason for lack of studies of trunk control might be due to the limited number of available clinical assessment tools (Heyrman et al., 2012).

Interventions proposed to improve trunk control in children with CP, include trunk targeted training (Butler, 1998), hippo therapy and horseback riding (Zadnikar & Kastrin, 2011), and adaptive seating (Ryan, 2012). However, due to lack of appropriate assessment tools (Ryan, 2012), and limited documentation of the measurement properties of existing tools, such studies should be interpreted with caution. Better documentation of existing assessment tools is therefore warranted (Saether, Helbostad, Riphagen, & Vik, 2013).

In a recent, systematic review of clinical balance tools in children and adults with CP (Saether et al., 2013), we found four clinical balance tools focusing on trunk control in children and adults with cerebral palsy. Among these tools the Sitting assessment for Children with Neuromotor Dysfunction (SACND) (Reid, Schuller, & Billson, 1996), the Trunk Control Measurement Scale (TCMS) (Heyrman et al., 2011), and the Trunk Impairment Scale (TIS) (Saether & Jorgensen, 2011) assess the quality of static and dynamic trunk control, while the Segmental Assessment of Trunk Control (SATCO) (Butler, Saavedra, Sofranac, Jarvis, & Woollacott, 2010) assesses the child's level of trunk control in one sitting position. The advantage of both the TIS and the TCMS compared with SACND is that both tools give more information about the dynamic trunk control, whereas compared with the TCMCS the TIS is less extensive and time-consuming.

In a previous study we found that the TIS had high intra- and inter-observer reliability in 5–12 year old children with CP (Saether & Jorgensen, 2011). The aim of the present study was to assess the construct validity of the TIS, and if the reliability of the tool would be equally good in adolescents with CP. We hypothesized that children with more severe gross motor impairments would have lower TIS scores, than those with less severe gross motor impairments, and that the TIS scores would be highly correlated ($\rho > 0.70$) with the Gross Motor Function Measure (GMFM) (Russell, Rosenbaum, & Lane, 2002).

2. Methods

2.1. Design

The present study is a reliability and validity study of the TIS including children and adolescents with CP with different gross motor function, according to the Gross Motor Function Classification System (GMFCS) levels. The TIS assessment was video recorded and the video scorings were used to assess intra- and inter-observer reliability. Construct validity of the TIS was evaluated by comparing the TIS total score with the different (GMFCS) levels (the ability to discriminate), as a classification of gross motor impairment, and the TIS total scores with the Gross Motor Function Measure (GMFM) (Russell et al., 2002). In this study, an expansion of our previous study of reliability in children 5–12 years old (Saether & Jorgensen, 2011), we have assessed reliability in adolescents 13–19 years old, and construct validity in the whole age group from 5 to 19 years.

2.2. Participants

Children, age 5–19 years old, able to sit on a bench without support, and to understand instructions were eligible for participation, and both children with CP, all subtypes within GMFCS level I–IV, as well as typical developing children were included. Children with no motor impairment were included in order to address the discriminative ability between children without apparent postural problems, and children with such problems (i.e. with CP). Children with CP were recruited from the neuro-orthopedic outpatient clinic at St. Olavs University Hospital (Trondheim, Norway), and children with no motor impairment were recruited from several mainstream schools. Exclusion criteria were surgical procedures, included botulinum toxin injections, performed during the preceding six months. Information about the diagnosis and classification of CP was provided by the physiotherapist responsible for the child's follow-up.

A total of 48 children were invited to participate. Two children did not meet the inclusion criteria (one could not sit without support and the other one could not understand instructions) and thus, the final study population comprised 46 participants; 37 with CP and nine children with typical development (controls). Among the 46 children, 25 had participated in our previous reliability study (Saether & Jorgensen, 2011), while in the present study, the reliability of the TIS in adolescents was assessed in 17 additionally recruited children with CP age 13–19 years old. However, construct validity was not addressed in our previous study, and all 46 children (5–19 years) were therefore included in the validity part of the present study.

2.3. Observers

Three physiotherapists, labeled A–C, were observers in the expanded reliability study, while observer B did the validity study. They were all working clinically with children with CP at St. Olavs University Hospital, and their clinical experience, were17 years for observer A, 20 years for observer B, and 0.5 years for observer C.

2.4. Assessment tools

2.4.1. TIS

The TIS is developed to evaluate trunk control in adult stroke patients, and assesses static and dynamic sitting balance and trunk coordination in a sitting position (Verheyden et al., 2004). *The subscale of static balance investigates*: (1) the ability of the subject to maintain a sitting position with feet supported; (2) the ability to maintain a sitting position while the legs are passively crossed, and (3) the ability to maintain a sitting position when the subject crosses the legs actively. In the present study, the children crossed their strongest leg over their weakest leg. *The subscale of dynamic balance investigates*: lateral flexion of the trunk and unilateral lifting of the hip. *The subscale of coordination investigates*: coordination of the trunk, the subject is asked to rotate the upper or lower part of his or her trunk 6 times, initiating the movements either from the shoulder girdle or from the pelvic girdle, respectively. For each item, a 2-, 3- or 4-point ordinal scale is used. Maximal scores for the subscales of static- and dynamic sitting balance and coordination are 7, 10 and 6 points, respectively. The total score for the TIS ranges between zero for a low performance to 23 for a high performance (Verheyden et al., 2006).

2.4.2. GMFM

Gross motor function was assessed by the GMFM (Russell et al., 2002), on the same day as the TIS assessment, by observer B. The GMFM consists of 88 items, divided into 5 dimensions: (A) lying and rolling, (B) sitting, (C) crawling and kneeling, (D) standing and (E) walking, running and jumping. GMFM was only performed in the children with CP. Percentages of maximum scores for dimension (B), (D), and (E) were calculated. Reliability and validity of the GMFM has earlier been found to be good in children with CP (Ko & Kim, 2012; Russell et al., 2000).

2.5. Assessment procedure

All children were assessed in a single session, by observer B, in the same physical environment at St. Olavs University Hospital. The children were sitting on a bench with support for their feet. The children were permitted to wear a tight shirt/no shirt, shorts/tights and regular footwear (orthoses, shoes), but were allowed to be barefoot if this was preferred. The test items were carried out in accordance with the test manual (Saether & Jorgensen, 2011). The test session was recorded using a video camera. The children were video recorded in the frontal plane for all of the tasks with the exception of static sitting balance, items 2 and 3 (therapist/child crosses the strongest leg over the weakest). These items were recorded in the sagittal plane. For item 3 (child crosses the strongest leg over the weakest) a red mark at 10 cm distance to the rear of the child's pelvis was placed on the bench, to make observations of trunk movement more than 10 cm backward easier.

The video recordings were edited by observer B, using the Pinnacle Studio 11.0. In accordance with the manual, each child had three attempts to complete a task, and the best attempt was selected (Saether & Jorgensen, 2011).

2.6. Training

To become familiar with the TIS and the criteria for definitions of the scorings the observers watched an instruction video provided by Verheyden (Verheyden, 2009) for 30 min. In addition, video clips of children, not included in the study, were scored collectively by the observers, in order to achieve a common understanding of the criteria for scoring. Both training sessions took about an hour.

2.7. Scoring

The scoring of the GMFM was completed by observer B, unaware of the TIS score, on the same day as the patient visited the hospital. The TIS scores of all participants were then done four weeks after the examination of the last participant. To study *inter-observer reliability*, the observers were sitting in the same room, independently assessing the 17 video recordings on a video-screen. Each assessment session started by observing a video-clip of a child with normal motor development who

was not included in the study. Video-clips of the included children included were then shown in random order. Each observer could watch a video-clip several times, but they were not allowed to see the scores of the other observers. For *intra-observer reliability*, the video-clips were rescored 4 weeks later by the same observers, following the same procedure.

2.8. Data analysis

All variables were examined for normality using the Kolmogorov–Smirnov test (Pallant, 2007). The TIS total score showed normal distribution, and parametric statistics were employed. *Relative reliability* was assessed by calculating an intraclass correlation coefficient (ICC). ICC (1,1) statistics were used because the observers were strategically chosen. In this model all within-subject variability is assumed to be measurement error. ICC (3,1) was also calculated, to study if there were any systematic shift between observers. In this model systematic shift is not considered part of measurement error. When no systematic error is present, ICC (1,1) is equal to ICC (3,1) (Shrout & Fleiss, 1979). According to Munro, reliability is considered to be high when the ICC is .70 or higher (Domholdt, 2005). The intra- (A1–A2, B1–B2, C1–C2) and inter-observer (A1–B1, A1–C1, B1–C1) reliability of total TIS score was assessed using ICC (1,1) and (3,1). For the items in the "static sitting balance-", "dynamic sitting balance-", and "coordination-subscale score", kappa and weighted kappa statistic (κ) were used to test agreement, or agreement was expressed as percent agreement if the κ value could not be calculated. Interpretations of results were done according to guidelines adapted from Landis and Koch (Landis & Koch, 1977), where a κ value of $<$0.20 is described as poor agreement, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 good, and 0.81–1.00 very good agreement.

To describe *absolute reliability* the standard error of measurement (SEM) was calculated as the square root of the mean within-subject variance. Low SEM expresses a small degree of measurement error (Bland & Altman, 1996). The difference between a child's score made by an observer and the true value would be expected to be less than 1.96 SEM for 95% of the observations (Bland & Altman, 1996). The difference between two measurement for the same subject, the smallest detectable difference (SDD), was then expected to be less than $\sqrt{2} \times 1.96 \times \text{SEM} = 2.77$ SEM for 95% of the pairs of observations (Bland & Altman, 1996).

The consistency of the measurements was also verified graphically using the Bland and Altman plots (Bland & Altman, 1986) for the total TIS score. This method plots differences between two observations against the average of the two observations, allowing scoring distribution, and possible measurement bias to be assessed visually (Bland & Altman, 1986).

Construct validity was evaluated by the Spearman rank correlation coefficient, comparing the TIS total and subscale scores, and the dimension B (sitting), dimension D (standing), and dimension E (walking, running and jumping) of the GMFM. The overall interpretation of the results of these correlation analyses were done in accordance with the criteria proposed by Terwee, whereby 75% of the results should be in correspondence with the hypothesis (Terwee et al., 2007).

The study was performed according to the Helsinki Declaration and approved by The Regional Committee for Medical Health Research Ethics. Written informed consent was obtained from the participants and their parents prior to participation in the study. The analyses were performed using SPSS (Statistical Package for Social Sciences, Chicago, IL, USA), version 19.

3. Results

3.1. The participants

The children's mean age was 11.6 (SD 3.6) years, ranging from 5 to 19 years. One of the participants turned 19 one week before the assessment. The distribution of sex, CP-subtypes, and GMFCS levels, are shown in Table 1.

3.2. Validity

3.2.1. Construct validity

The children scored between 4 and 23 points on the total TIS score. Decreasing GMFCS levels were associated with increasing total TIS scores, although the TIS scores for GMFCS level II and III did not differ (Table 2). The mean of the total TIS score for children with no motor impairment was 22.8 (SD 0.3) while it varied from 18.7 (SD 2.9) for children with CP and

Table 1
Characteristics of the children included in the study according to Gross Motor Function Classification System (GMFCS) levels.

Classification	Frequency	Mean age yrs/SD	Min-max	Spastic unilateral CP	Spastic bilateral CP	Dyskinetic CP	Sex	
				n	n	n	Male	Female
Children with no motor impairment	9	11.0 (4.03)	5–18	0	0	0	3	6
GMFCS, level 1	11	12.9 (3.11)	7–17	8	3	0	7	4
GMFCS, level 2	9	12.1 (3.30)	8–17	3	6	0	3	6
GMFCS, level 3	9	12.0 (5.15)	5–19	1	8	0	5	4
GMFCS, level 4	8	10.4 (2.13)	7–13	0	6	2	5	3
Total	46	11.6 (3.54)	5–19	12	23	2	23	23

Table 2
Total score of the Trunk Impairment Scale, observers A1, A2, B1, B2, C1, and C2, related to Gross Motor Functioning Classification System (GMFCS) levels in children aged 13–19 years.

	All observers			Children with no motor impairment			Children with cerebral palsy											
							GMFCS I			GMFCS II			GMFCS III			GMFCS IV		
Observer	Mean	SD	Total range	Mean	SD	Total range	Mean	SD	Total range	Mean	SD	Total range	Mean	SD	Total range	Mean	SD	Total range
A1	16.86	5.46	4.0–23.0	23.00	0.00	23.0–23.0	18.83	3.06	16.0–23.0	15.50	2.65	12.0–18.0	17.00	0.82	16.0–18.0	6.33	2.52	4.0–9.0
A2	16.71	5.40	4.0–23.0	23.00	0.00	23.0–23.0	18.50	3.15	16.0–23.0	16.50	1.73	14.0–18.0	15.75	1.50	14.0–17.0	6.33	2.52	4.0–9.0
B1	15.00	6.17	4.0–23.0	23.00	0.00	23.0–23.0	17.33	2.66	14.0–21.0	13.50	3.00	9.0–15.0	13.25	2.99	9.0–16.0	4.00	0.00	4.0–9.0
B2	15.67	5.70	4.0–23.0	23.00	0.00	23.0–23.0	18.50	3.15	16.0–23.0	14.00	2.00	11.0–15.0	13.75	2.63	10.0–16.0	5.67	2.89	4.0–9.0
C1	17.71	5.19	4.0–23.0	22.75	0.50	22.0–23.0	20.00	2.28	18.0–23.0	15.75	1.51	15.0–18.0	19.00	1.83	17.0–21.0	7.33	2.89	4.0–9.0
C2	16.95	5.27	4.0–23.0	22.50	1.00	21.0–23.0	19.33	3.01	15.0–23.0	16.25	1.89	15.0–19.0	16.00	2.70	12.0–18.0	7.00	2.65	4.0–9.0
All observers	16.48	5.53	4.0–23.0	22.88	0.25	21.0–23.0	18.74	2.89	14.0–23.0	15.21	2.13	9.0–19.0	15.79	2.08	9.0–21.0	6.11	2.08	4.0–9.0

Table 3
Spearman rank correlation coefficients between total and subscale scores of the Trunk Impairment Scale (TIS), and Gross Motor Function Measure (GMFM) dimension scores in children with CP aged 5–19 years.

TIS	GMFM		
	Dimension B	Dimension D	Dimension E
Total	0.80[a]	0.87[a]	0.84[a]
Static sitting balance	0.73[a]	0.72[a]	0.68[a]
Dynamic sitting balance	0.71[a]	0.82[a]	0.81[a]
Coordination	0.70[a]	0.62[a]	0.63[a]

Trunk Impairment Scale (TIS), Gross Motor Function Measure (GMFM).
Dimension B – sitting, Dimension D – standing, Dimension E – walking, running and jumping.
[a] P value < 0.01.

GMFCS level I to 6.1 (SD 2.1) for those with GMFCS level IV (Table 2). Mean total score for the children with spastic-bilateral and – unilateral CP were 11.32 (SD 5.6) and 16.11(SD 3.7) points. There were no missing values.

Spearman rank correlation coefficients between the TIS and the GMFM are summarized in Table 3. The TIS total score was highly and significantly correlated with the dimensions of the GMFM, with correlation coefficients varying between 0.80–0.87. The correlation between the TIS subscale scores and the dimensions of the GMFM varied between 0.62–0.82, the correlation with the static sitting balance subscale varied between 0.68–0.73, the dynamic sitting balance subscale between 0.71 and 0.82, and the coordination subscale between 0.62 and 0.70.

3.3. Reliability

3.3.1. Relative reliability total score and items

Intra- and inter-observer reliability of the total TIS score for the ICC (1,1) and ICC (3,1) varied between 0.96 and 0.98 and 0.82 and 0.97, respectively (Table 4). The ICC (3,1) values were higher than the ICC (1,1) values for the inter-observer reliability between observer A and B and between observer B and C. The TIS scores obtained by observer B were consistently lower than the scores obtained by observer A and C (Table 4).

Intra- and inter-observer agreement was also tested for individual test items with kappa statistics (Table 5). A total of 86% of all observations showed κ values between 0.61 and 1.00, suggesting good to very good agreement. The κ values for *intra-observer agreement* varied between 0.96 and 1.00 for the static sitting balance subscale, 0.31–1.00 for the dynamic sitting balance subscale, and 0.60–0.92 for the coordination subscale. The κ values for *inter-observer agreement* varied between 0.90 and 0.96 for the static sitting balance subscale, 0.15–0.90 for the dynamic sitting balance subscale, and 0.56–0.94 for the coordination subscale. Percentage agreement, for those items where a κ value could not be calculated, varied between 95 and 100%.

3.3.2. Absolute reliability, measurement error

The SEM values (Table 4) for the total TIS score varied between 0.92 and 1.38 for intra-observer reliability and between 1.47 and 2.97 for inter-observer reliability. The SDD varied between 2.55 and 3.82 for intra-observer reliability, and between 4.07 and 8.23 for inter-observer reliability.

3.4. Bland–Altman Plot

The Bland–Altman plots for intra- and inter-observer agreement of the total TIS score are shown in Fig. 1. The Bland–Altman plots demonstrated less consistent intra- and inter-observer agreement for the scores in the middle to upper range (12–20 points) of the TIS total score.

Table 4
Intra- and inter-observer reliability of the total score of the Trunk Impairment Scale (TIS) in children with CP aged 13–19 years.

Observer	TIS (total score 0–23)							
	Observation 1 mean (SD)	Observation 2 mean (SD)	ICC (1,1) (95% CI)	SEM	SDD	ICC (3,1) (95% CI)	SEM	SDD
A	16.86 (5.46)	16.71 (5.40)	0.98 (0.95–1.00)	0.92	2.55	0.98 (0.95–1.00)	0.92	2.55
B	15.00 (6.17)	15.67 (5.70)	0.98 (0.94–0.99)	1.08	2.99	0.98 (0.95–0.99)	1.08	2.99
C	17.71 (5.12)	16.95 (5.27)	0.96 (0.90–0.99)	1.38	3.82	0.97 (0.91–0.99)	1.38	3.82
A-B			0.91 (0.75–0.97)	2.17	6.01	0.96 (0.88–0.98)	2.17	6.01
A-C			0.96 (0.88–0.98)	1.47	4.07	0.97 (0.91–0.99)	1.47	4.07
B-C			0.82 (0.52–0.94)	2.97	8.23	0.94 (0.85–0.98)	2.97	8.23

CI, confidence interval; ICC, intraclass correlation coefficient; SDD, smallest detectable difference; SEM, standard error of measurement.

Table 5
Intra-observer and inter-observer agreement of the items of the Trunk Impairment Scale (TIS) in children with CP aged 13–19 years.

Item	Observer A1–A2 κ (*se* κ)	Observer B1–B2 κ (*se* κ)	Observer C1–C2 κ (*se* κ)	Observer A1–B1 κ (*se* κ)	Observer A1–C1 κ (*se* κ)	Observer B1–C1 κ (*se* κ)
Static sitting balance						
1. Keep sitting balance	100%	100%	100%	100%	100%	100%
2. Keep sitting balance with legs crossed	100%	95%	100%	95%	100%	95%
3. Keep sitting balance while crossing legs[a]	1.00 (0.00)	0.96 (.06)	1.00 (0.00)	0.90 (0.08)	0.90 (0.06)	0.96 (0.06)
Dynamic sitting balance						
Touch seat with elbow, most affected side (task achieved or not)	0.83 (0.02)	0.62 (0.24)	0.77 (0.22)	0.70 (0.19)	0.63 (0.23)	0.62 (0.24)
2. Touch seat with elbow, most affected side (trunk movement)	0.86 (0.14)	1.00 (0.00)	0.83 (0.16)	0.86 (0.14)	0.83 (0.16)	0.70 (0.20)
3. Touch seat with elbow (compensation strategies)[a]	0.72 (0.15)	0.70 (0.16)	0.53 (0.18)	0.61 (0.16)	0.72 (0.15)	0.35 (0.18)
4. Touch seat with elbow, less affected side (task achieved or not)	1.00 (0.00)	1.00 (0.00)	1.00 (0.00)	0.62 (0.24)	0.77 (0.22)	0.83 (0.16)
5. Touch seat with elbow, less affected side (trunk movement)	1.00 (0.00)	1.00 (0.00)	0.83 (0.16)	0.86 (0.14)	0.83 (0.16)	0.70 (0.20)
6. Touch seat with elbow, less affected side (compensation strategies)	0.79 (0.14)	0.62 (0.17)	0.70 (0.16)	0.47 (0.16)	0.90 (0.10)	0.40 (0.18)
7. Lift pelvis from seat, most affected side (task achieved or not)	1.00 (0.00)	0.77 (0.22)	0.64 (0.33)	0.77 (0.22)	0.45 (0.33)	0.32 (0.30)
8. Lift pelvis from seat, most affected side (compensation strategies)	0.63 (0.19)	0.72 (0.15)	0.69 (0.20)	0.35 (0.18)	0.48 (0.22)	0.17 (0.16)
9. Lift pelvis from seat, less affected side (task achieved or not)	0.65 (0.33)	0.69 (0.20)	1.00 (0.00)	0.62 (0.24)	0.64 (0.33)	0.35 (0.26)
10. Lift pelvis from seat, less affected side (compensation strategies)	0.63 (0.19)	0.90 (0.10)	0.31 (0.23)	0.15 (0.15)	0.70 (0.20)	0.19 (0.11)
Coordination						
1. Rotate shoulder girdle 6 times	0.83 (0.11)	0.92 (0.08)	0.74 (0.14)	0.76 (0.13)	0.65 (0.14)	0.75 (0.13)
2. Rotate shoulder girdle 6 times within 6 s	0.80 (0.14)	0.90 (0.09)	0.60 (0.18)	0.80 (0.13)	0.80 (0.14)	0.81 (0.13)
3. Rotate pelvic girdle 6 times[a]	0.79 (0.13)	0.92 (0.08)	0.81 (0.09)	0.94 (0.09)	0.63 (0.17)	0.56 (0.18)
4. Rotate pelvic girdle 6 times within 6 s	0.89 (0.11)	0.88 (0.12)	0.80 (0.13)	0.88 (0.12)	0.71 (0.16)	0.59 (0.17)

κ – kappa values, (*se* κ) – (standard error kappa).
[a] Weighted kappa.

4. Discussion

In this study we found high intra- and inter-observer reliability for the TIS in children 13–19 years of age. Consistent with our hypotheses, decreasing GMFCS levels were associated with increasing total TIS scores, and the total scores and the dynamic balance subscale scores of the TIS correlated highly with the GMFM scores.

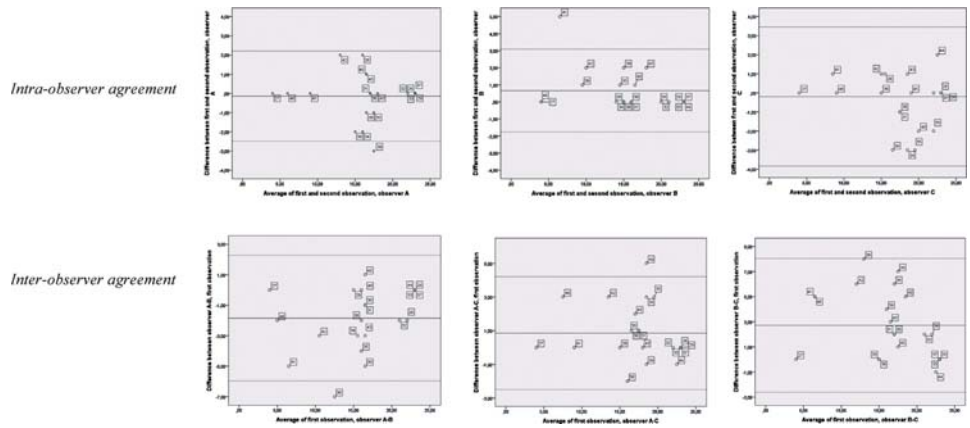

Fig. 1. Bland–Altman plots of the difference against the average of the total TIS score of 21 children, 17 with CP and 4 typical developing, aged 13–19 years, measured by the same observer (A1-A2, B1-B2, C1-C2) on two separate occasions and the two different observers (A1-B1, A1-C1, B1-C1) on the same occasion, with mean difference (solid line) and ±2SD (95% of agreement) (broken lines).

It may be considered a strength of the present study that we have adhered to the Consensus-based Standards for selection of Health Measurement Instruments (COSMIN) checklist (Mokkink et al., 2010). Even if the checklist was not published when our study was planned, we were able to interpret the results in accordance with these criteria. Furthermore, the included subjects with CP comprised a heterogeneous population which is important for the ability to generalize the results (de Vet, Terwee, Mokkink, & Knol, 2011). It may be considered a strength that we used video assessments when we examined the reliability, since this ensured that the variability of the scoring was unrelated to the child's performance or the instructions. A limitation of our study according to the COSMIN criteria could be that we included less than 30 participants in the reliability part of the study (Terwee et al., 2012). However, the number of participants mainly affects the precision of the estimates (Benfer, Weir, & Boyd, 2012), and since in our study the correlation coefficients, even taking into consideration the relatively broad confidence intervals, suggested high correlation (Domholdt, 2005), we consider the sample size of the present study to be sufficient to justify the conclusion that TIS has high intra- and inter-rater reliability (de Vet et al., 2011). Another limitation of our study may be that the physiotherapist who obtained the video recording of the TIS also participated in the video-based scoring. The consistently lower TIS scores obtained by this observer (B), compared with the two other observers (A and C) may, thus at least partly be due to the fact that observer B had instructed the children during the video-recording. Since inter-rater reliability was good between the two independent observers, this limitation most probably has led to an underestimation of the reliability of TIS in adolescents.

Compared to our previous reliability study of the TIS in children (Saether & Jorgensen, 2011), the mean TIS total score in adolescents was 2.8 points higher, and was observed within all GMFCS levels. This finding is consistent with other studies suggesting that the development of trunk control continues into adolescence (de Graaf-Peters et al., 2007; Mallau, Vaugoyeau, & Assaiante, 2010). The higher TIS scores in adolescents than in children (Saether & Jorgensen, 2011) may be due to the fact that children with CP spend more time in sitting than typical developing children, and that both typical developing and children with CP spend more time in sitting as they grow older (Maher, Williams, Olds, & Lane, 2007; OECD, 2010). In children with CP increasing time in sitting position with increasing age may in addition be due to a decline in gross motor function (GMFCS level III–V) from the age of six/seven (Hanna et al., 2009). Thus, this increased time in sitting with increasing age may lead to better postural control in the sitting position, and may explain the higher TIS scores obtained in adolescents than in younger children. The intra- and inter-observer ICC values were high in both our previous and present studies, supporting the high reliability for the age range 5–19 years, equal to the findings reported in stroke patients, and in subjects with multiple sclerosis, as reported by Verheyden et al. (2004, 2006). Like in our previous study (Saether & Jorgensen, 2011), there was a ceiling effect of TIS among children with no motor impairments (mean total score was 22.88; range 21–23 points). However, among children with CP there was no ceiling or floor effect.

Although the ICC values were high, the inter-observer SEM- and consequently the SDD values, varied between 4.1 and 8.2 points on the scale from 0 to 23 points. Systematic differences between the observers, as shown by the differences in the ICC (1,1) and (3,1) values (Table 4), may partly explain some of the high SDD values, and they were mainly due to lower scores by observer B (Table 4 and Fig. 1), the one conducting the TIS assessments. Between the observers A and C, who based their scoring solely on the videos, there was no systematic difference, and the inter-observer ICC values were as good as the intra-observer values. Although guidelines regarding acceptable SEM values are lacking, this large variation could be worrying, since it could indicate that only very large changes in trunk control may be identified by the TIS (de Vet et al., 2011). The systematic differences between observer B and the two other observers could also explain some of the low inter-observer kappa values shown in Table 5. Moreover the SEM value was higher for the least experienced observer (C), and lower for the most experienced observers (A and B) (Table 4). This finding is somewhat in contrast to our previous study, where experience did not seem to influence the scoring (Saether & Jorgensen, 2011).

Consistent with the recommendations proposed by the COSMIN group (Terwee et al., 2012), we formulated a specific hypothesis in order to examine the construct validity of the TIS. Our results suggested that the TIS has good construct validity, as the correlation coefficients between the TIS total score were high ($\rho > 0.70$).

Overall, the quality of our previous (Saether & Jorgensen, 2011) and present study and the results of the studies of the measurement properties (reliability and validity), suggest that the TIS can be used to assess trunk control in both children and adolescents with CP. Long experience as physiotherapist seems to be an advantage to achieve consistent measurements, and the scoring may differ if it is based upon video assessment or if it is based on a real examination. The TIS appeared to be fast (no more than 10 min) and easy to score. The newly developed TCMS is another reliable and valid tool to assess trunk control (Heyrman et al., 2011). However, the TCMS is a more comprehensive assessment of trunk control than the TIS, with a more narrow age range (8–15 years) and GMFCS level (I–III). For a discriminative purpose a less extensive assessment tool, like the TIS, might be as good as a more extensive and time consuming test. Moreover, future studies are needed to address the responsiveness, i.e. the ability to measure change, of both the TIS and the TCMS. Further assessments of the inter-observer reliability of the TIS may be warranted, including potential differences between live- and video-based scoring.

5. Conclusion

The TIS is a reliable and valid assessment tool of trunk control in children and adolescents 5–19 years. Further assessment of the test's ability to evaluate change is warranted.

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References

Benfer, K. A., Weir, K. A., & Boyd, R. N. (2012). Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: A systematic review. *Developmental Medicine and Child Neurology, 54*(9), 784–795.

Bertenthal, B., & Von Hofsten, C. (1998). Eye, head and trunk control: The foundation for manual development. *Neuroscience and Biobehavioral Reviews, 22*(4), 515–520.

Bigongiari, A., de Andrade e Souza, F., Franciulli, P. M., Neto Sel, R., Araujo, R. C., & Mochizuki, L. (2011). Anticipatory and compensatory postural adjustments in sitting in children with cerebral palsy. *Human Movement Science, 30*(3), 648–657.

Bland, J. M., & Altman, D. G. (1986). Statistical methods for assessing agreement between two methods of clinical measurement. *Lancet, 1*, 307–310.

Bland, J. M., & Altman, D. G. (1996). Statistics notes: Measurement error. *British Medical Journal, 313*, 744.

Brogren, E., Forssberg, H., & Hadders-Algra, M. (2001). Influence of two different sitting positions on postural adjustments in children with spastic diplegia. *Developmental Medicine and Child Neurology, 43*(8), 534–546.

Butler, P. B. (1998). A preliminary report on the effectiveness of trunk targeting in achieving independent sitting balance in children with cerebral palsy. *Clinical Rehabilitation, 12*(4), 281–293.

Butler, P. B., Saavedra, S., Sofranac, M., Jarvis, S. E., & Woollacott, M. H. (2010). Refinement, reliability, and validity of the Segmental Assessment of Trunk control. *Pediatric Physical Therapy, 22*(3), 246–257.

Carlberg, E. B., & Hadders-Algra, M. (2005). Postural dysfunction in children with cerebral palsy: Some implications for therapeutic guidance. *Neural Plasticity, 12*(2-3), 221–228.

Coluccini, M., Maini, E. S., Martelloni, C., Sgandurra, G., & Cioni, G. (2007). Kinematic characterization of functional reach to grasp in normal and in motor disabled children. *Gait and Posture, 25*(4), 493–501.

de Graaf-Peters, V. B., Blauw-Hospers, C. H., Dirks, T., Bakker, H., Bos, A. F., & Hadders-Algra, M. (2007). Development of postural control in typically developing children and children with cerebral palsy: Possibilities for intervention? *Neuroscience and Biobehavioral Reviews, 31*(8), 1191–1200.

de Vet, H., Terwee, C., Mokkink, L. B., & Knol, D. L. (2011). *Measurement in medicine* (1st ed.). New York: Camebridge University Press.

Domholdt, E. (2005). *Rehabilitation research: Principles and applications* (3rd ed.). St. Louis, MO Elsevier Saunders.

Forssberg, H. (1999). Neural control of human motor development. *Current Opinion in Neurobiology, 9*(6), 676–682.

Hadders-Algra, M., van der Fits, I. B., Stremmelaar, E. F., & Touwen, B. C. (1999). Development of postural adjustments during reaching in infants with CP. *Developmental Medicine and Child Neurology, 41*(11), 766–776.

Hanna, S. E., Rosenbaum, P. L., Bartlett, D. J., Palisano, R. J., Walter, S. D., Avery, L., et al. (2009). Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Developmental Medicine and Child Neurology, 51*(4), 295–302.

Heyrman, L., Desloovere, K., Molenaers, G., Verheyden, G., Klingels, K., Monbaliu, E., et al. (2012). Clinical characteristics of impaired trunk control in children with spastic cerebral palsy. *Research in Developmental Disabilities, 34*(1), 327–334.

Heyrman, L., Molenaers, G., Desloovere, K., Verheyden, G., De Cat, J., Monbaliu, E., et al. (2011). A clinical tool to measure trunk control in children with cerebral palsy: The Trunk Control Measurement Scale. *Research in Developmental Disabilities, 32*(6), 2624–2635.

Ju, Y. H., Hwang, I. S., & Cherng, R. J. (2012). Postural adjustment of children with spastic diplegic cerebral palsy during seated hand reaching in different directions. *Archives of Physical Medicine and Rehabilitation, 20*, 20.

Kavanagh, J., Barrett, R., & Morrison, S. (2006). The role of the neck and trunk in facilitating head stability during walking. *Experimental Brain Research, 172*(4), 454–463.

Ko, J., & Kim, M. (2012). Reliability and responsiveness of the Gross Motor Function Measure-88 in children with cerebral palsy. *Physical Therapy, 8*, 8.

Kyvelidou, A., Harbourne, R. T., Willett, S. L., & Stergiou, N. (2013). Sitting postural control in infants with typical development, motor delay, or cerebral palsy. *Pediatric Physical Therapy, 25*(1), 46–51.

Landis, J. R., & Koch, G. G. (1977). The measurement of observer agreement for categorical data. *Biometrics, 33*(1), 159–174.

Ledebt, A., & Bril, B. (2000). Acquisition of upper body stability during walking in toddlers. *Developmental Psychobiology, 36*(4), 311–324.

Liao, S. F., Yang, T. F., Hsu, T. C., Chan, R. C., & Wei, T. S. (2003). Differences in seated postural control in children with spastic cerebral palsy and children who are typically developing. *American Journal of Physical Medicine and Rehabilitation, 82*(8), 622–626.

Maher, C. A., Williams, M. T., Olds, T., & Lane, A. E. (2007). Physical and sedentary activity in adolescents with cerebral palsy. *Developmental Medicine and Child Neurology, 49*(6), 450–457.

Mallau, S., Vaugoyeau, M., & Assaiante, C. (2010). Postural strategies and sensory integration: No turning point between childhood and adolescence. *PLoS ONE5(9)*.

Mancini, M., & Horak, F. B. (2010). The relevance of clinical balance assessment tools to differentiate balance deficits. *European Journal of Physical Rehabilitation Medicine, 46*(2), 239–248, (Review).

Mokkink, L. B., Terwee, C. B., Knol, D. L., Stratford, P. W., Alonso, J., Patrick, D. L., et al. (2010). The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: A clarification of its content. *BMC Medical Research Methodology, 10*, 22.

OECD. (2010). *Health at a glance: Europe 2010* (No. ISBN 978-92-64-09031-6).

Pallant, J. (2007). *SPSS survival manual. A step-by-step guide to data analysis* (3rd ed.). New York: Mc Graw Hill Open University Press.

Patla, A. E., Adkin, A., & Ballard, T. (1999). Online steering: Coordination and control of body center of mass, head and body reorientation. *Experimental Brain Research, 129*(4), 629–634.

Prosser, L. A., Lee, S. C., VanSant, A. F., Barbe, M. F., & Lauer, R. T. (2010). Trunk and hip muscle activation patterns are different during walking in young children with and without cerebral palsy. *Physical Therapy, 90*(7), 986–997.

Redstone, F., & West, J. F. (2004). The importance of postural control for feeding. *Pediatric Nursing, 30*(2), 97–100.

Reid, D. T., Schuller, R., & Billson, N. (1996). Reliability of the Sitting Assessment for Children with Neuromotor Dysfunction (SACND). *Physical & Occupational Therapy in Pediatrics, 16*(3), 23–32.

Roncesvalles, M. N., Woollacott, M. W., & Burtner, P. A. (2002). Neural factors underlying reduced postural adaptability in children with cerebral palsy. *Neuroreport, 13*(18), 2407–2410.

Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., et al. (2007). A report: The definition and classification of cerebral palsy April 2006. *Developmental Medicine and Child Neurology. Supplement, 109*, 8–14.

Russell, D. J., Avery, L. M., Rosenbaum, P. L., Raina, P. S., Walter, S. D., & Palisano, R. J. (2000). Improved scaling of the Gross Motor Function Measure for children with cerebral palsy: Evidence of reliability and validity. *Physical Therapy, 80*(9), 873–885.

Russell, D. J., Rosenbaum, P. L., & Lane, L. M. (2002). *Gross Motor Function Measure (GMFM-66& GMFM-88) User's manual*. London, England: Mac Keith Press.

Ryan, S. E. (2012). An overview of systematic reviews of adaptive seating interventions for children with cerebral palsy: Where do we go from here? *Disability and Rehabilitation Assistive Technology, 7*(2), 104–111.

Saavedra, S., Joshi, A., Woollacott, M., & van Donkelaar, P. (2009). Eye hand coordination in children with cerebral palsy. *Experimental Brain Research, 192*(2), 155–165.

Saether, R., Helbostad, J. L., Riphagen, I., & Vik, T. (2013). Clinical tools to assess balance in children and adults with cerebral palsy: A systematic review. Developmental Medicine & Child Neurology, doi:10.111/dmcn12162, in press.

Saether, R., & Jorgensen, L. (2011). Intra- and inter-observer reliability of the Trunk Impairment Scale for children with cerebral palsy. *Research in Developmental Disabilities, 32*(2), 727–739.
Schmid, M., De Nunzio, A. M., & Schieppati, M. (2005). Trunk muscle proprioceptive input assists steering of locomotion. *Neuroscience Letters, 384*(1-2), 127–132.
Shrout, P. E., & Fleiss, J. L. (1979). Intraclass correlations: Uses in assessing rater reliability. *Psychological Bulletein, 86*(2), 420–428.
Terwee, C. B., Bot, S. D., de Boer, M. R., van der Windt, D. A., Knol, D. L., Dekker, J., et al. (2007). Quality criteria were proposed for measurement properties of health status questionnaires. *Journal of Clinical Epidemiology, 60*(1), 34–42.
Terwee, C. B., Mokkink, L. B., Knol, D. L., Ostelo, R. W., Bouter, L. M., & de Vet, H. C. (2012). Rating the methodological quality in systematic reviews of studies on measurement properties: A scoring system for the COSMIN checklist. *Quality of Life Research, 21*(4), 651–657.
Van de Walle, P., Hallemans, A., Truijen, S., Gosselink, R., Heyrman, L., Molenaers, G., et al. (2012). Increased mechanical cost of walking in children with diplegia: The role of the passenger unit cannot be neglected. *Research in Developmental Disabilities, 33*(6), 1996–2003.
Verheyden, G. (Writer). (2009). Instruction video of theTrunk Impairment Scale. Belgium: Verheyden, G.
Verheyden, G., Nieuwboer, A., Mertin, J., Preger, R., Kiekens, C., & De Weerdt, W. (2004). The Trunk Impairment Scale: A new tool to measure motor impairment of the trunk after stroke. *Clinical Rehabilitation, 18*(3), 326–334.
Verheyden, G., Nuyens, G., Nieuwboer, A., Van Asch, P., Ketelaer, P., & De Weerdt, W. (2006). Reliability and validity of trunk assessment for people with multiple sclerosis. *Physical Therapy, 86*(1), 66–76.
Zadnikar, M., & Kastrin, A. (2011). Effects of hippotherapy and therapeutic horseback riding on postural control or balance in children with cerebral palsy: A meta-analysis. *Developmental Medicine and Child Neurology, 53*(8), 684–691.

Paper III

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Gait characteristics in children and adolescents with cerebral palsy assessed with a trunk-worn accelerometer

Rannei Saether [a,b,*], Jorunn L. Helbostad [c,d], Lars Adde [a,d], Siri Brændvik [d,e], Stian Lydersen [b,f], Torstein Vik [a,b]

[a] Department of Laboratory Medicine, Children's and Women's Health, Norwegian University of Science and Technology, Trondheim, Norway
[b] Department of Pediatrics, St. Olavs Hospital, Trondheim University Hospital, Trondheim, Trondheim, Norway
[c] Department of Neuroscience, Norwegian University of Science and Technology, Trondheim, Norway
[d] Clinic for Clinical Services, St. Olavs Hospital, Trondheim University Hospital, Trondheim, Norway
[e] Human Movement Science Programme, Norwegian University of Science and Technology, Trondheim, Norway
[f] Regional Centre for Child and Youth Mental Health and Child Welfare – Central Norway, Norwegian University of Science and Technology, Trondheim, Norway

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ABSTRACT

This study aimed to investigate gait characteristics reflecting balance and progression in children and adolescents with cerebral palsy (CP) compared with typically developing (TD) children. Gait characteristics variables representing aspects of *balance* were trunk acceleration, interstride regularity and asymmetry of accelerations while gait characteristics representing *progression* were gait speed, cadence, step time and step length. Children in the age range 5–18 years (mean age 11.1 years) with spastic CP ($n = 41$) and a gross motor function corresponding to GMFCS I–III and children with TD ($n = 29$) were included. The children walked back and forth along a 5 m pathway with a tri-axial accelerometer worn on the lower back to allow assessment of their gait characteristics. Data were recorded along the anterioposterior (AP), mediolateral (ML), and vertical (*V*) axes. To assess the magnitude of potential differences in gait characteristics, standard deviation scores were calculated, using TD children as reference. Gait parameters related to balance, such as AP, ML, and *V* accelerations, were higher in the children with CP (*z*-scores between 0.4 and 0.7) and increased with increasing GMFCS levels. The differences in accelerations in the AP and *V* directions increased between children with CP and TD children with increasing speed. Also asymmetry in trunk accelerations differed significantly between the two groups in all three directions (*z*-scores between 0.8 and 1.8 higher in the CP group), while interstride regularity differed only slightly between children with CP and TD children, and only in the AP direction. Gait characteristics also differed between children with the spastic subtypes unilateral and bilateral CP, for accelerations and asymmetry in the AP and ML directions. Our results showed significant differences in gait characteristics between children with CP and TD children. The differences may be more related to balance than progression, and these problems seem to rise with increasing gross motor impairment and speed.

* Corresponding author at: Department of Laboratory Medicine, Children's and Women's Health, Faculty of Medicine, Norwegian University of Science and Technology, Post Box 8905, NO-7491 Trondheim, Norway. Tel.: +47 99248133.
E-mail addresses: rannei.sather@ntnu.no (R. Saether), jorunn.helbostad@ntnu.no (J.L. Helbostad), lars.adde@ntnu.no (L. Adde), siri.merete.brandvik@svt.ntnu.no (S. Brændvik), stian.lydersen@ntnu.no (S. Lydersen), torstein.vik@ntnu.no (T. Vik).

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1. Introduction

In children and adolescents with cerebral palsy (CP), gait is usually assessed in the decision processes leading to orthopedic surgery, botulinum toxin injections, and/or prescription orthoses. The main focus of such assessments has been the movements of the lower limbs (Dobson, Morris, Baker, & Graham, 2007). Several, but not all studies have reported that children with CP walk slower and with shorter steps compared with children with typical development (TD) (Abel & Damiano, 1996; Bourgeois, Mariani, Aminian, Zambelli, & Newman, 2014; Iosa, Marro, Paolucci, & Morelli, 2012; Norlin & Odenrick, 1986). These components of gait may be described as *progression*. Another important component of gait is *balance*. Balance during gait is particularly challenging since the body's center of mass (COM) is located outside the base of support in 80% of the gait cycle (Winter, 1995). Despite the high prevalence of balance problems in children with CP (de Graaf-Peters et al., 2007), relatively few studies have focused on balance during gait in this population, and even fewer have addressed the relationship between progression and balance (Bruijn et al., 2013).

Recently, some authors have studied balance during gait using optoelectronic and force plate motion analysis. A variety of parameters, such as kinematics of the COM, the trunk, the upper limbs and head, kinetics of the center of pressure (COP), Floquet analysis, and a foot placement estimator have been used to quantify balance. Several authors have reported displacement, velocity, accelerations, and variability in the anterioposterior (AP), mediolateral (ML) and vertical (*V*) directions of COM and COP in children with CP compared to TD children. More specifically, in children with CP, COM (displacement, velocity, and accelerations) was found to have increased in the AP, ML, and *V* directions (Cherng, Chou, Su, Shaughnessy, & Kaufman, 2007; Hsue, Miller, & Su, 2009a; Hsue, Miller, & Su, 2009b; Massaad, Dierick, van den Hecke, & Detrembleur, 2004), and COP (displacement, velocity and accelerations) in the ML direction (Feng, Pierce, Do, & Aiona, 2014; Hsue, Miller, & Su, 2009a; Hsue, Miller, & Su, 2009b). Moreover, kinematic analysis of the trunk showed increased forward tilt and increased range of motion in children with CP (Heyrman, Feys et al., 2013; Romkes et al., 2007). The kinematic analysis of the upper limbs showed decreased arm swing on the least affected side and the opposite on the most affected side (Bruijn, Meyns, Jonkers, Kaat, & Duysens, 2011), and a "guard position" with increased shoulder abduction and elbow flexion in children with CP (Meyns et al., 2012; Romkes et al., 2007). Kinematic analysis of the head showed greater variability of the head angle in the ML direction in children with CP (Wallard, Bril, Dietrich, Kerlirzin, & Bredin, 2012). Floquet analysis revealed that children with CP appear to utilize a wider step width and to modulate their step length (Kurz, Arpin, & Corr, 2012). Moreover, an assessment using a foot placement estimator showed marked instability in the AP and ML directions (Bruijn et al., 2013).

Trunk-worn accelerometers may provide an alternative approach to assessing the gait characteristics of both progression and balance (Kavanagh & Menz, 2008). The method is less time-consuming, less expensive, and not restricted to assessments conducted in a laboratory environment. The method is well established in assessments of adults with CP (Kavanagh & Menz, 2008), whereas we have identified only two studies using a trunk-worn accelerometer to assess gait in young children with CP (Iosa et al., 2012; Iosa, Morelli, Marro, Paolucci, & Fusco, 2013). The two latter studies, which were conducted by the same research group, found that children with CP had higher accelerations of COM, indicating impaired balance during gait, compared with TD children (Iosa et al., 2012, 2013). The authors reported that the children with CP were able to walk at speeds comparable to those of the TD children, but had higher trunk instability. However, the results regarding progression are inconsistent across studies, and few gait studies include both children and adolescents with CP, and to date no study including different CP subtypes has addressed both progression and balance.

The aim of our study was therefore to investigate, with the use of a trunk-worn accelerometer, the gait characteristics of children and adolescents (hereafter referred to as children) with CP compared with those with TD.

2. Materials and methods

2.1. Study design and subjects

In this cross-sectional study, gait was assessed with the use of a trunk-worn accelerometer. A consecutive sample of 70 children was included: 41 children with spastic CP (24 males) recruited from the neuro-orthopedic outpatient clinic at St. Olavs Hospital, Trondheim University Hospital (Trondheim, Norway), and 29 children (13 males) with no motor impairment were recruited from several public schools. To qualify for inclusion, participants had to be able to understand certain instructions and to walk at least 10 m without support, shoes, or orthoses. Exclusion criteria were treatment with botulinum toxin in the lower extremities during the preceding four months and/or surgery during the preceding 12 months. The characteristics of the included children are summarized in Table 1.

2.2. Instrumentation

In order to measure linear acceleration, a six degrees-of-freedom inertial sensor (MTx. XSens, Enschede, NL) (weight: 15 g) was attached over the L3 region of the participant's lower back. The sensor contains tri-axial units of accelerometers, gyroscopes, and magnetometers and is connected to a battery-operated communication unit (weight: 300 g), also worn by the participant. Data were acquired at a sampling frequency of 100 Hz and transmitted in real time to a laptop by Bluetooth technology (Aaslund, Helbostad, & Moe-Nilssen, 2011). Gait time was registered by photoelectric cells synchronized with the

Table 1
Characteristics of children with CP and TD children participating in the present study.

All children							Children with CP					
	All		TD		CP		GMFCS 1		GMFCS II		GMFCS III	
N	70		29		41		19		16		6	
Unilateral (*n*)	–	–	–	–	27	–	19	–	8	–	–	–
Bilateral (*n*)	–	–	–	–	14	–	–	–	8	–	6	–
Male gender, *n* (%)	37 (53)	–	13 (45)	–	24 (58)	–	10 (52)	–	10 (63)	–	4 (67)	–
Age (years), mean (SD)	11.1	(3.8)	10.3	(3.6)	11.7	(3.8)	11.4	(3.8)	11.8	(3.9)	13.1	(4.3)
Height (cm), mean (SD)	145.2	(21.7)	143.9	(22.6)	146.1	(21.3)	147.7	(24.9)	143	(17.6)	148.8	(20.5)
Weight (kg), mean (SD)	41.8	(18.9)	38.3	(15.5)	44.3	(21.3)	45.6	(25.1)	41.8	(17.1)	46.8	(16.8)
Body mass index (kg/m^2), mean (SD)	18.8	(4.0)	17.7	(2.5)	19.6	(4.7)	19.2	(4.8)	19.7	(5.3)	20.4	(2.9)

accelerometer device. Trunk-worn accelerometers have previously been tested for precision and accuracy (Moe-Nilssen, 1998b), test–retest reliability of accelerations (Henriksen, Lund, Moe-Nilssen, Bliddal, & Danneskiod-Samsoe, 2004; Kavanagh, Morrison, James, & Barrett, 2006), spatiotemporal parameters (Henriksen et al., 2004), and variability measures (Moe-Nilssen, Aaslund, Hodt-Billington, & Helbostad, 2010) during gait in healthy adults.

Information about motor functioning, described by the Gross Motor Functioning Classification System (GMFCS) for cerebral palsy (Palisano et al., 1997), and the classification of CP subtypes was obtained from each participating child's medical records.

The participants' height was measured to the nearest cm with a stadiometer, and their weight when wearing light clothing was measured to the nearest 0.5 kg using an electronic weight (Seca digital 770). Their body mass index (BMI) was then calculated as weight (kg)/height^2 (m^2).

2.3. Test procedure

The participants were instructed to walk barefoot back and forth along a 5 m pathway. To obtain data during steady state gait, the participants were instructed to start walking 2 m before reaching the photoelectric cells and to continue to walk 2 m beyond the photoelectric cells. The procedure was employed using three different instructions on gait speed, always followed by "go back and forth once": (1) "Walk normally, as you usually do," (2) "Walk as fast as you can, without running," and (3) "Walk slowly, as if you were strolling around." Thus, a series of six gait sequences, two sequences at each gait speed, was completed by each participant after two sequences used as habituation.

2.4. Data processing and variables

Gait characteristics variables representing aspects of *balance* were trunk acceleration, interstride regularity and asymmetry of accelerations in AP, ML and *V* directions, while gait characteristics representing *progression* were gait speed, cadence, step time and step length.

A customized software application TRASK, run under Matlab R2011b (Math Works Inc., Natrick, MA) was used for signal processing and calculation of gait variables. By applying the accelerometer as an inclinometer, the average tilt of the measuring axes could be calculated for each sample, thus eliminating gravity bias. The raw acceleration data were transformed into a horizontal–vertical coordinate system by a trigonometric algorithm and reported along anterioposterior (AP), mediolateral (ML), and vertical (*V*) axes (Moe-Nilssen, 1998a). Trunk acceleration amplitudes for each gait interval were expressed by root mean square (RMS) values (hereafter referred to as trunk accelerations). The periodicity of the acceleration curve enabled steps and strides to be registered. Interstep and interstride trunk acceleration regularity were calculated using an unbiased autocorrelation procedure, where an acceleration time series was correlated to the same series at a phase shift equivalent to one step and one stride, respectively. Perfect replication of the signals between consecutive steps or strides gives an autocorrelation coefficient of one (Moe-Nilssen & Helbostad, 2004). Trunk asymmetry was calculated for the AP, ML, and *V* axes separately by subtracting the interstep regularity from interstride regularity, reflecting differences in regularity between left and right steps beyond the regularity between strides. A perfect symmetry has the value of zero, whereas a positive value indicates an asymmetric gait pattern (Hodt-Billington, Helbostad, Vervaat, Rognsvag, & Moe-Nilssen, 2011). Step time and cadence for each walk was estimated from the autocorrelation curve of the vertical axis. Mean step length for each walk was calculated as gait distance/number of steps. The mean value of two walks (back and forth the pathway) at preferred speed, were used to compare gait variables between children with CP and children with TD. In this study sample a mean of 9.6 steps (SD 1.9) were used in the calculations.

2.5. Statistical analysis

Gait parameters for the CP and the TD group were compared using a three-step procedure: First unadjusted, using Student's *t*-test; second, adjusted for the potential confounders gender, age, height, and BMI, one at a time, in linear

regression, and with only confounders changing the coefficient for group more than 10% included in the model from this step; and third, since many gait variables have been shown to be associated with gait speed (Schwartz, Rozumalski, & Trost, 2008), we assessed the role of gait speed as a possible mediator by adding it as a covariate. The results from the unadjusted analysis were also calculated in terms of standard deviation scores ("*z*-scores") of each gait variable, using the mean value and SD in the control group as reference.

Differences between the CP and the TD group in gait strategy with increasing gait speed were assessed using a linear mixed model with acceleration at each of three velocities (slow, preferred, and fast gait) as a dependent variable, group, and measured velocity, and their interaction as fixed effects, and subject as random effect.

The TD children were also compared with children with different GMFCS levels with respect to the differences in gait parameters in two steps: first by One-Way Analysis of Variance (ANOVA); and second, if the ANOVA indicated a statistically significant difference between the groups, a Scheffe's post hoc test was applied to localize the difference. The same procedure was used to compare TD children with the two CP subtypes (unilateral and bilateral). Finally, the two CP subtypes were compared with respect to the differences in gait parameters using the Student's *t*-test.

Two-sided *p*-values < 0.05 were considered significant, and 95% confidence intervals (CI) are reported where relevant. Statistical analyses were performed using SPSS 19.

2.6. Ethics

All participants provided informed consent. The study was conducted in conformity with the Declaration of Helsinki, and was approved by Regional Committee for Medical Research Ethics in Central Norway.

3. Results

3.1. Gait characteristics of children with CP compared with TD children

Table 2 shows that there were no significant differences in the progression parameters mean preferred gait speed, cadence, mean step length and mean step time between children with CP and TD children.

However, regarding balance, children in the CP group had significantly higher mean accelerations and more asymmetry in all directions (AP, ML, and *V*) than the TD children, while stride regularity was significantly lower, but only in the AP direction (Table 2). Fig. 1 shows the size of the difference in *z*-scores for each of the gait parameters between TD children (mean *z*-score = 0) and the children with CP.

Among the potential confounders, only age was found to affect the difference in stride regularity and only in the AP direction between children with CP and the TD children (−0.10 (CI −0.17 to −0.04); $p \leq 0.002$). None of the other gait parameters were influenced by the potential confounders.

When gait speed as a potential mediator was included in the analyses, the differences in acceleration in all three directions between children with CP and TD children persisted. The mean difference in the AP direction was 0.47 (CI: 0.19–0.74), in the ML direction 0.67 (CI: 0.32–1.01), and in the *V* direction 0.99 (CI: 0.60–1.39); all $p \leq 0.001$). In addition the difference in the AP direction of stride persisted after adjusting for age and gait speed (mean difference: −0.10 (CI: −0.16 to −0.03); $p = 0.002$).

Table 3 shows larger increases in AP and *V* but not ML accelerations with increasing gait speed among children with CP compared with TD children.

Table 2
Gait characteristics among 41 children with cerebral palsy and 29 typical developing children.

Gait parameters	CP		TD		Mean diff	95% CI	*p*
	Mean	SD	Mean	SD			
Gait speed (m/s)	1.06	0.18	1.12	0.21	−0.06	−0.16 to 0.04	0.249
Cadence (steps/min)	120.73	19.58	120.80	15.10	−0.07	−8.66 to 8.52	0.987
Step length (m)	0.54	0.10	0.56	0.10	−0.02	−0.07 to 0.03	0.399
Step time (mean)	0.57	0.14	0.55	0.10	−0.02	−0.04 to 0.08	0.456
AP acceleration (RMS)	2.25	0.66	1.85	0.51	0.39	0.10 to 0.69	**0.009**
ML acceleration (RMS)	2.45	0.81	1.87	0.70	0.58	0.21 to 0.95	**0.003**
V acceleration (RMS)	3.05	1.02	2.18	0.71	0.88	0.44 to 1.31	**0.001**
AP regularity stride (ACORR)	0.62	0.14	0.70	0.14	−0.08	−0.15 to −0.02	**0.015**
ML regularity stride (ACORR)	0.51	0.19	0.47	0.19	0.04	−0.05 to 0.13	0.409
V regularity stride (ACORR)	0.71	0.14	0.66	0.16	0.05	−0.03 to 0.12	0.210
AP asymmetry (ACORR)	0.26	0.18	0.01	0.06	0.24	0.17 to 0.31	**0.001**
ML asymmetry (ACORR)	0.30	0.19	0.02	0.11	0.28	0.20 to 0.35	**0.001**
V asymmetry (ACORR)	0.24	0.20	0.02	0.08	0.22	0.14 to 0.30	**0.001**

AP, anteriorposterior; ML, mediolateral; *V*, vertical; RMS, root mean square; ACORR, autocorrelation.

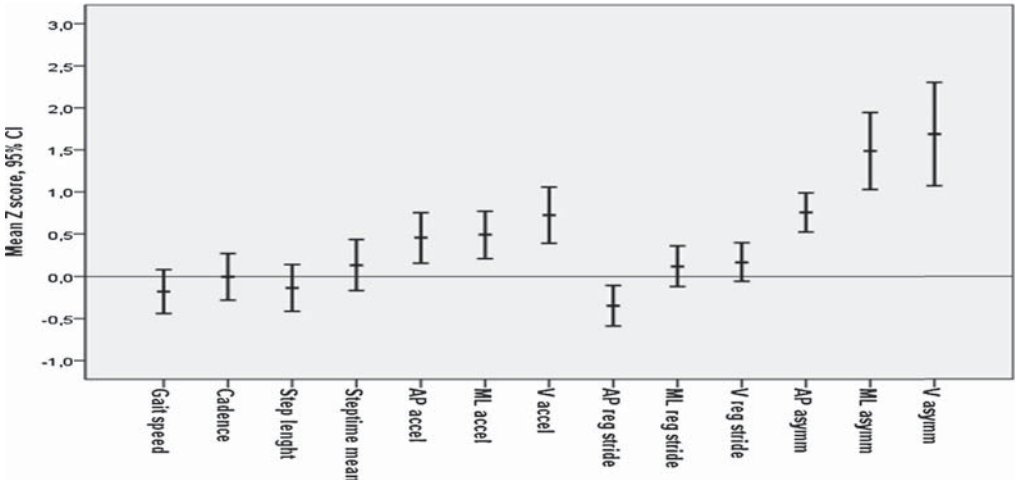

Fig. 1. Mean *z* scores, 95% CI, of gait parameters in children with cerebral palsy.
AP accel, anterioposterior acceleration; ML accel, mediolateral acceleration; *V* accel, vertical acceleration; AP reg stride, anterioposterior regularity between strides; ML reg stride, mediolateral regularity between strides; *V* reg stride, vertical regularity between strides; AP asymm, anterioposterior asymmetry; ML asymm, mediolateral asymmetry; *V* asymm, vertical asymmetry.

Table 3
Increase in acceleration with increasing gait speed in 41 children with cerebral palsy and 29 typical developing children assessed with a linear mixed model.

Gait parameters	CP		TD		Mean diff	95% CI	*p*
	Mean	Se	Mean	Se			
AP acceleration (RMS)	2.53	0.16	1.93	0.18	0.57	0.13 to 1.08	**0.013**
ML acceleration (RMS)	2.59	0.22	2.50	0.17	0.09	−0.38 to 0.57	0.698
V acceleration (RMS)	4.17	0.32	3.20	0.27	0.97	0.23 to 1.71	**0.011**

AP, anterioposterior; ML, mediolateral; *V*, vertical; RMS, root mean square.

3.2. Gait characteristics of children with different GMFCS levels compared with TD children

There were significant differences (ANOVA) in acceleration between children with CP (including the different GMFCS levels, I, II, and III) and TD children in the AP ($p = 0.022$), the ML ($p = 0.013$), and *V* directions ($p = 0.001$). Fig. 2a shows that acceleration *z*-scores increased with increasing GMFCS levels in all three directions.

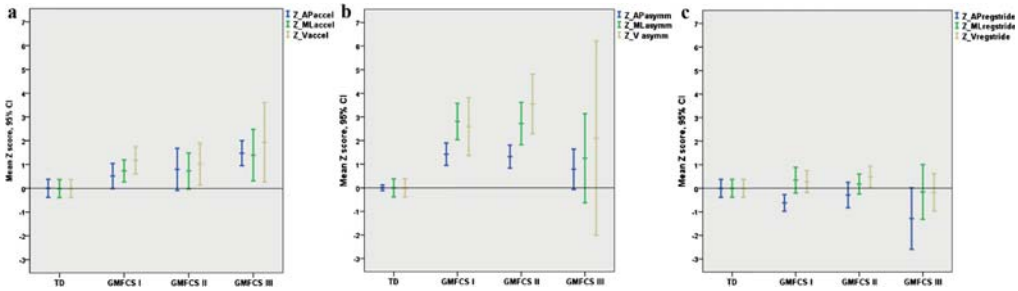

Fig. 2. Mean *z*-scores, 95% CI, of gait parameters for the different GMFCS levels.
TD, children with typical development; GMFCS, Gross Motor Functioning Classification System; AP accel, anterioposterior acceleration; ML accel, mediolateral acceleration; *V* accel, vertical acceleration; AP reg stride, anterioposterior regularity between strides; ML reg stride, mediolateral regularity between strides; *V* reg stride, vertical regularity between strides; AP asymm, anterioposterior asymmetry; ML asymm, mediolateral asymmetry; *V* asymm, vertical asymmetry.

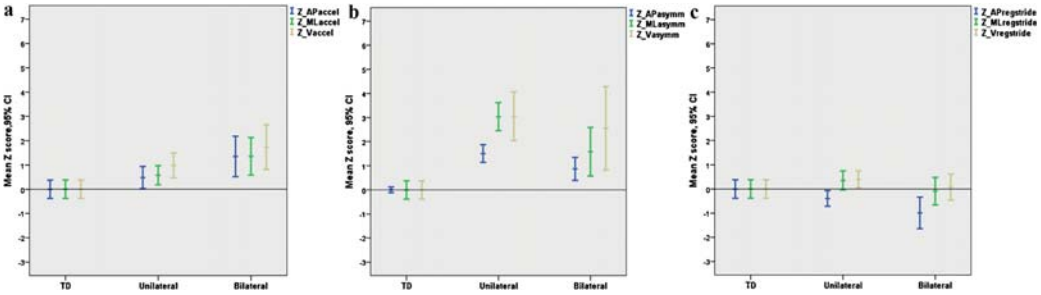

Fig. 3. Mean *z*-scores, 95% CI, of gait parameters for the unilateral and bilateral CP.
TD, children with typical development; AP accel, anterioposterior acceleration; ML accel, mediolateral acceleration; *V* accel, vertical acceleration; AP reg stride, anterioposterior regularity between strides; ML reg stride, mediolateral regularity between strides; *V* reg stride, vertical regularity between strides; AP asymm, anterioposterior asymmetry; ML asymm, mediolateral asymmetry; *V* asymm, vertical asymmetry.

The same increase was not found for asymmetry in any of the directions (Fig. 2b). Instead, there was a significant deviation in asymmetry between children with GMFCS I and TD children in all three directions ($p < 0.001$ versus TD children; Scheffe's post hoc test), but with no further deviation with increasing GMFCS levels. There were also statistically significant differences in asymmetry between children with GMFCS II and TD children in all directions, while asymmetry did not differ significantly between GMFCS III children and TD children. For regularity between strides (Fig. 2c), the only statistically significant difference was a lower AP stride regularity among GMFCS III children compared with TD children ($p = 0.037$; Scheffe's post hoc test).

The post hoc tests did not reveal any differences between the different GMFCS levels for any of the variables (progression and balance variables).

3.3. Gait characteristics of children with different CP subtypes compared with TD children

Fig. 3a shows that children with unilateral CP had higher accelerations only in the *V* direction ($p = 0.018$) compared with TD children, while children with bilateral CP had higher accelerations in all three directions ($p \leq 0.003$). Compared with TD children, asymmetry (Fig. 3b) differed significantly in all three directions both for children with unilateral CP ($p \leq 0.001$) and children with bilateral CP ($p \leq 0.003$). In contrast, regularity between strides (Fig. 3c) differed only between TD children and children with bilateral CP, and only in the AP direction ($p = 0.010$).

3.4. Gait characteristics in children with different CP subtypes

Compared to children with unilateral CP, children with bilateral CP had higher accelerations in the AP ($p = 0.043$) and ML directions ($p = 0.041$). In contrast, children with unilateral CP had a more asymmetric gait in the same directions ($p = 0.037$ and $p = 0.008$ in the AP and ML directions, respectively). Neither acceleration ($p = 0.111$) nor asymmetry ($p = 0.579$) differed between the two CP subtypes in the *V* direction. Regularity between strides did not differ statistically significant between the two groups in any direction.

4. Discussion

In the study, we found that children with CP had significant difficulties in balance control during gait, as reflected in higher trunk accelerations and asymmetry in the AP, ML, and *V* directions and lower regularity in the AP direction when compared with TD children. Moreover, the differences in balance control between the two groups increased with increasing gait speed. In contrast, the progression component of gait did not differ between children with CP and TD children when walking along a short pathway. Lastly, there were significant differences in balance control during gait between children with unilateral CP and children with bilateral CP.

4.1. Internal validity

It is unlikely that the main findings in this study were due to chance, as indicated by the low *p*-values. However, due to the relatively small sample size in some of the subgroup analyses, such as when comparing different GMFCS levels or CP subtypes, the lack of statistical significance should be interpreted with caution. The inclusion of a broad representation of the CP population is a strength of the present study, as it enabled us to study the gait characteristics according to severity and CP subtypes. The multivariable analyses indicated that the results could not be explained by differences in age, sex, height, BMI, or gait speed.

One limitation of our study is the lack of information on validity and reliability of using a trunk-worn accelerometer on children with CP, although high reliability has been reported in the case of healthy adults (Moe-Nilssen, 1998b). However, our findings demonstrating significant differences in balance parameters between children with CP and TD children indicate a face validity of the trunk-worn accelerometer in this population.

4.2. Causality

The differences in balance control between children with CP and TD children were significant, as suggested by differences in the asymmetry variables between 0.8 and 1.8 standard deviations, while deviations in the acceleration variables were between 0.4 and 0.7 standard deviations and for AP stride regulatory around 0.3 standard deviations (Fig. 1). The accelerations during gait increased among children with CP with increasing GMFCS levels (Fig. 2) when compared with the TD children, which is consistent with increasing difficulties in gait with increasing impairments in gross motor function. In contrast, the largest deviations in the asymmetry variables were observed among children with GMFCS level I. This finding may be explained by the fact that all children in the GMFCS I group had the spastic unilateral CP subtype which is consistent with the findings from a study conducted by Iosa et al. (2012). The asymmetry results were closest for TD children and children with GMFCS level III, which may be due to the fact that as all children in GMFCS level III were children with bilateral CP. Thus, although gait asymmetry is considered to be a good indicator of most gait abnormalities (Auvinet et al., 2002), our results suggest that it may also reflect other aspects than balance during gait.

In contrast to the marked differences in balance during gait between children with CP and TD children, all variables suggesting progression showed less than 0.5 standard deviations between the groups. Thus, our findings indicate that the main problem of gait control in children with CP is related to impaired balance control, and may to a lesser extent be related to progression.

4.3. Comparison with other studies

We are aware of only two very recent studies that have used trunk-worn accelerometers to assess gait in children with CP. However, these two studies, by the same group (Iosa et al., 2012, 2013), included children with a mean age of five years, and only children with unilateral CP. Thus, our study is the first to use a trunk-worn accelerometer to investigate gait characteristics in older children with CP, including children with spastic bilateral CP. Higher accelerations in all three directions in the CP group are consistent with the two studies of younger children with unilateral CP assessed by a trunk-worn accelerometer (Iosa et al., 2012, 2013), and with the findings of Hsue et al. (2009a), who assessed balance by optoelectronic motion analysis. In contrast, our study did not reveal any differences between CP and TD children in the variables indicating progression (gait speed), a finding that is consistent with the studies of Feng et al. (2014), Iosa et al. (2012), Dallmeijer & Brehm (2011), and Kurz et al. (2012), despite some differences in study design. However, other studies have reported lower gait speed means in children with CP (Abel & Damiano, 1996; Bruijn et al., 2013; Hsue et al., 2009b; Meyns et al., 2012; Norlin & Odenrick, 1986; Prosser, Lauer, VanSant, Barbe, & Lee, 2010). Possible explanations for the different results between those studies and our study may be that they included more children with the bilateral CP subtype (Abel & Damiano, 1996; Bregou Bourgeois et al., 2014; Norlin & Odenrick, 1986), younger children (Bruijn et al., 2013; Meyns et al., 2012; Norlin & Odenrick, 1986; Prosser et al., 2010), or significantly longer walking distances (Bregou Bourgeois et al., 2014). In our study the participants walked on a 5 m pathway, with a 2 m acceleration pathway before the start of the measurements and a 2 m deceleration pathway after the measurements were obtained. It should not be overlooked that children with CP and TD children need different times to reach their preferred speed, and that they also may start to decelerate differently. Such differences may have contributed to the similarity in the progression parameters.

We found that children with CP presented less regularity between strides (in the AP direction) than TD children. This finding is in agreement with earlier studies reporting increased variability in gait characteristics in children with CP (Hsue et al., 2009a; Katz-Leurer, Rotem, Keren, & Meyer, 2009; Kurz et al., 2012; Prosser et al., 2010).

4.3.1. Interpretation of findings

The increased trunk accelerations during gait observed in the present study may be a consequence of impaired trunk control, or a compensatory mechanism of impairments of the lower limbs, or both, consistent with the notation that the "*trunk may be oriented secondary to foot position or vice versa*" (Moe-Nilssen & Helbostad, 2005). A possible explanation for the higher accelerations may be the inability to attenuate the oscillations of the COM arising from abnormal muscle tone, muscle weakness, and joints stiffness in the lower limbs and in the trunk in the case of the children with CP (Hsue et al., 2009a; Kavanagh, Morrison, & Barrett, 2005). Increased accelerations of COM may also be a result of excessive reactive adjustments, which may be explained by inadequate anticipatory balance control (Moe-Nilssen, 1998a). Earlier studies have shown that children with CP have impaired anticipatory balance control in standing (Girolami, Shiratori, & Aruin, 2011; Tomita et al., 2011). Moreover, studies have shown that in healthy adults the proximal (hip/trunk) muscles are the primary contributors to anticipatory balance control during gait (Latash & Hadders-Algra, 2008; Tang, Woollacott, & Chong, 1998). Impaired trunk control has been reported in children at all GMFCS levels, and may thus be an explanation of impaired anticipatory control (Heyrman, Desloovere et al., 2013; Prosser et al., 2010; Saether, Helbostad, Adde, Jorgensen, & Vik, 2013; Saether & Jorgensen, 2011).

The steeper increase in accelerations with higher gait speed in children with CP compared with TD children may also be seen as compensatory mechanisms. Hence, children with CP have to "pay a price" in terms of a higher "cost" in order to be able to walk faster (Mulder, Zijlstra, & Geurts, 2002). The compensatory mechanisms may be due to insufficient muscle strength, such as in the plantar flexors, and that children with CP therefore may use their hips, pelvis, and trunk (proximal joints) to produce a momentum to propel their extremities forward, as suggested by some authors (Abel & Damiano, 1996; Feng et al., 2014; Gage, 2004; Hsue et al., 2009a). Gage (Gage, 2004) describes it like walking in deep mud or snow: "*one is forced to derive the power for mobility from 'pull up' from hips and knees, as opposed to 'push-off' from ankle and foot.*" It is likely that such proximal compensations may have contributed to the increased trunk accelerations in the children with CP, and hence challenged their balance control.

The differences between CP and TD children were largest in the *V* accelerations. In children, the value of *V* accelerations of COM at foot contact has been proposed as a developmental index of the postural capacity to control gravitational forces (Breniere & Bril, 1998). Impaired extensor muscle strength in children with CP (Eek, Tranberg, & Beckung, 2011) may be a reason for the differences in the *V* accelerations, as these muscles play an important role in the antigravity function (Bril & Breniere, 1992). Moreover, the results for regularity were closest to TD children. It may be speculated that the most affected children (GMFCS III) are more regular than children with less impairment (GMFCS I) because their strategy is to reduce the 'degrees of freedom' in order to be more stable. This strategy has been observed in earlier studies of fit and frail elderly people, where the frail were more regular than the fit group (Moe-Nilssen & Helbostad, 2005).

5. Conclusion

Our results showed significant differences in gait characteristics between children with CP and TD children. The differences may be more related to balance than to progression, and these problems seem to rise with increasing gross motor impairment and increasing speed. However, further studies are needed to explore the relation between progression and balance, and to unravel how (primary) impairments in trunk control and compensatory mechanisms due to impairments of the lower limbs affect balance during gait in children with CP.

References

Aaslund, M. K., Helbostad, J. L., & Moe-Nilssen, R. (2011). Familiarisation to body weight supported treadmill training for patients post-stroke. *Gait and Posture, 34*(4), 467–472.

Abel, M. F., & Damiano, D. L. (1996). Strategies for increasing walking speed in diplegic cerebral palsy. *Journal of Pediatric Orthopedics, 16*(6), 753–758.

Auvinet, B., Berrut, G., Touzard, C., Moutel, L., Collet, N., Chaleil, D., et al. (2002). Reference data for normal subjects obtained with an accelerometric device. *Gait and Posture, 16*(2), 124–134.

Bregou Bourgeois, A., Mariani, B., Aminian, K., Zambelli, P. Y., & Newman, C. J. (2014). Spatio-temporal gait analysis in children with cerebral palsy using foot-worn inertial sensors. *Gait and Posture, 39*(1), 436–442.

Breniere, Y., & Bril, B. (1998). Development of postural control of gravity forces in children during the first 5 years of walking. *Experimental Brain Research, 121*(3), 255–262.

Bril, B., & Breniere, Y. (1992). Postural requirements and progression velocity in young walkers. *Journal of Motor Behaviour, 24*(1), 105–116.

Bruijn, S. M., Meyns, P., Jonkers, I., Kaat, D., & Duysens, J. (2011). Control of angular momentum during walking in children with cerebral palsy. *Research in Developmental Disabilities, 4*, 4.

Bruijn, S. M., Millard, M., van Gestel, L., Meyns, P., Jonkers, I., & Desloovere, K. (2013). Gait stability in children with Cerebral Palsy. *Research in Developmental Disabilities, 34*(5), 1689–1699.

Cherng, R., Chou, L., Su, F., Shaughnessy, W. J., & Kaufman, K. R. (2007). Using motion of the whole-body center of mass to assess the balance during gait of children with spastic cerebral palsy. *Journal of Medical and Biological Engineering, 27*(3), 150–155.

Dallmeijer, A. J., & Brehm, M. A. (2011). Physical strain of comfortable walking in children with mild cerebral palsy. *Disability and Rehabilitation, 33*(15–16), 1351–1357.

de Graaf-Peters, V. B., Blauw-Hospers, C. H., Dirks, T., Bakker, H., Bos, A. F., & Hadders-Algra, M. (2007). Development of postural control in typically developing children and children with cerebral palsy: Possibilities for intervention? *Neuroscience and Biobehavioral Reviews, 31*(8), 1191–1200.

Dobson, F., Morris, M. E., Baker, R., & Graham, H. K. (2007). Gait classification in children with cerebral palsy: A systematic review. *Gait and Posture, 25*(1), 140–152.

Eek, M. N., Tranberg, R., & Beckung, E. (2011). Muscle strength and kinetic gait pattern in children with bilateral spastic CP. *Gait and Posture, 33*(3), 333–337.

Feng, J., Pierce, R., Do, K. P., & Aiona, M. (2014). Motion of the center of mass in children with spastic hemiplegia: Balance, energy transfer, and work performed by the affected leg vs. the unaffected leg. *Gait and Posture, 39*(1), 570–576.

Gage, J. R. (2004). Specific problems of the hips, knees and ankles. In Gage, J. R. (Ed.). *The treatment of gait problems in cerebral palsy* pp. 205–216). (vol. 1London: Mac Keith Press.

Girolami, G. L., Shiratori, T., & Aruin, A. S. (2011). Anticipatory postural adjustments in children with hemiplegia and diplegia. *Journal of Electromyography and Kinesiology, 21*(6), 988–997.

Henriksen, M., Lund, H., Moe-Nilssen, R., Bliddal, H., & Danneskiod-Samsoe, B. (2004). Test–retest reliability of trunk accelerometric gait analysis. *Gait and Posture, 19*(3), 288–297.

Heyrman, L., Desloovere, K., Molenaers, G., Verheyden, G., Klingels, K., Monbaliu, E., et al. (2013). Clinical characteristics of impaired trunk control in children with spastic cerebral palsy. *Research in Developmental Disabilities, 34*(1), 327–334.

Heyrman, L., Feys, H., Molenaers, G., Jaspers, E., Monari, D., Meyns, P., et al. (2013). Three-dimensional head and trunk movement characteristics during gait in children with spastic diplegia. *Gait and Posture, 38*(4), 770–776.

Hodt-Billington, C., Helbostad, J. L., Vervaat, W., Rognsvag, T., & Moe-Nilssen, R. (2011). Changes in gait symmetry, gait velocity and self-reported function following total hip replacement. *Journal of Rehabilitation Medicine, 43*(9), 787–793.

Hsue, B. J., Miller, F., & Su, F. C. (2009a). The dynamic balance of the children with cerebral palsy and typical developing during gait. Part II. Instantaneous velocity and acceleration of COM and COP and their relationship. *Gait and Posture, 29*(3), 471–476.

Hsue, B. J., Miller, F., & Su, F. C. (2009b). The dynamic balance of the children with cerebral palsy and typical developing during gait. Part I. Spatial relationship between COM and COP trajectories. *Gait and Posture, 29*(3), 465–470.

Iosa, M., Marro, T., Paolucci, S., & Morelli, D. (2012). Stability and harmony of gait in children with cerebral palsy. *Research in Developmental Disabilities, 33*(1), 129–135.

Iosa, M., Morelli, D., Marro, T., Paolucci, S., & Fusco, A. (2013). Ability and stability of running and walking in children with cerebral palsy. *Neuropediatrics, 44*(3), 147–154.
Katz-Leurer, M., Rotem, H., Keren, O., & Meyer, S. (2009). Balance abilities and gait characteristics in post-traumatic brain injury, cerebral palsy and typically developed children. *Developmental Neurorehabilitation, 12*(2), 100–105.
Kavanagh, J. J., & Menz, H. B. (2008). Accelerometry: A technique for quantifying movement patterns during walking. *Gait and Posture, 28*(1), 1–15.
Kavanagh, J. J., Morrison, S., & Barrett, R. S. (2005). Coordination of head and trunk accelerations during walking. *European Journal of Applied Physiology and Occupational Physiology, 94*(4), 468–475.
Kavanagh, J. J., Morrison, S., James, D. A., & Barrett, R. (2006). Reliability of segmental accelerations measured using a new wireless gait analysis system. *Journal of Biomechanics, 39*(15), 2863–2872.
Kurz, M. J., Arpin, D. J., & Corr, B. (2012). Differences in the dynamic gait stability of children with cerebral palsy and typically developing children. *Gait and Posture, 36*(3), 600–604.
Latash, M., & Hadders-Algra, M. (2008). What is posture and how is it controlled. In Hadders-Algra, M., & Carlberg, E. B. (Eds.), *Postural control: A key issue in developmental disorders* pp. 3–21). (vol. 1London: Mac Keith Press.
Massaad, F., Dierick, F., van den Hecke, A., & Detrembleur, C. (2004). Influence of gait pattern on the body's centre of mass displacement in children with cerebral palsy. *Developmental Medicine and Child Neurology, 46*(10), 674–680.
Meyns, P., Desloovere, K., Van Gestel, L., Massaad, F., Smits-Engelsman, B., & Duysens, J. (2012). Altered arm posture in children with cerebral palsy is related to instability during walking. *European Journal of Paediatric Neurology, 16*(5), 528–535.
Moe-Nilssen, R. (1998a). A new method for evaluating motor control in gait under real-life environmental conditions. Part 2. Gait analysis. *Clinical Biomechanics, 13*(4–5), 328–335.
Moe-Nilssen, R. (1998b). Test–retest reliability of trunk accelerometry during standing and walking. *Archives of Physical Medicine and Rehabilitation, 79*(11), 1377–1385.
Moe-Nilssen, R., Aaslund, M. K., Hodt-Billington, C., & Helbostad, J. L. (2010). Gait variability measures may represent different constructs. *Gait and Posture, 32*(1), 98–101.
Moe-Nilssen, R., & Helbostad, J. L. (2004). Estimation of gait cycle characteristics by trunk accelerometry. *Journal of Biomechanics, 37*(1), 121–126.
Moe-Nilssen, R., & Helbostad, J. L. (2005). Interstride trunk acceleration variability but not step width variability can differentiate between fit and frail older adults. *Gait and Posture, 21*(2), 164–170.
Mulder, T., Zijlstra, W., & Geurts, A. (2002). Assessment of motor recovery and decline. *Gait and Posture, 16*(2), 198–210.
Norlin, R., & Odenrick, P. (1986). Development of gait in spastic children with cerebral palsy. *Journal of Pediatric Orthopedics, 6*(6), 674–680.
Palisano, R., Rosenbaum, P., Walter, S., Rusell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology, 39*, 214–223.
Prosser, L. A., Lauer, R. T., VanSant, A. F., Barbe, M. F., & Lee, S. C. (2010). Variability and symmetry of gait in early walkers with and without bilateral cerebral palsy. *Gait and Posture, 31*(4), 522–526.
Romkes, J., Peeters, W., Oosterom, A. M., Molenaar, S., Bakels, I., & Brunner, R. (2007). Evaluating upper body movements during gait in healthy children and children with diplegic cerebral palsy. *Journal of Pediatric Orthopaedics. Part B, 16*(3), 175–180.
Saether, R., Helbostad, J. L., Adde, L., Jorgensen, L., & Vik, T. (2013). Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy. *Research in Developmental Disabilities, 34*(7), 2075–2084.
Saether, R., & Jorgensen, L. (2011). Intra- and inter-observer reliability of the Trunk Impairment Scale for children with cerebral palsy. *Research in Developmental Disabilities, 32*(2), 727–739.
Schwartz, M. H., Rozumalski, A., & Trost, J. P. (2008). The effect of walking speed on the gait of typically developing children. *Journal of Biomechanics, 41*(8), 1639–1650.
Tang, P. F., Woollacott, M. H., & Chong, R. K. (1998). Control of reactive balance adjustments in perturbed human walking: Roles of proximal and distal postural muscle activity. *Experimental Brain Research, 119*(2), 141–152.
Tomita, H., Fukaya, Y., Ueda, T., Honma, S., Yamashita, E., Yamamoto, Y., et al. (2011). Deficits in task-specific modulation of anticipatory postural adjustments in individuals with spastic diplegic cerebral palsy. *Journal of Neurophysiology, 105*(5), 2157–2168.
Wallard, L., Bril, B., Dietrich, G., Kerlirzin, Y., & Bredin, J. (2012). The role of head stabilization in locomotion in children with cerebral palsy. *Annals of Physical and Rehabilitation Medicine, 2*(12), 01286–01289.
Winter, D. A. (1995). Human balance and posture control during standing and walking. *Gait & Posture, 3*(4), 193–214.

Paper IV

The relationship between trunk control in sitting and during gait in children and adolescents with cerebral palsy

RANNEI SAETHER,[1,2] JORUNN L HELBOSTAD,[3,4] LARS ADDE,[1,4] SIRI BRÆNDVIK,[3,4] STIAN LYDERSEN,[2,5] TORSTEIN VIK [1,2]

1. Department of Laboratory Medicine, Children's and Women's Health, Norwegian University of Science and Technology, Trondheim, Norway

2. Department of Pediatrics, St. Olavs Hospital, University Hospital of Trondheim, Trondheim, Norway

3. Department of Neuroscience, Norwegian University of Science and Technology, Trondheim, Norway

4. Clinic for Clinical Services, St. Olavs University Hospital, Trondheim, Norway

5. Regional Centre for Child and Youth Mental Health and Child Welfare – Central Norway, Norwegian University of Science and Technology, Trondheim, Norway

Corresponding author:

Rannei Sæther

Norwegian University of Science and Technology
Faculty of Medicine
Department of Laboratory Medicine, Children's and Women's Health
Postboks 8905
7491 Trondheim

Phone: 0047 99248133

e-mail: rannei.sather@ntnu.no

Other authors:

Jorunn Helbostad: jorunn.helbostad@ntnu.no

Lars Adde: lars.adde@ntnu.no

Siri Brændvik: siri.merete.brandvik@svt.ntnu.no

Stian Lydersen: stian.lydersen@ntnu.no

Torstein Vik: torstein.vik@ntnu.no

Abstract

Aim: We wanted to assess the relationship between trunk control in sitting and trunk control during gait and to compare if this relationship differed, using two tests of trunk control in sitting.

Method: 26 children with spastic cerebral palsy (CP) (17 males, mean age 13.5 (range 8-18) years) were included. Trunk control in sitting was assessed with the Trunk Impairment Scale (TIS) and the Trunk Control Measurement Scale (TCMS), and trunk control during gait by a trunk-worn accelerometer.

Results: Trunk control in sitting assessed with the TCMS and the TIS total scores both correlated with trunk accelerations during gait (R_p= 0.67 and 0.60, respectively). Moreover, some subscale scores correlated equally well with trunk control during gait (the TCMS dynamic sitting balance reaching subscale score (DSB-R); R_p= 0.61) or even higher (TIS dynamic sitting balance subscale (DSB); R_p= 0.66).

Interpretation: Trunk control in sitting is moderate to good correlated with trunk control during gait. Our results suggest that two subscales of these tools, being less time consuming, may be applied in the clinical assessment of trunk control. Future studies are needed to explore how this information may be applied in the planning of 'gait interventions 'in children with CP.

What this paper adds

- This is the first study to show that trunk control in sitting correlates with trunk control during gait in children and adolescents with CP
- Two tests of trunk control in sitting control were equally related to trunk control during gait
- For clinical purposes dynamic subscales, being less time consuming than the complete tests may be applied

Introduction

Control of the trunk during gait is especially important for balance in man since two-thirds of the body mass (head, arms, and trunk) is located in the upper two-thirds of the body height, making the body unstable.[1] Moreover, the trunk serves a number of control functions during gait: it plays an important role in proactive balance control,[2] in steering (moving the center of mass (COM) to a new direction),[3] and in attenuation of gait-related oscillations to promote stability of the head.[4] Furthermore, the trunk interacts with the lower limb movements to achieve efficient locomotion.[5]

Poor trunk control is a primary impairment in children, adolescents and adults with cerebral palsy (CP) and may affect activities in daily life, such as sitting and walking.[1; 6] Treatment of the latter impairment, include orthopedic surgery, botulinum toxin injections, and/or application of orthoses. In the decision process leading to such treatment the main focus has been on the lower extremities,[7] while less attention has been paid to the trunk and to trunk control during gait.

The deviations observed in the trunk during gait are most often interpreted as compensations for impairments, such as weakness and impaired control in the lower limbs. However, the deviations may also be due to a primary impairment in the trunk.[6] Moreover, primary impairments in the trunk may cause compensatory movements in the lower limbs, for example by allowing the pelvis to rotate anteriorly and thereby cause increased hip flexion.[8] It may therefore be important to reveal primary trunk deficits in order to plan the most appropriate "gait treatment" in children with CP. This suggestion is supported by Rutz et al.,[9] who claim that the variations in the results of gait treatments are poorly understood.

However, it may be may be difficult to separate deviations in trunk control during gait due to "primary" impairments of trunk control from those due to "secondary" impairments caused by impairments in the lower extremities,[8] even when using advanced laboratory equipment. Since trunk control in sitting is less influenced by impairments in the lower extremities, assessment of trunk control in sitting may be used as a first step to identify primary impairments. Even though the two tasks are clearly different, trunk control in sitting may be assessed as an indicator of impaired (primary) trunk control during gait, if there is a relationship between trunk control in sitting and during gait. If such a relationship can be

documented, the implementation of a short test of trunk control in sitting may provide information about primary impairments in trunk control and thus may lead to improved decision processes regarding the choice of gait interventions. Moreover, the results of such a sitting assessment might be used to select children who need a thorough assessment of the relationship between the trunk and lower limbs, including full-body 3D gait analysis, before a treatment option is chosen.

The aim of this study was therefore to assess the relationship between trunk control in sitting, measured with the Trunk Impairment Scale (TIS) and the Trunk Control Measurement Scale (TCMS), and trunk control during gait, measured with a trunk-worn accelerometer in children and adolescents with CP (hereafter referred to as children with CP). A secondary aim was to assess which of the sitting trunk control tests (the less time-consuming TIS or its expanded version, the TCMS) were most related to trunk control during gait.

2. Materials and methods

2.1 Study design and participants

In this cross-sectional study, trunk control during gait was assessed using a trunk-worn accelerometer and trunk control in sitting with the TIS and the TCMS. A consecutive sample of 26 children with spastic CP (17 males), in the age group 8–18 years, were recruited from the Neuro-orthopedic outpatient clinic at St. Olavs University Hospital (Trondheim, Norway). To be eligible, participants had to be able to understand instructions and to walk at least 10 meters without support, shoes, or orthoses. Exclusion criteria were treatment with botulinum toxin in the lower extremities during the last 4 months and/or surgery during the last 12 months. All participants and/or their parents provided written informed consent. The study was conducted in conformity with the declaration of Helsinki, and was approved by Regional Committee for Medical and Health Research Ethics in Central Norway. The characteristics of the children are listed in Table 1.

2.2 Trunk control in sitting

Trunk control in sitting was tested with the TIS and the TCMS. The TIS was developed to evaluate trunk control in adults after stroke, through assessment of static sitting balance (SSB) and dynamic sitting balance (DSB) and trunk coordination (C) in sitting position.[10] The total score, which consist of the sum of three subscale scores, ranging from zero (lowest

performance) to 23 (best performance). The TIS has been tested for reliability and for validity in children and adolescents with CP in the age group 5–18 years.[11; 12]

The TCMS was developed from the TIS and was expanded to include assessments of selective trunk movements and dynamic reaching. The total score ranges from zero (lowest performance) to 58 (best performance), where the total score is the sum of three subscale scores: static sitting balance (SSB), dynamic sitting balance-selective movement control (DSB-S) and dynamic sitting balance-reaching (DSB-R). The TCMS has been tested for reliability and validity in children with CP in the age group 8–15 years.[13]

Since the TCMS is an expanded version of the TIS, the children were first tested with the use of the TIS, and then tested for the remaining tasks of the TCMS. The assessments of trunk control were video recorded, and the scoring of the test was performed through observation of the video by one of the authors (RS), without prior knowledge of the results of trunk control during gait.

2.3 Trunk control during gait

Trunk control during gait was tested with an accelerometer attached with double-sided tape to the L3 region of the lower back in order to acquire accelerometer and orientation data. The sensor, a six degrees-of-freedom inertial sensor (MTx. XSens, Enschede, NL) (weight: 15 grams), contains tri-axial units of accelerometers, gyroscopes, and magnetometers and is connected to a battery-operated communication unit (weight: 300 grams), which was also worn by the children. Data were acquired at a sampling frequency of 100 Hz and transmitted in real time to a laptop using Bluetooth technology. Gait time was registered by photoelectric cells synchronized with the accelerometer device. The trunk-worn accelerometer has previously been tested for test-retest reliability in healthy adults.[14]

Trunk control during gait was tested by asking the participants to walk back and forth along a 5 meter walkway at three different gait speeds according to the following instructions: (1) “Walk normally, walk as you usually do,” (2) “Walk as fast as you can, without running,” and (3) “Walk slowly, as if you were strolling around.” To obtain gait data during steady-state speed, the participants started walking 2 m in front of the photoelectric cells and continued to walk 2 m beyond the photoelectric cells. Thus, a series of six gait sequences was completed by each participant, after two sequences used for “warming-up.”

2.4 Data acquisition and analysis of gait variables

A customized software application TRASK, run under Matlab R2011b (Math Works Inc., Natrick, MA), was used for signal processing and calculation of gait variables. By applying the accelerometer as an inclinometer, the average tilt of the measuring axes could be calculated for each sample, thus eliminating gravity bias. The raw acceleration data were transformed into a horizontal-vertical coordinate system by a trigonometric algorithm and reported along anteroposterior (AP), mediolateral (ML), and vertical (V) axes. Trunk acceleration amplitudes for each gait interval were expressed by root mean square (RMS) values (hereafter referred to as trunk accelerations). The periodicity of the acceleration curve enabled steps and strides to be registered. Interstep and interstride trunk acceleration regularity were calculated using an unbiased autocorrelation procedure, whereby an acceleration time series was correlated to the same series at a phase shift equivalent to one step and one stride, respectively. Perfect replication of the signals between consecutive steps or strides gives an autocorrelation coefficient of 1.[15] Step time and cadence for each walk was estimated from the autocorrelation curve of the vertical axis. Mean step length for each walk was calculated as gait distance/number of steps. The mean value of two walks (back and forth along the pathway) (a mean of 9.4 steps (SD 1.7)) at preferred speed was used in the calculations.

In our previous study we found that among the gait parameters derived from a trunk worn sensor; trunk accelerations and regularity between strides best described trunk control during gait in children with CP.[16] These parameters were therefore used as the primary dependent variables in the assessment of the relationship between trunk control in sitting and trunk control during gait. However, we also studied the relationship between trunk control in sitting and spatiotemporal gait parameters derived from the sensor (cadence, step length, and step time).

2.5 Other variables

Information about motor functioning, described by the Gross Motor Functioning Classification System (GMFCS),[17] and the classification of CP subtypes was obtained from each child's hospital medical records.

The participants' height was measured to the nearest cm with a stadiometer, and their weight was measured to the nearest 0.5 kg using an electronic weight (Seca digital 770). Their body mass index (BMI) was then calculated as weight (kg)/height^2 (m^2).

2.6 Statistical analysis

The relationship between trunk control in sitting and during gait was first assessed by calculating the Pearson's (R) correlation coefficient. To control for the potential confounders age, height, BMI, and gait speed we calculated the partial correlation coefficient (R_p). As proposed by Portney,[18] correlation coefficients between 0 and 0.25 may be considered to indicate little or no relationship, between 0.25 and 0.50 low, between 0.50 and 0.75 moderate to good, and above 0.75 may be considered to indicate a good to excellent relationship. For the purpose of our study and in order to improve the readability of the results, we described correlation coefficients between 0.50 and 0.75 as "moderate" and those above 0.75 as "good."

Two-sided p-values < 0.05 were considered significant, and 95% confidence intervals (CI) were reported where relevant. Statistical analyses were performed using SPSS 19.

3. Results

The mean values and SD of general gait parameters and parameters representing trunk control in sitting and during gait are presented in Supplementary 1.

3.1 Total scores

The *TIS total score* correlated moderately with AP regularity (R = 0.60, p = 0.001) and after controlling for gait speed the correlation with AP acceleration increased from low to moderate (R_p = -0.51, p = 0.009) (Table 2 and Figure 1). The *TCMS total score* correlated moderately with AP acceleration, AP regularity, and step length (R = -0.51, 0.58, and 0.54 respectively, p $\leq$ 0.008). After controlling for gait speed, the correlation with AP acceleration increased (R_p = -0.67, p = 0.001). In addition, the correlation with V acceleration now improved from low to moderate (R_p = -0.58, p = 0.003) (Table 2 and Figure 2).

3.2 Subscale scores

The *TIS* subscale score SSB correlated at least as well or even better with AP and ML accelerations and the TIS total score. Another subscale, the *TIS DSB* correlated moderately with AP and V regularity (Table 2 and Figure 1).

The *TCMS* subscales SSB and DSB-R both correlated moderately with AP acceleration during gait, and SSB and DSB-R also correlated with V acceleration and AP regularity, respectively. These correlations for the subscale scores were almost as high as the correlation between TCMS total score and the same gait parameters. The third subscale, *DSB-S* correlated moderately with AP regularity (Table 2 and Figure 2).

4. Discussion

In the study we found that trunk control in sitting, assessed with the TCMS total score and TIS DSB, both correlated moderately with trunk control during gait. Moreover, some subscale scores correlated almost equivalent to (TCMS DSB-R subscale score) or even more highly (TIS DSB subscale score) with trunk control during gait.

4.1 Internal validity

It is unlikely that the main findings in the study were due to chance as indicated by the low p-values. However, due to the relatively small sample size, the results should be interpreted with caution. A limitation of our study is the lack of information on the validity and reliability of using a trunk-worn accelerometer in studies of children with CP, although high reliability has been reported in healthy adults. Moreover, the documented differences between subgroups in children with CP, assessed with a trunk-worn accelerometer, may indicate face validity. [16]

4.2 Causality

All functional tasks require trunk control, but the control strategies vary according to the task and the environment. Since trunk control in sitting and during gait are two different tasks it is not reasonable to expect excellent correlation between the two. Therefore, the moderate to good correlation found in our study may be considered high and suggests that trunk control in sitting, assessed with the TCMS total score and the TIS DSB, may potentially identify primary impairments of trunk control.

4.3 Comparison with other studies

To our knowledge, our study is the first to examine the relationship between trunk control in sitting and during gait in children with CP, except for a large retrospective study of children with CP (n = 5366), where the ability to sit by the age of 2 years was emphasized as a particularly strong predictor of ambulation.[19] The findings from the latter study may be consistent with those from our study because they underscore the relationship between trunk control in sitting and gait. However, this relationship has been explored in other patient groups. In post-stroke patients it has been found that trunk control in sitting, assessed with TIS, explained 52% of functional recovery (including gait),[20] and in people with knee osteoarthritis, trunk control has been found to discriminate between patients with and without "poor" gait speed (1.0 m/s).[21] However, the relation between trunk control in standing and during gait has been assessed in children with spastic diplegia.[22; 23] In the above mentioned studies, the dynamic balance tasks were found to be higher than the static balance tasks, although moderately correlated with spatiotemporal gait parameters (gait speed, step length, and cadence). This finding is in agreement with the finding from our study, where the dynamic subscales for both the TIS and the TCMS showed the highest correlations with the gait parameters. This may be explained by the "specificity of tasks"; gait is a task that requires more dynamic than static trunk control. Moreover, we found that trunk control in sitting was most strongly correlated with the gait parameters related to the direction of progression (AP acceleration and AP regularity). This finding is supported by the results of an earlier study in which we found these two parameters best reflected balance problems during gait. [16]

4.4 Interpretation of findings

Our results suggest that assessment of trunk control in sitting through an easily accomplished clinical test may also provide insights into trunk control during gait. Such insights could be valuable in clinical practice when planning gait treatments, as impaired trunk control may influence movement in the lower limbs,[24] thus making the prediction of the results of an intervention more uncertain. Further research is needed to document whether assessment of trunk control in sitting may be used to select patients who may need a more detailed examination of the relationship between their trunk and lower limbs, and whether, if trunk control in sitting is good, more advanced examinations could be omitted.

In the present study, we used two tests reflecting trunk control in sitting, the TIS and its expanded version, the TCMS, in addition to a trunk-worn accelerometer. The rationale for using two sitting tests was to explore whether the shorter and less time-consuming TIS would

show equally good correlation with gait parameters as the somewhat more time-consuming TCMS. The main results suggest that the two tests are equally well correlated with trunk control during gait. However, for the TIS, the DSB subscale score had the highest correlation, while this was the case for the total score for TCMS, although the TCMS DSB-R subscale score was not substantially lower than the total score. This suggests the possibility that in a busy clinical practice trunk control may be assessed with these subscale scores. Although there was no clear difference between the two sitting tests when correlated with balance during gait, we recommend that the TCMS DSB-R is the preferred test compared with TIS DSB, due to the better measurement properties of the TCMS.[25]

5. Conclusion

We found that trunk control in sitting, assessed with TIS and TCMS, was moderately to good correlated with trunk control during gait. Our results suggest that two subscales of these tools, being less time-consuming, may be applied in clinical assessment of trunk control. Future studies are needed to explore how this information may be applied in the planning of interventions aimed to improve the gait performance of children and adolescents with CP.

References

1 Winter DA. (1995) Human balance and posture control during standing and walking. *Gait Posture* **3**: 193-214.

2 Tang PF, Woollacott MH, Chong RK. (1998) Control of reactive balance adjustments in perturbed human walking: roles of proximal and distal postural muscle activity. *Exp. Brain Res.* **119**: 141-52.

3 Patla AE, Adkin A, Ballard T. (1999) Online steering: coordination and control of body center of mass, head and body reorientation. *Exp. Brain Res.* **129**: 629-34.

4 Kavanagh JJ, Morrison S, Barrett RS. (2005) Coordination of head and trunk accelerations during walking. *Eur. J. Appl. Physiol.* **94**: 468-75.

5 Thorstensson A, Nilsson J, Carlson H, Zomlefer MR. (1984) Trunk movements in human locomotion. *Acta Physiol. Scand.* **121**: 9-22.

6 Heyrman L, Feys H, Molenaers G, Jaspers E, Monari D, Meyns P, Desloovere K. (2013) Three-dimensional head and trunk movement characteristics during gait in children with spastic diplegia. *Gait Posture* **38**: 770-6.

7 Dobson F, Morris ME, Baker R, Graham HK. (2007) Gait classification in children with cerebral palsy: a systematic review. *Gait Posture* **25**: 140-52.

8 Trost J. (2004) Physical assessment and observational gait analysis. In: H Hart editor. *The treatment of gait problems in cerebral palsy* London: Mac Keith Press. p 71-98.

9 Rutz E, Tirosh O, Thomason P, Barg A, Graham HK. (2012) Stability of the Gross Motor Function Classification System after single-event multilevel surgery in children with cerebral palsy. *Dev. Med. Child Neurol.* **54**: 1109-13.

10 Verheyden G, Nieuwboer A, Mertin J, Preger R, Kiekens C, De Weerdt W. (2004) The Trunk Impairment Scale: a new tool to measure motor impairment of the trunk after stroke. *Clin. Rehabil.* **18**: 326-34.

11 Saether R, Jorgensen L. (2011) Intra- and inter-observer reliability of the Trunk Impairment Scale for children with cerebral palsy. *Res. Dev. Disabil.* **32**: 727-39.

12 Saether R, Helbostad JL, Adde L, Jorgensen L, Vik T. (2013) Reliability and validity of the Trunk Impairment Scale in children and adolescents with cerebral palsy. *Res. Dev. Disabil.* **34**: 2075-84.

13 Heyrman L, Molenaers G, Desloovere K, Verheyden G, De Cat J, Monbaliu E, Feys H. (2011) A clinical tool to measure trunk control in children with cerebral palsy: the Trunk Control Measurement Scale. *Res. Dev. Disabil.* **32**: 2624-35.

14 Moe-Nilssen R. (1998) Test-retest reliability of trunk accelerometry during standing and walking. *Arch. Phys. Med. Rehabil.* **79**: 1377-85.

15 Moe-Nilssen R, Helbostad JL. (2004) Estimation of gait cycle characteristics by trunk accelerometry. *J. Biomech.* **37**: 121-6.

16 Sæther R, Helbostad JL, Adde L, Brændvik S, Lydersen S, Vik T. (2013) Gait characteristics in children and adolescents with cerebral palsy assessed with a trunk-worn accelerometer. *Res. Dev. Disabil.* **Under revision**.

17 Palisano R, Rosenbaum P, Walter S, Rusell D, Wood E, Galuppi B. (1997) Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev. Med. Child Neurol.* **39**: 214-23.

18 Portney LG, Watkins MP. (2009) *Foundations of Clinical Research.* New Jersey: Pearson Education.

19 Wu YW, Day SM, Strauss DJ, Shavelle RM. (2004) Prognosis for ambulation in cerebral palsy: a population-based study. *Pediatrics* **114**: 1264-71.

20 Verheyden G, Nieuwboer A, De Wit L, Feys H, Schuback B, Baert I, Jenni W, Schupp W, Thijs V, De Weerdt W. (2007) Trunk performance after stroke: an eye catching predictor of functional outcome. *J. Neurol. Neurosurg. Psychiatry* **78**: 694-8.

21 Pua YH, Clark RA, Ong PH, Bryant AL, Lo NN, Liang Z. (2012) Association between seated postural control and gait speed in knee osteoarthritis. *Gait Posture* **4**: 4.

22 Liao HF, Jeng SF, Lai JS, Cheng CK, Hu MH. (1997) The relation between standing balance and walking function in children with spastic diplegic cerebral palsy. *Dev. Med. Child Neurol.* **39**: 106-12.
23 Moore JG. (2006) The relationship between dynamic balance and walking in children. Coral Gables, Florida: University of Miami.
24 Moe-Nilssen R, Helbostad JL. (2005) Interstride trunk acceleration variability but not step width variability can differentiate between fit and frail older adults. *Gait Posture* **21**: 164-70.
25 Saether R, Helbostad JL, Riphagen, II, Vik T. (2013) Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review. *Dev. Med. Child Neurol.* **16**: 12162.

Table 1. Characteristics of the 26 children with cerebral palsy (CP) included in the study

	All participants		GMFCS 1		GMFCS II		GMFCS III	
N	26		10		10		6	
Unilateral CP (n)	15	-	10	-	5	-	-	-
Bilateral CP (n)	11	-	-	-	5	-	6	-
Male gender, n (%)	17 (65)	-	6 (60)	-	7 (70)	-	4 (67)	-
Age (years), mean (SD)	13.5	(3.0)	13.7	(2.6)	13.6	(2.8)	13.1	(4.3)
Height (cm), mean (SD)	154.2	(17.3)	159.6	(20.3)	152.0	(11.7)	148.8	(20.5)
Weight (kg), mean (SD)	49.1	(17.7)	53.9	(22.0)	45.6	(13.7)	46.8	(16.8)
Body mass index (kg/m^2), mean (SD)	20.2	(4.3)	20.2	(4.7)	19.9	(4.9)	20.4	(2.9)

Table 2. Correlations of gait parameters and Trunk Impairment Scale and Trunk Control Measurement Scale total scores and subscale scores

	TIS	Total score				Static sitting balance (SSB)				Dynamic sitting balance (DSB)				Coordination (C)			
Gait parameters		Unadjusted		Adjusted		Unadjusted		Adjusted		Unadjusted		Adjusted		Unadjusted		Adjusted	
		R	p	R_p	p	R	p	R^p	p	R	p	R^p	p	R	p	R^p	p
Cadence (steps/min)		-0.08	0.695	-	-	-0.26	0.196	[c]-0.44	**0.027**	0.01	0.950	0.18	0.383	-0.11	0.606	[c]-0.36	0.081
Step length (m)-		0.39	**0.049**	-	-	0.48	**0.013**	-	-	0.21	0.311	-	-	0.44	**0.025**	-	-
Step time (mean)		-0.11	0.599	[c] -0.22	0.164	0.24	0.231	-	-	-0.03	0.894	-	-	-0.30	0.149	-	-
AP acceleration (RMS)		-0.36	0.067	[c]-0.51	**0.009**	-0.50	**0.009**	[c]-0.59	**0.002**	-0.30	0.145	[c]-0.39	**0.057**	-0.19	0.342	[c]-0.31	0.130
ML acceleration (RMS)		-0.31	0.119	[c]-0.42	**0.036**	-0.49	**0.012**	[c]-0.55	**0.004**	-0.37	0.064	[c]-0.45	**0.025**	-0.06	0.784	[c]-0.13	0.547
V acceleration (RMS)		-0.22	0.422	[c]-0.40	**0.049**	-0.33	0.106	[c]-0.44	**0.029**	-0.19	0.362	[c]-0.31	0.136	-0.06	0.761	[c]-0.21	0.311
AP regularity Stride (ACORR)		0.60	**0.001**	-	-	0.33	0.097	-	-	0.66	**0.001**	-	-	0.46	**0.018**	-	-
ML regularity Stride (ACORR)		0.44	**0.023**	-	-	0.30	0.144	-	-	0.46	**0.017**	-	-	0.41	**0.037**	-	-
V regularity Stride (ACORR)		0.41	**0.037**	-	-	0.21	0.307	-	-	0.51	**0.008**	-	-	0.27	0.186	-	-
	TCMS	Total score				Static sitting balance (SSB)				Dynamic sitting balance (DSB-S)				Dynamic sitting balance (DSB-R)			
Gait parameters		Unadjusted		Adjusted		Unadjusted		Adjusted		Unadjusted		Adjusted		Unadjusted		Adjusted	
		R	p	R_p	p	R	p	R_p	p	R	p	R_p	p	R	p	R_p	p
Cadence (steps/min)		-0.22	0.272	[c] 0.42	**0.035**	0.23	0.266	[c] 0.42	**0.036**	-0.09	0.650	[c] 0.34	0.094	-0.08	0.700	[c]-0.26	0.208
Step length (m)-		0.54	**0.005**	-	-	0.49	**0.011**	-	-	0.41	**0.037**	-	-	0.36	0.075	-	-
Step time (mean)		0.18	0.571	-	-	0.14	0.510	[a] 0.18	0.386	-0.10	0.620	[a]-0.27	0.898	0.03	0.895	-	-
AP acceleration (RMS)		-0.51	**0.008**	[c]- 0.67	**0.001**	-0.50	**0.010**	[c]-0.59	**0.002**	-0.28	0.161	[c]-0.42	**0.038**	-0.50	**0.010**	[c]-0.61	**0.001**
ML acceleration (RMS)		-0.31	0.125	[c] -0.49	**0.013**	-0.31	0.121	[c]-0.38	0.062	-0.19	0.354	[c]-0.28	0.176	-0.36	0.072	[c]-0.44	**0.029**
V acceleration (RMS)		-0.28	0.161	[c] -0.58	**0.003**	-0.43	**0.030**	[c]-0.56	**0.005**	-0.14	0.490	[c]-0.30	0.144	-0.32	0.117	[c]-0.45	**0.024**
AP regularity Stride (ACORR)		0.58	**0.002**	-	-	0.24	0.230	-	-	0.51	**0.001**	-	-	0.54	**0.004**	-	-
ML regularity Stride (ACORR)		0.43	**0.029**	-	-	0.16	0.436	-	-	0.41	**0.035**	-	-	0.47	**0.016**	-	-
V regularity Stride (ACORR)		0.48	**0.013**	-	-	0.13	0.519	-	-	0.36	0.069	-	-	0.38	0.058	-	-

Adjusted for: [a]Age, [b]Height, [c] Gait speed , [d]BMI, R; Pearson correlation coefficient, R_p partial correlation, AP; Anteriorposterior, ML; Mediolateral, V; Vertical, ACORR; Autocorrelation, Accel; acceleration, RMS; root mean square, ACORR; autocorrelation,. DSB-S; dynamic sitting balance-selective movement control, DSB-R; dynamic sitting balance-reaching, C; coordination

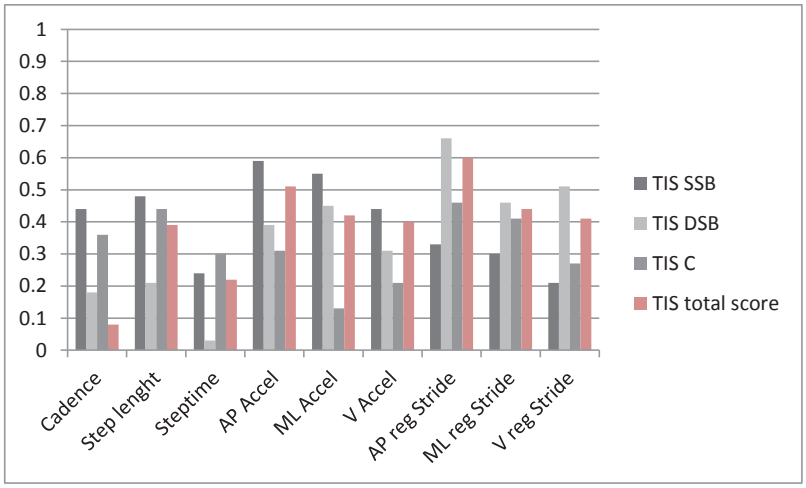

Figure 1. The partial correlation coefficients (R_p) (controlled for gait speed) (Y-axis) between gait parameters (X-axis) and Trunk Impairment Scale (red bars), as well as the subscale scores Static sitting balance (SSB; black bars), Dynamic sitting balance, (DSB; light grey bars), and Coordination (C; dark grey bars)

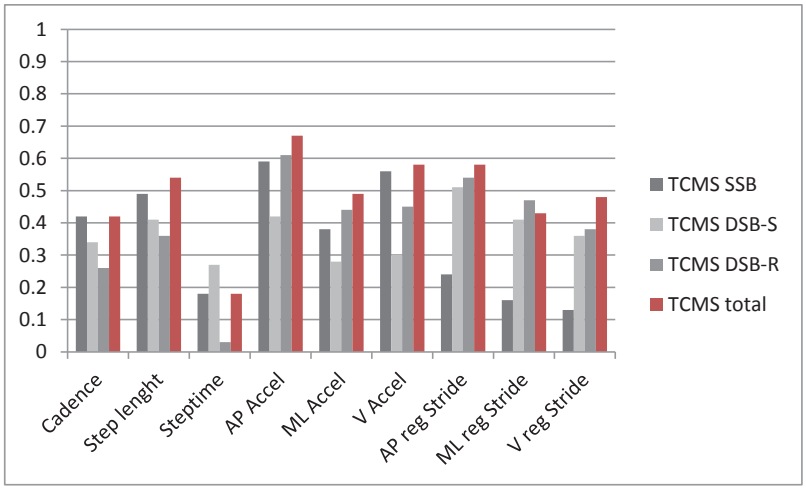

Figure 2. The partial correlation coefficients (R_p) (controlled for gait speed) (Y-axis) between gait parameters (X-axis) and Trunk Control Measurement Scale total score (red bars), as well as the subscale scores Static sitting balance (SSB; black bars), Dynamic sitting balance, selective movement control, (DSB-S; light grey bars), and Dynamic sitting balance, reaching (DSB-R; dark grey bars)