Postural Management

Assessment of posture

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Thesis for the degree of Philosophiae Doctor

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Ágrip

Inngangur: Fatlaðir fullorðnir einstaklingar með litla sem enga hreyfigetu, eru margir hverjir með miklar líkamlegar aflaganir. Stöðustjórnun er hugmyndafræði sem byggir á hugmyndum um að samhverfa í líkamsstöðu stöðvi eða seinki aflögun líkamans.

Markmið: Tilgangur ritgerðarinnar er að skapa grunn-matstækni, til að hægt sé að afla gagnreyndrar þekkingar á sviði stöðustjórnunar hjá fötluðum einstaklingum. Ritgerðin skiptist í tvo hluta. Annars vegar mat á réttmæti og áreiðanleika tveggja matstækja til að meta líkamsstöðu á hlutlægan hátt og hins vegar mat á tengslum þess að vera lengi í ósamhverfum líkamsstellingum, þess að vera með lélega stjórn á líkamshreyfingum og vera með aflagaðan líkama.

Aðferð: Gildi og áreiðanleiki matstækjanna var kannaður og líkindi á aflögunum vegna vanastellingar var metinn með þverskurðarrannsókn. Fyrra matstækið var metið með skoðun á líkamsstöðu og stjórn á líkamshreyfingum fullorðinna einstaklinga með heilalömun á myndum og myndskeiðum. Seinna matstækið var metið með samanburðarrannsókn á líkamsstöðum 7 heilbrigðra einstaklinga, þar sem niðurstöður þrívíddar myndavélar tengdri við iPad annars vegar og þrívíddar hreyfigreini (Qualisys) hins vegar voru borin saman. Fyrra matstækið ásamt gögnum frá 714/830 fullorðnum einstaklingum úr sænskum gagnagrunni sem geymir gögn um eftirfylgni með einstaklingum með heilalömun var notað til að meta tengsl á þess að dvelja lengi í slæmum líkamsstellingum, þess að vera með lélega stjórn á líkamshreyfingum, og þess að vera með aflaganir á líkamanum. Sérstök áhersla var lögð á að skoða ósamhverfa skerðingu á mjaðmabeygju, hryggskekkju og vindblásnar mjaðmir.

Niðurstaða: Bæði matstækin sýndu fram á réttmæti og áreiðanleika (weighted Kappa frá 0,85 og innri fylgni stuðull [ICC] frá 0,982). Einstaklingar með ósamhverfar skerðingar á mjaðmabeygju sýndu aukin líkindi til að vera með skakka mjaðmagrind (OR 2,6; 95% CI 1,6–2,1) og ósamhverfan bol (OR 2,1; 95% CI 1,1–4,2), aukin líkindi til að vera með hryggskekkju (OR 3,7; 95% CI 1,3–9,7) og vindblásnar mjaðmir (OR 2,6; 95% CI 1,2–5,4) samanborið við einstaklinga sem voru með samhverfa mjaðmabeygju. Einstaklingar sem ekki geta hreyft sig í liggjandi stöðu, af eigin rammleik, sýndu aukin líkindi á að vera með hryggskekkju (OR 5,7; 95% CI 3,5–9,1) og vindblásnar mjaðmir (OR 2,9; 95% CI 1,0–3,3), borið saman við þá sem geta hreyft sig í rúmi. Sama á við um þá sem ekki geta rétt úr hnjám, þeir sýndu aukin líkindi á að vera með hryggskekkju (OR 1,8; 95% CI 1,1–3,1) og vindblásnar mjaðmir (OR 1,6; 95% CI 1,1–2,3), miðað við þá sem geta rétt úr hnjám. Baklega jók líkindi á að vera með vindblásnar mjaðmir (OR 1,9; 95% CI 1,0–3,3) á meðan þeir sem dvöldu lengi í rúmlegu, voru með aukin líkindi á hryggskekkju (OR 2,9; 95% CI 1,6–2,1).

Ályktun: Niðurstöður á réttmæti og áreiðanleika matstækjanna gefa skýrt til kynna að þau henti vel í klínísku umhverfi. Ósamhverfar vanastellingar, ásamt því að dvelja lengi í sömu líkamsstöðu eykur líkindin á aflöguðum líkama.

Lykilorð:

Líkamstaða; Þrívíddar líkön; Heilalömun; Fullorðnir; Hryggskekkja; Vindblásnar mjaðmir;

Abstract

Introduction: Adult individuals with disability and low motor function frequently have body deformations, such as scoliosis and windswept hips. Posture management is a component of physical management, which aims to prevent or reduce the development of a deformed body shape by maintaining a symmetrical posture.

Aim: The purpose of this thesis is to establish valid and reliable methods of gathering evidence for postural management. This thesis is divided into two parts. The first focuses on the development of two tools to quantify posture, and the second focuses on the association between an asymmetrical posture, low postural ability and deformity.

Method: The validity and reliability of the two tools designed to evaluate posture was tested and the odds of developing deformity caused by a habitual posture was assessed in a cross-sectional study. The clinical tool for posture, the Posture and Postural Ability Scale (PPAS), was evaluated through rating of posture and postural ability from photos and video recordings of 30 adults with cerebral palsy (CP). The tool for posture evaluation, a three-dimensional (3-D) camera mounted on an iPad was evaluated in a comparative study on posture in seven healthy adults by comparing the output from an iPad 3-D model with an optical motion capture system (Qualisys). The PPAS was used along with registry data for 714/830 adults with CP to calculate the relationship between spending long periods of time in an asymmetrical posture, having low postural ability and the development of deformity. Special focus was paid to asymmetrical limited hip flexion, hip and knee contractures, scoliosis and windswept hips.

Results: Both posture evaluation tools (PPAS and a 3-D camera mounted on an iPad) were found to be reliable and valid (weighted Kappa values from 0.85 and intra-class correlation coefficient [ICC] from 0.982). Individuals with asymmetrical limited hip flexion showed higher odds for developing an oblique pelvis (odds ratio [OR] 2.6; 95% confidence interval [CI] 1.6–2.1) and an asymmetrical trunk (OR 2.1; 95% CI 1.1–4.2), scoliosis (OR 3.7; 95%CI 1.3–9.7) and windswept hips (OR 2.6; 95% CI 1.2–5.4), than those who had symmetrical hip flexion. Individuals who were unable to straighten their knees had higher odds of scoliosis (OR 1.8; 95% CI 1.1–3.1) and windswept hips (OR 1.6; 95% CI 1.1–2.3) compared with those who could do so. It was the same for those who were immobile when lying down; this increased the odds of windswept hips (OR 2.9; 95% CI 1.0–3.3) and scoliosis (OR 5.7; 95% CI 3.5–9.1). Similarly, lying only in the supine position increased the odds of having windswept hips (OR 1.9; 95% CI 1.0–3.3) and lying for long periods of time in bed increased the odds for scoliosis (OR 2.9; 95% CI 1.6–2.1).

Conclusion: Our publications on reliability and validity clearly indicate that both postural evaluation tools are suitable for use in clinical practise. Asymmetrical posture along with spending a long time in the same posture increases the odds of developing a deformity among adults with CP.

Keywords:

Posture; Cerebral palsy; PPAS; 3-D model; Adults; Scoliosis; Windswept hips.

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Contents

List of abbreviations

ALHF Asymmetrical limited hip flexion. When there is a difference in the range of hip flexion between the right and the left side.

ANOVA Analysis of variance is a statistic used to analyse the differences among group means in a sample.

CI Confidence interval is a statistic used to estimate the interval, which might hold the true value of an unknown population parameter.

CP Cerebral Palsy. A group of permanent disorders in the development of movement and posture causing limitations in activity that are attributed to non-progressive disturbances occurring in the developing foetal or infant brain.

CPUP Swedish cerebral palsy national surveillance program and quality registry.

GMFCS Gross Motor Function Classification System. A five-level classification that describes the gross motor function of individuals with CP based on self-initiated movement.

OR Odds Ratio. The odds ratio is a statistical measure of the association between the odds of a particular exposure occurring and an outcome, compared with the odds of an outcome occurring in the absence of that exposure.

PPAS Posture and Postural Ability Scale. Its aim is to assess posture and postural ability in individuals with severe physical disabilities, regardless of age.

List of figures

List of tables

List of original papers

I. Rodby-Bousquet, E., Agustsson, A., Jonsdottir, G., Czuba, T., Johansson, A. C., & Hägglund, G. (2014). Inter-rater reliability and construct validity of the Posture and Postural Ability Scale in adults with cerebral palsy in supine, prone, sitting and standing positions. *Clinical Rehabilitation, 28*(1), 82–90.

II. Agustsson, A., Sveinsson, T., & Rodby-Bousquet, E. (2017). The effect of asymmetrical limited hip flexion on seating posture, scoliosis and windswept hip distortion. *Research in Developmental Disabilities, 71*, 18–23.

III. Agustsson, A., Sveinsson, T., Pope, P., & Rodby-Bousquet, E. (2018). Preferred posture in lying and its association with scoliosis and windswept hips in adults with cerebral palsy. *Disability and Rehabilitation, 1–5.*

IV. Agustsson, A., Gislason MK., Ingvarsson P., Rodby-Bousquet E., Sveinsson T. (2019). Validity and reliability of an iPad with a three-dimensional camera for posture imaging. *Gait & Posture*, 68, 357-362

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Declaration of contributions

Below is a declaration of my contribution to each paper on which this thesis is based.

Paper I: Elisabet Rodby-Bousquet designed the study, developed the instrument, collected the data, rated the subjects, analysed the results and wrote the manuscript. I participated in the development of the instrument, rated the subjects, and actively revised and improved the manuscript.

Paper II: I designed the study with the aid of my supervisors Þórarinn Sveinsson and Elisabet Rodby-Bousquet. I prepared and coded the data, analysed the results and performed all statistical analysis with the help of Þórarinn Sveinsson. I wrote the manuscript, revised the manuscript based on contributions by the co-authors and made the final revisions of the manuscript before acceptance and publication.

Paper III: I designed the study with the aid of my supervisors Þórarinn Sveinsson and Elisabet Rodby-Bousquet. I prepared and coded the data, analysed the results and performed all statistical analysis with the help of Þórarinn Sveinsson. I wrote the manuscript, revised the manuscript based on contributions by the co-authors and made the final revisions of the manuscript before acceptance and publication.

Paper IV: I designed the study with the aid of my supervisors Þórarinn Sveinsson and Elisabet Rodby-Bousquet. I developed the instrument with the help of Magnús Gíslason and Þórarinn Sveinsson by combining the usage of premade devices, software packages and wrote a Matlab script. I collected the data, processed the data, analysed the results and performed all statistical analysis with the help of Þórarinn Sveinsson. I wrote the manuscript, revised the manuscript based on contributions by the co-authors and by Pauline Pope, and made the final revisions of the manuscript before acceptance and publication.

1 Introduction

Over 98% of babies are born without any musculoskeletal deformities (Dunn, 1972). In the remaining 2% born with deformities, nine out of ten resolve without any intervention. However, children with a severe and complex neurological disability frequently develop musculoskeletal deformities as early as in their first year (Fulford & Brown, 1976), which can progress throughout life.

An association has been found between the position of a fetus before delivery (prenatal posture) and the position that the infant adopts after birth (postnatal posture) (Dunn, 1974; Hamanishi & Tanaka, 1994; Porter, Michael, & Kirkwood, 2010). The fetus tends to lie with its back toward the mother's left side twice as often as toward her right (Dunn, 1974). When the back of the fetus is directed to the mother's left side, in vertex presentations, the left hip and the left side of the neck rest on the mother's spine, forcing the head and legs over to the right, which corresponds to the baby's supine lying posture after birth. The reverse occurs when the back of the fetus is directed to the mother's right side in the vertex presentation. After birth, this prenatal posture is quite often the infant's favourite or habitual sleeping posture.

Figure 1 Postural asymmetries (donated by Margrét Jónsdóttir)

Many babies born without impairment (Dunn, 1972) initially show some postural asymmetry before they start to sit up (Figure 1). These postural asymmetries decrease and disappear in healthy children, usually by the time they are able to sit independently (Fulford & Brown, 1976). Children with impaired motor and postural development continue to lie in their postnatal posture, usually asymmetrically, leading to established postural asymmetry within the first year of life.

1.1 Developmental biomechanics

In 1892, Julius Wolff published *"The law of transformation of the bone",* which describes the relationship between bone geometry and mechanical influences on bone remodelling (Wolff, 1986). The basis of this law states that any change in function of the tissues of the musculoskeletal system brings about definitive change in the internal and external architecture of the tissue in accordance with the direction and intensity of the new mechanical force acting on the tissue (LeVeau & Bernhardt, 1984). Based on this law, it can be reasoned that the postural asymmetry seen in newborn babies is a consequence of the time spent in their pre- or postnatal habitual posture. With time, the baby's musculoskeletal tissues adapt to this posture: i.e., one side of the joint is stretched while the opposite is relaxed. The deformation is active because the body's renewal process puts down newly synthesised collagen fibrils in the line of the tensile stress. The end result is a longer structure that has adapted to the new situation. The renewal process is also active on the relaxed side of the joint. Without stresses to guide them, the collagen fibrils are laid down in a random manner, with eventual shortening and thickening of the joint structures on the relaxed side (LeVeau & Bernhardt, 1984; Nordin & Frankel, 2012). The net result is permanent change or plastic adaption in the joint capsule, ligaments and tendons leading with time to established contractures and deformities.

The human body is constantly renewing itself, as the need to cut hair and nails regularly demonstrates. The skeletal system turnover is 10% per year in adults (4% cortical and 28% trabecular bone) and up to 100% per year in infants (Manolagas, 2000). Bone renewal includes preservation of mechanical strength by replacing older micro-damaged bone with newer and healthier bone, remodelling according to the stresses and strains within it (Nordin & Frankel, 2012). Collagen has unique characteristics of strength and stiffness. Its renewal rate is relatively fast, because it needs to respond quickly to changes in physical demand or replace damaged collagen. Thus, plastic adaptation occurs because of the human body's renewal processes in response to changing and sustained stresses and strains.

1.2 Contractures

A contracture is the result of plastic adaption that has occurred in the joint's musculoskeletal structures and is defined as the stage where the normal range of movement of the joint is limited (Pope, 2007). It is the established shortening of periarticular connective tissues and muscles (Trudel & Uhthoff, 2000). Lack of movement is the most common cause of joint contracture (Moseley et al., 2005; Nightingale, Moseley, & Herbert, 2007; Trudel & Uhthoff, 2000). This develops within 1 week and progresses in a time-dependent manner (Trudel, Uhthoff, & Brown, 1999). Within 2–4 weeks of immobilization, the joint contracture is largely induced by changes in muscles, tendons and fascia. If the joint immobilization is prolonged, the severity of joint contracture increases, in particular because of changes in the joint capsule, but also in ligaments, bones and cartilages (Chimoto, Hagiwara, Ando, & Itoi, 2007; Trudel, Laneuville, Coletta, Goudreau, & Uhthoff, 2014; Trudel & Uhthoff, 2000; Trudel et al., 1999; Trudel, Uhthoff, Goudreau, & Laneuville, 2014). The normal range of joint movement in the human body is maintained by repeated movements of all parts of the body throughout the day (Tardieu, Lespargot, Tabary, & Bret, 1988). One third of previously healthy individuals admitted to an intensive care unit for 2 weeks or longer, regardless of the reason for admission, experienced joint contracture that interfered with function. (Clavet, Hebert, Fergusson, Doucette, & Trudel, 2008).

The phrase “tissue adaptation” is the stage in which the articular joint and soft tissues feel tight, but a full range of motion is possible with slow passive stretch (Pope, 2007). If tissue adaptation continues, changes in joint musculoskeletal structures will lead to established shortening —contracture— e.g., when short hamstrings (muscle tendon unit) precede the development of knee contractures (Cloodt, Rosenblad, & Rodby-Bousquet, 2018). The increased fibrosis and stiffness of a spastic muscle in individuals with cerebral palsy (CP) could also be a compensatory mechanism to limit further muscle fibre injury in response to stretched sarcomeres (Patel, Das, Friden, Lutz, & Lieber, 2004), suggesting an inability of the affected myofibrils to add sarcomeres in series (Smith, Chambers, & Lieber, 2013).

The normal spine is straight in the frontal plane. Scoliosis refers to abnormal lateral curves of the spine combined with rotation of the vertebrae. There have been many explanations of the origin of scoliosis, such as spasticity, asymmetrical muscle tone, bony abnormalities and poor posture in lying and sitting (Gudjonsdottir & Mercer, 1997; LeVeau & Bernhardt, 1984; Pope, 2007; Roberts & Tsirikos, 2016). From a biomechanical perspective, the development of scoliosis is more readily explained than its cause. Whatever the cause of the scoliosis, the spine is held in a combination of lateral flexion and rotation. Over time, the musculoskeletal structure gradually shortens and thickens on the shorted concave side while the stretched musculoskeletal structure on the convex side lengthens and eventually relaxes as it adapts to the new position. The cartilage of the intervertebral disc degenerates because of excessive compression on the concave side and atrophies on the convex side in response to lack of loading, creating a wedge-shaped appearance. This is a classic description of contracture (Chimoto et al., 2007; Trudel, Laneuville, et al., 2014; Trudel & Uhthoff, 2000; Trudel et al., 1999; Trudel, Uhthoff, et al., 2014). In most cases, therefore, scoliosis can be modelled simply as a contracture of the trunk. Severe scoliosis has been linked to orthopaedic problems, such as hip dislocation, pelvic obliquity, windswept hips distortion, pain and inadequate support in sitting (Letts, Shapiro, Mulder, & Klassen, 1984; Roberts & Tsirikos, 2016). The prevalence of scoliosis is 11% in children with CP (Persson-Bunke, Hägglund, Lauge-Pedersen, Wagner, & Westbom, 2012), and it is slightly higher in adults with CP (Hägglund, Pettersson, Czuba, Persson-Bunke, & Rodby-Bousquet, 2018).

Windswept hips were defined by Lonstein and Beck (Lonstein & Beck, 1986) as “an abduction contractures on one side combined with an adduction contracture on the other”. This definition has been used by other researchers (Morrell, Pearson, & Sauser, 2002; Porter, Michael, & Kirkwood, 2007; Young et al., 1998) until Person-Bunke et al. (Persson-Bunke, Hägglund, & Lauge-Pedersen, 2006) refined it by describing windswept hips as a combination of abduction and external rotation of one hip with the opposite hip in adduction and internal rotation. The aetiology of windswept hips is unknown, though spasticity in the iliopsoas muscles and the hip adductors has been considered the prime candidate for a long time (Letts et al., 1984; Morrell et al., 2002). However, there has been an indication of knee contracture involvement that causes the legs to fall to one side in supine lying (Hägglund, Lauge-Pedersen, Bunke, & Rodby-Bousquet, 2016). With time, the hip joint adapts to this position as the stretched side lengthens and the relaxed side shortens, in response to the body’s renewal process. The side of hip dislocation is usually the adducted hip. The reasons given by some researchers for this adaption is asymmetrical muscle tone and severe spasticity (Letts et al., 1984; Morrell et al., 2002; Young et al., 1998). However, this view of asymmetrical muscle tone and severe spasticity overlooks the changed morphology of the hip joint and its reduced stability caused by changes in the hip joint capsule and its ligaments as a result of abnormal loading (Pope, 2007). The prevalence of windswept hips is up to 18% in persons with CP (Hägglund, Lauge-Pedersen, Bunke, & Rodby-Bousquet, 2016), and will lead to hip sub/dislocation in the severest cases (Letts et al., 1984; Porter et al., 2007).

1.3 Cerebral Palsy

Cerebral palsy (CP) or cerebral paresis, as it was called, was originally described in 1862 by an orthopaedic surgeon, William Little, in England (Dunn, 1995). Little reported contractures and deformities in children after a brain injury during infancy. These deformities were more common in children born pre-term or after complicated births causing asphyxia.

Today, CP is defined as:

> "A group of permanent disorders in the development of movement and posture causing activity limitation that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems" (Rosenbaum et al., 2007).

By definition, the core symptoms of CP are disorders of posture and movement limiting functional activity (Rosenbaum et al., 2007). The classification of CP subtypes currently in use has been adopted internationally and comes from the Surveillance of Cerebral Palsy in Europe (SCPE) (Cans et al., 2000). According to the SCPE, CP can be classified into spastic CP, dyskinetic CP, ataxic CP and non-classifiable CP. Spastic CP can be subdivided into unilateral spastic CP and bilateral spastic CP. Dyskinetic CP can be subdivided into dystonic CP and choreo-athetoid CP. In Europe the inclusion criteria for CP is: "a group of disorders of a permanent but not unchanging non-progressive interference that arises before the age of 2 years, that involves a disorder of movement and/or posture and of motor function" (Smithers-Sheedy et al., 2014). The brain injury can also be caused by different genetic syndromes. All progressive conditions are by definition excluded from CP (Rosenbaum et al., 2007). In the USA, hypotonia is classified as a subtype of CP, but not in Europe (Goldsmith et al., 2016). The Gross Motor Function Classification System – Expanded & Revised (GMFCS) (Figure 2) is used to assess gross motor function. The GMFCS is a five-level classification system that describes the gross motor function of individuals with CP based on their self-initiated movement. The emphasis is on sitting, walking and wheeled mobility. Distinctions between levels are based on functional abilities and the need for assistive technology (Palisano, Rosenbaum, Bartlett, & Livingston, 2008; Palisano et al., 1997). CP is the most common form of childhood disability with a worldwide prevalence of 2.11 individuals per 1000 live births (Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013). In Iceland, the prevalence of CP is 2.29 individuals per 1000 live births and the prevalence increases significantly with decreasing gestational age and birth weight (Sigurdardottir, Thorkelsson, Halldorsdottir, Thorarensen, & Vik, 2009). In more than 80% of the cases, CP is caused by brain lesions or maldevelopments occurring at different time periods in the developing brain. The extent and topography of the brain lesions determine the clinical subtype of CP and are also related to the presence and severity of associated disabilities (Krageloh-Mann & Cans, 2009). The life expectancy of people with CP can be similar to that of the general population, but for those with more severe disabilities, it can be reduced substantially (Hutton, 2006). A 2-year-old child with severe CP has about a 40% (Hutton, 2006) to 60% chance of living to age 20 (Westbom, Bergstrand, Wagner, & Nordmark, 2011), in contrast to a child with mild CP, for whom the chance is 99%.

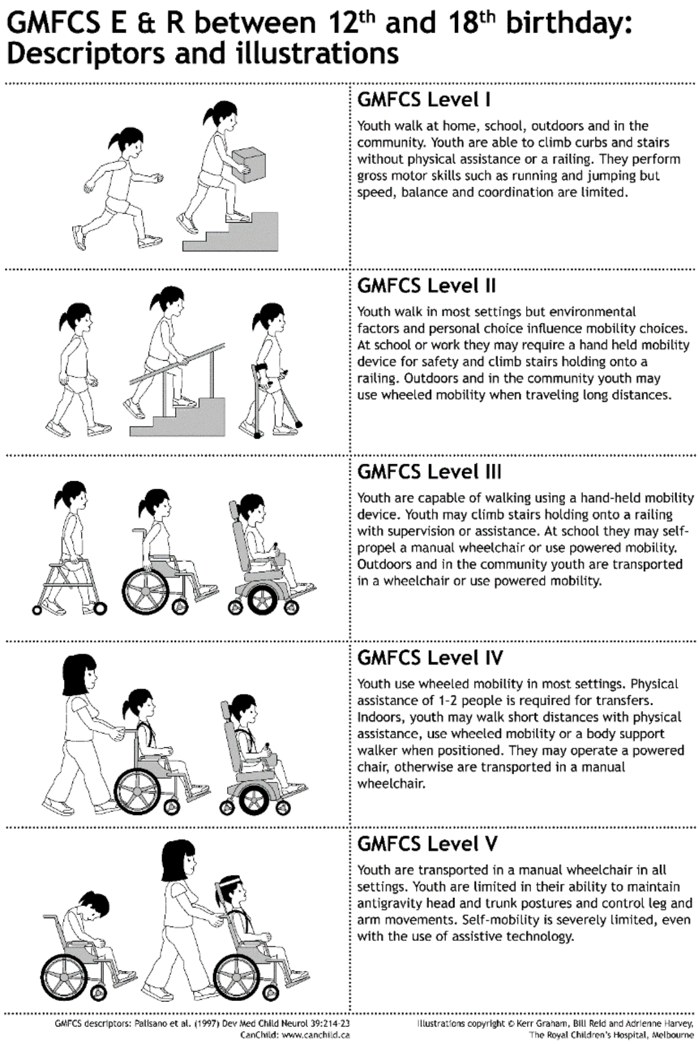

Figure 2 The Gross Motor Function Classification System - Expanded & Revised. Permission to use this illustration has been granted from the copyright holders: Graham, K., Reid, B., and Harvey, A. The Royal Children's Hospital, Melbourne, Australia.

It may be inappropriate to emphasise that the brain lesion in CP is static, without clearly stating that the musculoskeletal pathology will be progressive. The key feature of the CP musculoskeletal pathology is a failure of longitudinal growth of skeletal muscle (Graham & Selber, 2003). The musculoskeletal pathology is much more complex and damaging than simply the development of short muscles and joint contractures. As bone geometry changes due to abnormal stresses (see chapter 1.1), joints become unstable and premature degenerative changes happen in weight-bearing joints. CP involves upper motor neurone lesions that can have two different types of effects on the lower motor neuron pathways (Graham & Selber, 2003). These two types are, on the one hand, loss of inhibition, and on the other hand, loss of excitations. Loss of inhibition causes increased activity in the lower motor neurons and augmented features such as spasticity, hyper–reflexia, clonus and co–contraction. However, loss of excitation will cause decreased activity in the lower motor neurons and attenuated features such as muscle weakness, fatiguability, poor balance and sensory deficits, causing difficulty in moving effectively. Complications that are not a direct consequence of the pathological impairment are secondary complications (Pope, 2007). Those secondary complications are: contracture and deformity (see chapter 1.2), tissue damage, osteoporosis, pain, respiratory- and urinary infection, among others. These complications are largely the result of the inability to move effectively, stabilize posture and change position (Fulford & Brown, 1976; Pope, 2007). There is an association between CP, scoliosis and windswept hips (see chapter 1.2) as its prevalence increase with lesser functional ability of the individual with CP (Hägglund et al., 2018; Letts et al., 1984; Persson-Bunke et al., 2006; Persson-Bunke et al., 2012; Roberts & Tsirikos, 2016; Tsirikos & Spielmann, 2007), although the exact nature of the association is not recognised. Secondary musculoskeletal complications, such as joint contractures, tight muscles, scoliosis, pelvic obliquity, windswept and/or dislocated hips will result in problems with sitting, standing and walking (Agustsson & Jonsdottir, 2018; Letts et al., 1984; Roberts & Tsirikos, 2016). The problems with sitting, standing and walking may lead to asymmetrical posture within each position. Whenever the body or body parts deviate from midline, a gravitational moment is produced, and it compounds the deviation further.

1.4 Follow-up program

The Swedish cerebral palsy national surveillance program and quality registry (CPUP) is a national surveillance program and quality registry for children and adults with CP in Sweden, Norway, Denmark, parts of Scotland (Alriksson-Schmidt et al., 2017) and Iceland. The surveillance program began in 1994 in southern Sweden. The aim was to prevent hip dislocation and contractures by a standardized follow up of children's hips and spines from an early age and to initiate intervention if deterioration was found. CPUP has evolved since 1994; therefore, more than 95% of all children with CP in Sweden are now enrolled and participate in CPUP. In 2009, the program was expanded to include adults with CP. Further information is available on the CPUP web site (see www.cpup.se). CPUP is now a health-care program that "includes a continuing standardized follow-up with assessment of gross and fine motor functions, mobility, joint ranges of motion, clinical findings and treatments" (Rodby-Bousquet, 2012).

Inclusion criteria to participate in the Swedish CPUP registry is the diagnosis of CP according to Rosenbaum et al. (Rosenbaum et al., 2007). Inclusion and exclusion criteria are in accordance with the SCPE network (SCPE, 2000) with inclusion of all permanent but not unchanging disorder of movement, posture and of motor function due to a non-progressive brain injury before the age of 2 years. Individuals who do not fulfil the criteria of CP, with similar conditions manifested after the age of 2 years or with progressive brain injuries do not participate in CPUP. Adults are followed based on their GMFCS level, where those classified at GMFCS I are invited for examination every third year, GMFCS level II every second year and GMFCS levels III-IV every year.

1.5 Postural management

"Physical management" is a comprehensive plan for managing the physical status of severely disabled individuals (Pope, 1997b). It is a term used to describe the maintenance of "optimum physical condition" of an individual with a severe and complex neurological disability through postural management, physiotherapy, handling techniques and attention to nutrition. The objective of management of posture is to prevent or minimize secondary complications. It is a major component in a 24-hour approach encompassing all activities and interventions that impact on a person's posture and function by supporting the body in a symmetrical but comfortable posture. It might include a wheelchair with a custom-moulded seating system, support while standing and during the night, active exercise, orthotics, medical and surgical interventions, and individual therapy sessions (Gericke, 2006; Pope, 2007). Procedures are integrated into daily routine and incorporated into everyday life to improve individuals' quality of life and participation in society (Pope, 2007). Complications that are not a direct consequence of the pathological impairment are secondary complications, such as those arising from the motor and postural deficits (Pope, 2007). Contractures and deformity of the body are classic examples of secondary complications, which are largely the result of immobility: i.e., the individual's inability to stabilize posture and change position.

Actions taken to prevent the development of a secondary complication, or to correct one that has already occurred, usually involves support and time spent in corrected postures. These actions can either be a set of postures to be used during the day or "counter-strategies" used to counteract the deleterious effects of a posture that cannot be changed without compromising function or participation in the community (Pope, 2007). Management of posture is a team effort. For example, individuals with tight muscles, deformities or increased spasticity may need medical interventions such as botulinum toxin injections, baclofen pumps or corrective surgeries to aid the use of orthotics or adaptive support, in order for the individual to tolerate time spent in non-harmful positions. Neither the medical- or the non-invasive intervention would on its own have a long-term effect. Prevention of secondary complications is expected to improve the quality of life and maintain maximum functional ability. In addition, it is known that every secondary complication prevented is a cost-saving initiative and will reduce the effort of care (Bail et al., 2015).

1.6 Measurements of posture

Posture management has been practised for decades, from treating polio patient in the 1950s (Gossman, Sahrmann, & Rose, 1982) and children with a severe disabilities in the 1970s (Pope, 2007), to quite sophisticated and expensive sleeping system, used today. It was learned the hard way, that articular joints of polio patients needed to be in a zero position, to minimize contracture development (Gossman et al., 1982). But there is no quantitative evidence supporting the use of those expensive sleeping systems. One reason for this lack of evidence is the difficulty to do a full body measurement in a standardized way and lack of valid and reliable assessment tools and outcome measures.

The natural development of individuals with CP is known, which is decline in mobility and function, increasing pain and fatigue, and severe postural deformities (Letts et al., 1984; Saito, Ebara, Ohotsuka, Kumeta, & Takaoka, 1998; Tosi, Maher, Moore, Goldstein, & Aisen, 2009). The classic appearance of the preferred posture of an individual with CP at GMFCS level V in supine lying is: head is out of midline (rotated and laterally flexed to the same side), trunk asymmetrical (scoliosis) and hips windswept (Agustsson & Jonsdottir, 2018; Fulford & Brown, 1976; Letts et al., 1984; Porter et al., 2007). There are three different types of the orientation of the shoulders. The first type has the shoulders abducted and externally rotated. The second has the shoulders adducted and internally rotated, and the third type is a combination of the two former ones, i.e. with one shoulder in abduction and external rotation while the other is in adduction and internal rotation. In almost all cases, the elbows are flexed, and wrist and fingers are often flexed too. The hips are flexed with a combination of abduction and external rotation on one side while the other is in adduction and internal rotation. The knees are flexed and the ankles plantarflexed.

There is no universal definition of posture (Rodby Bousquet, 2012). When evaluating posture, it is practical to define it as the relative position of body segments with respect to an external reference frame (Hadders-Algra & Brogren-Carlberg, 2008). Postural ability is the ability to perform efficient movements in an energy conserving way without damaging the body in the process (Bouisset & Do, 2008; Pope, 2007; Rodby Bousquet, 2012). The Posture and Postural Ability Scale (PPAS) was modified between 2009 and 2011 under the supervision of Pauline Pope. The PPAS originated from the Postural Ability Scale (PAS) developed by Pauline Pope in the early 1990’s (Pope, 2007). The basis of the scale can be traced back to the Physical Ability Scale for assessment of postural ability in children with severe disability originally developed by Noreen Hare in the 1970s. The aim of the PPAS is to assess posture and postural abilities in individuals with severe physical disabilities, regardless of age or diagnosis. The PPAS evaluates the symmetry and alignment of posture and postural ability in standing, sitting and lying, both supine and prone. It has not been previously tested for reliability or validity.

To determine the effect of postural intervention, there is a need to measure body posture quantitatively. The PPAS identifies the presence of postural asymmetries or deviations but does not specify the magnitude. The preferred posture of an individual with severe and complex neurological disability is often so complex and deformed that it is difficult to measure with a goniometer. The difficulties relate to the inability to accurately distinguish between the movements in the cardinal planes, flexion/extension, abduction/adduction and internal/external rotation, needed to describe the orientation of the body part in question, in degrees. Three-dimensional (3-D) motion capture systems exist which describe the direction of extremities in degrees, with the help of markers placed on anatomical landmarks (Fortin, Feldman, Cheriet, & Labelle, 2011). These systems are complex, requiring a special laboratory and an experienced technician for operation; thus, they are expensive and not handy for usage in a clinical environment. An inexpensive two-camera system (Sato, Kondo, Ojima, Fukasawa, & Higuchi, 2017) for 3-D posture capture has been used for quantitative measurements of deformation of the trunk, but it is complicated to operate and too time consuming for use in a clinical setting. In addition, systems such as these require a dedicated space in the clinic, as once the cameras are calibrated they cannot be moved or changed without ruining the calibration (Cohen, Bravi, & Minciacchi, 2017). In a fixed two-camera system, the individual must be placed so that both cameras can image all the markers used for postural evaluation simultaneously. A movable two-camera system would require a complicated process of measurements and calculations of direct linear transformation parameters, which are needed for calibration purposes of the system (Abdel-Aziz & Karara, 2015). The calibrated volume of a two-camera system is so small that it can only capture a part of the body each time, rather than the full body 3-D posture required for clinical use.

1.7 Significance of this thesis

Despite advances in conventional physiotherapy and occupational therapy, better orthosis and medical science, vulnerable individuals are still developing contracture and deformity (Clavet et al., 2008; Offenbacher et al., 2014). Even though it has been known for decades that a zero joint position is crucial for physical recovery after polio to minimize contracture development (Gossman et al., 1982) and that tissue adaptation theories support physical management, there is a considerable lack of robust evidence to support postural management in clinical settings.

High technology postural intervention is often costly and the demand for evidence-based practice is growing to justify the funding of such intervention. Evidence-based practice is a clinical practice where medical treatment or intervention is based on the best available clinical experience, high-quality research and clinical trials. High-quality studies require valid and reliable instruments for objective and quantifiable measurements in these trials, which are currently not available for measuring posture. This thesis presents and tests the reliability and validity of two instruments for measuring posture that are simple to use in the clinic. It will facilitate further high-quality studies of the effect of treatments on posture in the disabled population and provide information for evidence-based practice.

The aim of postural intervention is preventing or reducing secondary complications and facilitate function. Scoliosis and windswept hips are common secondary complications affecting individuals with complex neurological disabilities. In chapter 1.1, “Developmental biomechanics”, the combined effects of forces and the body’s renewal process on the progression of trunk asymmetry of an infant spending too long a time in one position, are discussed. The prenatal position becomes the postnatal position (Dunn, 1974; Hamanishi & Tanaka, 1994; Porter et al., 2010), which is associated with established deformity later in adult life (Porter, Michael, & Kirkwood, 2008; Porter et al., 2010). There are indications that knee contractures are involved in the development of windswept hips deformity (Hägglund et al., 2016). Time and immobility are the major factors in the development of contractures in general (Clavet et al., 2008). It is not known which type of posture, position or limb contracture enhance the development and progression of scoliosis and windswept hips deformity. Therefore, it is not known which postural intervention is appropriate for counter-measures. This thesis provides evidence for the association between posture and position on the one hand, and between scoliosis and windswept on the other, using data from adults with CP. It provides further information for evidence-based practice in the management of people with motor impairments such as in those with CP.

2 Aims

The overall aim of the thesis is to provide means by which evidence is gathered to support postural management theories in the prevention of deformity in individuals with motor impairments, such as CP. This was achieved by two separate projects: 1) establishing the validity and reliability of an existing tool designed to evaluate posture and postural ability (Paper I), and the use of a 3-D model for quantitatively measuring posture in clinical settings (Paper IV); and 2) to use data from adults with CP to shed light on the observation that individuals who have low postural ability and spend long periods in one position have more deformities than those who are more able and have better postural ability (Papers II and III).

The specific aims for each Paper were as follows:

Paper I evaluated the PPAS for adults with CP in terms of:

- Inter-rater reliability
- Internal consistency
- Construct validity

Paper II determined the prevalence of asymmetrical limited hip flexion of <90° in individuals with CP and evaluated the association between asymmetrical limited hip flexion of <90° with:

- Asymmetrical seating posture
- Occurrence of scoliosis
- Occurrence of windswept hip deformity

Paper III examined the association of scoliosis and windswept hips in adults with CP to:

- Immobility
- Lying position
- Time spent lying down

Paper IV evaluated the use of an iPad with a 3-D camera to assess posture and postural deformities in a clinical environment in terms of:

- Inter-rater reliability
- Intra-rater reliability
- Criterion-related validity

3 Materials and methods

3.1 Design and participants

Paper I evaluated the properties of the PPAS for adults with CP from photos and video recordings of individuals with CP at GMFCS levels I–V. Papers II and III were cross-sectional studies of adults with CP, reported to the CPUP, focusing on the effect of asymmetrical limited hip flexion (Paper II) and the preferred/habitual position in lying (Paper III) on the development of deformity. Paper IV compared two systems used to produce 3-D models for quantifying lying posture.

Participants (Table 1) in Paper I were recruited in October 2009 to October 2010. In total, 30 adults were recruited: 29 in Sweden and one from the Rehabilitation Centre of Excellence in Kópavogur, Iceland. Written consent was collected from all participants or by proxy where the participant was unable to give such consent. The details of participants in studies of Papers II and III were extracted from the most recent reports for all adults with CP registered with CPUP between 1 January 2013 and 31 December 2015 (Paper II) and 31 December 2016 (Paper III). The participants in Paper IV were seven healthy adult volunteers, recruited among colleagues and friends.

Table 1. Participants from Papers I–IV

Participants	Paper I	Paper II	Paper III	Paper IV
Number	30	714	830	7
Female /Male	15/15	357/357	361/469	4/3
Age (years)	19–22	16–73	16–73	19–57
GMFCS I	6	159	159	
GMFCS II	6	150	185	
GMFCS III	6	114	130	
GMFCS IV	6	121	155	
GMFCS V	6	170	201	
Unilateral spastic CP	–	156	169	
Bilateral spastic CP	–	385	461	
Dyskinetic CP	–	92	29	
Ataxic CP	–	25	103	
Unclassified/mixed CP	–	42	69	

GMFCS: Gross motor function classification system; CP: Cerebral palsy.

3.2 Data Collection

For Paper I, posture and postural ability was rated from photographs and video-recordings by three experienced physiotherapists independently, corresponding to the points on the PPAS (Table 2–4).

For Papers II and III, anonymised data were extracted from the CPUP register after permission from the registry holder, and transferred into an excel file, based on the latest report for adults with CP in Sweden. All assessments were performed by local physiotherapists and/or occupational therapists in a standardised manner employing a CPUP assessment form and an accompanying manual (www.cpup.se). Data collected from the CPUP database were: gender, age, GMFCS level, CP subtypes, PPAS scores, scoliosis and hip range of movement. For Paper III, additional data for knee extension, lying position and time spent lying down were also collected from the CPUP database. All variables were coded, and separate variables of limited hip flexion and windswept hips were calculated (see section 3.3.5 and 3.3.6) based on passive range of hip motion. Anonymised data was transferred from an excel file into a statistical software.

For Paper IV, the orientations of markers in global space were simultaneously recorded using the Qualisys motion analysis system and a 3-D camera mounted on an iPad.

3.3 Classifications and measurements

CP was defined according to Rosenbaum et al. (Rosenbaum et al., 2007). Inclusion and exclusion criteria were in accordance with the SCPE network (SCPE, 2000) with inclusion of all permanent but not unchanging disorder of movement, posture and of motor function due to a non-progressive brain injury before the age of 2 years, exclusion of similar conditions manifested after the age of 2 years and progressive brain injuries. Gross motor function was assessed by local physiotherapists, according to the expanded and revised version of the GMFCS (Palisano et al., 2008) (Papers I–III).

Table 2. Symmetry and alignment of posture: frontal view.

Quality of posture, frontal view, (Yes = 1 point, No = 0 points)			
Standing	**Sitting**	**Supine**	**Prone**
Head midline	Head midline	Head midline	Head to one side
Trunk symmetrical	Trunk symmetrical	Trunk symmetrical	Trunk symmetrical
Pelvis neutral	Pelvis neutral	Pelvis neutral	Pelvis neutral
Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis
Arms resting by side	Arms resting by side	Arms resting by side	Arms resting (elevated, mid-position)
Weight evenly distributed	Weight evenly distributed	Weight evenly distributed	Weight evenly distributed

Table 3. Symmetry and alignment of posture: sagittal view

Quality of posture, sagittal view, (Yes = 1 point, No = 0 points)			
Standing	**Sitting**	**Supine**	**Prone**
Head midline	Head midline	Head midline	Trunk in neutral position
Trunk in neutral position	Trunk in neutral position	Trunk in neutral position	Pelvis neutral
Pelvis neutral	Pelvis neutral	Pelvis neutral	Hips extended
Legs straight, hips/knees extended	Hips mid-position (90°)	Legs straight, hips/knees extended	Knees extended
Feet mid-position/flat on floor	Knees mid-position (90°)	Feet resting in normal position	Arms resting (elevated, mid-position)
Weight evenly distributed	Feet mid-position/flat on floor	Weight evenly distributed	Weight evenly distributed

Table 4. Postural ability

	PPAS Levels of postural ability
Level 1	Unplaceable in an aligned posture
Level 2	Placeable in an aligned posture but needs support
Level 3	Able to maintain position when placed but cannot move
Level 4	Able to initiate flexion/extension of trunk
Level 5	Able to transfer weight laterally and regain posture
Level 6	Able to move out of position
Level 7	Able to move into and out of position

3.3.1 Inclusion and exclusion criteria

Paper I – young adults with CP born 1988-1991 were recruited in Sweden and Iceland, from October 2009 to October 2010 until six at each GMFCS-level were included.

Paper II – all adults with CP at all GMFCS-levels and neurological subtypes reported into the Swedish CPUP from 2013 to 2015 were included. Only the last assessment of each individual during this time period was included in the analyses. Exclusion criteria were adults reported before 2013 and individuals under the age of 16 years.

Paper III – all adults with CP at all GMFCS-levels and neurological subtypes reported into the Swedish CPUP from 2013 to 2016 were included. Only the last assessment of each individual during this time period was included in the analyses. Exclusion criteria were adults reported before 2013 and individuals under the age of 16 years.

Paper IV – Inclusion criteria were healthy adults' volunteers, recruited among colleagues and friends in Iceland.

3.3.2 The Gross Motor Function Classification System (GMFCS)

Gross motor function was assessed by local physiotherapists, according to the expanded and revised version of the GMFCS (Palisano et al., 2008).

Paper I – GMFCS level was assessed by local physiotherapists in Sweden and Iceland.

Paper II and III – Data on GMFCS level was extracted from the Swedish CPUP registry.

3.3.3 Posture and Postural Ability Scale (PPAS)

Paper I – The evaluation of the PPAS was based on ratings from photographs and video recordings of 30 adult individuals with CP (six at each GMFCS level). The video recordings were of adults getting into and out of supine, prone, sitting on a plinth, and into and out of standing positions. The same adults were photographed in frontal and sagittal planes when supine and lying prone, sitting on a plinth with feet on the floor, and when standing. The instruction given to the individual for the photographs, was to lie, sit or stand as straight as possible. If unable to do so they were placed in the position. Time to relax and settle into the position was allowed. Arms were relaxed by the side, except when lying prone, where the head was to the side and arms in an elevated position (90° abduction and external rotation in the shoulder joint and 90° flexion in the elbow). If support was needed to get into a position or maintain a position, it was provided, and noted. In the studies of Papers II and III, the PPAS was rated according to the guidelines of the CPUP manual (www.cpup.se). For assessment of posture, there are six items in the frontal plane (Table 2) and six items in the sagittal plane (Table 3). Symmetry and alignment scores one point while asymmetry or deviation from midline scores 0 points. For assessment of postural ability (Table 4), PPAS uses a seven-point ordinal scale. Levels one and two indicate no postural ability: thus, level one defines an inability to be placed in a symmetrical alignment and level two defines an ability to be placed in symmetrical alignment but needing support to hold the position. Level three indicates an ability to maintain a position when placed and level four indicates a change of position. Levels five to seven indicate the ability to move within a given position or change position independently. It is possible to have a subject with a posture score of 0, but with a much higher postural ability score: thus, the alignment score might be 0 but the subject still has some ability to move within a position and change it.

Paper II – Data on three items of PPAS sitting posture rating symmetry and alignment in a frontal plane was extracted from the CPUP registry. The items were trunk symmetrical, pelvis neutral and weight distribution frontal plane (Roberts & Tsirikos, 2016; Tsirikos & Spielmann, 2007). A score of one indicates symmetrical trunk, neutral pelvis or even weight distribution whereas zero indicates asymmetrical trunk, oblique pelvis or uneven weight distribution. All assessments were performed in a standardised manner by local physiotherapists and occupational therapists throughout Sweden. Individuals missing data on trunk symmetry pelvis position or weight distribution were excluded from statistical analysis regarding symmetry and alignment of posture.

Paper III – Data on PPAS level of postural ability in supine lying was extracted from the CPUP registry. Individuals at PPAS supine levels one to three have no ability to change position on their own when placed in supine lying and where therefore defined as immobile in lying. Individuals at level 4–7 have the ability to move within a position and ultimately, change position. All assessments were performed in a standardised manner by local physiotherapists and occupational therapists throughout Sweden. Individuals missing postural ability in supine lying data were excluded from statistical analysis regarding postural ability.

3.3.4 Passive Joint Ranges of Motion

The passive joint ranges of motion were measured using a goniometer in a standardised supine position according to the CPUP manual. All assessments were performed in a standardised manner by local physiotherapists and occupational therapists throughout Sweden.

Paper II – From the joint range of motion section in the CPUP register, data on hip flexion, hip abduction, and hip external and internal rotation was extracted. Asymmetrical limited hip flexion was defined as a passive range of hip movement less than 90° of flexion and was reduced by at least 5° compared with the contralateral side. Individuals missing hip flexion data were excluded from statistical analysis regarding limited hip flexion. The selection of 5° difference was based on results from Boone et al. (Boone et al., 1978). The data on hip abduction, and hip external and internal rotation was used for windswept hips calculation (see chapter 3.3.5).

Paper III – From the joint range of motion section in the CPUP register, data on hip extension, knee extension, hip abduction, and hip external and internal rotation was extracted. Lack of hip and knee extension was defined as inability to passively straighten hips and knees to zero degrees. Individuals missing hip extension or knee extension data were excluded from statistical analysis regarding lack of hip extension or lack of knee extension. The data on hip abduction, and hip external and internal rotation was used for windswept hips calculation (see chapter 3.3.5)

3.3.5 Windswept hips

Radiographic data of the hips are not available for the majority of the adults reported into CPUP. Windswept hips were defined by the range of hip movement measurement (Papers II and III). This was defined as at least 50% difference in abduction, internal and/or external rotation between the hips (Persson-Bunke et al., 2006). The direction of windswept distortion was defined using a modified version of Porters et al. method (Porter et al., 2007) and was defined as the sum of abduction, internal and external rotation angles of the right hip, divided by those of the left hip. The Porters et al. method, which is a modification of the Young et al. (Young et al., 1998) method, used the sum of abduction and adduction on one side divided by the sum of the other side. Hip adduction is not recorded in the CPUP registry, so internal and external rotation angles were used instead, as they were properties of the Young et al. original method. The threshold value was set at 20°, which is a 10° increase from the Porters et al method due to increase in the potential margin of error in measurements of abduction, internal and external rotation angles. A positive value of 20° or more indicated a windswept distortion to the right and a negative value of –20° or lower indicated a windswept distortion to the left. Individuals missing hip abduction, and hip external and internal rotation data were excluded from statistical analysis regarding windswept hips.

3.3.6 Scoliosis

Radiographic data of the spine and Cobb-angles for scoliosis are not available for the majority of the adults reported into CPUP. In CPUP a clinical examination of the spine is used to classify scoliosis. Scoliosis is graded as either severe, moderate, mild or no scoliosis. Severe scoliosis is defined as a scoliosis that requires a lateral support in upright sitting or standing. Moderate scoliosis is defined as a scoliosis that is visible both in forward bending and in upright sitting. Mild scoliosis is defined as a discrete curve only visible at forward bending of the spine. Severe and moderate scoliosis has a high sensitivity and specificity compared to a radiographic Cobb angle of at least 20 degrees. (Persson-Bunke, Czuba, Hägglund, & Rodby-Bousquet, 2015).

Paper II – From the scoliosis section in the CPUP registry, data on scoliosis (yes/no), severity of scoliosis (mild, moderate or severe), scoliosis surgery (yes/no), and the convex direction of their scoliotic curves, (thoracic convex right, thoracic convex left, thoracolumbal convex right, thoracolumbal convex left, lumbal convex right or lumbal convex left) was extracted. Moderate or severe scoliosis were treated as scoliosis and mild scoliosis was treated as no scoliosis. Individuals with missing data about having scoliosis, scoliosis surgery, severity of scoliosis or on the convex direction of their scoliotic curves were excluded from statistical analysis regarding scoliosis.

Paper III – From the scoliosis section in the CPUP registry, data on scoliosis (yes/no) and severity of scoliosis, was extracted. Moderate or severe scoliosis were treated as scoliosis and mild scoliosis was treated as no scoliosis. Individuals missing data about having scoliosis or severity of scoliosis were excluded from statistical analysis regarding scoliosis.

3.3.7 Lying

Paper III – From the lying section in the CPUP registry, data on lying position (supine, prone and sides) and time spent in lying, were extracted. Individuals missing data about lying position or time spent in lying were excluded from statistical analysis regarding lying position and time spent in lying.

3.3.8 Local frame of reference

Paper IV – The markers used to identify each anatomical landmark were Qualisys (Göteborg, Sweden) super-spherical beads, 8 mm in diameter. The markers were used to highlight the following anatomical landmarks on both the right and left sides: the coracoid processes, the costal margin around the 10th rib and the anterior superior iliac spine. Markers on the left leg were placed on the greater trochanter, the lateral and medial epicondyle of the femur, and the lateral and medial malleolus. Trunk symmetry measures (Pope, 2007) for quantifying scoliosis, by measuring distance, vertically and diagonally, between the coracoid processes and the ASIS. Local frames of reference were the upper trunk, the lower trunk, pelvis, thigh and shank. The 3–D joint angles were calculated between the upper and lower trunk, lower trunk and pelvis (Sato et al., 2017) and between thigh and shank, as the relative 3-D rotation between interconnecting local frames in global space.

3.3.9 Three-dimensional instrumentation

Paper IV – The position of the markers in the global space (X Y and Z coordinates) were recorded simultaneously by two systems, a Qualisys motion analysis system (Qualisys AB, Sweden) and an iPad system. As a gold standard for validation, an 8-camera three-dimensional Qualisys Oqus 300 motion capture system (Qualisys AB, Sweden) was used to record the markers position. A computer software, Qualisys Track Manager (QTM), digitised the markers for the Qualisys system semi automatically. The iPad system consists of a 3-D camera (Structure Sensor, Occipital Inc, USA), integrated with a 4th generation version of iPad (Apple, USA), an iPad app (Structure, Occipital Inc, USA) and two computer software’s (Skanect (Occipital Inc, USA) and Cloud Compare (CloudCompare v2.9.1 software, http://www.cloudcompare.org). The CloudCompare software was used to manually digitise the markers’ position for the iPad system. A Matlab script (Version R2017b; Mathworks, Inc, USA) defined one set of local frames of reference (upper trunk, lower trunk, pelvis, thigh and shank) from each of the two systems, using the position of markers in the global space, recorded by each system. The output from the Matlab scripts were thus two sets of trunk symmetry length measurements and two sets of angles between the local frames. Both sets were calculated using the same algorithm, using data from the two systems, i.e. the Qualisys QTM and the iPad/CloudCompare data.

3.4 Statistics

In Paper I, the R software environment (The R Foundation for Statistical Computing, https://www.r-project.org) was used in the statistical analysis. The significance level was set at $p < 0.05$ and 95% confidence intervals (95% CI) were used.

- Inter-rater reliability was calculated using weighted Kappa scores.
- Internal consistency was evaluated using Cronbach's alpha.
- For analysis of reliability and consistency, 95% non-parametric bootstrap CIs were generated based on 1000 re-samples of all GMFCS levels combined.
- The construct validity was evaluated by the rank-based non-parametric Jonckheere–Terpstra test, by comparing the trend between PPAS scores and GMFCS levels (scores).

In Paper II, SAS software (version 9.4; SAS Institute Inc., Cary, NC, USA) was used in the statistical analysis. The significance level was set at $p < 0.05$.

- The chi-squared (χ^2) test was used to analyse relationship between variables.
- Logistic regression was used to estimate the association between variables to estimate odds ratios (ORs).

In Paper III, SAS software was used in the statistical analysis. The significance level was set at $p < 0.05$.

- Spearman's correlation coefficient (r_s) values were calculated in analysing relationships between variables.
- Logistic regression analysis was used to investigate associations between variables as ORs.

In Paper IV, SAS software was used to evaluate criterion-related validity, and inter-rater and intra-rater reliability in the statistical analysis. The significance level was set at $p < 0.05$.

- Limits of agreement plots were used to evaluate any bias between the mean differences of the measurements and to estimate the 95% agreement interval.
- Mixed-model analysis of variance (ANOVA) was used to evaluate the effects of the mean differences of the measurements: i.e., systematic errors and their significance levels.
- Intra-class correlation coefficients (ICC(2,1) and ICC(3,1) (McGraw & Wong, 1996), and the standard error of measurement (SEM) were calculated from the mixed-model ANOVA.

In Paper IV, the intra–rater reliability evaluation was based on data from three repeated digitises of the markers' position using the CloudCompare software by the same physiotherapist. Inter–rater reliability analysis was based on digitised markers' positions done by four experienced physiotherapists.

3.5 Ethical considerations

Ethical approvals for Paper I was granted by the Medical Research Ethics Committee at Lund University (number 2009/361), for Papers II and III was granted by the Medical Research Ethics Committee at Lund University (LU 2009-341), and permission was obtained to extract data from the CPUP registry. All participants consented to research based on the reported data. No individual details are presented. As individuals in Paper IV were adult volunteers over the age of 18 years, no ethical approval was needed.

4 Results

4.1 Paper I

The PPAS showed an excellent inter-rater reliability for three independent raters with weighted Kappa values of 0.85–0.99. The internal consistency was high for all items with Cronbach's alpha of 0.96–0.97 (95% CI 0.93–0.98), and item-total correlation of 0.60–0.91. The rank-based non-parametric Jonckheere–Terpstra test showed construct validity, based on the PPAS ability to distinguish between the GMFCS levels (scores). Median values and range in terms of min and max values are presented together with p-values ($p < 0.02$) in Table 5. The PPAS could detect postural asymmetries at all GMFCS levels but did not distinguish postural ability between individuals at GMFCS levels I–II. The postural asymmetries for sagittal view in sitting had the lowest correlation between the three independent raters.

Table 5. Construct validity of the PPAS. Median values and range for GMFCS levels I–V and p-values of averaged values for the three raters.

		GMFCS										
		I		II		III		IV		V		P
Supine	Postural ability	7	(7–7)	7	(7–7)	7	(6–7)	4	(3–7)	1.5	(1–4)	<0.001
	Posture frontal	6	(2–6)	4	(2–5)	1	(0–6)	0	(0–1)	0	(0–1)	<0.001
	Posture sagittal	6	(4–6)	4	(1–6)	2.5	(0–6)	0.5	(0–3)	1	(0–3)	<0.001
Prone	Postural ability	7	(7–7)	7	(7–7)	6	(5–7)	4	(1–6)	1	(1–3)	<0.001
	Posture frontal	5	(2–6)	4	(2–5)	2	(0–5)	1	(0–3)	0	(0–3)	<0.001
	Posture sagittal	6	(2–6)	5	(2–6)	3	(0–6)	1	(0–4)	0	(0–4)	<0.001
Sitting	Postural ability	7	(7–7)	7	(7–7)	7	(2–7)	2	(2–6)	2	(1–2)	<0.001
	Posture frontal	6	(4–6)	4	(1–6)	3	(0–6)	0	(0–2)	0	(0–4)	<0.001
	Posture sagittal	3.5	(2–6)	2	(0–5)	3.5	(0–6)	0	(0–4)	1	(0–5)	0.019
Standing	Postural ability	7	(7–7)	7	(7–7)	4	(1–7)	1.5	(1–2)	1	(1–2)	<0.001
	Posture frontal	6	(3–6)	3	(0–5)	0	(0–3)	0	(0–2)	0	(0–2)	<0.001
	Posture sagittal	5	(4–6)	2	(0–6)	1	(0–4)	0	(0–3)	0	(0–3)	<0.001

4.2 Paper II

There were 63 (9%) individuals with CP, who had an asymmetrical limited hip flexion (ALHF): 93 (13%) presented with scoliosis and 163 (25%) with windswept hips. Most of the individuals missing hip flexion data (49) and data for windswept hip calculations (378), were at GMFCS level V (Table 6). Individuals with ALHF <90° had higher odds of an oblique pelvis, an asymmetrical trunk, scoliosis, and windswept hips (Table 7) compared with those who had bilateral hip flexion >=90°. No association was found between the side of limited hip flexion and either the direction of convexity of the lowest scoliotic curve (p = 0.808) or the windswept hips (p = 0.273).

Table 6. Numbers and percentages of deformities of the 714 individuals according to GMFCS levels. Deformities: asymmetrical limited hip flexion, and items from the Posture and Postural Ability Scale (trunk, pelvis and weight distribution) in sitting, scoliosis, and windswept hip deformity as well as complete data for each variable.

		Gross Motor Function Classification System Level					
	Complete data set N	I n = 159	II n = 150	III n = 114	IV n = 121	V n = 170	Total 714
ALHF < 90°[a]	665/714 (93%)	2/151 (1%)	5/141 (4%)	7/111 (6%)	11/113 (10%)	38 /150 (25%)	63/714 (9%)
Asymmetrical trunk	672/714 (94%)	24/159 (15%)	31/148 (21%)	41/114 (34%)	60/115 (52%)	96/136 (71%)	252/714 (35%)
Oblique pelvis	672/714 (94%)	24/159 (15%)	31/148 (21%)	41/114 (34%)	60/115 (52%)	96/136 (71%)	252/714 (35%)
Uneven weight	661/714 (93%)	15/153 (10%)	32/147 (22%)	39/112 (35%)	64/114 (56%)	99/135 (73%)	249/714 (35%)
Scoliosis	286/714 (40%)	2/46 (4%)	5/52 (10%)	10/60 (17%)	16/50 (32%)	60/75 (80%)	93/714 (13%)
Convex right		1/2 (50%)	3/5 (60%)	5/10 (50%)	6/16 (38%)	34/60 (57%)	49/714 (7%)
Convex left		1/2 (50%)	2/5 (40%)	5/10 (50%)	10/16 (62%)	26/60 (43%)	44/714 (6%)
Windswept hips	336/714 (47%)	14/36 (39%)	31/64 (48%)	26/76 (34%)	32/81 (40%)	60/70 (82%)	163/714 (23%)
Right direction		9/14 (64%)	17/31 (55%)	12/26 (46%)	17/32 (53%)	36/60 (60%)	91/714 (13%)
Left direction		5/14 (36%)	14/31 (45%)	14/26 (54%)	15/32 (47%)	24/60 (40%)	72/194 (10%)

a Asymmetrical limited hip flexion (ALHF < 90°)

Table 7. Odds Ratio (OR) for effects from asymmetrical limited hip flexion by the Gross Motor Function Classification System (GMFCS) scores on items from PPAS (trunk, pelvis, and weight) in sitting, scoliosis, and windswept hip deformity.

Dependent variable	GMFCS	Asymmetrical limited hip flexion	Asymmetrical limited hip flexion adjusted for GMFCS
Asymmetrical trunk	1.9 (1.8–2.4)[a]	4.5 (2.4–8.4)[a]	2.1 (1.1–4.2)[a]
Oblique pelvis	1.9 (1.7–2.2)[a]	4.9 (2.7–9.1)[a]	2.6 (1.6–2.1)[a]
Uneven weight distribution	2.2 (1.9–2.5)	3.5 (2.0–6.3)	1.5 (0.8–2.9)
Scoliosis	2.8 (2.2–3.7)[a]	7.4 (3.1–17.6)[a]	3.7 (1.3–9.7)[a]
Windswept hips	1.5 (1.3–1.8)[a]	3.5 (1.7–7.1)[a]	2.6 (1.2–5.4)[a]

The interaction between the GMFCS score and asymmetrical limited hip flexion was not significant. a Significant predictor $p < 0.05$

4.3 Paper III

Of the 830 adults with CP who participated in the study, 228 (27%), lay solely in one position during the night; 135 (16%) were immobile in that position; 215 (26%) were unable to actively or passively straighten their hips; and 445 (54%) were unable to actively or passively straighten their knees (Table 8). Individuals with scoliosis (n = 119), who were mainly at GMFCS level V, were unable to move in bed (immobile in lying position), spent more than 8 h in lying and could not straighten their hips or knees because of contractures. Individuals with windswept hips (n = 175) were unable to move in bed (immobile in lying position), lay mostly in a supine position in bed, and could not straighten hip or knee due to contracture. Seven individuals at GMFCS level I presented with windswept hips, according to the definition used in this thesis. The number of individuals with both windswept hips and scoliosis was 35. Immobility in lying and lack of knee extension were associated, both for scoliosis and windswept hips (Table 9 and Table 10).

Table 8. Numbers and percentages of deformities of the 830 individuals according to GMFCS levels. Deformities: asymmetrical limited hip flexion, and items from the Posture and Postural Ability Scale (trunk and pelvis) in sitting, scoliosis, and windswept hip deformity as well as complete data for each variable.

		Gross Motor Function Classification System Level					
	Complete data set N	**I** n = 159	**II** n = 185	**III** n = 130	**IV** n = 155	**V** n = 201	**Total** 830
Scoliosis [a]	767/830 (92%)	2/153 (1%)	6/177 (3%)	12/123 (10%)	29/138 (21%)	70/176 (39%)	119/830 (14%)
Windswept hips [b]	741/830 (89%)	7/146 (5%)	34/175 (19%)	30/126 (24%)	41/138 (30%)	63/156 (40%)	175/830 (21%)
Time in lying <8 h	791/830 (95%)	62/153 (40%)	49/174 (28%)	26/123 (21%)	23/145 (16%)	10/196 (5%)	170/830 (20%)
Time in lying 8-12 h	791/830 (95%)	90/153 (59%)	117/174 (67%)	88/123 (72%)	119/145 (82%)	143/196 (73%)	557/830 (67%)
Time in lying >12 h	791/830 (95%)	1/153 (1%)	8/174 (5%)	9/123 (7%)	3/145 (2%)	43/196 (22%)	64/830 (8%)
One lying position	791/830 (95%)	32/149 (21%)	57/176 (32%)	38/126 (30%)	48/146 (33%)	53/196 (27%)	228/830 (27%)
Supine	791/830 (95%)	8/149 (5%)	12/176 (7%)	7/126 (6%)	13/146 (9%)	21/196 (11%)	61/830 (7%)
Prone	791/830 (95%)	5/149 (3%)	12/176 (7%)	7/126 (6%)	9/146 (6%)	10/196 (5%)	43/830 (5%)
Right	791/830 (95%)	8/149 (5%)	15/176 (9%)	11/126 (9%)	18/146 (12%)	11/196 (6%)	63/830 (8%)
Left	791/830 (95%)	11/149 (7%)	18/176 (10%)	13/126 (10%)	8/146 (5%)	11/196 (6%)	61/830 (7%)
Lack of hip ext.	749/830 (95%)	9/146 (6%)	27/171 (16%)	41/125 (33%)	58/138 (42%)	80/169 (47%)	215/830 (26%)
Lack of knee ext.	794/830 (96%)	30/149 (20%)	67/178 (38%)	73/127 (57%)	119/147 (81%)	156/193 (81%)	445/830 (54%)
PPAS ability 1–3	794/830 (96%)	0/149 (0%)	0/178 (0%)	3/126 (2%)	16/146 (11%)	116/195 (59%)	135/830 (16%)

Table 9. Adjusted odds ratio (OR) values of having scoliosis for factors with a significant adjusted OR.

Effect (n)	OR	95% CI
PPAS lying <4 (116)	5.7	3.5–9.1
Lack of knee extension (116)	1.8	1.1–3.0
Time lying (116)		
>12 hours vs <8 hours	2.9	1.2–7.4
8–12 hours vs <8 hours	2.2	1.1–4.4

PPAS: Posture and Postural Ability Scale

Table 10. Adjusted odds ratio (OR) values of having windswept hips for those factors with significant adjusted OR.

Effect (n)	OR	95% CI
PPAS lying < 4 (174)	2.9	1.9–4.5
Supine (174)	1.9	1.0–3.3
Lack of knee extension (174)	1.6	1.1–2.3

PPAS: Posture and Postural Ability Scale

4.4 Paper IV

The seven volunteers aged 20 to 57 years (four women and three men), generated a total of 26 measurements of supine postures. All the intra-class correlation coefficients (ICCs) were >0.98, Table 11 shows the parameters of the intra-rater and inter-rater reliabilities (ICC, SEM and systemic error values) for the length measurements, and Table 12 shows angle measurements using the iPad 3-D and the Qualisys systems. The systematic error was 4.3 mm for the validity comparison, but less than half of that for the reliability comparisons. The systematic error was 0.2° or less and always non-significant.

Table 11. Trunk symmetry measurements.

Trunk symmetry measurements			
	Intra-rater reliabililty	Inter-rater Reliability	Validity
ICC(2,1)	0.997	0.994	0.995
ICC(3,1)	0.996	0.997	0.991
SEM	2.34	2.92	2.8
Systematic error	0.8 mm	1.9 mm	4.3 mm
p	<.001	<.001	<.001

Table 12 Trunk and knee angles.

	Upper-Lower trunk angle			Lower-Pelvis trunk angle			Thigh-Shank (knee) angle		
	Intra-rater reliability	Inter-rater reliability	Validity	Intra-rater reliability	Inter-rater reliability	Validity	Intra-rater reliability	Inter-rater reliability	Validity
ICC(2,1)	0.992	0.990	0.982	0.999	0.998	0.997	0.997	0.997	0.995
ICC(3,1)	0.992	0.991	0.982	0.999	0.998	0.997	0.997	0.997	0.995
SEM	0.45	0.51	0.69	0.45	0.62	0.72	1.29	1.25	1.5
Systematic error	0.1°	-0.1°	-0.1°	-0,1°	0.1°	0.2°	-0.1°	-0.2°	-0.1°
p	0.779	0.808	0.475	0.225	0.469	0.131	0.144	0.246	0.586

5 Discussion

The main achievement of this thesis is twofold. First, two methods of assessing posture were evaluated. Paper I found that the PPAS for rating posture and postural ability was valid and reliable. Paper IV introduced a valid and reliable way of quantifying posture, but further work is required to refine the procedure. Second, evidence was provided that an immobility and restricted joint range of motion is associated with the presence of scoliosis and windswept hip deformity. For this, data for all adults with CP in Sweden reported in the CPUP database were used. The PPAS (Paper I) played a key role in data collection for Papers II and III, i.e., it was used to identify asymmetries and quantify postural ability.

Paper I addressed the validity and reliability of a method of assessing posture and postural ability. The PPAS showed construct validity, internal consistency and excellent inter-rater reliability when used by experienced raters. Paper II discussed the relationship between asymmetrical limited hip flexion and seating posture, scoliosis and windswept hip deformity. The study confirmed the presence of asymmetrical limited hip flexion in the adult population with CP and its association with asymmetrical seating, scoliosis and windswept hip deformity. The direction of the lowest convex curve of the scoliosis or the direction of windswept hip deformity, was not linked with side of limited hip flexion. Paper III discussed the association of immobility, lying position and time spent in lying down with the presence of scoliosis and windswept hip deformity in adults with CP. Immobility and knee contractures were associated with scoliosis and windswept deformity. While the time lying down was associated with scoliosis, lying in a supine position was associated with windswept hip deformity. Lying only on the side or only in a prone position was not associated more with scoliosis or windswept hips than in those who had the other side to choose from. Paper IV addressed the criterion-related validity and reliability of the use of a 3-D camera mounted on an iPad for measuring posture. The results of the study showed excellent criterion-related validity, and inter- and intra-rater reliability, in measuring trunk symmetry length and in trunk and knee angle measurements.

5.1 Contracture

The ideology behind “physical management” concerns the individual’s right to health and participation in the community (see further in chapter 1.5, above). The coverage in this thesis has focused on the postural management component of physical management. The emphasis has been to aim at a symmetrical posture with joints in a neutral position focusing on prevention of deformity in the human body.

It has been believed that muscle shortening develops in response to spasticity (Hägglund & Wagner, 2011; Strobl & Grill, 2014). This view lacks robust evidence, as children with spastic CP who underwent successful selective dorsal rhizotomy to minimize spasticity had developed joint contractures at a 10-year follow-up (Clavet et al., 2008) and impaired muscle growth precedes the development of increased musculotendinous stiffness in children with CP (Willerslev-Olsen et al., 2018). Spasticity is thus a contributing factor, not a reason for contracture deformities. Muscle growth in children with spastic CP initially follows that of normally developing children, but the growth rate decreases significantly at 12–15 months of age (Herskind et al., 2016; Willerslev-Olsen et al., 2018). Muscles in children with spastic CP are shorter, and the tendons are longer than in normally developing children (Martin Lorenzo, Rocon, Martinez Caballero, & Lerma Lara, 2018). The fascicle length is also shorter, though the relative length is the same when normalised to the muscle belly length (Kruse, Schranz, Tilp, & Svehlik, 2018). Sarcomeres in a muscle sample taken from spastic muscle contractures in children with CP are in a stretched state (Smith, Lee, Ward, Chambers, & Lieber, 2011). This indicates a need for more sarcomeres in the muscle fibres. Satellite cell numbers in the muscle contracture samples were fewer than half of those seen in muscle samples from normally developing children (Smith et al., 2013). Furthermore, satellite cells in the shortened muscles of children with CP have impaired myogenic potential (Domenighetti et al., 2018). Satellite cells are essential for muscle growth and repair suggesting that the muscle fibre renewal process might be impaired with CP. There are indications from gene expression analysis that spastic muscles in individuals with CP have not fully reached a mature state (Smith et al., 2009). CP is a highly heterogeneous disorder. Its brain lesions occur at different time periods in the developing brain; therefore, these lesions will have different effects on the developing muscle. A lesion in the fetal brain will lead to an inactivity of muscles involved with that particular area of the brain and the properties of the fetal muscles remain immature because of the lack of prenatal stress. It is difficult to generalize about the properties of affected muscles in such a large and heterogeneous group as individuals with CP.

When an individual who cannot move on his own lies in a supine position, with bent knees (Paper III), gravity will bring the knees to the side (Hägglund et al., 2016) because the individual is not able to stabilize the legs. The strain on the hip joint musculoskeletal structures is higher on one side and lower on the other, which demands relatively fast remodelling on both sides of the hip joints through the body's renewal processes (LeVeau & Bernhardt, 1984). This results in a contracture in the hip joints (windswept hips). From biomechanical theory, it can be reasoned that as the knees fall to the side, the stretched musculoskeletal structures pull onto the pelvis, rotating it in the same direction as the knees. The rotated pelvis rotates the spine, by pulling onto the lumbar vertebra V, which in turn pulls onto the IV lumbar vertebra. This process continues up the spine until an equilibrium is reached. As the V lumbar vertebra follows the pelvis, the lumbar vertebra V will be backward rotated relative to the pelvis. Lumbar vertebrae IV will be backward rotated relative to the lumbar vertebrae V. This mechanism goes up the spine, causing a backward-rotated spine at its proximal end. From Fryette's law, it is known that rotation of the spine is combined with lateral flexion of the spine (Kapandji, 1974). The spine is made up of 33 mobile vertebrae, resulting in a great mobility of the structure as a whole, with minor movements at each level (Kapandji, 1974). The strain on the spine will be proportional to the degree of trunk asymmetry. Therefore, a small movement in the spine does not demand rapid remodelling of the musculoskeletal structures so the remodelling of the spine (scoliosis development) happens within the natural renewal of the body (LeVeau & Bernhardt, 1984). Scoliosis is usually diagnosed in children with CP after 8 years of age (Persson-Bunke et al., 2012), indicating that the development of scoliosis starts well before that age. Spending a long time in a lying position was associated with scoliosis (Paper III), which suggests that it is the lying posture, not the lying position, that influences the development of scoliosis. This enhances the need for symmetrical lying in individuals who do not move on their own. That knee contractures are associated with windswept hips and scoliosis (Paper III) supports the findings of Letts et al. (Letts et al., 1984) that the deformation of the spine in most individuals with CP starts with windswept hips.

5.2 Posture and Postural Ability Scale (PPAS)

The PPAS ratings (Paper I) are in descriptive form on an ordinal scale; therefore, weighted Kappa scores provided beyond-chance incorporation of ratio-scaled degrees of 0.85 to 0.99 agreements between raters, with values above 0.74 signifying excellent agreement (Fleiss, 1981). The participants in Paper I, 30 adults with CP, were recruited according to their GMFCS levels, with six individuals at each. The Jonckheere–Terpstra test was used to evaluate whether the PPAS tool could differentiate between known groups, defined as the five GMFCS levels. This showed that the median score of the PPAS increased with better gross motor function when analysing the arithmetic average values given by the raters. The PPAS could not identify differences in postural ability between individuals at GMFCS levels I–II, but was able to detect postural asymmetries at all GMFCS levels. Cronbach's alpha internal consistency values of the PPAS ranged from 0.96 to 0.97 when individual items were removed. The item-total correlation, the correlation between each item and the total score of the measurement ranged between 0.60 and 0.91. Cronbach's alpha becomes lower with any deleted item; the values of all alpha values exceeded the 0.8 threshold (Streiner & Norman, 2008), indicating high internal consistency of the PPAS. Only the item-total correlation of sitting in a sagittal plane did not exceed the 0.8 threshold, which was probably because of the lack of proper support under the feet for some participants. Those results indicate that PPAS is a valid and reliable tool to evaluate posture.

Since the PPAS (Paper I) was published, it has been applied for the assessment of posture and postural ability in individuals with severe and complex neurological disabilities, and has been recommended as a useful clinical tool (Holmes, Brock, & Morgan, 2018). In 2013, the PPAS was introduced into the national programs and quality registries for people with CP and into the national surveillance program and quality registry for people with myelomeningocele in Sweden (Alriksson-Schmidt et al., 2017), and was fully implemented in 2016. The PPAS was first evaluated for its properties in Paper I and then used in Papers II and III of this thesis. PPAS is the only postural evaluation tool that separates posture from postural ability, both in frontal and sagittal planes, in lying, sitting and standing. In 2015, the PPAS was evaluated for children by unexperienced raters with similar results as in Paper I (Rodby-Bousquet, Persson-Bunke, & Czuba, 2016). The PPAS evaluation was based on clinical assessment of children instead of photographs and video recordings of adults in Paper I. Therefore, PPAS has been evaluated both in standardized circumstances, using pictures and video recordings to minimize the disagreement arising from different performances (Paper I) and "live" in clinical settings where the performance could depend on uncontrollable factors, such as changed child mood or fatigue (Rodby-Bousquet et al., 2016). PPAS does not need any special equipment and only minor guidance for its use in a clinical environment. The posture component of PPAS, developed to identify postural deficits and asymmetries, was used in Paper II for evaluating trunk symmetry. The postural ability component of PPAS was used in Paper III to define individuals who were immobile when lying down.

The posture component of the PPAS was found to be a valuable clinical tool for assessing individuals with severe and complex neurological disabilities from day to day and to evaluate the effect of postural interventions. Deformities such as scoliosis and hip dislocation are associated with postural asymmetries (Rodby-Bousquet, Czuba, Hägglund, & Westbom, 2013). Identification of these are warnings, i.e., "red flags" (Novak, 2013), because they are indicators of developing deformity or limited movement (Rodby-Bousquet et al., 2013). The posture component of the PPAS is sensitive for detecting small postural asymmetries and deviations at an early stage, but does not differentiate between mild or severe deviations (Paper I). Early detection of postural asymmetries should lead to a thorough physical examination of the individual in question and early intervention where needed.

The postural ability component of the PPAS can be divided into two groups. Levels 1–3 includes individuals who cannot initiate or change position, while levels 4–7 relates to individuals who are able to do so. Individuals at level 3 can maintain a supine position when placed, but without being able to move, while individuals at level 4 can stabilize the trunk and be able to initiate flexion (lift the head and/or flex the knees). At level 5, the individual must be able to perform a controlled lateral weight shift onto the side of the trunk and be able to regain a supine position. Individuals who can perform a controlled lateral weight shift, but are unable to return to a supine position, are classified as level 4. In Paper III, adults with CP were divided into those two groups (levels 1–3 and levels 4–7) and were compared regarding scoliosis and windswept hips.

5.3 Asymmetric limited hip flexion

The effects of asymmetrical limited hip flexion on seating was the topic of a lecture given at the 1st International Posture and Mobility group conference in Dundee (Pope, 1997a). Since then has it been cited at various similar conferences in cities such as Exeter, Glasgow, Vancouver, Dublin, Oslo, Copenhagen and Stockholm, though this topic rarely appears in the literature. The effects of asymmetrical limited hip flexion on seating are only mentioned twice: i.e., in a book section on custom-moulded seating (Pope, 2007) and in an article where it is suggested that asymmetrical limited hip flexion explains hip subluxation being on the higher side of the pelvis instead of the lower side (Porter et al., 2007). Paper II was the first article to focus solely on asymmetrical limited hip flexion and its effect on seating posture, scoliosis and windswept hip deformity. This was a cross-sectional study, based on data of individuals included in the CPUP registry between January 1, 2013 and December 31, 2015. Those 714 individuals in the registry at that time participated voluntarily in the registry but were not the total population of adults with CP in Sweden and most of them have not been followed in the CPUP registry since when they were children. Chi-squared (χ^2) tests were used to test the hypothesis that there is an association between the side of asymmetrical limited hip flexion, the direction of windswept deformation and the direction of the lowest scoliotic curve. The OR is used as a measure of association between an exposure and an outcome. The OR in Paper II represented the odds that asymmetrical seating posture, windswept hips or scoliosis will occur in individuals with asymmetrical limited hip flexion, compared with the odds of an asymmetrical seating posture, windswept hips or scoliosis occurring in the absence of asymmetrical limited hip flexion.

Asymmetric limited hip flexion was more frequent in adults with spastic bilateral CP (78%) and at lower levels of motor function (GMFCS level V). That 22% of adults with CP at lower levels of motor function have asymmetrical limited hip flexion is concerning because this is associated with asymmetrical seating posture, having scoliosis and windswept hips. It is important to note that this might be an underestimate because most individual missing data points for hip flexion were classified at GMFCS level V. Typical contracture patterns demonstrate that when a joint is placed in full extension for sustained periods, it will cause a restriction in the range of movement in flexion (Trudel et al., 1999). The development of asymmetrical limited hip flexion contracture does not follow the typical contracture pattern in adults with CP, as they do not tend to spend a long time with the hips in extension. The aetiology or causes of this deformity are unknown, but there is probably more than one cause. From my clinical experience, I conclude that it occurs both in individuals with the pelvis in a posterior tilted position and in those with the pelvis in an anterior tilted position. Although a posterior tilted pelvis seems to be much more common in those with asymmetrical limited hip flexion, the severity of the clinical picture in seating is worse when the pelvis is tilted anteriorly.

The odds were higher that individuals with asymmetrical limited hip flexion had scoliosis, compared with those who did not (Paper II). It was expected from biomechanical reasoning that limited hip flexion on one side would result in as ipsilateral higher side of the pelvis and a contralateral convexity in scoliosis (Pope, 1997a, 2007; Porter et al., 2007). However, we did not confirm this in Paper II, as there was no association between the side of limited hip flexion and the direction of the lowest convex curve in scoliosis ($p = 0.808$). The severity of scoliosis is noted in the registry data, but not which curve is the primary one. It cannot be confirmed that the lowest convex curve in scoliosis in the registry was the primary curve in all cases. Therefore, I speculate that the results might have been different regarding the direction of the curve of the scoliosis if the primary curve had always been included in the analysis. The high prevalence of asymmetrical limited hip flexion in individuals with CP at GMFCS level V indicates the importance of appropriate seating with assessment of both posture and range of movement.

5.4 Lying down

Paper III focuses on lying positions and its association with scoliosis and windswept hip deformity. This was a cross-sectional study based on data of individuals with CP included in the CPUP registry. As for the participants, the 830 individuals in the registry at that time who participated voluntarily in the registry, were not the total population of adults in Sweden with CP and most of them have not been followed in the CPUP registry since they were children. As some of the data were ordinal, Spearman’s correlation coefficient (R_s) values were calculated for the association between lying positions and scoliosis and windswept hip deformity. The OR in Paper III represents the odds that windswept hips or scoliosis will occur in individuals with only one side to lie on, compared with the odds of these deformities occurring in individuals with more than one lying position. Over 50% of adult individuals with CP had a knee contracture. One in four had only one sleeping position to choose from and 16% could not move while lying in bed. Individuals who only lay in one position did not have higher odds of scoliosis compared with those with more lying positions. The results in Paper III clearly indicated that individuals unable to move or change their position in bed and/or have knee contractures have higher odds of having windswept hips and scoliosis (see further in chapter 5.1). This demonstrates the importance of incorporating corrective positions such as prone lying and standing in the daily routine.

Fulford & Brown’s (Fulford & Brown, 1976) findings indicated that the deformities in normal babies with a squint and babies with CP and windswept hips were the same. Normal babies with a squint develop windswept hips and scoliosis because they prefer a particular side when lying down and cry until they are put onto that side. Dunn (Dunn, 1974) demonstrated that the prenatal posture is quite often an infant's habitual or preferred postnatal sleeping posture, while (Porter et al., 2008) demonstrated that infants in supine lying, with their head consistently rotated to one side had convexity of scoliosis to the occipital side and windswept hips to the facial side as young adults (spastic bilateral CP). More recent studies on muscle function in individuals with spastic CP (see further in chapter 5.1), have indicated that the affected muscles have impaired function in that they do not grow or lengthen normally, suggesting an explanation for the results of Dunn (Dunn, 1972) and (Porter et al., 2008) that the affected infants prefer a particular side when lying, thereby not stretching the shorter muscles (Willerslev-Olsen et al., 2018). The effects arising from such impaired muscles along with the results from Paper III suggest that it is the posture of the lying position, but not the position itself, that is linked to deformity. This can best be demonstrated by the fact that the posture of individuals with severe and complex neurological disabilities in supine lying and seating is the same (Pope, 2007). That individuals who are immobile and spend a long time lying have higher odds of deformed spine (Paper III) was not so unexpected, as those are the two main ingredients or factors in the development of contractures (Trudel & Uhthoff, 2000).

5.5 3-D posture capture

The PPAS is clearly a valuable tool for assessing posture and detecting postural asymmetry early in children and adults. However, it does not quantify the asymmetry: i.e., it does not differentiate between mild or severe postural asymmetry. Quantitative measurements are required to measure postural asymmetry. The posture of an individual can roughly be described in terms of the orientation of 15 segments in space, rotating around 14 joints. The segments include the head, trunk and pelvis, hand, forearm, upper arm, thigh, shank, and foot. Optical motion capture systems, such as Qualisys, record the position of

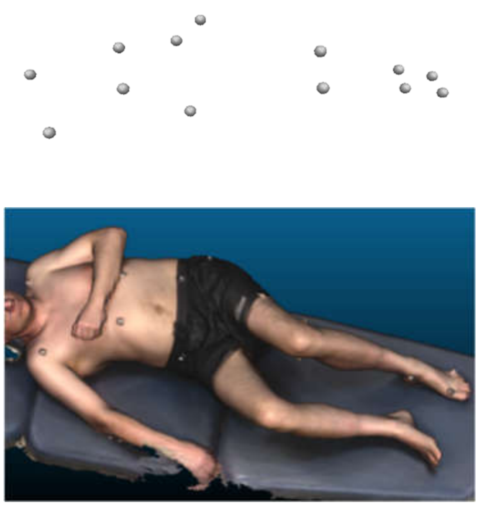

Figure 3 a) Output from the Qualisys system; b) Output from the iPad system (From Paper IV) markers placed on anatomical landmarks of the body, in a global frame of reference, without recording the body itself. Depending on the external instrument connected to the system, the optical motion capture system can export numerous data to be used in the kinematic and kinetic analysis. On the other hand, surface topography (digital mapping of environment/participant) creates a 3-D model of individuals' posture. Figure 3 demonstrates the difference from the visual output (image) from an optical motion capture system on the one hand and the image from a 3-D model on the other. The first is a picture of markers in space while the second is a picture of an individuals' posture with markers highlighting visible anatomical landmarks.

The study described in Paper IV is the first where surface topography has been used to analyse posture. That the participants in the study were seven healthy adult volunteers, recruited among colleagues and friends, is controversial. The aim was to use the iPad's 3-D camera to evaluate posture in individuals with severe postural deformities and it would have been ideal to use such individuals. However, the cost of bringing them to the laboratory along with their carers would have been high, the laboratory would have needed some adjustment and there would have been a time-consuming process gathering all the ethical permits. The timeframe of data collection would have to consider that every individual would have to have at least one backup appointment, if they were not comfortable at the first visit, thereby increasing the cost further. However, a 3-D model of an individual for evaluating posture and postural deformity depends on the visibility of anatomical landmarks, not on whether the subject is able or disabled. There is no reason to expect that the validity and reliability of a 3-D model differ between individuals with or without motor impairments, but this has yet to be established.

The 3-D coordinates of markers in space from the posture evaluation with a 3-D model (Paper IV) are in the form of continuous data. The descriptive statistic of an ICC(2,1)) was used to compare the intra-rater reliability between different ratings. ICC(2,1) values ranging from 0.992 to 0.999 indicate excellent agreement between the same rater, as a value of 1 indicates a perfect correlation. This descriptive statistic was used to compare the inter-rater reliability between different raters. ICC(2,1) values ranging from 0.990 to 0.997 indicate excellent agreement between the different raters. ICC(2,1) values were calculated to assess the validity between the iPad and the Qualisys method (Paper IV) and values ranging from 0.982 to 0.997 indicate excellent agreement between the two systems. The ICC(3,1) descriptive statistic was used to assess the internal consistency between the same rater, between raters and between the iPad and the Qualisys methods (Paper IV). ICC (3,1) values ranging from 0.982 to 0.997 indicate excellent agreement within the iPad system.

The usefulness of an iPad with 3-D camera is limited to a static posture, but its novelty lies in the simplicity of creating a 3-D model of body posture. The study in Paper IV confirmed the use this system as a valid and reliable way to quantify posture. Whatever kind of system is developed for such an analysis, local reference frames for angle calculations are required, with markers on anatomical landmarks to define each of the local reference frames. The markers need to be digitized and angles between segments calculated. Digitization of markers used in the 3-D model was the most challenging part of the study described in Paper IV. Currently, there is free software available to digitize 3-D models. With the appropriate software in place, digitization was simple. Matlab was used in Paper IV to define each of the local frames of reference, with vector calculations using marker coordinates from anatomical landmarks and to calculate angles and distances. These calculations could be done using any type of spreadsheet software.

5.6 Strengths and limitations

The use of the Swedish CPUP was a major strength of this thesis thanks to its vast and varied information. Data have been collected in a standardized way from a large population of adults with CP including all levels of motor function and cognitive abilities. The adult dataset is combined with data from many centres nationwide, covering all kinds of motor function and cognitive levels. Therefore, the results are likely to reflect the general population of adults with CP in Sweden. Another strength of this thesis is the long time-frame covered by the work. In 2009, studies began with the PPAS. While work on the thesis was progressing, the technology for creating a full body 3-D model expanded rapidly, turning a 3-D camera almost into a "plug and play" gadget. Although the use of the CPUP registry is one of the strengths of this thesis, it is also a major limitation. As there are many therapists in different centres who carry out physical examinations for the registry, there will always be some variations in the data reported, but the standardized positions for measurement, manuals and training sessions aim to minimize this.

The strength of Paper I, "Inter-rater reliability and construct validity of the PPAS in adults with CP in supine, prone, sitting and standing positions", lies in the quality of the individuals who modified the instrument and the quality of the instrument's predecessors indicating face validity. The PPAS is a modified version of the PAS, with the latter having been used successfully in clinical practice for many years, though it was not evaluated for reliability or validity. The use of pictures and video recordings for inter-rater evaluation standardized the occasion and minimized disagreements arising from different performances and circumstances. A limitation of the study lies in the methodology that the three raters who participated in the development of the instrument also rated the clients. Thus, there was a potential bias in terms of interrater reliability, but a newer study with unexperienced raters from different professions (Rodby-Bousquet et al., 2016) obtained similar results to Paper I, indicating good inter-rater reliability.

The strength of Papers II and III, “The effect of asymmetrical limited hip flexion on seating posture, scoliosis and windswept hip distortion” and “Preferred posture in lying and its association with scoliosis and windswept hips in adults with CP“, is that they were based on more subjects than in most other studies, using data from 714 and 830 adults participating in the CPUP register, respectively. A limitation of both those studies was in how the occurrence of a windswept hip deformity was established. Thus, an algorithm was used to determine this deformity instead of the use of a clinical examination. The original algorithm, using date on the hip range of motion was presented by Young et al. in 1998, and since then some modified versions of it have been used by Porter et al. (Porter et al., 2007, 2008, 2010), Persson-Bunke (Persson-Bunke et al., 2006) and Hägglund et al. (Hägglund et al., 2016). Only Young et al. (Young et al., 1998) evaluated the correspondence between the range of motion algorithm scores and clinically observed windswept hip deformity, with only moderate correlations. Knowing this, in Paper II and III the occurrence of windswept hip deformity was determined using two types of algorithms: a modified version of Porter’s algorithm and the Persson-Bunke algorithm. A threshold value of 20° further minimized the risk of including an individual without this deformity.

Limitation to Paper II was in the vague nature of the information in the CPUP manual regarding difference between one or more curves of scoliosis. In a S-shaped scoliosis there are two contiguous curves, and the lower end vertebra of the upper curve will represent the upper end vertebra of the lower curve. There is no proper guideline on how steep the connection between the lower end vertebrae of a scoliotic curve and the lower section of the spinal column is when deciding there is an S-shaped scoliosis. It is known that mild curve in the CPUP registry, is not a proper curve (Persson-Bunke et al., 2015). This can lead to an overestimation of the numbers of curves, as a mild pseudo apex follows a moderate or severe curve, in the opposite direction. However, scoliotic curves rated as moderate or severe should be examined by spinal radiography. Another limitation was how much missing values was in scoliosis and windswept data from the CPUP registry.

The strength of Paper IV, “Validity and reliability of an iPad with a three-dimensional camera for posture imaging” was in the use of a relatively simple tool to create a full body 3-D model of posture for use in a clinical environment. The limitation of that study was the reflective markers used in the study which were not suited for the 3-D camera and iPad. The 3-D camera emits a grid of laser light onto the environment in front of the iPad while the 3-D camera infrared sensor gathers the reflected light. The depth data are generated to build a digital map of the environment. The laser lights up the reflective markers which constantly reflect light to the infrared sensor, causing “shorter” depth to the markers. New surface layers are laid down on the digital map while the markers are illuminated, changing the shape of the markers in the 3-D model. This will not be a problem in clinical environments, where reflective markers are not needed. A further limitation lay in the complexity of work required after the full 3-D model had been created. For digitization, the 3-D model is imported into a 3-D point cloud and mesh processing software is used to construct the 3-D model by creating a point mesh, which is used for manual digitisation of markers. The marker locations in 3-D space are then exported into software where the locations of markers are used to calculate angles between segments and distances between markers. Further work in this area is required to reduce the amount of software needed to create a user-friendly system, with templates for calculating full and partial body posture angles and distances. The selection of healthy participants was also a limitation of the study. Although there were no differences between the 3-D models of abled and disabled individuals, it raises questions of possible errors in the 3-D model arising from movements of the subject.

5.7 Implications

This thesis has modified and validated a means of quantifying posture based on the PPAS (Paper I) and developed and validated a means of using a 3-D model of an individual’s posture created by a 3-D camera on an iPad system (Paper IV). This work has produced two reliable and valid instruments for quantifying posture. Both of those instruments can be used for research purposes, but the main significance of this work is the potential for measuring the effectiveness of postural interventions in a clinical environment. The PPAS is ideal for use in clinical settings. It does not require any particular equipment and it is easy to assess posture and postural ability. It provides information about postural deformities, if present, and where to apply postural support, if required, in lying, sitting and standing. On the other hand, the 3-D camera with an iPad provided a 3-D model of posture, which can be used to quantify the posture, but can also serve as a visual baseline for later comparisons.

The presence of asymmetrical limited hip flexion (<90°) was established (Paper II) in individuals whose level of gross motor function is low (GMFCS level V), together with its association with asymmetrical seating posture, scoliosis and windswept hips. The daily routine of an individual at GMFCS level V, is divided between lying, sitting and sometimes standing, with more than 20 hours of that time divided between lying and sitting (Rodby-Bousquet et al., 2013). It can be reasoned from biomechanical theories and is frequently observed in practice, that an asymmetrical limitation of hip flexion (<90°) produces an oblique pelvis when the subject is sitting. The ipsilateral side of the pelvis is higher and rotated forward, thereby accommodating the limited range of hip movement. As a result of pelvic obliquity, the upper body is directed towards the contralateral side, requiring compensatory lateral flexion of the spine to re-establish alignment of the thorax and the head. Paper III confirmed an association between the time spent in lying and scoliosis. It can be resonated that same applies with spending a long time in asymmetrical seating posture, as association is between asymmetrical limited hip flexion and scoliosis.

Paper III confirmed an association between adult individuals with CP who are immobile and cannot straighten their knees, and deformity of their body. This establishes a scientific foundation on which clinical guidelines can be set for postural management in lying (Ellis, 2016; Gericke, 2006; Pope, 1997b, 2007). As mentioned earlier in this thesis (see chapter 1.7), it has been known for decades that a zero joint position is crucial for the prevention of deformity in the treatment of subjects with polio. Paper III indicated an association between asymmetrical posture, not position and deformity, from which it can be reasoned that similar practice is required in postural management. Symmetrical posture with joints in as neutral position as possible, should be the aim of every postural management intervention.

5.8 Future studies

It is difficult to evaluate the effects of postural interventions. Randomised controlled trials would usually be ideal to make a fair comparison between individuals with severe neurological disability who get postural management along with conventional treatment and those who only get conventional treatment. In this situation, however, this would not be ideal because individuals with disability are a heterogeneous group living in various situations. This would make such studies hard to control especially because they would be a long-term study (10–20 years) and there are many ethical concerns that would need to be addressed. One possible way to evaluate the effect of postural management would be to compare the participants' postural management program to a known baseline. Clinical tools for the measurement of posture with high inter-rater reliability and validity such as the PPAS or an iPad with 3-D camera can be used to evaluate posture and changes over time. Creating baseline data on posture, postural ability and joint range of motion among adults with disabilities would be a good way to evaluate the effect of postural intervention.

It is clear that asymmetrical limited hip flexion does not follow the classical pattern of the development of contractures (Trudel et al., 1999). Further studies are needed to establish the cause or causes of asymmetrical limited hip flexion, its association to the higher side in the oblique pelvis and the main convex scoliotic curve. Further work is also needed regarding the definition of windswept hips, both clinically as a postural deformity and in the range of movement algorithm. Questions arise, such as how much abduction range of movement is allowed in the adducted hip? How much external rotation is allowed in the internal rotated hip? Does it need to be both adduction and internal rotation on the same leg? Do both legs need to be involved? All of these questions and more need to be answered.

The ideology that the implementation of symmetrical posture and neutral joints, will prevent deformity depends on normal development of the body. There are indications that the affected muscles of individuals with CP are not "normal" and do not grow or elongate normally. CP is a highly heterogeneous disorder, which makes any generalizations of affected muscles of little value. Therefore, there is a need to establish which muscle properties (see chapter 5.1) have the greatest clinical relevance and to develop an instrument that can accurately measure those muscle properties. The development of a system to quantify posture using a 3-D camera requires further work to create a protocol for full-body posture quantification together with special protocols for scoliosis and windswept hips. The aim of these templates is to simplify the process of evaluating posture quantitatively using a 3-D model.

6 Conclusions

The aim of this thesis was to find ways to quantify posture and to shed light on the association of posture and postural ability on one hand, and the presence of deformity in persons with CP on the other. In the postural section of this thesis, two methods of evaluating posture were presented. One method was the PPAS, which showed an excellent inter-rater reliability for experienced raters, with good internal consistency and construct validity. The PPAS detected postural asymmetries in adults with CP at all gross motor functional levels. The second evaluation method used a 3-D camera mounted of an iPad to create a 3-D model for quantifying posture and any postural deformity. The intra- and inter-rater reliabilities were high and showed good correlation and validity when compared with the output from the Qualisys system. The results from the PPAS on one hand and the use of a 3-D model created by a 3-D camera on the other, clearly indicate their potential for reliable use in the clinical environment for assessing posture, early signs of developing postural deformity and evaluating the outcomes of interventions. The PPAS is the optimal method to be used in the day-to-day evaluation of posture and postural ability in clinical settings, while the iPad-based method is to be used when there is a need to quantify posture.

In the epidemiological part of this thesis, the association of an individual's posture and postural ability with deformity was established. Sitting in an asymmetrical posture because of asymmetrical limited hip flexion <90° and being immobile in a lying position, lead to deformities. A high prevalence of asymmetrical limited hip flexion <90° was found in individuals with CP at GMFCS level V, which was associated with higher odds of having an oblique pelvis, asymmetrical trunk, scoliosis, and windswept hips: all clinical signs of asymmetrical seating posture. Being immobile (level III or less in the PPAS supine lying postural ability score) in a lying position and unable to straighten the knees, was associated with higher odds of both scoliosis and having windswept hips. Spending a long time lying increases the odds of scoliosis while a supine position increases the odds of windswept hips.

It can be concluded from this thesis that spending a long time immobile in an asymmetrical posture predisposes deformity development to some degree. It can be reasoned that being in different symmetrical positions during the day will at least minimize the development of deformities. It is important to have assessment tools to screen the posture of at-risk individuals that can be used for children and adults throughout life to identify their needs for support and evaluate interventions. It is especially important to screen posture from an early age in those children with disabilities who do not start to sit independently and are therefore at risk of developing asymmetrical postures.

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Interrater reliability and construct validity of the Posture and Postural Ability Scale in adults with cerebral palsy in supine, prone, sitting and standing positions

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Abstract

Objective: To evaluate reliability, internal consistency and construct validity of the Posture and Postural Ability Scale for adults with cerebral palsy.

Design: Psychometric evaluation of a clinical assessment tool.

Setting: Rehabilitation centres in Sweden and Iceland.

Subjects: Thirty adults with cerebral palsy aged 19–22 years, six people at each level I–V of the Gross Motor Function Classification System.

Main measures: The Posture and Postural Ability Scale contains a 7-point ordinal scale for postural ability in supine, prone, sitting and standing, and items for assessment of posture. Posture and postural ability was rated from photos and videos by three independent assessors. Interrater reliability was calculated using weighted kappa. Internal consistency was analysed with Cronbach's alpha if item deleted and corrected item–total correlation. Construct validity was evaluated based on known groups, using Jonckheere Terpstra for averaged values of the three raters relative to the Gross Motor Function Classification System.

Results: There was an excellent interrater reliability (kappa = 0.85–0.99) and a high internal consistency (alpha = 0.96–0.97, item–total correlation = 0.60–0.91). Median values differed ($P < 0.02$) between known groups represented by the levels of gross motor function, showing construct validity for all items.

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Conclusion: The Posture and Postural Ability Scale showed an excellent interrater reliability for experienced raters, a high internal consistency and construct validity. It can detect postural asymmetries in adults with cerebral palsy at all levels of gross motor function.

Keywords
Posture, postural control, assessment, cerebral palsy, reliability, validity

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Introduction

There are few assessment tools for posture and postural ability in lying, sitting and standing for people with severe physical disabilities, and none of them has been evaluated regarding its psychometric properties for adults with cerebral palsy, even though posture is a key problem in adult cerebral palsy.[1]

Asymmetric posture is known to cause progressive deformities in immobile people with cerebral palsy due to the effect of gravity.[2–4] However, early detection and preventive treatment can reduce the number of severe contractures, hip dislocations, scoliosis and other fixed deformities.[5–8] As tissue adaptations often occur slowly over time, standardized measurement is crucial to identify the need for treatment and postural support in order to minimize progressive deformities and also to evaluate therapeutic interventions.

The Physical Ability Scale[9] for assessment of postural ability in children with severe disabilities was developed by Noreen Hare during the 1970s and 1980s. This inspired Pountney and co-workers to develop the Chailey Levels of Ability[10] to describe stages of motor development in normal infants and children with motor impairments. Its validity has been evaluated for children and youth with cerebral palsy.[10] These instruments form the basis of the Postural Ability Scale developed by Pauline Pope in the early 1990s[11] to assess both posture and postural ability in people with severe physical disabilities regardless of age and diagnosis. This assessment tool allows postural ability and posture to be assessed separately. It is in clinical use for trained professionals but has not been evaluated for its psychometric properties. During the years 2009–2011, the Postural Ability Scale was developed further by Pope and co-workers (ERB, AA, GJ). The levels of ability were slightly modified and items added to the quality of posture to allow assessment of posture from a sagittal as well as a frontal view. This modified and expanded version. called the Posture and Postural Ability Scale (PPAS) (Table 1), has not previously been tested for reliability or validity.

There is no universal definition of posture and postural ability. In this paper, the term ‘posture’, relates to the shape of the body (i.e. the anatomical alignment of the body segments in relation to each other and the supporting surface) and also to the relationship between the body and the environment.[11–13] ‘Postural ability’ refers to the ability to stabilize the body segments relative to each other and to the supporting surface; to get into the most appropriate body configuration for the performance of the particular task and environment. This means control of the centre of gravity relative to the base of support during both static and dynamic conditions.[11,12]

Our purpose was to evaluate the interrater reliability, internal consistency and construct validity of the Posture and Postural Ability Scale for adults with cerebral palsy.

Methods

Participants were recruited during a project in the south of Sweden in October 2009–October 2010 to expand a National Health Care Program and Quality Register for cerebral palsy,[5,6,14] to include adults with cerebral palsy. The subjects who agreed to join the Cerebral Palsy Health Care Program were examined by a physiotherapist and occupational

Table 1. The Posture and Postural Ability Scale for assessment of postural ability in standing, sitting, supine and prone, followed by assessment of *quality of posture* in frontal and sagittal view

	Levels of postural ability
Level 1	Unplaceable in an aligned posture
Level 2	Placeable in an aligned posture but needs support
Level 3	Able to maintain position when placed but cannot move
Level 4	Able to initiate flexion/extension of trunk
Level 5	Able to transfer weight laterally and regain posture
Level 6	Able to move out of position
Level 7	Able to move into and out of position

Quality of posture, frontal view, (Yes = 1 point, No = 0 points)

Standing	Sitting	Supine	Prone
Head midline	Head midline	Head midline	Head to one side
Trunk symmetrical	Trunk symmetrical	Trunk symmetrical	Trunk symmetrical
Pelvis neutral	Pelvis neutral	Pelvis neutral	Pelvis neutral
Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis	Legs separated and straight relative to pelvis
Arms resting by side	Arms resting by side	Arms resting by side	Arms resting (elevated, mid-position)
Weight evenly distributed	Weight evenly distributed	Weight evenly distributed	Weight evenly distributed

Quality of posture, sagittal view, (Yes = 1 point, No = 0 points)

Standing	Sitting	Supine	Prone
Head midline	Head midline	Head midline	Trunk in neutral position
Trunk in neutral position	Trunk in neutral position	Trunk in neutral position	Pelvis neutral
Pelvis neutral	Pelvis neutral	Pelvis neutral	Hips extended
Legs straight, hips/knees extended	Hips mid-position (90°)	Legs straight, hips/knees extended	Knees extended
Feet mid-position/flat on floor	Knees mid-position (90°)	Feet resting in normal position	Arms resting (elevated, mid-position)
Weight evenly distributed	Feet mid-position/flat on floor	Weight evenly distributed	Weight evenly distributed

therapist. At the examination the subjects were also asked if they would agree to participate in the psychometric evaluation of the Posture and Postural Ability Scale. All subjects who agreed to participate were included, until six people at each level of the Gross Motor Function Classification System had accepted. One additional client with cerebral palsy was recruited from the Rehabilitation Centre of Excellence in Kópavogur, Iceland. Written consent was collected from all participants or by proxy where the participant was unable to give such consent. Ethical approval for the study was granted by the Medical Research Ethics Committee at Lund University, number 2009/361.

Cerebral palsy was defined according to Rosenbaum et al.[1] Exclusion and inclusion criteria were in accordance with the Surveillance of Cerebral Palsy in Europe (SCPE).[15] People with motor impairment and specific neurological signs (ataxia, dyskinesia and/or spasticity) caused by different genetic syndromes before the age of 2 without progressive brain dysfunction were included. Gross motor function was determined according to the expanded and revised version of the Gross Motor Function Classification System, which comprises five levels I–V.[16] The most severe functional limitation is level V, with difficulties controlling head and trunk posture in most positions, and severely limited or no voluntary control of movement.

Posture and postural ability was recorded by photos and videos of the participants. Photos of habitual posture of each individual were taken from a frontal and sagittal view of the whole body in supine, prone, sitting and standing position. Habitual refers to the posture customarily adopted by the individual when instructed to sit, stand or lay down in prone or supine as straight as possible or the posture the body assumes when placed as straight as possible in any of these positions and allowed to settle. The positions were: supine lying on a plinth with arms resting by side; prone lying on a plinth with the head to one side and arms resting in an elevated position (flexion in elbows and abduction, external rotation of shoulders); sitting on a plinth with feet on the floor; standing on the floor. Those who were unable to maintain position independently were provided the manual support needed to stay in position. People who required total body support in standing, such as those classified gross motor function level V, were assessed in a standing brace or on a tilt table. Videos recorded the participants' postural ability, from the lateral view of the plinth, while instructed to assume and get out of the four positions. If unable to do this they were placed in each position. Assessment of ability was then carried out sequentially corresponding to the points on the Posture and Postural Ability Scale (Table 1).

The Posture and Postural Ability Scale contains a 7-point ordinal scale for the assessment of postural ability in standing, sitting, supine and prone and six items for assessment of *quality of posture* in the frontal plane and another six items in the sagittal plane (Table 1). Postural symmetry and alignment gives 1 point for each item while asymmetry or deviation from midline gives 0 points. The total score of 0–6 points is calculated separately for each position in the frontal and sagittal plane. Postural ability is rated according to the ordinal scale ranging from unplaceable (level 1) to able to move into and out of position (level 7). The two lower levels of postural ability are in fact a rating of no ability, that is, they are unable to maintain or change position by themselves. The difference between those two levels is whether the person can (level 2) or cannot (level 1) conform to the position when placed by another person (i.e in anatomical alignment when supported). When a person cannot be placed in prone and standing due to hip dislocation or severe contractures, especially of the hip flexors, postural ability is scored as level 1 = unplaceable and posture is scored 0.

Postures and postural abilities of the 30 subjects were assessed from the photos and videos during February 2012 by three experienced physiotherapists independently using the Posture and Postural Ability Scale.

Statistical analysis

Interrater reliability for three independent raters was calculated using weighted kappa scores which takes the degree of disagreement into account.[17] The magnitude of weighted kappa indicates the agreement beyond chance and was interpreted according to the method of Fleiss,[18] where ≤ 0.40 signifies poor agreement, 0.40–0.75 fair to good agreement and ≥ 0.75 signifies excellent agreement. The internal consistency of the assessment tool was evaluated through 'Cronbach's alpha if item is deleted' and 'corrected item–total correlation' based on averaged values for the three raters. Cronbach's alpha if item is deleted corresponds to the value achieved if a specific item is removed.[19] The corrected item–total correlation shows the correlation between each item and the total score of the measurement and any item with a value <0.2 should be discarded.[19] For analysis of reliability and consistency all levels of the Gross Motor Function Classification System were combined and 95%

non-parametric bootstrap confidence intervals were added based on 1000 re-samples.[20,21] Construct validity was evaluated for known groups based on the five levels of gross motor function with median and range. Jonckheere-Terpstra was used for analysis of arithmetic average values given by the raters, and P-values <0.05 were considered significant. The alternative hypothesis of the test is that median values of the scores decrease with decreasing gross motor function. For all statistical computing, R software environment was used.

Results

Postures and postural abilities of 30 adults (15 males, 15 females) with cerebral palsy born 1988–1991 (age range 19–22) were recorded with photos and videos during October 2009–October 2010. The scores varied between participants at different levels of the Gross Motor Function Classification System, with decreasing values at lower levels of gross motor function such as level IV and V. The distribution of scores across the 30 participants were described as median, mean, standard deviation, min, max values, 25th and 75th percentile in all four positions (Table 2). Since every person was assessed by three raters, each missing assessment (photo/position) generated three missing values (Table 2).

The Posture and Postural Ability Scale showed excellent interrater reliability for the three independent raters with weighted kappa values of 0.85–0.99 (Table 3). There was a high internal consistency for the Posture and Postural Ability Scale for all items (Table 4). Cronbach's alpha if item deleted was 0.97 for sitting posture sagittal and 0.96 for all other items with a 95% confidence interval of 0.93–0.98 for all items. Corrected item–total correlation varied between 0.60 and 0.91 with the lowest correlation for sitting posture in the sagittal view.

The Posture and Postural Ability Scale showed construct validity based on the ability of the assessment tool to differ between known groups represented by the gross motor function levels I–V (Table 5, Figure 1 online). Median values and range in terms of min and max values are presented together with P-values ($P < 0.02$) calculated with Jonckheere-Terpstra for averaged values (Table 5). Distribution of scores at each level of gross motor function in all four positions is provided for all three raters (Figure 1 online). The Posture and Postural Ability Scale could not identify differences in postural ability between individuals at levels I–II but was able to detect postural asymmetries at all levels.

Table 2. Scores for the Posture and Postural Ability Scale across the 30 participants

		Median	Mean	SD	Min/max		Percentile		Valid/missing	
Supine	Postural ability	7	5.5	2.1	1	7	4	7	30	0
	Posture frontal	1	2.4	2.4	0	6	0	4.3	30	0
	Posture sagittal	2	2.7	2.3	0	6	1	5	30	0
Prone	Postural ability	6	5.0	2.4	1	7	4	7	29	1
	Posture frontal	2	2.4	2.0	0	6	0	4	27	3
	Posture sagittal	3	3.2	2.3	0	6	1	5	28	2
Sitting	Postural ability	7	5.0	2.4	1	7	2	7	30	0
	Posture frontal	2	2.7	2.4	0	6	0	5	30	0
	Posture sagittal	3	2.4	1.9	0	6	1	4	30	0
Standing	Postural ability	2	4.0	2.7	1	7	1	7	29	1
	Posture frontal	1	1.9	2.2	0	6	0	3.3	30	0
	Posture sagittal	1	2.2	2.1	0	6	1	4	30	0

Median, mean, standard deviation (SD), min and max values, 25th and 75th percentile, valid and missing values in supine, prone, sitting and standing.

Table 3. Interrater reliability of the Posture and Postural Ability Scale

		Weighted kappa	95% CI	
Supine	Postural ability	0.98	0.93	0.99
	Posture frontal	0.94	0.87	0.98
	Posture sagittal	0.88	0.79	0.92
Prone	Postural ability	0.99	0.98	1.00
	Posture frontal	0.89	0.79	0.96
	Posture sagittal	0.85	0.71	0.93
Sitting	Postural ability	0.99	0.96	1.00
	Posture frontal	0.91	0.83	0.95
	Posture sagittal	0.87	0.75	0.93
Standing	Postural ability	0.99	0.94	1.00
	Posture frontal	0.95	0.90	0.97
	Posture sagittal	0.95	0.90	0.97

Weighted kappa with 95% confidence interval (95% CI) for three independent raters.

Discussion

The Posture and Postural Ability Scale showed an excellent interrater reliability, a high internal consistency and good construct validity for adults with cerebral palsy. This is, to our knowledge, the first study evaluating an assessment tool for posture and postural ability in lying, sitting and standing position for adults.

A limitation of the study is that all three raters participated in the development of the Posture and Postural Ability Scale; they have long clinical experience and are specialized in posture management. Further research is needed to examine interrater reliability for trained practitioners not involved in the modification of the assessment tool. A further limitation is that the ratings were based on photos and videos. This removes some variability that arises in clinical practice. Photos provide reflection of the posture at just one point in time. However, the condition of people with severe disabilities may alter during the day due to fatigue, pain, etc. It may also change over time, making a measurement on different occasions such as test–retest and intrarater reliability more difficult to interpret. Therefore we chose to evaluate agreement between different raters and used photos and videos to standardize the occasion and minimize disagreement due to different performances and circumstances.

Internal consistency represents the average of the correlations among all items. The scale demonstrated a high internal consistency for all items, where Cronbach's alpha if item deleted was

Table 4. Internal consistency of the Posture and Postural Ability Scale

		Cronbach's α if item deleted	95% CI		Item–total correlation	95% CI	
Supine	Postural ability	0.96	0.94	0.98	0.72	0.58	0.85
	Posture frontal	0.96	0.93	0.98	0.91	0.84	0.96
	Posture sagittal	0.96	0.94	0.98	0.87	0.74	0.95
Prone	Postural ability	0.96	0.94	0.98	0.79	0.65	0.89
	Posture frontal	0.96	0.93	0.98	0.88	0.76	0.95
	Posture sagittal	0.96	0.94	0.98	0.87	0.75	0.96
Sitting	Postural ability	0.96	0.94	0.98	0.83	0.73	0.92
	Posture frontal	0.96	0.94	0.98	0.84	0.69	0.94
	Posture sagittal	0.97	0.94	0.98	0.60	0.32	0.81
Standing	Postural ability	0.96	0.93	0.98	0.91	0.84	0.96
	Posture frontal	0.96	0.94	0.98	0.82	0.68	0.92
	Posture sagittal	0.96	0.94	0.98	0.81	0.66	0.92

Cronbach's alpha if item deleted and corrected item–total correlation showing the correlation between each item and the total score when averaged scores for all raters are considered.

Table 5. Construct validity of the Posture and Postural Ability Scale

		GMFCS					
		I	II	III	IV	V	*P*-value
Supine	Postural ability	7 (7–7)	7 (7–7)	7 (6–7)	4 (3–7)	1.5 (1–4)	<0.001
	Posture frontal	6 (2–6)	4 (2–5)	1 (0–6)	0 (0–1)	0 (0–1)	<0.001
	Posture sagittal	6 (4–6)	4 (1–6)	2.5 (0–6)	0.5 (0–3)	1 (0–3)	<0.001
Prone	Postural ability	7 (7–7)	7 (7–7)	6 (5–7)	4 (1–6)	1 (1–3)	<0.001
	Posture frontal	5 (2–6)	4 (2–5)	2 (0–5)	1 (0–3)	0 (0–3)	<0.001
	Posture sagittal	6 (2–6)	5 (2–6)	3 (0–6)	1 (0–4)	0 (0–4)	<0.001
Sitting	Postural ability	7 (7–7)	7 (7–7)	7 (2–7)	2 (2–6)	2 (1–2)	<0.001
	Posture frontal	6 (4–6)	4 (1–6)	3 (0–6)	0 (0–2)	0 (0–4)	<0.001
	Posture sagittal	3.5 (2–6)	2 (0–5)	3.5 (0–6)	0 (0–4)	1 (0–5)	0.019
Standing	Postural ability	7 (7–7)	7 (7–7)	4 (1–7)	1.5 (1–2)	1 (1–2)	<0.001
	Posture frontal	6 (3–6)	3 (0–5)	0 (0–3)	0 (0–2)	0 (0–2)	<0.001
	Posture sagittal	5 (4–6)	2 (0–6)	1 (0–4)	0 (0–3)	0 (0–3)	<0.001

Median values and range (min–max values) for levels I–V of the Gross Motor Function Classification System (GMFCS) and *P*-values calculated with Jonckheere-Terpstra of averaged values for the three raters.

0.96–0.97, which exceeds the 0.8 recommended by Streiner and Norman.[19] Corrected item–total correlation showed a slightly lower value for sitting posture in the sagittal view compared to the other items. This is probably explained by the fact that the height of the plinth was not optimal in some photos, which affected the ratings of hips and knees mid-position. An adjustable plinth is not always available in clinical practice. If the plinth is not adjustable or if using a chair, it would be best to provide additional support for the feet when needed.

Construct validity was evaluated through its ability to differ between known groups in terms of the gross motor function levels in cerebral palsy. The expanded and revised version of the Gross Motor Classification System has been developed for children with cerebral palsy with the oldest age band 12–18 years, but some studies have shown validity and reliability for use of this classification in adults with cerebral palsy as well.[22–24] According to this classification, individuals at levels I and II can walk and stand unsupported. The highest level of the Posture and Postural Ability Scale is to move into and out of position and therefore the assessment tool was not expected to differ between gross motor function levels I and II in postural ability. The distribution of individuals at maximum and minimum score showed an anticipated ceiling effect for postural ability in all four positions for adults at level I–II, while the floor effect was higher for posture, indicating a better quantity in terms of ability than quality of posture. This indicates a need for assessing posture and postural ability separately and as distinct from gross motor function, in order to detect postural asymmetries and identify need for postural support. A strength of the Posture and Postural Ability Scale is that it identified postural asymmetries and deviations at all levels of gross motor function in this study of adults with cerebral palsy.

The Posture and Postural Ability Scale was sensitive to detect small postural asymmetries and deviations and is likely to detect postural asymmetries at an early stage. The assessment tool has no grading and cannot differ between a mild, moderate or severe deviation. The rationale is that any deviation will increase by forces imposed by gravity so it is clinically relevant to detect asymmetric posture early in order to apply the appropriate intervention to minimize progressive deformities.

The Posture and Postural Ability Scale does not require any special equipment and is easy to use in a clinical setting. It provides important information of the need for postural support and where it needs to be applied. Although the instrument has been used in clinical practice for different client groups, further research to evaluate its psychometric properties for people with diagnoses other than cerebral palsy is desirable. All assessment tools for posture and postural ability currently used in clinical practice require additional training of the professional intending to use them.

In conclusion, the Posture and Postural Ability Scale shows an excellent interrater reliability for experienced raters, a high internal consistency and good construct validity. It can detect postural asymmetries at all levels of gross motor function in adults with cerebral palsy. The results show an anticipated ceiling effect for postural ability at gross motor function level I–II. Further research is needed to examine interrater reliability for trained professionals not involved with modification of the assessment tool, as well as its application to other client groups.

Clinical messages

- This evaluation is based on photos and videos of posture and postural ability in adults with cerebral palsy.
- The Posture and Postural Ability Scale shows excellent interrater reliability, high internal consistency and construct validity.
- It can detect postural asymmetries at all levels of gross motor function in adults with cerebral palsy.

Authors' contributions

ERB designed the study, developed the instrument, collected the data, rated the subjects, analysed the results and wrote the manuscript. AA and GJ developed the instrument, rated the subjects and revised the manuscript. TC made the statistical calculations, analysed the data, provided the dot plots and revised the manuscript. ACJ revised the manuscript. GH designed the study and actively improved and revised the manuscript. All authors approved the final draft.

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Conflict of interest

The authors declare that they have no conflicts of interests.

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Paper II

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Research Paper

The effect of asymmetrical limited hip flexion on seating posture, scoliosis and windswept hip distortion

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ABSTRACT

Background: Postural asymmetries with seating problems are common in adults with cerebral palsy.
Aims: To analyse the prevalence of asymmetrical limited hip flexion (< 90°) in adults with CP, and to evaluate the association between asymmetrical limited hip flexion and postural asymmetries in the sitting position.
Methods and procedures: Cross-sectional data of 714 adults with CP, 16–73 years, GMFCS level I–V, reported to CPUP, the Swedish cerebral palsy national surveillance program and quality registry, from 2013 to 2015. Hip range of motion was analysed in relation to pelvic obliquity, trunk asymmetry, weight distribution, scoliosis and windswept hip distortion.
Outcomes and results: The prevalence of asymmetrical limited hip flexion increased as GMFCS level decreased. Of adults at GMFCS level V, 22% had asymmetrical limited hip flexion (< 90°). The odds of having an oblique pelvis (OR 2.6, 95% CI:1.6–2.1), an asymmetrical trunk (OR 2.1, 95% CI:1.1–4.2), scoliosis (OR 3.7, 95% CI:1.3–9.7), and windswept hip distortion (OR 2.6, 95% CI:1.2–5.4) were higher for adults with asymmetrical limited hip flexion compared with those with bilateral hip flexion > 90°.
Conclusions and implications: Asymmetrical limited hip flexion affects the seating posture and is associated with scoliosis and windswept hip distortion.

What this paper adds?

This paper contributes to the field of seating analysis for individuals in wheelchairs. It shows that asymmetrical limited hip flexion (< 90°) is present in about 22% of individuals with cerebral palsy who are classified at GMFCS level V. It confirms that the presence of asymmetrical limited hip flexion (< 90°), increases the odds of pelvic obliquity, trunk asymmetry, scoliosis, and windswept hip distortion. Therefore, asymmetrical limited hip flexion needs to be ruled out or compensated, especially in individuals with spastic bilateral CP at GMFCS level V, who are in a poor seating posture.

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1. Introduction

1.1. Cerebral palsy

Cerebral palsy is a well-recognised neurodevelopmental condition that manifests in early childhood and persists throughout life (Rosenbaum et al., 2007). It has been attributed to a non-progressive disturbance that occurs in the developing fetal or infant brain. Cerebral palsy is the most common physical disability in childhood, with a prevalence of 2–2.4/1000 live births in Europe (Nordmark, Hägglund, & Lagergren, 2001; SCPE, 2002). In Sweden, the survival rate to 20 years of age is 60% among the most severely disabled children with cerebral palsy (Westbom, Bergstrand, Wagner, & Nordmark, 2011), although the survival rate in Britain is lower (Hutton & Pharoah, 2006).

1.2. Contractures and asymmetries

The term "joint contractures" is used to describe the loss of passive range of movement in diarthrodial joints (Wong, Trudel, & Laneuville, 2015). Long-lasting reduction of spasticity does not prevent contracture development (Tedroff, Lowing, Jacobson, & Astrom, 2011), and in pure immobilisation, the role of arthrogenic structures in contracture development increases with time, in such a way that immobilisation in flexion leads to limited extension but allows more flexion (Trudel & Uhthoff, 2000). Pope (Pope, 2007) described the effect of sitting with asymmetrical limited hip flexion (< 90°), where the ipsilateral side of the pelvis will go up and in a forward direction, directing the trunk to the contralateral side. Lateral spinal curvature is needed to compensate for the asymmetry caused by pelvic obliquity (Porter, Michael, & Kirkwood, 2007). In adults with cerebral palsy who have lower levels of motor function, more postural asymmetries are present in the sitting position than when standing, and these asymmetries are associated with a limited range of motion, scoliosis, and the inability to change position (Rodby-Bousquet, Czuba, Hägglund, & Westbom, 2013).

1.3. Aim

The aim of this study was to analyse the prevalence of asymmetrical limited hip flexion less than 90° (ALHF < 90°) in individuals with cerebral palsy and to evaluate the association between ALHF < 90° and asymmetrical seating posture, the occurrence of scoliosis, and windswept hip distortion.

2. Methods

2.1. Ethical approval and consent

Ethical approval was granted by the Medical Research Ethics Committee at Lund University (LU 2009-341), and permission was obtained to extract data from the CPUP registry. All participants consent to research based on reported data. No individual details are presented.

2.2. Data collection and participants

This cross-sectional study was performed based on data from the national surveillance program and quality registry for cerebral palsy in Sweden (CPUP) (Alriksson-Schmidt et al., 2017). Data were extracted from the most recent reports for all adults with cerebral palsy included in the registry between 1st of January 2013 and 31st of December 2015. Inclusion and exclusion criteria were defined by the Surveillance of Cerebral Palsy in Europe (SCPE, 2002), and subtypes were classified into spastic unilateral, spastic bilateral, ataxic, and dyskinetic cerebral palsy. A total of 714 adults comprising 357 men and 357 women with a median age of 23 years (range 16–73 years) were reported to the CPUP registry. The subjects' gross motor function ranged from GMFCS level I (n = 159), II (n = 150), III (n = 114), IV (n = 122) to level V (n = 170) (Table 1). The distribution of participants' neurological subtypes was as follows: spastic unilateral (n = 156), spastic bilateral (n = 385), ataxic (n = 25), dyskinetic (n = 92), and mixed or unclassified subtypes (n = 42). Subtypes were not reported for 14 adults.

2.3. Classifications and measurements

All assessments were performed by local physiotherapists and occupational therapists in a standardised manner employing an assessment form and an accompanying manual (www.cpup.se).

2.3.1. Gross motor function classification system

Gross motor function was classified using the expanded and revised version of the Gross Motor Function Classification System (GMFCS) levels I–V, age band 12–18 years (Palisano, Rosenbaum, Bartlett, & Livingston, 2008). Even though GMFCS was developed for children, it has also been shown to be accurate for use in adults with cerebral palsy (Jahnsen, Aamodt, & Rosenbaum, 2006; McCormick et al., 2007).

Table 1
Numbers and percentages of the 714 individuals with distribution of the GMFCS levels, asymmetrical limited hip flexion, and items from the Posture and Postural Ability Scale (trunk, pelvis, and weight distribution) in sitting, direction of scoliosis, and windswept hip distortion as well as complete data for each variable.

	Complete data set	Gross Motor Function Classification System Level					
		I N = 159	II N = 150	III N = 114	IV N = 121	V N = 170	Total 714
ALHF < 90°[a]	N = 665 (93%)	2 (1%)	5 (3%)	7 (6%)	11 (9%)	38 (22%)	63 (9%)
Asymmetrical trunk	N = 672 (94%)	24 (15%)	31 (21%)	41 (36%)	60 (50%)	96 (56%)	252 (35%)
Oblique pelvis	N = 672 (94%)	24 (15%)	31 (21%)	41 (36%)	60 (50%)	96 (56%)	252 (35%)
Uneven weight distribution	N = 661 (93%)	15 (9%)	32 (21%)	39 (34%)	64 (53%)	99 (58%)	249 (15%)
Scoliosis[b]	N = 286 (40%)	2 (1%)	5 (3%)	10 (9%)	16 (13%)	60 (35%)	93 (13%)
Convex right		1 (1%)	3 (2%)	5 (4%)	6 (5%)	34 (20%)	49 (7%)
Convex left		1 (1%)	2 (1%)	5 (4%)	10 (8%)	26 (15%)	44 (6%)
Windswept hip distortion[c]	N = 336 (47%)	14 (9%)	31 (21%)	26 (23%)	32 (26%)	60 (35%)	163 (25%)
To the right		9 (6%)	17 (11%)	12 (11%)	17 (14%)	36 (21%)	91 (13%)
To the left		5 (3%)	14 (9%)	14 (12%)	15 (12%)	24 (14%)	72 (10%)

[a] Asymmetrical limited hip flexion (ALHF < 90°): including those with no missing hip flexion value.
[b] Scoliosis: including those with moderate and severe scoliosis.
[c] Windswept hip: distortion including those with no missing values for hip abduction or internal and external rotation.

2.3.2. Posture and postural ability scale

Seated posture was assessed using items from the Posture and Postural Ability Scale (PPAS) (Rodby-Bousquet et al., 2014), which has high psychometric properties for adults with cerebral palsy (Rodby-Bousquet et al., 2014). The items that were used were neutral pelvis, symmetrical trunk, and even weight distribution. Symmetry was noted as yes, and asymmetry was noted as no. In the CPUP registry, scoliosis was rated as no scoliosis, mild, moderate, or severe using a clinical spinal examination that has high psychometric properties for neuromuscular scoliosis in individuals with cerebral palsy (Persson-Bunke, Czuba, Hägglund, & Rodby-Bousquet, 2015). In this study, mild or no scoliosis was treated as not having scoliosis. Individuals operated for scoliosis were included in the study. If they were reported to have a remaining moderate or severe scoliosis after surgery they were included in the scoliosis group, if not, they were treated as not having a scoliosis. The direction of the lowest lateral curve of scoliosis was noted. Those with missing values for either the degree or the direction of scoliosis were excluded.

2.3.3. Range of motion

Passive joint range of movement for hip flexion, abduction, and external and internal rotation were measured with a goniometer in a standardised supine position according to the manual. Asymmetrical limited hip flexion was defined as a range of hip movement that did not exceed 90° of hip flexion and was reduced by at least 5° compared with the contralateral side. The selection of 5° difference was based on results from Boone et al. (1978).

2.3.4. Windswept hip distortion

The windswept hip distortion consists of abduction and external rotation of one hip, with the opposite hip in adduction and internal rotation. The presence of windswept hip distortion was confirmed by using Persson–Bunke's method (Persson-Bunke, Hägglund, & Lauge-Pedersen, 2006) and the direction of the windswept distortion was confirmed by using modified version of Porters method (Porter et al., 2007). Both methods are modified from Young's work (Young et al., 1998). Those who were not windswept according to both methods were determined not windswept. In Persson-Bunkes method at least 50% difference were needed to be between right and left side in either: hip abduction, hip internal or hip external rotation, to define the presence of windswept hip distortion. Values lower than 0.5 or higher than 2, were considered windswept. In Porters modified version the hip range of movement (hip abduction, hip external rotation and hip internal rotation on each side were added together. Then left side were subtracted from the right side. Threshold value was 20°, plus value 20° or higher indicated windswept over to the right and a negative value −20° or lower indicated windswept over to the left. Hip internal rotation is in opposite direction to hip abduction and hip external rotation and has therefore a minus value. Values for each side are added together.

2.4. Statistical analyses

The chi-square (χ^2) test, a nonparametric test, was used to analyse the relationship between variables. The significance level was set at $p < 0.05$. Logistic regression was used to estimate the association between ALHF < 90° and the GMFCS levels, postural asymmetries in sitting, scoliosis, windswept hip distortion, the direction of lateral spinal curvature caused by scoliosis, and the direction of windswept hip distortion. The results were presented as odds ratios (OR), which are ratios between two odds and express the probability that an event will occur, with 95% confidence intervals (95% CI). SAS Enterprise Guide 7.11 was used for the statistical analyses (SAS Institute Inc., Cary, NC, USA).

Table 2
Percentage of individuals with more than 90° bilateral hip flexion or asymmetrical limited hip flexion, relative to asymmetrical trunk, oblique pelvis, uneven weight distribution, scoliosis, and windswept hip distortion.

	Complete data set	> 90° bilateral hip flexion	Asymmetrical limited hip flexion (< 90°)	Chi square test
Asymmetrical trunk	N = 631	211 (37%) N = 577	39 (72%) N = 54	$p < 0.001$
Oblique pelvis	N = 621	199 (35%) N = 566	40 (72%) N = 55	$p < 0.001$
Uneven weight distribution	N = 629	200 (35%) N = 574	36 (65%) N = 55	$p < 0.001$
Scoliosis	N = 286	67 (26%) N = 257	21 (72%) N = 29	$p = 0.019$
Windswept hip distortion	N = 336	81 (27%) N = 302	19 (56%) N = 34	$p < 0.001$

The percentage of individuals, within each group, who have an oblique pelvis, an asymmetrical trunk, a scoliosis, and a windswept hip distortion.
Total number of individuals who have an oblique pelvis, asymmetrical trunk, moderate or severe scoliosis, or windswept hip distortion, and information about the hip flexion range of movement are available.

3. Results

3.1. Prevalence of asymmetrical limited hip flexion

A clear escalation of asymmetries was seen with increasing GMFCS level (Table 1); i.e., they became more frequent as function decreased. Hip flexion was reported for 665 individuals, 63 of whom (9%) had ALHF < 90°. Seven individuals had symmetrical limited hip flexion (i.e. < 90° of hip flexion on both sides) and were excluded from the analyses. Of the 172 individuals younger than 20 years of age, four (2%) had an asymmetric limited hip flexion, versus 9% in the total group. No one under the age of 19 years (n = 113) had ALHF < 90°. The majority of individuals with asymmetrical limited hip flexion were classified at GMFCS level V (60%), and the right hip (n = 39) was more often limited than the left hip (n = 24). All individuals were included in the analyses, independent of their GMFCS level. A majority of the individuals for whom hip flexion values were missing in the registry were at GMFCS level V. Of the individuals with ALHF < 90°, 49 (78%) were classified as having spastic bilateral cerebral palsy, and five (8%) were classified as having spastic unilateral cerebral palsy. Values for calculation of windswept hip distortion: abduction, and internal and external rotation were reported for 336 individuals; the majority of missing values were for adults at functional level V.

3.2. Asymmetrical limited hip flexion and postural asymmetries

More individuals with ALHF < 90° had an oblique pelvis, an asymmetrical trunk, an uneven weight distribution, scoliosis, and a windswept hip distortion (Table 2) compared with those who had bilateral hip flexion > 90°. No association was found between the side of limited hip flexion and either the direction of convexity of the lower scoliotic curve ($p = 0.808$) or the windswept hip distortion ($p = 0.273$). Of those subjects with limited right hip flexion, the direction of convexity of the lower scoliotic curve was to the right in six and to the left in six. Of those subjects with limited left hip flexion, the direction of convexity of the lower scoliotic curve was to the right in five and to the left in four. The odds of having an ALHF < 90° were higher (OR 2.2, 95% CI: 1.7–2.8) for each GMFCS level compared with that for the functional level above. The odds of pelvic obliquity, trunk asymmetry, uneven weight distribution, scoliosis, and windswept hips in sitting were almost four times higher for adults with ALHF < 90° compared with those who had bilateral hip flexion > 90° (Table 3). After adjustment for GMFCS level, the odds of having an oblique pelvis, an asymmetrical trunk, a scoliosis, and windswept hip distortion were still significantly higher for those with ALHF < 90° compared with those who had bilateral hip flexion > 90° (Table 3).

4. Discussion

There was a clear association between ALHF < 90° and an asymmetric sitting posture, scoliosis, and windswept hip distortion in adults with cerebral palsy. ALHF < 90° was more frequent in adults with spastic bilateral cerebral palsy (78%) and at lower levels of motor function (level V).

Table 3
Odds Ratio (OR) for effect from asymmetrical limited hip flexion and the Gross Motor Function Classification System (GMFCS) score on items from PPAS (trunk, pelvis, and weight) in sitting, scoliosis, and windswept hip distortion.

Dependent variable	GMFCS	Asymmetrical limited hip flexion	Asymmetrical limited hip flexion adjusted for GMFCS
Asymmetrical trunk	1.9 (1.8–2.4)[a]	4.5 (2.4–8.4)[a]	2.1 (1.1–4.2)[a]
Oblique pelvis	1.9 (1.7–2.2)[a]	4.9 (2.7–9.1)[a]	2.6 (1.6–2.1)[a]
Uneven weight distribution	2.2 (1.9–2.5)[a]	3.5 (2.0–6.3)[a]	1.5 (0.8–2.9)
Scoliosis	2.8 (2,2–3.7)[a]	7.4 (3.1–17.6)[a]	3.7 (1.3–9.7)[a]
Windswept hip distortion	1.5 (1.3–1.8)[a]	3.5 (1.7–7.1)[a]	2.6 (1.2–5.4)[a]

The interaction between the GMFCS score and asymmetrical limited hip flexion was not significant.
[a] Significant predictor $p < 0.05$.

The odds were higher of having an oblique pelvis and asymmetrical trunk in those individuals who had ALHF < 90° than in those with bilateral hip flexion > 90°. The results support Pope's (Pope, 2007) theory that ALHF < 90° is associated with an oblique pelvis and an asymmetry in the trunk. Furthermore, it supports Porter's (Porter et al., 2007) hypothesis that lateral curvature of the spine is needed to straighten up the trunk. Individuals with a spinal fusion, cannot straighten up the trunk and will lean to the side while sitting, if ALHF < 90°, has not been compensated for in the seating system. Depending on the level of the fusion, they have a tendency to compensate this with a lateral flexion of the cervical spine. Therefore it is essential to accommodate the seating system for any restricted hip motion in order to align the posture.

The fact that 22% of individuals at GMFCS level V had ALHF < 90° might explain Rodby-Bousquet's (Rodby-Bousquet et al., 2013) unexpected findings that adults with cerebral palsy who had lower levels of motor function had more postural asymmetries present in the sitting position than when standing. However, in either case, postural support is necessary. ALHF < 90° is easily overlooked in the sitting position, because it may be compensated for by mobility in the pelvis and the spine, causing an oblique pelvis and an asymmetrical trunk. Limited hip flexion has no effect on standing posture; therefore, it will not enhance any postural asymmetries while standing.

The pelvis is the anatomical structure that is directly linked to the hip joint and can therefore be expected to be most affected by ALHF < 90° in the sitting position. From a purely biomechanical aspect, it can be speculated that ALHF < 90°, when present, is the main contributor to the development of an ipsilateral higher side of the pelvis (Pope, 2007). According to Letts (Letts, Shapiro, Mulder, & Klassen, 1984), there is a clear association between total hip dislocation and the ipsilateral high side in oblique pelvis, but this effect has not been demonstrated for a subluxated hip (Porter et al., 2007).

Previous studies have suggested that in one-fourth of cases, the windswept hip distortion originates from the spine rather than the hips (Hägglund, Lauge-Pedersen, Bunke, & Rodby-Bousquet, 2016; Letts et al., 1984), but the presence of ALHF < 90° as a causative factor of oblique pelvis was not eliminated in those studies.

ALHF < 90° does not show the typical contracture pattern because the hip would need to be immobilised in extension to result in limited hip flexion (Trudel & Uhthoff, 2000). A majority of those with ALHF < 90° were individuals with bilateral spastic cerebral palsy at GMFCS levels IV and V. These are the individuals expected to have little or no postural ability, which can cause a slumped seating posture with a posteriorly tilted pelvis leading to an open hip joint angle. Their inability to resist the effect of gravity will usually require additional support in the seating system to maintain an aligned posture.

The direction of windswept hip distortion was independent of the side of limited hip flexion, but, consistent with previous findings (Persson-Bunke et al., 2006), the highest number of individuals with windswept hip distortion were classified at GMFCS level V. Coxa valga is often seen in spastic hips, and the proportion of valgus-deformed proximal femoral epiphyses increases as GMFCS level decreases (Lee et al., 2010). It can be speculated that the hip joint accommodates deteriorating forces from spasticity and lack of hip extension because of prolonged periods spent sitting and lying, resulting in limited hip flexion movement.

There were several limitations to this study. The majority of participants with missing values for the calculation of limited hip flexion and windswept hip distortion were at GMFCS level V; over 50% had missing values for the windswept hip calculation. The reason for these missing values is likely the fact that individuals at GMFCS level V have more severe contractures and distortion of body parts, resulting in difficulties in performing an appropriate range of movement for measurements. Because a majority of the individuals in this study with asymmetrical limited hip flexion were at GMFCS level V, the number with ALHF < 90° and windswept hips was most likely underestimated. No hip radiographs or information about the direction of the oblique pelvis, windswept hips, or primary curvature was available. This limited the possibility of estimating the causal effect of asymmetrical limited hip flexion.

Further studies investigating the combined factors that limit hip flexion movement and the causal factors for the high side in oblique pelvis are required. From the above, it can be concluded that ALHF < 90° greatly affects the parameters of the seating posture (pelvis and trunk) and is associated with scoliosis and windswept hip distortion. No association was found between the side of ALHF < 90° and the direction of the lowest scoliotic curve or the direction of windswept hip distortion. The fact that individuals younger than 20 years are less likely to have ALHF < 90°, indicates that ALHF < 90° either develops after 20 years of age, or that younger individuals receive more appropriate interventions to maintain range of motion.

5. Conclusions

Individuals with cerebral palsy, who has asymmetrical limited hip flexion < 90°, are likely to be spastic bilateral at GMFCS level V. The odds of pelvic obliquity, trunk asymmetry, scoliosis, and windswept hip distortion in adults with ALHF are higher than in those with bilateral hip flexion exceeding 90°. Oblique pelvis, asymmetric trunk, scoliosis, and windswept hip distortion are clinical signs of detrimental seating posture. ALHF need to be ruled out or compensated, especially in individuals with spastic bilateral CP at GMFCS level V, who are in a poor seating posture.

Competing interests

The authors declare that they have no competing interests.

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Paper III

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Preferred posture in lying and its association with scoliosis and windswept hips in adults with cerebral palsy

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ABSTRACT

Objective: The aim of this study was to clarify the association of scoliosis and windswept hips with immobility, lying position, and time in lying, in adults with cerebral palsy (CP).
Methods: This cross-sectional study included 830 adults (469 males and 361 females) with a diagnosis of CP, 16–73 years, and classified at levels I–V according to the Gross Motor Function Classification System (GMFCS). Subjects' Gross motor function classification system level, presence and severity of scoliosis, hip and knee joint range of movement, lying position, postural ability in lying, and time in lying were used to identify connections between them.
Results: Adults who are immobile in the lying position have higher odds of both scoliosis and windswept hips. Spending more than 8 h daily in the same lying position, increased the odds of having scoliosis, while lying solely in a supine position, resulted in higher odds of windswept hips.
Conclusions: The "preferred" habitual posture frequently observed in immobile adults with CP, leads to established distortion of their body shape. The results indicate the need for early introduction of appropriate posture control, in immobile individuals with CP, from a young age.

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Adult; cerebral palsy; scoliosis; wind-sweeping; hip

➤ IMPLICATIONS FOR REHABILITATION

- The preferred posture, observed in immobile adults with cerebral palsy, leads to a distortion of their body shape.
- One in four adults with cerebral palsy use only one position when in bed.
- The results indicate the need for early introduction of appropriate posture control in individuals unable to change position.

Introduction

Cerebral palsy (CP) is a neurological disorder, caused by a non-progressive brain injury, in the developing foetal or infant brain [1]. Even though the brain injury is non-progressive, secondary complications are prevalent. Limited mobility and postural asymmetry predispose to, for example, tissue adaptation, scoliosis, pelvic obliquity, hip sub/dislocation, and wind-sweeping particularly in the non-ambulant individual [2]. These complications increase the risk of further problems such as pain, pressure ulcers, respiratory, and urinary tract infections [3].

Preferred posture is used to describe a habitual posture, that is, one where the body returns to its original attitude after correction or change of position [3]. It indicates that the tissues of the body have adapted physiologically (plastic adaption) to a particular posture. It is deemed to be the result of extended periods of time in the same position. In a person with disabilities unable to change position the preferred posture is compounded by gravity, leading to further tissue adaptation. There are indications that these preferred postures are established in unborn babies [4], and maintained after birth [5] in babies unable to change from a lying position.

It is the asymmetries of posture, resulting in unequal forces acting on the tissues, compounded by long periods in one position that are thought to predispose to the problems of wind-sweeping, scoliosis, and other distortions of body structure [3,6,7]. Evidence of an association between the severity of CP, hip dislocation, pelvic obliquity, and scoliosis [8] is weak. Others have shown that a child's preferred lying posture is associated with the direction of wind-sweeping, hip dislocation, and lateral scoliosis curve [2,7]. It has been suggested that prone and side-lying may increase the risk of scoliosis and recommend use of the supine position instead [9]. Rodby-Bousquet et al. found that the time spent in one position predisposes to the development of contractures and distortion [10] due to plastic adaptation of the body structures.

Letts et al. [11] defined a windblown hip syndrome as a triad of scoliosis, pelvic obliquity, and wind-swept hips, without defining windswept hips specifically. Lonstein and Beck [12] defined windswept hips as an abduction contracture in one hip combined with an adduction contracture of the other hip. This definition has been used by other researchers [2,13] until Persson-Bunke et al. [14] refined it by describing windswept hips as a combination of abduction and external rotation of one hip with the

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opposite hip in adduction and internal rotation. The prevalence of windswept hips is 18% in the CP population [15], and will, in its severest cases, lead to hip dislocation [2,11].

In a total population of children with CP in Sweden, the prevalence of scoliosis is 11%, which increases with Gross Motor Function Classification System (GMFCS) level and, most significantly, with age [16]. In young adults, with GMFCS levels IV–V, there is a 50% risk of scoliosis by the age of 18 years. When analysing the development of scoliosis in individuals with CP and windswept hips, it was found that a hip dislocation was followed by pelvic obliquity and then scoliosis in three out of four non-ambulatory individuals [11]. Contracture in the knee joint is frequently reported among adults with CP [17], up to 60% cannot fully extend one or both knees, and is associated with postural asymmetries in both supine and standing position [10].

The aim of this study was to examine the association of scoliosis and windswept hips with immobility, lying position, and time in lying, in all adults, 16 years and older, with CP reported into the Swedish national CP surveillance programme and registry (CPUP) [18]. Immobility was defined as inability to move within or change position. Lying position was defined as their default sleeping and resting position.

Methods

Ethical approval was granted by the Medical Research Ethics Committee at Lund University (LU 2009–341), and permission was obtained to extract data from the Swedish national CP surveillance Programme and registry (CPUP). All participants on the register consent to research based on reported data.

Design

This cross-sectional study was based on data from the CPUP registry in Sweden [18]. Data were extracted from the most recent reports for all adults with CP registered between 1 January 2013 and 31 December 2016. Inclusion and exclusion criteria for the CP-diagnosis were defined by the Surveillance of Cerebral Palsy in Europe [19] with the brain injury occurring before the age of two years. Subtypes were classified as spastic unilateral, spastic bilateral, ataxic, and dyskinetic CP.

Assessment protocol

All assessments were performed in a standardised manner by local physiotherapists and occupational therapists throughout the country. They were trained in using the standardised CPUP assessment form and its accompanying manual. Both are attainable on the CPUP website (http://cpup.se/in-english/manuals-and-evaluation-forms/). These assessments are included as part of the national surveillance programme and entered into the CPUP web-based registry. The schedule of assessments is based on age and levels of motor function using the expanded and revised version of the GMFCS, age band 12–18 years [20]. Adults classified at GMFCS III–V are examined every year, GMFCS II every second year, and adults at GMFCS I every third year. Even though GMFCS was developed for children, it has also been shown to be accurate for use in adults with CP [21,22].

Passive joint range of movement for knee extension, hip extension, abduction, and external and internal rotation was measured with a goniometer in a standardised supine position. Lying positions and time in lying were either self-reported by the individual or reported by caregivers who know the respective individuals very well. Lying positions were defined as sleeping and resting positions over a 24 h cycle as supine, prone, right side lying, or left side-lying with the possibility of choosing 1–4 positions. Time in lying in any one position was reported as less than 8 h, 8–12 h, or more than 12 h, within a 24 h period, with the possibility of choosing just one item.

Scoliosis

In CPUP, scoliosis was rated as no scoliosis, mild, moderate, or severe using a clinical spinal examination with high psychometric properties for neuromuscular scoliosis in individuals with CP [23]. In this study, mild scoliosis (a discrete curve only visible in forward bending) was treated as not having scoliosis. Individuals operated for scoliosis were included in the study. Those reported to have a remaining moderate or severe scoliosis after surgery were included in the scoliosis group, if not, they were treated as not having scoliosis.

Posture and Postural Ability Scale

The preferred posture and postural ability in lying were assessed using the 7-point ordinal scale of the Posture and Postural Ability Scale (PPAS) [24], which has high psychometric properties for adults with CP. Postural ability ranges from level "1 – unplaceable" to level "7 – able to move into and out of position". PPAS level 1–3 indicates that the individual has little or no ability to counteract gravity or change position and is referred to as immobile in this study. Individuals at level 3 can maintain a position when placed by another person but cannot move. Individuals at level 4–7 have the ability to move within a position and ultimately, change position.

Windswept hips

Windswept hips consist of abduction and external rotation of one hip, with the opposite hip in adduction and internal rotation. The presence of windswept hips was calculated from hip range of movement values using Persson-Bunke's formula for calculation, not through a clinical analysis of posture [14]. The direction of the windswept hips was confirmed by using a modified version of Porter's formula [2]. Both formulas are modified versions from Young's work [13]. Those who were not windswept according to both methods were determined as not windswept. In Persson-Bunke's method, at least 50% difference was needed between right and left side in either: hip abduction, hip internal, or hip external rotation, to define the presence of windswept hips. Values lower than 0.5 or higher than 2 were considered windswept. In Porter's modified version, the hip range of movement (hip abduction, hip external rotation, and hip internal rotation) on each side was added together (hip internal rotation, being in opposite direction to hip abduction and external rotation, having a minus value). The left side was then subtracted from the right side. Threshold value was 20°, plus value of 20° or higher indicated windswept over to the right, and a minus value of $-20°$ or lower indicated windswept to the left.

Statistical analyses

Spearman's correlation coefficients (r_s) were calculated for the association between GMFCS levels on one hand, and scoliosis and windswept hips on the other. The interpretation of the strength of the Spearman's correlation coefficient was: $r_s > 0.70$ strong relationship; $r_s = 0.70–0.30$ moderate relationship; and $r_s < 0.30$ weak relationship. Logistic regression analysis was used to investigate the association between variables. Scoliosis and windswept hips were used as outcome variables. Explanatory variables used for scoliosis were: windswept hips, immobility in lying position (PPAS level < 4), having only one lying position, inability to passively straighten legs, and spending >12 h in lying. Explanatory variables used for windswept hips were: scoliosis, immobility in lying position (PPAS level < 4), having only one lying position, inability to passively straighten legs, and spending >12 h in lying. The results were presented as odds ratios (OR), which are ratios between two odds that an event will occur, with 95% confidence intervals (95% CI). SAS Enterprise Guide 7.11 was used for the statistical analyses (SAS Institute Inc., Cary, NC). The first step in the logistic regression process was to calculate estimates of unadjusted OR for each outcome variable and all the explanatory variables. Those explanatory variables, that demonstrated significant unadjusted OR for each of the outcome variable, were used in a backward elimination regression process. For each of the outcome variables, the explanatory variables with the least non-significant *p* values were eliminated from the next regression model, until only significant values were left in the model. As the variables "passively straighten hips" and "passively straighten knees" were strongly correlated, only ability to "passively straighten knees" were used in the multiple regression models to avoid collinearity problems.

Results

In all, 830 adults with CP participated in the study (469 men and 361 women) at a median age of 23 years (range 16–73 years). The subjects' GMFCS ranged from level I ($n = 159$), II ($n = 185$), III ($n = 130$), IV ($n = 155$) to level V ($n = 201$). The distribution of participants' neurological subtypes was as follows: spastic unilateral ($n = 169$), spastic bilateral ($n = 461$), ataxic ($n = 29$), dyskinetic ($n = 103$), and mixed or unclassified subtypes ($n = 69$). Of the 830 adults in the CPUP database, 228 (27%) lay solely in one position when in bed, 135 (16%) were immobile in lying position, in 216 (26%) passive straightening of the hips (to zero degrees) was not possible and in (449 (54%) passive straightening of the knees was not possible (to zero degrees).

Of the 830 adults, 119 (14%) individuals had a scoliosis, 175 (21%) had windswept hips, and 35 (4%) individuals had both windswept hips and a scoliosis. With respect to passive straightening of hips and knees to zero degrees, passive straightening was not possible in 215 (26%) of hips and in 445 (54%) of knees. GMFCS levels showed a moderate correlation with scoliosis ($r_s = -0.39$, $p < 0.001$) but the correlation with windswept hips was weak ($r_s = -0.27$, $p < 0.001$).

A majority of those with scoliosis were classified at GMFCS level V and spent more than 8 h lying (Table 1). Almost one-third of individuals with scoliosis had solely one lying position, where the most frequent position was side-lying on the right side. In the majority of the individuals with scoliosis the knees could not be straightened passively (due to contracture) and half of the group with scoliosis were unable to change their lying position or passively straighten their hips (due to contracture) (Table 1).

The highest number of individuals with windswept hips was at GMFCS level V and the majority of those with windswept hips spent more than 8 h lying daily (Table 2). Of those with windswept hips, 21% had only one lying position, where the most frequent position was side-lying. In half the individuals with windswept hips, passive straightening of the knees was not possible and in 30% passive straightening of the hips was not possible (due to contracture in both cases). Of those with windswept hips, 30% were unable to change position (Table 2). Four of the seven with windswept hips at GMFCS level I, were classified as spastic unilateral.

Of the nine factors tested for association with scoliosis (Table 3), individuals who were immobile in lying position (PPAS < 4), had the highest unadjusted OR (8.4) of having scoliosis, when compared to those who had some movement in lying position. Those who only used one lying position did not have higher odds of scoliosis, compared to those who used alternative

Table 1. Distribution of adults with scoliosis at GMFCS-levels I–V, relative to their time spent in lying, lying position, hip and knee range of motion, and inability to move or change position as measured with PPAS.

	GMFCS level					
	I	II	III	IV	V	Total number
Scoliosis	2	6	12	29	70	119
Time in lying <8 h	1	3	3	2	1	10
Time in lying 8–12 h	1	3	8	25	53	90
Time in lying >2 h	0	0	1	2	16	19
One lying position	0	3	5	12	17	37
Supine	0	2	2	1	4	9
Prone	0	0	0	5	5	10
Side-lying (right)	0	0	3	6	3	12
Side-lying (left)	0	1	0	0	5	6
Lack of hip extension	0	1	5	11	33	50
Lack of knee extension	0	4	9	20	32	65
PPAS ability 1–3 (unable to change position)	0	0	1	3	53	57

GMFCS: gross motor function classification system; PPAS: posture and postural ability scale.

Table 2. Distribution of adults with windswept hip distortion at GMFCS-levels I–V, relative to their time spent in lying, lying position, hip and knee joint range of motion, and inability to change position as measured with PPAS.

	GMFCS level					
	I	II	III	IV	V	Total number
Windswept hip distortion	7	34	30	41	63	175
Time in lying <8 h	1	3	3	2	1	10
Time in lying 8–12 h	1	3	8	25	53	90
Time in lying >12 h	0	0	1	2	16	19
One lying position	0	2	3	13	17	35
Supine	0	1	2	1	4	8
Prone	0	0	0	5	5	10
Side-lying (right)	0	0	1	6	3	10
Side-lying (left)	0	1	0	0	5	6
Lack of hip extension	0	1	6	11	33	51
Lack of knee extension	0	4	9	22	53	88
PPAS ability 1–3	0	0	1	3	53	57

GMFCS: gross motor function classification system; PPAS: posture and postural ability scale.

Table 3. Unadjusted odds ratios (OR) values of having scoliosis for nine independent factors.

Effect (*n*)	OR	95 % CI	
Windswept (96)	2.18	1.38	3.45
Supine lying (119)	0.84	0.39	1.83
Prone lying (119)	1.89	0.90	3.98
Side lying (119)	0.94	0.61	1.45
Time lying (119)			
>12 h vs. <8 h	7.55	3.25	17.53
8–12 h vs. <8 h	3.27	1.66	6.44
Lack of hip extension (106)	2.78	1.82	4.25
Lack of knee extension (116)	2.99	1.90	4.70
PPAS lying <4 (119)	8.39	5.39	13.06

PPAS: posture and postural ability scale.

Table 4. Adjusted odds ratios (OR) values of having scoliosis for factors with significant adjusted OR.

Effect (n)	OR	95% CI	
PPAS lying <4 (116)	5.68	3.53	9.14
Lack of knee extension (116)	1.84	1.12	3.00
Time lying (116)			
>12 h vs. <8 h	2.93	1.16	7.39
8–12 h vs. <8 h	2.20	1.08	4.44

PPAS: posture and postural ability scale.

Table 5. Unadjusted odds ratios (OR) of having windswept hips for nine independent factors.

Effect (*n*)	OR	95% CI	
Scoliosis (160)	2.18	1.38	3.45
Supine (175)	2.18	1.24	3.83
Prone (175)	1.34	0.65	2.76
Side lying (175)	0.83	0.57	1.21
Time lying (170)			
>12 h vs. <8 h	3.30	1.69	6.44
8–12 h vs. <8 h	1.01	0.66	1.56
Lack of hip extension (160)	1.99	1.37	2.90
Lack of knee extension (174)	1.97	1.38	2.81
PPAS lying <4 (175)	3.60	2.36	5.51

PPAS: posture and postural ability scale.

Table 6. Adjusted odds ratios (OR) of having windswept hips for factors with significant adjusted OR.

Effect (*n*)	OR	95% CI	
PPAS lying <4 (174)	2.90	1.86	4.53
Supine (174)	1.86	1.03	3.34
Lack of knee extension (174)	1.58	1.09	2.29

PPAS: posture and postural ability scale.

lying positions. When adjusting the scoliosis OR values, for all other factors having a significant adjusted OR value (Table 4), those who could not change position independently still had the highest OR (5.7). In addition, in those individuals where passive straightening of the knees was not possible and in those who spent more than 8 h daily in lying, the adjusted OR values for scoliosis were significant.

Of the nine factors tested for association with windswept hips (Table 5), individuals who were immobile in lying position (PPAS <4), had the highest unadjusted OR (3.6), compared to those who had at least some movement in a lying position. Those who lay solely in supine position; also had higher odds of windswept hips compared with those who used other positions. When adjusting the OR values of windswept hips, for all other factors having a significant adjusted OR value (Table 6), those who were unable to change position in lying, passively straighten knees and who lay only in a supine position, had significant OR for windswept hips. GMFCS level, which is a predictor of scoliosis (OR =2.6, 95% CI =2.1–3.1 for each increase in level by one) and windswept hips (OR =1.6, 95% CI =1.4–1.8 for each increase in level by one), was left out of the regression analysis, as the impact of other predictors, on scoliosis and windswept hips, were the focus of this study.

Discussion

The foremost finding of this study indicates that adults with CP who are immobile in a lying position have higher odds of both scoliosis and windswept hips than those who are more mobile. Lying (in one or more positions) for more than 8 h, increased the odds of having scoliosis, while lying solely in a supine position, gives higher odds of windswept hips. The results from this study are consistent with the findings, in previous work [23,24] in babies and young children with disabilities, that inability to change position in lying results in higher odds of developing both scoliosis and windswept hips. Those who spend longer time in lying, have higher odds of scoliosis.

It has been suggested that side-lying increases the risk and severity of scoliosis [9] due to instability resulting from a narrow base of support and the degree of mobility between body segments. The advice given is to avoid side lying in immobile individuals and use the supine position instead. The results of this study do not support the view that individuals with CP, who only lie on their side have higher odds of scoliosis, than those who only lie supine or use alternative positions. Knee contracture with inability to passively straighten the knees (to zero degrees) was the most common secondary complication of the body in this study. Those whose knees could not be straightened passively have higher odds of scoliosis and windswept hips. In the disabled person with CP lying supine and unable to move or change position, the knees tend to flex and fall to the same side resulting in tissue adaptation, established contracture over time and predisposing to windswept hips. Even the smallest unilateral flexion contracture of a knee, creates an apparent leg length discrepancy, which is standing and walking, leads to an oblique pelvis and eventual scoliosis.

Only a small difference was found between the 21% prevalence of windswept hips in this study and the previously reported prevalence (18%) of windswept hips in children with CP [15]. This study included both ambulatory and non-ambulatory individuals with CP, which is a novelty in studies of windswept hips. Although unexpected, seven individuals at GMFCS level I had windswept hips, according to the range of movement criteria used in this study, which suggests, that a more specific definition of windswept hips is required. Neither Lonstein and Beck [12] nor Persson-Bunke et al. [14] included a rotation of pelvis to the same side as the knees, in their definition of windswept hips. The findings in this study support the conclusion of Hägglund et al. [15] that development of windswept hips is caused initially by knee flexion with fall of the legs to one side in a supine position. Thus, it is not the knee flexion per se, that causes the windswept hips and it is likely the persistent fall of the knees to one side which, in consequence, rotates the pelvis to the same side.

In this study, the prevalence for scoliosis was found to be 14% in adults with CP compared with 11% in the total Swedish population of children with CP [16]. It was unexpected to find that only 30% of the individuals with scoliosis also had windswept hips, compared to 43% in Young et al.'s study [13], resulting in a weaker correlation between scoliosis and windswept hips than anticipated. This may be explained by the CPUP surveillance programme, as it was founded to prevent hip dislocations. The lower number of windswept hips, among individuals with scoliosis, might be partially explained by the reduced number of hip dislocations [25].

Conclusion

The preferred posture, frequently observed in immobile adults with CP, leads to a distortion of their body shape. This posture becomes habitual and is compounded with time in lying and gravity. The results strengthen the need for adequate and appropriate postural support in lying from a young age, in individuals unable to change their posture and position.

Disclosure statement

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Paper IV

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Full length article

Validity and reliability of an iPad with a three-dimensional camera for posture imaging[☆]

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ABSTRACT

Background: It is important to quantify a static posture to evaluate the need for and effectiveness of interventions such as physical management, physiotherapy, spinal orthosis or surgical treatment on the alignment of body segments. Motion analysis systems can be used for this purpose, but they are expensive, require a high degree of technical experience and are not easily accessible. A simpler method is needed to quantify static posture. Research objective: Assess validity and inter and intra rater reliability using an iPad with a 3-D camera to evaluate posture and postural deformity.
Method: A 3-D model of a lying posture, created using an iPad with a 3-D camera, was compared to a Qualisys motion analysis system of the same lying posture, the latter used as the gold standard. Markers on the trunk and the leg were captured by both systems, and results from distance and angle measurements were compared.
Results: All intra-class correlation coefficient values were above 0.98, the highest systematic error was 4.3 mm for length measurements and 0.2° for angle measurements. Significance: A 3-D model of a person, with markers on anatomical landmarks, created with an iPad with a 3-D camera, is a valid and reliable method of quantifying static posture.
Conclusion: An iPad with a 3-D camera is a relatively inexpensive, valid and reliable method to quantify static posture in a clinical environment.

1. Introduction

Posture refers to the position and shape that the body adopts when it is relaxed or during activities. “Good” posture is generally perceived as one that is erect and symmetrically aligned, but it is more accurately defined as a posture that facilitates effective energy conserving function without damaging the body [1]. Posture can have a huge impact on health, as it is potentially damaging to the body system. Any posture, adopted for sustained periods, will put some tissue under stress leading to tissue adaptation, and ultimately contracture and deformity. In individuals with cerebral palsy and lower levels of motor function, postural asymmetries are frequently observed and these asymmetries are associated with secondary complications such as limited range of motion, and scoliosis [2].

The importance of quantifying posture in health and in disease, has been highlighted by several authors [3]. This is required to evaluate the effectiveness of interventions such as physical management, physiotherapy, spinal orthosis or surgical treatment on the alignment of body segments. In order to quantify posture in supine- prone- or side-lying, a reference posture is needed. Usually the anatomical position is considered as neutral (zero-positions) when standing straight with arms by side, thumbs directed forwards, the functional longitudinal axes of the feet parallel and separated by a space equal to the distance between the hips, and the gaze directed forwards and horizontally [4].

In practice, measuring people with deformity and/or displaced body parts in the lying position, such as scoliosis and windswept hips, is not easy to perform with accuracy. This is due to difficulties in identifying the normal anatomical points in the person with significant deformity

[☆] This study focuses on the validity and reliability of the use of an iPad with 3-D camera, to evaluate posture in a quantifying way.

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with reference to the corresponding normal zero-position within each of the cardinal planes. Clinical experience has found that angle or distance measurements of deformed body parts cannot be carried out accurately from photographs, due to errors within the camera system, the parallax error and the projection error [5]. Motion analysis system, such as Qualisys, Optotrak, Vicon and Motion Analysis, have been used to assess posture in three dimensions, in a reliable and valid way [3]. However, these systems are not easily accessible for most clinicians since they are expensive, require specialized trained technicians and complex data processing. It is essential for clinicians to have access to valid and reliable clinical measures to properly evaluate effectiveness of interventions used to improve posture [3]. Sato [6] used a two-camera system for evaluating trunk deformity, based on 3-D measurements of body surface landmarks in the frontal plane, expressing rotational and lateral flexion angles between the upper and the lower thorax and between the lower thorax and pelvis. This method is too complicated for use in the clinical environment, as accuracy depends on the camera's resolution, distance to the imaging volume, camera separation and quality of calibration. Total body exposure is not possible with high image resolution and the process involves complex calculation of direct linear transformation parameters [7], which reflects the relationship between the 3-D coordinates of the calibrated volume and the 2D coordinate of the markers within the lens of each camera.

Few years ago, Occipital Inc (USA) made available 3-D scanner (Structure Sensor) that uses a grid of laser to generate a 3-D image of a stationary object within the visual field. This has been marketed as a "3-D camera for iPad". iPad with a 3-D camera is a handheld system that digitally maps the environment (surface topography). The system has the potential to be ideal for the use in clinical environments as the iPad with the 3-D camera does not need any calibration and is designed to be moved around while capturing surface topography. The purpose of the present study was to assess validity, and inter and intra rater reliability, in the use of iPad with a 3-D camera to evaluate posture and postural deformity, for use in the clinical environment.

2. Methods

2.1. Participants

Seven healthy adult volunteers were recruited among colleagues and friends to test validity and reliability of a new method used to evaluate posture and postural deformity. The first participant was used as a pilot to identify landmarks and reference points in two different postures. For the remaining six participants the measuring protocol included four supine postures, starting with supine lying in neutral zero-position; bent hips and knees in the second posture; progressing to an imitated windswept posture in the third and adding scoliosis to the imitated windswept hip deformity in the fourth measurement.

2.2. Instrumentation

The new system consists of a 3-D camera (Structure Sensor, Occipital Inc, USA), integrated with 4th generation version of iPad (Apple, USA), an iPad app (Structure, Occipital Inc, USA) and two computer software Skanect (Occipital Inc, USA) and Cloud Compare (CloudCompare v2.9.1 software, http://www.cloudcompare.org) on a PC computer. The 3-D camera resolution is 320 × 240 pixels (QVGA), sampling at 60 Hz with a precision of 0.5 mm at 400 mm distance from the iPad to the subject. As a gold standard for validation, an 8-camera three-dimensional Qualisys Oqus 300 motion capture system (Qualisys AB, Sweden) and computer software (Qualisys track manager (QTM) Qualisys AB, Sweden) simultaneously recorded markers position, was used for comparison. Qualisys system resolution was 1280 × 1024 pixels sampling at 200 Hz with a precision of ± 1 mm inside the calibrated volume.

2.3. Local frame of reference

The markers used to identify each landmark were Qualisys super spherical reflective markers, 8 mm in diameter, though reflective markers are not feasible to be used with the 3-D camera on the iPad. The 3-D camera sends out a grid of laser onto the environment in front of the iPad while the 3-D camera infrared sensor gathers the reflective light and depth data is generated, from which a digital map of the environment is built. The laser lights up the reflective markers which constantly reflect light to the infrared sensor, causing "shorter" depth to the markers. New layers of surface are laid down on the digital map, while the markers are illuminated. Markers that are not in full view will need longer exposure time using the 3-D camera, as it takes longer time to orient the 3-D camera to capture those markers. However, longer exposure time tends to lead to bigger out of shape marker in the 3-D model.

The 3-D coordinates of the following anatomical landmarks were used to evaluate the orientation of the upper trunk, the lower trunk and the pelvis: right and left: coracoid process, costal margin around the 10th rib and anterior superior iliac spine (ASIS) [1,6]. The x-axis within the local frame was defined as the direction from posterior to anterior when a participant was lying in a supine position, the y-axis was the distal to proximal direction and the z-axis was from medial to lateral (left to right direction).

For the upper trunk local frame, the z-axis was defined as the normalized vector between the left and right coracoid process markers. For the lower trunk local frame, the z-axis was defined as the normalized vector between the left and right lower rib markers. For both the upper trunk and lower trunk local frames, the temporary y-axis was defined as the normalized vector between the mid-shoulder markers and the mid-rib markers. For pelvis local frame, the z-axis was defined as the normalized vector between the left and right ASIS markers; the temporary y-axis was defined as the normalized vector between the mid-rib markers and the mid-pelvis markers. For all three local frames (upper trunk, lower trunk and pelvis), the x-axis was defined by the cross product of the temporary y-axis and the z-axis, and the final y-axis was then defined by the cross product of the x-axis and the z-axis.

Trunk symmetry [1] measures for quantifying scoliosis, is a standardized measurement in the Swedish national surveillance program and quality registry for cerebral palsy (CPUP) [8], by measuring distance, vertically and diagonally, between the coracoid processes and the ASIS.

The 3-D coordinates of following anatomical landmarks were used for evaluating the orientation of left thigh and shank: the greater trochanter, the lateral and medial epicondyle of the femur, the lateral and medial malleolus. For thigh local frame, the z-axis was defined as the normalized vector between the lateral and medial epicondyle markers. The temporary y-axis was defined as the normalized vector between the greater trochanter marker and the lateral epicondyle marker, and the x-axis was defined by the cross product of the temporary y-axis and the z-axis. The final y-axis was then defined by the cross product between the x-axis and the z-axis. For shank local frame, the z-axis was defined as the normalized vector between the lateral and medial malleolus markers. The y-axis was defined as the normalized vector between the lateral epicondyle marker and the lateral malleolus marker. The x-axis was defined by the cross product of the y-axis and the z-axis.

2.4. Protocol

Each participant lay supine on a plinth. The measurements were taken at the movement analysis laboratory by the same experienced physiotherapist and biomedical engineer. Although the recording time was different, five seconds for the Qualisys and more than a minute for the iPad, both systems started recording participants simultaneously. The output from the two systems have different formats, the Qualisys system output are markers location in 3-D space while the iPad systems

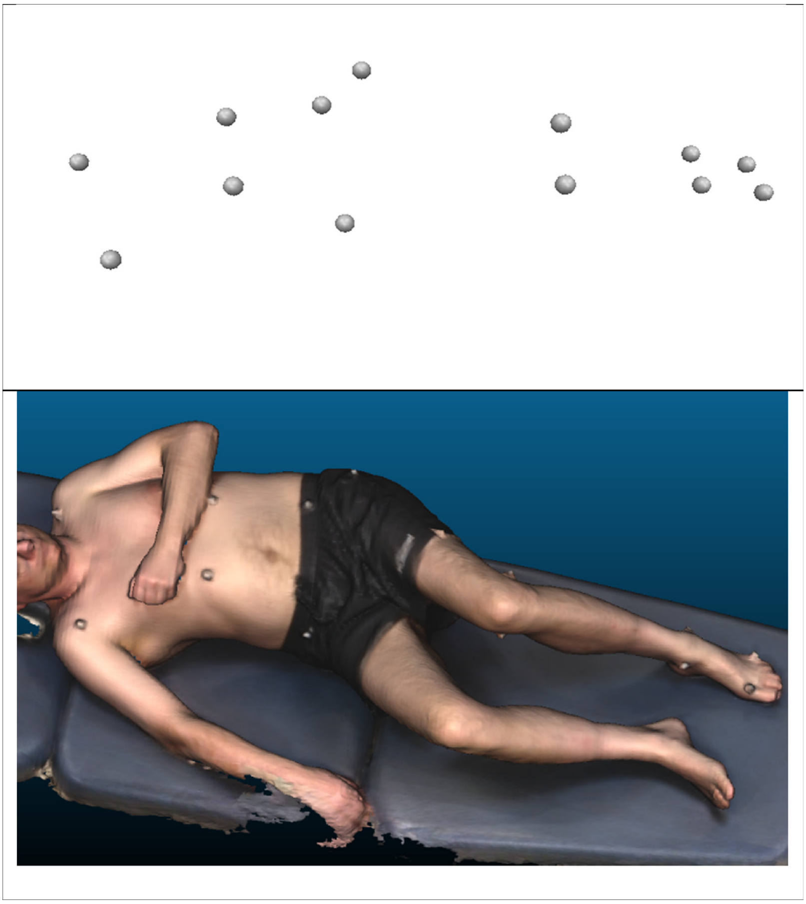

Fig. 1. a) Output from the Qualisys system; b) Output from the iPad system.

output is a 3-D model of the individual posture (Fig. 1).

During the iPad 3-D scanning, the iPad with the 3-D camera, was hand-held and carried two times around the plinth as a surface topography of the participant lying in supine appeared on the iPad screen. The distance from the iPad to the participant varied within every session, between 400 mm to 1000 mm. The recording time was about 1 min. The surface topography was simultaneously streamed into the Skanect software on a PC computer, via Wi-Fi uplink, for storage. During post processing in the Skanect software was the surface topography reconstructed and fused into a 3-D model and exported on a PLY format. The PLY file was opened in CloudCompare v2.9.1 software, (http://www.cloudcompare.org) where the markers were identified and its position in 3-D space manually digitised on the surface of every marker and exported as an ASCII file.

The Qualisys Oqus 300 captured the markers position in 3-D space and the QTM digitised the markers position in the centre of each of the marker. Recording time 5 s. During post processing in QTM markers in every posture (session) were manually identified and each session was exported on a C3D format (https://www.c3d.org/). Both the manual identification of markers in the QTM and the manually digitisation of markers in the Cloud Compare software, are the most critical factors to accurately determine the markers location.

2.5. Data analysis

Matlab (Version R2017b, Mathworks Inc, Natick, USA) script calculated trunk and knee angles (degrees) and trunk symmetry lengths (mm) from data in the C3D and ASCII files. One frame, frame 100 in the C3D file, was used in the calculation. Three-dimensional coordinate systems were created for each segment based on the marker position. Unit direction vectors of each segment were placed into a matrix representing a three-dimensional rotation matrix for each segment. The rotation matrix was a 3 × 3 matrix on the form

$$R = \begin{bmatrix} R_{11} & R_{12} & R_{13} \\ R_{21} & R_{22} & R_{23} \\ R_{31} & R_{32} & R_{33} \end{bmatrix}$$

The rotation of each segment was calculated using the formula

$$\varphi = \tan^{-1}\left(\frac{R_{21}}{R_{11}}\right)$$

$$\theta = \tan^{-1}\left(\frac{-R_{31}}{\sqrt{R_{11}^2 + R_{21}^2}}\right)$$

$$\psi = \tan^{-1}\left(\frac{R_{32}}{R_{33}}\right)$$

where φ represented flexion/extension, θ represented ab/adduction and ψ represented internal/external rotation.

The joint angles were calculated as the relative three-dimensional rotation between interconnecting segments using the set of equations above. The joint angles were angles in degrees between upper trunk and lower trunk, angles between lower trunk and pelvis and angles between thigh and shank. The distance extracted from the analysis was the length in millimetres between the right coracoid process and right ASIS, right coracoid process and left ASIS, left coracoid process and left ASIS, left coracoid process and right ASIS. The output from Matlab was copied into Excel (Excel, Microsoft, USA) where the data was organized for statistical analysis.

A systematic digitizing bias at each marker (8 mm in diameter) can theoretically be between minus 4 mm to plus 4 mm, and thus ± 8 mm for calculation of a length between two markers. The Qualisys system digitizes in the centre of the marker while the iPad system digitises on the marker surface. Criterion related validity was established using the Qualisys system as gold standard, evaluated as concurrent validity since they were measured at the same time. The evaluation of intra-rater reliability was based on data from three repeated measurements when digitising the markers of the same 3-D model in the CloudCompare software by the same physiotherapist. The analysis of inter-rater reliability was based on data from three experienced physiotherapists, who digitised the markers in the CloudCompare software, for each measurement. A limits of agreement plot (Excel, Microsoft, USA) was used to evaluate bias between the mean differences of the measurements and to estimate the agreement interval, within which 95% of the differences of the mean lie. Mixed model ANOVA SAS 9.4 (SAS Institute Inc, USA) was used to evaluate the effects of the mean differences of the measurements, i.e. the systematic error and its significance level, and to calculate the intraclass correlation coefficient (ICC(2,k) and ICC(3,k) [9] together with the standard error of measurement (SEM).

3. Results

The seven volunteers, aged 20–57 years (4 females, 3 males), generated a total of 26 measurements of supine postures. Four of these measurements had missing markers in the Qualisys system data and were not included in the analysis, leaving 22 complete measurements. Table 1 shows the parameters of the intra-rater and inter-rater reliabilities (ICC, SEM and systemic error) for four selected measurements using the iPad system, as well as the validity parameters (iPad - Qualisys comparison). These four measurements are the length measurements (Fig. 2), Upper-Lower trunk angles (Fig. 3), Lower-Pelvis angles (Fig. 4) and knee angles (Fig. 5).

All ICCs were above 0.98. SEM for the length measurements was lowest for intra-rater reliability, 2.3 mm, and slightly higher for the inter-rater and validity comparisons. Systematic error was 4.3 mm for the validity comparison but less than half of that for the reliability comparisons. For the three angle measurements, SEM were less than 1° for Upper-Lower and Lower-Pelvis angles and 1.5° or less for knee angles. Systematic error was 0.2° or less and non-significant in all analyses.

4. Discussion

The aim of this study was to assess intra- and inter-rater reliability and validity in the use of an iPad with a 3-D camera to evaluate posture and postural deformity. Our results show that, in quantifying posture, there is a high correlation between the outcome from iPad with a 3-D camera and the Qualisys system. The result thus clearly indicates that the use of iPad with 3-D camera can be used in clinical environments, with sufficient accuracy for quantified evaluation of posture and postural deformity. There is no need for calibration of the volume when using 3-D camera and iPad and total body 3-D models are readily analysed. Furthermore, 3-D models of an individual can be saved for reference later. The intraclass correlation coefficient (ICC) (Table 1-4) indicates excellent assessment of accuracy and consistency, both between the Qualisys system and the iPad system (validity), and the intra- and inter-rater reliability. ICC values from 0.98 indicate that there is an excellent reproducibility by the iPad system.

Table 1
Agreement between raters and measurements.

	ICC(2,1)	ICC(3,1)	SEM	Systematic error	p
Trunk symmetry measurements					
Intra-Reliability	0.997	0.996	2.34	0.8 mm[a]	< .001
Inter-Reliability	0.994	0.997	2.92	1.9 mm[b]	< .001
Validity	0.995	0.991	2.80	4.3 mm[c]	< .001
Upper-Lower trunk angle					
Intra-Reliability	0.992	0.992	0.45	0.1°	0.779
Inter-Reliability	0.990	0.991	0.51	−0.1°	0.808
Validity	0.982	0.982	0.69	−0.1°	0.475
Lower-Pelvis trunk angle					
Intra-Reliability	0.999	0.999	0.45	−0.1°	0.225
Inter-Reliability	0.998	0.998	0.62	0.1°	0.469
Validity	0.997	0.997	0.72	0.2°	0.131
Thigh-Shank (knee) angle					
Intra-Reliability	0.997	0.997	1.29	−0.1°	0.144
Inter-Reliability	0.997	0.997	1.25	−0.2°	0.246
Validity	0.995	0.995	1.50	−0.1°	0.586

All trunk symmetry measurements are pooled together, all Thorax upper-lower angles are pooled together, all Thorax lower-pelvis angles are pooled together and all thigh-shank angles are pooled together.

[a] The systematic error is the average difference between the first and the second measurement and the first and the third measurement of the 3-D models.

[b] The systematic error is the average difference between Rater 1 and Rater 2, Rater 1 and Rater 3 and Rater 1 and Rater 4, measurement of the 3-D models.

[c] The systematic error is the difference between Qualisys system measurement and the IPad 3-D model measurement.

Cerebral palsy is a well-recognised neurodevelopmental condition that manifests in early childhood and persists throughout life [10]. It is the most common physical disability in childhood, with a prevalence of 2.0–2.4/1000 live births in Europe [11,12]. Although the need for quantification is common to a wide range of diseases and types of disability, it is especially valid in cerebral palsy due to the frequent occurrence of postural deformities. Even though the main objective of this study is to compare the accuracy and correlations between two methods quantifying posture in general, the test positions were particularly chosen with some of the most common postural problems of cerebral palsy in mind.

The difference in right and left vertical length, in the trunk symmetry measurement, indicates the degree of asymmetry in lateral flexion, whereas the difference in the right and left diagonal length indicates the degree of asymmetry in rotation of the trunk. Using surface topography reconstructed by iPad, the shortest distance between landmarks is automatically selected, filtering out error effect from the body itself, that will affect the measurement when using a standard measuring tape. The systemic error is less than 2 mm when assessing the reliability and is smaller than SEM (Fig. 2) indicating good precision of the system. In the validity measurement, the mean difference is 4.3 mm indicating that the output from the two systems, iPad with 3-D camera and Qualisys, are not too different. The reason for this difference arises from the systematic digitizing bias between the two systems, as they localize differently the reference point on the marker (see the method section for further details).

The conventional method to evaluate posture is to measure the joint angles of the human body [4] with a goniometer. The results from these conventional methods of measuring joint angles and calculating 3-D joint angles are usually not the same, due to different local frames of reference and the effect from the order of rotation in 3-D angle calculation [13]. In all angular measurements, the difference of mean

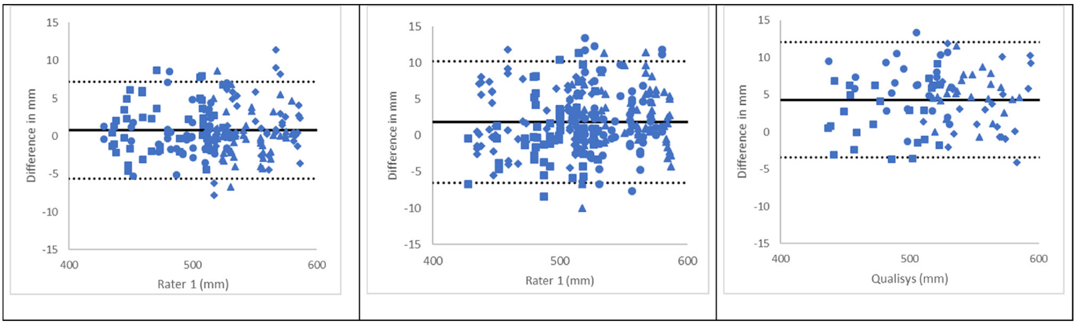

Fig. 2. Distances between: right coracoid process and right ASIS ■, right coracoid process and left ASIS▲, left coracoid process and left ASIS •, left coracoid process and right ASIS ♦. a) Intra-rater reliability (difference in length measurement between the first and the second measurement and the first and the third measurement of the IPads 3-D models, pooled in the graph); b) Inter-Rater reliability (difference in length measurement between Rater 1 and Rater 2, Rater 1 and Rater 3 and Rater 1 and Rater 4, of the IPads 3-D models, pooled in the graph); c) Validity (the difference of length measuremetns between the IPads 3-D model measurement and the Qualisys system measurement). Solid line shows the mean difference for the pooled data. Dashed lines show the 95% upper and lower limits of agreement representing two standard deviations above and below the mean difference.

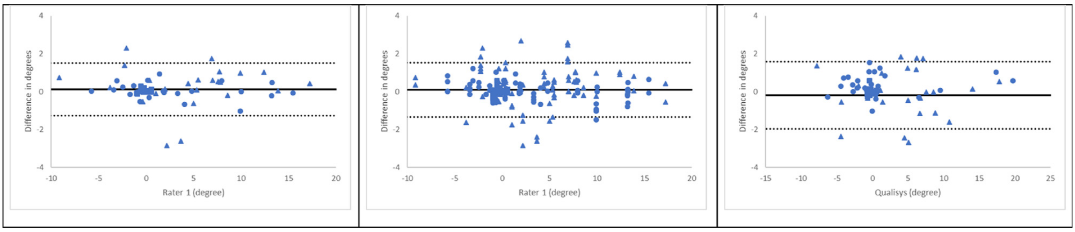

Fig. 3. Thorax Upper-Lower angles. Pooled angles: Flexion / Extension ■, Lateral flexion▲, Rotation •. a) See Fig. 2a for further explanation; b) See Fig. 2b for further explanation; c) Validity. The difference of calculated Upper-Lower angles between the IPads 3-D model measurement and the Qualisys system measurement. See Fig. 2c for further explanation.

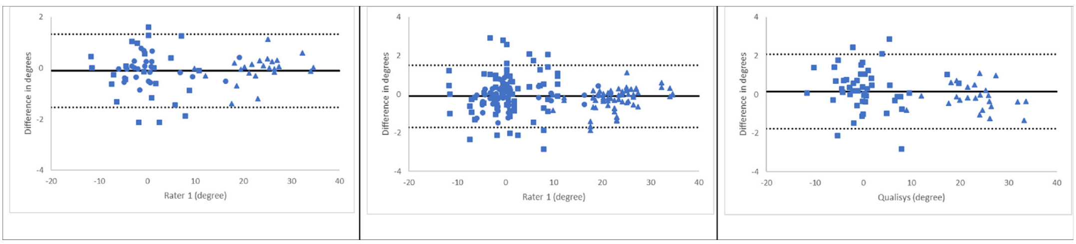

Fig. 4. Thorax Upper-Lower angles. Pooled angles: Flexion / Extension ■, Lateral flexion ▲, Rotation •. a) See Fig. 2a for further explanation; b) See Fig. 2b for further explanation; c) See Fig. 3c for further explanation.

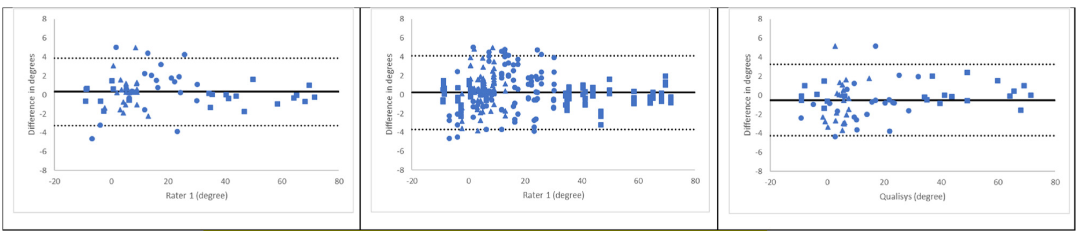

Fig. 5. Knee angles. Pooled angles: Flexion / Extension ■, Lateral flexion ▲, Rotation •. a). See Fig. 2a for further explanation; b) See Fig. 2b for further explanation; c) See Fig. 3c for further explanation.

between the gold standard (Qualisys) and the new iPad system, was negligent (Fig. 3–5). Each local reference segment, constructed from three markers on each segment, filters out the effect from the discrepancy of individual marker location, during angle calculation. In both trunk angle measurements, the SEM of the difference was lowest during intra-rater reliability test and highest during the validity test.

The range of the limits of agreement, in the trunk measurements, were around four degrees, which is well within any clinical environment error [14]. The SEM of the difference in the knee angle measurement results are higher than in the trunk angle measurement results, with the range of limits agreement being around eight degrees. The high digitizing error found at the knees and ankles, during imposed windswept posture, is due to big out of shape markers next to the plinth in the 3-D model. As the marker gets bigger and more out of shape, the digitizing bias increases as the distance from the markers surface to its centre gets longer.

Though the use of reflective marker was needed in this study for comparison with the gold standard measurement (Qualisys), it is not necessary in the clinical environment when the iPad system is used. In the clinical environment, any type of marker that is not reflective will hold its form during the surface topography. Markers that are not out of shape will lessen the digitizing error and smaller digitizing error would lead to similar reliability (SEM) in the knee angles as is in the trunk angles. This study suggests that the ideal marker for an iPad with 3-D camera postural evaluation would be a half spherical with a checkerboard pattern, in order to pinpoint accurate digitization.

A limitation of this study lies in the use of healthy subjects to evaluate the validity and reliability of the method for quantifying posture proposed for use with motor impairments such as CP. However, a 3-D model of an individual (Fig. 1), for evaluating posture and postural deformity, depends on visibility of anatomical landmarks, not on being able or disabled. There is no reason to expect that the validity and reliability of a 3-D model, differ between individuals with or without motor impairments. However, subjects unable to stay still while constructing surface topography are not candidates for this type of static posture evaluation.

5. Conclusion

The use of an iPad with a 3-D camera to evaluate posture and postural deformity shows a high correlation and good validity with the output from the Qualisys system, along with high intra- and inter-rater reliability. The results clearly indicate that the use of iPad with 3-D camera can be used in the clinical environment, with good accuracy, for evaluating posture and postural deformity.

Conflict of interest

The authors report no conflict of interest. The authors alone are responsible for the content and writing of the paper and the manuscript is not under consideration for publication elsewhere.

Author contribution

Atli Ágústsson: Designed the study, collected the data and analyzed the result, drafted, improved and revised the manuscript and approved the final draft.

Magnús Kjartan Gíslason: Designed the study, improved and revised the manuscript, and approved the final draft.

Páll Ingvarsson: Improved and revised the manuscript and approved the final draft.

Elisabet Rodby-Bousquet: Designed the study, improved and revised the manuscript, and approved the final draft.

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