# Assessment and management of the common postural characteristics of non-ambulant adults with cerebral palsy

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# Abstract

Adults with cerebral palsy (CP) are living longer and experience progressive musculoskeletal issues, impacting on pain, quality of life, function and carer burden. Non-ambulant adults with CP exhibit the highest risk for secondary complications and potential for shorter life expectancy.

In this thesis postural symmetry refers to a normally developed musculoskeletal system with no observable or functional deviation from symmetry. Postural asymmetry, thus refers to, any observable or functional deviation from symmetry with a particular focus on the ribcage, pelvis and hips.

An initial scoping review (**study 1**) explored the nature, prevalence and conservative management of postural asymmetries in non-ambulant adults with CP. It also synthesized measurement tools used to identify postural asymmetry and described conservative interventions selected for management. The review revealed that non-ambulant adults with CP suffer from significant postural asymmetry associated with windswept hips, scoliosis, pelvic obliquity and limb contractures and that there is a lack of standardised measurement tools and documented interventions.

The second study investigated the reliability (**study 2**) of a postural measurement tool, known as the Goldsmith Indices of Body Symmetry (GIofBS). The GIofBS provides a measure of thoracic shape and symmetry, hip and pelvic mobility and a windswept index (abduction /external rotation of one hip and adduction / internal rotation of the other hip). A cohort of 30 non-ambulant adults with CP participated in this study in which the GIofBS was found to have good to excellent inter and intra rater reliability. A control study was conducted which provided valuable comparative data, suggesting less thoracic cage symmetry and greater variation in hip joint range in those with CP.

The third study explored the relationship between postural asymmetry, as measured with the GIofBS, and radiographs of a similar anatomical area (**study 3**). Minimal correlation was found between the two measurement approaches, suggesting GIofBS constructs and radiographs are measuring two different aspects of postural asymmetry, both of which may have a place in the comprehensive management of non-ambulant adults with CP. The final two studies explored the stability of posture over an 18-month period (**study 4**) and the relationship between pain and postural asymmetry (**study 5**) in a subset of this cohort. Study 4 explored postural change in a case series of 10 non-ambulant adults with CP. Considerable postural asymmetry was evident in all participants at study onset with minimal change in some

GIofBS outcomes and fluctuations in other GIofBS outcomes between time points. Repeated measures of posture over the lifespan, using the GIofBS, would more accurately reflect the musculoskeletal status of individuals, potentially enabling evaluation of the effectiveness of interventions. Study 5 revealed that high levels of pain (both day and night) are evident in this population, as measured by proxy report using the Non-Communicating Adult Pain Checklist, and that pain may be related to more extreme measures of postural asymmetry. Pain remains challenging to assess and manage in adults with cognitive and communication deficits.

In conclusion, this thesis, provides new knowledge regarding measurement of asymmetry of the thoracic cage, pelvis and hips in non-ambulant adults with CP. This population of adults with CP experience disabling postural asymmetry which may be measured over the lifespan using the GIofBS as a suitable assessment tool. Measurement and monitoring of posture may aid in the evaluation of treatment efficacy and implementation, potentially improving the care for this vulnerable population.

Publications and presentations during candidature

## Journal Publications:

**Holmes C,** Kim Brock & Prue Morgan (2019) Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review, Disability and Rehabilitation, 41:9, 1079-1088, DOI: 10.1080/09638288.2017.1422037

**Holmes C**, Fredrickson E, Brock K, Morgan P (2020). The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy. Disability and Rehabilitation. 7:1-7. DOI: 10.1080/09638288.2019.1708979

**Holmes C,** Kim Brock & Prue Morgan (2020): Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study, Pediatric Physical Therapy, *In Press*, DOI: 10.1097/PEP.0000000000000787

**Holmes C,** Kim Brock & Prue Morgan (2021): Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study. Disability and Health Journal, *In Press,* DOI: 10.1016/j.dhjo.2021.101063 Online availability 20 January 2021

**Holmes C,** Kim Brock & Prue Morgan (2021): The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy. New Zealand Journal of Physiotherapy, *In Press,* DOI: 10.15619/NZJP/49.1.03

## Poster Presentations:

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**Holmes C**, Brock K, Morgan P. Identification of postural abnormality in adults with cerebral palsy. Research Week, St. Vincent's Hospital, Melbourne, Australia, August 2017.

**Holmes C**, Brock K, Morgan P. Postural asymmetry in non–ambulant adults with cerebral palsy: a scoping review. Research Week, St. Vincent's Hospital, Melbourne, Australia, August 2018.

**Holmes C**, Brock K, Morgan P. Identification of postural abnormality in young adults with cerebral palsy. American Academy for Cerebral Palsy and Developmental Medicine. 72nd Annual Meeting, Cincinnati, Ohio, October 2018.

**Holmes C**, Fredrickson E, Brock K, Morgan P. The intra and inter-rater reliability of a postural measurement tool in non-ambulant adults with cerebral palsy. Research Week, **St**. Vincent's Hospital, Melbourne, Australia, August 2019.

## Oral Presentations:

**Holmes C**, Fredrickson E, Brock K, Morgan P. The intra and inter-rater reliability of a postural measurement tool in non-ambulant adults with cerebral palsy. Research Week, St. Vincent's Hospital, Melbourne, Australia, August 2019.

**Holmes C**, Fredrickson E, Brock K, Morgan P. Identification of postural abnormality in young adults with cerebral palsy. Oceania Seating Symposium, Melbourne, Australia, November 2019.

**Holmes C**, Fredrickson E, Brock K, Morgan P. The intra and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy. Australasian Academy of Cerebral Palsy and Developmental Medicine 10th Biennial Scientific Conference, Perth, Australia, March 2020

**Holmes C**, Fredrickson E, Brock K, Morgan P. Reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy. Research Week, St. Vincent's Hospital Sydney, August 2020.

## Workshops:

**Holmes C,** Luscombe D. Identification, prevention and measurement of postural asymmetry. Oceania Seating Symposium, Melbourne, Australia, November 2019.

Morgan P, **Holmes C,** Tracy J. Enhancing the health of adults with cerebral palsy after transition from paediatric services. Australasian Academy of Cerebral Palsy and Developmental Medicine 10th Biennial Scientific Conference, Perth, Australia, March 2020.

Thesis including published works declaration

I hereby declare that this thesis contains no material which has been accepted for the award of any other degree or diploma at any university or equivalent institution and that, to the best of my knowledge and belief, this thesis contains no material previously published or written by another person, except where due reference is made in the text of the thesis.

This thesis includes 2 original papers published in peer reviewed journals, 2 publications *In Press* and 1 publication under final review. The core theme of the thesis is postural asymmetry in non-ambulant adults with cerebral palsy. The ideas, development and writing up of all the papers in the thesis were the principal responsibility of myself, the student, working within the Department of Physiotherapy under the supervision of Professor Prue Morgan and Dr. Kim Brock.

In the case of *Chapters 3, 5, 7, 9 and 11* my contribution to the work involved the following:

| **Thesis Chapter** | **Publication Title** | **Status** *(published, in press, accepted or returned for revision, submitted)* | **Nature and % of student contribution** | **Co-author name(s) Nature and % of Co-author's contribution*** | **Co-author(s), Monash student Y/N*** |
|---|---|---|---|---|---|
| 3 | Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review | Published | Led the study conception, methodological design, search strategy, study selection, data extraction, analysis, synthesis, and preparation and drafting of manuscript for publication. 70% | **Prue Morgan:** Contributed to conception, methodological design, data review, and preparation and drafting of manuscript for publication 20%<br>**Kim Brock:** Contributed to conception, methodological design, and preparation and drafting of manuscript for publication 10% | No<br>No |

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| 5 | The intra and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy | Published | Led the study conception, methodological design, data collection data, analysis, and preparation and drafting of manuscript for publication.<br>65% | **Prue Morgan:**<br>Contributed to conception, methodological design, data review, and preparation and drafting of manuscript for publication<br>20%<br>**Kim Brock:**<br>Contributed to conception, methodological design, and preparation and drafting of manuscript for publication<br>10%<br>**Emma Fredrickson:**<br>Contributed to data collection and review of manuscript for publication<br>5% | No<br><br>No<br><br>No |
| 7 | The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy. | Returned for revision | Led the study conception, methodological design, data collection data, analysis, and preparation and drafting of manuscript for publication.<br>70% | **Prue Morgan:**<br>Contributed to conception, methodological design, data review, and preparation and drafting of manuscript for publication<br>20%<br>**Kim Brock:**<br>Contributed to conception, methodological design, and preparation and drafting of manuscript for publication<br>10% | No<br><br>No |

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| 9 | Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study | In Press | Led the study conception, methodological design, data collection data, analysis, and preparation and drafting of manuscript for publication.<br>70% | **Prue Morgan:** Contributed to conception, methodological design, data review, and preparation and drafting of manuscript for publication<br>20%<br>**Kim Brock:** Contributed to conception, methodological design, and preparation and drafting of manuscript for publication<br>10% | No<br><br>No |
| 11 | Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study | In Press | Led the study conception, methodological design, data collection data, analysis, and preparation and drafting of manuscript for publication.<br>70% | **Prue Morgan:** Contributed to conception, methodological design, data review, and preparation and drafting of manuscript for publication<br>20%<br>**Kim Brock:** Contributed to conception, methodological design, and preparation and drafting of manuscript for publication<br>10% | No<br><br>No |

I have renumbered sections of submitted or published papers in order to generate a consistent presentation within the thesis.

**Student name:** Carlee Holmes

**Student signature:** **Date:** 23/03/2021

I hereby certify that the above declaration correctly reflects the nature and extent of the student's and co-authors' contributions to this work. In instances where I am not the responsible author I have consulted with the responsible author to agree on the respective contributions of the authors.

**Main Supervisor name:** Professor Prue Morgan

**Main Supervisor signature:** **Date:** 18/03/2021

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Acknowledgement must be given to the many participants, families and carers who readily gave up their time to attend the measurement appointments. This participation has provided knowledge on the measurement of postural asymmetry in non-ambulant adults with CP which will provide clinicians with an opportunity to measure posture and monitor the effect of interventions in this complex population across the lifespan.

On a personal level I would like to thank my family, Gwen and Frank Holmes, Andrea, Dave, Sam and Joel Hodder for believing in me and for the IT support. Thank you Dave. A special thank you to my boys, Jack and Max, for keeping me grounded and busy and not forgetting Roy the kelpie. Distraction is a good thing.

# Glossary of Abbreviations

| Abbreviation | Description |
|---|---|
| ABLAP | Angle Between Legs and Pelvis |
| CFCS | Communication Function Classification System |
| CP | Cerebral palsy |
| CT | Computed Tomography |
| ER/Abd | External Rotation / Abduction |
| GMFCS | Gross Motor Function Classification System |
| GIofBS | Goldsmith Indices of Body Symmetry |
| ICC | Intraclass Correlation Co-efficient |
| ICF | International Classification of Functioning, Disability and Health Framework |
| ITB | Intrathecal baclofen |
| IQR | Inter Quartile Range |
| JBI | Joanna Briggs Institute |
| MACS | Manual Ability Classification System |
| MCPHCS | Melbourne Cerebral Palsy Hip Classification System |
| MP | Migration percentage |
| MRI | Magnetic Resonance Imaging |
| NCAPC | Non-Communicating Adult Pain Checklist |
| NICE | National Institute for Health and Care Excellence |
| PPAS | Posture and Postural Ability Scale |
| RCH | Royal Children's Hospital |

| | |
|---|---|
| RESNA | Rehabilitation Engineering and Assistive Technology Society of North America |
| SD | Standard deviation |
| SVHM | St. Vincent's Hospital, Melbourne |
| YACDS | Young Adult Complex Disability Service |

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# Chapter 1:  General Introduction

This thesis has been completed by publication. All chapters have either been published in peer reviewed journals or in press. All manuscripts (published and in press) have been included in word format for consistency of formatting, with the provision of linking chapters to provide the reader with context to the publications and manuscripts. Relevant references are provided at the end of each chapter throughout the thesis, except for Introductory, Discussion/Conclusion and linking chapter references which are consolidated at the end of the thesis. PDF versions of published articles from the relevant journals are available in Appendix C.

## 1.1 Background

Cerebral palsy (CP) is a non-progressive disorder of movement and posture attributable to an impairment in the developing fetal or infant brain at a stage when brain maturation is rapid and thus, damage due to prematurity, hypoxia or infection is a high risk (1, 2) The rate of live births of infants with CP has declined in recent years from 2.1 to 1.4 per 1000 live births in Australia (3). In 2018 35,522 Australians were estimated to have CP, with this figure expected to increase to 46,000 by 2060 as the population grows (4). Of note, 3 in every 4 people with CP in Australia are currently over the age of 18 (5). Associated impairments are common with spasticity reported as the most frequent motor disorder noted in up to 92% of individuals. Dyskinetic CP (includes dystonic and athetoid/choreoathetoid CP) occurs in 7% of individuals with unilateral spasticity and in close to 20% of individuals with bilateral spasticity. In children with the predominant motor type of dyskinesia 41% were also described has having spasticity indicative of the prevalence of secondary and or co-occurring motor types Nearly half of those with CP have associated intellectual impairment, 29% have epilepsy, >60% have speech impairment, 35% have visual impairment and 11 % experience hearing impairments (4).

Children with CP are likely to live into adulthood (6) and despite the initial neurological lesions being static in nature, the secondary musculoskeletal consequences are progressive (7). Secondary impairments are most common in the more severely affected with the effects of ageing and functional decline occurring earlier in adults with CP than the general population (6). This may be related to early development of osteoporosis and arthritis, poor nutrition, sedentary lifestyles and resultant sarcopenia and fatigue (6, 8) setting up an ongoing cycle of progression and decline. Hospital admissions due to respiratory disease are more common in adults compared to children with CP (20% and 16% respectively) (9). This may be related to progressive asymmetry of the thoracic cage and contribute to adult mortality from diseases of the respiratory system (10). The effects of ageing impact earlier on adults with CP (6). Hence neither paediatric CP knowledge, nor knowledge gleaned from those without disability can necessarily be applied to the adult CP population who require specific guidelines for best practice throughout adulthood.

The impairments observed in people with CP result in activity limitations, that is, an inability to participate in activities of one's choice. Activity limitations for people with CP are further classified according to the extent of the underlying motor abnormalities, accompanying impairments, anatomical and neuro-imaging findings and causation and timing of the brain injury (4). Within these classifications, rating scales relevant to the impacted system are

employed such as the Gross Motor function Classification System (GMFCS) for functional mobility, the Manual Ability Classification System (MACS) for hand and arm function, and the Communication Function Classification System (CFCS) (4). Such classifications provide further insights into the extent to which activity limitations are affected.

The Gross Motor Function Classification System (GMFCS) is a standardised measure of gross motor function. Its use is well established in paediatric care (11). A later extended and revised version for adolescents (GMFCS E & R) (12) was developed, which is also accepted for use in adult CP populations (Figure 1.1). Use of the term GMFCS in this thesis thus refers to the extended and revised GMFCS E & R version. The GMFCS is a 5-level clinical classification system used by health professionals to describe the gross motor function of people with CP on the basis of self-initiated movement abilities. Particular emphasis is on evaluating sitting, walking, and wheeled mobility. Distinctions between levels are based on functional abilities; the need for walkers, crutches, wheelchairs, or canes / walking sticks; and to a much lesser extent, the actual quality of movement. This thesis focuses on non-ambulant young adults with CP in the non-ambulant categories of GMFCS levels IV and V.

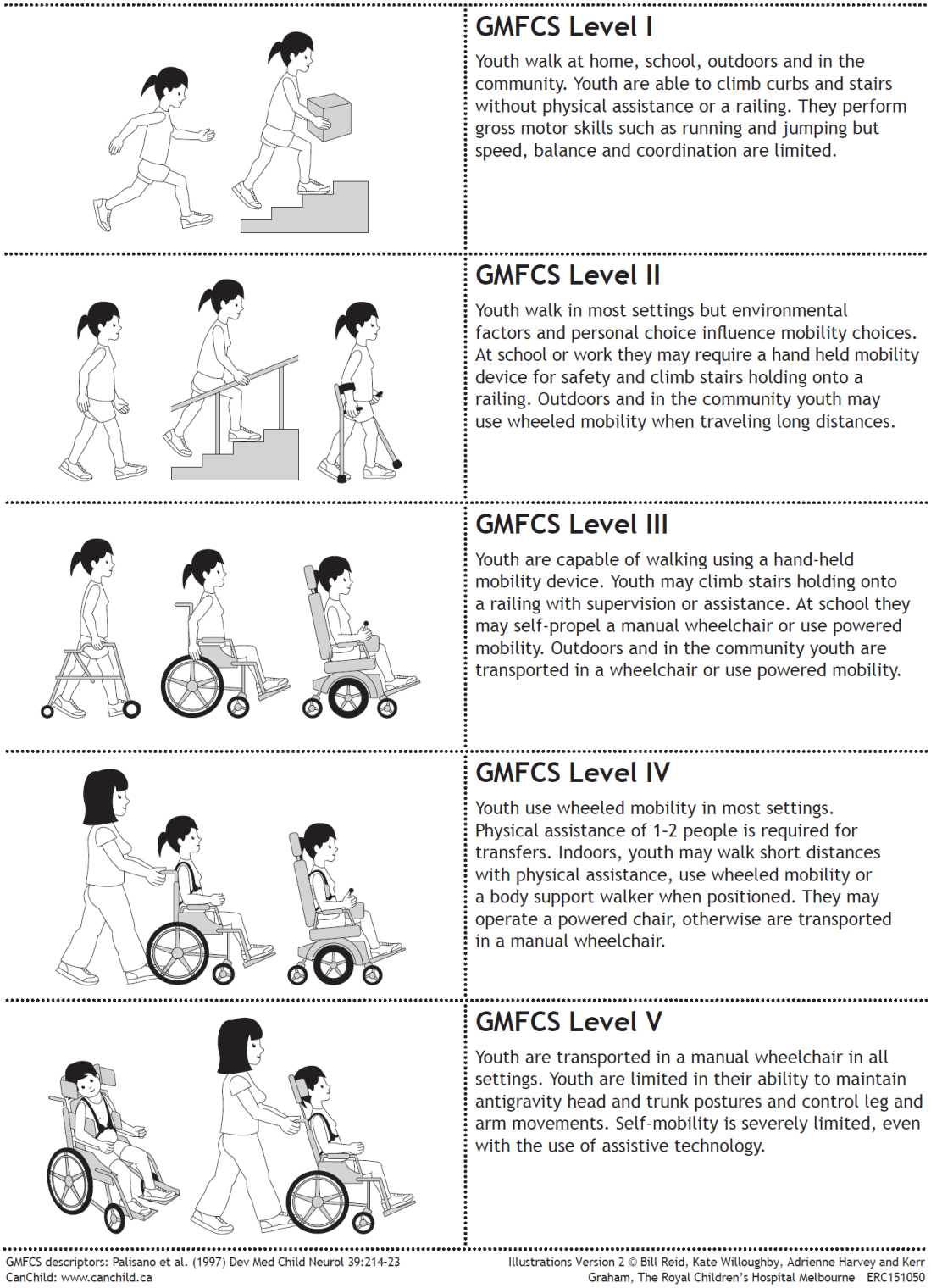


Figure 1.1. Gross Motor function Classification System

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## 1.2 Postural asymmetry in cerebral palsy

Postural asymmetries affecting the spine, pelvis and hips, inclusive of hip dislocation, are common in non-ambulant individuals with CP (13). The postural asymmetries in CP have been noted to be progressive, despite the relatively static nature of CP. Adults with complex disabilities are vulnerable to increasing postural asymmetry and the secondary complications associated with postural asymmetry. A snap shot of data suggests that at least 30% of non-ambulant adults with CP have a hip migration (migration percentage; MP) of greater than 30%, more than 75% have some degree of "scoliosis", and more than 40% demonstrate some amount of pelvic obliquity (13).

A further common postural problem in the adult CP population, known as "windsweeping" is characterised by abduction and external rotation of one lower limb and adduction and internal rotation of the opposite lower limb (Figure 1.3). Windswept hips are often accompanied by pelvic rotation in the transverse plane in the opposite direction to which the legs fall (14, 15). Windsweeping can be accompanied by associated postural asymmetries including scoliosis, pelvic obliquity and hip displacement (7) although contradictory evidence exists as to the specific relationships between the individual components.

Contractures of the hip and knee joints have been reported in up to 100% and 32% to 87% of non-ambulant adults with CP respectively (13). Contractures are defined as limited joint range of motion resulting from a shortening of muscles, tendons, ligaments and soft tissue with spasticity and immobility contributing factors (16)

Any one of these impairments on its own, or existing in a triad of deformities (scoliosis, pelvic obliquity and windswept hips), present as a significant treatment dilemma once established (17). These postural asymmetries increase the risk of soft tissue adaptation and pressure injuries, and the formation of contractures and progressive deformities, being most prevalent in non-ambulant people with CP (18). The inability to change position and the time spent in an asymmetrical posture contribute to the escalating problem in this population (18) with children who are unable to change position in supine reportedly having twice the risk of developing postural asymmetries (19). The postural asymmetries increase with age and with lower motor function (19). The temporal sequencing of scoliosis, pelvic obliquity, hip displacement, and the development of contractures is unclear.

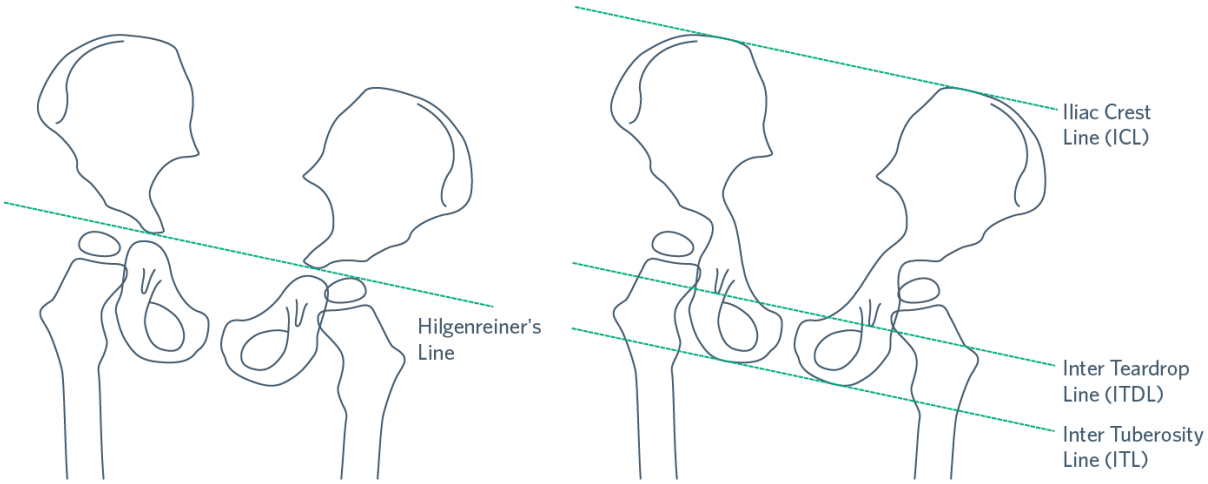


Figure 1.2. Pelvic Obliquity

Source: Australian Hip Surveillance Guidelines for Children with Cerebral Palsy: 2014, https://www.ausacpdm.org.au/resources/australian-hip-surveillance-guidelines/; used with permission of the Australian National Hip Surveillance Working Group.

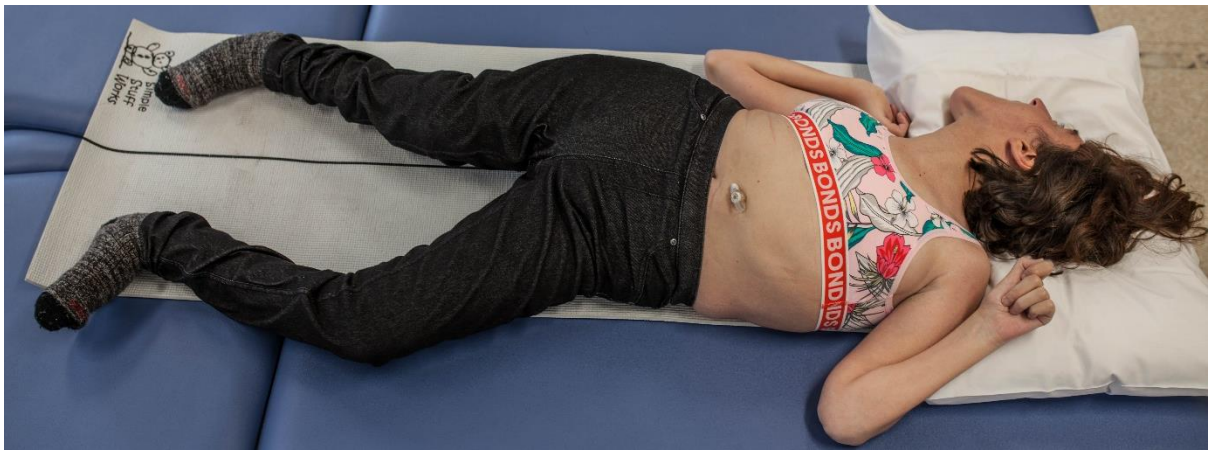

Figure 1.3 Windsweeping posture to the left

Photograph reproduced with permission

People with CP who are wheelchair dependent are particularly vulnerable to hip pathology. There is a strong linear relationship between GMFCS classification and hip dysfunction (20), with children classified at GMFCS IV and V having the highest risk of developing hip displacement. It has also been reported that progression and hip dislocation can occur after

skeletal maturity when other risk factors are present such as scoliosis and pelvic obliquity (20). There are established relationships between hip dysfunction, pelvic obliquity and scoliosis (7) and reports of pain associated with these musculoskeletal issues (21, 22). Ongoing hip surveillance is thus crucial to prevent adverse postural consequences. 'The Australian Hip Surveillance Guidelines for children with Cerebral Palsy: 2014' (23) recommends that those with motor function classified at GMFCS levels IV and V (i.e. non-ambulant) should "continue annual hip radiographs following skeletal maturity if hip displacement remains abnormal or in the presence of progressive scoliosis, significant pelvic obliquity, pain or deteriorating function". However the required position for the radiograph in hip surveillance may be difficult in adults with CP who have fixed postural asymmetries (24), or with significant behavioural problems limiting compliance in radiographic positioning. The requirement for ongoing hip surveillance can thus be difficult to achieve for adults with CP and postural asymmetry, impacting the ability for health professionals to monitor hip status, and potentially delaying appropriate interventions to minimise progressive hip dysfunction.

### 1.3 Consequences of postural asymmetry in cerebral palsy

Postural asymmetry may cause pressure on internal organs (Figure 4.4) resulting in secondary complications adversely affecting respiration, circulation and digestion. Respiratory dysfunction is a leading cause of premature death in those with more severe forms of CP, with a 14-fold increased risk of mortality attributed to diseases of the respiratory system and a 3-fold increase attributed to diseases of the circulatory system (10). Some of the common respiratory risk factors that may lead to premature death due to respiratory failure in those with CP include airway clearance dysfunction, saliva management difficulties, immobility, reflux, thoracic deformity and sleep-disordered breathing (25).

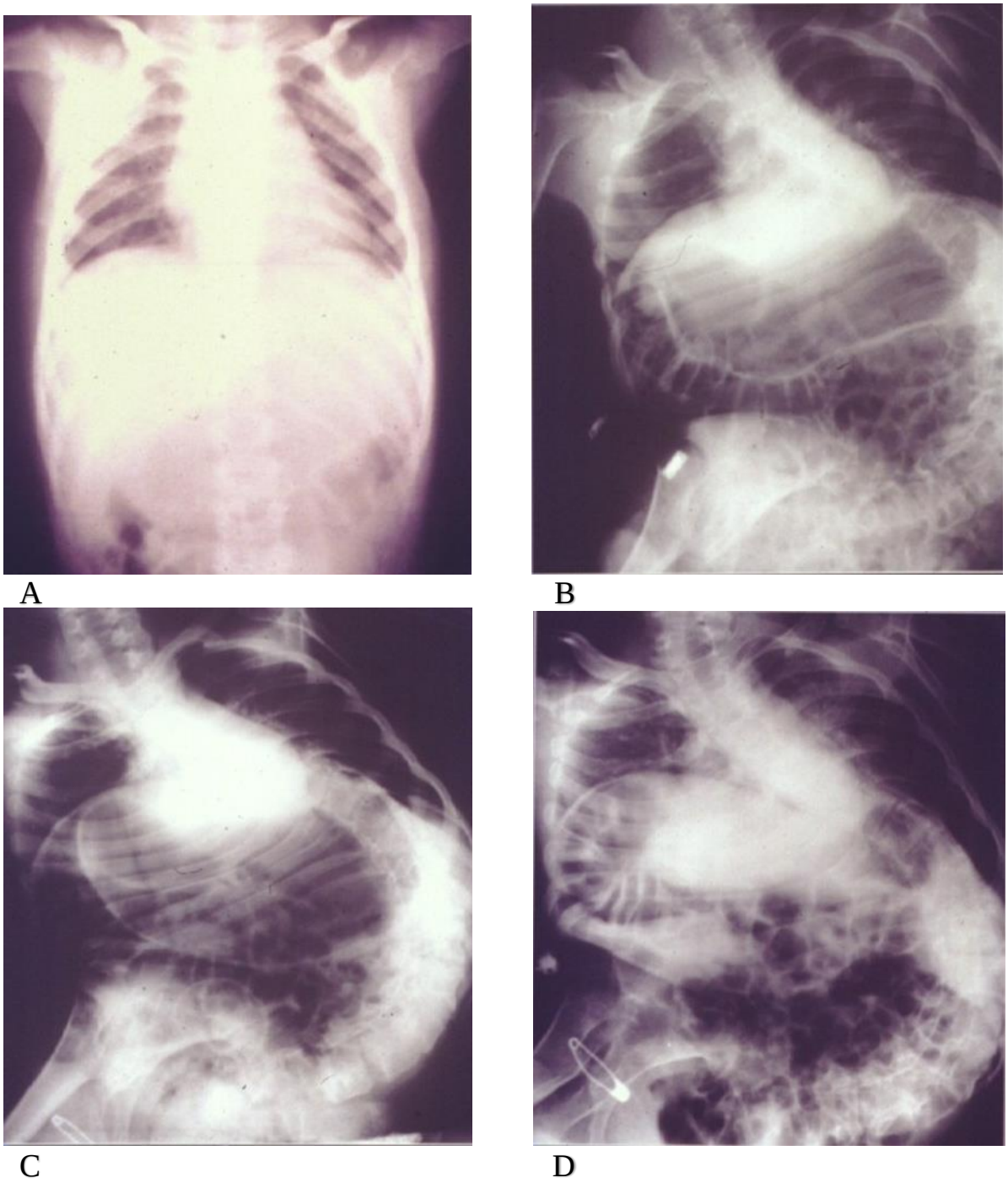


Figure 1.4 Radiographs depicting progression of postural asymmetry at age 3 (A), 17 (B), 21 (C) and 23 years old (D) in a young man with CP, GMFCS Level V

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Sixty-two to 87% of adults with CP (all levels of disability) report chronic pain with a strong association between severity of deformity and musculoskeletal pain (21). Experience of pain in adults with CP may be associated with contractures, orthopaedic deformity, spasticity, fractures, asymmetrical posture, poor nutrition and fatigue alongside gastrointestinal factors (21) leading to difficulty with seating, decreased participation and increased carer burden (17). Commonly reported sites of pain for adults with CP (all GMFCS levels) include back (59%), neck (44%), shoulder (43%), foot (44%), knee (39%), and hip (36%) (22). For adults with disorders of cognition and communication, reporting and localisation of pain is more challenging, with greater risk of pain neglect and under-management (26). Studies evaluating self-reported pain usually exclude the experience of people with cognitive and communication disorders who are unable to complete questionnaires. The incidence of pain in children with severe postural asymmetries is twice as likely as in those with less severe deformity (19) however the specific contribution of postural asymmetry to the pain experience of non-ambulant adults with CP is unknown.

### 1.4 Postural asymmetry and the International Classification of Functioning, Disability and Health framework

Postural asymmetry of the thoracic cage, pelvis and hips as explored in this thesis falls under the ICF domain of body structure and is closely interrelated with function. The International Classification of Functioning, Disability and Health (ICF) provides a framework for classifying function and disability over the lifespan (Figure 1.5). The ICF consists of two interacting domains: 1. Function and Disability involving body functions and structures, and activities and participation: and 2. Contextual factors involving environmental and personal factors. (27). Health outcomes are defined in terms of impairments in body structure, restriction in body function and limitations in activity and participation (28). To overcome limitations in activity and participation, adults with CP (GMFCS IV and V), require appropriate bed supports and seating with consideration of postural asymmetries (i.e. ICF Body functions).

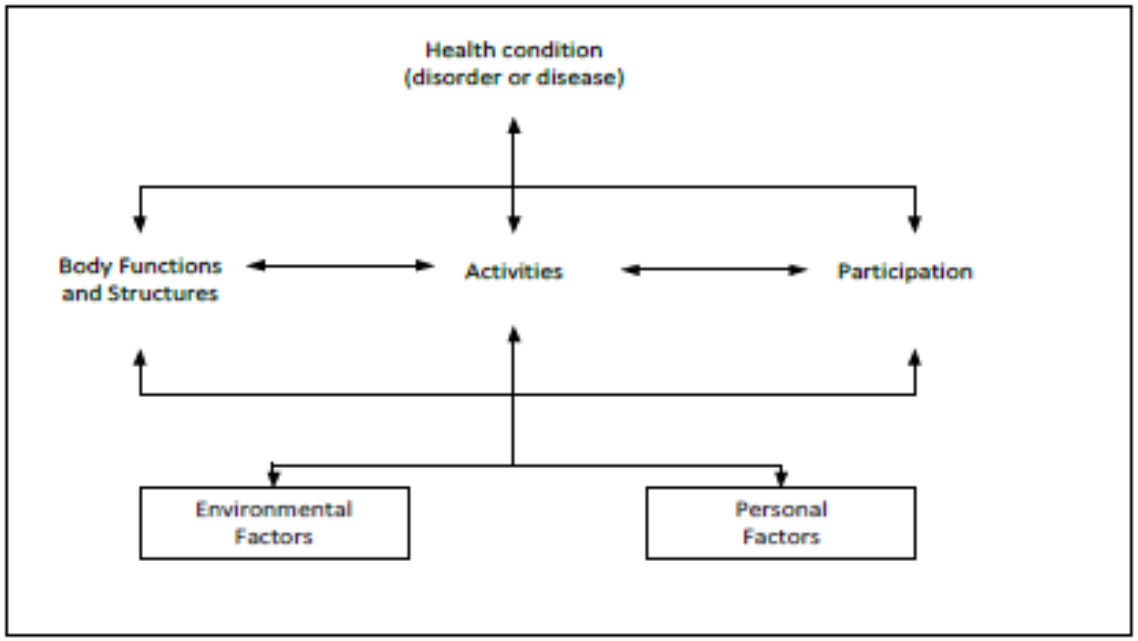



Figure 1.5 The International classification of Functioning, Disability and Health Model

Postural asymmetry impacts on a person, not only at the body function and structure level but also influences activities and participation with further interactions at an environmental and personal level as depicted in figure 1.5 above. This figure illustrates, as applied to a hypothetical non-ambulant adult ageing with CP and cognitive disability, the impact of the body structure on the domains of activity and participation, and the influence of environmental and personal factors (figure 1.6).

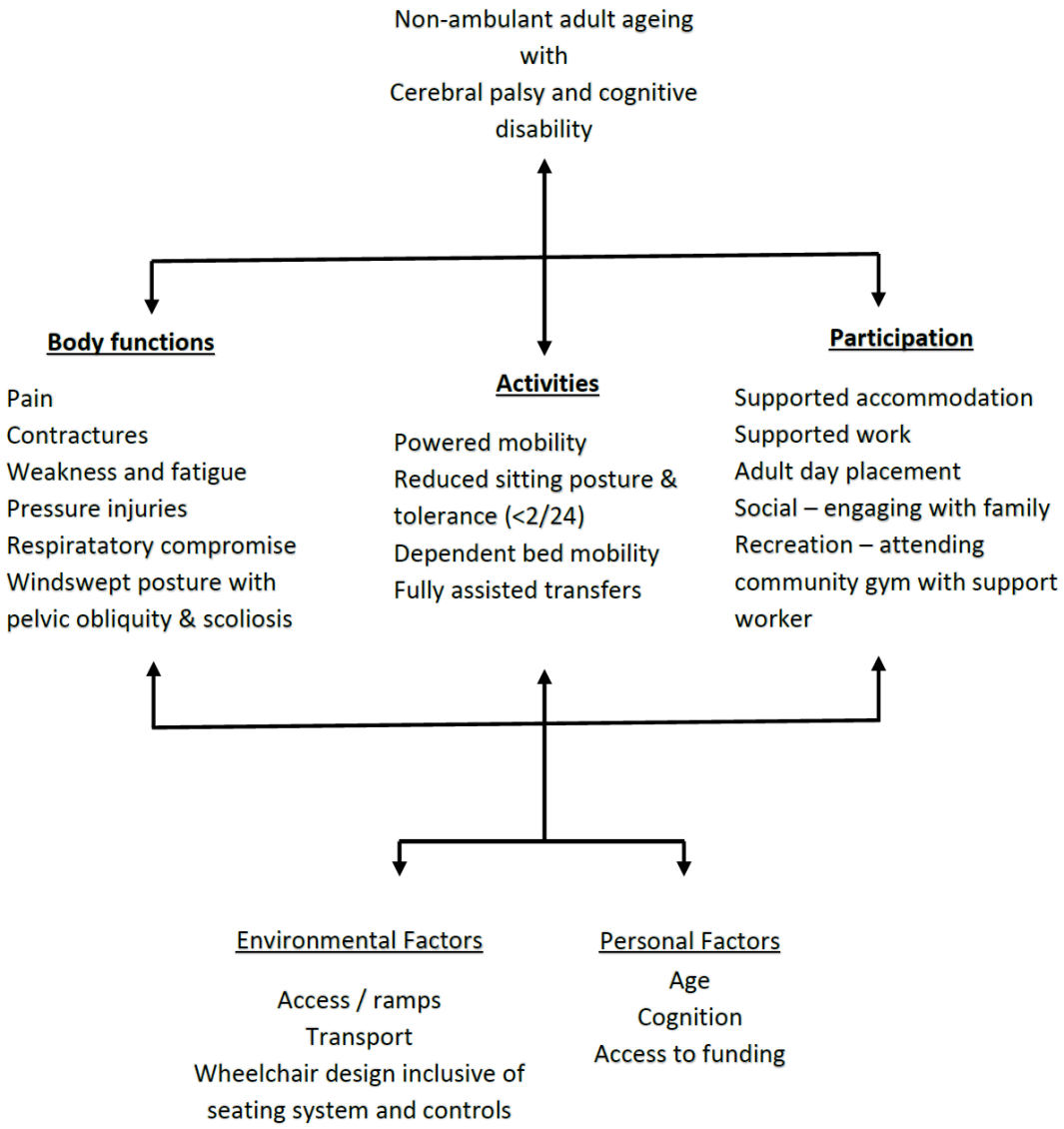



Figure 1.6 Adapted International Classification of Functioning illustrating the relationship between domains for a hypothetical non-ambulant adult with CP

## 1.5 Measurement of Postural Asymmetry

Postural asymmetry of the spine, pelvis and hips are most commonly measured radiographically for children and adolescents. Ongoing annual surveillance is encouraged post-skeletal maturity for those who are at higher risk of postural deterioration (20). Cobb angle, MP and pelvic obliquity are radiographic measurements used to assess status of scoliosis, hip displacement and lateral pelvic displacement respectively. The Cobb angle is an objective measure of the extent of spinal curvature (14), reported in degrees, with scoliosis defined as Cobb angle ≥ 10° (15). Migration percentage (MP) is a radiographic measure of

the amount of ossified femoral head not covered by the ossified acetabulum (16) Pelvic obliquity (Figure 1.3) may be measured as the angle between the inter tear drop line (ITDL) and a horizontal reference line parallel to the frame of the radiograph (17). The Melbourne Cerebral Palsy Hip Classification Scale (MCPHCS) is a six level radiographic ordinal scale used to classify morphology of the skeletally mature hip (29). The classification covers a wide range of radiographic features, from a Grade I (normal hip), through to a Grade V (dislocated hip) and Grade VI (dislocated hip that required salvage surgery). The classification includes sub-classifications for femoral head deformity, acetabular deformity and pelvic obliquity (29, 30)

Radiographs (plain X-rays) provide a two-dimensional image of what is a complex three-dimensional problem. Further, as previously indicated, there are issues of standardisation and compliance for those with contractures and behavioural issues. More complex methods which have capacity to provide greater detailed information regarding skeletal asymmetry and impact on soft tissues, such as CT scanning and MRI, also have limitations in ongoing screening for this population such as patient access and compliance. Further, the relevance of radiographical results to lying and seated activities and interventions is unclear. These radiographical measurements of posture focus on body structures at an impairment level as defined by the ICF.

Standard clinical assessment tools for bedside identification of joint range and movement include goniometers and tape measures. Goniometric tools may provide two-dimensional information about specific joint angles and spinal alignment in different planes (31) but are limited in ability to capture the multi- dimensional, multi-joint asymmetries observed in CP and are only able to capture movement in a single plane at a time whereas the GIofBS is able to measure movement across multiple planes simultaneously. Given the limitations of radiography and these clinical assessment tools alternative clinical measurement systems have been developed at the impairment level of the ICF model to measure complex three-dimensional postural asymmetry to enable evaluation of the effect of interventions. The Goldsmith Indices of Body Symmetry (GIofBS) is a simple clinical measurement tool which captures quantitative objective data of rib cage shape, pelvic alignment and hip orientation providing a three-dimensional understanding of the rotary nature of these asymmetries The GIofBS, using customised measurement apparatus, captures the segmental inter-relationships of the thoracic cage, pelvis and hips. The information obtained through the GIofBS can aid in problem solving complex seating and bed positioning requirements. Obtaining postural

symmetry is also essential for non-ambulant adults to maximise vision, communication and swallow in a seated position, as well as to enable community access and engagement. To date, the GIofBS has most commonly been used in the paediatric population in the United Kingdom.

The Posture and Posture Ability Scale (PPAS) is a 7 point ordinal scale describing postural ability in supine, prone, sitting and standing, with a focus on symmetry and weight bearing, alongside the ability to move in and out of postures (32). For posture, the scale scores deviations from midline of specific body segments as present or absent. This scale utilises an activity-based framework in contrast to the impairment-based focus of the GIofBS an interval scale,. The PPAS has been used as an outcome measure for postural asymmetries as a part of a long term follow up of joint motion and musculoskeletal status in the Swedish Cerebral Palsy Follow Up Program (CPUP) (33) The CPUP registry includes both clinical and radiological data providing access to a wealth of longitudinal data that can be used in the evidence for lifespan care of individuals with CP (34-37).

### 1.6 Interventions to improve postural asymmetry in adults with CP

There is a paucity of evidence regarding conservative interventions aimed at addressing the development and progression of postural asymmetry (13). While the effectiveness of interventions has been evaluated in children with CP (38) a similar comprehensive review for adults has not occurred. To enable evaluation of the effectiveness of postural interventions in adults with CP, appropriate measurement tools linked to the observed postural asymmetries and with clinical utility in relation to seating and bed positioning is required. Non-ambulant adults with CP rely on customised wheelchair seating and positioning to engage in daily activities, participate in the community, and 'live an ordinary life' (39). Postural assessment, monitoring and interventions to maximise comfort and function are an important aspect of lifespan care for this population

### 1.7 Thesis direction

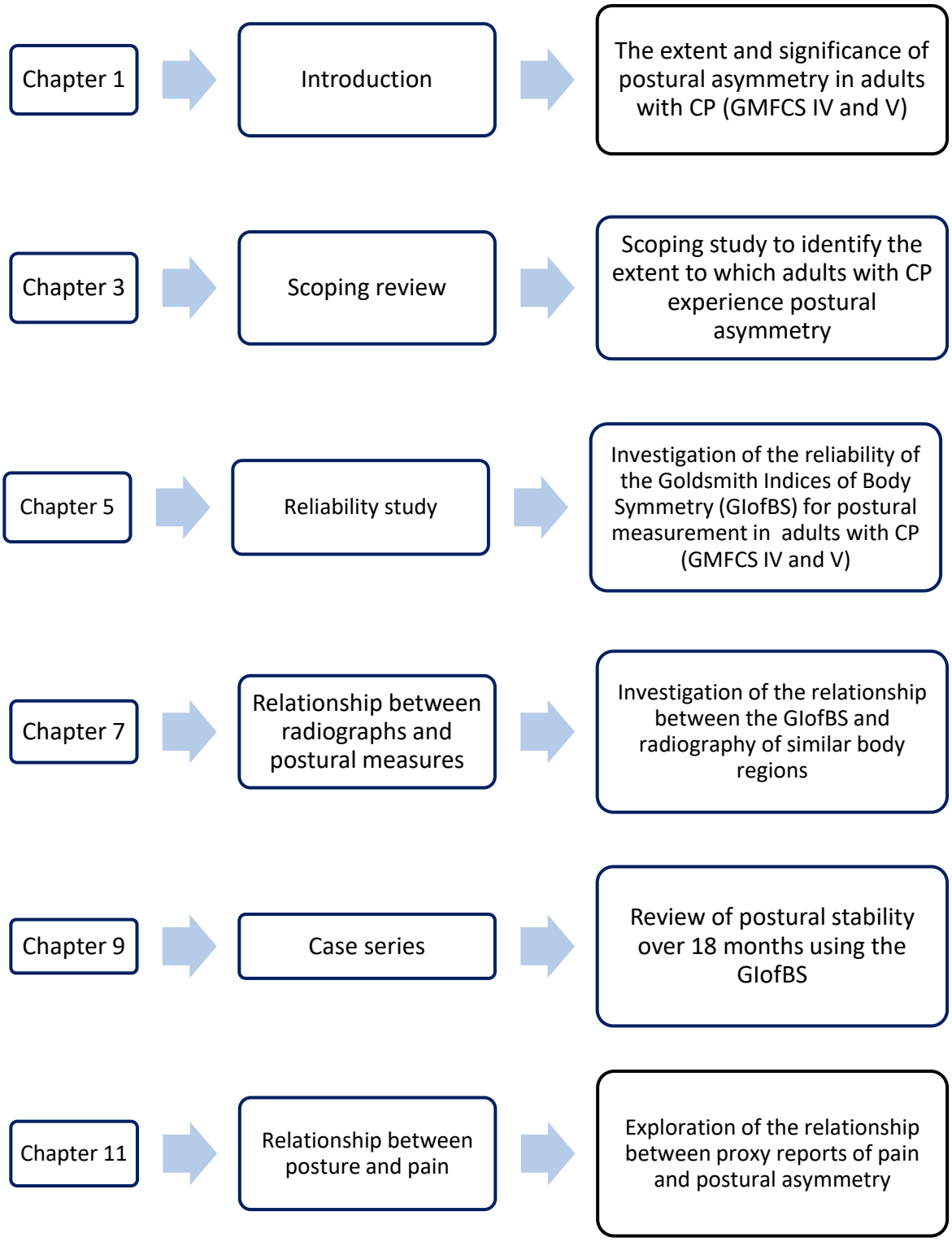
Non-ambulant adults with CP (GMFCS IV and V) are likely to have specific health care needs relating to neuromuscular dysfunction arising from lifelong disordered movement. We know that the majority of children diagnosed with CP will survive into adulthood but we do not thoroughly understand the impact of the ageing process on the musculoskeletal system or other functional domains. A lack of lifespan studies limit our knowledge of the known secondary postural complications involving the ribcage cage, spine, pelvis and hips in this population. This thesis describes a series of studies investigating postural asymmetry in adults with CP.

The following research questions regarding postural asymmetry in non-ambulant adults with CP are posed:

- What is the nature and prevalence of postural deformities in non-ambulant adults with CP?
    - What measurement tools for assessing and monitoring postural deformities are commonly used?
- Are the psychometric properties of the clinical measurement tool, the GIofBS sufficient to support clinical use?
    - Do GIofBS measurements differ between adults with CP and healthy controls?
    - Are GIofBS measures reliable?
- Is there a relationship between the GIofBS outcomes, (anthropometric measures of posture), and radiographs?
- Does postural asymmetry, as measured by the GIofBS, change over time?
- Is there a relationship between postural asymmetry and pain?

## 1.8 Thesis structure

A concept map outlining the thesis structure is described in Figure 1.7, and a brief summary of each thesis component is provided below.


Chapter 1
Introduction
The extent and significance of postural asymmetry in adults with CP (GMFCS IV and V)
Chapter 3
Scoping review
Scoping study to identify the extent to which adults with CP experience postural asymmetry
Chapter 5
Reliability study
Investigation of the reliability of the Goldsmith Indices of Body Symmetry (GIofBS) for postural measurement in adults with CP (GMFCS IV and V)
Chapter 7
Relationship between radiographs and postural measures
Investigation of the relationship between the GIofBS and radiography of similar body regions
Chapter 9
Case series
Review of postural stability over 18 months using the GIofBS
Chapter 11
Relationship between posture and pain
Exploration of the relationship between proxy reports of pain and postural asymmetry

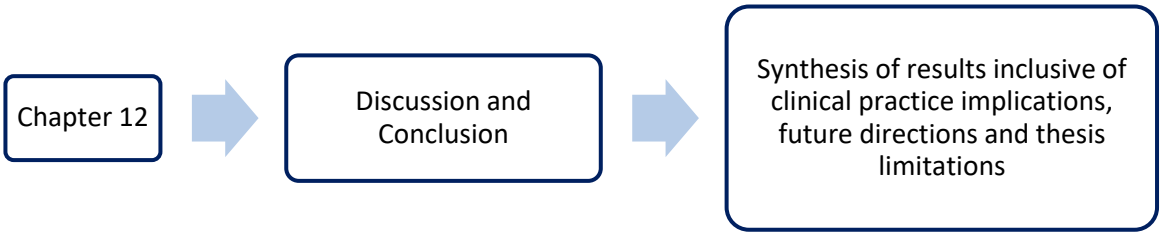



Figure 1.7 Thesis structure

*Note – brief linking chapters (linking one publication/chapter to the subsequent publication/chapter) and appendices have not been included in this figure*

### 1.8.1 Study 1

Study 1 (chapter 3) identifies and maps the available evidence, according to the Arksey and O'Malley framework (40), pertaining to the assessment of posture and conservative interventions for young adults with CP classified at GMFCS levels IV and V. A lack of evidence exists as to the breadth and extent of this issue despite improved mortality with many children now surviving well into adulthood (6) and the need for an understanding of lifespan issues and management. The scoping review identified prevalence, conservative management and measurement tools used for postural asymmetries in adults with CP (GMFCS IV and V). This scoping review was published in Disability and Rehabilitation in January 2018 (13). Carlee Holmes, Kim Brock & Prue Morgan (2018): Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review, Disability and Rehabilitation, DOI:10.1080/09638288.2017.1422037

To link to this article: https://doi.org/10.1080/09638288.2017.1422037

### 1.8.2 Study 2

Study 2 (chapter 5) investigates a clinical measurement tool known as the Goldsmith Indices of Body Symmetry for use in adults with CP (GMFCS IV and V). The intra and inter-rater reliability of the GIofBS to measure chest shape and symmetry, hip and pelvic mobility and windsweeping was investigated. Quantitative analysis methods were employed to investigate the reliability and efficacy of the GIofBS in this complex cohort of adults with CP. The establishment of comparative data from healthy controls (Appendix A) provided valuable

information to further understand the extent of asymmetry in adults with CP (GMFCS IV and V). This study was published in Disability and Rehabilitation in January 2020 (41). Carlee Holmes, Emma Fredrickson, Kim Brock & Prue Morgan (2020): The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy, Disability and Rehabilitation, DOI: 10.1080/09638288.2019.1708979

To link to this article: https://doi.org/10.1080/09638288.2019.1708979

### 1.8.3 Studies 3, 4 and 5

Having established the reliability of the GIofBS (chapter 5), Studies 3, 4 and 5 (chapters 7, 9 and 11) further investigated the relevance of the GIofBS in respect to relationships with common radiographic measures (chapter 7), ability to detect change in posture over time (chapter 9) and pain (chapter 11).

Further investigations were warranted to investigate the relationship between radiographs and GIofBS constructs of a similar body region due to limitations with radiography for those with established contractures and / or cognitive or behavioural challenges. This study was accepted for publication in The New Zealand Journal of Physiotherapy. Carlee Holmes, Kim Brock & Prue Morgan (2021): The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy, New Zealand Journal of Physiotherapy, In Press.

To link to this article: https://doi.org/10.15619/NZJP/49.1.03

Study 4 (Chapter 9) is a case series study investigating change in posture over an 18 month period (4 measurements) in a cohort of 10 young adults with CP (GMFCS IV and V). It has been established that this group of adults experience extreme postural asymmetry yet the rate of change is unknown. This study helps to identify the stability of posture and progression of any postural change. This study was accepted for publication in Pediatric Physical Therapy. Carlee Holmes, Kim Brock & Prue Morgan (2020): Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study, Pediatric Physical Therapy, In Press. DOI: 10.1097/PEP.0000000000000787

To link to this article: https://doi.org/10.1097/PEP.0000000000000787

Study 5 (Chapter 11) investigates the relationship of proxy reported pain to measures of postural asymmetry (radiographs and GIofBS) alongside conservative interventions that may impact on pain. Pain in adults with communication and cognitive disorders is challenging to

localise and accurately measure and may be impacted by the extreme postural asymmetries seen in adults with CP (GMFCS IV and V). Exploring this relationship may provide clinicians with insights into pain management strategies for this complex group of adults. This study was accepted for publication in Disability and Health (42). Carlee Holmes, Kim Brock & Prue Morgan (2021): Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study, Disability and Health Journal, In Press.

To link to this article: https://doi.org/10.1016/j.dhjo.2021.101063

## 1.9 Thesis Significance

Adults with CP functioning at GMFCS levels IV and V experience significant postural asymmetries of the spine pelvis and hips affecting many domains of health and functioning. There are also carer burden and economic impacts, including specialised equipment, support workers, hospital admissions and premature mortality associated with postural asymmetry. Consistent and reliable clinical measurement of posture and the impact of interventions has been lacking, with radiographic studies proving to be challenging for those with contractures and/or behavioural or movement disorders. Evidence for reliable and valid measurement tools would provide a means to monitor posture and assess the effectiveness of interventions. This thesis aims to establish the clinical utility of the GIofBS for use in adults with CP (GMFCS IV and V) and improve understanding of the impact of postural asymmetry and how it may be measured clinically, thus guiding the evaluation of interventions.

# Chapter 2: Background and Rationale

The following scoping review (study 1) was undertaken to explore the literature regarding postural deformities in adults with CP classified at GMFCS levels IV and V. The purpose of the scoping review was to identify gaps in the literature and provide a synthesis of available knowledge regarding the measurement of posture in the non-ambulant population of adults with CP (GMFCS IV and V). A paucity of high quality evidence on this topic guided the decision to conduct a scoping review rather than a systematic review. The review followed the guidelines of Arksey and O'Malley (40). This review was published in Disability and Rehabilitation in January 2018 (13). Carlee Holmes, Emma Fredrickson, Kim Brock & Prue Morgan (2020): The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy, Disability and Rehabilitation, DOI: 10.1080/09638288.2019.1708979

# Chapter 3: Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review

3.1 Abstract

**Purpose**: Non-ambulant adults with cerebral palsy are vulnerable to development of postural asymmetry and associated complications. The primary aim of this scoping review was to identify postural deformities in non-ambulant adults with cerebral palsy

**Materials and methods**: Comprehensive searches were undertaken in EMBASE, CINAHL, AMED, Cochrane, Psych INFO, and Joanna Briggs (1986-Jan 2017), supplemented by hand searching**.** Two reviewers independently extracted data using a customised tool focusing on study design, participant characteristics, postural descriptors, measurement tools, and interventions.

**Results**: From 2546 potential records, 17 studies were included.Variability in populations, reporting methodology and measurement systems was evident. Data suggests more than 30% of this population have hip migration percentage in excess of 30%, more than 75% experience 'scoliosis', and more than 40% demonstrate pelvic obliquity. Estimates ranged from 14-100% hip and 32-87% knee contracture incidence. Conservative interventions were infrequently and poorly described.

**Conclusion**: Many non-ambulant adults with cerebral palsy experience postural asymmetry associated with windswept hips, scoliosis, pelvic obliquity and limb contracture. Options for non-radiographic monitoring of postural asymmetry should be identified, and conservative interventions formally evaluated in this population.

**Key words**

Cerebral palsy, posture, deformity, scoliosis, windswept, migration percentage

## 3.2 Introduction

Cerebral Palsy (CP) describes a group of permanent disorders of the development of movement and posture that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain [1]. The lifelong motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour, and epilepsy further adding to the heterogeneity of this population [1]. Although the initial neurological lesion is static in nature, the extent of the motor disability ranges from minimal to profound, with a deterioration in function reported in many adults with CP along with secondary consequences such as musculoskeletal deformities, pain, fatigue and falls [2, 3].

Management of secondary complications, particularly postural asymmetry such as scoliosis and pelvic obliquity, is an important aspect of healthcare for adults with CP. The development of postural asymmetry of the thoracic cage can affect respiration and lung function resulting in pneumonia and respiratory diseases for the most severely affected adults [4]. Reports of pain, difficulty seating, and decreased participation have been described as a consequence of an asymmetrical posture in this population [5]. Further complications such as hygiene issues, compromised swallow, sleep disturbance and pressure injuries can also arise [6]. Postural asymmetries may be most evident in non-ambulant adults with CP with low motor function typically described as Gross Motor Function Classification System (GMFCS) Levels IV and V [7]. This predominantly wheelchair user subgroup makes up around twenty percent of the total population of people with CP [8], exhibiting the highest risk for secondary complications and potential for a shorter life expectancy [9]. With improved access to medical care, the CP population with severe and multiple disabilities, do survive into adulthood [9], and thus assessment and management of the secondary complications is essential to ensure quality of life is maximised.

Diagnosing postural asymmetry such as scoliosis and pelvic obliquity may be particularly challenging in adults with CP who have profound disability, which frequently includes limitations in communication and cognition. As such, the identification, monitoring and management of secondary complications for adults with CP becomes critical given the potential for adverse consequences to rapidly escalate. Although hip surveillance guidelines are generally followed for at-risk children with cerebral palsy, effective methods such as radiography to monitor posture over time is problematic for many adults with severe disability who cannot cognitively tolerate radiographical procedures or cannot achieve a standardised supine position for radiographs due to postural asymmetry and contractures. Further, the

complex three-dimensional postural asymmetry frequently encountered in adults with severe CP is difficult to quantify by standard methods such as goniometry and calculation of Cobb angles without using resource-intensive computerised tomography. Consequently there is limited evidence available regarding the nature of the postural asymmetry, progression of postural asymmetry over time, and effective interventions to prevent, minimise and/or remediate postural asymmetry in the adult population with CP.

Both the extent of scoliosis and pelvic obliquity may progress following skeletal maturity in adults with CP, but the rate and timing of such progression is not well recognised [10, 11]. Although there is considerable evidence surrounding the proportion of children with displaced hips and scoliosis [12, 13] information tracking the progress of these children and their posture into adulthood is scarce. Published evidence tends to focus on the outcomes of hip or spinal surgical techniques in various cohorts, and/or excludes those with most severe disability due to them being a higher surgical risk due to respiratory compromise [4]. Conservative interventions to address postural asymmetry such as prescription of customised sleep, seating and pressure management systems may also be poorly described, and lack rigorous methods of evaluation. Clinicians thus lack guidelines to assist them in managing postural asymmetry in this complex population.

Nevertheless, the population of adults with severe cerebral palsy (GMFCS Level IV and V) need careful and regular monitoring, and deserve timely and affordable interventions to manage postural asymmetry to enhance the quality of their life. Given the lack of high quality evidence available, a scoping review was thus initiated following the guidelines of Arksey and O'Malley [14]. The key foundations of a scoping review are to examine broad areas, to identify gaps in the evidence, clarify key concepts, and report on the types of evidence that address and inform practice in a topic area [14]. The primary aim of this scoping review was thus to investigate the nature, prevalence and conservative management of postural deformities in non-ambulant adults with CP (GMFCS levels IV and V). A secondary aim of this review was to synthesise measurement tools used to document postural deformities in non-ambulant adults with CP, and describe conservative interventions selected for the management of postural deformities in this population.

## 3.3 Method

An initial search strategy was created in Ovid Medline and search terms customised as necessary for the following databases: EMBASE, CINAHL, AMED, Cochrane, Psych INFO, and Joanna Briggs, until January 2017. In addition, targeted hand searching of reference lists supplemented the strategy. The search terms included the following keywords, with Boolean operators applied to combine domains: cerebral palsy, scoliosis, Cobb angle, hip dislocation, hip subluxation, hip dysplasia, migration percentage, pelvic obliquity, windblown, windsweep, asymmetry, posture, deformity. Adolescent and adult age group filters were applied. The full search strategy is included in Supplementary Material 1.

The titles and abstracts identified by the initial search strategy were screened to identify potentially eligible reports and retrieve full-text reports. When the title or abstract did not clearly indicate whether an article should be included, the complete article was retrieved and reviewed and consensus achieved by two authors (PM and CH). Full-text reports were independently evaluated by two authors (CH and PM) for the following inclusion criteria: (1) all participants with a diagnosis of cerebral palsy, or if mixed cohorts included, data pertaining to those with CP able to be extracted; (2) all participants GMFCS Level IV and/or V (or described as non-ambulant), or if mixed cohorts included, data pertaining to GMFCS Level IV and/or V able to be extracted; (3) adult age group, or ≥80% of participants over the age of 16, or if wider age groups included, data pertaining to adult ages able to be extracted; (4) data included measurement of any aspect of posture; (5) intervention data collected described as conservative, inclusive of spasticity management; (6) English language. Any trial design was accepted, including case series. Given the advances in medical and surgical management of CP over the last 30 years [15] a decision was made to restrict studies to those published from 1986 onwards.

*Charting the data and data extraction*

A standardised data extraction form was constructed to identify and detail key features of each study using recommended information from the Joanna Briggs Institute (JBI) [14]. Two reviewers (CH and PM) initially independently pilot tested the form with a small subset of representative studies to confirm the content and ensure consistency in its application. The extracted study details focused on study design, sample participant characteristics, descriptors of posture, measurement tools utilised, and any interventions applied. As this was a scoping

study, an appraisal of study quality was not included. Results are presented in descriptive format.

### 3.4 Results

Database searching identified 3491 potential records, and an additional 39 records were identified through other sources (hand searching reference lists). After duplicates were removed, 2546 potential records remained. Following exclusion of records based on title and abstract, 93 records remain to be screened based on full text. Seventeen studies were included in the final synthesis (figure 3.1).

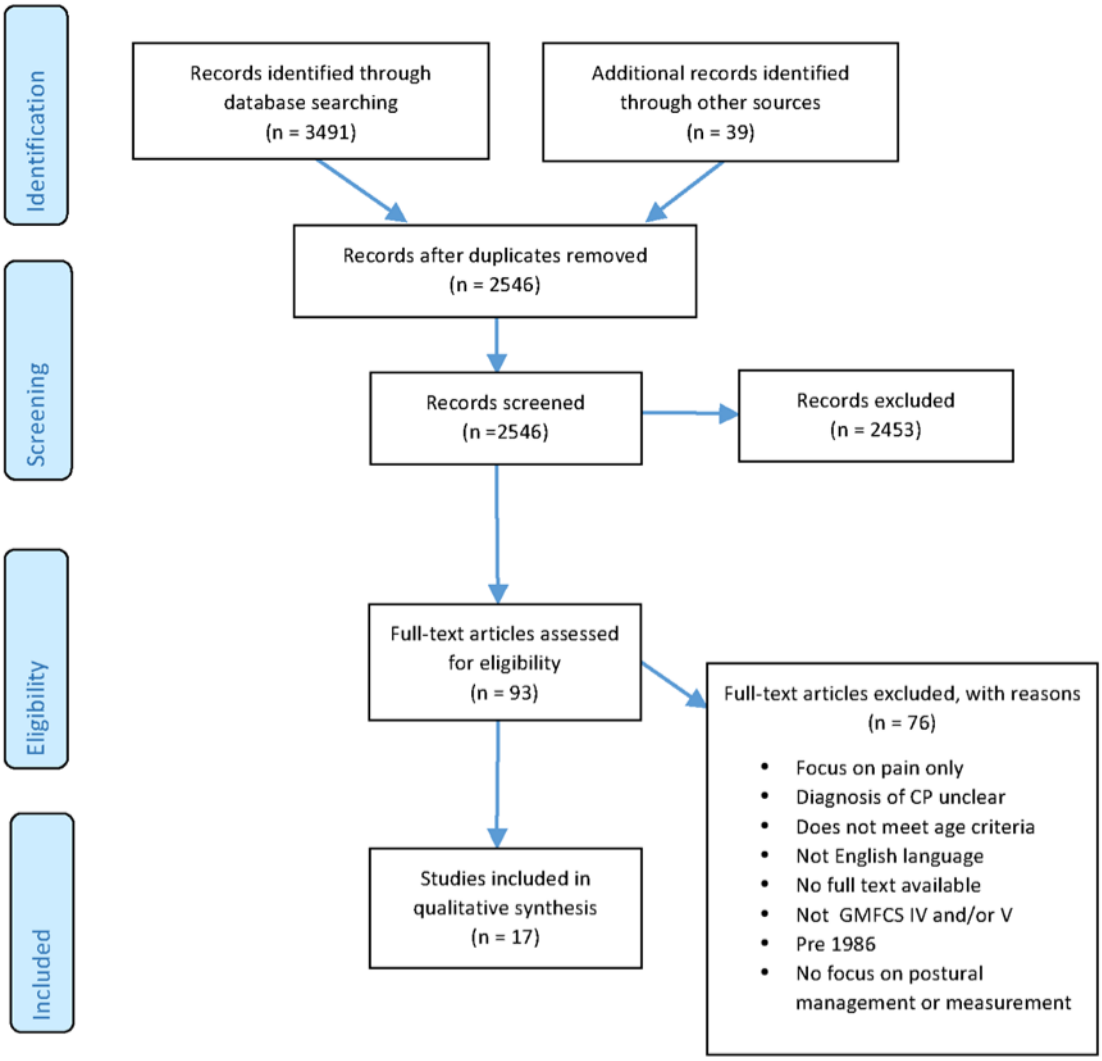



Figure 3.1. PRISMA flow diagram

Flow chart mapping the results of records identified throughout the database searching process and reason for exclusion

3.5 Study samples

The majority of studies arose from USA (n=7), with four from Japanese groups, and three studies arising from Sweden (table 3.1). Five studies were identified as being led by a physiotherapist [4, 16, 17, 18]. Six studies used targeted recruitment methods aimed at selecting participants with identified postural abnormalities [10, 12, 16, 19, 20, 21].

Study participants ranged from five [16] to 234 [22]. Most studies were retrospective or observational cohort studies involving medical record (chart) reviews (n=13), and/or radiographs (n=12), with 11 studies including additional physical examination components. Two studies recruited participants from a CP registry [17, 18] whilst four studies recruited participants exclusively from residential institutions [19, 20, 23, 24]. The remaining studies recruited from the community and tertiary hospitals. Only three studies included longitudinal data [11, 12, 21].

Not surprisingly, more recent studies used the GMFCS to describe their populations as Levels IV and/or V [2, 4, 10, 11, 16- 18, 23- 25] with generally older studies using terms such as 'bed ridden', 'propped sitters', or 'non ambulatory' to describe the motor function of participants [12, 19- 21, 22, 26, 27]. In these instances, the review authors converted descriptions to equivalent GMFCS Levels. Four studies [4, 16, 23, 24] exclusively recruited those at GMFCS Level V, with the remaining studies including both GMFCS Level IV and V participants. Where reported, spastic quadriplegia was the dominant motor type and distribution. As expected, many participants had additional impairments such as epilepsy, intellectual impairment, and were non-verbal. All but one study [24] recruited exclusively those with a diagnosis of CP. Sato's study [24] included 2 of 22 participants with other neurological disorders. Where reported (n=14), the mean age was less than 40 years old in 11 studies. A further three of these 14 studies included participants with a mean age between 40 and 45 years [4, 24, 27]. Eleven studies included broader inclusion criterion of which relevant data pertaining to postural deformities, adult age groups and/or GMFCS Levels IV and V was extracted. Table 3.1 synthesises the study type and demographic information regarding included study participants.

Table 3.1 Study details and demographic information of included participants

| First author, year | Country | Study type | Study setting | Specific inclusion or exclusion criterion related to postural abnormalities | n (entire cohort) | Age: mean (sd), range | Gender n (%) | GMFCS Level | CP subtype/ motor distribution | Reported comorbidities |
|---|---|---|---|---|---|---|---|---|---|---|
| Boldingh 2007 | Netherlands | Cross sectional | 58% residential nursing facility | Femoral head able to be identified on X-ray | 140 (160) | *36 (16), 16-84 | *M: 54% F: 46% | Level V | Spastic, quadriplegia | Minimum mental age 4 years |
| Heidt 2015 | Australia | Prospective cohort | Transition clinic | MP>30% , history of adductor release | 67(98) | *19, 15-24 | *M:65 F:33 | 32 Level IV; 35 Level V | N/R | N/R |
| Hodgkinson 2001 | France | Cross sectional, multi-centre | 32 centres – including some residential | Nil reported | 234 | 28, 18-38 (95% CI) | M:59 %; F:41% | Level IV and V[1] | 100% spastic quadriplegia; 26.9% athetosis | 51.1% multiple disabilities; 49.1% epilepsy; 47.2% hip pain |
| Horimoto 2012 | Japan | Case series | Acute hospital | Previous CT scan for diagnosis of pneumonia | 20 | 43 (18), 20-70 | M 16; F 4 | Level V | N/R | N/R |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| Kalen 1992 | USA | Cohort study | Single residential institution | Nil reported | 56 (62) | *39, 29-67 | *M 26: F 36 | Level IV and V[1] | *54 spastic quadriplegia; 6 spastic diplegia; 1 hemiplegia; 1 not categorised | Subset of 14 with scoliosis >45°: 100% mental retardation; 22% pressure ulcers |
| Knapp, 2002 | USA | Retrospective cohort study | 4 residential institutions | established hip dislocations or subluxations | 29 (117) | 34, 21-52 | M: 17; F: 12 | Level IV and V[1] | Spastic quadriplegia | N/R |
| Littleton, 2011 | USA | Single subject research design | Institutional and community | severe scoliosis (fixed deformity), history of respiratory illness | 5 | 17-37 (37, 22, 25, 26 & 17) | M: 3; F: 2 | Level V | Spastic quadriplegia | 4 gastrostomy tubes<br>4 hip surgery<br>3 seizure disorders<br>2 sleep apnoea |
| Murphy 1995 | USA | Observational cohort study | Community | Nil reported | 67 (101) | *43 (19-74) | *M: 53; F: 48 | Level IV and V[1] | 2 hemiplegia; 5 diplegia; 26 quadriplegia; 34 dyskinesia (non ambulatory groups) | N= 44 prior orthopaedic procedures; 50% incontinent (urinary); 43% back pain; pain in lower limb joints 44% ; leg |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | | | contractures 91% |
| Oda 2017 | Japan | Observational cohort study | Single tertiary hospital | excluded prior scoliosis surgery | 47 (92) | 20-69 | N/R | Level IV and V | N/R | N/R |
| Pritchett 1990 | USA | Observational cohort study | Group homes | 50 treated and 50 untreated with unstable hips | 100 | Untreated:26 (21-50); treated: 25 (20-42) | Untreated: M:23 F:27 Treated: M:24 F:26 | Level IV and V[1] | spastic | profoundly mentally retarded and incontinent; 75% seizure disorder; 85% non verbal |
| Rodby-Bousquet 2012 | Sweden | Reliability and tool validation study | CPUP program (registry) | Nil reported | 12 (30) | *19-22 | *M:15 F: 15 | 6 Level IV 6 Level V | N/R | N/R |
| Rodby-Bousquet 2013 | Sweden | Cross sectional study | CPUP program (registry) | Nil reported | 30 (102) | *Md 21 (19-22) | *M: 62 F:39 | 10 Level IV; 20 Level V | *unilateral spastic (n=26), bilateral spastic (n=45), ataxic (n=12), dyskinetic (n=19). | N/R |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| Saito 1998 | Japan | Retrospective cohort study | Single residential institution | X-ray before 15yo, scoliosis at least 10° | 31 (37) | 25, 15-36 | *M: 18; F: 19 | Level IV and V[1] | spastic | *High levels of intellectual impairment & seizures<br>31 had contractures;<br>35 non verbal |
| Sandström 2004 | Sweden | observational cohort study | Institution and community | Nil reported | 18 (48) | *33 (8.2) | *M: 48% F: 52% | 9 Level IV 9 Level V | *13 hemiplegia; 19 diplegia; 9 quadriplegia; 7 dyskinesia | N/R |
| Sato 2016 | Japan | reliability and tool validation study | Single residential institution | Excluded prior spinal surgery | 20 (22) | 35-52 | M 11<br>F 11 | Level V | N/R | N/R |
| Senaran 2007 | USA | retrospective cohort study with matched control group | Single tertiary hospital | Prior ITB implantation and age/gender/ GMFCS control with scoliosis | ITB group 11, Control 14 | 18; 16-21 | ITB group: 6M:5F Control: 10M: 4F | ITB group: all Level V, Control: 2 Level IV & 12 Level V | spastic | N/R |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| Thometz 1998 | USA | Retrospective cohort study | Previous students of special school | Scoliosis - X-rays available after skeletal maturity and at least 4 year follow up X-Ray | 40 (51) | 20-63 | N/R | Level IV and V[1] | 23 spastic quadriplegia, 7 spastic diplegia, 5 spastic hemiplegia, 16 miscellaneous = ataxia, athetosis, hypotonia | Intellectual impairment |

*entire cohort, not age/GMFCS level subset; [1] author interpretation; MP: migration percentage; N/R not reported; ITB: intrathecal baclofen; CPUP: Sweden's follow up surveillance program for CP

### 3.5.1 Impairments

The postural impairments of hip deformity, scoliosis, pelvic obliquity and lower limb contractures often co-occur. These four musculoskeletal deformities (impairments) were reported by 16/17 included studies, with 11 describing information regarding more than one postural deformity. Descriptors of scoliosis or thoracic deformity (n=12) were most frequent (table 3.2). Information regarding migration percentage / hip dislocation / hip subluxation was reported in seven studies, and pelvic obliquity in six studies (table 3.2). Of note, where the study included broader age groups or level of disability, only postural deformities that are specifically attributable to the population of interest are included in tables 3.2 and 3.3.

Table 3.2 Total number of postural deformities (impairments) described in scoping review studies

| Postural Deformity | Incidence of Description (n=16 studies*) |
|---|---|
| MP/hip dislocation, subluxation, femoral head deformity | 7 [10][17][19][20[22][23][26] |
| Scoliosis and thoracic deformity | 12 [4][11][12][16][17][19][20][21][24][25][26][27] |
| Pelvic obliquity | 6 [10][16][19][20][25][26] |
| Hip and/or knee contracture | 6 [2][17][19][23][26][27] |

MP: migration percentage; *studies may have described more than one postural deformity

Migration percentage is the percentage of the bony width of the femoral capital epiphysis which falls lateral to a line drawn vertically from the bony lateral margin of the acetabulum, on an AP pelvis radiograph. In the included studies, migration percentage (MP) was reported as the proportion exhibiting either hip enlocation or not (0 or 100%) [18, 20, 26], or proportions of participants exhibiting a specified range of migration percentage [10, 22, 23] predominantly using Reimer's index from radiographs to calculate. Only Kalen's study [26] did not specify the use of Reimer's index for MP estimation. Heidt and colleagues reported median values of MP for high side and low side hips [10]. Those who did not specifically recruit participants with hip dysfunction, revealed MP proportions greater than 30% (suggesting subluxation) of 24% [19], 25% [22]and 32% [23] of their cohort.

Twelve studies described evidence of scoliosis and/or associated thoracic deformity in participants. Eight of these studies described the severity of the scoliosis by calculation of Cobb angle from standard radiographs [11, 12, 19-21, 24- 26]. Overall synthesis of data pertaining

to scoliosis was not possible due to variation in reporting methods. For example, Oda and colleagues [11] described mean scoliosis relative to age groups, Saito [21] and Thometz [12] described mean scoliosis relative to functional ability, and others described proportions of participants exhibiting a specific Cobb angle range [19, 20, 26]. Studies led by Rodby-Bousquet [18] and Murphy [27] reported proportion of participants demonstrating any scoliosis (77% and 90% respectively), and Senaran's study [25] reported the mean Cobb angle of group participants (57° ITB group, 66° control group). Other authors provided additional descriptors of the associated thoracic diameter using calculations from CT scans or digital photography [4, 24], curvature [24], and impact of the scoliotic deformity [16]. Where radiographic outcomes were described, Senaran et al. [25] measured scoliosis in supported sitting while studies led by Oda and that by Thometz used supine as the measurement position [11, 12]. A "standard upright" position was used in Kalen's study [26] and the measurement position was not stated by four authors [4, 19- 21, 24]

Pelvic obliquity was similarly reported using a range of methods, most commonly either proportion of participants exhibiting pelvic obliquity (%) [16, 19, 20, 26] or the mean pelvic obliquity in the cohort (degrees) [10, 25]. Where reported (n = 6), the presence of any pelvic obliquity in the cohort ranged from 41% [19] to 63% [20] and the amount of pelvic obliquity ranged from 1-14° (GMFCS Level IV), or 1-22° (GMFCS Level V) [10], and 0-20° (ITB group) or 0-45° (control group) [25].

Table 3.3. Conservative interventions for the management of postural deformity

| First author, year | Interventions | Evidence of use |
|---|---|---|
| **Wheelchairs and seating** | | |
| Boldingh, 2005 | Custom moulded seat | 61% total cohort (n=140);<br>87% with serious femoral head deformity<br>51% with no femoral head deformity |
| Kalen, 1992 | Custom moulded seat | 19% in those with scoliosis<45° (n=42)<br>54% in those with scoliosis>45° (n=14) |
| Knapp, 2002 | Modified wheelchair | 100% usage |
| Murphy, 1995 | Solid seat or postural supports | 3/67 usage |
| Littleton, 2011 | Customised wheelchair | 5/5 usage |
| **Bed Positioning** | | |
| Littleton, 2011 | pillows or customised side lyer | 5/5 usage |
| **Spasticity Management** | | |
| Heidt, 2015 | Oral medications including baclofen and diazepam<br>Botulinum Toxin to hip adductors<br>ITB pumps<br>Phenolisation of obturator nerve | 'Majority' of cohort (prior intervention)<br>42/98* (prior intervention)<br>4/98*<br>Unspecified (prior intervention) |
| Senaran, 2007 | ITB pump | 100% of selected participants (n=50) in ITB group |
| **Contracture management** | | |
| Heidt, 2015 | Botulinum Toxin for hip flexion contractures with iliopsoas lengthening | Not specified (prior intervention) |

*total cohort including 29 participants GMFCS Level II and III; ITB: intrathecal baclofen

Information regarding hip or knee contractures was provided by six studies [2, 18, 19, 23, 26, 27] and generally described the proportion of participants exhibiting a contracture. Where reported, goniometry or visual estimates of joint angles was implemented. Where specified, estimates ranged from 14% hip and 32% incidence of knee contractures [23] to 100% hip [2] and 87% knee contractures [18] in the cohorts studied.

### 3.5.2 Activity limitations and Postural Asymmetries

Rodby-Bousquet [17, 18] was the only lead author who utilised a clinical assessment scale (the Posture and Postural Ability Scale) to describe the static and dynamic posture of adults with CP. This scale classifies posture in supine, prone, sitting and standing, with attention to the relationship between body parts (head, trunk, arms, legs), weight bearing, and symmetry. It also describes the participant's ability to move in and out of a static posture, thus utilising an activity limitation framework. Postural ability is described using a 7 point scale, from Level 1: unplaceable in an aligned posture, through to Level 7: able to move into and out of a position. The low median scores for postural ability for the six participants described as GMFCS Level IV (4, range 3-7) and six described as Level V (1.5, range 1-4) indicate that at best the person was able to initiate flexion/extension of trunk (level 4) and, if rated at level 1, was unplaceable in an aligned posture in supine [17]. In sitting, those at GMFCS Level IV and V were both rated as 2 (placeable in an aligned posture but needs support). The quality of the posture in both the frontal and sagittal planes was very low for participants with most scores of 0 indicating they were unable to achieve any of the six requirements of alignment of body parts in either supine or sitting [17]. In Rodby-Bousquet's 2013 study, 17/30 (participants GMFCS Level IV and V) were unable to achieve a symmetric trunk in supine, and 18/30 were unable to achieve a symmetric trunk in sitting as measured by the Posture and Postural Ability Scale [18].

### 3.5.3 Conservative interventions

As most studies were observational, descriptions of non-surgical interventions (conservative) and subsequent evaluation of any conservative interventions were scarce (table 3.4). Where conservative interventions were described, they may not have been specific to GMFCS Level IV and V, hence limited data was able to be extracted. Eight studies reported the use of customised wheelchairs (and/or customised seating inserts) in describing their cohorts, with some reporting percentage usage [23, 26]. Boldingh's study [23] related the need for special moulded seating to the incidence of femoral head deformity, with 87% of participants with serious femoral head deformity and 51% of participants with no femoral head deformity requiring special moulded seating [23]. Kalen's study related the need for customised moulded seating to the severity of scoliosis, with 54% of participants with Cobb angles greater than 45° requiring customised moulded seating [26]. Littleton [16] provided detail about the customisation of wheelchairs used, with description of head support, tilt in space feature,

custom cushion and lateral support, pelvic positioning device and foot support. Several studies noted the inadequacy of the current seating systems for study participants as a striking outcome from their findings in the discussion [20, 22, 27], or made reference to it as a potential management strategy [20, 22]. Only one study noted the use of customised side-lyers to maintain a side lie position in relation to respiratory function [16].

Two studies [10, 25] described prior or current spasticity management interventions in their cohorts as methods to reduce or slow progression of the postural deformities of the trunk, pelvis and hips. In Heidt's study [10] participants had previously undergone a variety of spasticity management techniques including oral medications, nerve phenolisation, Botulinum Toxin and intrathecal baclofen (ITB) pumps with the aim of reducing known causes of pelvic obliquity leading to windswept hips and late hip dislocation. Senaran and colleagues [25] in discussion proposed that the reduction in asymmetrical spasticity from the ITB pump, allowed greater balance in paraspinal and postural muscles facilitating better upright sitting posture, however concluded it had no significant effect on scoliosis progression in people with quadriplegic CP. Efficacy of these strategies ongoing throughout adulthood are not addressed. No studies specifically described strategies to manage contracture in the adult cohorts other than Heidt et al. who mentioned prior botulinum toxin and iliopsoas lengthening strategies to manage hip contracture in an unknown proportion of the cohort [10].

Table 3.4. Summary of postural asymmetries recorded

| First author, year | **Postural deformity** | **Method of assessment** |
|---|---|---|
| **Hip deformity** | | |
| Boldingh 2007 | MP: % of cohort<br><30: 56%<br>30-59: 22%<br>60-100: 10%<br>Femoral head deformity: % of cohort<br>None: 29%<br>Moderate: 41%<br>Severe: 29% | Standard AP radiograph of hips and pelvis (Reimer's)<br><br><br>From X-ray using 3 point scale – none, moderate, severe |
| Heidt 2015 | MP Md (IQR):<br>*High side hip* | Standard AP radiograph of hips and pelvis (Reimer's) |

| | | |
|---|---|---|
| | 31 (22-41) Level IV; 40 (29-88) Level V<br>*Low side hip*<br>15 (3-25) Level IV; 9 (1-15) Level V | |
| Hodgkinson 2001 | MP: % of cohort<br><33: 74.7%<br>33<100: 13.2%<br>100: 12.1% | Standard AP radiograph of hips and pelvis (Reimer's) |
| Kalen 1992 | MP: % of cohort<br>100: 64% (9/14)[1] | Not reported |
| Knapp 2002 | MP[2]: % of cohort<br>30<100: 24% (9/38)[3]<br>100: 76% (29/38)[3] | Standard AP radiograph of hips and pelvis (Reimer's) |
| Pritchett 1990 | MP: % of cohort<br>100: 100%[2]<br>50 untreated - 34 bilateral; 16 unilateral hips affected<br>50 treated – 33 bilateral; 17 unilateral hips affected | Standard AP radiograph of hips and pelvis (Reimer's) |
| Rodby Bousquet 2013 | MP: % of cohort<br>100: 30% | Standard AP radiograph of hips and pelvis (Reimer's) |
| **Scoliosis** | | |
| Horimoto 2012 | Protocol 1 (mm): AP diameter mean 153 (33); LL diameter 261 (38); AP/LL 0.6 (0.1)<br>Protocol 2 (mm): AP diameter mean 198 (43); LL diameter 242 (36); AP/LL 0.8 (0.2) | Calculation of anteroposterior and laterolateral diameter of thorax from CT scan using 2 different protocols |
| Kalen 1992 | Scoliosis: >45° 33% (14/42) | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Knapp 2002 | Scoliosis:<br><45° 17% (5/29)<br>45-135° 55% (16/29) | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Littleton 2011 | Scoliosis: 'severe with fixed deformity'[2] 100% (5/5)<br>Rib impingement against iliac crest (3/5); R thoracic convexity (3/5); L thoracic convexity (2/5); R rib hump (2/5); L rib hump (2/5) | Descriptive only |

| | | |
|---|---|---|
| Murphy 1995 | Scoliosis: 90% | N/R |
| Oda 2017 | Scoliosis (mean Cobb angle)*<br>Age 20 to 29: IV 34°, V 74°,<br>Age 30 to 39: IV 42°, V 38 °<br>Age 40+: IV 56°, V 64° | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Pritchett 1990 | Scoliosis:<br>0-10° 15%<br>10-30° 19%<br>30-60° 25%<br>>60° 41% | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Rodby Bousquet 2013 | Scoliosis present: 77% | Clinical examination or prior spinal fusion surgery |
| Saito 1998 | Population of "sitters" n=13<br><40° 2/13 15%<br>40–60° 7/13 54%<br>>60° 4/13 31%<br>Population of "bedridden" n=7<br>>60° 7/7 100% | Standard AP and lateral spine X-ray; calculation of Cobb angle* |
| Sato 2016 | 14/22 S-shaped curves<br>8/22 C-shaped curves<br>Mean thoracic curve 30.8°<br>Mean lumbar curve 41.1°<br>Mean Upper trunk lateral lean 5.5° Mean Lower trunk lateral lean 14.4°<br>Mean Upper trunk rotation 4.9°<br>Mean Lower trunk rotation 8.6° | Standard AP and lateral spine X-ray; calculation of Cobb angle PLUS<br><br>Calculation of 3D coordinates from anatomical landmarks for identification of lateral lean & upper and lower trunk rotation using digital cameras |
| Senaran 2007 | Scoliosis<br>ITB group: mean 56.8° (15-96°)[4]<br>Control group: mean 66.2° (24-113°)[4] | Standard AP and lateral spine X-ray; calculation of Cobb angle (in supported sitting) |
| Thometz 1998 | Scoliosis mean angle (range)<br>walks only in PT 68.3° (32-117°) n=6 indep sitter 68.7° (40-128°) n=9<br>dep sitter 76.1° (25-152°) n=21 bedridden 119.7° (59-147°) n=4 | Standard AP and lateral spine X-ray; calculation of Cobb angle (in supine) |

| | | |
|---|---|---|
| **Pelvic obliquity** | | |
| Heidt 2015 | GMFCS IV: PO range 1-14°; Md 4° IQR 2- 7°<br>GMFCS V: PO range 1– 22°; Md 7° IQR 4-11°<br><br>Geometric means of Sharp's angle using ITDL for **HIGH** side<br>GMFCS IV: 47.57 (SD 1.32),<br>GMFCS V: 53.85 (SD 1.71)<br>for **LOW** side<br>GMFCS IV: 37.43 (SD 1.39)<br>GMFCS V: 26.18 (SD 1.87) | From X-ray using intersect of horizontal reference line parallel to the frame of the radiograph with 3 reference lines: ITDL, ITL & IICL<br><br>ITDL – Line joining acetabular tear drops |
| Kalen 1992 | PO present in 57% (8/14) with scoliosis >45° | N/R |
| Knapp 2002 | PO present in 41% (12/29) | N/R |
| Littleton 2011 | R PO present (3/5);<br>L PO present (2/5); | Descriptive from physical examination |
| Pritchett 1990 | PO present in 63% (both treated hip & untreated hip groups) | Pelvic radiograph and physical examination |
| Senaran 2007 | ITB group: mean PO 10.5° (0-20°)[4]<br>Control group: mean PO 13.8° (0-45°)[4] | Radiograph: more than 10° angle between horizontal and a line connecting the most proximal points of the iliac crests |
| **Lower limb contracture** | | |
| Boldingh 2007 | 58% hip abduction limitation (>30°)<br>14% hip flexion contracture (loss of >30° extension)<br>32% knee flexion contracture (loss of >30° extension) | Goniometer |
| Kalen 1992 | hip flex contracture mean 26° for those with scoliosis >45° (n=14) | N/R |
| Knapp 2002 | 38% severe hip adduction contracture (11/29) | N/R |
| Murphy 1995 | 91% lower limb contractures | N/R |

| | | |
|---|---|---|
| Rodby Bousquet 2013 | 47% hip contractures (14/30)<br>87% knee contractures (26/30) | Goniometer |
| Sandstrom 2004 | 100% hip contracture (18/18)<br>78% knee contracture (14/18) | ROM – visually estimated scored using scale 0-2 where 0= a few degrees, 1= limited, 2= normal/almost normal ROM. |

[1]of subset with scoliosis >45°; [2]targeted recruitment; [3]9 participants had both hips affected; [4] author calculations form tables for ages 16 and over;*author calculation from graphed data; PO: pelvic obliquity; AP: antero posterior; LL: lower limbs; N/R: not reported; ITB: intrathecal baclofen; ITDL: inter-tear drop line, ITL: inter-tuberosity line, IICL: inter-iliac crest line, ROM: range of motion

### 3.6 Discussion

This scoping review was able to identify a small body of literature relating to common postural deformities in non-ambulant adults with CP (GMFCS IV and V) and describe prevalence, conservative management and measurement tools used. While it is clear that many non-ambulant adults with CP (GMFCS IV and V) experience significant and disabling postural asymmetries requiring standardised specialist health care services throughout the life span [28] there is a lack of evidence as to what interventions they require and how the efficacy of any interventions may be measured and implemented.

More specifically, this review has identified that deformities of scoliosis, pelvic obliquity and windswept hips inclusive of hip dislocation are common. A snap shot of data suggests that at least 30% of this population have a hip migration percentage of greater than 30%, more than 75% have some degree of 'scoliosis', and more than 40% demonstrate some amount of pelvic obliquity. As noted in table 3.2, either impairment on its own, or existing in a triad of deformities is a significant treatment dilemma once established [5]. These postural asymmetries increase the risk of soft tissue adaptation and the formation of contractures and progressive deformities, being most prevalent in non-ambulant people with CP [18]. The inability to change position and the time spent in an asymmetrical posture contribute to the escalating problem [18]. The temporal sequencing of scoliosis, pelvic obliquity, hip dislocation and the development of contractures is unclear [5]. More information on temporal sequencing may enhance the timing and applications of appropriate interventions. Paediatric studies have shown that hip dislocation and a severe windswept posture are preventable [10] but as this

scoping review revealed, many adults have existing asymmetries and it is unclear how to manage and monitor them once established.

This review also revealed a lack of standardised measurement techniques beyond radiographs for the monitoring of postural asymmetry. Even when radiography was used, there was a lack of standardised reporting methods, particularly for pelvic obliquity and hip outcomes. Further, patient positioning was also not standardised practice for radiography procedures, ranging from supine positioning to supported sitting for scoliosis and hip assessment. Of note, this population may not always be able to conform to the standardised positioning requirements of radiographs promoted by paediatric hip surveillance programs [30] due to extent of postural asymmetry, cognitive limitations, and/or pain.

Despite a high incidence of postural deformities described in this population, reporting and evaluation of conservative management methods are also variable and scarce. Interventions including physical therapy, seating and orthoses may be reported, but no detail was provided enabling replication, nor evaluation to identify effective strategies. Seating was most often described (as present or absent) but not in the context of a therapeutic intervention. The inadequacy of seating to meet the postural needs of the adults with CP was noted by several authors [19, 22, 27]. Given the dependency on this population for seated mobility, it would seem essential that effective postural management is considered. The Rehabilitation Engineering & Assistive Technology Society of North America (RESNA) position on seating for people with various disabilities including CP discusses the need for seating functions to realign posture, regulate spasticity, accommodate and prevent contractures and orthopaedic deformities, redistribute and relieve pressure, increase seating tolerance and comfort and to enable position change [30]. The authors would strongly support the implementation of these recommendations for all non-ambulant adults with CP.

Several limitations to this scoping review have been identified. By narrowing the study to only CP, assessment and interventions research in other fields of adult neurology, such as the RESNA guidelines already referred to, may have provided useful information for the adult CP population. However, we deliberately wished to focus on CP-specific literature given the relatively unique situation of lifelong developmental postural asymmetry experienced by this population. We also used search terms to return studies which were centred on the reporting of incidence of postural abnormalities in this population. This may have resulted in some omission of studies focused on assessment tools. Further, a focused strategy was implemented to only

review the adult CP literature. It is possible that paediatric interventions for those with CP which include the management of body shape distortion may be applicable to their adult counterparts [31, 32, 33]. However, given the potential acceleration of postural asymmetry following skeletal maturity, the efficacy of strategies that may be able to address and reverse shorter-lived asymmetry in children is unknown when applied to the long standing fixed postural asymmetries experienced by adults with CP. As with any lifelong developmental disability, management and interventions must constantly be reviewed to meet the unique needs of adults and the capacity of their (often) ageing carers to implement.

### 3.7 Implications for clinical practice and research

Valid and sensitive non-radiographic clinical measurement tools should be investigated that will enable regular monitoring of body shape over time, without the need to subject this vulnerable population to frequent radiographs. Standardised reporting of postural outcomes, via a minimum data set, should be mandated so as data from longitudinal observational studies can be pooled. The Posture and Postural Ability Scale has been identified as a valid non-radiographic clinical measurement tool for both lying, sitting and standing and should be given consideration in the management of body shape in adults with CP. Given that many adults with CP have not benefited from the now routine surveillance of impairments in paediatric practice, robust investigation of the efficacy of non-surgical interventions for the management of postural asymmetry in non-ambulant adults with CP is urgently needed.

### 3.8 Conclusion

Many non-ambulant adults with CP experience postural asymmetry associated with windswept hips, scoliosis, pelvic obliquity and limb contracture. Paediatric management of potential postural asymmetries in CP has resulted in a significant decrease in incidence of these problems over the last decade or so. This has occurred following concerted collaborative efforts to devise and implement screening programs such as hip surveillance guidelines [34, 35]. A lack of systematic, large scale follow up studies of children with CP into adulthood with thorough longitudinal assessments [28] has heightened our lack of understanding of the long term outcomes of postural asymmetries in this population. Future efforts should be targeted at how best postural care can be provided to non-ambulant adults with CP who have not benefited from such surveillance throughout their childhood, do not have access to these programs due

to resource limitations, and/or continue to progress in postural asymmetry despite surveillance and interventions.

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3.10 Supplementary Material 1

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19. 1 or 2

20. 3 or 4

21. 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18

22. 19 and 20 and 21

# Chapter 4: Background and Rationale

The scoping review (study 1) confirmed that many non-ambulant adults with CP experience postural asymmetry involving the thoracic cage, pelvis and hips. These postural asymmetries impact on the cardio respiratory and digestive systems, seating and bed positioning, pressure injury risk and pain impacting on function, quality of life and carer burden (43) Once established the asymmetries of the thorax, pelvis and hips are difficult to remediate (17). The identification, management and monitoring of the postural asymmetries through the lifespan may be challenging due to the complex three-dimensional nature of the asymmetries, which are difficult to measure through the commonly used radiographs and standard goniometry. The scoping review revealed only a small body of literature relevant to adults with CP and a lack of standardised assessment techniques.

This thesis has focused on the measurement of posture using a clinical measurement tool, the Goldsmith Indices of Body Symmetry (GIofBS) to provide objective information on chest shape and symmetry, pelvic orientation and hip mobility at an impairment level (15). The GIofBS is commonly used in England with a largely paediatric CP population, yet little information on the reliability of the GIofBS in adults with CP is available. The following chapter (study 2) explores the inter-rater and intra-rater reliability of the GIofBS in non-ambulant adults with CP and provides comparison with a group of non-disabled peers. This study was published in Disability and Rehabilitation in January 2020 (41). Carlee Holmes, Emma Fredrickson, Kim Brock & Prue Morgan (2020): The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy, Disability and Rehabilitation, DOI: 10.1080/09638288.2019.1708979

# Chapter 5: The intra and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy

5.1 Abstract

**Purpose:** Non-ambulant adults with cerebral palsy are at risk of developing asymmetry affecting thoracic cage, pelvis and hips. The primary aim of this study was to establish intra-rater and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy. The secondary aim was to establish comparative data for the Goldsmith Indices of Body Symmetry in healthy adults.

**Materials and method:** Thirty non-ambulant young adults with cerebral palsy (17 males), and 48 young healthy controls (19 males), were recruited. Thoracic shape and symmetry, pelvic orientation and hip range, was measured using the Goldsmith Indices of Body Symmetry. Intra-rater reliability was established by repeated measurement within a single session. Inter-rater reliability was established having two raters measure each participant on two sequential sessions. Analysis utilized intraclass correlation coefficients.

**Results:** The Goldsmith Indices of Body Symmetry has excellent intra-rater reliability (intraclass correlation coefficients $\geq 0.97$). Inter-rater reliability for all Goldsmith Indices of Body Symmetry measures was good to excellent (intraclass correlation coefficients $\geq 0.85$). Range and variability of results was greater for participants with cerebral palsy compared to comparative data.

**Conclusion:** The Goldsmith Indices of Body Symmetry has good inter and intra-rater reliability for measurement of thoracic shape and symmetry, pelvic orientation and hip range, allowing accurate tracking of postural changes over time in non-ambulant adults with cerebral palsy.

**Key words**

Cerebral palsy, clinical measurement, reliability, thorax, hips, pelvis, windswept

## 5.2 Introduction

Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture that are attributed to non-progressive disturbances in the developing fetal or infant brain (1). The core motor features of CP, abnormalities of movement and posture, can lead to secondary complications associated with behaviour, musculoskeletal function and participation (1). Management of musculoskeletal function leading to postural asymmetry involving spine, pelvis and hips, is an important aspect of healthcare for adults with CP. The development of postural asymmetry in this population impacts on cardiorespiratory (2, 3) and digestive function (2) and may cause pain and pressure injuries, negatively impacting carer burden (2). Windswept hips, consisting of abduction and external rotation of one hip and adduction and internal rotation of the contralateral hip (2, 4), are one element of the complex postural asymmetry most evident in non-ambulant adults with CP with low motor function typically described as Gross Motor Function Classification System (GMFCS) Levels IV and V (5). This predominantly wheelchair-user subgroup exhibit the highest risk for secondary complications and potential for shorter life expectancy (2, 6).

Diagnosing and measuring the extent of, and contributors to, postural asymmetry including scoliosis and hip mal-alignment may be particularly challenging in adults with CP who have profound disability. Although hip surveillance guidelines are generally followed for at-risk children (7), methods such as radiography to monitor spine, pelvis and hips, are problematic for adults with severe disability who cannot cognitively tolerate traditional radiographical procedures or cannot achieve a standardised supine position for radiographs due to postural asymmetry and contractures. Further, elements contributing to the complex three-dimensional postural asymmetry frequently encountered in adults with severe CP are difficult to quantify by standard methods such as goniometry and radiographs. A recent scoping review on postural asymmetry in non-ambulant adults with CP indicated there is no standardised impairment-focused clinical measurement suitable to accurately capture and objectively record common postural asymmetries of windswept hips, scoliosis, pelvic obliquity and limb contractures (8).

There is currently limited evidence available regarding the nature of postural asymmetry, progression over time, and effective interventions to prevent, minimise and/or remediate postural asymmetry in the adult CP population. Although there is considerable evidence surrounding the proportion of children with displaced hips and scoliosis (9, 10), information tracking progress of these children and their posture into adulthood is scarce. The population

of adults with severe CP need careful and regular monitoring, and deserve timely and affordable interventions to manage contributors to postural asymmetry to reduce pain and enhance life quality. The Posture and Postural Ability Scale (PPAS) (11) utilises an activity limitation framework to describe static and dynamic postures and the ability to move in and out of a static position. This scale has the potential to be utilised for long term monitoring purposes, however it does not allow close examination of contributing impairments, limiting potential for evaluation of specific interventions.

The Goldsmith Indices of Body Symmetry (12) is a clinical measurement tool providing objective information on thoracic shape and symmetry, pelvic orientation and hip mobility, enabling calculation of a Windswept Index at an impairment level. Windswept hips seriously impair seating comfort and function (13), as well as contributing to other secondary complications and functional limitations (2). The Goldsmith Indices of Body Symmetry has face validity, objectively measuring constructs relating to thoracic symmetry and shape, hip and pelvic orientation and range that contribute to posture, but lacks more robust validation. It was developed in 1992 (12) and has been predominantly used with the paediatric CP population in England. The Goldsmith Indices of Body Symmetry may be used as part of a comprehensive postural evaluation exploring key impairments in supine that contribute to postural dysfunction in activities such as lying and supported sitting. It was chosen as a clinical measurement tool potentially suited for adults with severe neuromuscular disability such as CP in this study due to the scarcity of alternate appropriate and consistent non-radiographical methods. However, little is known about the reliability of the Goldsmith Indices of Body Symmetry in documenting elements contributing to postural asymmetry often observed in non-ambulant adults with CP, and in adults without disability to provide comparative control data.

The primary aim of the study was to establish the intra-rater and inter-rater reliability of the Goldsmith Indices of Body Symmetry in adults with CP with the non-ambulant classification of GMFCS Level IV or V. The secondary aim of the study was to establish comparative data in adults without disability (control group) for the Goldsmith Indices of Body Symmetry to aid interpretation of data collected from adults with CP. Study outcomes are reported using the Guidelines for Reporting Reliability and Agreement Studies (14).

## 5.3 Methods

### 5.3.1 Setting

Participants with CP were recruited from the Young Adult Complex Disability Service (YACDS) at St. Vincent's Hospital, Melbourne, predominantly a CP transition clinic from paediatric to adult healthcare services. Participants for the comparative control data collection were recruited from the Monash University Peninsula Campus and staff from St. Vincent's Hospital Melbourne. Ethical approval was gained from St. Vincent's Hospital Melbourne and Monash University Human Research Ethics Committees.

### 5.3.2 Participants

Adults with CP: Participants aged between 17 and 40 were included if they had a diagnosis of CP and a motor classification of GMFCS level IV or V. Participants were excluded if they had a significant movement disorder, such as severe dystonia, that rendered them unable to remain relatively still during testing. Participants were also excluded if they were pregnant, or had behavioural issues that placed either raters or participant at risk.

Control data: Participants were young adults aged between 18 and 40 who self-declared no history of movement disorder and were not pregnant.

### 5.3.3 Recruitment

Adults with CP: Suitable participants attending YACDS between February 2017 and August 2018 were invited to participate and provided with study information. For participants unable to consent, the invitation was provided to next of kin / legal guardians.

Control data: Suitable participants were recruited from Monash University Peninsula Campus and St. Vincent's Hospital, Melbourne via advertisements on notice boards and online learning sites.

All participants (or next of kin) signed informed consent.

## 5.4 Outcome Measures

Adults with CP: The following demographic and medical data was retrieved: gender, date of birth, living situation, GMFCS level, CP subtype (according to Bax et al (15), usually sourced from paediatric referral facility), height and weight, prior scoliosis surgery, communication (verbal, non-verbal and nil consistent effective communication) and cognitive capacity (ability to provide own consent).

Control data: Participants' age, gender, height and weight were recorded. Any self-declared history of musculoskeletal issues potentially impacting spinal/hip alignment was sourced.

### 5.5 Goldsmith Indices of Body Symmetry

The Goldsmith Indices of Body Symmetry involves three testing procedures: chest measures; measurement of rotation of the pelvis as influenced by movement of the flexed knees; and measurement of combined hip external rotation and abduction with a stable pelvis. Data was collected using the Anatomical Measurement Instrument as per standard testing protocol (12, 16). Prior to commencement, an optimal standard starting position was established, being crook lie with 110° knee flexion and level pelvis. If this position was unable to be achieved an alternate starting position was identified, recorded, and used as the starting position for all measurement. The outcome measures collected, procedure and guide for data interpretation are described in Table 5.1.

Table 5.1. Outcome measure data obtained from Goldsmith Indices of Body Symmetry

| | Outcome Measure | Procedure | Instrument | Interpretation of results |
|---|---|---|---|---|
| **1** | **Chest Depth Width Ratio** | Chest depth value divided by total chest width value<br>(chest depth/R side chest + L side chest) | Chest frame | Values <1.0 suggest less chest depth relative to width; values >1.0 suggest greater chest depth relative to width |
| **2** | **Chest Right Left Ratio** | Right chest value divided by left chest value<br>(Right side chest / Left side chest) | Chest frame | A score of 1 is indicative of symmetry. A score of 0 to <1 is indicative of chest anticlockwise rotation. A score of >1 is indicative of chest clockwise rotation. |
| **3** | **Angle Between Legs and Pelvis (ABLAP)** | Angle of legs minus angle of pelvis<br>R ABLAP = R leg angle – R pelvic angle<br>L ABLAP = L leg angle – L pelvic angle | Leg paddle and pelvic bridge | The side (right or left) which has the higher value is the side to which the person is windswept towards |
| **4** | **Combined Hip External rotation / Abduction** | Hip joint range of motion | Leg paddle and pelvic bridge | R≠L suggests asymmetry |
| **5** | **Windswept Index (WI)** | Difference between the left and right ABLAP<br>WI = R ABLAP – L ABLAP | calculation | Higher values represent greater postural asymmetry |

[1]ABLAP, Angle Between the Legs and Pelvis; [2]ER /Abd, External Rotation / Abduction

[3]Testing of the R ABLAP and combined hip ER/Abd, and calculation of the windswept index was not possible for 3 participants; one did not have classic windsweeping (rotation of the pelvis and legs in opposite directions as opposed to legs and pelvis moving together) (12) and two participants had significant fixed lower limb contractures that prevented measurement.

[*]Significant results $p<0.05$

Note: Medians and IQR's are reported for variables that were not normally distributed. Square root transformations were used on these variables prior to ICC analysis.

## 5.6 Procedures

Adults with CP: Two experienced therapists (raters) undertook participant measurement, having undergone additional training in administration of the Goldsmith Indices of Body Symmetry Anatomical Measurement Indices. The training was provided by a physiotherapist who has gained certification of Level 3 Award in Measurement of Body Symmetry (Open College Network West Midlands).

Each participant was measured by each rater with a 15 to 30 minute rest between the two measurement sessions. Each Goldsmith Indices of Body Symmetry outcome was measured four times in succession prior to advancing to the next procedure according to Goldsmith Indices of Body Symmetry guidelines (16). Rater order was randomised using a randomisation application (http://www.randomization.com/). For inter-rater reliability, raters were blinded to results of previous testing. An overhead hoist was used to transfer participants from wheelchair to plinth where the Goldsmith Indices of Body Symmetry measurement was performed. An additional assistant and family member/carer of the participant assisted in the process as required. A testing session typically lasted 45 minutes.

Control data: One rater was involved in a single session of data collection following documented Goldsmith Indices of Body Symmetry procedure (14). Participants attended in pairs for measurement, and the assistant role was undertaken by the second participant. A testing session typically lasted up to 30 minutes.

## 5.7 Statistical Analysis

The SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for analysis. All calculations from raw data were performed according to Goldsmith Indices of Body Symmetry procedure (Table 5.1). Chest right/left ratio values were transformed to reflect magnitude of rotation rather than clockwise or anticlockwise direction. Normality of data was evaluated using visual inspection and skewness and kurtosis statistics, and descriptive statistics used to report Goldsmith Indices of Body Symmetry measures. Means and standard deviations for each Goldsmith Indices of Body Symmetry variable for data from adults with CP and control comparison were calculated. Medians and Interquartile ranges were calculated for data not following a normal distribution. Data that did not follow a normal distribution was transformed using the square root model (17) to enable further parametric data analysis.

Comparisons between adults with CP and control adults' demographic data were undertaken using Mann Whitney U or Chi Square tests. Additional exploration of any impact of scoliosis

surgery on Goldsmith Indices of Body Symmetry measurement was completed (Mann Whitney U test).

Adult with CP data was assessed for intra-rater reliability of individual Goldsmith Indices of Body Symmetry measurements (taken four times) using intraclass correlation coefficients (ICC [3,1]) and 95% confidence intervals (18) for each rater. A one-way repeated measures ANOVA was calculated to determine any difference between the four measures of individual Goldsmith Indices of Body Symmetry components for each rater. Inter-rater reliability for the CP participant data, using the mean of the four measures, was assessed using intraclass correlation coefficients (ICC [2,1]), with 95% confidence intervals (18, 19). A paired samples t-test was applied to determine any difference between the Goldsmith Indices of Body Symmetry measures by the two raters. Bland-Altman plots for each Goldsmith Indices of Body Symmetry variable were used to determine if any systematic differences across the range of values occurred between the two assessments completed by the two raters in the exploration of inter-rater reliability (20). Sample size estimates for the inter-rater reliability study phase were based on methods of Walter et al (21) which indicated that 27 participants would be required for a study with 2 observations per participant, assuming an expected ICC of 0.6, a null ICC of 0.2, a type I error of 0.05, and a type II error of 0.2. Level of significance for all analyses was set at $p<0.05$.

5.8 Results

After screening 166 potential participants, thirty adults with CP participated. Of the 136 excluded, 38 declined, 34 did not have a diagnosis of CP, 46 were not GMFCS Level IV or V, five had significant dystonia, and 13 had extreme cognitive/behavioural challenges. Forty-eight control adults participated in a single session of data collection. Demographic data describing all participants is in Table 5.2.

There was no significant difference in age nor gender distribution between adults with CP and control group ($p = 0.137$ and $p = 0.215$ respectively). Adults with CP had significantly lower weight and height than control group ($p < 0.0004$).

Table 5.2. Demographic data from all participants

| **Participant Demographics** | **Participants (CP) n=30** | **Participants (control) n=48** |
|---|---|---|
| **Age years[1], median (IQR); range** | 21.00 (18.75, 24.25); 17.00 – 38.00 | 21.50 (20.00, 26.00); 18.00 – 34.00 |
| **Gender (n)[2]** | M - 17 F - 13 | M - 19 F - 29 |
| **Living Situation** | Home with family: 27<br>Community Residential Unit: 3 | Data not collected |
| **Height cm[3], median (IQR); range** | 151.50 (144.75, 157.25); 119.00 -167.00 | 171.50 (165.25, 178.75); 151.00 – 192.00 |
| **Weight kg[3], median (IQR); range** | 46.9 (39.17, 59.27); 32.80 – 78.40 | 68.50 (60.25, 73.75 ); 51.00 – 100.00 |
| **GMFCS Level (n)** | Level IV 4<br>Level V 26 | N/A |
| **CP subtype (n)** | Spastic Quadriplegia 28<br>Spastic Diplegia 1<br>Dystonia 1 | N/A |
| **Prior scoliosis surgery (n)** | 10 | 0 |
| **Communication (n)** | Nil consistent communication 19<br>Communication device 2<br>Verbal 9 | N/A |

CP: cerebral palsy; IQR: interquartile range; M: male; F: female; GMFCS: Gross Motor Function Classification System; N/A: Not applicable

[1] no significant difference between groups (p = 0.137).

[2]no significant difference between groups (p = 0.215)

[3] significant difference between groups (p = 0.00).

5.9 Goldsmith Indices of Body Symmetry (descriptives) – Adults with CP and control data

In the CP population, all Goldsmith Indices of Body Symmetry variables were normally distributed, with exceptions being chest right left ratio and windswept index, which were not normally distributed. Transformations using square root model were utilised on these variables before conducting parametric analyses.

All individual components of the Goldsmith Indices of Body Symmetry for both adults with and without (control) CP are presented in Table 5.3. The results demonstrate that for most components, variability in measurements of adults with CP appears higher, with standard deviations more than double those observed for control participants, and both minimum and maximum scores beyond control range.

There was no significant difference in Goldsmith Indices of Body Symmetry outcomes for those who had received scoliosis surgery and those who had not ($p > 0.05$).

Table 5.3. Goldsmith Indices of Body Symmetry data for controls and adult with CP groups, with inter rater reliability Intra Class Correlations for adults with CP data

| | **Normative** | **CP Rater 1** | **CP Rater 2** | | |
|---|---|---|---|---|---|
| **Goldsmith Indices of Body Symmetry Procedures** | **Mean (SD)** Range | **Mean (SD)** Range | **Mean (SD)** Range | **ICC 95% CI P value** | **Paired t test CP data P value** |
| **Chest Depth Width Ratio** CP n = 30 | **0.62 (0.05)** 0.52–0.75 | **0.66 (0.09)** 0.50 – 0.85 | **0.67 (0.09)** 0.50 – 0.86 | 0.95 0.90 – 0.98 0.00 | 0.470 |
| **Chest Right Left Ratio (Magnitude)** CP n = 30 | **0.07 (0.05)** 0.00-0.27 | **0.16 (0.13)** 0.01–0.56 *Median (IQR) 0.12 (0.05, 0.23)* | **0.17 (0.14)** 0.03 – 0.57 *Median (IQR) 0.13 (0.06, 0.26 )* | 0.74 0.45 – 0.87 0.00 | 0.386 |
| **R ABLAP[1]** CP n = 27[3] | **122.11 (7.96)** 100.00- 135.25 | **125.47 (15.43)** 91.5-168.25 | **122.90 (14.70)** 101.00-164.50 | 0.97 0.93 – 0.98 0.00 | 0.021* |
| **L ABLAP[1]** CP n = 30 | **122.04 ( 8.89)** 96.75-139.75 | **117.73 (16.48)** 80.25-155.25 | **118.04 (12.90)** 87.25-154.25 | 0.85 0.66 – 0.93 0.00 | 0.884 |
| **Combined R ER/ Abd[2]** CP n = 28[3] | **53.61° (6.19°)** 35.00°-65.50° | **42.15°(18.08°)** 7.80°-79.00° | **43.96° (16.37°)** 15.50-78.00° | 0.95 0.88 – 0.97 0.00 | 0.229 |
| **Combined L ER/ Abd[2]** CP n = 29[3] | **55.79° (6.84°)** 41.75°69.00° | **43.14° ( 15.59°)** 2.25°-69.50° | **45.63° (14.99°)** 11.00°-71.50° | 0.94 0.87 – 0.97 0.00 | 0.075 |
| **Windswept Index** CP n = 27[3] | **3.59 (3.21)** 0.00-14.25 | **19.02 (22.74)** 0.75-81.00 *Median (IQR) 11.25 (3.75, 16.50)* | **17.19 (16.93)** 0.75-66.50 *Median (IQR) 10.75 (4.00, 28.50)* | 0.91 0.80 – 0.96 0.00 | 0.900 |

[1]ABLAP, Angle Between the Legs and Pelvis; [2]ER /Abd, External Rotation / Abduction

[3]Testing of the R ABLAP and combined hip ER/Abd, and calculation of the windswept index was not possible for 3 participants; one did not have classic windsweeping (rotation of the

pelvis and legs in opposite directions as opposed to legs and pelvis moving together) (12) and two participants had significant fixed lower limb contractures that prevented measurement.

*Significant results $p<0.05$

*Note: Medians and IQR's are reported for variables that were not normally distributed.*

5.10 Inter-rater reliability – Adults with CP

Results for inter-rater reliability of the Goldsmith Indices of Body Symmetry using mean of four measures across identified variables for Raters 1 and 2 are shown in Table 5.3. The ICC (2,1) for all variables was ≥0.85, indicating excellent agreement for all Goldsmith Indices of Body Symmetry components. Paired samples t-test indicated no significant difference between raters ($p >0.05$) for all procedures except R ABLAP ($p = 0.02$). Inspection of Bland Altman plots (Figures 5.1a-e) showed no systematic differences across the range of scores, with only between 1 (left external rotation/abduction) and 3 (windsweeping index) of all scores falling outside 95% limits of agreement.

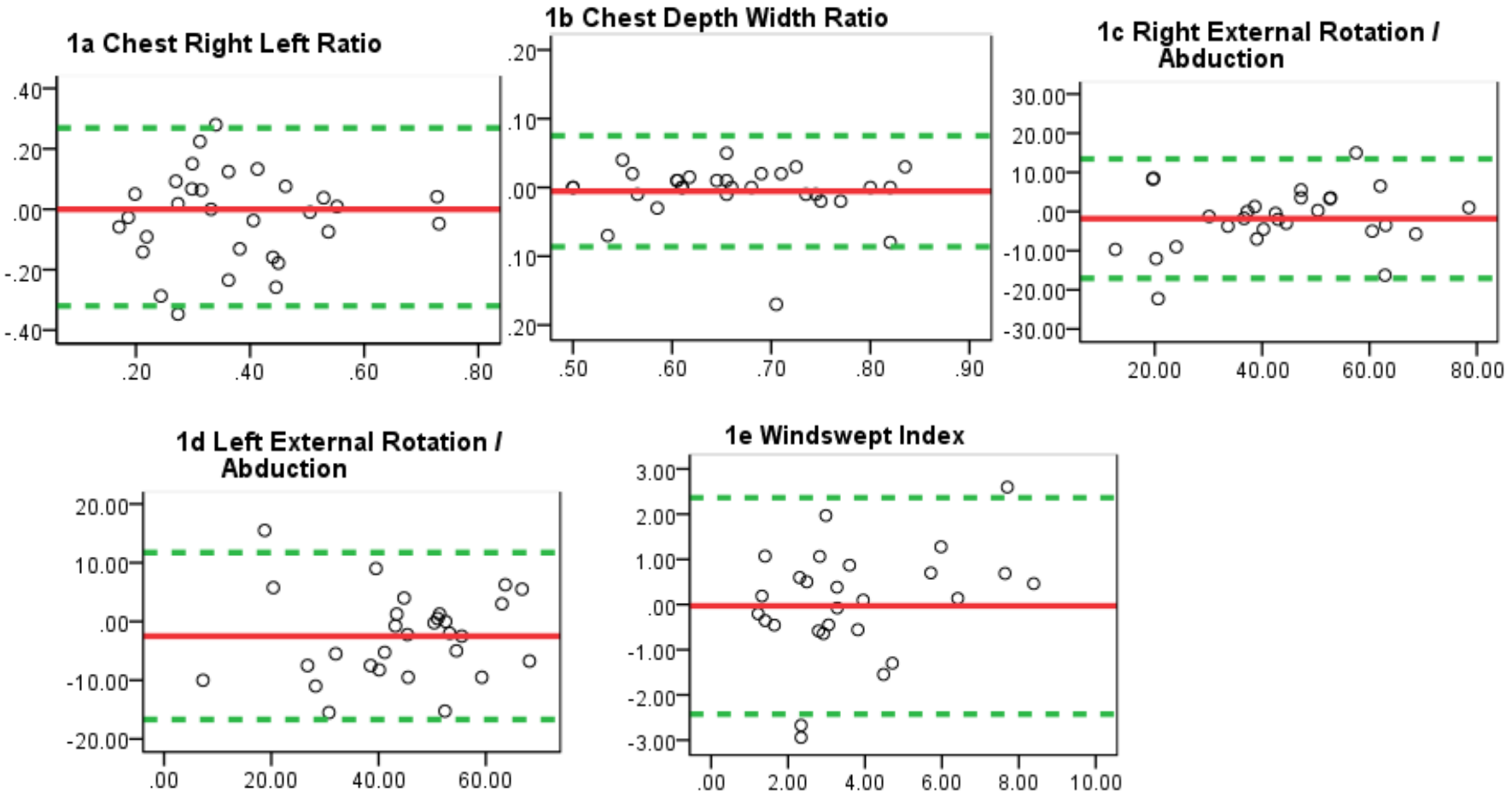


Figure 5.1. (a-e) Bland Altman plots of agreement between rater scores

Y Axis: Differences in scores (rater 1 – rater 2), X Axis: Averages of rater 1 and rater 2

Solid line: Mean Difference

Dashed line: differences +/- 95%

The ICC (2,1) using the first measure only (rather than mean of four measures) for each rater for each variable similarly resulted in high levels of inter-rater agreement (>0.80) for all components of the Goldsmith Indices of Body Symmetry (Supplementary table 5.4).

## 5.11 Intra-rater reliability – Adults with CP

Intra-rater reliability results of the four repeated measures contributing to calculation of Goldsmith Indices of Body Symmetry in adults with CP are shown in Supplementary table 5.5. The ICC (3,1) for all measurements was ≥0.97 for both raters indicating an excellent level of intra-rater agreement. A one-way repeated measures ANOVA indicated no significant difference between the four measures for each rater ($p > 0.05$) for all except Left Hip ER/Abd for Rater 1 ($p = 0.017$ between measures 3 and 4). Partial eta squared values indicate a small to very small effect size for the single significant result.

## 5.12 Discussion

This study is the first to explore the use of the Goldsmith Indices of Body Symmetry in adults with CP (GMFCS IV and V). In the present study we sought to establish the level of agreement within a single rater, and between two raters, when they measured the same variables in non-ambulant adults with CP using a standard protocol. The secondary aim of the study was to establish comparative control data for the Goldsmith Indices of Body Symmetry. Study outcomes demonstrated the Goldsmith Indices of Body Symmetry is a reliable tool for measurement of thoracic shape and symmetry, pelvic orientation and hip mobility and enables calculation of a Windswept Index in adults with CP classified as GMFCS Level IV or V. The intra-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with CP was excellent across all Goldsmith Indices of Body Symmetry domains. The inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with CP was similarly good to excellent across all Goldsmith Indices of Body Symmetry domains. Most Goldsmith Indices of Body Symmetry components had notably different distributions to those recorded from a control group.

Findings support the initial reliability study by Goldsmith and colleagues (12) and a later thesis (22) both which found the Goldsmith Indices of Body Symmetry to be a reliable measurement tool for postural asymmetry in primarily paediatric populations. The present study suggests similarly reliable outcomes can be achieved in adults with CP compared to those obtained in children using the Goldsmith Indices of Body Symmetry.

Comparison of control data to data from participants with CP demonstrated wide variability in the CP group, with minimum and maximum scores outside control range for most components. The range of scores for both the angle between legs and pelvis, and the combined hip external rotation/abduction in participants with CP varied greatly from person to person, indicating some participants had very little pelvic and hip mobility associated with fixed contractures and/or spasticity, and others had excessive mobility associated with hypotonus. As the Goldsmith Indices of Body Symmetry is not a measure of spasticity, testing movements were taken slowly to pain-free end of passive range and not to the point of a velocity dependent catch. Control data can be utilised to assist in Goldsmith Indices of Body Symmetry interpretation in people with CP, for example identifying scores that are more than two standard deviations higher or lower than control data means.

The literature provides clear definition of what constitutes a windswept hip deformity (2, 13) yet measurement of this deformity is not standardised. A mathematical algorithm was developed by Young and colleagues (13) and later used in a modified version (9, 23) whereby at least 50 % difference in abduction, internal, and/or external rotation between left and right hips was defined as 'windsweeping', a two dimensional outcome. The Goldsmith Indices of Body Symmetry Windswept Index offers a three-dimensional solution using combined measurement of hip mobility less the influence of the pelvic mobility, thus more accurately reflecting the functional problem. As expected, the control population's mean windswept index was close to symmetry (0) but the CP population showed a large departure from symmetry (>0). Contributing factors to an elevated windswept index may include an asymmetrical static sleeping posture with the influence of gravity on hip and knee flexion contractures causing lower limbs to fall to one side. The inability to change position and the influence of spasticity may also contribute in those with severe disability. Interestingly, the presence or absence of prior scoliosis surgery did not impact Goldsmith Indices of Body Symmetry results. This may be an area for future exploration. The relationship of Goldsmith Indices of Body Symmetry hip and pelvic measurements may enable ready monitoring of change over time and an opportunity to evaluate the effect of interventions.

According to the published testing protocol (16) each individual component of the Goldsmith Indices of Body Symmetry should be performed four times with data average and range calculated. Training in use and application of a standard protocol for instrument usage minimises error. Repeating the measurement four times may enable an aberrant result due to a momentary spasm or measurer error to be noted (12, 16). Despite exclusion of participants

with extreme behavioural challenges and severe dystonia, this study was conducted on a very challenging participant population of adults with CP with a high incidence of spasticity and cognitive deficits. Varying results between measures due to spasms, pain and behavioural challenges would not be unusual in this population. Despite this, high intra-rater reliability across the four measures for each rater (ICC ≥0.98) was achieved. An acceptable intra-rater reliability (ICC ≥ 0.80) based on the first of the four measures only was also achieved. Thus the Goldsmith Indices of Body Symmetry remains a reliable measurement tool for participants who may not be able to tolerate four measures. A high inter-rater reliability (ICC ≥0.85) for almost all components of the Goldsmith Indices of Body Symmetry further confirms different yet experienced raters can undertake testing and subsequent re-testing with confidence in reliability.

### 5.13 Limitations

A limitation of the study was that both raters were formally trained in the Goldsmith Indices of Body Symmetry. Further research is required to examine reliability in non-accredited therapists. It is also acknowledged that establishment of intra-rater reliability occurred within a single test session, using published Goldsmith Indices of Body Symmetry protocol of repeating measurements four times each. Although this resulted in assessor non-blinding, we would argue the need to reset participant's position and equipment between each measurement reduced any influence of prior measurement on subsequent measurement.

A further limitation was the inability to accurately capture pelvic obliquity with the tool, an abnormal tilt of the pelvic girdle that may occur in all three movement planes. It is associated with progressive scoliosis and hip dislocation, pain and necrosis of ischial tuberosities (24). Pelvic obliquity is part of a triad, together with scoliosis and windswept hips, which is extremely difficult to treat once established (4). The Goldsmith Indices of Body Symmetry captures elements of pelvic asymmetry through the pelvic bridge apparatus, specifically rotation in the frontal plane, however the contribution of pelvic obliquity is unable to be teased out from that of hip contracture. The potential to specifically measure pelvic obliquity using the Goldsmith Indices of Body Symmetry pelvic bridge requires additional investigation.

In some participants, severe lower limb contractures prevented complete measurement of all Goldsmith Indices of Body Symmetry elements. Also, potential participants with movement disorders such as severe dystonia, and extreme behavioural issues were excluded. In these cases alternate measurement processes remain challenging and require further investigation.

Finally, it is recognised that the Goldsmith Indices of Body Symmetry takes an impairment-focus to postural asymmetry measurement. The impact on a person's function, such as seating ability, is not described. As previously indicated, the PPAS describes static and dynamic posture of adults with CP using an activity-based focus (11). Although the PPAS can detect postural asymmetry (present or absent) in a range of functional positions, it does not quantify posture, nor postural directional change over time. The inclusion of both the PPAS and Goldsmith Indices of Body Symmetry would enable comprehensive clinical assessment for ongoing monitoring and treatment planning for postural management of adults with CP.

## 5.14 Conclusions

Many non-ambulant adults with CP experience asymmetry of thoracic cage, pelvis and hips which may progress after skeletal maturity. The Goldsmith Indices of Body Symmetry enables reliable quantification of this asymmetry. The provision of a Windswept Index, thoracic shape and symmetry, and combined influence of hip rotation and abduction/adduction provides valuable information regarding lifespan management of adults with CP. The Goldsmith Indices of Body Symmetry is a reliable measurement tool to measure and monitor asymmetry of the thorax, pelvis and hips in this population.

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Declaration of interest

The authors have no declaration of interest to disclose

Table 5.4. Supplementary table: GIofBS data adult with CP for first measure only with inter-rater reliability Intra Class Correlations

| | **CP Rater 1** | **CP Rater 2** | | |
|---|---|---|---|---|
| **GIofBS Procedures** | **Mean (SD)**<br>Range | **Mean (SD)**<br>Range | **ICC**<br>**95% CI**<br>**P value (0)** | **Paired t test**<br>**P value** |
| **Chest Depth Width Ratio** | **0.65 (0.10)**<br>0.50 0.83 | **0.65 (0.11)**<br>0.45 0.89 | 0.89 | 0.56 |
| **Chest Right Left Ratio (Magnitude)** | **0.35 (0.16)**<br>0.00 0.74<br>*Median (IQR)*<br>*0.37 (0.28 0.46)* | **0.38 (0.16)**<br>0.00 0.68<br>*Median (IQR)*<br>*0.39 (0.28 0.50)* | 0.81<br>0.60 0.90 | 0.21 |
| **R ABLAP[1]**<br>CP n = 27[3] | **124.32 ( 15.38)**<br>85.00 168.00 | **122.78( 15.39)**<br>101.00 164.00 | 0.93<br>0.84 0.97 | 0.28 |
| **L ABLAP[1]** | **116.98 (17.25)**<br>75.00 155.00 | **116.18 (14.04)**<br>85.00 152.00 | 0.80<br>0.55 0.90 | 0.90 |
| **R ER/ Abd[2]**<br>CP n = 28[3] | **41.84° (18.17°)**<br>8.50° 79.00° | **44.18° (16.70°)**<br>14.00° 76.00° | 0.92<br>0.82 0.96 | 0.21 |
| **L ER/ Abd[2]**<br>CP n = 29[3] | **43.14° (15.77°)**<br>2.00° 70.00° | **45.96° (15.52°)**<br>12.00° 72.00° | 0.93<br>0.84 0.96 | 0.07 |
| **Windswept Index**<br>CP n = 27[3] | **3.75 (2.36)**<br>0.00 8.94<br>*Median (IQR)*<br>*3.32 (2.45 4.58)* | **3.93 (1.91)**<br>1.00 8.37<br>*Median (IQR)*<br>*3.46 (2.65 5.00)* | 0.88<br>0.74 0.95 | 0.50 |

[1]ABLAP, Angle Between the Legs and Pelvis; [2]ER /Abd, External Rotation / Abduction

[3]Testing of the R ABLAP and hip ER/Abd, and calculation of the windswept index was not possible for 3 participants; one did not have classic windsweeping (rotation of the pelvis and

legs in opposite directions as opposed to legs and pelvis moving together) (33) and two participants had significant fixed lower limb contractures that prevented measurement.

*Significant results $p<0.05$

*Note: Medians and IQR's are reported for variables that were not normally distributed. Square root transformations were used on these variables prior to ICC analysis.*

Table 5.5 Supplementary table: Intra Class Correlations for intra-rater reliability

| **Variables** | | **Rater 1 ICC 95% CI** | **Rater 1 P value** | **Rater 2 ICC 95% CI** | **Rater 2 P value** |
|---|---|---|---|---|---|
| **Chest Width** | **Right** | 0.99 0.98, 0.99 | 0.125 | 0.98 0.97, 0.99 | 0.845 |
| | **Left** | 0.99 0.98, 0.99 | 0.244 | 0.99 0.97, 0.99 | 0.284 |
| **Chest depth** | | 1.00 0.99, 1.00 | 0.390 | 1.00 0.99, 1.00 | 0.270 |
| **Pelvic angle** | **Right** | 0.99 0.98, 0.99 | 0.747 | 0.99 | 0.173 |
| | **Left** | 0.99 0.98, 0.99 | 0.681 | 0.99 0.96, 0.99 | 0.653 |
| **Leg angle** | **Right** | 0.99 0.98, 0.99 | 0.522 | 0.99 0.97, 0.99 | 0.414 |
| | **Left** | 0.99 0.99, 1.00 | 0.152 | 0.99 0.98, 0.96 | 0.222 |
| **External Rotation /Abduction** | **Right** | 1.00 0.99, 1.00 | 0.089 | 0.99 0.98, 0.99 | 0.129 |
| | **Left** | 1.00 0.99, 1.00 | 0.017* | 0.99 0.98, 0.99 | 0.139 |

- Significant results $p<0.05$

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# Chapter 6: Background and Rationale

It has been established that non-ambulant adults with CP exhibit higher risks for secondary complications and potential for a shorter lifespan (10, 44) and would benefit from lifespan measurement and management of the progressive postural asymmetries affecting the thoracic cage, pelvis and hips. Radiographs are commonly used, especially in paediatric settings as a part of the recommended hip surveillance programs (23). In addition, due to established contractures, cognitive or behavioural challenges, the standardised positioning required for radiographs may be difficult to achieve.

The GIofBS is able to measure the three dimensional rotary nature of the asymmetries and was found in the previous chapter to have excellent intra-rater reliability and good to excellent intra-rater reliability (41). The constructs captured with the GIofBS have noted relevance to seated and sleeping positions impacting on pain, pressure risks and function in this population. Further, given the focus on key elements of postural asymmetry that need to be considered for functional management of adults with complex physical disability (such as chest shape, hip range) it has the potential to value add to the information available via radiography. The following chapter (Study 3) aimed to explore the relationship between selected components of the GIofBS and the specific radiographic measures relating to scoliosis and hip dislocation in non-ambulant adults with cerebral palsy. This study is in press in the New Zealand Journal of Physiotherapy. Carlee Holmes, Kim Brock & Prue Morgan (2021): The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy, New Zealand Journal of Physiotherapy, In Press, DOI: 10.15619/NZJP/49.1.03

# Chapter 7: The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy.

## 7.1 Abstract

Non-ambulant adults with cerebral palsy (CP) are commonly affected by progressive secondary, debilitating musculoskeletal issues which may be clinically measured using the Goldsmith Indices of Body Symmetry (GIofBS). The primary aim was to explore relationships between clinical outcomes and relevant radiographic measures in non-ambulant adults with CP. Thoracic shape and symmetry, pelvic orientation and hip range was measured using the GIofBS. Radiographs of pelvis and spine were reviewed. Pearson correlation (r) was undertaken to assess relationships between clinical and relevant radiographic measures. Positioning and readability of radiographic data in 30 non-ambulant adults with CP was variable. Minimal to no correlation between paired measures of radiographic and clinical data for trunk, pelvis and hips was found ranging from lowest correlation: r(15)= -0.09, p=0.620 (left migration percentage and hip range) to highest: r(15)= -0.25 , p=0.200 (right hip morphology scale and hip range). The complex three-dimensional nature of asymmetries of thorax, pelvis and hips, measured clinically with the GIofBS provides valuable yet different postural information to that obtained by radiographs. Inclusion of both radiographs and the GIofBS would enable comprehensive lifespan assessment for postural management of adults with CP.

Keywords

Cerebral palsy, Posture, Windswept hips, Scoliosis, Radiograph

## 7.2 Introduction

The secondary musculoskeletal consequences of cerebral palsy (CP), a permanent life-long condition acquired before, during or after birth, are progressive and often debilitating. The arising postural asymmetries affecting the spine, pelvis and hips may result in further adverse consequences such as pain and pressure injuries (Gudjonsdottir & Mercer, 1997), especially for non-ambulant adults with CP. The risk of mortality due to respiratory disease in adults with CP is much greater than the general population (Ryan et al., 2019) with postural asymmetry of the thoracic cage contributing to increased risk in the most severely affected adults (Horimoto et al., 2012). Postural asymmetry involving limited hip flexion, pelvic obliquity, trunk asymmetry, scoliosis and windswept hip posture is common in adults with CP with low motor function (Ágústsson et al., 2018) described as Gross Motor Function Classification System (GMFCS) levels IV and V (Palisano, 1997). These asymmetries typically occur alongside pain and spasticity, further adversely affecting function and participation (Benner et al., 2019).

The identification, monitoring and management of secondary postural complications for adults with CP is critical given the impact on many domains of health and functioning alongside carer burden and economic impacts including specialised equipment, support workers, hospital admissions and reliance on crisis services (Collis et al., 2008; Gudjonsdottir & Mercer, 1997). This presents particular challenges for non-ambulant adults with CP who frequently have limitations in communication and cognition. Despite potential importance, there is a lack of standardised measurement techniques beyond radiographs to record postural asymmetry in this population (Benner et al., 2019; Holmes et al., 2018). In addition, capturing the complex three-dimensional asymmetry of the thoracic cage and windswept hips with a reliable measurement tool can prove even more challenging. Physiotherapists are well placed to fill this critical surveillance role both within standard and advanced scope of practice roles (World Confederation for Physical Therapy, 2019).

When radiographic surveillance is possible, there are limitations in interpretation of objective findings for those with significant postural asymmetry. The Cobb angle and migration percentage (MP) are recommended radiographic measurements used to assess status of scoliosis and hip displacement respectively in those with CP. The Cobb angle is a radiographic objective measure of the extent of spinal curvature (Cobb, 1948), reported in degrees, with scoliosis defined as Cobb angle $\geq 10°$ (Oda et al., 2017). The MP is a radiographic measure (in percentage) of the amount of ossified femoral head not covered by the ossified acetabulum (Reimers, 1980). The Australian Hip Surveillance Guidelines considers MP of $\leq 10\%$ to be

normal and MP ≥ 30% as abnormal or 'at risk' (M Wynter et al., 2014). The Cobb angle and MP represent two-dimensional measures of complex three-dimensional skeletal deformities, and may be limited in effectively documenting functional postural deformity. The rotary components of thoracic asymmetry and windswept hips are thus difficult to ascertain with radiographic studies alone in non-ambulant adults with CP. Due to the potential for progression of scoliosis and hip displacement in this vulnerable population, and aforementioned limitations with radiographic monitoring, there is a need for an additional valid and reliable clinical measurement tool that can capture three-dimensional elements of posture to be used alongside radiographs to assist in functional management.

The Goldsmith Indices of Body Symmetry (GIofBS) is a clinical measurement tool providing a systematic, objective, three-dimensional approach to identification of asymmetry of chest, pelvis and hips (Goldsmith et al., 1992). The chest measures provide a component of axial rotation and the hip and pelvic measures occur across more than one plane of movement, as compared to the views obtained from plain radiographs and goniometer measures which only provide anterior / posterior or lateral measures. The GIofBS was chosen as a clinical measurement tool potentially suited for adults with severe neuromuscular disability due to the scarcity of alternate appropriate and reliable tools able to capture three-dimensional aspects of complex postural asymmetry in this population. The constructs captured with the GIofBS have noted relevance to seated and sleeping positions impacting on pain, pressure risks and function. Satisfactory intra- and inter-rater reliability of the GIofBS have recently been determined in non-ambulant adults with CP and control data established (Holmes et al.).

Exploration of the relationships between radiographic and anthropomorphic measurements of postural deformity in adults with CP may provide valuable information to assist clinical management of adults with complex disabilities and suggest effective assessment tools to identify specific elements of postural asymmetry. Continuity of care and specialist knowledge are two of the identified barriers to effective transitioning of young adults from paediatric to adult healthcare services (Burns et al., 2014) which may be addressed with use of the GIofBS across the lifespan.

### 7.3 Objectives

The study objectives were to establish the relationship between radiographic and anthropomorphic measurements of postural deformity in adults with CP, such as described with

the GIofBS. Specifically, this study aimed to explore any relationship between GIofBS measures and radiographs in adults with CP classified as GMFCS levels IV or V.

## 7.4 Methods

This cross-sectional study used data arising from 30 adults with CP, some of whom participated in a measurement tool reliability study to undertake secondary analysis of previously unreported radiographic data (Holmes et al., 2020).

### 7.4.1 Participants and Setting

All patients referred to the Young Adult Complex Disability Service (YACDS) between February 2017 and December 2018 were considered for inclusion. Participants who had a diagnosis of CP, GMFCS level IV or V were eligible for inclusion. Participants were excluded if they had a severe movement disorder or behavioural issues that placed either themselves or raters at risk during measurement, or if they were pregnant. A total of 30 adults with CP completed the study.

Ethical approval was gained from St Vincent's Hospital, Melbourne Human Research Ethics Committee (HREC/16/SVHM/148). All participants (or next of kin) signed informed consent.

## 7.5 Outcome Measures

### 7.5.1 Goldsmith Indices of Body Symmetry

The relevant GIofBS outcome measures collected for analysis were a) chest right left ratio (indicative of chest rotation); b) combined hip external rotation / abduction (left and right); and c) the Windswept index (indicative of degree of asymmetry between left and right pelvis / lower limbs) (Goldsmith et al., 1992; Goldsmith & Goldsmith, 2013) as per standard testing protocol. These measures have previously been shown to have excellent inter and intra rater reliability in this population (Holmes et al., 2020). The components of the GIofBS Anatomical Measurement Instrument are illustrated in Figure 7.1 with further illustrations of the measurement process provided in Figure 7.2.

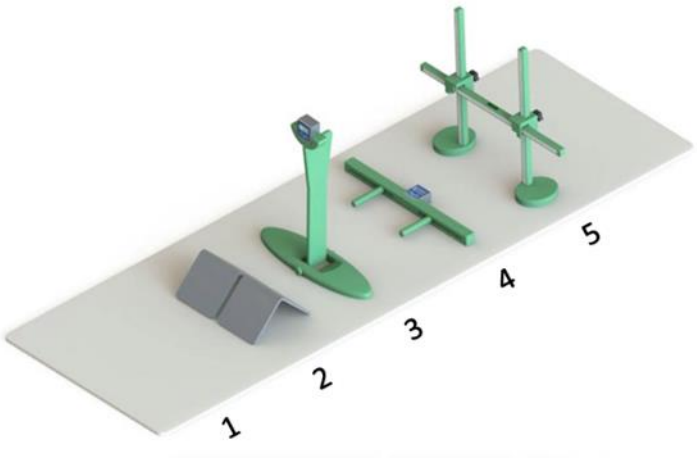


Figure 7.1. Anatomical Measurement Instrument

*Note.* Anatomical Measurement Instrument with equipment listed from left to right: 1 non slip mat; 2 foot brackets; 3 leg paddle with level box angle sensor; 4 pelvic bridge with level box angle sensor; 5 chest frame (27).


Figure 7.2. (a-c) Goldsmith Indices of Body Symmetry Measurement Process

*Note.* Depiction of the Goldsmith Indices of Body Symmetry measurement processes: a) chest frame to gain chest right left ratio and chest depth width ratio; b) pelvic bridge and leg paddle to gain measures used to calculate Windswept Index; c) leg paddle to gain right hip external rotation / abduction.

### 7.5.2 Hip/spine radiographs

Antero-posterior (AP) radiographs of the pelvis and spine obtained within 12 months of the collection of the GIofBS data were reviewed. The Australian Hip Surveillance Guidelines for Children with Cerebral Palsy recommend 12 monthly surveillance continue beyond skeletal maturity in the presence of abnormal MP, progressive scoliosis or significant pelvic obliquity (M Wynter et al., 2014), thus a 12 month time frame was considered acceptable. All radiographic measurements were completed using tools within a picture archiving and communication system (PACS) (Synapse™, Fujifilm Corp., Tokyo, Japan).

## 7.6 Procedures

GIofBS measurements were performed by an experienced therapist (rater) having undergone additional training in administration of the GIofBS Anatomical Measurement Indices as per standard testing protocol (Goldsmith & Goldsmith, 2013). A testing session typically lasted 45 minutes.

Radiographic evaluation and measurement for each participant was undertaken by a senior orthopaedic physiotherapist who is a postgraduate research fellow with 10 years experience and responsibility for radiographic evaluation and measurement in a clinical musculoskeletal surveillance service. Measurement of MP, Cobb angle and pelvic obliquity and grading hip status according to the Melbourne Cerebral Palsy Hip Classification Scale (MCPHCS) (Robin et al., 2009) were undertaken. The MCPHCS is a six level radiographic ordinal scale used to classify morphology of the skeletally mature hip. The classification covers a wide range of radiographic features, from a Grade I (normal hip), through to a Grade V (dislocated hip) and Grade VI (dislocated hip that required salvage surgery). The classification includes sub-classifications for femoral head deformity, acetabular deformity and pelvic obliquity (Robin et al., 2009; Shrader et al., 2017). Pelvic obliquity was measured as the angle between the inter tear drop line (ITDL) and a horizontal reference line parallel to the frame of the radiograph (Heidt et al., 2015). If the ITDL was obscured by gonadal shielding, the inter-ischial or inter-iliac crest line was used (Heidt et al., 2015). A quality rating was provided for each radiograph (not readable, readable, challenging to read), and the position in which the spine radiograph was obtained was recorded (supine, sitting, not reported). Any limitations in evaluating the radiograph and obtaining valid measurements was recorded, along with any reason for missing data (e.g. inadequate participant position invalidating measurement). If more than one spinal curve was present, the largest Cobb angle was selected. The highest migration percentage (left or right hip) was noted. The researcher was blinded to GIofBS results for the participant.

The following paired data were selected for exploration of any relationship between GIofBS and radiographic measures based on similarity of investigation of a specific skeletal area (i.e. spine, hip).

1. Chest right left ratio compared to Cobb angle (largest angle if >1 curve)

2. Combined hip external rotation and abduction compared to MP (right and left)

3. Combined hip external rotation and abduction compared to Melbourne Cerebral Palsy Hip Classification Scale (MCPHCS) (right and left)

4. Windswept Index compared to the highest MP (either right or left)

### 7.7 Statistical Analysis

The SPSS statistical software version 24 (SPSS Inc., Chicago, Illinois) was used for all quantitative analysis. Normality of all data was evaluated using visual inspection of the histogram and evaluation of the Kolmogorov-Smirnov statistic, with $p>0.05$ satisfying normal distribution. Mean scores and standard deviations for each variable were subsequently calculated once normative data was confirmed. The following adjustments to the data were made if required to facilitate analyses. The GIofBS chest right left ratio was adjusted to reflect the magnitude of the measure rather than a positive or negative value (indicative of rotation in a clockwise or anticlockwise direction). The Windswept index was adjusted to reflect absolute values rather than positive and negative values either side of zero.

Parametric analysis (Pearson's r) was undertaken to assess correlation between paired GIofBS outcomes against radiographic measures (Cobb angle, MP and MCPHCS). The strength and direction of any relationships (r) were established according to Cohen (1988), where 0.1 to 0.29 = small, 0.30 to 0.49 = medium and 0.50 to 1.0 = large effect size. Significance was set at $p<0.05$.

Scatterplots were constructed to visualise relationships between GIofBS variables and key radiographic data. Individual cases were identified that were outside the cut offs previously established from the literature for the Cobb angle and MP (Oda et al., 2017; M Wynter et al., 2014). For GIofBS measures, a priori decision was made to use control data to calculate cut off cases sitting above or below two standard deviations (95% of cases) from the mean (Holmes et al., 2020), described as very high or very low scores.

## 7.8 Results

After screening 165 potential participants, 30 adults participated in the study, median age 19 (range 17 – 38 years). Of the 135 excluded, 36 declined, 34 did not have a diagnosis of CP, 46 did not have function classified at GMFCS level IV or V, five had a severe movement disorder (dystonia), nine had significant cognitive/behavioural challenges, and five did not have radiographs available. Twenty-nine of the 30 included participants had CP sub-type quadriplegia and one had diplegia. Ten participants had previous surgery for scoliosis. Twenty participants had no consistent communication methods, two used communication devices and eight were verbal communicators.

Positioning for spinal radiographs was variable, with nine of 30 performed in supine, 13 in sitting, six with an undocumented position and two were missing spinal radiographs. The quality of the spinal radiographs also varied with five of 30 not readable, and five reported as challenging to read. Only two of the 30 hip radiographs were not readable in a valid and reliable manner due to extreme positioning of the hip into fixed abduction and abduction / external rotation (Figure 7.3).


Figure 7.3. Example of radiographic view of pelvis and hips from which migration percentage is calculated

*Note.* Right hip migration percentage: 100%. Left hip migration percentage: unable to complete valid and reliable assessment and measurement (Reimers, 1980) due to hip positioning in extreme abduction and external rotation.

Demographic and postural data for the cohort is presented in Table 7.1 and Table 7.2 respectively. The total number of participants included in the spinal and hip radiograph data was 28 and 30 respectively. Of note, from radiographs, 10 hips were reported as 'at risk', only eight hips were considered within normal limits (MP≤ 10%), 19 participants had a documented scoliosis and 27 participants had pelvic obliquity. Cobb angles as high as 93° were identified, and pelvic obliquity ranged from 0° (two participants) to 29° (two participants). The majority of hips (15 right / 16 left) were described as Grade III using the MCPHCS (Table 7.2).

Table 7.1. Patient Demographics

| Demographic data | |
|---|---|
| Gender, n | Males, 17<br>Females, 13 |
| Age, years Md, range | 19, 17-38 |
| GMFCS level, n | IV: 3<br>V: 27 |

Table 7.2. Participant Postural Data

| GIofBS Data* | Mean (sd)<br>Range | |
|---|---|---|
| Chest Right Left Ratio,<br>Magnitude | 0.16 (0.13)<br>0.01 – 0.56 | |
| Windswept index,<br>Absolute values | 19.02 (22.74)<br>0.75 – 81.00 | |
| Right ER/Abd | 43.79° (19.57°)<br>7.80° – 79.00° | |
| Left ER/Abd | 46.19° (16.43°)<br>2.25° – 70.00° | |
| Radiographic Data | Mean (sd)<br>Range | Incidence, n |
| Highest MP[1] | 31% (22%)<br>0% - 100% | <10°: 8<br>10°- 30°: 38<br>31°- 99°: 8<br>100°: 2 |
| Largest Cobb angle[2] | 32° (24°)<br>7° - 93° | < 10°: 3<br>10° - 30°: 10<br>30° - 60°: 6<br>> 60°: 3 |
| Pelvic Obliquity[3] | 8° ( 9°)<br>0° - 29° | < 10° : 21<br>11°- 20° : 5<br>21°- 30° : 3 |

| MCPHCS[4] Right Hip / Left Hip, n | | | | | |
|---|---|---|---|---|---|
| Grade I | Grade II | Grade III | Grade IV | Grade V | Grade VI |
| 2 / 0 | 6 /7 | 15 / 16 | 3 / 4 | 1 /1 | 1 / 0 |

[1] n=60; 4 unreadable quality, [2]n=2 missing spinal radiographs; n= 6 unreadable quality, [3]n = 1 unreadable quality, [4]2 participants unable to be graded due to poor radiograph quality

ER/Abd External Rotation / Abduction, MP Migration Percentage, MCPHCS Melbourne Cerebral Palsy Hip Classification Scale

*****Control data for comparison: Mean (sd), range:

Chest Right Left Ratio: 0.07 (0.05), 0.00 – 0.27,

Windswept index: 3.59 (3.21), 0.00 – 14.25,

Right ER/Abd 53.61° (6.19°), 35.00° – 65.50°; Left ER/Abd 55.79° (6.84°), 41.75° – 69.00°

Any association between radiographic and GIofBS outcomes for trunk, pelvis and hips was explored. Minimal to no correlation between paired measures of radiographic and GIofBS data was found ranging from lowest correlation: r(15)= -0.09, p=0.620 (left MP and left ER/Abd) to highest: r(15)= -0.25, p=0.200 (right MCPHCS and right ER/Abd) (Table 7.3).

Table 7.3. Correlations between Goldsmith Indices of Body Symmetry and radiographic Data

| Variables | Correlation r [Pearson] | *p* value |
|---|---|---|
| Cobb angle and Chest Right Left Ratio | 0.13 | 0.580 |
| Left MP and Left ER/Abd | -0.09 | 0.620 |
| Right MP and Right ER/ Abd | -0.19 | 0.330 |
| MP and WI | -0.23 | 0.240 |
| Right MCPHCS and Right ER/Abd | -0.25 | 0.200 |
| Left MCPHCS and Left ER /Abd | -0.19 | 0.350 |

MP, migration percentage; ER / Abd, External Rotation / Abduction; WI, Windswept Index; MCPHCS, Melbourne Cerebral Palsy Hip Classification Scale

Scatterplots were constructed to illustrate the relationship between windswept index and highest MP (Figure 7.4), right MP and right abduction/external rotation (Figure 7.5), and highest Cobb angle and Chest right left ratio (Figure 7.6). Scatterplots of these comparisons demonstrate the majority of participants had Cobb angles and MP values above control cut off values. In comparison, for GIofBS values, more people with CP fell within control range (+/- 2 standard deviation (sd)).

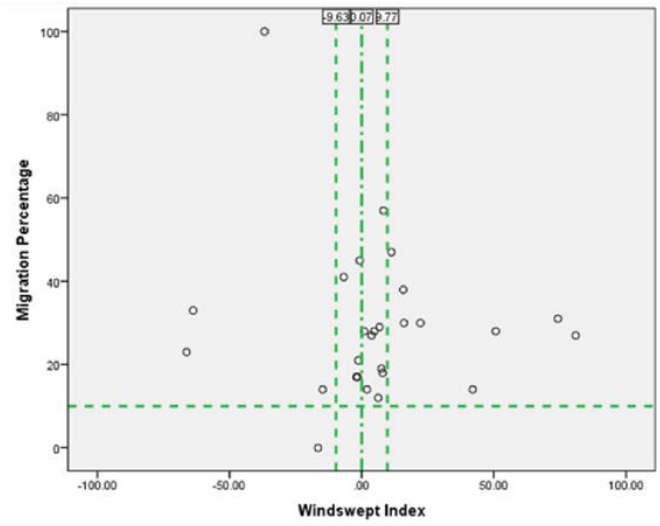


Figure 7.4. Scatter plot depicting relationship between Windswept Index and the highest Migration Percentage

*Note. The vertical dotted lines represent the mean and +/- 2 standard deviation and the horizontal represent 10° migration percentage.*

Figure 7.4 depicts the relationship between the Windswept Index and the highest MP. It illustrates that 28 of 29 participants had a MP of concern (MP ≥ 10°) yet only 12 participants had a Windswept Index of concern (outside 2 sd of control data).

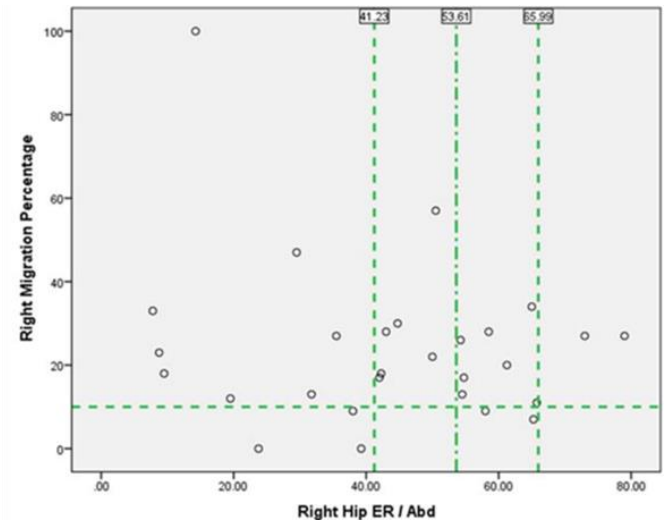


Figure 7.5. Scatter plot depicting relationship between the right Migration Percentage and right Hip External Rotation / Abduction

*Note. The vertical dotted lines represent the mean and +/- 2 standard deviation and the horizontal represent 10°migration percentage.*

Figure 7.5 depicts the relationship between right MP and right hip ER / Abd. It illustrates that 23 of 28 participants had right MP of concern (MP ≥ 10°) yet only 10 of these also had hip range of concern (outside two sd of control data).

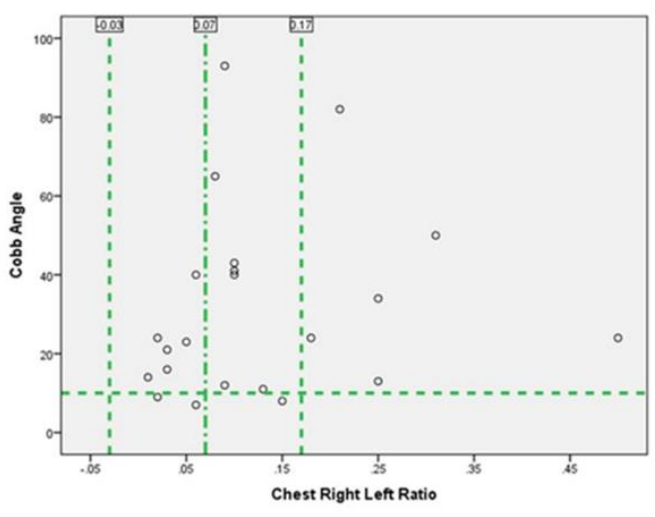


Figure 7.6. Scatter plot depicting the relationship between the highest Cobb Angle and Chest Right Left Ratio

*Note. The vertical dotted lines represent the mean and +/- 2 standard deviation and the horizontal represent 10° Cobb Angle.*

Figure 7.6 depicts the relationship between the highest Cobb angle and the chest right left ratio. It illustrates that while 19 participants had a Cobb angle of concern (≥10°) only six also had a chest right left ratio of concern (outside two sd of normative data).

## 7.9 Discussion

This study is the first to explore relationships between an anthropometric measurement tool (GIofBS) for thoracic shape and symmetry, pelvis and hips, and similar radiographic measures in a sample of non-ambulant young adults with CP attending a large metropolitan healthcare service. As found in previous studies (Holmes et al., 2018), the incidence of hip displacement and scoliosis was high with 19 of 22 participants demonstrating a scoliosis ≥ 10° and 48 hips demonstrating a MP ≥ 10°. All correlations between paired GIofBS variables and radiograph outcomes were small and not significant suggesting these tools are measuring two different constructs.

Study findings highlighted that measures of WI alone (using the GIofBS) cannot be used to predict the presence or absence of hip displacement (MP). Similarly, hip MP is not related to hip mobility range in this cohort, as those with higher MP values demonstrated hip range values both higher and lower than hip external rotation/abduction range observed in control comparisons. The radiographic MCPHCS also bore minimal relationship to anthropometric constructs as measured with the GIofBS. Previous studies have also found that physical examinations of joint range of motion via goniometry cannot replace information gleaned from radiographs in children with CP (Hägglund et al., 2007; Pruszczynski et al., 2016; Soo et al., 2006) as physical examinations of hip range in a paediatric study was a poor indicator of risk of hip displacement (Hägglund et al., 2007). In this study, excess hip movement range was typically unilateral, associated with windswept hips and fixed postural deformity, often of an extreme nature in most participants. It is therefore not surprising that the MCPHCS and the GIofBS Hip ER/Abd showed little relationship considering the MCPHCS is used to describe hip morphology as opposed to the GIofBS measure of hip mobility (GIofBS Hip ER/Abd).

GIofBS measures of chest asymmetry (rotation) in this study also showed little correlation with radiographic measures of Cobb angles, suggesting it is measuring a different spinal construct. Only six of 22 participants demonstrated both clinically significant Cobb angles (scoliosis) and extreme chest asymmetry. Previous studies exploring parameters correlated with the Cobb angle have demonstrated varying results (Sato et al., 2016; Suzuki et al., 1993), possibly indicative of the complex nature of the thoracic cage deformity. For example, three dimensional aspects of scoliosis involving lateral deviation and spinal rotation measured with ultrasound was found to have only a small (but significant) correlation to the Cobb angle in a cohort of 11 children with CP (Suzuki et al., 1993). Frequent documentation of the three dimensional nature of the rotational postural deformity of the thoracic cage will thus require additional clinical tools, beyond radiographs and ultrasound, such as the GIofBS to provide a comprehensive understanding of the stability of the thoracic asymmetry and any responsiveness to interventions.

A recent scoping review on postural asymmetry in adults with CP noted that non-standardisation of radiographic positioning is common in this population (Holmes et al., 2018). Australian Hip Surveillance Guidelines for children with Cerebral Palsy (M Wynter et al., 2014; M. Wynter et al., 2014) recommends a standardised position for AP pelvis radiographs (supine with neutral pelvic tilt and neutral hip rotation and abduction) yet this is not always achievable. As noted in this study, the feasibility of obtaining a standardised position for

imaging is often compromised in the complex adult CP population due to contracture, cognitive and movement disorder challenges. In this study, spinal radiograph start positioning was variable with supine recorded for nine participants and sitting recorded for 13 participants. The Cobb angles from five spinal radiographs were unable to be measured at all and five were noted to be challenging to accurately measure in this cohort because of image quality issues. Positioning of participants for AP pelvis radiographs was similarly variable, decreasing validity of MP measurement on some radiographs. A valid MP measurement was unable to be obtained at all for either hip for two participants due to either significant windswept positioning or wide hip abduction positioning ('frog leg' image) for another. Error may occur in radiographs due to positioning error and / or measurement error (Schmid et al., 2016), with measurement error +/- 5% for migration percentage measurement (Schmid et al., 2016) and 4% to 8 % for Cobb angle measurement (Gstoettner et al., 2007). Positioning error is largely unknown with suggestions that it may be up to 30% (Schmid et al., 2016). Mandatory recording of variance from standard radiographic position for hip surveillance and spinal monitoring would facilitate greater accuracy in ongoing management of adults with CP as required quality and accuracy of radiographs for standardised and accurate measurement is not always possible in this population. An additional measurement tool, with established reliability (Holmes et al., 2020) such as the GIofBS is of value.

A greater understanding of the observed rotary postural elements of the spine and hip/pelvis can be gleaned with the addition of the GIofBS to appropriate radiographic studies, where feasible, adding to optimal functional management of non-ambulant adults with CP. This enables treating therapists to regularly monitor effectiveness of non-surgical interventions such as tailored sleep systems (Public Health England, 2018). There is also the potential to use the GIofBS to monitor postural asymmetry in other populations ageing with significant neuromuscular dysfunction such as muscular dystrophy or multiple sclerosis. However, measurement of asymmetry of the spine, hips and pelvis remains particularly challenging for those patients with severe cognitive or extreme movement disorders who may not tolerate either radiographs or bedside measurement using GIofBS. Further research is required in this area to explore potential for digital photography monitoring or shape capture methods such as three-dimensional laser scanning systems for fabrication of customised seating systems (Tasker et al., 2011) or Dual energy X-Ray Absorptiometry (DXA), commonly used in athletic populations (Nana et al., 2016) and in those with eating disorders (Stewart et al., 2012) to track changes in body composition. Biomedical imaging, despite its many diagnostic, prognostic and

therapeutic applications (Farahani et al., 2017) remains unexplored in the measurement of postural asymmetry.

With the recent introduction of the National Disability Insurance Agency (NDIA) (National Disability Insurance Agency, 2019) in Australia enabling funding for those with significant and permanent disability, rigour of assessment and efficacy of interventions is paramount. Clinicians who have previously had little experience managing adults with CP are now providing much needed therapeutic interventions to this population. Study findings will provide a greater understanding of the impact of postural asymmetry and clinical measurement in non-ambulant adults with CP thus guiding interventions. Clinician knowledge of lifespan care is of extreme importance, ensuring adults with CP receive the best possible healthcare outcomes.

## 7.10 Limitations

A limitation of this study was a reliance on radiographs taken within one year of the GIofBS measurement which assumes postural stability within the year, or very slow rate of change. However, given the (often) >20-year history of abnormal forces impacting on postural symmetry in the cohort, we would argue a one-year time frame was justified. The recently published NICE guidance on the management of adults with CP recognises the requirement for regular assessment including that of posture for adults with CP (Bromham et al., 2019). It has been well established that musculoskeletal complications are progressive (Tosi et al., 2009) yet the rate of change due to a dearth of longitudinal studies is unknown. Until longitudinal evidence regarding the rate of change can be established annual reviews as recommended by hip surveillance and NICE guidelines should be considered (Bromham et al., 2019; M Wynter et al., 2014). It is for these reasons that a 12 month period between GIofBS measures and radiographs was considered appropriate.

Another limitation was the relatively high number of radiographs that were unable to be accurately read. This meant that missing data were evident for 17% (spine) and 10% (hips) of the cohort which may have influenced interpretation of any relationship between radiographic and anthropometric data.

Figure 7.3 highlights that radiographs alone may not be adequate to document posture due to adults with complex disabilities and contractures being unable to achieve standardised positioning, variation in participant's position for radiographs and potentially variation of expertise and knowledge amongst radiographers in attempting to obtain standardised alignment

for valid and reliable measurements. As previously indicated, strategies for optimising documented start position in radiographs in this population may enhance future radiographic quality and interpretation.

Participants in this study were non-ambulant adults with CP (GMFCS levels IV and V), managed by a specialist multidisciplinary team, for complex medical issues and co-morbidities, and findings may not be representative for those with less severe postural asymmetry (GMFCS Levels I –III).

Effective management of posture in non-ambulant adults with CP is extremely challenging due to the combination of skeletal, muscular and soft tissue distortion over a lifetime. Further, elements contributing to complex three-dimensional postural asymmetry of the thoracic cage, spine, pelvis and hips, frequently encountered in adults with severe CP are difficult to quantify by standard two-dimensional methods such as radiographs alone.

Use of the GIofBS highlights the nature of postural asymmetry complementary to that obtained by radiographs, and may be useful in guiding interventions while ensuring relevant objectivity of clinical assessment is met for this challenging and complex group of adults. Inclusion of both radiographs and GIofBS could facilitate comprehensive clinical assessment for lifetime postural care of non-ambulant adults with CP.

## 7.11 Conclusions

This study showed there was minimal to no relationship between GIofBS measures and radiographic data for similar body areas in clinical measurement posture metrics in non-ambulant adults with CP.

## 7.12 Key Points

1. Non-ambulant adults with cerebral palsy (CP) are commonly affected by progressive secondary musculoskeletal issues which are challenging to manage.
2. The three dimensional rotary nature of postural asymmetry is difficult to quantify by radiographs alone.
3. Postural asymmetry of the chest, pelvis and windswept hips is objectively measured using Goldsmith Indices of Body Symmetry
4. Use of radiographs and Goldsmith Indices of Body Symmetry facilitates lifespan care of non-ambulant adults with CP

## 7.13 Impact Statement

Adults with cerebral palsy (CP) often experience severe and debilitating musculoskeletal problems. These problems affect the spine and rib cage, pelvis and hips negatively impacting quality of life and carer burden. The measurement of posture can be difficult especially for non-ambulant adults with CP who may already have established changes to their musculoskeletal system. This study explored the relationship between X-Rays and a clinical measurement tool (Goldsmith Indices of Body Symmetry) that physiotherapists can accurately use to measure posture. Physiotherapists can use this tool to guide relevant treatment practices such as wheelchair prescription and bed positioning.

## Disclosures

No funding was obtained for this study. There are no conflicts of interest which may be perceived to interfere with or bias this study.

## Permissions

Ethical approval was gained from St. Vincent's Hospital, Melbourne. Human Research Ethics Committee (HREC/16/SVHM/148). All participants (or next of kin) signed informed consent.

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7.14 References

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# Chapter 8: Background and Rationale

Despite CP being a non-progressive disease, the secondary musculoskeletal problems are progressive and warrant lifespan assessment and management to minimise adverse consequences and enable appropriate interventions. Effective management of posture in non-ambulant adults with CP is extremely challenging due to the combination of skeletal, muscular and soft tissue distortion over a lifetime. Further, elements contributing to complex three-dimensional postural asymmetry of the thoracic cage, spine, pelvis and hips are difficult to quantify by standard two-dimensional methods such as goniometry and radiographs alone.

The previous chapter explored the relationship between selected GIofBS constructs and radiographs of a similar skeletal area (spine and hips). All correlations between paired GIofBS variables and radiograph outcomes were small and not significant suggesting these tools are measuring two different constructs rather than duplicating measurement processes.

The GIofBS provides valuable yet complementary postural information about the thoracic cage, pelvis and hips to that obtained by radiographs. Effective evaluation is necessary to manage and monitor interventions over the lifespan of these vulnerable adults with particular emphasis on issues affecting cardiorespiratory and digestive system, seating and sleeping postures, pressure injury risk, carer burden and function. Inclusion of both radiographs (where feasible) and GIofBS would facilitate comprehensive clinical assessment for lifetime postural care of non-ambulant adults with CP.

It has been established that adults with CP experience significant postural asymmetry (13) yet the rate of progression of these asymmetries following skeletal maturity, if progression indeed occurs, is unknown. The presence of windswept hips in a Swedish cohort of children and youth up to age 20 was noted alongside potential contributing factors including lower limb contractures, pelvic obliquity and scoliosis (45, 46). The following chapter (study 4) aims to explore the stability of spinal and pelvic/hip posture, over 18 months in a case series.

This study was accepted for publication in Pediatric Physical Therapy. Carlee Holmes, Kim Brock & Prue Morgan (2020): Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study, Pediatric Physical Therapy, In Press. DOI: 10.1097/PEP.0000000000000787

# Chapter 9: Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are non-ambulant: An exploratory study

9.1 Abstract

**Purpose:** Young adults with cerebral palsy (CP) who are non-ambulant are at risk of developing or increasing musculoskeletal asymmetries affecting the rib cage, spine, pelvis and hips. This longitudinal study aimed to explore postural change using the Goldsmith Indices of Body Symmetry (GIofBS) over an 18-month period in adults with CP who are non-ambulant.

**Summary of Key Points:** All participants demonstrated considerable postural asymmetry at study onset with evidence of minimal change in some GIofBS outcome measures and fluctuations in other outcomes over 18 months.

**Statement of Conclusions:** Repeated measurement of postural alignment and range of movement using the GIofBS at regular intervals can demonstrate whether musculoskeletal asymmetries are static or changing in this population.

**Recommendations for Clinical Practice:** Physiotherapists may use the GIofBS across the lifespan to screen for deterioration in musculoskeletal status or in assessing longer-term outcomes of interventions impacting posture in this complex population.

**What this paper adds:** This study highlights the value of the Goldsmith Indices of Body Symmetry (GIofBS) as a clinical bedside tool to monitor complex postural asymmetry in those ageing with CP who may already have established postural changes. Physiotherapists can accurately use this tool to measure and monitor posture and to guide relevant treatment practices. Lifespan measurement using the GIofBS addresses continuity of care throughout transition from paediatric to adult healthcare services.

**Key words**

Cerebral palsy, clinical measurement, posture, Goldsmith Indices of Body Symmetry

## 9.2 Introduction

Cerebral palsy affects the development of movement and posture with the occurrence of frequent and progressive orthopaedic problems occurring throughout development into adulthood [1]. Associated impairments, inclusive of sensation and perception, alongside cognition, communication, hearing, vision and behaviour increase in number and severity with higher levels on the Gross Motor Function Classification System (GMFCS) [2] and also in the CP subtype spastic quadriplegia [3]

With improving survival rates of children with CP the majority are surviving into adulthood [4] necessitating lifespan care inclusive of postural care. Musculoskeletal asymmetries are common in adults with cerebral palsy (CP) who are non-ambulant with low motor function typically described as GMFCS Levels IV and V [2]. This wheelchair-user subgroup makes up around 20% of those with CP [5] exhibiting the highest risk for secondary complications and potential for shorter life expectancy. Asymmetries of thoracic cage, spine, hips and pelvis, inclusive of windswept deformity whereby one lower limb lies in hip abduction and external rotation and the opposite lower limb lies in adduction and internal rotation resulting in difficulty bringing the legs back to a midline position as the deformity progresses, cause significant and disabling consequences for the adult with CP [6] who is non-ambulant. Respiratory conditions are the most common reason for hospital admissions and mortality in adults with CP [4,7]with kyphoscoliosis listed as one of the risk factors for respiratory dysfunction especially in adults functioning at GMFCS levels IV and V [8]. Lifespan measurement of posture in severely impaired people with CP (GMFCS levels IV and V) is challenging [9]. A scoping review on postural asymmetry in adults with CP who are non-ambulant revealed a lack of clinical measurement options able to measure the three-dimensional rotary nature of asymmetries of the thoracic cage, pelvis and hips in this population [9]. The Goldsmith Indices of Body Symmetry (GIofBS) is a reliable and valid clinical measurement tool for posture in both paediatric CP populations [10] and highly complex young adults with CP who are non-ambulant[11]. This tool provides objective information on the extent of asymmetry affecting thoracic cage, pelvis and hips, which is highly relevant to seated or lying function for this wheelchair-user population. Standard measurement procedures utilizing goniometers and tape measures are two-dimensional as are radiographs, and thus unable to capture the complex three-dimensional aspects of the asymmetries. The Posture and Postural Ability Scale (PPAS) [12], was the only clinical assessment scale utilized for adults with CP identified in this scoping review, but uses an activity framework rather than postural impairments [9]. In contrast the

GIofBS utilizes an impairment framework and when coupled with appropriately chosen additional measures, inclusive of the PPAS or radiographs, provides lifespan measurement options for the debilitating postural issues experienced by many adults with CP who are non-ambulant.

There is evidence to suggest that many people with CP experience mobility decline, with those with severe disability experiencing decline during late adolescence/early adulthood [13]. Despite lifelong functional impairments, maladapted growth and altered body composition [1,4], it is unknown the rate and extent to which postural dysfunction may change over time throughout adulthood once a person with CP is skeletally mature, or whether postural dysfunction becomes relatively static.

The purpose of this exploratory study was to investigate postural change using the GIofBS over an 18 month period in young adults with CP (GMFCS levels IV and V). It was hypothesized that there would be minimal change in posture over this period for the majority of participants.

This study used a subset of 36 adults with CP to undertake secondary analysis of previously unreported longitudinal postural data. There was some overlap with a cohort of 30 participants who were recruited into the inter and intra rater reliability study [11].

## 9.3 Methods

### 9.3.1 Participants and Recruitment

Participants referred to the Young Adult Complex Disability Service (YACDS) at St Vincent's Hospital, Melbourne between February 2017 and May 2019 were invited to participate and were provided with study information. Confirmation of interest and provision of an appointment was gained via telephone by the lead researcher. Next of kin or legal guardian provided written consent for study participation if appropriate. Ethical approval was gained from St Vincent's Hospital Melbourne and Monash University Human Research Ethics Committees.

Participants with a diagnosis of CP (any subtype), motor classification of GMFCS level IV or V, age 17 years or older (thus accessing adult-based services) attending the YACDS met inclusion criteria. Participants with significant movement disorder that rendered them unable to remain still during testing, who were pregnant, or with extreme behavioural issues (e.g. biting, lashing out) that placed testers or participant at risk were excluded. Participants undergoing postural surgical procedures within the time period were also excluded.

9.4 Outcome Measures

Demographic and medical data was accessed from participant's history and included: gender, age, CP sub-type, GMFCS level, postural tendency, most recent radiographic results (Cobb angle, hip migration percentage and presence of spinal fusion) and cognitive capacity (ability to provide own consent as determined by the signatory on the hospital consent form).

Posture was recorded using the GIofBS to collect the following data: a) Chest right left ratio (chest rotation; a score of 1.0 is indicative of symmetry); b) chest depth width ratio (chest shape: values <1.0 suggest less chest depth relative to width, values >1.0 suggest greater chest depth relative to width); c) combined hip external rotation/abduction (left and right; degrees); and d) Windswept index (degree of asymmetry between left and right pelvis / lower limbs). A higher Windswept Index suggests greater postural asymmetry [9]. Prior to commencement, an optimal standard starting position was established, being crook lie with 110° knee flexion and level pelvis. If this position was unable to be achieved an alternate starting position was identified, recorded, and used as the starting position for all measurement. These components of the GIofBS were identified in a previous study to have adequate reliability in non-ambulant adults with CP [11].

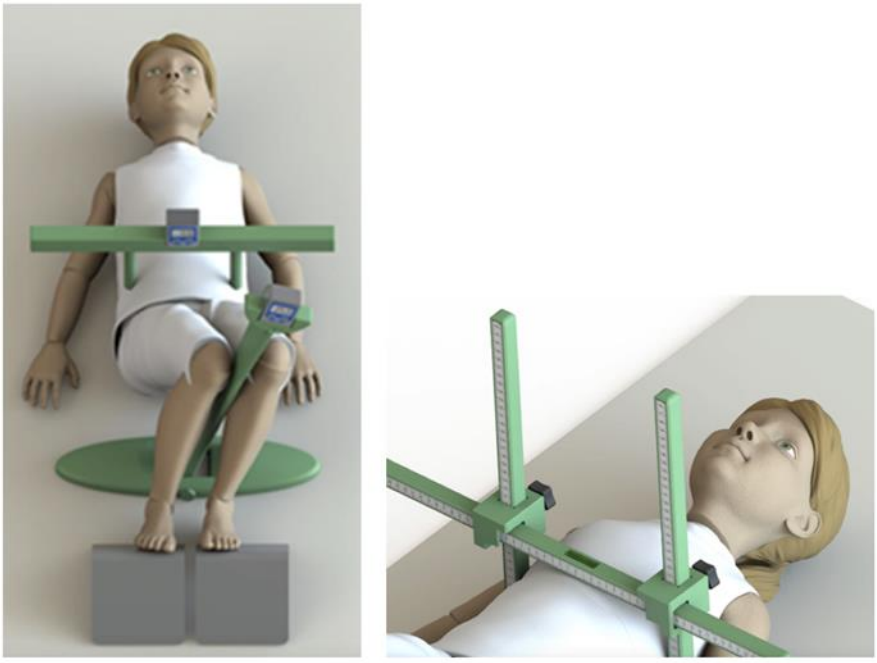

Figure 9.1. Goldsmith Indices of Body Symmetry measurement process using the Anatomical Measurement Instrument

Reproduced with permission [17].

Depiction of: a) the pelvic bridge and leg paddle with digital goniometers used to gain the hip abduction / external rotation and to calculate the windswept index, b) the chest frame used to gain chest measures used in the calculation of depth width ratio and left right ratio

9.5 Procedures

Participants attended four appointments at six monthly intervals over the 18-month period. An experienced physiotherapist trained and accredited in the use of the GIofBS (Level 3 Award in Measurement of Body Symmetry; Open College Network West Midlands, UK[14]) completed GIofBS measurements according to standard procedure [15]. Discomfort or pain was gauged through observations of reactions including spasm, withdrawal, facial or vocal reactions with input from the familiar carer standing at the head end of the participant. If participants were in obvious discomfort at any time during the assessment process, the measurement session was terminated. Forty-five minutes was allocated for each testing session.

9.6 Statistical Analysis

SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for quantitative analysis. Nonparametric descriptive statistics were employed due to the small sample size and skewed data. GIofBS chest right left ratio data was adjusted to reflect magnitude of the measure rather than a positive or negative value (indicative of clockwise or anticlockwise rotation).

Visual inspection of individual's GIofBS results was performed. Variance from control scores, derived from 48 healthy young adults without disability, for individual participants was visually depicted on line graphs using 2 SD of control data for comparison [11].

9.7 Results

Twenty-five potential participants met eligibility criteria and were invited to participate. Carers provided informed consent for all participants. Seventeen participants began the study, two did not continue after the initial appointment. Data collection was ceased after an 18-month period. A complete set of longitudinal data was gained for ten adults with CP. Of the 10 participants, seven were males, one was GMFCS Level IV and nine were GMFCS Level V, with a median age of 19, (IQR 18.00, 19.25), range 17 to 33 years. All participants had CP sub-type spastic quadriparesis. Radiographic data indicated that three participants had undergone spinal fusion. The median Cobb angle was 24° (IQR 8.5°, 37°), range 7° to 50°. Median hip migration percentage was 19% (IQR 7%, 29%), range 0% to 47%. The most common postural tendency in supine was windswept hips (n=6). The procedure, including the requirement to be positioned

in supine for measurement purposes, was well tolerated by all participants with nil participants requiring termination of the measurement process due to pain or distress.

Table 9.1. Participant and Demographic Data

| | Age (years) | Gender | GMFCS | Postural tendency in supine | Spinal fusion | Migration Percentage R/L (percentage) | Cobb Angle (degrees)* |
|---|---|---|---|---|---|---|---|
| 1 | 19 | M | 5 | Windswept to the right | no | 9/19 | 7 |
| 2 | 19 | F | 5 | Hip and knee flexion | no | 26/29 | 9 |
| 3 | 18 | M | 5 | Windswept to the right | no | 11/14 | 50 |
| 4 | 19 | M | 5 | Windswept to the right | yes | 7/31 | 24 |
| 5 | 20 | F | 5 | Windswept to the left | yes | 47/0 | 23 |
| 6 | 33 | M | 5 | Bilateral hip adduction | no | 2/12 | 8 |
| 7 | 17 | M | 5 | Windswept to the right | no | 34/45 | 44 |
| 8 | 18 | F | 5 | Bilateral hip abduction | yes | 27/19 | 34 |
| 9 | 18 | M | 4 | Hip and knee flexion | no | 20/30 | 24 |
| 10 | 19 | M | 5 | Windswept to the left | no | 0/0 | 40 |

*where more than one scoliotic curve was present, the Cobb angle of the most severe curve was recorded

Three participants were unable to achieve the optimal starting position of 110° knee flexion and a level pelvis in supine. These participants were unable to achieve a level pelvis in supine and required movement of their legs to the side to achieve a level pelvis. This leg position was noted as the alternate starting position and used for each procedure. All participants were able to achieve the required 110°of knee flexion.

The median (IQR), minimum and maximum values for hip abduction /external rotation and windswept index for CP indicated all participants were experiencing considerable postural asymmetry at the onset of the study. Hip mobility ranged from very little (1.5 °) to excessive (79 °). Minimum and maximum values for GIofBS variables indicated that participants with

CP demonstrated wide variability in chest shape and symmetry, hip mobility and windsweeping throughout the study period.

Many participants with CP fell either above or below the 2 SD derived from healthy control data [11] (Figure 9.2). This was most apparent for hip external rotation abduction with all CP participants beyond the 2 SD range of the controls for the left hip and six participants with CP for the right hip. Extreme variation from the control data was noted for the Windswept Index of 3 participants.

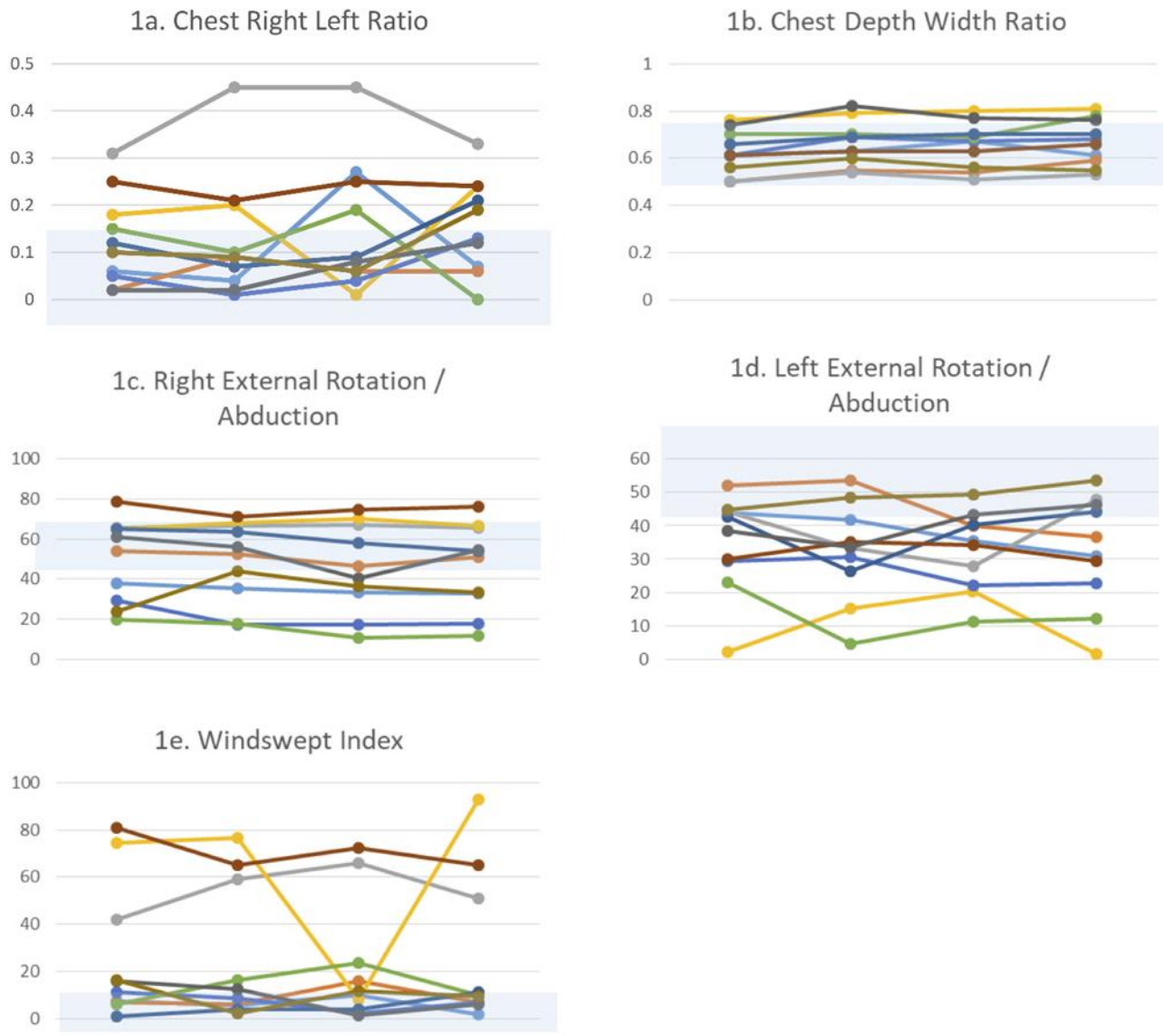


Figure 9.2 (a to e). Goldsmith Indices of Body Symmetry measures over 18months for individual participants

Figure legend

Y Axis: Goldsmith Indices of Body Symmetry results

X Axis: Time in months at 0, 6, 12 and 18 months

Participants

1 2 3 4 5 6 7 8 9 10

+/- 2 SD from control data

Visual inspection of each individual's measures over time (Figure 12.2) indicate that scores were relatively static over the 18-month period for most participants and for most GIofBS components. A trend towards reduction in hip external rotation/ abduction over time was observed for some cases (Right side, Participants 7 and 6; Left side, Participants 1, 2 and 6). between 6-monthly measurements (e.g. Participants 3, 4, 5 and 6 chest right left ratio, Participant 3 and 4 left external rotation /abduction, and Participant 2, 3 and 8 Windswept Index).

For Participant 4 a large decrease in the Windswept Index was noted at the 12-month measurement, likely attributed to a non-traumatic left femur fracture being conservatively managed in a brace coinciding with an increase in right hip and decrease in left hip mobility. Individual fluctuation in GIofBS outcomes was visually apparent for some participants

## 9.8 Discussion

This exploratory study investigated postural change across an 18-month period in a sample of young adults with CP who were non-ambulant. Results show asymmetry and postural abnormality in this cohort on initial assessment, and ongoing throughout the study period. For most participants, measurement of GIofBS components was generally stable over the study time. Some participants showed fluctuations, often with one time point being different to the general trend. Fluctuations in measurement may be related to personal factors (e.g. pain or mood) or changes in health or medication (e.g. fracture). If scores return towards baseline at subsequent measurement, they are of less concern. Repeated measures over time are necessary for an accurate lifespan picture as a measurement taken at a single point of time may not reflect the ongoing musculoskeletal status. Given the relative stability of scores over time in this population, evidence of either an upward or downward trend over subsequent measurements may suggest a more permanent change. If individuals show systematic and sustained deterioration in posture this may prompt the need for more urgent clinical review. Alternatively, if sustained improvement is noted following an intervention, compared to the trend observed prior to intervention, this may indicate the efficacy of the intervention.

## 9.9 Limitations

An 18 month time frame was selected for exploration given that this is longer than the recommended annual period for ongoing hip surveillance post skeletal maturity for those classified at GMFCS IV and V in the presence of an abnormal migration percentage, progressive scoliosis or significant pelvic obliquity [16], and hence any real change may be

apparent. Longer follow up period is advised to identify postural change progression in this population, and effects of any specific postural interventions. Minimal clinically important differences for GIofBS have not yet been established. As this sample were attending a specialist multidisciplinary healthcare facility, extrapolation of findings to those who do not have access to best practice care cannot be made. A further limitation identified was the inability to control for personal factors at the time of measurement, inclusive of mood, fatigue and injuries (e.g. fracture) which may influence results causing a one off fluctuation in outcomes. The case series findings highlight the extreme postural asymmetry young adults with cerebral palsy who are non-ambulant experience and the need for lifespan measurement and monitoring. Posture was relatively stable in this cohort, although often beyond that of a control group [11] (Figure 9.1) which may be indicative of the slow and insidious nature of progression of asymmetry. For this reason measurement across the lifespan using the GIofBS would provide longitudinal insights into the debilitating nature of asymmetries affecting the thoracic cage, pelvis and hips. Clinical measurement can be incorporated into physiotherapy assessment and value add to standard assessments for wheelchair prescription and bed positioning.

## 9.10 Conclusion

Findings from this exploratory study suggests posture is relatively static post skeletal maturity over an 18-month period in small cohort of young adults with CP who are non-ambulant. Repeated measurement of the GIofBS over time provides a more accurate picture than a single time point measurement. Observation of trends in postural outcomes over time can highlight individuals where deterioration may be occurring, and be used to assess effectiveness and sustainability of interventions in this complex population. Further longitudinal research tracking lifespan postural change is warranted.

## 9.11 What this paper adds

This study highlights the value of the Goldsmith Indices of Body Symmetry (GIofBS) as a clinical bedside tool to monitor complex postural asymmetry in those ageing with CP who may already have established postural changes. Physiotherapists can accurately use this tool to measure and monitor posture and to guide relevant treatment practices. Lifespan measurement using the GIofBS addresses continuity of care throughout transition from paediatric to adult healthcare services.

9.12 References


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# Chapter 10: Background and Rationale

There is evidence to suggest that many people with CP experience mobility decline, with those with severe disability experiencing decline during late adolescence/early adulthood (47). Despite lifelong functional impairments, maladapted growth and altered body composition (6, 22), information regarding the rate and extent to which postural dysfunction may change over an extended period throughout adulthood once a person with CP is skeletally mature, and/or whether postural dysfunction becomes relatively static is emerging.

The case series reported in the previous chapter confirmed that many non-ambulant adults with CP demonstrated considerable postural asymmetry in measurements of hip abduction /external rotation and Windswept Index. This study also found that overall posture, as measured with the GIofBS, remained relatively stable over the 18-month time frame. It remains unknown however if the posture of adults with CP who do not have access to best practice care would also remain relatively stable over 18 months. Further, it is also unknown as to any progression in postural changes in those with less severe (e.g. GMFCS Level I-III), or less complex CP.

It also remains unknown as to any relationship between postural asymmetries and pain in this population with complex, lifelong postural dysfunction (21). Pain has been reported in up to 87% of adults with CP (21) and is the most common secondary issue associated with CP (48) increasing with age from 18% in 30 year olds to 40 % in 60 year olds (22). This is a much higher incidence of pain than is reported in the adult general population where chronic pain ranges from 10 % to 30 % (49). Pain in CP may be attributed to a range of causes such as spasticity/tone, contracture, gastrointestinal problems, joint degeneration and dental causes (3, 50). We know little however about the pain experience in non-ambulant and non-communicating adults with CP, who may have lived their life with significant neuromuscular dysfunction resulting in postural asymmetry, and how they (and their carers) may manage their pain. The following chapter (study 5) will explore the relationship between pain using the Non-Communicating Adult Pain Checklist and postural data obtained from the GIofBS and radiographs in a cohort of young adults with CP. Conservative pain management strategies including medications and the use of assistive technology will be also be identified. This review was accepted for publication to Disability and Health (42). Carlee Holmes, Kim Brock & Prue Morgan (2021): Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study, Disability and Health Journal, In Press. DOI: 10.1016/j.dhjo.2021.101063

# Chapter 11: Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study

11.1 Abstract

**Background:**

Pain in adults with cerebral palsy (CP) is commonly reported, with muscular and skeletal dysfunction resulting in postural asymmetry as potential contributors to multifactorial causes of pain. The relationship between pain and postural asymmetry of the thoracic cage, pelvis and hips in non-ambulatory adults with CP however is unknown, particularly in those with cognitive and communication limitations.

**Objective:**

The primary aim of this study was to describe and quantify day and night pain in non-ambulatory adults with CP. Secondary aims were to investigate any relationship between pain and postural asymmetry and to describe current pain management strategies utilised.

**Methods**

Pain was measured using the Non-Communicating Adult Pain Checklist (NCAPC). Posture was measured using the Goldsmith Indices of Body Symmetry (GIofBS) and radiographs. Correlations between pain scores and posture (GIofBS and radiographs) were assessed using non-parametric analysis. Information regarding pain management strategies was gained from medical records and carer interviews.

**Results**

Seventeen non-ambulatory adults with CP were recruited. High levels of day pain were experienced by ≥50% of participants with a high incidence of prescribed medications targeting pain. Strong positive correlations between day and night NCAPC scores, chest right left ratio and night pain, Cobb angle and day pain and between Cobb angle and night pain were evident.

**Conclusion**

The incidence and severity of pain in non-ambulatory adults with CP is high with postural asymmetry a potential contributor. Pain remains difficult to assess and manage in adults with significant cognitive and communication impairments and warrants further investigation.

**Keywords**

Cerebral palsy, pain, posture, windswept hips, scoliosis, radiographs

## 11.2 Introduction

Cerebral Palsy (CP) is a lifelong disability with most children now surviving into adult hood (1). Pain has been reported in 62% to 87% of all adults with CP (2) with greater pain associated with greater disability and is the most common secondary condition associated with CP (3). Despite this, understanding pain in those with complex disability and significant communication and/or cognitive limitations remains challenging. A focus on complex healthcare needs, and specifically identifying and managing pain in adults with CP is not common, and may be overlooked due to communication difficulties, difficulties accessing health services and diagnostic overshadowing (4).

Pain in those with CP is likely to be multifactorial, originating from acute and/or chronic system dysfunction such as skeletal, neuromuscular, respiratory and gastrointestinal origins. Muscular and skeletal issues which may contribute to postural asymmetry (i.e. asymmetry affecting thoracic cage, pelvis and hips) are two of several potential causes of pain in adults with CP (2, 5), with high proportions of non-ambulatory adults with CP experiencing hip dislocation, fixed hip/knee contractures and profound scoliosis (6). Any relationship between postural asymmetry and pain is unknown in this population, as is the relationship between pain and postural dysfunction over a 24-hour period.

Adults with CP (GMFCS IV and V) experience significant postural asymmetry affecting the thoracic cage, pelvis and hips (6) which may be associated with pain and cause activity limitation. Measurement of postural asymmetry utilising an impairment framework is an important aspect of management which, when coupled with a clinical assessment focused at an activity level (e.g. Posture and Postural Ability Scale) (7) provides a holistic postural assessment across the lifespan.

An extensive systematic review by Novak et al (2020) revealed a lack of evidence for efficacy of interventions for management of pain in children with CP other than pharmacological agents targeting spasticity (8). High level evidence exists to support the use of Botulinum Toxin, Intrathecal Baclofen and oral medications for spasticity reduction as a pain minimisation strategy (9). The efficacy of non-pharmacological postural management interventions are less conclusive (10). Conservative interventions aimed at managing postural asymmetry and potentially minimising pain include the prescription of assistive technologies (e.g. adaptive seating and bed positioning equipment) aimed at 24-hour postural care, handling techniques and education of families and involved professionals (11). However while the evidence

regarding efficacy of these interventions in children is relatively limited (11, 12) there is almost no published literature relating specifically to conservative pain interventions for adults with CP (2) whose needs across their lifespan may well be different to those of children. As a starting point, it would be worthwhile to sample the interventions currently being utilised for pain management in non-ambulatory adults with CP attending a specialist adult transition clinic.

Engaging people with cognitive and communication limitations in research studies may be difficult (13, 14), and they may be unable to participate due to not meeting inclusion criterion. The Non-Communicating Adult Pain Checklist (NCAPC) (15) has been developed to capture pain behaviours in adults with significant intellectual and developmental disabilities (15). The scale explores both physiological and behavioural elements of pain and has satisfactory construct validity, internal consistency and sensitivity to pain alongside high reliability for this population (16) using proxy reporting. Despite well recognised limitations with proxy reporting (3, 5) for those with severe cognitive and communication difficulties, proxy reporting may be the best available option (17). The primary aim of this study was thus to describe and quantify day and night pain experienced using the NCAPC in non-ambulatory adults with CP. The secondary aims were to investigate any relationship between pain and postural asymmetry, and describe pain management strategies in this cohort. It was hypothesised that those with greater 24-hour pain would also have greater postural asymmetry.

## 11.3 Methods

### 11.3.1 Design

This study is a component of an observational cohort study of non-ambulatory young adults with CP investigating a postural measurement tool (18). The data regarding pain is previously unreported.

### 11.3.2 Participants

Participants who had a diagnosis of CP classified at the non-ambulatory categories of Gross Motor Function Classification System (GMFCS) levels IV and V (19), newly referred to the Young Adult Complex Disability Service (YACDS) during the period February 2017 to May 2019 met inclusion criteria and were invited to participate. Participants were excluded who had a significant movement disorder that rendered them unable to remain relatively still during postural testing, were pregnant, or had extreme behavioural issues that placed either testers or the participant at risk. Next of kin or a legal guardian of suitable participants were given study

information and provided written consent for all participants due to severe cognitive impairment. Ethical approval was gained from St. Vincent's Hospital Melbourne Human Research Ethics Committee (HREC/16/SVHM/148).

### 11.4 Outcome Measures

Demographic and medical data was accessed from the participant's medical history and included gender, date of birth and GMFCS level. Control data (Table 11.2) was obtained from 48 young adults with no self-declared disability (18).

### 11.5 Pain

The Non-Communicating Adult Pain Checklist (NCAPC) is an 18 item scale with each item rated from 0 – 3 referring to the frequency of the observed behaviour (0 = not at all, 1 = just a little, 2 = fairly often, 3 = very often). It describes six sub dimensions of pain behaviour: vocal reaction, emotional reaction, facial expression, body language, protective reaction, and physiological reaction (15, 16). A score of ≥10/54 is indicative of a high pain level (16). As pain may vary between night and day due to factors including seating, bed positioning, spasticity and medications, both night and day time NCAPC were completed by the carer for a typical day / night in the week prior to the research appointment. Night pain scores were recorded either to reflect pain levels when tending to participant overnight, or at initial contact with the participant first thing in the morning. Carers were not expected to report on the participant's overnight pain unless it was usual care to review or tend to the participant overnight.

### 11.6 Postural Alignment

### 11.7 Anthropometric Measurement

The Goldsmith Indices of Body Symmetry (GIofBS) is a clinical measurement tool providing objective information on posture including thoracic shape and symmetry, pelvic orientation and hip mobility with demonstrated reliability in non-ambulatory adults with CP (18). The GIofBS, according to standard testing procedure (20), was used to collect a) Chest right left ratio (indicative of chest rotation); b) combined hip external rotation/abduction (left and right); and c) the Windswept index (indicative of degree of asymmetry between left and right pelvis/lower limbs). These measurements were selected as they have been shown to vary significantly in non-ambulatory adults with CP compared to those without disability (18).

### 11.8 Radiographic Measurement

Antero-posterior (AP) radiographs of the pelvis and spine obtained within 12 months of the collection of the GIofBS data were reviewed and calculations of highest hip migration percentage (MP), a measurement of the amount of ossified femoral head not covered by the ossified acetabulum (21), Cobb angle, spinal curvature measured in degrees (22) and pelvic obliquity, an abnormal tilt of the pelvis in the frontal plane were completed. This time frame was selected as it meets published recommendations for surveillance in those post skeletal maturity in the presence of abnormal radiographic findings (23). All radiographic measurements were completed using tools within a picture archiving and communication system (Synapse™; Fujifilm Corp., Tokyo, Japan).

### 11.9 Procedure

Participants attended an appointment at which they were measured by an experienced and trained physiotherapist (Level 3 Award in Measurement of Body Symmetry; Open College Network West Midlands) using the GIofBS Anatomical Measurement Instrument according to published practice (20). Forty-five minutes was allocated for a testing session.

### 11.10 Pain management strategies

The commonly prescribed medications and conservative interventions impacting spasticity and / or pain were identified via carer interview and researcher review of medical chart (routine correspondence between YACDS and General Practitioners) at the time of measurement. Spasticity medications were included in the data extractions due to the recognised relationship between spasticity and pain (2, 24) and the difficulty in extrapolating the exact cause of pain in non-communicating adults with CP.

### 11.11 Statistical Analysis

The SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for all quantitative analysis. Descriptive statistics were used to report demographic data, NCAPC measures (day and night), and GIofBS and radiographic measures. All continuous variables were analysed for distribution and skew. The GIofBS chest right left ratio was adjusted to reflect the magnitude of the measure rather than a positive or negative value (indicative of rotation in a clockwise or anticlockwise direction). Pain management strategies were categorised and proportions counted.

Non Parametric analysis (Spearman's rho) was undertaken to assess any correlation between total day and night pain scores. The correlation between pain scores and both GIofBS

constructs and radiographs (MP, Cobb angle and pelvic obliquity) were also explored. The strength and direction of any relationships were established according to Cohen (25), where r = 0.1 to 0.29 = small, r = 0.30 to 0.49 = medium and r = 0.50 to 1.0 = large effect size. Significance was set at $p<0.05$.

11.12 Results

After screening 25 potential participants, 17 adults with CP (10 males), median age 19 (range 17 to 33 years), GMFCS Level IV/V (n = 1/16) consented to participate (via carer/guardian). Nil participants were able to self-consent. Of those who did not consent, two reported transport issues, five declined and one had significant behavioural challenges.

NCAPC total and sub scores for day and night pain are provided in Table 1. Day and night NCAPC total scores (median, IQR, outliers) are illustrated in Figure 1. Ten participants displayed high levels of pain behaviours (scores ≥10 ) (16) for day pain. Six participants displayed high levels of pain behaviours (scores ≥10 ) (16) for night pain. The NCAPC subcategory of 'body language' was rated highest for both day and night pain (median 2/6; range 0-5 day; range 0-6 night). A strong positive correlation between total day and night NCAPC scores was found (rho = 0.76, p = 0.000).

Table 11.1. NCAPC Sub-categories for day and night pain

| **NCAPC Subsets** | **NCAPC Day time Median (IQR) range** | **NCAPC Night time Median (IQR) range** |
|---|---|---|
| Vocal reaction /12 | 2 (0, 4) 0 - 10 | 0 (0, 3.5) 0 - 10 |
| Emotional reaction /6 | 1 (0, 2) 0 - 4 | 0 (0, 1.0) 0 - 4 |
| Facial expression /12 | 3 (0, 4) 0 - 10 | 1 (0, 4.5) 0 - 10 |
| Body language /6 | 2 (1, 3.5) 0 - 5 | 2 (0, 3.5) 0 - 6 |
| Protective reaction /12 | 2 (0.5, 3.5) 0 -12 | 0 (0, 2.5) 0 – 12 |
| Physiological reaction /6 | 1 (0, 2) 0 - 4 | 0 (0, 1.5) 0 - 4 |
| Total /54 | 12 (5.5, 15) 0 - 41 | 5 (1.5, 17.0 ) 0 - 43 |

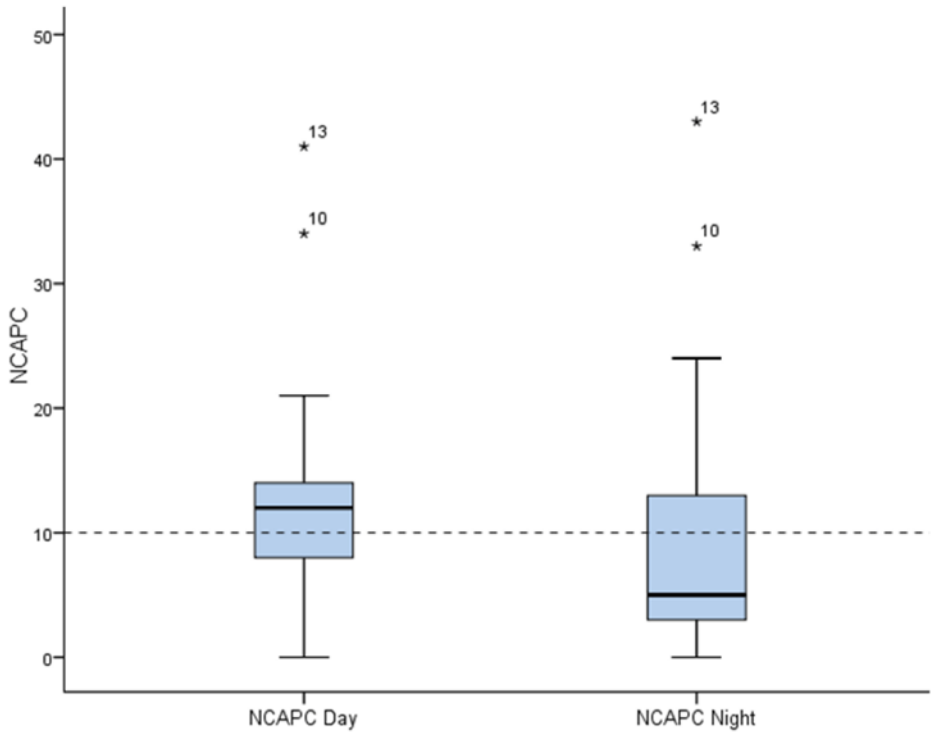


Figure 11.1. Comparison of day and night NCAPC total scores

*Note:* Box contains median (centre line) and 50% of cases (Q1 – Q3). Outliers are indicated by asterisks. The highest and lowest scores, excluding outliers, are represented by the whiskers. Dotted line indicates a value of 10, indicative of severe pain in the NCAPC (16).

GIofBS outcomes and radiographic data (median, IQR, range) are presented in table 11.2 for CP participants and healthy adult controls (18).

Table 11.2. Descriptive data for Goldsmith Indices of Body Symmetry and radiographs for participants with cerebral palsy

| **GIofBS Data** | **Median (IQR) Range (CP)** |
|---|---|
| Windswept Index | 11.25 (6.50, 32.12) 1.25- 81.00 |
| Chest Right Left Ratio | 0.08 (0.03, 0.16) 0.02 - 0.31 |
| Right Hip External Rotation/Abduction (degrees) | 44.75° (33.75°, 63.25°) 19.50° - 79.00° |
| Left Hip External Rotation/Abduction (degrees) | 38.50° (29.62°, 47.50°) 2.25° - 54.50° |
| **Radiographic Data** | **Median (IQR) Range (CP)** |
| Migration percentage[1] | 27.00% (14.00%, 37.50%) 0.00% – 47.00% |
| Cobb angle (degrees)[2] | 23.00° (10.50°, 37.00°) 7.00° - 65.00° |
| Pelvic obliquity (degrees) | 3.00° (2.00°, 11.50°) 0.00° - 21.00° |

[1] MP of ≤ 10% normal and MP ≥ 30% abnormal (23), [2] scoliosis defined as Cobb angle ≥ 10° (28)

Control data for GIofBS (Median (IQR) range): Windswept Index 2.87,(1.0, 4.69) 0.00-14.25; Chest Right Left Ratio 0.05 (0.03, 0.09) 0.00-0.27; Right Hip External Rotation/Abduction (degrees) 53.12° (49.87°, 58.94°) 35.00°-65.50°; Left Hip External Rotation/Abduction (degrees) 56.37°(51.00°; 60.44°) 41.75°-69.00°.

## 11.13 Relationship between pain and posture

GIofBS outcomes: Other than a significant correlation between chest right left ratio (chest rotation) and night pain (rho = 0.521; p=0.032), all remaining correlations between pain and GIofBS outcomes were not strong and failed to reach significance (Table 11.3).

Radiographic outcomes: Other than a significant correlation between the Cobb angle and day pain (rho = 0.611, p = 0.027), and between the Cobb angle and night pain (rho = 0.593, p = 0.033) all remaining correlations between radiographic measures and pain measures were not strong and failed to reach significance (Table 11.3).

Table 11.3. Correlations between NCAPC pain (day and night), and GIofBS or radiographic outcomes

| **GIofBS Correlations** | **rho value** | **P value** |
|---|---|---|
| Windswept Index and NCAPC Day | 0.304 | 0.236 |
| Windswept Index and NCAPC Night | 0.373 | 0.140 |
| Chest RL Ratio and NCAPC Day | 0.419 | 0.094 |
| Chest RL Ratio and NCAPC Night | 0.521 | 0.032* |
| Right Hip ER/Abd and NCAPC Day | 0.188 | 0.470 |
| Right Hip ER/Abd and NCAPC Night | 0.321 | 0.208 |
| Left Hip ER/Abd and NCAPC Day | -0.224 | 0.388 |
| Left Hip ER/Abd and NCAPC Night | -0.163 | 0.531 |
| **Radiographic Correlations** | | |
| Highest MP and NCAPC Day[1] | 0.023 | 0.933 |
| Highest MP and NCAPC Night | -0.118 | 0.665 |
| Cobb angle and NCAPC Day[2] | 0.611 | 0.027* |
| Cobb angle and NCAPC Night | 0.593 | 0.033* |
| Pelvic Obliquity and NCAPC Day[3] | 0.327 | 0.216 |
| Pelvic Obliquity and NCAPC Night | 0.135 | 0.617 |

[1]n = 16, [2]n = 13, [3]n = 16 due to poor radiograph quality

NCAPC: Non-Communicating Adult pain Checklist, RL: Right left, ER/Abd: External rotation / Abduction, MP: Migration Percentage, *significant at $p<0.05$

## 11.14 Interventions

Prescribed medications, assistive technology relating to postural management and NCAPC totals scores are presented for individual participants in table 11.4. Of the prescribed medications, 13 participants were taking medications for reducing spasticity or spasm, and five participants were prescribed gabapentin which may be recommended for spasticity / dystonia, but is not a first line treatment. Gabapentin may be prescribed as an anticonvulsant or for neuropathic pain. The reason for prescription in this study was unable to be ascertained. Of the four participants with no prescribed medications targeting pain, two had marked pain (scores ≥10) and two had minimal pain. All participants were seated in customised wheelchairs yet 12 were awaiting new or modified seating systems. Only one participant had bed positioning postural supports in place, with nine participants waiting for prescribed bed positioning equipment.

Table 11.4. Pharmacological and conservative management interventions

| Participant | Age | Baclofen[1] | Gabapentin[2] | Artane[3] | Benzodiazepines[4] | Wheelchair and seating system[5] | Night time positioning system | NCAPC Day Total / 54 | NCAPC Night Total / 54 |
|---|---|---|---|---|---|---|---|---|---|
| **1** | 19 | ✓ | ✓ | | | * | | 0 | 0 |
| **2** | 18 | ✓ | | ✓ | ✓ | * | # | 14 | 11 |
| **3** | 19 | | | | | * | | 1 | 3 |
| **4** | 18 | ✓ | ✓ | | | * | # | 10 | 4 |
| **5** | 19 | ✓ | ✓ | | | * | | 14 | 6 |
| **6** | 20 | ✓ | ✓ | | ✓ | * | # | 9 | 0 |
| **7** | 33 | ✓ | | | | * | # | 3 | 3 |
| **8** | 17 | | | | | * | # | 16 | 24 |
| **9** | 17 | ✓ | | ✓ | ✓ | * | | 13 | 0 |
| **10** | 18 | | | | ✓ | * | ✓ | 34 | 33 |
| **11** | 21 | ✓ | | | | ✓ | | 13 | 3 |
| **12** | 18 | | | | ✓ | ✓ | | 8 | 5 |
| **13** | 19 | | | | ✓ | * | # | 41 | 43 |
| **14** | 18 | ✓ | ✓ | | ✓ | * | # | 12 | 21 |
| **15** | 19 | ✓ | | | ✓ | ✓ | # | 9 | 8 |
| **16** | 19 | | | | | ✓ | | 1 | 0 |
| **17** | 28 | | | | | * | # | 21 | 13 |

Drug management: [1]Baclofen: spasticity and pain management, [2]Gabapentin: anticonvulsant and pain management, [3]Artane: spasm management, [4]Benzodiazepines: spasm management, [5]All participants used customised wheelchairs

✓Equipment utilised and meets current needs, * Equipment utilised yet insufficient for current needs,

# Equipment requirement identified but not yet provided

## 11.15 Discussion

This study explored the assessment of pain in cognitively impaired, non-ambulatory adults with CP attending a multidisciplinary specialist transition service, the relationship between pain and postural asymmetry and pain management strategies employed. Proxy reporting of pain revealed that more than half the participants experienced high levels of day pain (NCAPC ≥10) (16) as may be expected in a cohort most vulnerable to issues that contribute to pain.

Assessing pain in adults with significant physical, cognitive and communication impairments presents a challenge. The proxy reporting of pain in this study revealed that all except one participant displayed pain behaviours during the day and all except four participants demonstrated some degree of night pain. The true incidence of pain in the participants is unknown however due to the limitations of proxy reporting (3, 5), yet suitable alternatives are not available for those with severe communication and or cognitive disabilities. In the paediatric literature, under-reporting of pain in children with complex disabilities results in the potential for greater risk of pain neglect and management (26). Reliance on interpreting behavioural aspects of pain behaviour in the NCAPC such as body language (rated more highly by proxies in this study), may be more indicative of the underlying neurological pathology rather than actual pain (26), or personal experience of pain by the proxy. Although the majority of study participants displayed pain, the reporting of pain to health care practitioners in the presence of motor impairment, cognitive and communication dysfunction is not common (26) and may require a concerted effort by healthcare practitioners to investigate thoroughly.

The strong correlation between night and for day pain scores is suggestive of a consistent pain picture for participants irrespective of time and postural position (e.g. sitting versus lying down). This may suggest that pain is arising from intrinsic musculoskeletal distortion and subsequent impact on the underlying body structure and function.

The radiographic measures of Cobb angle and the GIofBS measure of Chest right left ratio both had strong correlations to pain. Postural asymmetry resulting in a progressive body shape distortion of the spine and thoracic cage will impact underlying cardiorespiratory and gastrointestinal organs. This not only impacts function, but also increases the likelihood of pain due to distortion of the organs and altered function (i.e. pain associated with constipation or bladder dysfunction) (3). With changes to the alignment of the spinal column, as measured by the Cobb angle, the somatosensory system may potentially be impacted at the level of the intervertebral foramen where the nerve roots enter and leave the spinal cord resulting in pain

or sensory changes to the affected areas. Coupled with a dysfunctional truncal musculoskeletal system, the inability to change posture and gravity may further exacerbate malalignment and pain. Participants in this study were experiencing significant and established postural asymmetry which is not uncommon in adults functioning at GMFCS levels IV and V (6), as compared to a control group without disability (Table 11.2). In this study many participants were using inadequate postural support both in wheelchairs and in bed which may have contributed to the overall high pain picture. Seating is very challenging for those with postural asymmetry of the thoracic cage and spine and it is noteworthy that 12 participants were awaiting new seating systems and thus potentially experiencing pain due to current inadequate postural support. A lack of night time postural support was also evident, potentially contributing to night pain with nine participants awaiting funding for appropriate customised bed positioning equipment. Although measures of windswept hips had only a moderate non-significant correlation to day and night pain (rho = 0.304 and 0.373 respectively), in conjunction with scoliosis and pelvic obliquity, this creates a disabling postural asymmetry (27) presenting challenges for seating and bed positioning. However, it is difficult to ascertain the specific cause of pain in this population due to the interrelated nature of the pelvis, hips and spine in conjunction with limited communication. In this small sample it is of interest that pain appeared greater during the day when participants were sitting and gravity was impacting postural alignment. Further investigation is required to identify the impact of assistive technology (seating and bed positioning) on observed pain behaviours. Repeating the NCAPC after customised postural support delivery to ascertain any impact on pain might be valuable.

The majority of participants were taking prescribed medications for spasticity or spasm, which may reduce musculoskeletal pain. The routine use of a pain measurement tool, such as the NCAPC, pre and post staged changes to medications (trial and error), may enhance pain management in adults with CP, although it is acknowledged that NCAPC does not allow the identification of pain source. Further investigation into the exact cause of pain in adults with severe and complex disabilities is required to enable lifespan planning and evaluation of efficacy of interventions

## 11.16 Limitations

In this preliminary exploratory study, the use of a participant group who were referred from a tertiary paediatric healthcare facility to a specialist adult transition clinic may not be truly representative of adults with CP being managed in the community and thus results should be

extrapolated with caution. Also, the sample size is relatively small, larger samples may reveal additional significant correlations between pain and postural measures.

A further study limitation was the inability to identify whether gabapentin was prescribed for the anticonvulsive or neuropathic pain relieving effects or both. Finally, it is acknowledged that measures of pain were sought from a 'moment in time', that is, a typical day and night and may not necessarily be reflective of a participant's chronic pain behaviours over a longer time period.

## 11.17 Conclusion

The incidence and severity of pain in non-ambulatory adults with CP is high. Postural asymmetries may be associated with pain. Pain remains difficult to assess in adults with significant cognitive and communication impairments. Further investigation into assessment and management strategies for the lifespan management of non-ambulatory adults with CP is warranted.

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# Chapter 12: Discussion and Conclusion

Cerebral Palsy (CP) is an umbrella term describing permanent and lifelong disorders affecting movement and posture that occurred in the developing fetal or infant brain (2). Despite CP being a non-progressive disease, the secondary musculoskeletal issues may increase over time and impact many domains of health and body function, limiting the ability to participate in activities of choice. These participation restrictions and risks are particularly problematic for those with low motor function (GMFCS IV and V), including limitations to the ability to be seated or lie in bed free from discomfort, and associated risks such as pain, pressure injury and respiratory compromise. Many also experience increasing disability with ageing and earlier onset of ageing (4).

The thesis explored the extent of postural asymmetry in non-ambulant adults with CP, a population who have attracted little research interest to date. The reliability of a clinical measurement tool, the Goldsmith Indices of Body Symmetry (GIofBS) was investigated followed by further exploration of this tool and its relationship to postural radiographic data. Stability of posture over an 18-month period using the GIofBS was investigated alongside an exploration of the relationship between posture and pain in non-ambulant adults with CP. A control data set of GIofBS outcomes was also established in healthy young adults.

A series of investigations were designed to accomplish this goal and were outlined in the initial concept map (Thesis structure). The existing literature was reviewed, optimal study methods developed and a series of experimental studies investigating posture, postural measurement and associated pain were conducted.

The following specific research questions relating to non-ambulant adults with CP provided the framework for the studies included in this thesis:

- What is the nature and prevalence of postural asymmetries in non-ambulant adults with CP?
    - What measurement tools for assessing and monitoring postural asymmetries are commonly used?
- Are the psychometric properties of the clinical measurement tool, GIofBS sufficient to support clinical use?
    - Do GIofBS measurements differ between adults with CP and healthy controls?
    - Are GIofBS measures reliable?
- Is there a relationship between the GIofBS (anthropometric measures of posture), and radiographs?

- Does postural asymmetry, as measured by the GIofBS, change over time?
- Is there a relationship between postural asymmetry and pain?

This chapter summarises the results of these studies and provides recommendations for future research based on noted study limitations and gaps in knowledge.

The first publication in this thesis (Chapter 3) identified the common postural asymmetries associated with windswept hips, scoliosis, pelvic obliquity, and limb contracture in non-ambulant adults with CP. It demonstrated that a large proportion of non-ambulant adults with CP have extensive postural asymmetries with hip migration percentage (MP) over 30% present in 30%, scoliosis present in 75%, pelvic obliquity present in more than 40%, hip contracture in up to 100% and knee contracture in up to 87% of this population. Variability in populations, reporting methodology, and measurement systems was evident in reviewed studies with conservative interventions infrequently and poorly described. A lack of systematic, large scale follow-up studies of children with CP into adulthood with thorough longitudinal assessments has contributed to our lack of understanding of the long-term outcomes of postural asymmetries in this population.

With a lack of standardised clinical postural measurement tools for use in non-ambulant adults with CP evident, the second publication in this thesis (Chapter 5) subsequently investigated a clinical postural measurement tool, the Goldsmith Indices of Body Symmetry (GIofBS). This tool is in use in the paediatric CP population, largely in some areas of the United Kingdom and Western Australia, and demonstrated potential value as a non-invasive clinical measurement tool that may be able to capture the complex three-dimensional characteristics of postural asymmetry in adults with complex disability. However, there was no published information regarding its use in adults (either with or without any form of disability). This study therefore investigated the intra and inter-rater reliability in the non-ambulant adult CP population. The GIofBS was found to have excellent intra-rater reliability (ICC's $\geq$ 0.97). Inter-rater reliability for GIofBS was also found to be good to excellent (ICC's $\geq$ 0.85). The range and variability of measures of postural asymmetry was greater for all participants with CP, and participants with CP generally had much higher windswept indices compared to data from a control cohort without disability. The control data was presented in Appendix A. Further, the measurement process was well tolerated by this population who demonstrate considerable complexity and medical fragility, adding to its clinical utility.

Given the predominance to date of using radiography to track change in skeletal form in those aging with CP (where this is possible to achieve), it was unknown as to whether radiographic measures of scoliosis or hip dysfunction were reflecting similar measures of postural asymmetry captured by the GIofBS. Exploration of any relationships between GIofBS outcomes and radiographs was thus investigated in Chapter 7 (Chapter 7). Minimal to no correlation between paired measures of radiographic and GIofBS data was found (all $r \leq 0.25$; all $p \geq 0.20$). Clearly the GIofBS did not yield similar information to the radiographic measures of the spine and pelvis. It also can't be utilised as a substitute measure for surveillance of hip enlocation. It is possible that the GIofBS provides information on the three-dimensional rotary nature of asymmetry of the ribcage, pelvis and hips which may not be gained from two-dimensional radiographs. The WI in conjunction with subcomponent GIofBS measures (rib cage deformity and combined hip external rotation / abduction) may facilitate clinical interpretation by the therapist to determine optimal bed and seating positioning supports to support thoracic asymmetry, enhance pelvic alignment, and hip range in addition to that obtained from two-dimensional radiographs aiding in the comprehensive understanding and management of postural asymmetry in the complex population of non-ambulant adults with CP.

Changes in postural asymmetry post skeletal maturity may have relevance to functional ability in those ageing with CP. The subsequent study (Chapter 9) provided an overview of the stability of the asymmetries of the ribcage, pelvis and hips in a case series of non-ambulant adults with CP over an 18-month period using the GIofBS to track change. As this was an exploratory study with a small sample size results must be interpreted with caution. Not surprisingly, many non-ambulant adults with CP demonstrated considerable postural asymmetry in hip abduction /external rotation and Windswept Index outcomes on initial measurement. This study found however that there was no consistent pattern in either postural stability or postural decline as measured with the GIofBS in the participants over an 18-month period. No reproducible patterns or rates of change were identified across the group suggesting that change in posture is highly individual necessitating regular surveillance. Individual point to point (six monthly) fluctuation may be attributed to personal or environmental factors in this population, such as change in pain status, spasticity, or mood. Given the lifelong neuromuscular dysfunction evident in this population, it suggests that permanent postural change may not occur rapidly, and more sustained change (observed on more than one testing occasion) in

outcomes across the lifespan may indicate true change and warrant the commencement or modification of a postural intervention.

The published evidence demonstrates that those with CP experience significant chronic pain often associated with skeletal deformity (21), and pain is frequently poorly investigated and managed in those with cognitive/communication problems (10). The relationship between pain and postural asymmetry was investigated in the final study (Chapter 11). Asymmetry of the ribcage, pelvis and hips was measured with the GIofBS and of the spine and hips with radiographs. Proxy (carer) reporting of pain was recorded using the Non-Communicating Adult Pain Checklist (NCAPC). Information on conservative management including medications related to pain management and postural assistive technology (wheelchairs and bed positioning devices) was documented with the majority of participants receiving prescribed medications targeting pain. All participants were seated in customized wheelchairs yet only one was using postural supports in bed. Aligned with previous evidence, most study participants demonstrated high levels of day and night time pain. Chronic pain behaviour across a 24-hour period was evident. More extreme scoliosis (Cobb angle) and chest rotary asymmetry (GIofBS chest right left ratio) was linked to higher pain levels. Thus extreme scoliosis and rotary asymmetry of the ribcage may be associated with chronic and severe pain in non-ambulant adults with CP but warrants further investigation with a larger sample size. In lieu of a lack of adequate alternate reporting methods, proxy reporting of pain was considered satisfactory in those with cognitive and communication deficits, in this and other studies including similar populations – particularly when provided by a family member (50).

In conclusion adults with CP functioning at GMFCS levels IV and V experience significant postural asymmetries of the ribcage, pelvis and hips affecting many domains of health and functioning (43,51). Consistent and reliable clinical measurement of posture of the ribcage, pelvis and hips and the impact of interventions has been lacking in this population with radiographic studies proving to be challenging for those with contractures and/or significant behavioural or movement disorders. The GIofBS is a valid and reliable measurement tool and coupled with ongoing appropriate radiographic studies (where possible) provides a means to monitor posture and assess the effectiveness of interventions.

### 12.1 Clinical management landscape and service delivery

The body of research described in this thesis was born out of the author's initial frustration with a lack of information to guide clinical management of adults with complex cerebral palsy.

Since that time, there is a growing awareness of the lifespan needs of those aging with CP, with recognition that most children born with CP now survive into adulthood (6). This recognition is evident in the sharp increase in published literature relevant to adults with CP, rather than paediatric-focused (52), with a noticeable increase in adult-focused publications in the decade from 2009 to 2019 (300 and 442 annual publications respectively; Figure 12.1).

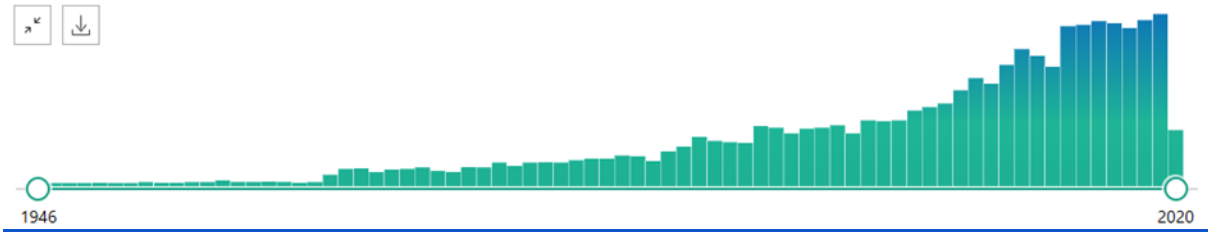


Figure 12.1. Graphical representation of growth in number of publications pertaining to adults with cerebral palsy over the period 1946 - 2020. *Note:* The Y-axis represents the number of publications, ranging from 1 in 1946 to 442 in 2019.

Source :
https://pubmed.ncbi.nlm.nih.gov/?term=adults%20cerebral%20palsy&timeline=expanded

The funding landscape in Australia has also changed significantly over the duration of this thesis with the roll out of the National Disability Insurance Scheme (NDIS) from 2013 onwards in Australia affording eligible adults with CP previously non-existent funded opportunities for allied health intervention. The NDIS is a publicly funded insurance scheme providing eligible Australians, with permanent and significant disability, access to funding for specialist disability supports inclusive of therapy support (39). Prior to 2013, children with CP were able to receive publicly funded allied health services via school-based programs and associated community therapy initiatives. However after the age of 18, these services were no longer available, and unless adults with CP had private health insurance, they had to access generic therapies for short term acute issues via community services, many of which did not accept referrals due to patient complexity. This increase in those accessing funded therapies through the NDIS has seen the creation and expansion of many private allied health therapy practices. Whereas an increase in funding and thus employment opportunities for graduating therapists is welcomed, an unintended consequence has been inexperienced therapists working with complex adults with disability, often with a lack of specialist multidisciplinary support. The percentage of NDIS participants (children and adults) with CP as a proportion of all NDIS participants has

remained relatively constant at 4 to 5% Australia-wide (39), yet a large increase in actual participants has occurred. NDIS participants with CP in 2015 was less than 1000 and this has increased to 15,635 in 2019/2020 (39). Not surprisingly, anecdotal reports from Melbourne-based neurological therapy private practices suggest a large increase in referrals for allied health services for adults with CP since the advent of NDIS funded therapy. This situation necessitates the need for therapist education in management of those with complex CP, of which postural management is a key area.

The acknowledgement of the effects of ageing across the lifespan on the person with CP has also become evident throughout the course of this thesis with a recent 5 year Australian government NHMRC-funded program of research, ('CP-Achieve'; 2020-2024), aimed at investigating effective strategies to improve the lives of young people with CP (up to the age of 30) based at the Murdoch Children's Research Institute, Melbourne (https://www.mcri.edu.au/research/centres/centre-research-excellence-cerebral-palsy). For the first time in Australia, the need for a comprehensive program of research into issues impacting young adults with CP will be acknowledged and championed.

Both funding initiatives open the landscape for greater access to clinical services, and improved clinical practice supported by a strong evidence base. This thesis by publication on postural asymmetry in non-ambulant adults with CP aligns with these initiatives and adds to the growing body of evidence pertaining to adults with CP.

In the following sections, key findings regarding postural asymmetry will be reviewed, implications for interventions to ameliorate postural asymmetry are discussed, and identification and management of pain in adults with CP who have cognitive and communication impairments will be considered. Recommendations will be made regarding management of postural asymmetry over the lifespan and support for knowledge translation for better health care for non-ambulant adults living with CP.

## 12.2 Postural asymmetry

The emergence of several themes pertaining to postural asymmetry in non-ambulant adults with CP became evident throughout the series of research studies within this thesis.

### 12.2.1 Prevalence of postural asymmetry

The high prevalence of postural asymmetries in adults with CP classified at GMFCS levels IV and V was confirmed– arising from both the scoping review (Chapter 3) and asymmetry prevalence in study participants (Chapter 5). Previous studies have highlighted the presence of

contractures of the hips and knees and radiographic measures of scoliosis, hip subluxation and pelvic obliquity in adults with CP, as documented in the scoping review (13). The distribution of postural asymmetries of the ribcage, pelvis and hips in study participants (Chapter 5, 7 and 9) further supported this data. With most children with CP now living into adulthood a greater understanding of posture, inclusive of prevention and management strategies, and the lifelong impact on function is required.

### 12.2.2 Measurement of postural asymmetry

The scoping review identified a lack of consensus amongst researchers and clinicians regarding appropriate clinical measurement of posture in adults with CP (GMFCS IV and V). Radiographs and goniometry may be implemented - with some limitations due to population complexity - however, both are two-dimensional measures which fail to adequately describe the more complex three-dimensional rotary asymmetries affecting the rib cage, pelvis and hips. The only clinical assessment scale of posture in evidence from the scoping review for use in non-ambulant adults with CP was the Posture and Postural Ability Scale. This scale measures both posture and function, inclusive of the ability to move in and out of postures. However, with regard to posture, the PPAS is limited to identifying whether postural asymmetries are present or not but does not indicate the severity (impairment level) of the asymmetries, such as degree of scoliosis or hip contracture contributing to postural (dis)ability, essential to the evaluation of postural interventions. The thesis findings support the use of the GIofBS to measure the complex three-dimensional rotary nature of postural impairments of the ribcage, pelvis and hips in adults with CP classified at GMFCS levels IV and V. It is recommended that the GIofBS, with an impairment-based focus, coupled with the PPAS (32), with an activity-based focus, would enable comprehensive clinical assessment for ongoing monitoring and treatment planning in this population utilizing the ICF framework (Figure 1.5, page 34). This would provide therapists valid and reliable measurement tools for use in non-ambulant adults with CP.

### 12.2.3 Postural measurement ‘tool box’

Given the high prevalence and progressive nature of musculoskeletal deformities there is a need for widespread adoption of a measurement tool(s) that will capture these changes and cross the lifespan between paediatric and adult health care provision. An awareness amongst clinicians in regards to both the need for measurement of posture and the use of one or more purpose-built tools is required to accurately capture the three-dimensional postural asymmetries of the rib cage, pelvis and hips experienced by many adults with CP.

The concept of a ‘tool box’ of assessments for measurement is not a new concept with manuals such as the ‘Clinical Outcome Measurement in Adult Neurological Physiotherapy’ (53) summarising a range of potentially suitable tools aligned with ICF categories for a range of acquired neurological conditions. This concept has gained popularity in recent years in complex health phenomenon with specific measurement and interventional guidelines and manuals provided for CP focusing on pain in children (54) and overall adult management (1). Despite providing an excellent overview of associated problems for adults with CP, the recently published National Institute for Health and Care Excellence (NICE) guidelines for adults with CP (1) does not adequately address the postural and pain issues experienced by adults functioning at GMFCS IV and V. This is particularly pertinent for those with cognitive and communications disorders. The pain measures in the NICE guidelines rely on the ability to self-report, thus missing 20% of people with CP with moderate to severe intellectual disability and up to 24% of people with non-verbal communication (4). Posture is listed as a factor in the NICE guidelines that may exacerbate abnormal muscle tone, with recommendations for specialist orthopaedic or musculoskeletal referral if posture is affected (1), yet there is no information provided on how to assess for changes in posture. The options to measure the complex three-dimensional rotary nature of asymmetries affecting the thoracic cage, pelvis and hips are limited (6, 13), thus leaving a gap in how the decision is made to refer to a specialist service.

A “tool box” of assessments to address the needs of adults living with CP, inclusive of the most severely affected adults is thus proposed. The inclusion of additional measures investigated and used within this thesis, the GIofBS (Chapter 5) and the NCAPC (Chapter 11) within paediatric CP guidelines (55) and the NICE guidelines (1) would address the complex postural and pain issues, across the lifespan, experienced by non-ambulant adults, many of whom present with cognitive and / or communication challenges. In addition, use of the PPAS (32) would greatly improve objectivity and rationale for treatment. Based on the ICF framework, the proposed tool box would enable a focus on high risk impairments (e.g. pain, spasticity, weakness, fatigue and pressure from sitting on bony prominences) and activities (e.g. seating, bed positioning, ability to change position and independent movement and postural control) adding to the clinical utility of the current guidelines.

A traffic light system similar to that employed in the Swedish Cerebral Palsy Follow-up Program (CPUP) which guides clinicians based on joint ranges and cut off points may also be considered for scores and ranges gained in the GIofBS (33). Three-dimensional impairment based measures of the ribcage, pelvis and hips, as obtained from the GIofBS, are not included in the CPUP adult assessment manual (56). Given the prevalence and profound disabling impact of such deformities, the inclusion of the GIofBS to enable measurement and guide interventions may be of great benefit to an assessment toolbox. In the CPUP traffic light system, "green" indicates "clear" with no deterioration noted, "yellow" indicates vigilant observation is required with either modification or initiation of treatment recommended, and "red" indicates "alert: with a need for urgent treatment" (33). Ranges of motion or cut off points are assigned to each traffic light.

Figure 12.2 provides a hypothetical example of how the traffic light system may be applied using the GIofBS Windswept Index. As with the CPUP system, traffic lights would indicate a need for a change or initiation of treatment but not dictate the type of intervention, which is left to the treating clinician's clinical judgement and expertise. Care pathways and referral indicators may guide the less experienced in intervention options. For example, a person with a consistent increase in windswept index may require one or more of: more frequent measures, change in treatment, investigation of contributing factors, or orthopaedic referral. Further research is required to establish target zones.

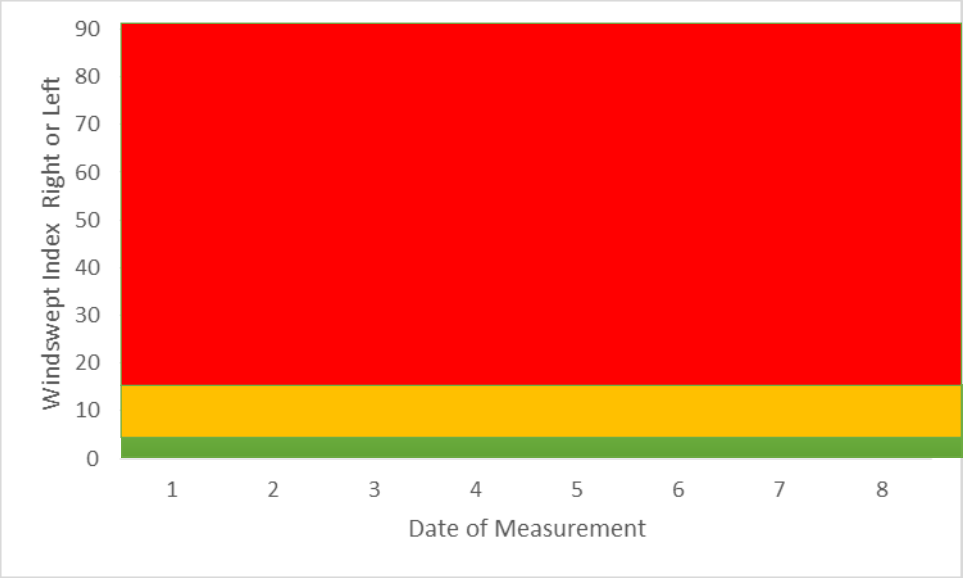


Figure 12.2. Hypothetical example of the CPUP traffic light system (51) applied to Windswept Index from the GIofBS scores obtained from adult control data using percentiles (25, 50 and 75) (41)

*Note:* ‘Red’ suggests a need for urgent treatment. Windswept Index is beyond the 100 percentile range of control data (41).

## 12.3 Implications for Interventions

Non-ambulant adults with CP often require customised seating and bed positioning systems to provide adequate postural support, maximise comfort and function and reduce the risk of pain and pressure injury as evidenced in study 5 (Chapter 11). These postural interventions are readily available, yet adequate measurement of the complex three-dimensional rotary postural asymmetries experienced by many adults with CP functioning at GMFCS levels IV and V to guide and monitor prescription of such interventions is lacking. The GIofBS, has the ability to measure these complex asymmetries affecting the ribcage, pelvis and hips providing clinicians with relevant objective postural information upon which to base postural interventions. In Figure 12.3 below, the case of a hypothetical young woman with severe CP, ‘Stella’ who is poorly managed with a consequent tragic outcome is illustrated. A number of opportunities to provide appropriate interventions to address and monitor outcomes evident from the proposed assessment ‘toolbox’ are illustrated.

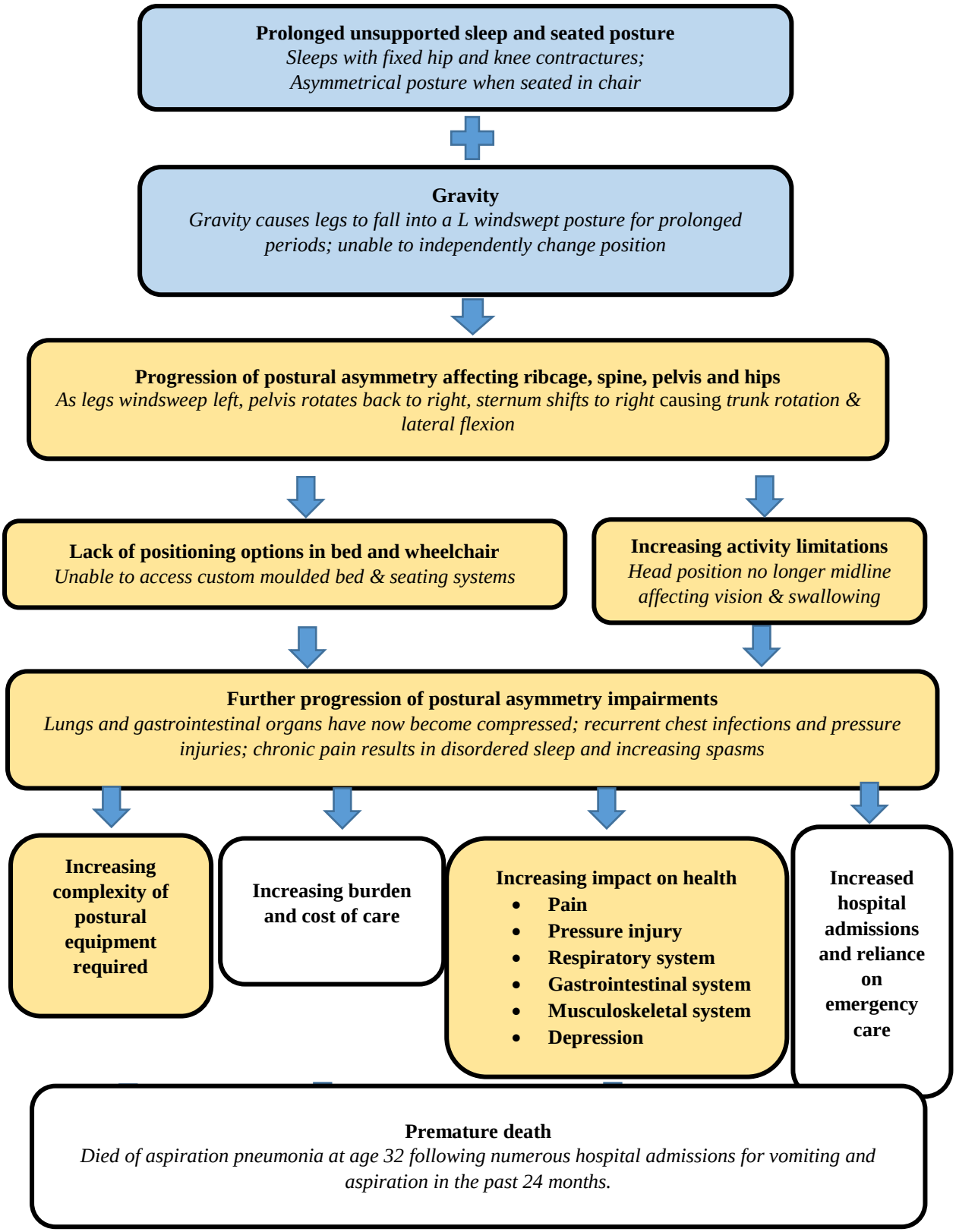


Figure 12.3. Consequences of failing to provide postural care: Case example of a young female with CP illustrating in (orange) missed opportunities for assessment and interventions to manage postural impairments or postural activity limitations. *Adapted from Simplestuffworks*

## 12.4 Pain in adults with CP and relationship with postural asymmetry

Findings from a workshop held in 2008 (21) involving the Cerebral Palsy Research Foundation, the American Academy for Cerebral Palsy and Developmental Medicine, and a consumer group, highlighted that up to 87% of adults experience pain with 33% using medications that influence pain (anti-spasticity medication or narcotics). Musculoskeletal impairments commonly impact adults with CP with associations between severity of deformity and pain (21) yet the relationship between the three-dimensional rotary postural asymmetries of the rib cage, pelvis and hips and pain remains unknown.

It is disappointing that more than 10 years later, similar findings arose from research within this thesis. The incidence of high pain scores in this research (>50% of participants), supports the 2008 workshop findings (22) in regards to a high incidence and severity of pain in adults with CP (GMFCS IV and V) yet the exact cause of the pain remains unclear which may potentially limit the understanding and effectiveness of interventions.

This study (chapter 11) confirmed that pain can be routinely assessed in adults with cognitive and communication issues using the NCAPC (proxy reported) enabling clinical evaluation of interventions including wheelchair and bed positioning equipment alongside pharmacological interventions that target pain and / or spasticity. Due to the links between postural asymmetry and pain routine and appropriate pain assessment for adults with CP should be included in all baseline assessments of adults with CP. Adults with CP functioning at GMFCS levels IV and V with communication and cognitive disorders are frequently neglected in terms of appropriate pain assessment strategies. Despite noting this difficulty for the most severely affected adults, the objective pain measures provided in the NICE guidelines for adults with CP (1) only provide examples of self-report scales (numerical rating scales, visual analogue scales, faces pain scale and body maps) all of which do not meet the needs for the adults with significant communication and cognitive issues. Thesis findings indicate a clear need for pain assessment in the complex adult with CP who is vulnerable to progressive musculoskeletal issues that have secondary consequences impacting on pain ranging from pressure injuries to gastrointestinal issues (21) issues. The NCAPC is a simple and quickly administered reliable and valid tool with noted clinical utility (in the paediatric version) for use in adults with cognitive and communication issues. Routine adoption of the NCAPC in adults with cognitive and /or communication difficulties will provide clinicians with the means to measure efficacy of interventions and justify intervention spending to funding bodies and health care systems.

In this thesis the relationship between postural asymmetries and pain were not strong and limited to a weak association between asymmetry of the ribcage (GIofBS) and scoliosis (radiograph). This may be due to a combination of a small sample size and the known limitations of proxy reporting. Many participants were also on medications targeting spasticity and / or pain thus the true relationship between pain and postural asymmetry may be underestimated by these findings. Despite this, this thesis highlights the role therapists can adopt in recognising, assessing and providing and evaluating interventions in a multidisciplinary setting, alongside pharmacological management strategies targeting spasticity and / or pain. In adopting this role, therapists will begin a much-needed conversation with carers and other clinicians in regards to pain and postural management. A multidisciplinary approach is recommended due to the high incidence of pharmacological interventions targeting pain and / or spasticity arising from study 5 (Chapter 11) and the impact of pain and / or spasticity on posture and thus positioning requirements.

## 12.5 Lifespan management

Despite CP being a non-progressive neurological disorder, the associated musculoskeletal problems are progressive. This thesis has added to our understanding of the rate and progression of postural asymmetries over time in young adults living with CP. Over an 18-month period, trunk and pelvic/hip posture in a small cohort of adults with complex CP (GMFCS IV and V) attending a tertiary multidisciplinary clinic was shown to be relatively stable, with only some fluctuation evident. However, due to the inability of participants to independently change their posture, the adoption of prolonged static asymmetrical postures poses a risk for gradual lifelong progression and secondary associated complications. It is clear that in some young adults with CP, deterioration of posture has occurred to a severe degree earlier in their life, and in others less so, but it remains unknown as to what the exact causative factors are, who may be at greater risk, and when. With lifelong measurement of posture in those with CP using the GIofBS, clinicians will gain a greater understanding of progression and be able to rapidly implement multi-disciplinary management interventions in children, adolescents and adults, with the aim to minimise or halt adverse changes. Larger cohort longitudinal studies beginning in adolescence and continuing throughout adulthood are required to gain a more comprehensive insight into progression of postural asymmetries and efficacy of interventions.

The current situation in most of Australia and internationally involves the transition of children with CP from paediatric to adult healthcare services at approximately 18 years of age. The

challenges of managing the complex adult with CP in the adult healthcare system includes a lack of specialist knowledge alongside time, access and communication barriers (51, 57). This vulnerable and often medically fragile population of adults with CP experience increased hospital admissions (9, 44), crisis care and emergency department admissions (9), and increased mortality (10). Recommendations from the NICE guidelines for adults with CP recognises the ongoing specialist healthcare needs of adults with CP, especially those functioning at GMFCS levels IV and V(1). Increasing the awareness and assessment options for postural management will improve specialist adult CP services and enable them to work alongside general practitioners to provide optimal lifespan management of this complex group of adults as they age. A lifespan model of care has the potential to reduce crisis care and hospital admissions providing cost savings to the public healthcare system and improving quality of life for adults with CP.

### 12.6 Knowledge Translation

Many non-ambulant adults with CP experience inter-related systemic dysfunction (e.g. neuromuscular, gastrointestinal, cardiorespiratory) resulting in a high degree of medical complexity and fragility. The need for prescription of complex customised assistive technology (wheelchairs and bed positioning equipment) to support these adults adds a further challenge for therapists. This thesis has focused on the progressive musculoskeletal issues affecting the rib cage, pelvis and hips and how these asymmetries can be objectively and reliably measured using the GIofBS, thus providing funding bodies with evidence, and therapists with knowledge required to initiate or change interventions.

Urgent dissemination of knowledge to those working in the field including NDIS support coordinators and workers, families, therapists and general practitioners (and other primary care providers) is required to optimally support adults with complex disability living in the community. Support coordinators, disability support workers and general practitioners are often the key people who are involved on a regular basis and who may be involved in referrals to therapists thus need to be aware of the progressive nature of postural asymmetry (including flags indicating the need for attention), speciality practices available and referral pathways to these specialists. Families and young adults living with CP require accessible knowledge to enhance informed decision making and choice.

Avenues for knowledge translation are wide and varied and must be inclusive of the usual pathways for the NDIS support coordinators and workers, families, therapists and general

medical practitioners and be appropriately targeted. Alternate options for publicly available education suited to a wider audience should also be considered such as freely accessible podcasts or CP/disability organisation newsletters. Accessible evidence will improve clinician knowledge and confidence to manage non-ambulant adults with CP.

## 12.7 Thesis Limitations

### 12.7.1 Ethical Considerations

Research involving participants with complex cognition and communication problems and experiencing significant medical fragility requires careful ethical consideration, specifically issues pertaining to consent. No participants in the series of studies described in this thesis were able to consent for themselves. Complex patients (and their families) managed by tertiary specialist facilities frequently develop close relationships with expert clinicians providing care over an extended period of time. Conflict of interest between researchers/ clinicians seeking an expression of interest to participate was managed by clearly stating there would be no impact on clinical care. Many research trials selectively recruit people with CP who are cognitively intact and hence less likely to be GMFCS level IV and V. The inclusion of this population was essential to this research. It was also clearly acknowledged to families that we recognised that they may have many competing appointments and commitments, or must travel several hours to attend the clinic, impacting on their capacity to be involved over an extended period. Finally, it was stated that all patients of YACDS who met the inclusion criteria were invited, to ensure we were not seen to be selective in who was or was not included in the research.

### 12.7.2 Participant bias

Recruitment from a specialised metropolitan tertiary care hospital with a state-wide catchment is likely to skew recruitment to more complex participants, as those with less complex needs may be accessing services (if services are required) in generic primary care services within their local metropolitan and regional areas. There are limited specialist centres for management of young adults with complex CP in Melbourne, Australia. All participants within the series of studies reported in this thesis were recruited from a single large metropolitan transition service, Young Adult Complex Disability Service, St. Vincent's Hospital, Melbourne. They were undergoing assessment and treatment by a specialist multidisciplinary team involving the disciplines of physiotherapy, occupational therapy, speech pathology, social work, dietetics, prosthetics and orthotics and rehabilitation medicine with ready access to referral for additional

specialist services such as gastroenterology and epilepsy clinic as required. The YACDS, a transition service between paediatric and adult healthcare, has a cut off age of 40 years (compared to the more typical 25 years). The higher cut off age for inclusion into the YACDS may similarly skew results towards those exhibiting greater postural asymmetry.

Given the specialist care available to the participants in these studies, adults with CP with similar disability profiles who have not received expert care may present with more severe postural asymmetries and pain issues than those observed in this study. Due to the difference in service provision within Australia (e.g. community and rural settings) and internationally, inclusive of under resourced countries, the results from this research may impact generalisability. Despite this potential limitation, this thesis provides valuable evidence contributing to the evidence base regarding adults with CP.

### 12.7.3 Recruitment Issues

As often occurs in pragmatic clinical studies, participants were not able to be recruited simultaneously, thus time taken to recruit participants meeting the inclusion criteria was a limitation that reduced participant numbers. Recruitment from a non-homogenous population in which there was variability in diagnosis of patients referred to the service (e.g. Retts Syndrome, Hereditary Spastic Paraplegia), GMFCS level, and behaviour challenges limited the availability of participants meeting the inclusion criteria. Of the 166 potential participants screened for inclusion in any of the thesis studies, 136 were excluded due to not meeting the inclusion criteria and a further 38 declined to participate.

Other issues related to recruitment included gaining consent for those living in community residential units or under guardianship orders, competing healthcare appointments, cancellations and rescheduling due to health issues. Flexibility in timing and rescheduling helped to overcome some of these recruitment issues, including scheduling alongside other healthcare appointments to reduce time and costs for families needing to take time off work and engaging support staff early in the process.

Many potentially suitable participants were familiar with participation in studies through paediatric services and, where possible, were willing to be involved. As one parent noted, “being involved in research is the one thing that he can do”.

## 12.8 Confidence in study findings

The series of studies included in this thesis were necessarily exploratory in nature. There were few comparative studies that include adults with such complex CP to guide methodology.

Calculation of statistical power required for recruitment was not typically undertaken due to lack of knowledge about the GIofBS scores in this population. The strength of the study findings therefore are presented as preliminary only. Confidence intervals for most findings, would be typically wide as depicted in the significant results (Table 12.1), of correlations between posture and pain.

Table 12.1. Correlations between NCAPC (day and night), and GIofBS or radiographic outcomes

| **GIofBS Correlations** | **rho value** | **P value** | **0.95%CI Lower limit** | **0.95%CI Upper limit** |
|---|---|---|---|---|
| Chest RL Ratio and NCAPC Night | 0.521 | 0.032 | 0.054 | 0.801 |
| Cobb angle and NCAPC Night | 0.593 | 0.033 | 0.063 | 0.862 |
| Cobb angle and NCAPC Day | 0.611 | 0.027 | 0.091 | 0.869 |

GIofBS: Goldsmith Indices of Body Symmetry, CI: Confidence Interval, Chest RL Ratio: Chest Right Left Ratio, NCAPC: Non-Communicating Adult Pain Checklist

## 12.9 Comprehensive multidisciplinary assessment

This thesis focused on the assessment of impairments contributing to postural asymmetry, using the GIofBS. It should be noted that this is only one aspect of assessment that is performed in a comprehensive multi-disciplinary patient appointment, for adults attending specialist clinics such as the YACDS, and consideration of the patient's presenting issues across a broad range of activities and participation are typically included. GIofBS adds to our understanding of contributing factors to postural asymmetry, alongside measures of peripheral joint range of motion, spasticity, pain, postural control and ability to change position, alongside client centred goals and functional capacity.

## 12.10 Future Research Directions

Postural asymmetries experienced by adults with CP functioning at GMFCS levels IV and V should not be considered as "par for the course". This thesis has added to the growing knowledge base on adults with CP and in particular, postural asymmetry and measurement utilizing the GIofBS. With emerging interest and funding for research into adults with CP, opportunities to continue with this important work into postural asymmetry in non-ambulant adults with CP are becoming available. Funding and research interest previously limited to studies related to children with disabilities is gradually increasing in the adult CP population as evidenced through NDIS funding, the government-funded Australian Centre for Health,

Independence, Economic Participation and Value Enhanced Care for adolescents and young adults with cerebral palsy ("CP-Achieve") 2020, and the large increase in evidence-based practice and publications identified earlier.

Future directions may include:

- Validation of the GIofBS as a measure of postural asymmetry.
    - Investigation of the accuracy of the GIofBS for measuring three dimensional postural alignments against the gold standard, Computed Tomography (CT) scan would enable improved validation. For thoracic cage distortion, GIofBS measures could be compared with CT scan using the methods developed by Horimoto etal 2012 (58). For validation of the Windswept Index, comparison with the methods used by Persson-Bunke etal 2006 (45) and Porter etal 2007 (7) could be explored.
- Explore the relationship between GIofBS measures and spasticity, including management of spasticity
- Further explore the relationships between GIofBS measures and pain, with consideration of pharmacological management of pain.
- Expansion of current care guidelines to incorporate measurement of postural impairments targeted at adults with CP (GMFCS IV and V)
    - Current guidelines recognise the negative impact of posture on an adult living with CP yet there is a lack of recognised measurement tools that adequately measure the complex three-dimensional rotary nature of asymmetry affecting the thoracic cage, pelvis and hips. The GIofBS addresses these issues and would be an important addition to all current guidelines.
- Identify GIofBS cut off scores for severity of postural asymmetry to prompt review/action with use of a traffic light system
    - Provision of a simple system to alert clinicians to the needs for action (monitor versus change intervention) in regards to severity of postural asymmetry may assist in the halting of debilitating progression of postural asymmetry.
- Development of a 'postural care tool box'
    - Due to the complexity of postural care of adults with CP and the limitations in the adult healthcare system (expertise, time and access) a toolbox, similar to the Australian Hip Surveillance Guidelines for Children with CP (23), to guide and

educate clinicians on available options for lifespan postural management may assist in achieving appropriate assessment and guide interventions.

- Larger cohort longitudinal lifespan studies beginning in childhood to gain better insight into progression of postural asymmetries and efficacy of interventions.
    - This thesis has highlighted that adults with CP (GMFCS IV and V) experience significant postural asymmetry affecting the ribcage cage, pelvis and hips yet it is unknown at what stage of development these asymmetries began and the rate of progression. It is also unknown which measurement activities (radiological measures or the three-dimensional assessment using the GIofBS) are more strongly associated with poorer long-term outcomes. A greater understanding, and identification of risk factors, will enable early intervention with the aim of reducing the severity of postural asymmetry and rate of progression.

- Development of a national/international database
    - This thesis highlighted the difficulty in gaining sufficient participants numbers to provide information other than preliminary exploratory data. A database of standardised assessments would allow for larger cohort studies from which stronger findings could be gained. Expansion of the database to other healthcare services nationally and internationally would enable pooling of standardised measurement outcomes and facilitate research into the lifespan care needs of adults with CP functioning at GMFCS levels IV and V

The clinical and research implications for lifespan management of non-ambulant adults with CP are summarised in table 12.2.

Table 12.2. Summary of clinical and research implications for non-ambulant adults with CP

| Theme | Clinical Implication | Research Implication |
|---|---|---|
| Many adults with CP (GMFCS IV and V) experience postural asymmetry | Clinicians need greater understanding of postural care with respect to:<br>the implications on function, quality of life and carer burden, the rate of progression, and potential assessment strategies and interventions | Translation of research findings required to increase use of the GIofBS.<br>Larger cohort studies across the health and disability sector needed to understand the impact of postural asymmetry on all adults with CP (GMFCS levels I – V)<br>Research trials required to evaluate interventions to address postural asymmetry. |
| A lack of standardised clinical postural measurement tools | The GIofBS provides reliable and relevant three-dimensional clinical postural information (impairment-focus) suited for use in those with complex CP<br>Radiographs provide complementary information to that obtained with clinical measurement (GIofBS).<br><br>A lifespan approach is required. | Integration of research findings into wider clinical practice.<br>Creation and evaluation of a postural measurement toolbox to inform clinicians of reliable and valid tools to use in this population. |
| There is a relationship between postural asymmetry and pain | Pain is highly prevalent in adults with CP (GMFCS IV and V)<br>Pain remains difficult to measure in those with cognitive/ communication difficulties<br>Pain may be related to postural asymmetry.<br>Clinicians should ensure they assess pain in all people with CP, including those with communication problems.<br>Greater education of families and clinicians into the recognition and management of pain. | Increased recognition into pain for adults with cognitive and communications difficulties is required with larger cohort studies established investigating the use of the NCAPC for adults with cognitive and or communication disorders.<br>Further investigation into the relationship between pain and posture is required in large scale observational studies. |

| | | |
|---|---|---|
| Postural asymmetry is progressive but change may be slow and insidious. | The GIofBS is able to detect change in posture over time across the lifespan.<br>Ongoing and regular measurement is required to detect trends in change.<br>Ongoing lifespan measurement and intervention for postural asymmetry is recommended as standard care practice for adults with CP.<br><br>Postural measurement should be included pre-adulthood for all individuals with CP, including those who have not yet developed fixed postures.<br><br>Education of families and clinicians in lifespan need for postural vigilance is required. | Development of care guidelines and recommendations for appropriate measurement tools for all adults with CP inclusive of postural and pain assessment and management. |
| Contribution to clinical education and knowledge base | Clinician education regarding use of GIofBS in lifespan care.<br>Adoption of the *toolbox* to guide clinicians in appropriate measurement of posture and pain.<br>Promote adult-specific literature guiding clinicians in assessment and management of posture in adults with CP.<br>Support the use of tertiary specialist clinics to provide patient review and guidance to community clinicians and lead clinical education. | Ongoing collection of GIofBS data to establish cut off scores in the 'traffic light' system. |

## 12.11 Conclusion

This thesis investigated the complex three-dimensional rotary nature of postural asymmetry affecting the ribcage, pelvis and hips in adults with CP classified as functioning at GMFCS levels IV and V. With many clinicians now working with this group of adults in community and private settings, these findings will raise awareness of the postural issues, provide knowledge on clinical measurement and introduce potential intervention concepts.

The key findings from these studies are:

1. A scoping review explored the extent of postural asymmetry in adults with CP (GMFCS IV and V), measurement techniques and conservative interventions identifying that:
    a. Adults with CP experienced considerable postural asymmetry
    b. Variability existed in reporting methods and measurement systems
    c. There was a lack of information regarding conservative postural interventions
2. A reliability study of a clinical measurement tool, the Goldsmith Indices of Body Symmetry (GIofBS), indicated that:
    a. The GIofBS was a reliable clinical tool for measurement of asymmetries of the ribcage, pelvis and hips in adults with CP (GMFCS IV and V)
    b. The provision of a control data set allowed for valuable comparative information and indicated that the control group had less variability and a smaller range of GIofBS outcomes.
3. An investigation of the relationship between radiographs and GIofBS constructs of similar anatomical regions indicated:
    a. The GIofBS and radiographs measured two different constructs
    b. Where possible, the inclusion of both GIofBS and radiographs provided useful postural information for monitoring impairments and management of adults with CP (GMFCS IV and V).
4. A case series investigating the stability of posture over 18-months indicated that:
    a. Despite some fluctuations between time points, posture was relatively stable over an 18-month period in a cohort of adults with CP (GMFCS IV and V) who were actively engaged in a specialist transition service.
5. An investigation of the relationship between posture (GIofBS and radiographs) and pain using a proxy reported scale (NCAPC) indicated that:
    a. Adults with CP (GMFCS IV and V) experienced high levels of pain

b. Assessment of pain remained challenging in adults with cognitive and / or communication difficulties yet a strong correlation existed between pain and measures relating to the spine and thoracic cage.

The changing funding landscape that the NDIS has brought to Australia is enabling adults with CP to access therapy services that have previously been unavailable to them. With appropriate assessment and intervention the destructive cycle of a lack of provision of postural care may be avoided (Figure 12.3). Findings from this thesis equips health professionals with the knowledge and tools to manage the challenging postural asymmetry evident in this complex population, non-ambulant adults with CP.

In summary, many adults with CP functioning at GMFCS levels IV and V experience considerable and debilitating postural asymmetry. Specific measurement of the three-dimensional aspects of postural asymmetry impairments affecting the ribcage, pelvis and hips is lacking but can be captured with the GIofBS in clinical practice. Physiotherapists with their expertise in anatomy, measurement, function and clinical reasoning are well placed to lead the measurement process and liaise with other involved clinicians, families and support workers regarding results and potential interventions. This is an exciting opportunity to make significant impacts on the lives of those living with CP.

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# Appendix A: Control Data

## Background

The Goldsmith Indices of Body Symmetry (GIofBS) is a simple and relatively inexpensive clinical measurement tool providing objective information on the extent of postural asymmetry such as chest, pelvic and hip deformities. It was developed in 1992 (10) and has been predominantly used with the paediatric cerebral palsy (CP) population in the United Kingdom. A recent study (1) has found that the GIofBS enables reliable quantification of the extent of windsweeping, or asymmetrical posture that is influenced by the angle between the legs and pelvis, and the combined influence of hip rotation and abduction/adduction on posture. In addition the GIofBS also provides information on chest asymmetry and shape. The GIofBS is thus a reliable measurement tool to measure and monitor postural asymmetry in non-ambulant people with CP (1). However, comparative control data is not available. Thoracic cage dimensions have been reported for different ages, gender and at various vertebral levels using the Haller Index (2, 3). The Haller index is a ratio of thoracic width and height, measured from an axial CT image and used to describe the internal dimensions of the thoracic cage. As the internal dimensions may differ significantly from the external dimensions these values cannot be used as comparative data for the GIofBS. The primary aim of the study was to establish comparative control data for the GIofBS to aid in the interpretation of postural data collected from non-ambulant adults with CP.

Clinical decision making regarding treatment efficacy and disease progression is aided by comparison with age-matched healthy subjects (4). Numeric health outcome measures enable statistical analysis that are not only useful in clinical decision making but also research and policy making (5). In the development of clinical measurement tools or use of a tool on a different population, as is the case with the proposal to utilise the GIofBS in adults with CP, it is essential to ensure any data collected is meaningful and relevant to the population (6). To date, there is no published evidence regarding normative data gained from the GIofBS in an adult population. This makes it difficult therefore to interpret measures of the GIofBS in clinical populations such as non-ambulant adults with CP. We do not know what measures of the GIofBS suggest problematic postural pathology, and what measures of the GIofBS could be considered asymptomatic and within normal variability.

## Aims

The specific aims of this study were to establish comparative control values for the GIofBS in a sample of young adults without self-identified disability.

## Method

### *Research Design*

Observational case control study

### *Participants*

Young adults aged between 18 and 40 who self-declared no movement disorder and were not pregnant were invited to participate.

### *Recruitment*

Advertisements (flyers) were placed in the Monash University Physiotherapy Building, Peninsula Campus and on the Physiotherapy notice board at St. Vincent's Hospital Melbourne. In addition, electronic invitations were distributed on the online learning sites of the Monash Bachelor of Physiotherapy units and via email to the physiotherapy department at St. Vincent's Hospital Melbourne. Interested participants were invited to contact researchers and were provided with the explanatory statement and informed consent for completion. A mutually convenient appointment was scheduled for data collection.

### *Outcome measures*

Data was collected using the GIofBS anatomical measurement instrument (Figure A.1) and followed the published guidelines for application of the GIofBS (7), excluding the use of a third person typically required for the support and wellbeing of a disabled person. The measures collected using the GIofBS are shown in Table A.1.

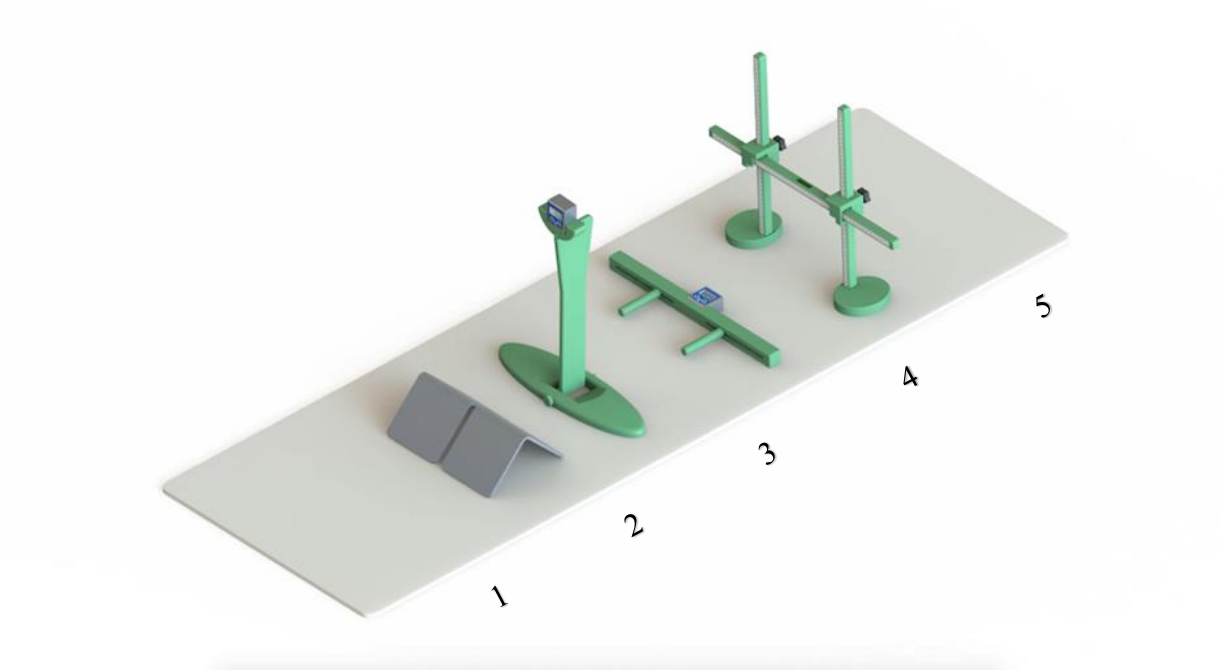


Figure A.1. Anatomical Measurement Instrument

*Note:* Equipment listed from left to right: 1 Non slip mat. 2 Foot brackets. 3 Leg paddle with level box angle sensor. 4 pelvic bridge with level box angle sensor. 5 Chest frame (8)

Table A.1. Outcome measures obtained from the GIofBS

| | **Outcome Measure** | **Procedure** | **Instrument** | Interpretation of results |
|---|---|---|---|---|
| **1** | **Chest Depth Width Ratio** | Chest depth value divided by total chest width value (chest depth/R side chest + L side chest) | Chest frame | Values <1.0 suggest less chest depth relative to width; values >1.0 suggest greater chest depth relative to width |
| **2** | **Chest Right Left Ratio** | Right chest value divided by left chest value (Right side chest / Left side chest) | Chest frame | A score of 1 is indicative of symmetry. A score of 0 to <1 is indicative of chest anticlockwise rotation. A score of >1 is indicative of chest clockwise rotation. |
| **3** | **Angle Between Legs and Pelvis (ABLAP)** | Angle of legs minus angle of pelvis<br>R ABLAP = R leg angle – R pelvic angle<br>L ABLAP = L leg angle – L pelvic angle | Leg paddle and pelvic bridge | The side (right or left) which has the higher value is the side to which the person is windswept towards |
| **4** | **Hip External rotation / Abduction** | Hip joint range of motion | Leg paddle and pelvic bridge | R≠L suggests asymmetry |
| **5** | **Windswept Index** | Difference between the left and right ABLAP<br>WI = R ABLAP – L ABLAP | calculation | Higher values represent greater postural asymmetry |

In addition to data collection from the GIofBS, age, weight, height, gender and any self-reported existing injuries or postural conditions were documented.

Statistical Analysis

The SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for all quantitative analysis. Chest right/left ratio values were transformed to reflect the magnitude of the rotation rather than a clockwise or anticlockwise direction. Normality of all data was evaluated using visual inspection and skewness and kurtosis statistics, and descriptive statistics used to report all GIofBS measures. Data that did not follow a normal distribution was transformed using the square root model (9) to enable further parametric data analysis. Spearman (non parametric) product-moment correlation co efficient was used to determine relationships between age and the GIofBS measures where non normal distribution of data was identified. The strength of any relationship was described as ranging between r value of -1 and 1, with a correlation of 0 indicating no relationship, and a correlation of 1 or -1 indicating a perfect relationship in either a positive or negative direction (10). The following guidelines regarding the (absolute) strength of the correlation were applied: r = .10 to .29 indicative of a small relationship, r = .30 to .49 indicative of a medium relationship and r = .50 to 1.0 indicative of a large relationship between variables (11).

Significant correlations were identified as $p<0.05$.

Results

Institutional ethics approval was sought and received for this study (MUHREC 2016-1458-1343).

Forty–eight young adults (29 females), participated with a median age of 21.50 years (Table A.2). The cohort was generally healthy with only four participants reporting a minor injury at the time of data collection. Reported injuries included hip pain from sports injuries (2 right hip, 1 left hip) and lower back pain which was currently non-problematic. In addition, one participant reported a leg length difference, one reported bilateral hip dysplasia and one reported a mild scoliosis (self-reported at 4 degrees). Of note the participant with hip dysplasia competed at high level sports, suggesting no functional limitation. Demographic data of participants and GIofBS results are documented in table A.2 and table A.3 respectively.

Table A.2. Demographic data from all participants

| **Participant Demographics** | **Participants n=48** |
|---|---|
| **Gender (n)** | M - 19 F - 29 |
| **Age years, median (IQR); range** | 21.50 (20.00, 26.00); 18.00 – 34.00 |
| **Height cm, median (IQR); range** | 171.50 (165.25, 178.75); 151.00 – 192.00 |
| **Weight kg, median (IQR); range** | 68.50 (60.25, 73.75 ); 51.00 – 100.00 |

Table A.3. Goldsmith Indices of Body Symmetry data from all participants

| **GIofBS Procedures** | **Mean (SD)**<br>Range |
|---|---|
| **Chest Depth Width Ratio** | **0.62 (0.05)**<br>0.52 – 0.75 |
| **Chest Right Left Ratio (Magnitude)** | **0.07 (0.05)**<br>0.00 – 0.27<br>Median (IQR)<br>0.05 (0.03, 0.09) |
| **R ABLAP[1]** | **122.11(7.96)**<br>100.00 – 135.25 |
| **L ABLAP[1]** | **122.04 ( 8.89)**<br>96.75 -139.75 |
| **R ER/ Abd[2]** | **53.61° (6.19°)**<br>35.00° – 65.50° |
| **L ER/ Abd[2]** | **55.79° (6.84°)**<br>41.75° – 69.00° |
| **Windswept Index** | **3.59 (3.21)**<br>0.00 – 14.25<br>Median (IQR)<br>2.87 (1, 4.69) |

[1]ABLAP, Angle Between the Legs and Pelvis; [2]ER /Abd, External Rotation / Abduction

The spread of Windswept index scores for the cohort are illustrated in Figure A.2, suggesting a low median of 2.87 with 50 % of the values falling between 1.00 and 4.69. The lowest Windswept index value was 0.00 (indicating perfect symmetry) and the highest was 10.0 excluding the three outliers. All three outliers were male aged 30, 22 and 21 years respectively, and did not self-report any prior postural abnormality or injuries.

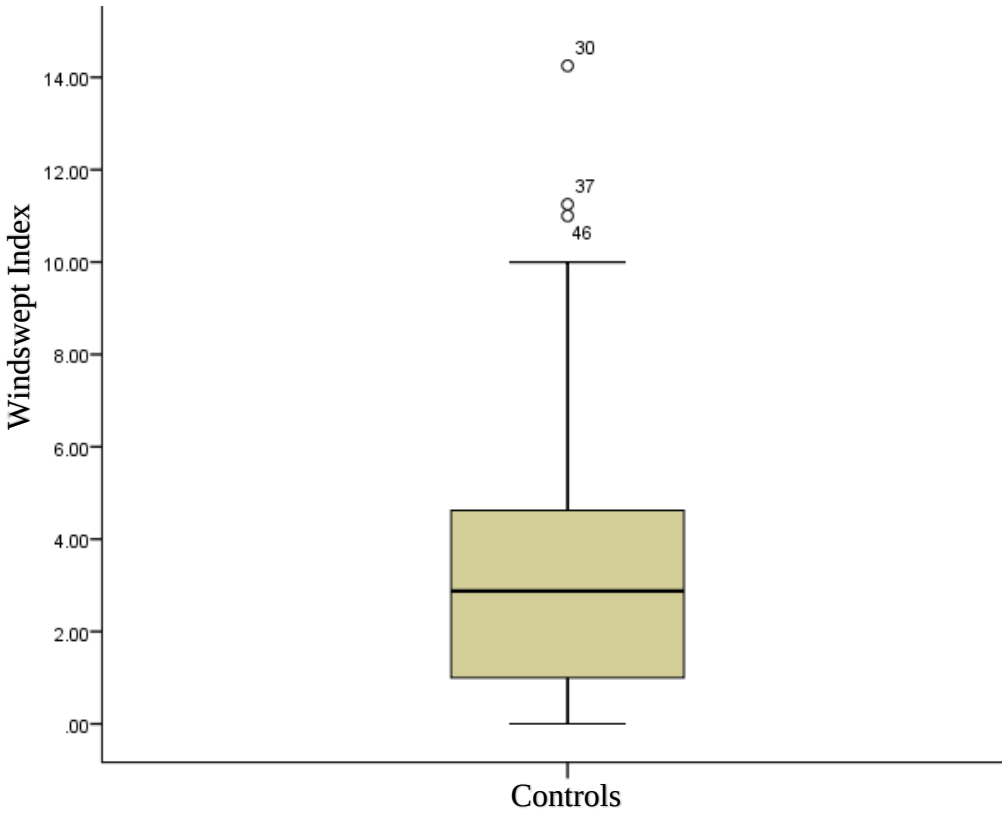


Figure A.2. Windswept Index

*Note:* Windswept Index. The upper and lower margins of the box plot represent the interquartile range (Q3 – Q1) demarcating the 25$^{th}$ and 75$^{th}$ percentiles and contains 50 per cent of the cases. The centre line sits at the median score (50$^{th}$ percentile). The outer bars or whiskers represent the range of scores at each end of the distribution with circles indicating outliers beyond 3 standard deviations from the mean.

*Correlation*

There was a small negative significant correlation between age and the chest depth width ratio (r=-0.3, p=0.038). All remaining correlations between age and GIofBS outcomes failed to reach significance (Table A.4).

Table A.4. Correlations between age and the Goldsmith Indices of Body Symmetry

| **GIofBS n = 48** | **r value** | **p value** |
|---|---|---|
| Chest right left ratio | -0.005 | 0.975 |
| Chest depth width ratio | -0.300 | 0.038* |
| Windswept Index | 0.121 | 0.414 |
| Right external rotation / abduction | -0.039 | 0.793 |
| Left external rotation / abduction | -0.167 | 0.255 |

*significant at $p<0.05$ level

Discussion

This study described control data for the GIofBS in a cohort of healthy adults with no movement disorders. Not surprisingly, the GIofBS median scores are close to those indicative of symmetry for the chest right left ratio, there is minor difference between left and right hip abduction/external rotation ranges, and the Windswept Index is relatively low, suggesting some minor variation in healthy subjects.

The depth width ratio is a measure of the chest shape. A score between 0.65 and 0.85 has previously been reported by the GIofBS developers as being within normal limits with scores towards the lower end of the range indicative of a flattened chest shape in the anterior-posterior direction (12). However, this data reportedly was taken from standard anthropometric data in a text book for the UK population (personal communication). The chest shape scores obtained from this study had a similar spread, yet were typically lower (0.52 to 0.75). This may be due to greater chest width in this population as a result of more muscle mass in a young, fit and active cohort. In particular the latissimus dorsi was noted to be well developed in many of the cohort, significantly adding to the measure of chest width. A study by Laurin and colleagues (13) used a pelvic calliper to gain anterior-posterior rib cage diameters (13). The median depth-width ratio result obtained in this study is very similar to that obtained by Laurin (13) who included hospital employees, medical students and medical residents, potentially a similar demographic to this study as opposed to the unknown participants included in Goldsmiths'

study (7). The contemporary data gathered in this study may thus be used to compare against adults with CP who may have a different chest shape as a result of developmental disability.

Scoliosis presents with a rotation of the thoracic cage and the development of a rib hump (14) and is typically measured using the Cobb technique (15). Due to the three dimensional nature of vertebral rotation the Cobb Angle alone cannot be used to evaluate the degree of vertebral rotation (16). The right left ratio which is indicative of chest rotation in the GIofBS may more accurately reflect the three dimensional nature of chest asymmetry. The control data in this study revealed a median very close to 1 indicating symmetry of the chest with minimal rotation. The range of values in this study shows an expected variation amongst healthy subjects. This suggests that the GIofBS may be a useful clinical tool to describe the amount of scoliotic rotation in adults with movement dysfunction.

The range of external rotation / abduction was variable in this study, reflecting a typical range in flexibility across the cohort of young healthy adults. The minor yet statistically significant asymmetry between L and R hip range found in this study may be reflective of lower limb dominance, and had no functional significance.

Participants in this study ranged in age from 18 to 34 whereby skeletal growth is complete and they are yet to experience significant musculoskeletal issues associated with ageing. A small negative correlation between age and chest depth width ratio was found in this study indicating that the younger participants had a higher depth width ratio indicative of a raised chest shape. Due to the small correlation in only one of the GIofBS measures, and the relatively narrow age range included in this cohort, age did not influence GIofBS outcomes.

Limitations and Strengths

Limitations of this study include a relatively small sample size (n=48). Although the sample size for collection of normative data in health instruments is typically over 100, some have been as low as 32 (6). A larger sample size would provide greater representation across an extended age group (beyond 40 years old) allowing for reference across the lifespan for people with CP, however given the focus on younger adults presenting to complex disability clinics, this age range was considered representative of that population. A further limitation was the relatively homogenous sample of fit, healthy active young adults recruited largely from undergraduate and employed physiotherapists, being a sample of convenience. However, although inclusion of young adults who are less active may impact on the chest shape/size and hip flexibility outcomes, inactivity is unlikely to be linked to greater postural asymmetry in

those without movement dysfunction. A strength of the study is that it is the first to provide control data that may be relevant to reference to adults with CP, enabling normative information against which to evaluate symptom risk, status change and response to interventions.

## Conclusion

This study demonstrated that a cohort of young, healthy subjects exhibit some variation in postural range, but minimal asymmetry. Although a few outliers demonstrated more extreme values of postural symmetry, as they are able to freely move out of any postural set they are not a risk of it becoming an abhorrent posture causing destructive body shape and adverse consequences.

Some small variation in postural symmetry and range of movement is common amongst healthy participants with no movement disorders. This data will aid in the interpretation of measures of postural symmetry in young adults with CP.

## Acknowledgements

The participation of staff, students and friends of Monash University Peninsula Campus and St. Vincent's Hospital Melbourne is gratefully acknowledged. A grant received from the School of Primary Health Care Research and Research degrees committee (RRDS) inter-professional industry seeding grant scheme facilitated data collection.

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# Appendix B: Funding Acknowledgements

A number of funding sources have been accessed to support this research, as well as unfunded time through St. Vincent's Hospital, Melbourne. Acknowledgement of the funding received is presented in Table B.1 below. Remaining outputs resulted from unfunded research.

Table B.1. Details of funding sources, amount and applicants relative to thesis outputs

| **Funding source** | **Amount** | **Applicants** | **Thesis output** |
|---|---|---|---|
| St. Vincent's Hospital, Melbourne Research Endowment Fund | $22, 406.00 | **C Holmes**<br>K Brock<br>P Morgan | Chapter 3 |
| School of Primary health care research and research degrees committee (RRDS) inter-professional industry seeding grant scheme. | $4,914.00 | P Morgan<br>N Dalwood<br>K Bowles<br>**C Holmes** | Appendix 1 |
| OrthoPediatrics Travel Scholarship from the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) | US $1500 and conference registration | **C Holmes** | Poster presentation at the AACPDM Meeting in Cincinnati, Ohio from October 10th-13th, 2018. |

# Appendix C: PDF Versions of Published Manuscripts

Disability and Rehabilitation

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# Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review

Carlee Holmes, Kim Brock & Prue Morgan

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REVIEW ARTICLE



# Postural asymmetry in non-ambulant adults with cerebral palsy: a scoping review

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**ABSTRACT**

**Purpose**: Non-ambulant adults with cerebral palsy are vulnerable to development of postural asymmetry and associated complications. The primary aim of this scoping review was to identify postural deformities in non-ambulant adults with cerebral palsy.

**Materials and methods**: Comprehensive searches were undertaken in EMBASE, CINAHL, AMED, Cochrane, Psych INFO, and Joanna Briggs (1986–Jan 2017), supplemented by hand searching. Two reviewers independently extracted data using a customised tool focusing on study design, participant characteristics, postural descriptors, measurement tools, and interventions.

**Results**: From 2546 potential records, 17 studies were included. Variability in populations, reporting methodology, and measurement systems was evident. Data suggest more than 30% of this population have hip migration percentage in excess of 30%, more than 75% experience "scoliosis", and more than 40% demonstrate pelvic obliquity. Estimates ranged from 14% to 100% hip and 32% to 87% knee contracture incidence. Conservative interventions were infrequently and poorly described.

**Conclusion**: Many non-ambulant adults with cerebral palsy experience postural asymmetry associated with windswept hips, scoliosis, pelvic obliquity, and limb contracture. Options for non-radiographic monitoring of postural asymmetry should be identified, and conservative interventions formally were evaluated in this population.

**➤ IMPLICATIONS FOR REHABILITATION**

- The common postural asymmetries of windswept hips, scoliosis, pelvic obliquity, and limb contracture require standardised clinical measurement.
- Radiography is most commonly used to monitor postural asymmetry in this population, but standardised positioning is not applied and may not be feasible indicating a need for alternate methods and rigorous documentation.
- The Posture and Postural Ability Scale may be considered for use in the management of body shape in adults with CP.

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**KEYWORDS**
Cerebral palsy; posture; deformity; scoliosis; windswept; migration percentage

## Introduction

Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain [1]. The lifelong motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, and epilepsy further adding to the heterogeneity of this population [1]. Although the initial neurological lesion is static in nature, the extent of the motor disability ranges from minimal to profound, with a deterioration in function reported in many adults with CP along with secondary consequences such as musculoskeletal deformities, pain, fatigue, and falls [2,3].

Management of secondary complications, particularly postural asymmetry such as scoliosis and pelvic obliquity, is an important aspect of healthcare for adults with CP. The development of postural asymmetry of the thoracic cage can affect respiration and lung function resulting in pneumonia and respiratory diseases for the most severely affected adults [4]. Reports of pain, difficulty seating, and decreased participation have been described as a consequence of an asymmetrical posture in this population [5]. Further complications such as hygiene issues, compromised swallow, sleep disturbance, and pressure injuries can also arise [6]. Postural asymmetries may be most evident in non-ambulant adults with CP with low motor function typically described as Gross Motor Function Classification System (GMFCS) Levels IV and V [7]. This predominantly wheelchair user subgroup makes up around 20% of the total population of people with CP [8], exhibiting the highest risk for secondary complications and potential for a shorter life expectancy [9]. With improved access to medical care, the CP population with severe and multiple disabilities, do survive into adulthood [9], and thus assessment and management of the secondary complications is essential to ensure quality of life is maximised.

Diagnosing postural asymmetry such as scoliosis and pelvic obliquity may be particularly challenging in adults with CP who have profound disability, which frequently includes limitations in communication and cognition. As such, the identification, monitoring, and management of secondary complications for adults with CP becomes critical given the potential for adverse consequences to rapidly escalate. Although hip surveillance guidelines are generally followed for at-risk children with CP, effective

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Supplemental data for this article can be accessed here.

methods such as radiography to monitor posture over time is problematic for many adults with severe disability who cannot cognitively tolerate radiographical procedures or cannot achieve a standardised supine position for radiographs due to postural asymmetry and contractures. Further, the complex three-dimensional postural asymmetry frequently encountered in adults with severe CP is difficult to quantify by standard methods such as goniometry and calculation of Cobb angles without using resource-intensive computerised tomography. Consequently there is limited evidence available regarding the nature of the postural asymmetry, progression of postural asymmetry over time, and effective interventions to prevent, minimise, and/or remediate postural asymmetry in the adult population with CP.

Both the extent of scoliosis and pelvic obliquity may progress following skeletal maturity in adults with CP, but the rate and timing of such progression is not well recognised [10,11]. Although there is considerable evidence surrounding the proportion of children with displaced hips and scoliosis [12,13] information tracking the progress of these children and their posture into adulthood is scarce. Published evidence tends to focus on the outcomes of hip or spinal surgical techniques in various cohorts, and/or excludes those with most severe disability due to them being a higher surgical risk due to respiratory compromise [4]. Conservative interventions to address postural asymmetry such as prescription of customised sleep, seating, and pressure management systems may also be poorly described, and lack rigorous methods of evaluation. Clinicians thus lack guidelines to assist them in managing postural asymmetry in this complex population.

Nevertheless, the population of adults with severe CP (GMFCS Levels IV and V) need careful and regular monitoring, and deserve timely and affordable interventions to manage postural asymmetry to enhance the quality of their life. Given the lack of high quality evidence available, a scoping review was thus initiated following the guidelines of Arksey and O'Malley [14]. The key foundations of a scoping review are to examine broad areas, to identify gaps in the evidence, clarify key concepts, and report on the types of evidence that address and inform practice in a topic area [14]. The primary aim of this scoping review was thus to investigate the nature, prevalence, and conservative management of postural deformities in non-ambulant adults with CP (GMFCS Levels IV and V). A secondary aim of this review was to synthesise measurement tools used to document postural deformities in non-ambulant adults with CP, and describe conservative interventions selected for the management of postural deformities in this population.

## Method

An initial search strategy was created in Ovid Medline and search terms customised as necessary for the following databases: EMBASE, CINAHL, AMED, Cochrane, Psych INFO, and Joanna Briggs, until January 2017. In addition, targeted hand searching of reference lists supplemented the strategy. The search terms included the following keywords, with Boolean operators applied to combine domains: CP, scoliosis, Cobb angle, hip dislocation, hip subluxation, hip dysplasia, migration percentage (MP), pelvic obliquity, windblown, windsweep, asymmetry, posture, deformity. Adolescent and adult age group filters were applied. The full search strategy is included in Supplementary Material 1.

The titles and abstracts identified by the initial search strategy were screened to identify potentially eligible reports and retrieve full-text reports. When the title or abstract did not clearly indicate whether an article should be included, the complete article was retrieved and reviewed and consensus achieved by two authors (PM and CH). Full-text reports were independently evaluated by two authors (CH and PM) for the following inclusion criteria: (1) all participants with a diagnosis of CP, or if mixed cohorts included, data pertaining to those with CP able to be extracted; (2) all participants GMFCS Levels IV and/or V (or described as non-ambulant), or if mixed cohorts included, data pertaining to GMFCS Levels IV and/or V able to be extracted; (3) adult age group, or $\geq$80% of participants over the age of 16, or if wider age groups included, data pertaining to adult ages able to be extracted; (4) data included measurement of any aspect of posture; (5) intervention data collected described as conservative, inclusive of spasticity management; (6) English language. Any trial design was accepted, including case series. Given the advances in medical and surgical management of CP over the last 30 years [15] a decision was made to restrict studies to those published from 1986 onwards.

### *Charting the data and data extraction*

A standardised data extraction form was constructed to identify and detail key features of each study using recommended information from the Joanna Briggs Institute (JBI) [14]. Two reviewers (CH and PM) initially independently pilot tested the form with a small subset of representative studies to confirm the content and ensure consistency in its application. The extracted study details focused on study design, sample participant characteristics, descriptors of posture, measurement tools utilised, and any interventions applied. As this was a scoping study, an appraisal of study quality was not included. Results are presented in descriptive format.

## Results

Database searching identified 3491 potential records, and an additional 39 records were identified through other sources (hand searching reference lists). After duplicates were removed, 2546 potential records remained. Following exclusion of records based on title and abstract, 93 records remain to be screened based on full text. Seventeen studies were included in the final synthesis (Figure 1).

## Study samples

The majority of studies arose from USA ($n = 7$), with four from Japanese groups, and three studies arising from Sweden (Table 1). Five studies were identified as being led by a physiotherapist [4,16–18]. Six studies used targeted recruitment methods aimed at selecting participants with identified postural abnormalities [10,12,16,19–21].

Study participants ranged from 5 [16] to 234 [22]. Most studies were retrospective or observational cohort studies involving medical record (chart) reviews ($n = 13$), and/or radiographs ($n = 12$), with 11 studies including additional physical examination components. Two studies recruited participants from a CP registry [17,18] whilst four studies recruited participants exclusively from residential institutions [19,20,23,24]. The remaining studies recruited from the community and tertiary hospitals. Only three studies included longitudinal data [11,12,21].

Not surprisingly, more recent studies used the GMFCS to describe their populations as Levels IV and/or V [2,4,10,11, 16–18,23–25] with generally older studies using terms such as "bed ridden", "propped sitters", or "non-ambulatory" to describe the motor function of participants [12,19–22,26,27]. In these instances, the review authors converted descriptions to equivalent

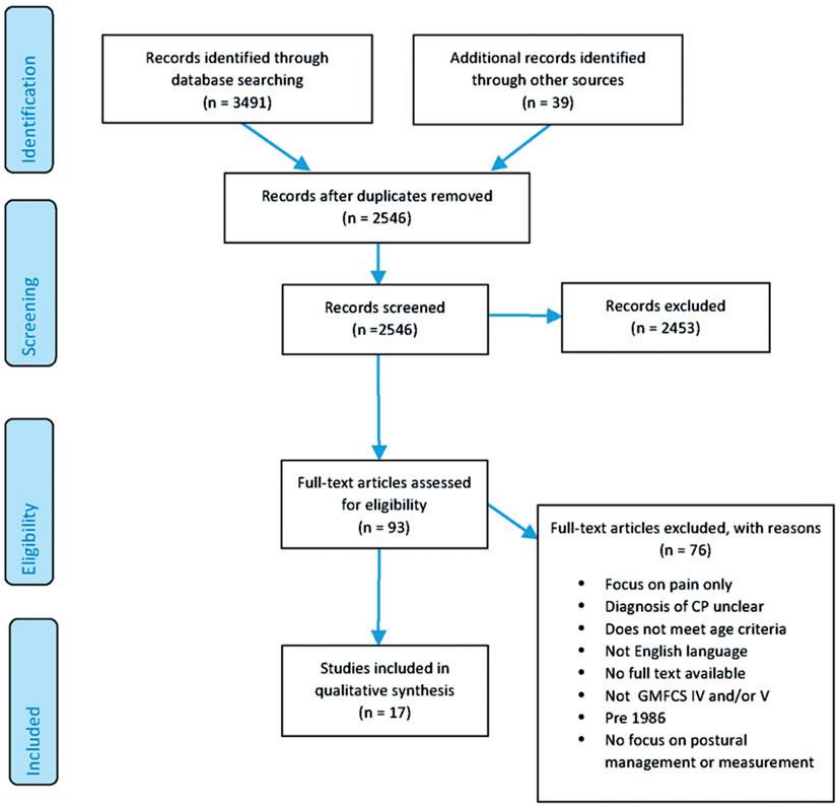


Figure 1. PRISMA flow diagram. Flow chart mapping the results of records identified throughout the database searching process and reason for exclusion.

GMFCS Levels. Four studies [4,16,23,24] exclusively recruited those at GMFCS Level V, with the remaining studies including both GMFCS Levels IV and V participants. Where reported, spastic quadriplegia was the dominant motor type and distribution. As expected, many participants had additional impairments such as epilepsy, intellectual impairment, and were non-verbal. All but one study [24] recruited exclusively those with a diagnosis of CP. Sato's study [24] included two of 22 participants with other neurological disorders. Where reported ($n = 14$), the mean age was less than 40 years old in 11 studies. A further three of these 14 studies included participants with a mean age between 40 and 45 years [4,24,27]. Eleven studies included broader inclusion criterion of which relevant data pertaining to postural deformities, adult age groups and/or GMFCS Levels IV and V was extracted. Table 1 synthesises the study type and demographic information regarding included study participants.

## Postural deformities

### *Impairments*

The postural impairments of hip deformity, scoliosis, pelvic obliquity, and lower limb contractures often co-occur. These four musculoskeletal deformities (impairments) were reported by 16/17 included studies, with 11 describing information regarding more than one postural deformity. Descriptors of scoliosis or thoracic deformity ($n = 12$) were most frequent (Table 2). Information regarding MP/hip dislocation/hip subluxation was reported in seven studies, and pelvic obliquity in six studies (Table 2). Of note, where the study included broader age groups or level of disability, only postural deformities that are specifically attributable to the population of interest are included in Tables 2 and 3.

MP is the percentage of the bony width of the femoral capital epiphysis which falls lateral to a line drawn vertically from the bony lateral margin of the acetabulum, on an AP pelvis radiograph. In the included studies, MP was reported as the proportion exhibiting either hip enlocation or not (0 or 100%) [18,20,26], or proportions of participants exhibiting a specified range of MP [10,22,23] predominantly using Reimer's index from radiographs to calculate. Only Kalen's study [26] did not specify the use of Reimer's index for MP estimation. Heidt and colleagues reported median values of MP for high side and low side hips [10]. Those who did not specifically recruit participants with hip dysfunction, revealed MP proportions greater than 30% (suggesting subluxation) of 24% [19], 25% [22], and 32% [23] of their cohort.

Twelve studies described evidence of scoliosis and/or associated thoracic deformity in participants. Eight of these studies described the severity of the scoliosis by calculation of Cobb angle from standard radiographs [11,12,19–21,24–26]. Overall synthesis of data pertaining to scoliosis was not possible due to variation in reporting methods. For example, Oda and colleagues [11] described mean scoliosis relative to age groups, Saito [21] and Thometz [12] described mean scoliosis relative to functional

**Table 1.** Study details and demographic information of included participants.

| First author, year | Country | Study type | Study setting | Specific inclusion or exclusion criterion related to postural abnormalities | *n* (entire cohort) | Age: mean (sd), range | Gender *n* (%) | GMFCS Level | CP subtype/motor distribution | Reported comorbidities |
|---|---|---|---|---|---|---|---|---|---|---|
| Boldingh, 2007 | Netherlands | Cross-sectional | 58% residential nursing facility | Femoral head able to be identified on X-ray | 140 (160) | 36 (16)[a], 16–84 | M: 54%[a] F: 46% | Level V | Spastic, quadriplegia | Minimum mental age 4 years |
| Heidt, 2015 | Australia | Prospective cohort | Transition clinic | MP >30%, history of adductor release | 67 (98) | 19[a], 15–24 | M: 65[a] F: 33 | 32 Level IV; 35 Level V | N/R | N/R |
| Hodgkinson, 2001 | France | Cross-sectional, multi-centre | 32 centres – including some residential | Nil reported | 234 | 28, 18–38 (95% CI) | M: 59% F: 41% | Level IV and V[b] | 100% spastic quadriplegia; 26.9% athetosis | 51.1% multiple disabilities; 49.1% epilepsy; 47.2% hip pain |
| Horimoto, 2012 | Japan | Case series | Acute hospital | Previous CT scan for diagnosis of pneumonia | 20 | 43 (18), 20–70 | M: 16 F: 4 | Level V | N/R | N/R |
| Kalen, 1992 | USA | Cohort study | Single residential institution | Nil reported | 56 (62) | 39[a], 29–67 | M: 26[a] F: 36 | Level IV and V[b] | 54 spastic quadriplegia; six spastic diplegia; one hemiplegia; one not categorised[a] | Subset of 14 with scoliosis >45°: 100% mental retardation; 22% pressure ulcers |
| Knapp, 2002 | USA | Retrospective cohort study | Four residential institutions | Established hip dislocations or subluxations | 29 (117) | 34, 21–52 | M: 17 F: 12 | Level IV and V[b] | Spastic quadriplegia | N/R |
| Littleton, 2011 | USA | Single subject research design | Institutional and community | Severe scoliosis (fixed deformity), history of respiratory illness | 5 | 17–37 (37, 22, 25, 26, & 17) | M: 3 F: 2 | Level V | Spastic quadriplegia | Four gastrostomy tubes, four hip surgery, three seizure disorders, two sleep apnoea |
| Murphy, 1995 | USA | Observational cohort study | Community | Nil reported | 67 (101) | 43 (19–74)[a] | M: 53[a] F: 48 | Level IV and V[b] | Two hemiplegia; five diplegia; 26 quadriplegia; 34 dyskinesia (non-ambulatory groups) | $N = 44$ prior orthopaedic procedures; 50% incontinent (urinary); 43% back pain; pain in lower limb joints 44%; leg contractures 91% |
| Oda, 2017 | Japan | Observational cohort study | Single tertiary hospital | Excluded prior scoliosis surgery | 47 (92) | 20–69 | N/R | Level IV and V | N/R | N/R |
| Pritchett, 1990 | USA | Observational cohort study | Group homes | 50 treated and 50 untreated with unstable hips | 100 | Untreated: 26 (21–50); treated: 25 (20–42) | Untreated: M: 23 F: 27 Treated: M: 24 F: 26 | Level IV and V[b] | spastic | Profoundly mentally retarded and incontinent; 75% seizure disorder; 85% non-verbal |
| Rodby-Bousquet, 2012 | Sweden | Reliability and tool validation study | CPUP program (registry) | Nil reported | 12 (30) | 19–22[a] | M: 15[a] F: 15 | Six Level IV Six Level V | N/R | N/R |
| Rodby-Bousquet, 2013 | Sweden | Cross-sectional study | CPUP program (registry) | Nil reported | 30 (102) | Md 21[a] (19–22) | M: 62[a] F: 39 | 10 Level IV; 20 Level V | Unilateral spastic ($n = 26$), bilateral spastic ($n = 45$), ataxic ($n = 12$), dyskinetic ($n = 19$)[a]. | N/R |

(*continued*)

**Table 1.** Continued

| First author, year | Country | Study type | Study setting | Specific inclusion or exclusion criterion related to postural abnormalities | *n* (entire cohort) | Age: mean (sd), range | Gender *n* (%) | GMFCS Level | CP subtype/motor distribution | Reported comorbidities |
|---|---|---|---|---|---|---|---|---|---|---|
| Saito, 1998 | Japan | Retrospective cohort study | Single residential institution | X-ray before 15 years, scoliosis at least 10° | 31 (37) | 25, 15–36 | M: 18[a] F: 19 | Level IV and V[b] | Spastic | High levels of intellectual impairment & seizures 31 had contractures; 35 non-verbal[a] |
| Sandström, 2004 | Sweden | Observational cohort study | Institution and community | Nil reported | 18 (48) | 33 (8.2)[a] | M: 48%[a] F: 52% | 9 Level IV 9 Level V | 13 hemiplegia; 19 diplegia; 9 quadriplegia; 7 dyskinesia[a] | N/R |
| Sato, 2016 | Japan | Reliability and tool validation study | Single residential institution | Excluded prior spinal surgery | 20 (22) | 35–52 | M: 11% F: 11% | Level V | N/R | N/R |
| Senaran, 2007 | USA | Retrospective cohort study with matched control group | Single tertiary hospital | Prior ITB implantation and age/gender/GMFCS control with scoliosis | ITB group 11, Control 14 | 18; 16–21 | ITB group: 6 M: 5 F Control: 10 M: 4 F | ITB group: all Level V, Control: 2 Level IV & 12 Level V | Spastic | N/R |
| Thometz, 1998 | USA | Retrospective cohort study | Previous students of special school | Scoliosis – X-rays available after skeletal maturity and at least 4 year follow up X-ray | 40 (51) | 20–63 | N/R | Level IV and V[b] | 23 spastic quadriplegia, 7 spastic diplegia, 5 spastic hemiplegia, 16 miscellaneous = ataxia, athetosis, hypotonia | Intellectual impairment |

MP, migration percentage; N/R, not reported; ITB, intrathecal baclofen; CPUP, Sweden's follow-up surveillance program for CP.
[a]Entire cohort, not age/GMFCS Level subset.
[b]Author interpretation.

ability, and others described proportions of participants exhibiting a specific Cobb angle range [19,20,26]. Studies led by Rodby-Bousquet et al. [18] and Murphy [27] reported proportion of participants demonstrating any scoliosis (77% and 90%, respectively), and Senaran's study [25] reported the mean Cobb angle of group participants (57° ITB group, 66° control group). Other authors provided additional descriptors of the associated thoracic diameter using calculations from CT scans or digital photography [4,24], curvature [24], and impact of the scoliotic deformity [16]. Where radiographic outcomes were described, Senaran et al. [25] measured scoliosis in supported sitting while studies led by Oda and that by Thometz used supine as the measurement position [11,12]. A "standard upright" position was used in Kalen's study [26] and the measurement position was not stated by four authors [4,19–21,24]

Pelvic obliquity was similarly reported using a range of methods, most commonly either proportion of participants exhibiting pelvic obliquity (%) [16,19,20,26] or the mean pelvic obliquity in the cohort (°) [10,25]. Where reported ($n = 6$), the presence of any pelvic obliquity in the cohort ranged from 41% [19] to 63% [20] and the amount of pelvic obliquity ranged from 1° to 14° (GMFCS Level IV) or 1° to 22° (GMFCS Level V) [10], and 0° to 20° (ITB group) or 0° to 45° (control group) [25].

Information regarding hip or knee contractures was provided by six studies [2,18,19,23,26,27] and generally described the proportion of participants exhibiting a contracture. Where reported, goniometry or visual estimates of joint angles was implemented. Where specified, estimates ranged from 14% hip and 32% incidence of knee contractures [23] to 100% hip [2] and 87% knee contractures [18] in the cohorts studied.

### *Activity limitations and postural asymmetries*

Rodby-Bousquet et al. [17,18] was the only lead author who utilised a clinical assessment scale (the Posture and Postural Ability Scale) to describe the static and dynamic posture of adults with CP. This scale classifies posture in supine, prone, sitting, and standing, with attention to the relationship between body parts (head, trunk, arms, and legs), weight bearing, and symmetry. It also describes the participant's ability to move in and out of a static posture, thus utilising an activity limitation framework. Postural ability is described using a 7-point scale, from Level 1: unplaceable in an aligned posture, through to Level 7: able to move into and out of a position. The low median scores for postural ability for the six participants described as GMFCS Level IV (4, range: 3–7) and six described as Level V (1.5, range: 1–4) indicate that at best the person was able to initiate flexion/extension of trunk (Level 4) and, if rated at Level 1, was unplaceable in an aligned posture in supine [17]. In sitting, those at GMFCS Levels IV and V were both rated as 2 (placeable in an aligned posture but needs support). The quality of the posture in both the frontal and sagittal planes was very low for participants with most scores of 0 indicating they were unable to achieve any of the six requirements of alignment of body parts in either supine or sitting [17]. In Rodby-Bousquet et al.'s 2013 study, 17/30 (participants GMFCS Levels IV and V) were unable to achieve a symmetric trunk in supine, and 18/30 were unable to achieve a symmetric trunk in sitting as measured by the Posture and Postural Ability Scale [18].

### *Conservative interventions*

As most studies were observational, descriptions of non-surgical interventions (conservative) and subsequent evaluation of any conservative interventions were scarce (Table 4). Where conservative interventions were described, they may not have been specific to GMFCS Levels IV and V, hence limited data were able to be extracted. Eight studies reported the use of customised wheelchairs (and/or customised seating inserts) in describing their cohorts, with some reporting percentage usage [23,26]. Boldingh et al.'s study [23] related the need for special moulded seating to the incidence of femoral head deformity, with 87% of participants with serious femoral head deformity and 51% of participants with no femoral head deformity requiring special moulded seating [23]. Kalen's study related the need for customised moulded seating to the severity of scoliosis, with 54% of participants with Cobb angles greater than 45° requiring customised moulded seating [26]. Littleton et al. [16] provided detail about the customisation of wheelchairs used, with description of head support, tilt in space feature, custom cushion, and lateral support, pelvic positioning device, and foot support. Several studies noted the inadequacy of the current seating systems for study participants as a striking outcome from their findings in the discussion [20,22,27], or made reference to it as a potential management strategy [20,22]. Only one study noted the use of customised side-lyers to maintain a side lie position in relation to respiratory function [16].

Two studies [10,25] described prior or current spasticity management interventions in their cohorts as methods to reduce or slow progression of the postural deformities of the trunk, pelvis, and hips. In Heidt et al.'s study [10] participants had previously undergone a variety of spasticity management techniques including oral medications, nerve phenolisation, botulinum toxin, and intrathecal baclofen (ITB) pumps with the aim of reducing known causes of pelvic obliquity leading to windswept hips and late hip dislocation. Senaran and colleagues [25] in discussion proposed that the reduction in asymmetrical spasticity from the ITB pump, allowed greater balance in paraspinal and postural muscles facilitating better upright sitting posture, however concluded it had no significant effect on scoliosis progression in people with quadriplegic CP. Efficacy of these strategies ongoing throughout adulthood are not addressed. No studies specifically described strategies to manage contracture in the adult cohorts other than Heidt et al. who mentioned prior botulinum toxin and iliopsoas lengthening strategies to manage hip contracture in an unknown proportion of the cohort [10].

## Discussion

This scoping review was able to identify a small body of literature relating to common postural deformities in non-ambulant adults with CP (GMFCS IV and V) and describe prevalence, conservative management, and measurement tools used. While it is clear that many non-ambulant adults with CP (GMFCS IV and V) experience significant and disabling postural asymmetries requiring standardised specialist health care services throughout the life span [28] there is a lack of evidence as to what interventions they require

Table 2. Total number of postural deformities (impairments) described in scoping review studies.

| Postural deformity | Incidence of description ($n = 16$ studies[a]) |
|---|---|
| MP/hip dislocation, subluxation, femoral head deformity | 7 [10,17,19,20,22,23,26] |
| Scoliosis and thoracic deformity | 12 [4,11,12,16,17,19,20,21,24–27] |
| Pelvic obliquity | 6 [10,16,19,20,25,26] |
| Hip and/or knee contracture | 6 [2,17,19,23,26,27] |

MP, migration percentage.
[a]Studies may have described more than one postural deformity.

**Table 3.** Summary of postural asymmetries recorded.

| First author, year | Postural deformity | Method of assessment |
|---|---|---|
| *Hip deformity* | | |
| Boldingh, 2007 | MP: % of cohort<br><30: 56%<br>30–59: 22%<br>60–100: 10%<br>Femoral head deformity: % of cohort<br>None: 29%<br>Moderate: 41%<br>Severe: 29% | Standard AP radiograph of hips and pelvis (Reimer's)<br>From X-ray using 3-point scale – none, moderate, severe |
| Heidt, 2015 | MP Md (IQR):<br>*High side hip*<br>31 (22–41) Level IV; 40 (29–88) Level V<br>*Low side hip*<br>15 (3–25) Level IV; 9 (1–15) Level V | Standard AP radiograph of hips and pelvis (Reimer's) |
| Hodgkinson, 2001 | MP: % of cohort<br><33: 74.7%<br>33 < 100: 13.2%<br>100: 12.1% | Standard AP radiograph of hips and pelvis (Reimer's) |
| Kalen, 1992 | MP: % of cohort<br>100: 64% (9/14)[a] | Not reported |
| Knapp, 2002 | MP[b]: % of cohort<br>30 < 100: 24% (9/38)[c]<br>100: 76% (29/38)[c] | Standard AP radiograph of hips and pelvis (Reimer's) |
| Pritchett, 1990 | MP: % of cohort<br>100: 100%[b]<br>50 untreated – 34 bilateral; 16 unilateral hips affected<br>50 treated – 33 bilateral; 17 unilateral hips affected | Standard AP radiograph of hips and pelvis (Reimer's) |
| Rodby-Bousquet, 2013 | MP: % of cohort<br>100: 30% | Standard AP radiograph of hips and pelvis (Reimer's) |
| *Scoliosis* | | |
| Horimoto, 2012 | Protocol 1 (mm): AP diameter mean 153 (33); LL diameter 261 (38); AP/LL 0.6 (0.1)<br>Protocol 2 (mm): AP diameter mean 198 (43); LL diameter 242 (36); AP/LL 0.8 (0.2) | Calculation of anteroposterior and laterolateral diameter of thorax from CT scan using two different protocols |
| Kalen, 1992 | Scoliosis: >45° – 33% (14/42) | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Knapp, 2002 | Scoliosis:<br><45° – 17% (5/29)<br>45°–135° – 55% (16/29) | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Littleton, 2011 | Scoliosis: "severe with fixed deformity"[b] 100% (5/5)<br>Rib impingement against iliac crest (3/5); R thoracic convexity (3/5); L thoracic convexity (2/5); R rib hump (2/5); L rib hump (2/5) | Descriptive only |
| Murphy, 1995 | Scoliosis: 90% | N/R |
| Oda, 2017 | Scoliosis (mean Cobb angle)[e]<br>Age 20–29: IV 34°, V 74°<br>Age 30–39: IV 42°, V 38°<br>Age 40+: IV 56°, V 64° | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Pritchett, 1990 | Scoliosis:<br>0°–10°: 15%<br>10°–30°: 19%<br>30°–60°: 25%<br>>60°: 41% | Standard AP and lateral spine X-ray; calculation of Cobb angle |
| Rodby-Bousquet, 2013 | Scoliosis present: 77% | Clinical examination or prior spinal fusion surgery |
| Saito, 1998 | Population of "sitters" $n = 13$<br><40°: 2/13, 15%<br>40°–60°: 7/13, 54%<br>>60°: 4/13, 31%<br>Population of "bedridden" $n = 7$<br>>60°: 7/7, 100% | Standard AP and lateral spine X-ray; calculation of Cobb angle[e] |
| Sato, 2016 | 14/22 S-shaped curves<br>8/22 C-shaped curves<br>Mean thoracic curve 30.8°<br>Mean lumbar curve 41.1°<br>Mean Upper trunk lateral lean 5.5°<br>Mean Lower trunk lateral lean 14.4°<br>Mean Upper trunk rotation 4.9°<br>Mean Lower trunk rotation 8.6° | Standard AP and lateral spine X-ray; calculation of Cobb angle PLUS<br>Calculation of 3D coordinates from anatomical landmarks for identification of lateral lean & upper and lower trunk rotation using digital cameras |
| Senaran, 2007 | Scoliosis<br>ITB group: mean 56.8° (15–96°)[d]<br>Control group: mean 66.2° (24–113°)[d] | Standard AP and lateral spine X-ray; calculation of Cobb angle (in supported sitting) |

(*continued*)

**Table 3.** Continued

| First author, year | Postural deformity | Method of assessment |
|---|---|---|
| Thometz, 1998 | Scoliosis mean angle (range)<br>Walks only in PT: 68.3° (32°–1 17°), $n = 6$<br>Indep sitter: 68.7° (40°–128°), $n = 9$<br>Dep sitter: 76.1° (25°–152°), $n = 21$<br>Bedridden: 119.7° (59°–147°), $n = 4$ | Standard AP and lateral spine X-ray; calculation of Cobb angle (in supine) |
| *Pelvic obliquity* | | |
| Heidt, 2015 | GMFCS IV: PO range 1°–14°; Md: 4°, IQR: 2°–7°<br>GMFCS V: PO range 1°–22°; Md: 7°, IQR: 4°–11°<br>Geometric means of Sharp's angle using ITDL for<br>**HIGH** side<br>GMFCS IV: 47.57 (SD 1.32),<br>GMFCS V: 53.85 (SD 1.71)<br>for **LOW** side<br>GMFCS IV: 37.43 (SD 1.39)<br>GMFCS V: 26.18 (SD 1.87) | From X-ray using intersect of horizontal reference line parallel to the frame of the radiograph with three reference lines: ITDL, ITL & IICL<br>ITDL – Line joining acetabular tear drops |
| Kalen, 1992 | PO present in 57% (8/14) with scoliosis >45° | N/R |
| Knapp, 2002 | PO present in 41% (12/29) | N/R |
| Littleton, 2011 | R PO present (3/5)<br>L PO present (2/5) | Descriptive from physical examination |
| Pritchett, 1990 | PO present in 63% (both treated hip & untreated hip groups) | Pelvic radiograph and physical examination |
| Senaran, 2007 | ITB group: mean PO 10.5° (0°–20°)[d]<br>Control group: mean PO 13.8° (0°–45°)[d] | Radiograph: more than 10° angle between horizontal and a line connecting the most proximal points of the iliac crests |
| *Lower limb contracture* | | |
| Boldingh, 2007 | 58% hip abduction limitation (>30°)<br>14% hip flexion contracture (loss of >30° extension)<br>32% knee flexion contracture (loss of >30° extension) | Goniometer |
| Kalen, 1992 | Hip flex contracture mean 26° for those with scoliosis >45° ($n = 14$) | N/R |
| Knapp, 2002 | 38% severe hip adduction contracture (11/29) | N/R |
| Murphy, 1995 | 91% lower limb contractures | N/R |
| Rodby-Bousquet, 2013 | 47% hip contractures (14/30)<br>87% knee contractures (26/30) | Goniometer |
| Sandstrom, 2004 | 100% hip contracture (18/18)<br>78% knee contracture (14/18) | ROM – visually estimated<br>scored using scale 0–2, where 0 = a few degrees, 1 = limited, 2 = normal/almost normal ROM |

PO, pelvic obliquity; AP, antero-posterior; LL, lower limbs; N/R, not reported; ITB, intrathecal baclofen; ITDL, inter-tear drop line, ITL, inter-tuberosity line, IICL, inter-iliac crest line, ROM, range of motion.
[a]Of subset with scoliosis >45°.
[b]Targeted recruitment.
[c]Nine participants had both hips affected.
[d]Author calculations from tables for ages 16 and over.
[e]Author calculation from graphed data.

and how the efficacy of any interventions may be measured and implemented.

More specifically, this review has identified that deformities of scoliosis, pelvic obliquity, and windswept hips inclusive of hip dislocation are common. A snap shot of data suggests that at least 30% of this population have a hip MP of greater than 30%, more than 75% have some degree of "scoliosis", and more than 40% demonstrate some amount of pelvic obliquity. As noted in Table 2, either impairment on its own, or existing in a triad of deformities is a significant treatment dilemma once established [5]. These postural asymmetries increase the risk of soft tissue adaptation and the formation of contractures and progressive deformities, being most prevalent in non-ambulant people with CP [18]. The inability to change position and the time spent in an asymmetrical posture contribute to the escalating problem [18]. The temporal sequencing of scoliosis, pelvic obliquity, hip dislocation, and the development of contractures is unclear [5]. More information on temporal sequencing may enhance the timing and applications of appropriate interventions. Paediatric studies have shown that hip dislocation and a severe windswept posture are preventable [10] but as this scoping review revealed, many adults have existing asymmetries and it is unclear how to manage and monitor them once established.

This review also revealed a lack of standardised measurement techniques beyond radiographs for the monitoring of postural asymmetry. Even when radiography was used, there was a lack of standardised reporting methods, particularly for pelvic obliquity and hip outcomes. Further, patient positioning was also not standardised practice for radiography procedures, ranging from supine positioning to supported sitting for scoliosis and hip assessment. Of note, this population may not always be able to conform to the standardised positioning requirements of radiographs promoted by paediatric hip surveillance programs [29] due to extent of postural asymmetry, cognitive limitations, and/or pain.

Despite a high incidence of postural deformities described in this population, reporting and evaluation of conservative management methods are also variable and scarce. Interventions including physical therapy, seating, and orthoses may be reported, but no detail was provided enabling replication, nor evaluation to identify effective strategies. Seating was most often described (as present or absent) but not in the context of a therapeutic intervention. The inadequacy of seating to meet the postural needs of the adults with CP was noted by several authors [19,22,27]. Given the dependency on this population for seated mobility, it would seem essential that effective postural management is considered. The Rehabilitation Engineering & Assistive Technology Society of

Table 4. Conservative interventions for the management of postural deformity.

| First author, year | Interventions | Evidence of use |
|---|---|---|
| Wheelchairs and seating | | |
| Boldingh, 2005 | Custom moulded seat | 61% total cohort ($n = 140$)<br>87% with serious femoral head deformity<br>51% with no femoral head deformity |
| Kalen, 1992 | Custom moulded seat | 19% in those with scoliosis $<45^{\circ}$ ($n = 42$)<br>54% in those with scoliosis $>45^{\circ}$ ($n = 14$) |
| Knapp, 2002 | Modified wheelchair | 100% usage |
| Murphy, 1995 | Solid seat or postural supports | 3/67 usage |
| Littleton, 2011 | Customised wheelchair | 5/5 usage |
| Bed positioning | | |
| Littleton, 2011 | Pillows or customised side lyer | 5/5 usage |
| Spasticity management | | |
| Heidt, 2015 | Oral medications including baclofen and diazepam | "Majority" of cohort (prior intervention) |
| | Botulinum toxin to hip adductors | 42/98[a] (prior intervention) |
| | ITB pumps | 4/98[a] |
| | Phenolisation of obturator nerve | Unspecified (prior intervention) |
| Senaran, 2007 | ITB pump | 100% of selected participants ($n = 50$) in ITB group |
| Contracture management | | |
| Heidt, 2015 | Botulinum toxin for hip flexion contractures with iliopsoas lengthening | Not specified (prior intervention) |

ITB, intrathecal baclofen.
[a]Total cohort including 29 participants GMFCS Levels II and III.

North America (RESNA) position on seating for people with various disabilities including CP discusses the need for seating functions to realign posture, regulate spasticity, accommodate, and prevent contractures and orthopaedic deformities, redistribute, and relieve pressure, increase seating tolerance and comfort and to enable position change [29]. The authors would strongly support the implementation of these recommendations for all non-ambulant adults with CP.

Several limitations to this scoping review have been identified. By narrowing the study to only CP, assessment and interventions research in other fields of adult neurology, such as the RESNA guidelines already referred to, may have provided useful information for the adult CP population. However, we deliberately wished to focus on CP-specific literature given the relatively unique situation of lifelong developmental postural asymmetry experienced by this population. We also used search terms to return studies which were centred on the reporting of incidence of postural abnormalities in this population. This may have resulted in some omission of studies focused on assessment tools. Further, a focused strategy was implemented to only review the adult CP literature. It is possible that paediatric interventions for those with CP which include the management of body shape distortion may be applicable to their adult counterparts [30–32]. However, given the potential acceleration of postural asymmetry following skeletal maturity, the efficacy of strategies that may be able to address and reverse shorter-lived asymmetry in children is unknown when applied to the long standing fixed postural asymmetries experienced by adults with CP. As with any lifelong developmental disability, management, and interventions must constantly be reviewed to meet the unique needs of adults and the capacity of their (often) ageing carers to implement.

## Implications for clinical practice and research

Valid and sensitive non-radiographic clinical measurement tools should be investigated that will enable regular monitoring of body shape over time, without the need to subject this vulnerable population to frequent radiographs. Standardised reporting of postural outcomes, *via* a minimum data set, should be mandated so as data from longitudinal observational studies can be pooled. The Posture and Postural Ability Scale has been identified as a valid non-radiographic clinical measurement tool for both lying, sitting, and standing and should be given consideration in the management of body shape in adults with CP. Given that many adults with CP have not benefited from the now routine surveillance of impairments in paediatric practice, robust investigation of the efficacy of non-surgical interventions for the management of postural asymmetry in non-ambulant adults with CP is urgently needed.

## Conclusion

Many non-ambulant adults with CP experience postural asymmetry associated with windswept hips, scoliosis, pelvic obliquity, and limb contracture. Paediatric management of potential postural asymmetries in CP has resulted in a significant decrease in incidence of these problems over the last decade or so. This has occurred following concerted collaborative efforts to devise and implement screening programs such as hip surveillance guidelines [33,34,35]. A lack of systematic, large scale follow-up studies of children with CP into adulthood with thorough longitudinal assessments [28] has heightened our lack of understanding of the long-term outcomes of postural asymmetries in this population. Future efforts should be targeted at how best postural care can be provided to non-ambulant adults with CP who have not benefited from such surveillance throughout their childhood, do not have access to these programs due to resource limitations, and/or continue to progress in postural asymmetry despite surveillance and interventions.

## Disclosure statement

The authors report no declarations of interest.

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# The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy

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ORIGINAL ARTICLE



# The intra- and inter-rater reliability of the Goldsmith indices of body symmetry in non-ambulant adults with cerebral palsy

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**ABSTRACT**

**Purpose:** Non-ambulant adults with cerebral palsy are at risk of developing asymmetry affecting thoracic cage, pelvis and hips. The primary aim of this study was to establish intra-rater and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy. The secondary aim was to establish comparative data for the Goldsmith Indices of Body Symmetry in healthy adults.

**Materials and method:** Thirty non-ambulant young adults with cerebral palsy (17 males), and 48 young healthy controls (19 males), were recruited. Thoracic shape and symmetry, pelvic orientation and hip range, was measured using the Goldsmith Indices of Body Symmetry. Intra-rater reliability was established by repeated measurement within a single session. Inter-rater reliability was established having two raters measure each participant on two sequential sessions. Analysis utilised intraclass correlation coefficients.

**Results:** The Goldsmith Indices of Body Symmetry has excellent intra-rater reliability (intraclass correlation coefficients $\geq$0.97). Inter-rater reliability for all Goldsmith Indices of Body Symmetry measures was good to excellent (intraclass correlation coefficients $\geq$0.85). Range and variability of results was greater for participants with cerebral palsy compared to comparative data.

**Conclusion:** The Goldsmith Indices of Body Symmetry has good inter and intra-rater reliability for measurement of thoracic shape and symmetry, pelvic orientation and hip range, allowing accurate tracking of postural changes over time in non-ambulant adults with cerebral palsy.

**➤ IMPLICATIONS FOR REHABILITATION**

- The Goldsmith Indices of Body Symmetry is a reliable clinical measurement tool to enable measurement of the thoracic shape and symmetry, pelvic and hip orientation in adults with cerebral palsy.
- The Goldsmith Indices of Body Symmetry, with an impairment focus, is a useful adjunct to the assessment and management of postural asymmetry in adults with cerebral palsy.

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Cerebral palsy; clinical measurement; reliability; thorax; hips; pelvis; windswept

## Introduction

Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture that are attributed to non-progressive disturbances in the developing foetal or infant brain [1]. The core motor features of CP, abnormalities of movement and posture, can lead to secondary complications associated with behaviour, musculoskeletal function and participation [1]. Management of musculoskeletal function leading to postural asymmetry involving spine, pelvis and hips, is an important aspect of healthcare for adults with CP. The development of postural asymmetry in this population impacts on cardiorespiratory [2,3] and digestive function [2] and may cause pain and pressure injuries, negatively impacting carer burden [2]. Windswept hips, consisting of abduction and external rotation of one hip and adduction and internal rotation of the contralateral hip [2,4], are one element of the complex postural asymmetry most evident in non-ambulant adults with CP with low motor function typically described as Gross Motor Function Classification System (GMFCS) Levels IV and V [5]. This predominantly wheelchair-user subgroup exhibit the highest risk for secondary complications and potential for shorter life expectancy [2,6].

Diagnosing and measuring the extent of, and contributors to, postural asymmetry including scoliosis and hip mal-alignment may be particularly challenging in adults with CP who have profound disability. Although hip surveillance guidelines are generally followed for at-risk children [7], methods such as radiography to monitor spine, pelvis and hips, are problematic for adults with severe disability who cannot cognitively tolerate traditional radiographical procedures or cannot achieve a standardised supine position for radiographs due to postural asymmetry and contractures. Further, elements contributing to the complex three-dimensional postural asymmetry frequently encountered in adults with severe CP are difficult to quantify by standard methods such as goniometry and radiographs. A recent scoping review on postural asymmetry in non-ambulant adults with CP indicated there is no standardised impairment-focused clinical measurement suitable to accurately capture and objectively record common postural asymmetries of windswept hips, scoliosis, pelvic obliquity and limb contractures [8].

There is currently limited evidence available regarding the nature of postural asymmetry, progression over time, and effective interventions to prevent, minimise and/or remediate postural asymmetry in the adult CP population. Although there is

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Supplemental data for this article can be accessed here.

considerable evidence surrounding the proportion of children with displaced hips and scoliosis [9,10], information tracking progress of these children and their posture into adulthood is scarce. The population of adults with severe CP need careful and regular monitoring, and deserve timely and affordable interventions to manage contributors to postural asymmetry to reduce pain and enhance life quality. The Posture and Postural Ability Scale (PPAS) [11] utilises an activity limitation framework to describe static and dynamic postures and the ability to move in and out of a static position. This scale has the potential to be utilised for long term monitoring purposes, however it does not allow close examination of contributing impairments, limiting potential for evaluation of specific interventions.

The Goldsmith Indices of Body Symmetry [12] is a clinical measurement tool providing objective information on thoracic shape and symmetry, pelvic orientation and hip mobility, enabling calculation of a Windswept Index at an impairment level. Windswept hips seriously impair seating comfort and function [13], as well as contributing to other secondary complications and functional limitations [2]. The Goldsmith Indices of Body Symmetry has face validity, objectively measuring constructs relating to thoracic symmetry and shape, hip and pelvic orientation and range that contribute to posture, but lacks more robust validation. It was developed in 1992 [12] and has been predominantly used with the paediatric CP population in England. The Goldsmith Indices of Body Symmetry may be used as part of a comprehensive postural evaluation exploring key impairments in supine that contribute to postural dysfunction in activities such as lying and supported sitting. It was chosen as a clinical measurement tool potentially suited for adults with severe neuromuscular disability such as CP in this study due to the scarcity of alternate appropriate and consistent non-radiographical methods. However, little is known about the reliability of the Goldsmith Indices of Body Symmetry in documenting elements contributing to postural asymmetry often observed in non-ambulant adults with CP, and in adults without disability to provide comparative control data.

The primary aim of the study was to establish the intra-rater and inter-rater reliability of the Goldsmith Indices of Body Symmetry in adults with CP with the non-ambulant classification of GMFCS Level IV or V. The secondary aim of the study was to establish comparative data in adults without disability (control group) for the Goldsmith Indices of Body Symmetry to aid interpretation of data collected from adults with CP. Study outcomes are reported using the Guidelines for Reporting Reliability and Agreement Studies [14].

## Methods

### *Setting*

Participants with CP were recruited from the Young Adult Complex Disability Service (YACDS) at St. Vincent's Hospital, Melbourne, predominantly a CP transition clinic from paediatric to adult healthcare services. Participants for the comparative control data collection were recruited from the Monash University Peninsula Campus and staff from St. Vincent's Hospital Melbourne. Ethical approval was gained from St. Vincent's Hospital Melbourne and Monash University Human Research Ethics Committees.

### *Participants*

Adults with CP: Participants aged between 17 and 40 were included if they had a diagnosis of CP and a motor classification of GMFCS level IV or V. Participants were excluded if they had a significant movement disorder, such as severe dystonia, that rendered them unable to remain relatively still during testing. Participants were also excluded if they were pregnant, or had behavioural issues that placed either raters or participant at risk.

Control data: Participants were young adults aged between 18 and 40 who self-declared no history of movement disorder and were not pregnant.

### *Recruitment*

Adults with CP: Suitable participants attending YACDS between February 2017 and August 2018 were invited to participate and provided with study information. For participants unable to consent, the invitation was provided to next of kin/legal guardians.

Control data: Suitable participants were recruited from Monash University Peninsula Campus and St. Vincent's Hospital, Melbourne *via* advertisements on notice boards and online learning sites.

All participants (or next of kin) signed informed consent.

## Outcome measures

Adults with CP: The following demographic and medical data was retrieved: gender, date of birth, living situation, GMFCS level, CP subtype (according to Bax et al. [15], usually sourced from paediatric referral facility), height and weight, prior scoliosis surgery, communication (verbal, non-verbal and nil consistent effective communication) and cognitive capacity (ability to provide own consent).

Control data: Participants' age, gender, height and weight were recorded. Any self-declared history of musculoskeletal issues potentially impacting spinal/hip alignment was sourced.

### *Goldsmith indices of body symmetry*

The Goldsmith Indices of Body Symmetry involves three testing procedures: chest measures; measurement of rotation of the pelvis as influenced by movement of the flexed knees; and measurement of combined hip external rotation and abduction with a stable pelvis. Data was collected using the Anatomical Measurement Instrument as per standard testing protocol [12,16]. Prior to commencement, an optimal standard starting position was established, being crook lie with 110° knee flexion and level pelvis. If this position was unable to be achieved an alternate starting position was identified, recorded, and used as the starting position for all measurement. The outcome measures collected, procedure and guide for data interpretation are described in Table 1.

## Procedures

Adults with CP: Two experienced therapists (raters) undertook participant measurement, having undergone additional training in administration of the Goldsmith Indices of Body Symmetry Anatomical Measurement Indices. The training was provided by a physiotherapist who has gained certification of Level 3 Award in Measurement of Body Symmetry (Open College Network West Midlands).

Each participant was measured by each rater with a 15 to 30 min rest between the two measurement sessions. Each Goldsmith Indices of Body Symmetry outcome was measured four times in succession prior to advancing to the next procedure

Table 1. Outcome measure data obtained from the Goldsmith Indices of Body Symmetry.

| | Outcome Measure | Procedure | Instrument | Interpretation of results |
|---|---|---|---|---|
| 1 | Chest Depth Width Ratio | Chest depth value divided by total chest width value (chest depth/R side chest + L side chest) | Chest frame | Values <1.0 suggest less chest depth relative to width; values >1.0 suggest greater chest depth relative to width |
| 2 | Chest Right Left Ratio | Right chest value divided by left chest value (Right side chest/Left side chest) | Chest frame | A score of 1 is indicative of symmetry. A score of 0 to <1 is indicative of chest anticlockwise rotation. A score of >1 is indicative of chest clockwise rotation. |
| 3 | Angle Between Legs and Pelvis (ABLAP) | Angle of legs minus angle of pelvis<br>R ABLAP = R leg angle – R pelvic angle<br>L ABLAP = L leg angle – L pelvic angle | Leg paddle and pelvic bridge | The side (right or left) which has the higher value is the side to which the person is windswept towards |
| 4 | Combined Hip External rotation / Abduction | Hip joint range of motion | Leg paddle and pelvic bridge | R ≠ L suggests asymmetry |
| 5 | Windswept Index (WI) | Difference between the left and right ABLAP<br>WI = R ABLAP – L ABLAP | calculation | Higher values represent greater postural asymmetry |

according to Goldsmith Indices of Body Symmetry guidelines [16]. Rater order was randomised using a randomisation application (http://www.randomisation.com/). For inter-rater reliability, raters were blinded to results of previous testing. An overhead hoist was used to transfer participants from wheelchair to plinth where the Goldsmith Indices of Body Symmetry measurement was performed. An additional assistant and family member/carer of the participant assisted in the process as required. A testing session typically lasted 45 min.

Control data: One rater was involved in a single session of data collection following documented Goldsmith Indices of Body Symmetry procedure [14]. Participants attended in pairs for measurement, and the assistant role was undertaken by the second participant. A testing session typically lasted up to 30 min.

### *Statistical analysis*

The SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for analysis. All calculations from raw data were performed according to Goldsmith Indices of Body Symmetry procedure (Table 1). Chest right/left ratio values were transformed to reflect magnitude of rotation rather than clockwise or anticlockwise direction. Normality of data was evaluated using visual inspection and skewness and kurtosis statistics, and descriptive statistics used to report Goldsmith Indices of Body Symmetry measures. Means and standard deviations for each Goldsmith Indices of Body Symmetry variable for data from adults with CP and control comparison were calculated. Medians and Interquartile ranges were calculated for data not following a normal distribution. Data that did not follow a normal distribution was transformed using the square root model [17] to enable further parametric data analysis.

Comparisons between adults with CP and control adults' demographic data were undertaken using Mann–Whitney *U* or Chi Square tests. Additional exploration of any impact of scoliosis surgery on Goldsmith Indices of Body Symmetry measurement was completed (Mann–Whitney *U*-test).

Adult with CP data was assessed for intra-rater reliability of individual Goldsmith Indices of Body Symmetry measurements (taken four times) using intraclass correlation coefficients (ICC [3,1]) and 95% confidence intervals [18] for each rater. A one-way repeated measures ANOVA was calculated to determine any difference between the four measures of individual Goldsmith Indices of Body Symmetry components for each rater. Inter-rater reliability for the CP participant data, using the mean of the four measures, was assessed using intraclass correlation coefficients (ICC [2,1]), with 95% confidence intervals [18,19]. A paired samples *t*-test was applied to determine any difference between the Goldsmith Indices of Body Symmetry measures by the two raters. Bland-Altman plots for each Goldsmith Indices of Body Symmetry variable were used to determine if any systematic differences across the range of values occurred between the two assessments completed by the two raters in the exploration of inter-rater reliability [20]. Sample size estimates for the inter-rater reliability study phase were based on methods of Walter et al. [21] which indicated that 27 participants would be required for a study with two observations per participant, assuming an expected ICC of 0.6, a null ICC of 0.2, a type-I error of 0.05, and a type-II error of 0.2. Level of significance for all analyses was set at $p < 0.05$.

## Results

After screening 166 potential participants, thirty adults with CP participated. Of the 136 excluded, 38 declined, 34 did not have a diagnosis of CP, 46 were not GMFCS Level IV or V, five had significant dystonia, and 13 had extreme cognitive/behavioural challenges. Forty-eight control adults participated in a single session of data collection. Demographic data describing all participants is in Table 2.

There was no significant difference in age nor gender distribution between adults with CP and control group ($p = 0.137$ and $p = 0.215$ respectively). Adults with CP had significantly lower weight and height than control group ($p < 0.0004$).

### *Goldsmith indices of body symmetry (descriptives) – adults with CP and control data*

In the CP population, all Goldsmith Indices of Body Symmetry variables were normally distributed, with exceptions being chest right left ratio and windswept index, which were not normally distributed. Transformations using square root model were utilised on these variables before conducting parametric analyses.

**Table 2.** Demographic data from all participants.

| Participant Demographics | Participants (CP) $n = 30$ | Participants (control) $n = 48$ |
|---|---|---|
| Age years[a], median (IQR); range | 21.00 (18.75, 24.25); 17.00–38.00 | 21.50 (20.00, 26.00); 18.00–34.00 |
| Gender (*n*)[b] | M - 17 F - 13 | M - 19 F - 29 |
| Living Situation | Home with family: 27<br>Community Residential Unit: 3 | Data not collected |
| Height cm[c], median (IQR); range | 151.50 (144.75, 157.25); 119.00–167.00 | 171.50 (165.25, 178.75); 151.00–192.00 |
| Weight kg[c], median (IQR); range | 46.9 (39.17, 59.27); 32.80–78.40 | 68.50 (60.25, 73.75 ); 51.00–100.00 |
| GMFCS Level (*n*) | Level IV 4<br>Level V 26 | N/A |
| CP subtype (*n*) | Spastic Quadriplegia 28<br>Spastic Diplegia 1<br>Dystonia 1 | N/A |
| Prior scoliosis surgery (*n*) | 10 | 0 |
| Communication (*n*) | Nil consistent communication 19<br>Communication device 2<br>Verbal 9 | N/A |

CP: cerebral palsy; F: female; GMFCS: Gross Motor Function Classification System;IQR: interquartile range; M: male; N/A: Not applicable.
[a]no significant difference between groups ($p = 0.137$).
[b]no significant difference between groups ($p = 0.215$).
[c]significant difference between groups ($p = 0.00$).

**Table 3.** Goldsmith Indices of Body Symmetry data for controls and adult with CP groups, with inter rater reliability Intra Class Correlations for adults with CP data.

| Goldsmith Indices of Body Symmetry Procedures | Normative Mean (SD) Range | CP Rater 1 Mean (SD) Range | CP Rater 2 Mean (SD) Range | ICC 95% CI *p* value | Paired t test CP data P value |
|---|---|---|---|---|---|
| **Chest Depth Width Ratio** | **0.62 (0.05)** | **0.66 (0.09)** | **0.67 (0.09)** | 0.95 | 0.470 |
| | 0.52–0.75 | 0.50–0.85 | 0.50–0.86 | 0.90–0.98 | |
| CP $n = 30$ | | | | 0.00 | |
| **Chest Right Left Ratio (Magnitude)** | **0.07 (0.05)** | **0.16 (0.13)** | **0.17 (0.14)** | 0.74 | 0.386 |
| | 0.00-0.27 | 0.01–0.56 | 0.03 – 0.57 | 0.45–0.87 | |
| | | *Median (IQR)* | *Median (IQR)* | 0.001 | |
| CP $n = 30$ | | *0.12 (0.05, 0.23)* | *0.13 (0.06, 0.26)* | | |
| **R ABLAP[a]** | **122.11 (7.96)** | **125.47 (15.43)** | **122.90 (14.70)** | 0.97 | 0.021* |
| CP $n = 27$[c] | 100.00–135.25 | 91.5–168.25 | 101.00–164.50 | 0.93–0.98 | |
| | | | | 0.00 | |
| **L ABLAP[a]** | **122.04 ( 8.89)** | **117.73 (16.48)** | **118.04 (12.90)** | 0.85 | 0.884 |
| CP n = 30 | 96.75–139.75 | 80.25–155.25 | 87.25–154.25 | 0.66–0.93 | |
| | | | | 0.00 | |
| **Combined R ER/ Abd[b]** | **53.61° (6.19°)** | **42.15°(18.08°)** | **43.96° (16.37°)** | 0.95 | 0.229 |
| | 35.00°-65.50° | 7.80°–79.00° | 15.50–78.00° | 0.88–0.97 | |
| CP $n = 28$[c] | | | | 0.00 | |
| **Combined L ER/ Abd[b]** | **55.79° (6.84°)** | **43.14° ( 15.59°)** | **45.63° (14.99°)** | 0.94 | 0.075 |
| | 41.75°69.00° | 2.25°–69.50° | 11.00°–71.50° | 0.87–0.97 | |
| CP $n = 29$[c] | | | | 0.00 | |
| **Windswept Index** | **3.59 (3.21)** | **19.02 (22.74)** | **17.19 (16.93)** | 0.91 | 0.900 |
| CP $n = 27$[c] | 0.00–14.25 | 0.75-81.00 | 0.75-66.50 | 0.80–0.96 | |
| | | *Median (IQR)* | *Median (IQR)* | 0.00 | |
| | | *11.25 (3.75, 16.50)* | *10.75 (4.00, 28.50 )* | | |

[a]ABLAP: Angle Between the Legs and Pelvis.
[b]ER/Abd: External Rotation/Abduction.
[c]Testing of the R ABLAP and combined hip ER/Abd, and calculation of the windswept index was not possible for 3 participants; one did not have classic wind-sweeping (rotation of the pelvis and legs in opposite directions as opposed to legs and pelvis moving together) [12] and two participants had significant fixed lower limb contractures that prevented measurement.
*Significant results $p < 0.05$.
Medians and IQR's are reported for variables that were not normally distributed. Square root transformations were used on these variables prior to ICC analysis.
Bold texts and values are used to highlight the Goldsmith procedures and the Mean and SD.

All individual components of the Goldsmith Indices of Body Symmetry for both adults with and without (control) CP are presented in Table 3. The results demonstrate that for most components, variability in measurements of adults with CP appears higher, with standard deviations more than double those observed for control participants, and both minimum and maximum scores beyond control range.

There was no significant difference in Goldsmith Indices of Body Symmetry outcomes for those who had received scoliosis surgery and those who had not ($p > 0.05$).

### *Inter-rater reliability – adults with CP*

Results for inter-rater reliability of the Goldsmith Indices of Body Symmetry using mean of four measures across identified variables for Raters 1 and 2 are shown in Table 3. The ICC [1,2] for all variables was $\geq$0.85, indicating excellent agreement for all Goldsmith Indices of Body Symmetry components. Paired samples t-test indicated no significant difference between raters ($p > 0.05$) for all procedures except R ABLAP ($p = 0.02$). Inspection of Bland Altman plots (Figure 1(a–e)) showed no systematic differences across the range of scores, with only between 1 (left external rotation/

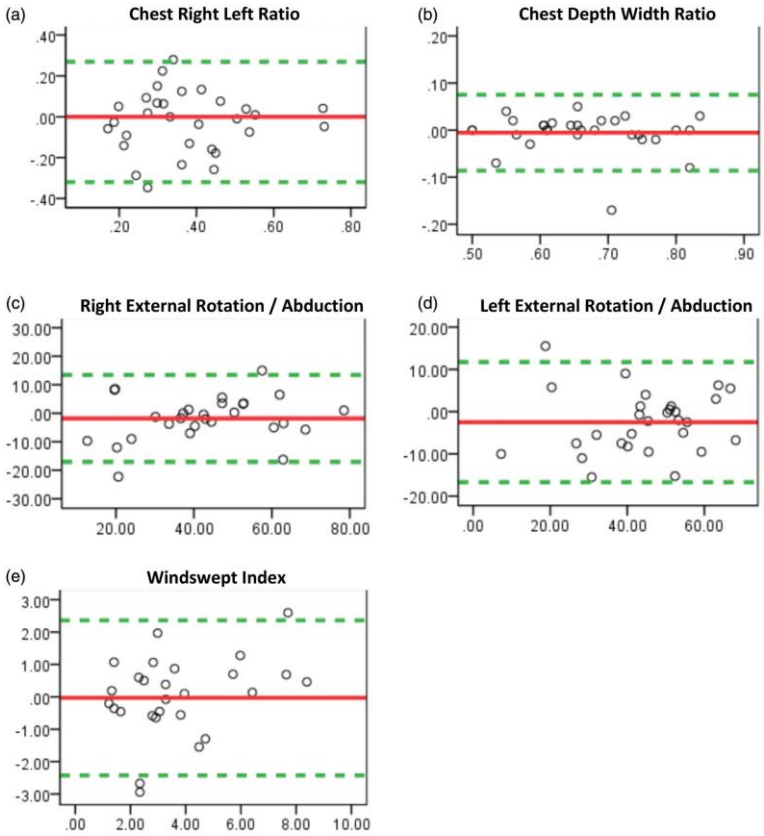


Figure 1. (a–e) Bland–Altman plots of agreement between rater scores. Y Axis: Differences in scores (rater 1 – rater 2), X Axis: Averages of rater 1 and rater 2. Solid line: Mean Difference; Dashed line: differences +/− 95%.

abduction) and 3 (windsweeping index) of all scores falling outside 95% limits of agreement.

The ICC [1,2] using the first measure only (rather than mean of four measures) for each rater for each variable similarly resulted in high levels of inter-rater agreement (>0.80) for all components of the Goldsmith Indices of Body Symmetry (Supplementary Table S1).

### *Intra-rater reliability – adults with CP*

Intra-rater reliability results of the four repeated measures contributing to calculation of Goldsmith Indices of Body Symmetry in adults with CP are shown in Supplementary Table S2. The ICC [1,3] for all measurements was ≥0.97 for both raters indicating an excellent level of intra-rater agreement. A one-way repeated measures ANOVA indicated no significant difference between the four measures for each rater ($p > 0.05$) for all except Left Hip ER/Abd for Rater 1 ($p = 0.017$ between measures 3 and 4). Partial eta squared values indicate a small to very small effect size for the single significant result.

## Discussion

This study is the first to explore the use of the Goldsmith Indices of Body Symmetry in adults with CP (GMFCS IV and V). In the present study we sought to establish the level of agreement within a single rater, and between two raters, when they measured the same variables in non-ambulant adults with CP using a standard protocol. The secondary aim of the study was to establish comparative control data for the Goldsmith Indices of Body Symmetry. Study outcomes demonstrated the Goldsmith Indices of Body Symmetry is a reliable tool for measurement of thoracic shape and symmetry, pelvic orientation and hip mobility and enables calculation of a Windswept Index in adults with CP classified as GMFCS Level IV or V. The intra-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with CP was excellent across all Goldsmith Indices of Body Symmetry domains. The inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with CP was similarly good to excellent across all Goldsmith Indices of Body Symmetry domains. Most Goldsmith Indices of Body Symmetry components had notably different distributions to those recorded from a control group.

Findings support the initial reliability study by Goldsmith and colleagues [12] and a later thesis [22] both which found the Goldsmith Indices of Body Symmetry to be a reliable measurement tool for postural asymmetry in primarily paediatric populations. The present study suggests similarly reliable outcomes can be achieved in adults with CP compared to those obtained in children using the Goldsmith Indices of Body Symmetry.

Comparison of control data to data from participants with CP demonstrated wide variability in the CP group, with minimum and maximum scores outside control range for most components. The range of scores for both the angle between legs and pelvis, and the combined hip external rotation/abduction in participants with CP varied greatly from person to person, indicating some participants had very little pelvic and hip mobility associated with fixed contractures and/or spasticity, and others had excessive mobility associated with hypotonus. As the Goldsmith Indices of Body Symmetry is not a measure of spasticity, testing movements were taken slowly to pain-free end of passive range and not to the point of a velocity dependent catch. Control data can be utilised to assist in Goldsmith Indices of Body Symmetry interpretation in people with CP, for example identifying scores that are more than two standard deviations higher or lower than control data means.

The literature provides clear definition of what constitutes a windswept hip deformity [2,13] yet measurement of this deformity is not standardised. A mathematical algorithm was developed by Young and colleagues [13] and later used in a modified version [9,23] whereby at least 50% difference in abduction, internal, and/or external rotation between left and right hips was defined as "windsweeping", a two dimensional outcome. The Goldsmith Indices of Body Symmetry Windswept Index offers a three-dimensional solution using combined measurement of hip mobility less the influence of the pelvic mobility, thus more accurately reflecting the functional problem. As expected, the control population's mean windswept index was close to symmetry (0) but the CP population showed a large departure from symmetry ($>0$). Contributing factors to an elevated windswept index may include an asymmetrical static sleeping posture with the influence of gravity on hip and knee flexion contractures causing lower limbs to fall to one side. The inability to change position and the influence of spasticity may also contribute in those with severe disability. Interestingly, the presence or absence of prior scoliosis surgery did not impact Goldsmith Indices of Body Symmetry results. This may be an area for future exploration. The relationship of Goldsmith Indices of Body Symmetry hip and pelvic measurements may enable ready monitoring of change over time and an opportunity to evaluate the effect of interventions.

According to the published testing protocol [16] each individual component of the Goldsmith Indices of Body Symmetry should be performed four times with data average and range calculated. Training in use and application of a standard protocol for instrument usage minimises error. Repeating the measurement four times may enable an aberrant result due to a momentary spasm or measurer error to be noted [12,16]. Despite exclusion of participants with extreme behavioural challenges and severe dystonia, this study was conducted on a very challenging participant population of adults with CP with a high incidence of spasticity and cognitive deficits. Varying results between measures due to spasms, pain and behavioural challenges would not be unusual in this population. Despite this, high intra-rater reliability across the four measures for each rater (ICC $\geq$ 0.98) was achieved. An acceptable intra-rater reliability (ICC $\geq$ 0.80) based on the first of the four measures only was also achieved. Thus the Goldsmith Indices of Body Symmetry remains a reliable measurement tool for participants who may not be able to tolerate four measures. A high inter-rater reliability (ICC $\geq$0.85) for almost all components of the Goldsmith Indices of Body Symmetry further confirms different yet experienced raters can undertake testing and subsequent re-testing with confidence in reliability.

### *Limitations*

A limitation of the study was that both raters were formally trained in the Goldsmith Indices of Body Symmetry. Further research is required to examine reliability in non-accredited therapists. It is also acknowledged that establishment of intra-rater reliability occurred within a single test session, using published Goldsmith Indices of Body Symmetry protocol of repeating measurements four times each. Although this resulted in assessor non-blinding, we would argue the need to reset participant's position and equipment between each measurement reduced any influence of prior measurement on subsequent measurement.

A further limitation was the inability to accurately capture pelvic obliquity with the tool, an abnormal tilt of the pelvic girdle that may occur in all three movement planes. It is associated with progressive scoliosis and hip dislocation, pain and necrosis of ischial tuberosities [24]. Pelvic obliquity is part of a triad, together with scoliosis and windswept hips, which is extremely difficult to treat once established [4]. The Goldsmith Indices of Body Symmetry captures elements of pelvic asymmetry through the pelvic bridge apparatus, specifically rotation in the frontal plane, however the contribution of pelvic obliquity is unable to be teased out from that of hip contracture. The potential to specifically measure pelvic obliquity using the Goldsmith Indices of Body Symmetry pelvic bridge requires additional investigation.

In some participants, severe lower limb contractures prevented complete measurement of all Goldsmith Indices of Body Symmetry elements. Also, potential participants with movement disorders such as severe dystonia, and extreme behavioural issues were excluded. In these cases alternate measurement processes remain challenging and require further investigation.

Finally, it is recognised that the Goldsmith Indices of Body Symmetry takes an impairment-focus to postural asymmetry measurement. The impact on a person's function, such as seating ability, is not described. As previously indicated, the PPAS describes static and dynamic posture of adults with CP using an activity-based focus [11]. Although the PPAS can detect postural asymmetry (present or absent) in a range of functional positions, it does not quantify posture, nor postural directional change over time. The inclusion of both the PPAS and Goldsmith Indices of Body Symmetry would enable comprehensive clinical assessment for ongoing monitoring and treatment planning for postural management of adults with CP.

## Conclusions

Many non-ambulant adults with CP experience asymmetry of thoracic cage, pelvis and hips which may progress after skeletal maturity. The Goldsmith Indices of Body Symmetry enables reliable quantification of this asymmetry. The provision of a Windswept Index, thoracic shape and symmetry, and combined influence of hip rotation and abduction/adduction provides valuable information regarding lifespan management of adults with CP. The Goldsmith Indices of Body Symmetry is a reliable measurement tool to measure and monitor asymmetry of the thorax, pelvis and hips in this population.

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## Disclosure statement

No potential conflict of interest was reported by the authors.

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# Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study

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ABSTRACT

*Background:* Pain in adults with cerebral palsy (CP) is commonly reported, with muscular and skeletal dysfunction resulting in postural asymmetry as potential contributors to multifactorial causes of pain. The relationship between pain and postural asymmetry of the thoracic cage, pelvis and hips in non-ambulatory adults with CP however is unknown, particularly in those with cognitive and communication limitations.
*Objective:* The primary aim of this study was to describe and quantify day and night pain in non-ambulatory adults with CP. Secondary aims were to investigate any relationship between pain and postural asymmetry and to describe current pain management strategies utilised.
*Methods:* Pain was measured using the Non Communicating Adult Pain Checklist (NCAPC). Posture was measured using the Goldsmith Indices of Body Symmetry (GIofBS) and radiographs. Correlations between pain scores and posture (GIofBS and radiographs) were assessed using non-parametric analysis. Information regarding pain management strategies was gained from medical records and carer interviews.
*Results:* Seventeen non-ambulatory adults with CP were recruited. High levels of day pain were experienced by $\geq$ 50% of participants with a high incidence of prescribed medications targeting pain. Strong positive correlations between day and night NCAPC scores, chest right left ratio and night pain, Cobb angle and day pain and between Cobb angle and night pain were evident.
*Conclusion:* The incidence and severity of pain in non-ambulatory adults with CP is high with postural asymmetry a potential contributor. Pain remains difficult to assess and manage in adults with significant cognitive and communication impairments and warrants further investigation.


## Introduction

Cerebral Palsy (CP) is a lifelong disability with most children now surviving into adult hood.[1] Pain has been reported in 62%–87% of all adults with CP[2] with greater pain associated with greater disability and is the most common secondary condition associated with CP.[3] Despite this, understanding pain in those with complex disability and significant communication and/or cognitive limitations remains challenging. A focus on complex healthcare needs, and specifically identifying and managing pain in adults with CP is not common, and may be overlooked due to communication difficulties, difficulties accessing health services and diagnostic overshadowing.[4]

Pain in those with CP is likely to be multifactorial, originating from acute and/or chronic system dysfunction such as skeletal, neuromuscular, respiratory and gastrointestinal origins. Muscular and skeletal issues which may contribute to postural asymmetry (i.e. asymmetry affecting thoracic cage, pelvis and hips) are two of several potential causes of pain in adults with CP,[2,5] with high proportions of non-ambulatory adults with CP experiencing hip dislocation, fixed hip/knee contractures and profound scoliosis.[6] Any relationship between postural asymmetry and pain is unknown in this population, as is the relationship between pain and postural dysfunction over a 24-h period.

Adults with CP (GMFCS IV and V) experience significant postural asymmetry affecting the thoracic cage, pelvis and hips[6] which may

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be associated with pain and cause activity limitation. Measurement of postural asymmetry utilising an impairment framework is an important aspect of management which, when coupled with a clinical assessment focused at an activity level (e.g. Posture and Postural Ability Scale)[7] provides a holistic postural assessment across the lifespan.

An extensive systematic review by Novak et al. (2020) revealed a lack of evidence for efficacy of interventions for management of pain in children with CP other than pharmacological agents targeting spasticity.[8] High level evidence exists to support the use of Botulinum Toxin, Intrathecal Baclofen and oral medications for spasticity reduction as a pain minimisation strategy.[9] The efficacy of non-pharmacological postural management interventions are less conclusive.[10] Conservative interventions aimed at managing postural asymmetry and potentially minimising pain include the prescription of assistive technologies (e.g. adaptive seating and bed positioning equipment) aimed at 24-h postural care, handling techniques and education of families and involved professionals.[11] However while the evidence regarding efficacy of these interventions in children is relatively limited[11,12] there is almost no published literature relating specifically to conservative pain interventions for adults with CP[2] whose needs across their lifespan may well be different to those of children. As a starting point, it would be worthwhile to explore the interventions currently being utilised for pain management in non-ambulatory adults with CP.

Engaging people with cognitive and communication limitations in research studies may be difficult,[13,14] and they may be unable to participate due to not meeting inclusion criterion. The Non Communicating Adult Pain Checklist (NCAPC)[15] has been developed to capture pain behaviours in adults with significant intellectual and developmental disabilities.[15] The scale explores both physiological and behavioural elements of pain and has satisfactory construct validity, internal consistency and sensitivity to pain alongside high reliability for this population[16] using proxy reporting. Despite well recognised limitations with proxy reporting[3,5] for those with severe cognitive and communication difficulties, proxy reporting may be the best available option.[17] The primary aim of this study was thus to describe and quantify day and night pain experienced using the NCAPC in non-ambulatory adults with CP. The secondary aims were to investigate any relationship between pain and postural asymmetry, and describe pain management strategies in this cohort. It was hypothesised that those with greater 24-h pain would also have greater postural asymmetry.

## Methods

### *Design*

This study is a component of an observational cohort study of non-ambulatory young adults with CP investigating a postural measurement tool.[18] The data regarding pain is previously unreported.

### *Participants*

Participants who had a diagnosis of CP classified at the non-ambulatory categories of Gross Motor Function Classification System (GMFCS) levels IV and V,[19] newly referred to the Young Adult Complex Disability Service (YACDS) during the period February 2017 to May 2019 met inclusion criteria and were invited to participate. Participants were excluded who had a significant movement disorder that rendered them unable to remain relatively still during postural testing, were pregnant, or had extreme behavioural issues that placed either testers or the participant at risk. Next of kin or a legal guardian of suitable participants were given study information and provided written consent for all participants due to severe cognitive impairment. Ethical approval was gained from St. Vincent's Hospital Melbourne Human Research Ethics Committee (HREC/16/SVHM/148).

### *Outcome measures*

Demographic and medical data was accessed from the participant's medical history and included gender, date of birth and GMFCS level. Control data (Table 2) was obtained from 48 young adults with no self-declared disability.[18]

### *Pain*

The Non Communicating Adult Pain Checklist (NCAPC) is an 18 item scale with each item rated from 0 to 3 referring to the frequency of the observed behaviour (0 = not at all, 1 = just a little, 2 = fairly often, 3 = very often). It describes six sub dimensions of pain behaviour: vocal reaction, emotional reaction, facial expression, body language, protective reaction, and physiological reaction.[15,16] A score of $\geq$10/54 is indicative of a high pain level.[16] As pain may vary between night and day due to factors including seating, bed positioning, spasticity and medications, both night and day time NCAPC were completed by the carer for a typical day/night in the week prior to the research appointment. Night pain scores were recorded either to reflect pain levels when tending to participant overnight, or at initial contact with the participant first thing in the morning. Carers were not expected to report on the participant's overnight pain unless it was usual care to review or tend to the participant overnight.

## Postural alignment

### *Anthropometric measurement*

The Goldsmith Indices of Body Symmetry (GIofBS) is a clinical measurement tool providing objective information on posture including thoracic shape and symmetry, pelvic orientation and hip mobility with demonstrated reliability in non-ambulatory adults with CP.[18] The GIofBS, according to standard testing procedure,[20] was used to collect a) Chest right left ratio (indicative of chest rotation); b) combined hip external rotation/abduction (left and right); and c) the Windswept index (indicative of degree of asymmetry between left and right pelvis/lower limbs). These measurements were selected as they have been shown to vary significantly in non-ambulatory adults with CP compared to those without disability.[18]

### *Radiographic measurement*

Antero-posterior (AP) radiographs of the pelvis and spine obtained within 12 months of the collection of the GIofBS data were

**Table 1**
NCAPC Sub-categories for day and night pain.

| NCAPC Subsets | NCAPC Day time | NCAPC Night time |
|---|---|---|
| | Median (IQR) range | Median (IQR) range |
| Vocal reaction/12 | 2 (0, 4) 0 - 10 | 0 (0, 3.5) 0 - 10 |
| Emotional reaction/6 | 1 (0, 2) 0 - 4 | 0 (0, 1.0) 0 - 4 |
| Facial expression/12 | 3 (0, 4) 0 - 10 | 1 (0, 4.5) 0 - 10 |
| Body language/6 | 2 (1, 3.5) 0 - 5 | 2 (0, 3.5) 0 - 6 |
| Protective reaction/12 | 2 (0.5, 3.5) 0 -12 | 0 (0, 2.5) 0–12 |
| Physiological reaction/6 | 1 (0, 2) 0 - 4 | 0 (0, 1.5) 0 - 4 |
| Total/54 | 12 (5.5, 15) 0 - 41 | 5 (1.5, 17.0) 0 - 43 |

**Table 2**
Descriptive data for Goldsmith Indices of Body Symmetry and Radiographs for participants with cerebral palsy.

| GIofBS Data | Median (IQR) Range (CP) |
|---|---|
| Windswept Index | 11.25 (6.50, 32.12) 1.25–81.00 |
| Chest Right Left Ratio | 0.08 (0.03, 0.16) 0.02–0.31 |
| Right Hip External Rotation/Abduction (degrees) | 44.75° (33.75°, 63.25°) 19.50°–79.00° |
| Left Hip External Rotation/Abduction (degrees) | 38.50° (29.62°, 47.50°) 2.25°–54.50° |
| **Radiographic Data** | **Median (IQR) Range (CP)** |
| Migration percentage[a] | 27.00% (14.00%, 37.50%) 0.00%–47.00% |
| Cobb angle (degrees)[b] | 23.00° (10.50°, 37.00°) 7.00°–65.00° |
| Pelvic obliquity (degrees) | 3.00° (2.00°, 11.50°) 0.00°–21.00° |

Control data for GIofBS (Median (IQR) range): Windswept Index 2.87,(1.0, 4.69) 0.00–14.25; Chest Right Left Ratio 0.05 (0.03, 0.09) 0.00–0.27; Right Hip External Rotation/Abduction (degrees) 53.12° (49.87°, 58.94°) 35.00°–65.50°; Left Hip External Rotation/Abduction (degrees) 56.37°(51.00°; 60.44°) 41.75°–69.00°.

[a] MP of ≤10% normal and MP ≥ 30% abnormal,.[23]

[b] scoliosis defined as Cobb angle ≥ 10°.[28]

reviewed and calculations of highest hip migration percentage (MP), a measurement of the amount of ossified femoral head not covered by the ossified acetabulum,[21] Cobb angle, spinal curvature measured in degrees[22] and pelvic obliquity, an abnormal tilt of the pelvis in the frontal plane were completed. This time frame was selected as it meets published recommendations for surveillance in those post skeletal maturity in the presence of abnormal radiographic findings.[23] All radiographic measurements were completed using tools within a picture archiving and communication system (Synapse™; Fujifilm Corp., Tokyo, Japan).

## Procedure

Participants attended an appointment at which they were measured by an experienced and trained physiotherapist (Level 3 Award in Measurement of Body Symmetry; Open College Network West Midlands) using the GIofBS Anatomical Measurement Instrument according to published practice.[20] Forty-five minutes was allocated for a testing session.

### *Pain management strategies*

The commonly prescribed medications and conservative interventions impacting spasticity and/or pain were identified via carer interview and researcher review of medical chart (routine correspondence between YACDS and General Practitioners) at the time of measurement. Spasticity medications were included in the data extractions due to the recognised relationship between spasticity and pain[2,24] and the difficulty in extrapolating the exact cause of pain in non-communicating adults with CP.

### *Statistical analysis*

The SPSS statistical software version 24 (SPSS Inc, Chicago, Illinois) was used for all quantitative analysis. Descriptive statistics were used to report demographic data, NCAPC measures (day and night), and GIofBS and radiographic measures. All continuous variables were analysed for distribution and skew. The GIofBS chest right left ratio was adjusted to reflect the magnitude of the measure rather than a positive or negative value (indicative of rotation in a clockwise or anticlockwise direction). Pain management strategies were categorised and proportions counted.

Non Parametric analysis (Spearman's rho) was undertaken to assess any correlation between total day and night pain scores. The correlation between pain scores and both GIofBS constructs and radiographs (MP, Cobb angle and pelvic obliquity) were also explored. The strength and direction of any relationships were established according to Cohen,[25] where r = 0.1 to 0.29 = small, r = 0.30 to 0.49 = medium and r = 0.50 to 1.0 = large effect size. Significance was set at $p < 0.05$.

## Results

After screening 25 potential participants, 17 adults with CP (10 males), median age 19 (range 17–33 years), GMFCS Level IV/V (n = 1/16) consented to participate (via carer/guardian). Nil participants were able to self-consent. Of those who did not consent, two reported transport issues, five declined and one had significant behavioural challenges.

NCAPC total and sub scores for day and night pain are provided in Table 1. Day and night NCAPC total scores (median, IQR, outliers) are illustrated in Fig. 1. Ten participants displayed high levels of pain behaviours (scores ≥10)[16] for day pain. Six participants displayed high levels of pain behaviours (scores ≥10)[16] for night pain. The NCAPC subcategory of 'body language' was rated highest for both day and night pain (median 2/6; range 0–5 day; range 0–6 night). A strong positive correlation between total day and night NCAPC scores was found (rho = 0.76, p = 0.000).

GIofBS outcomes and radiographic data (median, IQR, range) are presented in Table 2 for CP participants and healthy adult controls.[18]

### *Relationship between pain and posture*

GIofBS outcomes: Other than a significant correlation between chest right left ratio (chest rotation) and night pain (rho = 0.521; p = 0.032), all remaining correlations between pain and GIofBS outcomes were not strong and failed to reach significance (Table 3).

Radiographic outcomes: Other than a significant correlation between the Cobb angle and day pain (rho = 0.611, p = 0.027), and between the Cobb angle and night pain (rho = 0.593, p = 0.033) all remaining correlations between radiographic measures and pain measures were not strong and failed to reach significance (Table 3).

### *Interventions*

Prescribed medications, assistive technology relating to postural management and NCAPC totals scores are presented for individual participants in Table 4. Of the prescribed medications, 13 participants were taking medications for reducing spasticity or spasm, and five participants were prescribed gabapentin which may be recommended for spasticity/dystonia, but is not a first line treatment. Of the four participants with no prescribed medications targeting pain, two had marked pain (scores ≥10) and two had minimal pain. All participants were seated in customised wheelchairs yet 12 were awaiting new or modified seating systems. Only

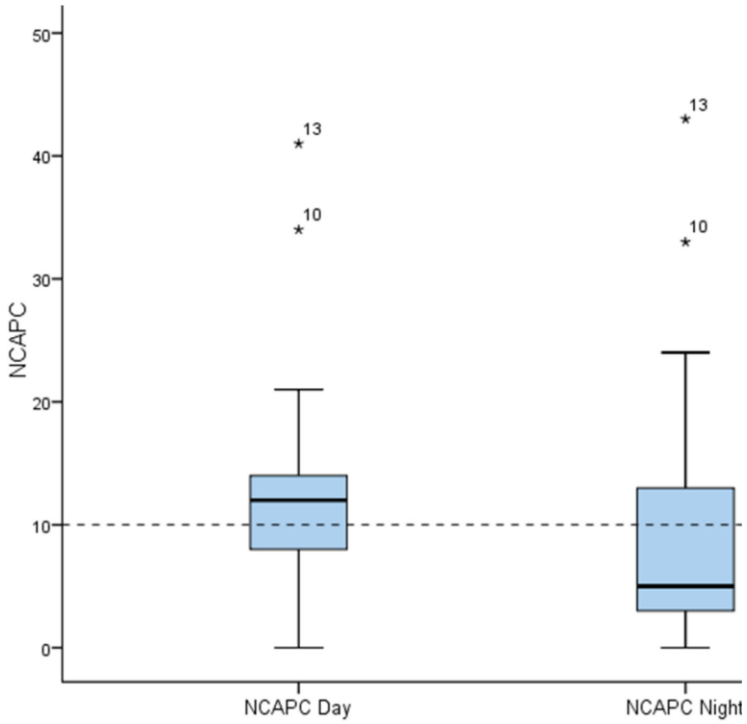


**Fig. 1.** Comparison of day and night NCAPC total scores. Box contains median (centre line) and 50% of cases (Q1 – Q3). Outliers are indicated by asterisks. The highest and lowest scores, excluding outliers, are represented by the whiskers. Dotted line indicates a value of 10, indicative of severe pain in the NCAPC.[16]

one participant had bed positioning postural supports in place, with nine participants waiting for prescribed bed positioning equipment.

**Table 3**
Correlations between NCAPC pain (day and night), and GlofBS or radiographic outcomes.

| GlofBS Correlations | rho value | P value |
|---|---|---|
| Windswept Index and NCAPC Day | 0.304 | 0.236 |
| Windswept Index and NCAPC Night | 0.373 | 0.140 |
| Chest RL Ratio and NCAPC Day | 0.419 | 0.094 |
| Chest RL Ratio and NCAPC Night | 0.521 | 0.032* |
| Right Hip ER/Abd and NCAPC Day | 0.188 | 0.470 |
| Right Hip ER/Abd and NCAPC Night | 0.321 | 0.208 |
| Left Hip ER/Abd and NCAPC Day | −0.224 | 0.388 |
| Left Hip ER/Abd and NCAPC Night | −0.163 | 0.531 |
| **Radiographic Correlations** | | |
| Highest MP and NCAPC Day[a] | 0.023 | 0.933 |
| Highest MP and NCAPC Night | −0.118 | 0.665 |
| Cobb angle and NCAPC Day[b] | 0.611 | 0.027* |
| Cobb angle and NCAPC Night | 0.593 | 0.033* |
| Pelvic Obliquity and NCAPC Day[c] | 0.327 | 0.216 |
| Pelvic Obliquity and NCAPC Night | 0.135 | 0.617 |

NCAPC: Non Communicating Adult pain Checklist, RL: Right left, ER/Abd: External rotation/Abduction, MP: Migration Percentage, *significant at $p < 0.05$.
[a] n = 16.
[b] n = 13.
[c] n = 16 due to poor radiograph quality.

## Discussion

This study explored the assessment of pain in cognitively impaired, non-ambulatory adults with CP attending a multidisciplinary specialist transition service, the relationship between pain and postural asymmetry and pain management strategies employed. Proxy reporting of pain revealed that more than half the participants experienced high levels of day pain (NCAPC $\geq$10)[16] as may be expected in a cohort most vulnerable to issues that contribute to pain.

Assessing pain in adults with significant physical, cognitive and communication impairments presents a challenge. The proxy reporting of pain in this study revealed that all except one participant displayed pain behaviours during the day and all except four participants demonstrated some degree of night pain. The true incidence of pain in the participants is unknown however due to the limitations of proxy reporting,[3,5] yet suitable alternatives are not available for those with severe communication and or cognitive disabilities. In the paediatric literature, under-reporting of pain in children with complex disabilities results in the potential for greater risk of pain neglect and management.[26] Reliance on interpreting behavioural aspects of pain behaviour in the NCAPC such as body language (rated more highly by proxies in this study), may be more indicative of the underlying neurological pathology rather than actual pain[26], or personal experience of pain by the proxy. Although the majority of study participants displayed pain, the reporting of pain to health care practitioners in the presence of motor impairment, cognitive and communication dysfunction is

**Table 4**
Pharmacological and conservative management interventions.

| Participant | Age | Baclofen[1] | Gabapentin[2] | Artane[3] | Benzodiazepines[4] | Wheelchair and seating system[5] | Night time positioning system | NCAPC Day Total/54 | NCAPC Night Total/54 |
|---|---|---|---|---|---|---|---|---|---|
| 1 | 19 | ✓ | ✓ | | | * | | 0 | 0 |
| 2 | 18 | ✓ | | ✓ | ✓ | * | # | 14 | 11 |
| 3 | 19 | | | | | * | | 1 | 3 |
| 4 | 18 | ✓ | ✓ | | | * | # | 10 | 4 |
| 5 | 19 | ✓ | ✓ | | | * | | 14 | 6 |
| 6 | 20 | ✓ | ✓ | | ✓ | * | # | 9 | 0 |
| 7 | 33 | ✓ | | | | * | # | 3 | 3 |
| 8 | 17 | | | | | * | # | 16 | 24 |
| 9 | 17 | ✓ | | ✓ | ✓ | * | | 13 | 0 |
| 10 | 18 | | | | ✓ | * | ✓ | 34 | 33 |
| 11 | 21 | ✓ | | | | ✓ | | 13 | 3 |
| 12 | 18 | | | | ✓ | ✓ | | 8 | 5 |
| 13 | 19 | | | | ✓ | * | # | 41 | 43 |
| 14 | 18 | ✓ | ✓ | | ✓ | * | # | 12 | 21 |
| 15 | 19 | ✓ | | | ✓ | ✓ | # | 9 | 8 |
| 16 | 19 | | | | | ✓ | | 1 | 0 |
| 17 | 28 | | | | | * | # | 21 | 13 |

Drug management: [1]Baclofen: spasticity and pain management, [2]Gabapentin: anticonvulsant and pain management, [3]Artane: spasm management, [4]Benzodiazepines: spasm management, [5]All participants used customised wheelchairs.
✓Equipment utilised and meets current needs, * Equipment utilised yet insufficient for current needs, # Equipment requirement identified but not yet provided.

not common[26] and may require a concerted effort by healthcare practitioners to investigate thoroughly.

The strong correlation between night and day pain scores is suggestive of a consistent pain picture for participants irrespective of time and postural position (e.g. sitting versus lying down). This may suggest that pain is arising from intrinsic musculoskeletal distortion and subsequent impact on the underlying body structure and function.

The radiographic measures of Cobb angle and the GIofBS measure of Chest right left ratio both had strong correlations to pain. Postural asymmetry resulting in a progressive body shape distortion of the spine and thoracic cage will impact underlying cardiorespiratory and gastrointestinal organs. This not only impacts function, but also increases the likelihood of pain due to distortion of the organs and altered function (i.e. pain associated with constipation or bladder dysfunction).[3] With changes to the alignment of the spinal column, as measured by the Cobb angle, the somatosensory system is likely to be impacted at the level of the intervertebral foramen where the nerve roots enter and leave the spinal cord resulting in pain or sensory changes to the affected areas. Coupled with a dysfunctional truncal musculoskeletal system, the inability to change posture and gravity may further exacerbate malalignment and pain. Participants in this study were experiencing significant and established postural asymmetry which is not uncommon in adults functioning at GMFCS levels IV and V,[6] as compared to a control group without disability (Table 2). In this study many participants were using inadequate postural support both in wheelchairs and in bed which may have contributed to the overall high pain picture. Seating is very challenging for those with postural asymmetry of the thoracic cage and spine and it is noteworthy that 12 participants were awaiting new seating systems and thus potentially experiencing pain due to current inadequate postural support. A lack of night time postural support was also evident, potentially contributing to night pain with nine participants awaiting funding for appropriate customised bed positioning equipment. Although measures of windswept hips had only a moderate non-significant correlation to day and night pain (rho = 0.304 and 0.373 respectively), in conjunction with scoliosis and pelvic obliquity, this creates a disabling postural asymmetry[27] presenting challenges for seating and bed positioning. However, it is difficult to ascertain the specific cause of pain in this population due to the interrelated nature of the pelvis, hips and spine in conjunction with limited communication. In this small sample it is of interest that pain appeared greater during the day when participants were sitting and gravity was impacting postural alignment. Further investigation is required to identify the impact of assistive technology (seating and bed positioning) on observed pain behaviours. Repeating the NCAPC after customised postural support delivery to ascertain any impact on pain might be valuable.

The majority of participants were taking prescribed medications for spasticity or spasm, which may reduce musculoskeletal pain. The routine use of a pain measurement tool, such as the NCAPC, pre and post staged changes to medications (trial and error), may enhance pain management in adults with CP, although it is acknowledged that NCAPC does not allow the identification of pain source. Further investigation into the exact cause of pain in adults with severe and complex disabilities is required to enable lifespan planning and evaluation of efficacy of interventions.

## Limitations

In this preliminary exploratory study, the use of a participant group who were referred from a tertiary paediatric healthcare facility to a specialist adult transition clinic may not be truly representative of adults with CP being managed in the community and thus results should be extrapolated with caution. Also, the sample size is relatively small, larger samples may reveal additional significant correlations between pain and postural measures.

A further study limitation was the inability to identify whether gabapentin was prescribed for the anticonvulsive or neuropathic pain relieving effects or both. Finally, it is acknowledged that measures of pain were sought from a ‘moment in time’, that is, a typical day and night and may not necessarily be reflective of a participant’s chronic pain behaviours over a longer time period.

## Conclusion

The incidence and severity of pain in non-ambulatory adults with CP is high. Postural asymmetries may be associated with pain. Pain remains difficult to assess in adults with significant cognitive and communication impairments. Further investigation into assessment and management strategies for the lifespan management of non-ambulatory adults with CP is warranted.

## Ethical considerations

The authors have no ethical issues to report and the study has received ethics approval.

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## Declaration of competing interest

None.

The manuscript has not been published and will not be submitted elsewhere for publication while being considered by the journal Disability Health Journal. No prior presentation of the content has occurred.

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# Appendix D: Chapter Declarations

# Declaration of thesis chapter 3

**Publication title: Postural asymmetry in non-ambulant adults with cerebral palsy: A scoping review**

In the case of the publication, **"Postural asymmetry in non-ambulant adults with cerebral palsy: A scoping review"**, the nature and extent of my contribution to the work was the following:

| **Nature of contribution** | **Extent of contribution (%)** |
|---|---|
| Led the conception, methodological design, search strategy, study selection quality and risk of bias assessment, data extraction, analysis, synthesis, and preparation and drafting of manuscript for publication. | 70% |

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the candidate's and co-authors' contributions to this work.

| | | |
|---|---|---|
| **Candidate's signature** | Signature | 23/03/2021 |
| **Prue Morgan Co-author's Signature** | Signature | 18/03/2021 |
| **Kim Brock Co-author's Signature** | Signature | 01/03/2021 |

# Declaration of thesis chapter 5

**Publication title: The intra and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy**

In the case of the publication, **"The intra and inter-rater reliability of the Goldsmith Indices of Body Symmetry in non-ambulant adults with cerebral palsy",** the nature and extent of my contribution to the work was the following:

| **Nature of contribution** | **Extent of contribution (%)** |
|---|---|
| Led the conception, methodological design, data collection, data analysis, and preparation and drafting of manuscript for publication | 65% |

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the candidate's and co-authors' contributions to this work.

| | |
|---|---|
| **Candidate's signature** | Signature 23/03/2021 |
| **Prue Morgan Co-author's Signature** | Signature 18/03/2021 |
| **Kim Brock Co-author's Signature** | Signature 01/03/2021<br>01/03/2021 |
| **Emma Fredrickson Co-author's Signature** | Signature 01/03/2021<br>Signature, date |

# Declaration of thesis chapter 7

**Publication title: The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy.**

In the case of the publication, "**The relationship between radiographic and anthropomorphic measurements of deformity of the thorax, hips and pelvis in adults with cerebral palsy**", the nature and extent of my contribution to the work was the following:

| **Nature of contribution** | **Extent of contribution (%)** |
|---|---|
| Led the conception, methodological design, data collection, data analysis, and preparation and drafting of manuscript for publication | 70% |

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the candidate's and co-authors' contributions to this work.

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|---|---|
| **Candidate's signature** | Signature 23/03/2021 |
| **Prue Morgan Co-author's Signature** | Signature 18/03/2021 |
| **Kim Brock Co-author's Signature** | Signature 01/03/2021 |

# Declaration of thesis chapter 9

**Publication title: Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study**

In the case of the publication, "**Progression of Postural Asymmetry in Young Adults with Cerebral Palsy who are Non-ambulant: An exploratory study**", the nature and extent of my contribution to the work was the following:

| **Nature of contribution** | **Extent of contribution (%)** |
|---|---|
| Led the conception, methodological design, data collection, data analysis, and preparation and drafting of manuscript for publication | 70% |

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the candidate's and co-authors' contributions to this work.

| | | |
|---|---|---|
| **Candidate's signature** | Signature | 23/03/2021 |
| **Prue Morgan Co-author's Signature** | Signature | 18/03/2021 |
| **Kim Brock Co-author's Signature** | Signature | 01/03/2021 |

# Declaration of thesis chapter 11

**Publication title: Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study**

In the case of the publication, **"Pain and its relationship with postural asymmetry in adults with cerebral palsy: A preliminary exploratory study",** the nature and extent of my contribution to the work was the following:

| **Nature of contribution** | **Extent of contribution (%)** |
|---|---|
| Led the conception, methodological design, data collection, data analysis, and preparation and drafting of manuscript for publication | 70% |

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the candidate's and co-authors' contributions to this work.

| | | |
|---|---|---|
| **Candidate's signature** | Signature | 23/03/2021 |
| **Prue Morgan Co-author's Signature** | Signature | 18/03/2021 |
| **Kim Brock Co-author's Signature** | Signature | 01/03/2021 |