Prescribing upper limb orthoses for children with cerebral palsy

Exploring decision making and a hand deformity classification to guide orthosis prescription

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Declaration

This thesis contains no material that has been extracted in whole or in part from a thesis that I have submitted towards the award of any other degree or diploma in any other tertiary institution.

No other person's work has been used without due acknowledgment in the main text of the thesis.

All research procedures reported in the thesis received the approval of the relevant Ethics/Safety Committees.

[redacted]

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Abstract

Hand deformities in children with cerebral palsy (CP) result from an imbalance between wrist, finger and thumb musculature secondary to central neurological disturbances. Upper limb orthotic intervention aims to minimise muscular imbalance to improve function and/or prevent the progression of deformity from becoming fixed and painful. A lack of clarity in literature regarding upper limb orthosis prescription has led to insufficient evidence to support or refute the efficacy of this intervention. The paucity of evidence may result in part from difficulty researching a complex and multi-faceted intervention. This doctoral program aimed to explore clinicians' decision making when prescribing upper limb orthoses for children with CP, and investigate the validity and reliability of a hand deformity classification designed to aid clinical decision making.

A systematic review (Study 1) examined 18 intervention studies. Study 1 found that the connection between the reported rationale for orthosis prescription and the effect described was not transparent nor well defined. A child's manual ability and movement of the wrist, fingers and thumb were not described or considered when prescribing upper limb orthoses. Consequently, a Q methodology study (Study 2) explored what guides an occupational therapist's clinical decision making when prescribing upper limb orthoses for children with CP. Study 2 yielded three viewpoints: (i) the potential effect of the orthosis; (ii) biomechanical presentation; and (iii) the client-therapist relationship. These viewpoints were supported by the primary regard occupational therapists place on the client's goals when deciding on upper limb orthosis prescription. There is strong evidence for client-centred approaches that value the therapeutic relations and these perspectives are not unique to orthosis prescription. The challenge for clinicians and researchers in this field of enquiry is to connect the purpose and potential effect of an orthosis as informed by the biomechanical presentation of the client.

The Neurological Hand Deformity Classification (NHDC), previously known as the Wilton Classification of Deformities of the Hand in the Presence of Neurological Dysfunction, was originally designed in 1986. The NHDC was developed to guide a therapist's decision making of upper limb intervention based on the dynamic interplay of the wrist and hand musculature. A lack of procedure to classify hand deformity using the NHDC was identified in 2014 following an initial reliability study. As part of this doctoral program a manual and website were developed to describe a standardised procedure for the NHDC, to ensure consistency in observation and recording of hand deformity (Chapter 5). The NHDC provides seven categories for classifying

hand deformity. The categories are differentiated by the presence or absence of active wrist and finger movement, relative to the flexed or extended wrist position. For the NHDC to be useful, evidence of validity and reliability is essential. In Study 3 (Chapter 6), construct validity of the NHDC was established through comparison of hypothesised and actual relationships between the NHDC and the body function and structure and the activity domains of the International Classification of Functioning, Disability and Health. Reliability of the NHDC was assessed by calculating agreement between repeated measures (test–retest) and paired raters (inter-rater). Expected relationships between the NHDC and other measures, stability between repeated measures, and acceptable inter-rater agreement provided confidence in classifying hand deformity with the NHDC. Case studies (Chapter 7) illustrate the clinical application of the NHDC. The NHDC provides clinicians with a framework to structure and organise the presentations of hand deformity in children with CP. The case studies were used as a pilot to test the usefulness of the classification in supporting orthotic prescription.

The research findings have the potential to: provide a valid and reliable tool for clinicians to classify hand deformity based on muscular imbalance; support clinicians' decision making regarding upper limb intervention according to an individual's classified hand deformity; and contribute to guidelines for upper limb orthosis prescription. Ultimately, the goal is to improve the management and outcomes of upper limb intervention for children with CP.

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Abbreviations

ABILHAND	ABILHAND-Kids questionnaire
ACROBAT-NRSI	A Cochrane risk of bias assessment tool: for non-randomised studies of intervention
AHA	Assisting Hand Assessment
AHTA	Australian Hand Therapy Association
AP	Adductor Pollicis
AROM	Active range of motion
ASHT	American Society of Hand Therapists
AusACPDM	Australasian Academy of Cerebral Palsy and Developmental Medicine
BFMF	Bimanual Fine Motor Function Classification
Box and Blocks	Box and Blocks Test of Manual Dexterity
CARE	Case report guidelines
CFCS	Communication Function Classification System
CMC	Carpometacarpal
CO-OP	Cognitive Orientation to daily Occupational Performance
COPM	Canadian Occupational Performance Measure
COSMIN	Consensus-based standards for the selection of health measurement instruments
CP	Cerebral palsy
DIP	Distal interphalangeal
DPA	Dynamic Positional Analysis (component of the Shriner's Hospital Upper Extremity Evaluation)
ECRB	Extensor Carpi Radialis Brevis
ECRL	Extensor Carpi Radialis Longus
ECU	Extensor Carpi Ulnaris
EPL	Extensor Pollicis Longus
FCR	Flexor Carpi Radialis
FCU	Flexor Carpi Ulnaris
FDP	Flexor Digitorum Profundus
FDS	Flexor Digitorum Superficialis
FPB	Flexor Pollicis Brevis
FPL	Flexor Pollicis Longus
GAS	Goal Attainment Scaling

GMFCS	Gross Motor Function Classification System
HIPM	Hypertonicity Intervention Planning Model
HREC	Health Research Ethics Committee
ICF	International Classification of Functioning, Disability and Health
ICF-CY	International Classification of Functioning, Disability and Health Child and Youth Version
IP	Interphalangeal
ISO	International organization for standardization
iWHOT	Infant Wrist Hand Orthosis Trial
MACS	Manual Ability Classification System
MAS	Modified Ashworth Scale
MCP	Metacarpophalangeal
MIT	Minimising Impairment Trial
MTS	Modified Tardieu Scale
NHDC	Neurological Hand Deformity Classification
PEDI-CAT	Pediatric Evaluation of Disability – Computer Aided Test
PEDro	Physiotherapy Evidence Database scale
PIP	Proximal interphalangeal
PL	Palmaris Longus
PRISMA	Preferred reporting items for systematic reviews and meta-analyses
PROM	Passive range of motion
PROSPERO	International Prospective Register of Systematic Reviews
QUEST	Quality of Upper Extremity Skills Test
RCT	Randomised Controlled Trial
RoBiNT	Risk of bias in n of 1 trials scale
ROM	Range of motion
SHUEE	Shriner's Hospital Upper Extremity Evaluation
STROBE	Strengthening the reporting of observational studies in epidemiology

List of Publications

Publication 1

Garbellini, S., Robert, Y., Randall, M., Elliott, C., & Imms, C. (2018). Rationale for prescription, and effectiveness of, upper limb orthotic intervention for children with cerebral palsy: a systematic review. *Disability and Rehabilitation*, 40 (12)1361–1371. Published online March 2017.

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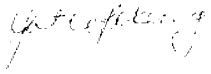
Study selection, quality assessment and data extraction Review of draft manuscript pre-submission	10%	 11/06/2020
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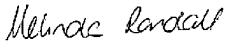
Development and review of study protocol Discussion of content and findings Review and editing of draft manuscript pre-submission	5%	 10/06/2020
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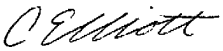
Development and review of study protocol Discussion of content and findings Review and editing of draft manuscript pre-submission	5%	 11/06/2020
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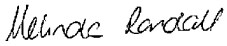
Development and review of study protocol Discussion of content and findings Review and editing of draft manuscript pre-submission	5%	 10/06/2020
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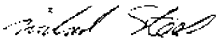
Development and review of study protocol Data analysis and interpretation Discussion of content and findings Review and editing of draft manuscript pre-submission	10%	 10/06/2020
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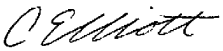
Development and review of study protocol Discussion of content and findings Review and editing of draft manuscript pre-submission	5%	 11/06/2020
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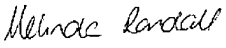
Discussion of content and concepts Review and editing of draft manuscript pre-submission	5%	 10/06/2020
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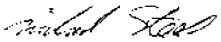
Discussion of content and concepts Review and editing of draft manuscript pre-submission	5%	 11/06/2020
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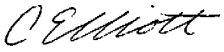
Discussion of content and concepts Review and editing of draft manuscript pre-submission	5%	 11/06/2020
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1.1 Overview

Prescribing upper limb orthoses for children with cerebral palsy (CP) is multi-faceted and can challenge even the most experienced therapist. There is little evidence about the effectiveness of upper limb orthotic intervention and a lack of domain-specific guidelines to assist therapists' decision making (Copley & Kuipers, 2014b; Jackman et al., 2014b; Novak et al., 2013). Despite low level evidence supporting the effectiveness of upper limb orthotic intervention (Novak et al., 2013; 2020) orthoses continue to be used in clinical practice.

Lannin and Ada (2011) proposed that an understanding of the capabilities and limitations of each orthosis, and clearly identifying the reason for orthotic intervention, would provide a basis for clinical decision making and potentially reduce controversy surrounding its use. This focus is on the orthosis itself, which is only one component to be considered when prescribing upper limb orthoses. The very presence of an orthosis can do harm (Brand & Hollister, 1999). A critical part of decision making for upper limb orthosis prescription is the therapist's: understanding of the capabilities and limitations of each orthosis; identification of deforming forces on the individual client's wrist and hand; and consideration, if indicated, of how to resolve these forces with the appropriate orthotic intervention. The problem of lack of evidence might sit with the difficulty in researching a complex intervention and the possible prescription of orthoses for children who are not suitable for, and therefore do not benefit from, the intervention.

The focus of this doctoral program was to:

1. Explore clinician's decision making when prescribing upper limb orthoses for children with CP.
2. Investigate the validity and reliability of a hand deformity classification designed to aid clinical decision making when prescribing appropriate upper limb interventions.

The overarching objective is to improve outcomes of upper limb intervention for children with CP. The ability to accurately classify hand deformity may be particularly helpful in identifying young children who might need a variety of upper limb interventions to maintain range of motion (ROM) and develop functional hand use. A reliable tool to classify hand deformity may provide clinicians with a structure to analyse patterns of dynamic movement, assist in planning intervention and add to practice guidelines for use of upper limb orthoses based on hand deformity classification.

This chapter provides:

- an overview of CP and its clinical presentation in the upper limb;
- a description of possible musculoskeletal change;
- the use of upper limb orthotic intervention aimed at managing this change;
- a summary of upper limb classifications to differentiate between different clinical presentations;
- an introduction to hand deformity classifications;
- the theoretical frameworks used to guide the doctoral program; and
- a description of the aims and objectives reported in this thesis.

1.2 Cerebral palsy and its clinical presentation in the upper limb

Childhood physical disability is mostly represented by children with CP, occurring in approximately two in every 1000 live births (Australian Cerebral Palsy Register, 2013; Reddihough & Collins, 2003). An accepted definition of what constitutes CP, used by CP Surveillance Programs in the United Kingdom, Europe and Australia (Smithers-Sheedy et al., 2014) is:

> A group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems (Rosenbaum et al., 2007; p 9).

Movement control is dependent on the planning and organisation of the movement by the central nervous system (Eliasson, 2005). Impaired voluntary movement control and involuntary muscle contraction, caused by lesions to upper motor neurons of descending spinal tracts in the brain and spinal cord, are referred to as an upper motor neuron syndrome (Esquenazi et al., 2013; Ivanhoe & Reistetter, 2004; Mayer et al., 1997; Sanger et al., 2006). In children, upper motor neuron syndromes will usually be associated with a diagnosis of CP (Graham & Selber, 2003). The upper motor neuron syndrome has both positive features (spasticity, hyper-reflexia, clonus and co-contraction) and negative features (loss of dexterity and motor control, weakness, poor coordination and sensory deficits) (Chin et al., 2005; Copley & Kuipers, 2014b; Ivanhoe & Reistetter, 2004; Sanger et al., 2006). These result in an imbalance of muscle activity in the arm and hand, inadequate joint stability and reduced sensory and motor experiences (Bartlett & Palisano, 2000; Kreulen et al., 2006; Morris, 2002; Scholtes et al., 2006).

The clinical presentation and predominant movement patterns of the child with CP are dictated by the interaction of positive and negative features of the upper motor neuron syndrome and differ between individuals (Koman et al., 2008). CP is classified by topographical distribution (unilateral or bilateral limb involvement) and motor type or movement abnormality (spastic, dyskinetic, ataxic and mixed movement disorders) (Cans, 2000; Gainsborough et al., 2008; Graham, 2005; Howard et al., 2005; Stewart & Harvey, 2018). In a consecutively sampled cross sectional survey exploring the prevalence and pattern of upper limb involvement of children with CP, Makki et al. (2014) found that upper limb involvement was seen in 83% of the children. The level of upper limb impairment was related to topology and motor type. Those children with total body involvement and dystonia and athetosis had poorer function (Makki et al., 2014).

Movement patterns of the upper limb vary according to the severity of the classified motor type (such as spasticity or dystonia), the distribution of muscles affected, and age (Makki et al., 2014). The most common upper limb movement patterns are internal rotation at the shoulder, elbow flexion, forearm pronation, wrist and finger flexion, and thumb-in-palm (Burtner et al., 2008; Casey & Kratz, 1988; Chin et al., 2005; Eliasson et al., 1998; Kanellopoulos et al., 2009; Koman et al., 2008; Kreulen et al., 2006). This characteristic upper limb posture seen in children with CP could be attributed to a combination of changes in muscular control and musculoskeletal components (de Bruin et al., 2013).

Joints and muscles are the effectors of planned movements required for daily activities (Eliasson, 2005). In children with CP, imbalance of muscle activity and changes in musculoskeletal structures can reduce motor experiences and impair upper limb motor development (Yasukawa & Cassar, 2009). Even though the primary lesion is in the central nervous system, significant musculoskeletal changes occur secondary to the lesion (Pontén et al., 2005). At birth, most children with CP do not have musculoskeletal deformity and exhibit full passive range of motion (Graham, 2004; Hedberg-Graff et al., 2019). Musculoskeletal change develops gradually and sometimes is not recognized until it is pronounced, with adolescents and adults presenting with joint deformity and fixed contractures (Chin et al., 2005; Hedberg-Graff et al., 2019). Therefore, the associated musculoskeletal deformity that occurs is progressive (Basu et al., 2014; Chin et al., 2005; Georgiades et al., 2014; Graham, 2004; Graham & Selber, 2003).

1.3 Secondary musculoskeletal change

Children with CP may present with secondary musculoskeletal problems, including: shortening of muscle fibres, deformity and contractures. These problems develop and progress as a result of growth, spasticity, the effects of gravity, and ageing (Chin et al., 2005; de Bruin et al.,

2013; Fulford & Brown, 1976; Graham & Selber, 2003; Hanna et al., 2003; Rosenbaum et al., 2007; Vaz et al., 2006). Muscle shortening may occur due to an inability to move a joint through its full range of motion. In addition, as the tissues adapt to functional demands and atypical movement patterns, changes in connective tissue significantly contribute to the development of hand deformities and contractures (Tardieu & Tardieu, 1987; Vaz et al., 2006; Wilton, 2013b).

Hand deformity is defined as a structural deviation of the wrist, hand, fingers and thumb from normal size, shape or alignment (Farlex Medical Dictionary, 2012). Deformities may be dynamic or fixed. Dynamic deformities can be modified by active or passive motion (Georgiades et al., 2014). Fixed deformities, or loss of joint motion, usually require intervention through surgery (Copley & Kuipers, 2014b). Hand deformities vary according to the underlying pathology to the developing brain (Arner et al., 2008) and are determined by sensory impairment, secondary musculoskeletal change, severity of imbalance between spastic and paretic muscles and voluntary ability to grasp and release objects (Graham, 2004; Zancolli et al., 1983).

Park et al. (2011) explored the effect of upper limb deformities on upper limb function in 234 children with spastic CP. Of these 234 participants, 147 (62.8%) presented with flexion deformities of the fingers and wrist, with deformity noted in 11 children under the age of two years. The degree of deformity was strongly related to the amount of active upper limb movement measured by the Upper Limb Physician's Rating Scale and Upper Extremity Rating Scale (Park et al., 2011).

In a prospective study of 81 children with unilateral CP (mean age 9 years 11 months, SD 3 years 3 months), mapping the five-year time course of upper limb function and influencing factors, Klingels et al. (2018) found more passive ROM limitations in children nine years of age and older than those aged less than nine. The most pronounced limitation was reported for wrist extension. Klingels et al. (2018) discussed the importance of continued intervention to lengthen wrist and finger flexor muscles for children within this age group, including use of orthoses and Botulinum Toxin followed by intensive therapy and surgical intervention.

In a population-based study exploring upper limb contracture in 771 children with CP, Hedberg-Graff et al. (2019) found that 19.4% of the children had significantly decreased passive wrist and/or wrist and finger extension at age four years compared to children aged one to four years. Seventeen children, aged one to three years, had upper limb contracture. The number of children with contractures increased with age, with greater deterioration between 12 to13 years of age, a time that commonly includes a growth spurt (Hedberg-Graff et al., 2019). There is evidence that children from all Manual Ability Classification System (MACS) levels

(Eliasson et al., 2006), including high ability levels (MACS levels I and II), may develop contractures (Hedberg-Graff et al., 2019).

As secondary musculoskeletal changes progress and the degree of hand deformity and contracture increases, so do the functional deficits of the hand (Arner et al., 2008; Law et al., 2008). Impaired arm and hand function has been reported as a problem in about half the children who present with CP (Arnould et al., 2004; Fedrizzi et al., 2003). Varied coordinated movements of the hands are required in daily activities for independence, for communication and for social contact (Arner et al., 2008; Basu et al., 2014).

With the best available evidence, the management options aimed at improving upper limb function or correcting deformity include: bimanual training; constraint-induced movement therapy; context-focused therapy; goal directed training; occupational therapy following Botulinum Toxin injections; and surgery (Boyd et al., 2001; Novak et al., 2013; 2020). The provision of orthoses has not been well evaluated even though they are the mainstay of treatment (Basu et al., 2014; Shierk et al., 2016; Stanger & Oresic, 2003).

1.4 Upper limb orthotic intervention

The International Organization for Standardization (ISO) (1989) defines an orthosis as a device used to modify neuro-musculoskeletal components of the body through the application of external forces. The term orthosis may also be interchanged with splint and/or brace (Australian Hand Therapy Association, 2012). The application of metal orthoses to manage joint contracture can be dated back to the 1500s (Lannin & Ada, 2011).

The aims of upper limb orthotic intervention for children with CP include: management of the effect of hypertonicity; prevention of deformity and contractures; management of pain; maintenance of tissue and joint integrity; and improvement of function and participation in activity (Arnould et al., 2014; Barroso et al., 2011; Chin et al., 2005; Copley & Kuipers, 1999; Gormley Jr, 2001; Hughes et al., 2017; Wilton, 2013b). Both clients and therapists have identified that the most frequent reason for the prescription of upper limb orthoses was to prevent deformity and contracture (Kuipers et al., 2009).

Although upper limb orthoses continue to be used in clinical practice, there is insufficient evidence to support use of this intervention for children with CP. Novak et al. (2020), in a systematic review on the state of the evidence about interventions for children with CP, reported a lack of evidence on the use of orthoses to improve upper limb function or prevent contracture for the CP population. The lack of evidence has resulted in continued controversies about the

effectiveness of upper limb orthoses, when they should be considered, what type of orthosis to use, and timing and length of wear (Burtner et al., 2008; Copley & Kuipers, 1999; Jackman et al., 2014b; Langlois et al., 1989; Lannin & Ada, 2011; Novak et al., 2013; 2020). Despite this, there is evidence that upper limb orthoses are being prescribed to significant numbers of children with CP (Andersson et al., 2019; Russo et al., 2009).

As part of a cross-sectional survey of function, pain, burden of care, self-concept and quality-of-life in children with hemiplegic CP (n=107) in Australia, Russo et al. (2009) reported findings on orthosis prescription. Fifty-six percent of children were prescribed upper limb orthoses, with approximately half of the 56% using the orthosis. Children who had higher levels of spasticity across the wrist (a score ≥ 2 on the Modified Ashworth Scale) were almost 40 times more likely to be prescribed an upper limb orthosis than those with lower levels of spasticity. Reasons for poor adherence to orthoses included: the orthosis interfering with completion of functional tasks; children with less functional impairment abandoning the orthosis; the benefits failing to offset the perceived stigma associated with its use (Russo et al., 2009). These authors acknowledged that little was known about the child characteristics associated with prescription, the prescribing practices of clinicians, adherence to wearing, and the benefits from orthosis wear.

In a randomised controlled trial investigating the effectiveness of cognitive orientation to daily occupational performance over and above functional hand orthoses for children with CP, Jackman et al. (2018) reported that if given the choice, 16 out of 25 children (64%) would not have worn the orthosis during daily practice of their identified goals. Reasons for the choice included: restriction of movement; increased difficulty with grasp and release; practice of goals with the orthosis being more difficult; and the orthosis being poorly tolerated (Jackman et al., 2018).

In a descriptive study investigating upper extremity spasticity-reducing treatment in adjunct to movement training and orthoses, Andersson et al. (2019) reported that out of 25 participants with CP (MACS levels IV-V): 56% adhered to wearing a wrist and hand orthosis at night as prescribed; 20% wore their wrist and hand orthosis during the day so as not to affect sleep; 12% did not use their prescribed orthosis due to changes in muscle tone; and 12% used an alternate thumb or wrist orthosis (Andersson et al., 2019).

It is crucial to recognise that children with CP vary in musculoskeletal impairment and hand deformity and this may affect the degree of response to upper limb orthotic intervention. In an overview of review articles Autti-Ramo et al. (2006) stated that the number of children with CP in upper limb orthosis studies was small, the prescribed orthoses differed among studies and were not always described sufficiently. In rehabilitation settings, the evidence generated by

traditional experimental methods, such as randomised controlled trials, are seen as having limited application (Copley & Kuipers, 2014a). Those methods describe outcomes in terms of the 'average effect' and may not adequately represent the heterogeneity or variability of functional ability of children with CP or the complexity of clinical practice (Burgess et al., 2008; Copley & Kuipers, 2014a; Teplicky et al., 2002). In considering the individual needs of the child, the type of orthosis may vary between children with similar clinical characteristics (Autti-Ramo et al., 2006; Goodman & Bazyk, 1991). The benefits gained from an orthosis must outweigh any restriction of function while it is worn (Exner & Bonder, 1983). There is a lack of studies specifically linking biomechanical characteristics and the dynamic interplay of wrist and finger musculature with upper limb orthotic intervention. Consideration of the individualised pattern of movement and hand deformity and an understanding of biomechanical principles is essential to guide prescription, design and fabrication of any orthoses applied to the upper limb (Fess et al., 2004; Wilton, 1983, 2003, 2013a). The initial focus of this doctoral program is to explore the reasons for upper limb orthosis use, and if hand deformity and biomechanical factors are considered in the clinician's decision making regarding upper limb orthosis prescription for children with CP.

1.5 Upper limb classification

The purpose of classification systems is to discriminate among variations of the presentation of a health condition (Rosenbaum et al., 2014). Classification systems distinguish between individuals with and without a particular characteristic (Kirshner & Guyatt, 1985; Rosenbaum et al., 1990) and provide an external framework for organising complex, unfamiliar or unstructured clinical problems (Copley & Kuipers, 2014a). It is an accepted international standard in CP to use the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997), the MACS (Eliasson et al., 2006) and the Communication Function Classification System (CFCS) (Hidecker et al., 2011) to classify gross motor, manual ability and communication function respectively. These systems do not substitute for more detailed assessment of function and performance, but provide: a common language; a reliable 'snapshot' of an individual's presentation; an aid for clinical reasoning regarding intervention choices; and complementary information to more detailed assessment and measurement (Kuipers & Grice, 2009; McConnell et al., 2011).

For children with CP, different upper limb classifications are available for classifying function or hand deformity. Clinicians may need to use more than one system to capture a complete clinical picture of the upper limb of a child with CP (McConnell et al., 2011). Valid and reliable classifications of upper limb function include the MACS (Eliasson et al., 2006), which

classifies the child's typical use of their upper limb during observed manual activity, and the Bimanual Fine Motor Function (BFMF) classification, which classifies how each of the child's hands grasp, hold and manipulate objects (Elvrum et al., 2016). Information provided by these classifications differs between fine motor capacity and actual manual performance (Elvrum et al., 2016). Classification of hand deformity is more problematic, as the biomechanical classifications used in clinical practice have clinical utility and some evidence of reliability (Klingels et al., 2010) but little evidence of validity (Arner et al., 2008; McConnell et al., 2011). McConnell et al. (2011) reported it difficult to recommend a classification system for hand deformity in the CP population.

1.6 Classification of hand deformity

Classifications of hand deformity used in clinical practice include the wrist classification developed by Zancolli (2003) and the thumb classification developed by House et al. (1981). Both classification systems were developed for pre and post-operative assessment in CP, and aim to describe the habitual position of the wrist and thumb respectively (McConnell et al., 2011). Classification of spastic hand deformity using this system is based on the patient's ability to voluntarily straighten their fingers by actively flexing the wrist, and ability to extend the wrist with flexed fingers. Surgical intervention is closely related to the classification of wrist and finger deformities (Zancolli, 2003).

Clinical experience in classifying hand deformity in children with CP using the Zancolli (2003) and House et al. (1981) classifications led Wilton (2003) to identify limitations in their use. The levels of the Zancolli and House classifications relate to surgical options for the specific deformity in each of the levels. The spectrum of hand deformities seen in clinical practice during activity are not reflected in these classifications and they therefore have less useful therapeutic application. Neither wrist extension deformities nor consideration for thumb posture is included in the Zancolli classification (Zancolli, 2003). There is no reference in the House thumb classification (House et al., 1981) to considering the position of the wrist when thumb deformity classification is made.

As a result of the limitations noted in the Zancolli and House classifications, Wilton (2003) developed a wrist and hand deformity classification to assist therapists to identify the various presentations of hand deformity and guide decision making for intervention, including orthosis prescription. After further development by Wilton (2013b), this classification was named the Neurological Hand Deformity Classification (NHDC). An initial reliability study has been published (Georgiades et al., 2014), which demonstrated high levels of inter-observer and intra-observer reliability of the NHDC (κ=0.87 and κ=0.91 respectively). Following that study by

Georgiades et al. (2014), a lack of guidelines in the procedure to classify hand deformity with the NHDC and its clinical application was identified by the author of this thesis. As part of this doctoral program a manual and website (Garbellini & Wilton, 2017) was developed to describe a standardised procedure for the NHDC to ensure consistency in observation and recording of hand deformity. A subsequent focus of this doctoral program is to further test the psychometric properties of the NHDC.

1.7 Theoretical framework

Hand deformity, decision making and upper limb orthosis prescription are the key foci of this doctoral program. A discussion of these concepts and frameworks – International Classification of Functioning, Disability and Health, biomechanical frame of reference and professional reasoning – that were used to support and guide this research follow.

1.7.1 International Classification of Functioning, Disability and Health (ICF)

The ICF (World Health Organisation, 2001) has re-defined the way clinicians think about CP and expanded clinical focus from managing impairments to promoting activity and participation intervention options (Novak et al., 2013; Rosenbaum & Stewart, 2004). The ICF frames the impact a health condition has on an individual's functioning. An individual's functioning is a dynamic inter-relationship between the body structure and function, activity and participation domains, within the context of their personal and environmental factors (Bartlett & Palisano, 2000; Hoare et al., 2011).

According to the ICF, hand deformity sits within the body structure and functions domain, as an impairment or negative consequence of CP (Ohrvall et al., 2013; World Health Organisation, 2007). The presence of CP may impact the body structures of the hand (muscle, joint, bone) as well as body functions (coordinated control of movement, muscle strength), which may limit the ability to complete a necessary daily living task, for example, eating and drinking (Arnould et al., 2014). Worsening impairment of body structures such as muscle shortening, joint contracture and increased hand deformity and their impact on body functions including movement, have the potential to negatively affect activity and participation (Copley et al., 2013).

1.7.2 Biomechanical frame of reference

A biomechanical frame of reference in occupational therapy is concerned with an individual's capacity for movement during daily occupations (McMillan, 2011; Rybski, 2012). The biomechanical approach uses anatomical and physiological knowledge, such as kinetics,

anatomy and kinematics to explain movement. (Rybski, 2012; Wilby, 2007). Movement is produced by a synthesis of forces coordinated by the nervous and musculoskeletal systems (McMillan, 2011). How a person moves and the effect that movement has on their occupational performance can be analysed. This information can be used to determine appropriate intervention strategies (McMillan, 2011; Rybski, 2012; Wilby, 2007). Orthosis use is an example of a body structure intervention intended to alter musculoskeletal components such as muscle stiffness, muscle length, balance and strength (Wallen & Stewart, 2016) through the application of external forces (Australian Hand Therapy Association, 2012). The biomechanical frame of reference is used to provide context for analysing the forces a person's movement creates and how an orthosis might counteract these forces.

1.7.3 Professional reasoning

Professional reasoning in occupational therapy is a complex and multi-faceted process that clinicians use to plan, direct, implement and reflect on client care (Mattingly & Fleming, 1994; Rogers, 1983; Schell & Cervero, 1993). Therapists do not always use a linear form of logic but use various methods of thinking in response to features of a clinical problem (Fleming, 1991a, 1991b). Factors that comprise each client's condition, client's contextual factors, the clinician's world view and the skill of the individual clinician and what they know how to do, continuously influence reasoning (Cohn, 1991; Hooper, 1997; Schell & Cervero, 1993). The most commonly agreed aspects of professional reasoning as summarized by Schell (2019) are scientific, diagnostic, procedural, narrative, pragmatic, ethical, interactive, and conditional. Most research about reasoning in occupational therapy practice suggests that all these aspects overlap and interact (Schell, 2019). An aim of this doctoral program is to explore decision making for upper limb orthosis prescription and the use of a hand deformity classification tool to aid decision making. The professional reasoning aspects will be used as a guide to frame the decision making component of this research.

1.8 Statement of the research problem

Upper limb movement and function is impaired in about half the children who are diagnosed with CP (Arnould et al., 2014). The clinical presentation of their upper limb positioning and movement is dictated by the influence of positive and negative factors of the upper motor neuron syndrome. Many children develop secondary musculoskeletal changes over time, which may limit use of their hands in daily tasks. Upper limb orthotic intervention is used to manage or prevent musculoskeletal changes, however, evidence to support orthosis use for children with CP is limited. There is a lack of clarity in the literature and a lack of guidelines supporting

therapists' decision making regarding upper limb orthotic intervention. Hand deformity classifications may identify the deforming hand postures in ways that guide further assessment and intervention of the body structures of concern. Limitations in the clinical use of surgical hand deformity classifications such as the Zancolli and House classifications have been identified. As a result, the NHDC was developed as a tool to guide decision making for therapists working in the clinical area. The NHDC has preliminary evidence of reliability but evidence about validity is limited. Further investigation of the reasons for upper limb orthosis prescription is required. What guides therapists' decision making for orthosis prescription needs further exploration. The psychometric properties of the NHDC, that has the potential to be used as a guide for decision making, requires exploration.

1.9 Statement of study purpose

The purpose of this research was to conduct a series of studies investigating the reasoning behind upper limb orthosis prescription for children with CP, to add to specific information within this clinical area, and to develop a hand deformity classification tool. This research addresses the following questions:

- What are the reasons for upper limb orthosis prescription for children with CP?
- Is there a documented link between the reason for an upper limb orthosis according to its intended effect, and the outcome measure used to determine its effect?
- Is standard nomenclature used in the literature to describe upper limb orthoses?
- What guides the decision making of occupational therapists in Australia when prescribing upper limb orthoses?
- What are the psychometric properties of the NHDC, and can the NHDC be used with confidence in clinical practice to classify hand deformity?
- How can the NHDC be applied in clinical practice?

1.10 Objectives of the doctoral program

In response to the research questions posed above the objectives of this doctoral program are:

1. To systematically review the literature
    - To explore the rationale and effect of upper limb orthosis prescription for children with CP
    - To identify any links between the reason for upper limb orthosis prescription, according to the intended outcome of the orthosis, and outcome measure used
    - To identify if any standard nomenclature is used when describing upper limb orthoses used in published research

2. To use a qualitative method to explore what guides occupational therapists' decision making when prescribing upper limb orthoses for children with CP

3. To establish validity and reliability of the NHDC by demonstrating
    - Construct validity as evaluated using hypothesis testing against specific criteria
    - Test–retest stability between occasions of assessment when no change is expected
    - Inter-rater reliability when different raters classify the same participants
    - Clinical utility using case studies

These specific objectives are a precursor to the future aim of exploring whether a biomechanical intervention, wrist hand orthoses, can be guided by a hand deformity classification (the NHDC) to improve targeting of prescription of the intervention and subsequently the effectiveness of upper limb orthotic intervention for children with CP.

1.11 Structure of the thesis

This doctoral program is presented in eight chapters:

- Chapter 2 presents the findings from a systematic review exploring the rationale for orthosis prescription and the effect of upper limb orthotic intervention
- Chapter 3 unpacks information from the systematic review and current literature in more detail to identify what domain-specific evidence exists to guide upper limb orthosis prescription
- Chapter 4 is comprised of two sections. The first section presents the findings from a Q method study, exploring what guides Australian occupational therapists' upper limb orthosis prescription decision making. The second section presents a critical reflection on the use of Q methodology. Findings of the Q method study support the need for clinical tools which aid decision making
- Chapter 5 introduces the Neurological Hand Deformity Classification as an aid to decision making
- The focus of Chapter 6 is on investigating the psychometric properties of the NHDC. This chapter is also comprised of two sections. The first presents the extended methodology of a study to establish validity and reliability of the NHDC. The second provides results of construct validity and test–retest and inter-rater reliability study of the NHDC
- In Chapter 7, case studies are presented to explore and explain how the NHDC can be applied clinically

- The final chapter, Chapter 8, provides an extended discussion of the findings from this doctoral program, its strengths and limitations, and implications for clinical practice and further research

Note: The manuscripts accepted for publication are presented in the chapters of this thesis (Chapters 2 and 4). See Appendix B for publications.

1.12 Context of research

This doctoral research was embedded within two multi-centred randomised controlled trials of upper limb orthoses for children (Imms et al., 2016); the Minimising Impairment Trial (MIT) and infant Wrist-Hand Orthosis Trial (iWHOT). The MIT and iWHOT investigator group comprise occupational therapists with clinical expertise in the area of CP and upper limb orthotic intervention. Consumer representatives were engaged to guide the MIT and iWHOT. These consumers participated in regular site meetings and provided firsthand experience of participating in the trials. Data from the MIT and iWHOT were used by the PhD candidate to explore the psychometric properties of the NHDC. The candidate, a senior occupational therapist with more than 20 years of clinical experience in this area, was an associate investigator for the MIT and iWHOT and was directly involved in data collection at the Perth Children's Hospital site. A summary of the research methods used during the candidate's research journey is presented in Table 1.1.

Table 1.1 A summary of the methods used in each study in this doctoral program

Aspect	Study 1	Study 2	Study 3	Study 4
Design	Systematic review	Qualitative cross- sectional survey utilising Q methodology	Measurement study design – Construct validity, test–retest and inter-rater reliability	Case study design
Sample	815 records were identified; 18 studies met selection criteria	39 occupational therapists who were currently active or had treated children with CP	Children with CP (age range 0–15 years) with abnormal or persistent postures of the wrist and fingers; Construct validity n=127 Test–retest reliability n=36 Inter-rater reliability n=113	Two children with CP purposely selected
Sample Source	Cochrane central register, MEDLINE, CINAHL, Embase, SCOPUS and Web of Science databases	Australian occupational therapists from community, not-for-profit and tertiary health services were approached	Two Australian multi-centre randomised controlled trials investigating the use of rigid wrist and hand orthoses, iWHOT and MIT	iWHOT and MIT
Data analysis	Narrative synthesis with tabulation of data for each study	By-person factor analysis specific to Q methodology (Ken-Q Analysis software)	Validity: Chi-squared test of independence; t-test; Spearman correlation co-efficient Reliability: Percentage of absolute agreement between repeated measures (test-retest) and raters (inter-rater); Cohen's kappa with 95% confidence intervals	Narrative synthesis of data
Publication status	Published in *Disability and Rehabilitation* 2017	Results published in *Disability and Rehabilitation* 2018 Critical review published in the *Scandinavian Journal of Occupational Therapy* 2020	Under review in the *Journal of Hand Therapy*	Not prepared for publication
Relevant abstracts	Presented at the AusACPDM scientific conference 2016	Presented at the AHTA scientific conference 2018	Presented at the AusACPDM scientific conference 2020	Not yet presented

Chapter Two

A Systematic Review

Preamble 18

PUBLICATION 1 – RATIONALE FOR PRESCRIPTION, AND EFFECTIVENESS OF, UPPER LIMB ORTHOTIC INTERVENTION FOR CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW 19

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Preamble

The aims of the study reported in this chapter were to: explore why upper limb orthoses were prescribed for children with CP; explore the connection between reason and outcome from use of orthoses; and describe upper limb orthoses within the domains of the ICF and common terminology of the Australian and American Hand Therapy orthosis classification systems. These aims were achieved by systematically reviewing the literature with objective and transparent methods as per the preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement (Moher et al., 2009). The protocol for this systematic review was submitted and registered with the International Prospective Register of Systematic Reviews (PROSPERO) (Appendix C). The accepted manuscript as prepared for *Disability and Rehabilitation* is shown in this chapter. See Appendix B for publication.

As it has been three years since the systematic review was published, an updated search using methods consistent with the original review was conducted. The aim was to identify studies published between 2017–2019 that investigated the effect of an upper limb orthotic intervention for children with cerebral palsy. Data were extracted and tabulated from additional studies identified within the time period between initial publication and the end of the year 2019. Findings are integrated with the published review findings and implications for clinical practice and further research are discussed.

PUBLICATION 1 – RATIONALE FOR PRESCRIPTION, AND EFFECTIVENESS OF, UPPER LIMB ORTHOTIC INTERVENTION FOR CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW

Publication details

Title:	Rationale for prescription, and effectiveness of, upper limb orthotic intervention for children with cerebral palsy: a systematic review
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[1] See Appendix B, Section B.1 for the published edition of this paper

Abstract

Purpose:

i. To explore reasons for upper limb orthosis prescription for children with cerebral palsy
ii. To explore the link between reason and effect according to intended outcome and outcome measure utilised
iii. To classify the prescribed orthoses using standard terminology

Method: A prospectively registered (Centre for Reviews and Dissemination: 42015022067) systematic review searched for experimental and observational studies investigating rigid/thermoplastic upper limb orthotic intervention for children aged 0-18 with cerebral palsy. The Cochrane central register, MEDLINE, CINAHL, Embase, SCOPUS and Web of Science databases were searched. Included studies were assessed for risk of bias.

Results: Sixteen studies met selection criteria. Two studies described a specific reason for orthosis prescription, six prescribed orthoses to manage a clinical symptom and eight did not describe a reason. Eight studies were analysed for effect according to intended outcome with no clear connection found between reasons for prescription, outcome measures utilised and effect reported.

Interpretation: The lack of evidence for upper limb orthotic intervention for children with cerebral palsy leads to uncertainty when considering this treatment modality. Future research is needed to evaluate the effect of orthosis wear in relation to intended outcome utilising robust methods and valid and reliable outcome measures.

2.1 Introduction

Limitations in hand use due to musculoskeletal change impact people with cerebral palsy (CP) across their life span and can result in significant disability and pain (Blair, 2010; Chin et al., 2005; Foran et al., 2005; Park et al., 2011; Vargus-Adams, 2009). Musculoskeletal problems develop as a result of growth, ageing, spasticity and limitations of movement against gravity (Chin et al., 2005; de Bruin et al., 2013; Fulford & Brown, 1976; Graham & Selber, 2003; Hanna et al., 2003; Rosenbaum et al., 2007; Vaz et al., 2006). Secondary musculoskeletal deformities are, therefore, progressive (Chin et al., 2005; Georgiades et al., 2014; Graham, 2004; Graham & Selber, 2003) and important to control, if possible.

Orthoses are one intervention prescribed to children with CP to minimise secondary upper limb musculoskeletal deformities (Wilton, 2013a). An orthosis is defined as an externally applied device used to modify the structural and functional characteristics of the neuro-muscular and skeletal systems by applying forces to the body. In clinical practice, orthosis, splint and brace are interchangeable terms (Australian Hand Therapy Association, 2012). Orthosis is the term preferred by the International Organization for Standardization (ISO) (1989). The primary purpose of an orthosis fitted to the upper limb is to immobilise or mobilise tissues to achieve the reason for which it is prescribed (Boscheinen-Morrin & Conolly, 2001; Colditz, 1996; Wilton, 2013a). Orthoses used to mobilise tissues apply gentle forces aimed at increasing passive range of joint motion (Wilton, 2013a). The use of upper limb orthoses for children with upper limb impairment is not a stand-alone intervention within clinical practice. Prescription needs to be made in consideration of other therapeutic, pharmacological and surgical interventions specifically related to the individual (Wilton, 2013b). Orthoses may be prescribed to manage the effect of hypertonicity, prevent deformity and contractures, manage pain, maintain tissue and joint integrity and improve function and participation in activity (Boscheinen-Morrin & Conolly, 2001; Colditz, 1996; Copley & Kuipers, 1999; Fess, 2011; Wilton, 2013b). This range of reasons for orthosis prescription targets the body function and structure, activity and participation domains of the International Classification of Functioning, Disability and Health (World Health Organisation, 2007).

Practice guidelines for the prescription of orthoses in the management of neurologically-based upper limb impairments are scarce and great variability in prescription exists. Reported factors which influence therapists' prescription of orthoses include therapist training, familiarity and expertise in the orthoses used and the availability of resources such as time, equipment and materials (Fess et al., 2004). Other factors that influence prescription include client goals; client engagement in the intervention; consideration of the therapeutic relationship; use of other interventions; family and carer support of the intervention; and the client's environment (Kuipers et al., 2009; Wilton, 2013b). Although these many factors have been identified, there is a paucity of information about, and studies investigating, these factors in the literature.

Evidence of the effect of upper limb orthoses for children with CP has been reviewed in four publications between 1989 and 2014 (Autti-Ramo et al., 2006; Jackman et al., 2014b; Langlois et al., 1989; Teplicky et al., 2002). Langlois et al. (1989) conducted a literature review of 13 studies on the use of hand splints and cerebral spasticity, with three studies including children with CP. The orthoses applied in the three studies included Snook's spasticity reduction splint (McPherson, 1981); dorsal and volar resting hand splints (McPherson et al., 1982); Orthokinetic Cuff, Short Opponens Thumb Splint and MacKinnon splints (Exner & Bonder, 1983). Langlois et al. (1989) reported that clinical observation and scientific investigation supported spasticity reduction using orthotic intervention. No conclusion was reached regarding the most effective orthosis design.

Teplicky et al. (2002) conducted a critical review of the literature on the use of upper limb orthoses for children with CP and traumatic brain injury. The review included four papers utilising (i) Orthokinetic Cuff, Short Opponens Thumb Splint and MacKinnon splints (Exner & Bonder, 1983); (ii) MacKinnon Splint (Flegle & Leibowitz, 1988); (iii) Hand Positioning Device (Reid & Sochaniwskyj, 1992); (iv) Dynamic Proximal Stability (Lycra®) Splint (Blair et al., 1995). The reviewers concluded that hand splints improved grasp. Autti-Ramo et al. (2006) completed an overview of review articles to summarise evidence on the effect of upper and lower limb casting or orthoses provided to children with CP. Only one review by Teplicky et al. (2002) was included that investigated the use of upper limb orthoses. Autti-Ramo et al. (2006) found inconclusive evidence about the management of upper extremity problems using casting or splinting.

Jackman et al. (2014b) assessed evidence of the effectiveness of orthotic intervention for improving hand function for children with CP using a systematic review with meta-analysis. Only randomised and quasi-randomised trials were included in the review. Although the authors reported study limitations made it difficult to draw clear conclusions, their findings suggested that upper limb orthoses may have small benefits for hand skills however improvements diminished after wearing ceased. The authors identified four studies as being sufficiently homogenous in regard to participants, reason for orthosis, impairment-based outcome measures and wearing regimes, to include in a meta-analysis. On closer inspection of these factors, particularly the reason for the orthosis and wearing regimes, they may not be sufficiently homogenous for inclusion in a meta-analysis. The intervention of a non-functional orthosis included those constructed of thermoplastic material, plaster and fibreglass and metal lockable hinges. The joints on which the orthoses acted were described but there was no comment as to whether the orthoses were designed to immobilise joint movement or mobilise tissue. Regimen of wear varied from nightly for 6 months, at least 4 hours daily for 6 months and two thirty-minute sessions daily for 6 months.

2.2 Rationale and aims

Upper limb orthoses are used to treat upper limb impairment in children with CP; however, there is little evidence about their effect and no clear guidelines regarding when the intervention should be considered, what orthosis to use, and timing and length of wear (Copley & Kuipers, 1999; Jackman et al., 2014b; Novak et al., 2013). The lack of guidelines for practice may relate to the lack of systematic investigation as to the reason for orthosis prescription and whether any observed effects meet the intended outcomes of the prescribed orthoses. In addition, there is inadequate use of standardised terminology and inconsistent reporting of orthosis descriptions in clinical practice.

The Australian Hand Therapy Association (AHTA) and American Society of Hand Therapists (ASHT) endorse the use of orthosis classification systems to provide standard nomenclature to clearly describe an orthosis (Australian Hand Therapy Association, 2012; Fess, 2011; American Society of Hand Therapists, 1992). Classification of orthoses can be made regardless of the condition for which the orthosis is prescribed, theoretical approach or material used to fabricate the orthosis. Defined language from the classification systems eliminates confusion caused by colloquial jargon, improving communication between clinicians. The more defined the orthosis description the greater potential there is for advancing knowledge, improving patient outcomes and research (Fess, 2011).

This study aimed to:

1. Explore the reasons for upper limb orthosis prescription for children with CP;
2. Explore the link between reason and effect of upper limb orthoses for children with CP according to the orthoses' intended outcome and outcome measures utilised; and
3. Classify the upper limb orthoses using the ICF-CY (World Health Organisation, 2007) framework and common criteria of the AHTA and ASHT orthosis classification systems (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992).

2.3 Methods

2.3.1 Design

A systematic review was undertaken using objective and transparent methods as per the PRISMA guidelines (Moher et al., 2009), to identify, evaluate and summarise all relevant research findings (Centre for Reviews and Dissemination, 2009). The protocol for this review was submitted to the Prosepero register and accepted on the 16/07/2015 (CRD42015022067).

2.3.2 Eligibility

Inclusion criteria

Studies investigating the effect of an upper limb orthosis intervention, regardless of publication date, that met the following criteria were included in the review:

1. Included participants were children aged 0–18 years. In instances where a paper also included participants older than 18 years, the study was included if 50% or more of the participants were between 0–18 years;
2. Participants had a diagnosis of CP. In instances where a paper also included participants with other neurological diagnoses, such as acquired brain injury, the study was included if 50% or more of the participants had a diagnosis of CP;
3. The intervention included a rigid/thermoplastic orthosis applied to the upper limb (regardless of duration or dosage). An orthosis was considered rigid if it did not allow dynamic movement of the joints it acted upon, even if it was fabricated with a combination of thermoplastic and flexible materials; and
4. The research design was a randomised controlled, quasi experimental study, or an observational study using a cohort or case-control or single case experimental study design.

Exclusion criteria

Studies were excluded based on the following criteria:

1. The participants had neuro-degenerative conditions;
2. The orthotic intervention was designed to constrain the unaffected upper limb for the purpose of improving movement of the affected upper limb;
3. The orthotic intervention was a cast applied to the upper limb;
4. The orthosis used in the intervention was fully constructed of soft, non-rigid materials for example, neoprene or Lycra®;
5. The study design was qualitative or a systematic review; and
6. Full papers not written in English requiring translation.

2.3.3 Information sources

Databases searched included the Cochrane central register, MEDLINE, CINAHL, Embase, SCOPUS and Web of Science. Database searches were conducted in June 2015 and again in May 2016. Hand searches of reference lists of included studies were conducted to ensure additional relevant references were identified. Although systematic reviews were excluded,

reference lists were checked to ensure all primary research was located for inclusion. There was no limit placed on the publication dates of the included studies. Only full papers written in English were included. Full papers that required translation were excluded due to a lack of resources. Where multiple publications reporting on the same study existed, data from the study were extracted and reviewed only once.

2.3.4 Search

A full electronic advanced search strategy conducted in the MEDLINE complete database, utilising the EBSCOhost Research Databases interface, is presented Table 2.1. This strategy was modified as needed and applied to the remaining databases.

Table 2.1 Electronic search strategy in MEDLINE complete database

Search ID	Search Terms
S1	(MH "Cerebral Palsy") OR (MH "Hemiplegia") OR (MH "Brain Injuries") OR (MH "Craniocerebral Trauma") OR (MH "Brain Damage, Chronic") OR (MH "Hypoxia, Brain")
S2	(MH "Brain Hemorrhage, Traumatic") OR (MH "Brain Stem Hemorrhage, Traumatic") OR (MH "Cerebral Hemorrhage, Traumatic")
S3	AB (cerebral palsy" OR CP OR hemiplegi* OR "brain injur*" OR "head injur*" OR "brain damage" OR hypoxi*) OR TI ("cerebral palsy" OR CP OR hemiplegi* OR "brain injur*" OR "head injur*" OR "brain damage" OR hypoxi*)
S4	S1 OR S2 OR S3
S5	(MH "Child") OR (MH "Child, Preschool") OR (MH "Adolescent") OR (MH "Infant") OR (MH "Infant, Newborn+")
S6	AB (child* OR infant* OR toddler* OR babies OR baby OR adoles* OR youth OR preschool) OR TI (child* OR infant* OR toddler* OR babies OR baby OR adoles* OR youth OR preschool)
S7	S5 OR S6
S8	S4 AND S7
S9	(MH "Splints") OR (MH "Orthotic Devices") OR (MH "Braces")
S10	AB (splint* OR orthot* OR orthos* OR brace*) OR TI (splint* OR orthot* OR orthos* OR brace*)
S11	S9 OR S10
S12	(MH "Hand") OR (MH "Fingers") OR (MH "Thumb") OR (MH "Wrist") OR (MH "Metacarpus") OR (MH "Upper Extremity") OR (MH "Elbow") OR (MH "Arm") OR (MH "Hand Deformities") OR (MH "Hand Deformities, Acquired") OR (MH "Hand Deformities, Congenital")
S13	AB (hand* OR arm* OR elbow* OR wrist* OR thumb* OR "upper limb*") OR TI (hand* OR arm* OR elbow* OR wrist* OR thumb* OR "upper limb*")
S14	S12 OR S13
S15	S8 AND S11
S16	S14 AND S15

Note: Terms were modified as needed and the search re-run in each of the Cochrane central register, MEDLINE, CINAHL, SCOPUS and Web of Science databases.

2.3.5 Study selection

Study selection was conducted in two stages utilising the Covidence systematic review software for data management (Veritas Health Innovation, 2013). In stage one two authors (SG, YR) independently screened titles and abstracts against the inclusion/exclusion criteria and identified relevant papers. In stage two, the same two authors independently reviewed the full text studies unable to be excluded by title and abstract alone. Comparison of papers was completed between the two authors with no disagreements regarding inclusion.

2.3.6 Data extraction process

Two authors (SG, YR) independently extracted data using a customised data extraction form developed for the purpose of the review. The data extraction form was piloted for use and tested for agreement with two studies. Once agreement between the authors was reached regarding completeness of the data extraction form, data were extracted from the remaining identified studies. Data extracted for each study were tabulated. Extracted data were compared and consensus was reached via discussion between the two independent reviewers where differences in data extraction were evident.

2.3.7 Data synthesis

A narrative synthesis with tabulation of the data for each study aim was completed. Comparison of the data relating to the reason for upper limb orthosis prescription, the link between reason and intended outcome, outcome measures utilised and effect, and orthosis classification is presented. To meet the aim of classifying orthoses, the domain(s) of the ICF-CY (World Health Organisation, 2007) which the orthosis was prescribed to influence were identified and common criteria adapted from the ASHT and AHTA orthoses classification systems were used to describe the anatomical focus and primary purpose of each orthosis (Australian Hand Therapy Association, 2012; Colditz, 1996; Fess, 2011; American Society of Hand Therapists, 1992). Figure 2.1 displays the common classification criteria.

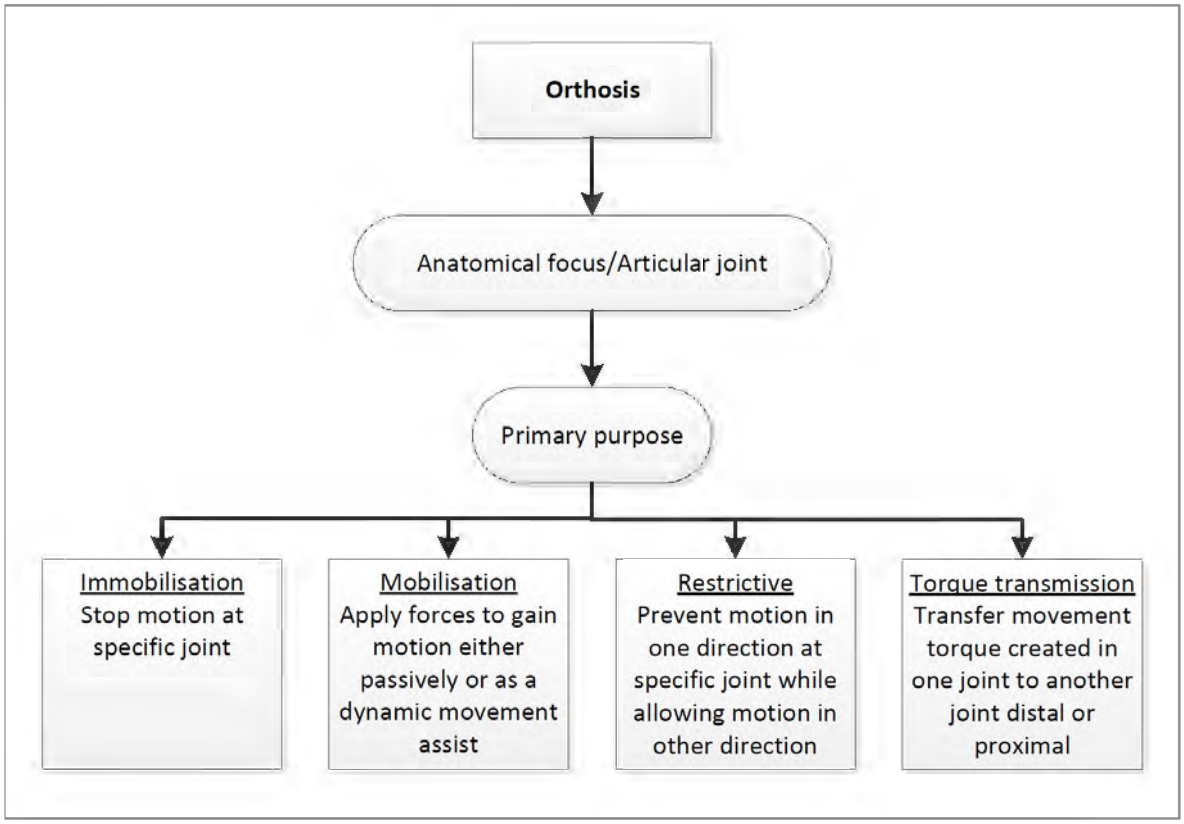

Figure 2.1 Common criteria of ASHT and AHTA Orthosis Classification Systems

2.3.8 Risk of bias in individual studies

All studies were assessed independently for risk of bias by two authors (SG, YR). The PEDro scale (Maher et al., 2003) was utilised to assess methodological quality of randomised controlled trials. Evaluation of methodological quality of intervention studies using single-case methodology was completed utilising the Risk of Bias in N of 1 Trials (RoBiNT) Scale (Tate et al., 2015). All non-randomised intervention studies were assessed using A Cochrane Risk of Bias Assessment Tool: for Non-Randomised Studies of Intervention (ACROBAT-NRSI) (Sterne et al., 2014). Using this tool, the highest assessment of risk of bias for an individual domain is used to determine the overall risk of bias for the study (Sterne et al., 2014). Moderate risk indicates the study is judged to be at low or moderate risk for all domains and is sound for a non-randomised study with regard to this domain but cannot be considered comparable to a well-performed randomised trial. Serious risk of bias indicates the study is judged to be at serious risk of bias for at least one domain (Sterne et al., 2014).

2.4 Results

A total of 16 studies were selected for inclusion in this review after the primary search was completed (see flow diagram of results in Figure 2.2). Only one study (Lidman et al., 2015) was selected for full text review following the second database search. The study did not meet inclusion criteria even though it utilised a rigid/thermoplastic wrist and hand orthosis, as the study aim was to evaluate and compare the effectiveness of repeated Botulinum Toxin-A injections plus occupational therapy versus occupational therapy alone (Lidman et al., 2015). The impact of orthosis in isolation use could not be determined by the study design (Russo, 2015).

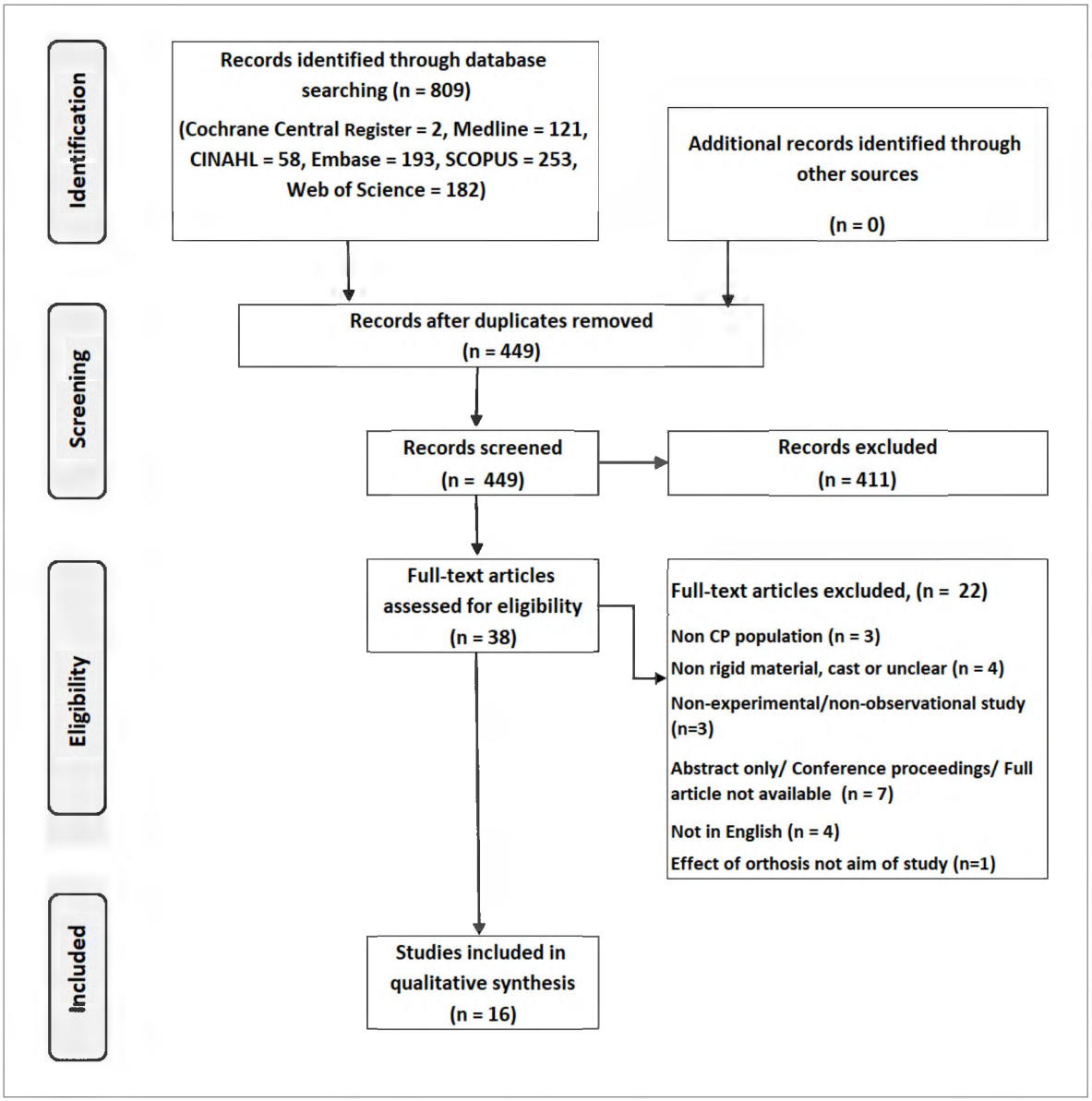

Figure 2.2 Study selection flow diagram

Two randomised controlled trials (Kanellopoulos et al., 2009; Ozer et al., 2006), two single-case experimental designs (Goodman & Bazyk, 1991; Kinghorn & Roberts, 1996) and 12 non-randomised intervention studies (Barroso et al., 2011; Burtner et al., 2008; Carmick, 1997; Chhawchhria, 2014; Currie & Mendiola, 1987; Exner & Bonder, 1983; Louwers et al., 2011; Postans et al., 2010; Scheker et al., 1999; Yasukawa & Cassar, 2009; Yasukawa et al., 2008; Yasukawa et al., 2003) were included in the review and assessed for risk of bias. The scores on the PEDro scale for the randomised controlled trials by Kanellopoulos et al. (2009) and Ozer et al. (2006) were 4/10 (fair quality) and 7/10 (high quality) respectively. Kanellopoulos et al. (2009) did not conceal allocation, blind assessors to the intervention or provide point measures and measures of variability for outcomes. Ozer et al. (2006) did not have a control group. Neither study was able to blind therapists administering therapy or blind subjects to their allocated group, as this is not possible in this intervention. The single-case experimental design studies by Goodman and Bazyk (1991) and Kinghorn and Roberts (1996) assessed with the RoBiNT scale (Tate et al., 2015) scored 11/30 and 12/30 respectively. Internal validity ratings suggested high risk of bias (3/14 and 4/14 respectively). It is difficult to attribute the reported improvement in hand function to use of the orthosis as neither study discussed random assignment of treatment times or blinding of assessors to the intervention (Goodman & Bazyk, 1991; Kinghorn & Roberts, 1996). The findings may not be generalizable as indicated by external validity scores of 8/16 for both studies. The risk of bias assessment of the 12 non-randomised studies is presented in Table 2.2. Nine of the non-randomised studies of intervention were assessed with an overall moderate risk of bias and three with serious risk of bias (see Table 2.2).

2.4.1 Reason for orthosis prescription

All children in the included studies who were prescribed an upper limb orthosis had a diagnosis of CP and all but one study (Yasukawa et al., 2008) reported that the children had spastic motor type. All children were seen in an outpatient setting. Table 2.3 outlines the reasons for orthosis prescription documented in the included studies. Information regarding study design, purpose of the study and inclusion criteria is also included in Table 2.3.

The reported reasons for orthosis prescription varied in the included studies as seen in Table 2.3. Two of the 16 studies described a specific reason for orthosis prescription. Barroso et al. (2011) reported the orthosis was to improve wrist and thumb joint positions. Carmick (1997) reported the orthosis was to stabilise the wrist in a functional position and 'replace the therapist's hands', to maintain an extended wrist position to encourage motor learning. Six studies (Burtner et al., 2008; Currie & Mendiola, 1987; Kanellopoulos et al., 2009; Louwers et al., 2011; Yasukawa et al., 2008; Yasukawa et al., 2003) prescribed orthoses to manage the presentation

of spasticity and increased muscle tone, altered arm and hand position or primary contracture that limited active and passive movement. The remaining eight studies (50%) (Chhawchhria, 2014; Exner & Bonder, 1983; Goodman & Bazyk, 1991; Kinghorn & Roberts, 1996; Ozer et al., 2006; Postans et al., 2010; Scheker et al., 1999; Yasukawa & Cassar, 2009) did not report the reason for orthosis prescription.

2.4.2 Link between reason for prescription and effect

Prescribed reason

For the eight studies reporting a reason for orthosis prescription, data describing the effect of the upper limb orthosis according to its intended outcome are presented in Table 2.4. For each of the eight studies listed, their analyses for orthosis effect found a positive effect. In the remaining eight studies where the reason for the orthosis was not reported, it was not possible to present results exploring the link between reason and effect.

Table 2.2 Risk of bias assessment of non-randomised studies (n=12)

Study/ *Study design*	Domain of risk of bias							
	Confounding	**Selection of participants**	**Measurement of intervention**	**Departure from intended intervention**	**Missing data**	**Measurement of outcomes**	**Selection of reported results**	**Overall Bias** [d]
Barroso (2011) *Pre/post test*	Moderate [b]	Moderate	Low [a]	No information	Low	Moderate	Moderate	Moderate
Burtner (2008) *Pre/post test*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Carmick 1997) *Case study*	Moderate	Serious [c]	Moderate	No information	Low	Serious	Moderate	Serious
Chhawchhria (2014) *Pre/post test*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Currie (1987) *Pre/post test*	Moderate	Moderate	Low	No information	Low	Serious	Moderate	Serious
Exner (1983) *Pre/post test*	Moderate	Moderate	Low	No information	Low	Serious	Serious	Serious
Louwers 2001) *Pre/post test*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Postans (2010) *Case series*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Scheker (1999) *Case series*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Yasukawa (2009) *Case study*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Yasukawa (2008) *Case series*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate
Yasukawa (2003) *Case series*	Moderate	Moderate	Low	No information	Low	Moderate	Moderate	Moderate

Note: Studies assessed for risk of bias with the ACROBAT-NRSI (Sterne et al., 2014) in alphabetical order. [a] Low risk of bias indicates the study is comparable to a well-performed randomised trial with regards to the domain; [b] Moderate risk of bias indicates the study is sound for a non-randomised study with regard to the domain but cannot be considered comparable to a well-performed randomised trial; [c] Serious risk of bias indicates the study has some important problems with regards to the domain. [d] Overall risk of bias is judged at the highest level of risk in any one of the domains (Sterne et al., 2014).

Table 2.3 Reason for orthosis prescription (n=16)

Study/ ***Study design***	**Study purpose**	**Inclusion criteria**	**Reason for orthosis prescription**
Barroso (2011) *Pre/post test*	To investigate indicators of hand improvement when a wrist extending/thumb abduction (WETA) orthosis is used.	5 to 12 years; Spastic hemi CP; Limited wrist and thumb ROM; MACS levels I/II; Able to follows instructions	To improve position of hypertonic hand Indicated by treating OT or PT
Carmick (1997) *Case study*	To describe a task-specific physical therapy approach using NMES and a dorsal wrist splint to aid upper limb function.	Not reported	To reduce wrist flexion 'Replace therapist's hands' for motor learning
Burtner (2008) *Pre/post test*	To compare grip, pinch, dexterity and EMG activity of selected UE muscles while participants with and without CP wore (i) no splint or (ii) static or (iii) dynamic splints.	Not reported	Moderate spasticity warranting splinting
Currie (1987) *Pre/post test*	To note whether thumb position improved while wearing the orthosis. To monitor any change in function of the paretic hand while wearing the orthosis.	Not reported	Mild to moderate hemi CP 'Cortical' thumb position limiting use
Kanellopoulos (2009) *RCT*	To evaluate the necessity and effectiveness of a static night splint following outpatient botulinum toxin A treatment in children with upper limb spastic CP	Not reported	Primary contracture in the upper limb
Louwers (2001) *Pre/post test*	To investigate immediate effects of a static wrist and thumb brace on the spontaneous use of the affected upper limb in bimanual activities in children with hemi CP	4 to 12 years; Spastic hemi CP; Zancolli classification of I, IIA, or IIB.	Hemiplegic pattern of the upper limb
Yasukawa (2008) *Case series*	To determine the efficacy of an Ultraflex orthosis with dynamic elbow and static wrist components in maintaining or improving PROM of the elbow and wrist	CP; Limited ROM; Abnormal muscle tone on MAS	Abnormal muscle tone interfering with range of motion or functional movement
Yasukawa (2003) *Case series*	To measure the efficacy of maintaining range of motion with the use of the bivalve long arm cast and the Ultraflex orthosis following botulinim toxin type A injection and serial casting when worn for an 8 month period.	Limited ROM; Severe to moderate spasticity at the elbow	Severe to moderate spasticity interfering with passive or active movements at the elbow

Study/ ***Study design***	**Study purpose**	**Inclusion criteria**	**Reason for orthosis prescription**
Chhawchhria (2014) *Pre/post test*	To determine the effect of and compare an above elbow wrist hand orthosis (AEWHO) and elbow gutter with below elbow wrist hand orthosis (BEWHO) on spasticity and hand function	18 months to 8 years; Spastic CP any topography; MAS of 1+ or greater	Not reported
Exner (1983) *Pre/post test*	To investigate the effects of three hand splints on bilateral hand use, grasp skills, and arm-hand posture in children with hemiplegia	2 to 16 years; Spastic type; at least 18 months Developmental level; Able to follow instructions.	Not reported
Goodman (1991) *Single case design*	To measure the effects of a short opponens splint on hand function and on the underlying aspects of hand function in a child with moderate spastic quadriparesis	Not documented	Not reported
Kinghorn (1996) *Single case design*	To investigate the use of an upper-extremity weight-bearing splint on muscle tone and functional hand skills in a child with cerebral palsy.	20 month old; spastic quadriplegia (age and motoric involvement resembling Smelt's (1989) subject) ().	Not reported
Ozer (2006) *RCT*	To investigate whether the improvement in the hand function was due to the effect of NMES, dynamic bracing or a combination of the two methods.	3 to 18 years; Spastic hemi; Zancolli classification type II or III; Follows directions; Sensation to light touch	Not reported
Postans (2010) *Case series*	To investigate the feasibility of applying a custom-made dynamic splint with NMES, to the wrist or elbow joint (or both).	Spastic CP; Limited ROM; Follows orders	Not reported
Scheker (1999) Case series	To assess the effects of dynamic orthotic and static traction and NMES on upper limb spasticity in cerebral palsy patients	3 to 21 years; Spastic hemi CP, mild to moderate spasticity; Follows directions; Good sensation	Not reported
Yasukawa (2009) Case study	Not reported	Not reported	Not reported

Note: The order of the studies in the table reflects the progression from specific documented reasons to absence of documented reasons for orthosis prescription. Hemi, hemiplegic; CP, cerebral palsy; ROM, range of motion; MACS, Manual Ability Classification System; OT, occupational therapy; PT physical therapy; NMES, neuromuscular electrical stimulation; EMG, electromyography; UE, upper extremity; PROM, passive range of motion; MAS, modified Ashworth scale. The order of the studies reflects the progression from well documented specific reasons to absence of documented reasons for orthosis prescription.

Table 2.4 Effect of upper limb orthoses according to intended outcome (n=8)

	Participants			Orthosis			Orthosis wear			Outcome measures		Conclusion
Study/ *Study design*	**Sample size (M/F)**	**Age range (Mean)**	**Topographical distribution**	**Reason**	**Name of Orthosis**	**Joint(s) position in orthosis**	**Hours of wear**	**Timing of wear**	**Duration of wear**	**Measure**	**Frequency**	**Study conclusion**
Currie (1987)* *Pre/post test*	5 (Not reported)	20 to 26 months (Not reported)	Hemiplegia	Mild to moderate hemiplegic CP, "Cortical" thumb position limiting use	The cortical thumb orthosis	Thumb abduction	During all waking hours	During all waking hours	1 week	Clinical observations, photos, prehension pattern	Pre orthosis and one week later with orthosis	Improved thumb position, hand function and prehension
Yasukawa (2008)* *Case series*	6 (4/2)	7 to 16 years (12 years)	Not reported	Abnormal muscle tone interfering with ROM or functional movement	Ultraflex orthosis	Not described	Not reported	As much as possible requested, report of day wear only	10 months	MAS, PROM, Caregiver interview	Baseline, 2, 3, 6 and 10 months	Positive correlation of wear and ROM
Yasukawa (2003)* *Case series*	3 (1/2)	7 years (7 years)	Quadriplegia	Severe to moderate spasticity interfering with passive/ active ROM at the elbow	Ultraflex orthosis	Elbow extension, forearm and wrist in neutral alignment	Average of 4 or more hours each day	Night wear	8 months	MAS, PROM, Caregiver questionnaire	Pre BoNT-A, 2, 6 and 8 months post injection	Improved ROM from baseline
Barroso (2011)† *Pre/post test*	32 (15/17)	5 to 11 years (8.65 years)	Hemiplegia	To improve hypertonic hand position; Indicated by OT or PT	WETA orthosis	Wrist joint 20º extension; thumb abduction and extension	Not reported	During data collection with the orthosis on	Not reported	Digital images for ROM, Muscle strength, JTTHF	With and without orthosis at data collection	Improved hand function

	Participants			Orthosis				Orthosis wear		Outcome measures		Conclusion
Study/ *Study design*	**Sample size (M/F)**	**Age range (Mean)**	**Topographical distribution**	**Reason**	**Name of Orthosis**	**Joint(s) position in orthosis**	**Hours of wear**	**Timing of wear**	**Duration of wear**	**Measure**	**Frequency**	**Study conclusion**
Burtner (2008)† *Pre/post test*	15 (6/9)	4 to 13 years (8.5 years)	Hemiplegia	Moderate spasticity warranting splinting	1. Volar wrist immobilizing splint 2. Dynamic spiral wrist hand splint	15-20º wrist extension	Not reported	During testing only	Not reported	Grip and pinch strength, Adapted nine hole peg test, EMG	With and without the orthosis at data collection	Greater grip dexterity with dynamic spiral splint
Carmick (1997)† *Case study*	1 (1/0)	7 years 7 months (7years 7 months)	Hemiplegia	To reduce wrist flexion; 'Replace therapist's hands'	Dorsal wrist splint	10º wrist extension	6 hours/ day at school	During home program at least 3 times a week	Nine months	Mowery's functional classification	Pre and post	Functional gains
Louwers (2011)† *Pre/post test*	25 (16/9)	4 to 11 years (8 years 4 months)	Hemiplegia	Hemiplegic pattern of the upper limb	Children's wrist and thumb support by Otto Bock	Neutral wrist, thumb abduction and opposition	During assessment only	During the second AHA only	During assessment only	AHA	Pre-test; 1 week later with brace; post-test (1 week later)	Positive effect for 9 of 22 AHA items
Kanellopoulos (2009)† *RCT*	20 (13/7)	2.5 to 12 years (7 years)	Hemiplegia	Primary contracture in the upper limb	Static night splint	Neutral wrist, thumb abduction, fingers extended	Not reported	Night time	6 months	QUEST	Baseline, 2 and 6 months post injection	Improved muscle tone, ROM and cosmesis

Note: Studies have been presented in order of those * with a link between between orthosis reason, outcome measure and effect and those † without a link. (M/F), (Male/Female); ROM, range of motion; MAS, modified Ashworth scale; PROM, passive range of motion; BoNT-A, Botulinum toxin type-A; OT, occupational therapy; PT, physical therapy; WETA, wrist extending thumb abduction; JTTHF, Jebsen Taylor Test of Hand Function; EMG, electromyography; CP, cerebral palsy; AHA, Assisting Hand Assessment; QUEST, Quality of Upper Extremity Skills Test.

Outcome measures of effect

Of the eight studies in which a reason for orthosis prescription was provided, only three studies (Currie & Mendiola, 1987; Yasukawa et al., 2008; Yasukawa et al., 2003) had a direct link between the reason for the orthosis, the outcome measure utilised and effect (see Table 2.4). For example, Yasukawa et al. (2003) prescribed an elbow orthosis for children where spasticity was interfering with range of motion (ROM) at the elbow. The outcome measures utilised included the Modified Ashworth Scale (MAS) (a spasticity measure) and ROM measures. The conclusion was a positive effect of improved ROM from baseline.

In the five remaining studies there was no clear link between the documented reasons for orthosis prescription, intended outcome, outcome measures utilised and effect (see Table 2.4). For example, the reason described for orthosis prescription by Burtner et al. (2008) was spasticity warranting orthotic intervention, but this was not supported by any spasticity measure, only the conclusion that grip improved. Kanellopoulos et al. (2009) used the Quality of Upper Extremity Skills Test (QUEST) as the sole outcome measure to evaluate quality of upper extremity function in four domains: dissociated movement, grasp, protective extension and weight bearing (DeMatteo et al., 1993). No goniometric range of motion measures or spasticity measure were utilised to support the conclusion reported by Kanellopoulos et al. (2009) of improvement in muscle tone, range of motion and hand cosmesis.

2.4.3 Classification of orthoses

No studies classified orthoses using the domains of the ICF-CY (World Health Organisation, 2007) framework or referred to an orthosis schedule when describing orthoses. Table 2.5 presents the review author's classification of the orthoses prescribed in the studies according to anatomical focus and primary purpose of the orthoses.

Table 2.5 Review author's classification of orthoses according to anatomical focus and primary purpose

		Reviewer classification		
			Orthosis classification system	
Study	**Orthosis**	**ICF domain(s)**	**Articular Joint/ Anatomical focus**	**Primary purpose**
Postans (2010)	Dynamic splint (Elbow)	BF & S and Activity	Elbow	Mobilising
Yasukawa (2009)	The forearm rotation elbow orthosis (FREO)	BF & S and Activity	Elbow and forearm	Mobilising
Yasukawa (2008)	Ultraflex orthosis	BF & S	Elbow and wrist	Mobilising
Yasukawa (2003)	Ultraflex orthosis	BF & S	Elbow and wrist	Mobilising
Ozer (2006)	Ultraflex orthotic device	BF & S and Activity	Elbow, forearm, wrist and fingers	Immobilising
Chhawchhria (2014)	Above elbow wrist hand orthosis (AEWHO)	BF & S and Activity	Elbow, wrist, fingers and thumb	Immobilising
Burtner (2008)	Volar wrist immobilization splint	BF & S and Activity	Wrist	Immobilising
	Dynamic spiral wrist hand splint	BF & S and Activity	Wrist	Restrictive
Carmick (1997)	Dorsal wrist splint	BF & S and Activity	Wrist	Immobilising
Postans (2010)	Dynamic splint (Wrist)	BF & S and Activity	Wrist	Mobilising
Chhawchhria (2014)	Below elbow wrist hand orthosis (BEWHO)	BF & S and Activity	Wrist, fingers and thumb	Immobilising
Kanellopoulos (2009)	Static night splint	BF & S and Activity	Wrist, fingers and thumb	Immobilising
Barroso (2011)	Wrist extending/thumb abduction (WETA)	BF & S and Activity	Wrist and thumb	Immobilising
Louwers (2011)	Children's wrist and thumb support	BF & S and Activity	Wrist and thumb	Immobilising
Scheker (1999)	Dynamic orthotic traction	BF & S	Fingers (MCP joints)	Mobilising
Kinghorn (1996)	Inhibitive weight-bearing splint	BF & S and Activity	Fingers and thumb	Immobilising
Currie (1987)	The cortical thumb orthosis	BF & S and Activity	Thumb	Immobilising
Exner (1983)	Short opponens thumb splint	BF & S and Activity	Thumb	Immobilising
Goodman (1991)	Volar-type short thumb opponens splint	BF & S and Activity	Thumb	Immobilising

Note: Studies have been organised by the classification of joint(s) addressed by the orthosis. Exner and Bonder (1983) compared the effects of three different orthoses on hand use, grasp and posture. The short opponens thumb splint was the only orthosis of Exner and Bonder's study that met the rigid/thermoplastic inclusion criteria of this review and therefore was the only orthosis in the study classified by the reviewers. BF & S, Body Function and Structure; Body functions are physiological functions of body systems; Body structures are anatomical parts of the body; Activity is an execution of a task or action by an individual (World Health Organisation, 2007); MCP, Metacarpophalangeal joints. Mobilising orthoses aimed to gain specific passive and active range of motion; Immobilising orthoses prevent movement at joints; Restrictive orthoses restrict ranges of movement (Australian Hand Therapy Association, 2012; Fess, 2011; American Society of Hand Therapists, 1992).

In all studies, the orthoses were classified as having an impact on the body function and structure domain of the ICF-CY as all were applied directly to the child and positioned the joints of the arm and hand. Only three studies (Scheker et al., 1999; Yasukawa et al., 2008; Yasukawa et al., 2003) were classified as having potential to impact the activity domain of the ICF-CY as the authors of these three studies included assessment of the orthosis effect on task execution.

Four of six elbow orthoses prescribed (Postans et al., 2010; Yasukawa & Cassar, 2009; Yasukawa et al., 2008; Yasukawa et al., 2003) were mobilising orthoses aimed at increasing the passive or active range of motion at the elbow joint. The dynamic spiral wrist splint (Burtner et al., 2008) was a restrictive orthosis as it allowed 30 degrees of movement but limited active wrist flexion range of motion. This reflects consideration of the dynamic interplay between wrist and finger musculature aimed at improving grasp, however the connection between this factor and orthosis design is not described in the study. The orthoses acting on the wrist and/or wrist and thumb joints (Barroso et al., 2011; Burtner et al., 2008; Carmick, 1997; Louwers et al., 2011) were classified as immobilising orthoses aimed at improving grip for task specific or bimanual use. Again, there was no connection between the movement of the wrist and fingers and orthosis design evident in the study authors' descriptions. All of the orthoses acting on the thumb (Currie & Mendiola, 1987; Exner & Bonder, 1983; Goodman & Bazyk, 1991) were hand based and immobilised the thumb in a position out of the palm aimed at developing improved grasp.

2.5 Discussion

This review aimed to explore reasons for upper limb orthosis prescription, explore the link between reason and effect according to the intended outcome and outcome measures utilised, and to classify orthoses within the domains of the ICF-CY (World Health Organisation, 2007) and common criteria of the ASHT and AHTA orthosis classification systems. Our findings suggest that connection between the reported rationale for orthosis prescription, the selection of outcome measures utilised according to intended outcomes and the effect described, is not transparent, nor well defined. All interventional study designs were included in this review to maximise the information available to explore the rationale for orthosis prescription. If only randomised controlled trials were included, only one study would have met selection criteria.

The systematic review undertaken by Jackman et al. (2014b) included randomised and quasi-randomised trials of upper limb orthoses. These authors reported there is no evidence supporting the use of orthoses for children with CP in isolation. In contrast three studies included in this review (Barroso et al., 2011; Burtner et al., 2008; Louwers et al., 2011) assessed the effect of upper limb orthoses while the child wore the orthoses only during testing. No additional

therapy or adjunct interventions were reported. The frequency of outcome measures were with and without the orthosis within 30 minutes (Barroso et al., 2011); twice with and without the orthosis within a two week period (Burtner et al., 2008); at baseline, a week later with orthosis wear and a week following without orthosis wear (Louwers et al., 2011). The reported effect was improved hand function (Barroso et al., 2011), greater grip dexterity (Burtner et al., 2008) and the ability to grasp objects of different shapes without assistance of the dominant hand (Louwers et al., 2011). It is not possible to determine the effect of adjunct interventions on the reported effectiveness of upper limb orthoses in the studies in this review.

The lack of clearly documented reasons for upper limb orthosis prescription and the effect of other interventions on upper limb impairment increases assumptions made regarding the clinical decisions for orthosis use and decreases capacity to use the evidence to inform clinical practice. The absence of a reason for orthosis prescription impacts the selection of appropriate outcome measure(s) utilised to determine effect, as it is not clear if the intended outcome is effectively measured. Often there was no clear link between the stated reason of the orthosis, the way the results were measured and the stated outcome of the study.

Manual ability and the influence of spasticity on the movement of the wrist and fingers of the affected limb(s) are important factors to consider in orthosis prescription (Wilton, 2013b), yet the connection between these factors and reason for prescription and description of orthoses is not well described in the included studies. Only three studies (Barroso et al., 2011; Louwers et al., 2011; Ozer et al., 2006) included a description of either the child's manual ability and/or active hand movement of the affected limb(s). A tool to classify manual ability for children with CP, the Manual Ability Classification System (MACS) (Eliasson et al., 2006), was published in 2006 however only two of the seven studies published after this date utilised the MACS to report the manual ability level of children in their study. Barroso et al. (2011) included children at MACS levels I and II and Louwers et al. (2011) included children at MACS levels I to III. The Zancolli classification (Zancolli, 2003) is another classification tool which reports the action of spasticity on the muscles of the wrist and fingers and classifies wrist and finger deformity according to observed movements. Only two studies, Ozer et al. (2006) and Louwers et al. (2011) classified children using this classification. All children in these two studies had voluntary extension of their wrist and/or fingers. Understanding how manual ability and movement limitations of the wrist and finger musculature of the affected limb(s) inform the reason for and choice of orthosis is critical.

All studies included in the review were evaluated to have some risk of bias. Therefore, the assessment of the effect of the orthoses should be taken in context of the quality of the individual

studies. Caution is required in generalising the reported effect of orthotic intervention in this review. The positive effects reported in the studies did not match the initial reason for the orthosis. This consistent mismatch between reason for prescription and measured effect significantly limits our understanding of the utility of orthoses for children with CP. It is critical in any intervention research (or intervention prescription) that the mechanism by which the intervention is intended to effect change is demonstrated. If associated, or additional, effects are anticipated, for example, improved use of the hand following orthosis wearing, it is certainly appropriate to also measure those effects. However, in the absence of knowing whether the primary mechanism of the orthosis is supported, it is difficult to interpret subsequent potential impacts.

2.5.1 Limitations of review

There were limitations of this review related to the search criteria, study design selection and evaluation of effect of intervention. Because the search was restricted to studies written in English, four studies were excluded that may have provided additional evidence. Study design selection included observational studies to determine the reason for the use of upper limb orthoses but, given the lower level of evidence this limited the evaluation and generalisation of effectiveness of the intervention. It was not possible to synthesise the data using statistical analyses given the heterogeneity and lower level of evidence within the included studies.

2.5.2 Recommendations for practice and research

Evidence to inform the use of upper limb orthoses for children with CP in clinical practice is limited. This may be because it is difficult to research a complex multi-factorial intervention rather than because the intervention lacks effect. Under these circumstances it is strongly recommended that continued clinical use of upper limb orthotic intervention should only occur with the inclusion of the following practice recommendations:

- A description of the dynamic interplay of the wrist and finger musculature as part of a comprehensive upper limb assessment;
- Clear documentation of the use of the orthosis as an intervention including the aim(s) of the orthosis based upon assessment findings;
- A description of the orthosis using consistent terminology applying an orthosis classification schedule such as those endorsed by the AHTA or ASHT; and
- Application (pre, during and post) of intervention outcome measures that are consistent with the reasons for the orthosis when evaluating the effect of the orthotic intervention.

Future clinical research is required to understand the factors that influence clinicians' rationale for prescribing orthoses as well as establishing the effect of upper limb orthotic intervention through connecting the reason for prescribing the orthosis, intended outcomes and effect utilising valid and reliable outcome measures.

2.6 Conclusion

The lack of clear evidence about the effect of upper limb orthoses leads to many challenges for clinicians who are considering offering upper limb orthotic intervention for children with CP. The variability in design, materials and presumed mechanism of effect of upper limb orthoses further increases the difficulty clinicians have in determining whether or not the intervention has value. Variability in terminology and stated rationale of orthoses used creates a communication barrier and introduces considerable difficulty in synthesising research evidence to determine the outcomes of orthosis prescription.

2.6.1 Acknowledgement

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2.6.2 Disclosure statement

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

END OF PUBLICATION 1

SYSTEMATIC REVIEW UPDATE

2.7 Background

In November 2019, the same electronic databases were searched with the same search terms as for the original review. The search yielded six papers that were not previously identified. One author (Garbellini) screened the titles and abstracts of the additional papers against the original inclusion and exclusion criteria. Four papers were excluded as three did not include an upper limb orthotic intervention and one used a non-rigid orthosis as the intervention. Two randomised controlled trials (RCTs) were identified: Jackman et al. (2018; 2019) as meeting criteria for inclusion. The same author (Garbellini) extracted data using the original data extraction form developed and piloted for the original review and tabulated the data. Quality of the RCTs were assessed with the PEDro scale (Maher et al., 2003), again by the same author.

Prior to publication of the two RCTs by Jackman et al. (2018; 2019), a protocol paper was published outlining both trials (Jackman et al., 2014a). The protocol outlined that enrolled participants would participate in both trials (Jackman et al., 2014a). To manage the similarities across both trials, data extraction was tabulated as if there was: one trial when determining the reason for orthosis prescription and classification according to anatomical focus and primary purpose, as the orthotic intervention was the same; and two trials when determining the effect of the intervention and quality assessment of the RCTs. To remain consistent with the tables in the published systematic review, the same format was used.

2.8 Results of systematic review update

PEDro scale scores for both trials (Jackman et al., 2018; 2019) were 8/10. Due to the nature of the intervention (orthosis prescription) blinding of participants and therapists to the intervention was not possible. A score of 8/10 indicated high quality on the PEDro scale for both studies. Table 2.6, Table 2.7 and Table 2.8 respectively present: the reason for orthosis prescription; effect of the orthoses; and the review author's (Garbellini) classification of orthoses according to anatomical focus and primary purpose.

Table 2.6 Reason for orthosis prescription

Study/ ***Study design***	**Study purpose**	**Inclusion criteria**	**Reason for orthosis prescription**
Jackman et al. (2018; 2019) *RCT*	1. To investigate whether CO-OP led to greater achievement of goals over and above functional orthotic intervention and in combination with orthotic intervention. 2. To investigate whether a functional orthosis led to an immediate improvement in hand function.	1. Aged 4 to 15 years 2. Diagnosed with cerebral palsy or brain injury 3. MACS level I to IV 4. Impaired hand function as a result of neurological condition 5. Child-set goals focused on improving hand function 6. Sufficient language, cognition and behavioural skills to participate 7. Parent commitment to a two-week block of therapy	To support one or more joints to maximise function of the upper limb during a task. For example, a wrist orthosis aimed at optimising wrist position for grasp and release.

Note: RCT, Randomised controlled trial; CO-OP, Cognitive Orientation to daily Occupational Performance; MACS, Manual Ability Classification System

Table 2.7 Effect of upper limb orthoses according to intended outcome

	Participants			Orthosis			Orthosis wear			Outcome measures		Conclusion
Study/ *Study design*	**Sample size (M/F)**	**Age range (Mean)**	**Topographical distribution**	**Reason**	**Name of Orthosis**	**Joint(s) position in orthosis**	**Hours of wear**	**Timing of wear**	**Duration of wear**	**Measure**	**Frequency**	**Study conclusion**
Jackman et al. (2018) *3 group parallel design RCT*	45 (23/22)	Range not reported (8.4 years)	Unilateral/ Bilateral 32/12	Support joint(s) to maximise function during a task	Wrist cock up orthosis	Wrist extension (20-30°) Customised to participant's finger flexion and extension status				*Primary*: COPM and GAS *Secondary*: Box & Blocks ROM	Baseline, post intervention and 8 weeks post	No difference in achieving goals. Orthoses generally not tolerated
Group												
1. Orthosis only	n=15	8.3	14/1				1 hour daily	Goal practice at home	2 weeks			
2. CO-OP only	n=15	8.1	11/4				Nil	N/A	N/A			
3. Orthosis and CO-OP	n=15	8.8	7/8				2 hours daily	During hourly CO-OP sessions and goal practice at home	2 weeks			

	Participants			Orthosis			Orthosis wear			Outcome measures		Conclusion
Study/ *Study design*	**Sample size (M/F)**	**Age range (Mean)**	**Topographical distribution**	**Reason**	**Name of Orthosis**	**Joint(s) position in orthosis**	**Hours of wear**	**Timing of wear**	**Duration of wear**	**Measure**	**Frequency**	**Study conclusion**
Jackman et al. (2019) *2 group parallel design RCT*	30 of the 45 above (16/14)	Range not reported (8.2 years)	Unilateral/ Bilateral 25/5	Support joint(s) to maximise function during a task	Wrist cock up orthosis	Wrist extension (20-30^{o}) Customised to participant's finger flexion and extension status				Box & Blocks	Baseline and one-hour post experiment-al orthosis wear	No between group difference. No immediate improve-ment in grasp and release
Group												
1. Orthosis	n=15	8.3	14/1				1 hour daily	Goal practice at home	For one hour post experimenta l baseline			
2. No orthosis	n=15	8.1	11/4				Nil	N/A	N/A			

Note: (M/F), (Male/Female); CO-OP, Cognitive Approach to Occupational Performance; N/A, Not applicable; COPM, Canadian Occupational Performance Measure; GAS, Goal Attainment Scale; ROM, range of motion.

Table 2.8 Review author's classification of orthoses according to anatomical focus and primary purpose

		Reviewer classification		
			Orthosis classification system	
Study	**Orthosis**	**ICF domain(s)**	**Articular Joint/ Anatomical focus**	**Primary purpose**
Jackman et al. (2018; 2019)	Wrist cock up orthosis	BF & S and Activity	Wrist	Immobilising

Note: BF & S, Body Function and Structure; Body functions are physiological functions of body systems; Body structures are anatomical parts of the body; Activity is an execution of a task or action by an individual.

2.9 Discussion

The information extracted from the new RCTs by Jackman et al. (2018; 2019) reinforced the findings of the original systematic review. The specific reason for orthosis prescription in both RCTs was described — to support the wrist in an optimal position to improve function, that is, grasp and release — however, the connection between the reason for prescription and specific goal-directed functional task for improvement was not well defined. Both trials refer to the wrist orthosis as being customised to allow maximal possible wrist extension with full finger extension (Jackman et al., 2018) and customised to the participant's finger flexion and extension status (Jackman et al., 2019), but further information describing the interplay of the wrist and finger musculature was lacking. There was no protocol or standard operating procedure presented for orthosis fabrication. For those participants with bilateral involvement, there was no information about the limb for which the orthosis was fabricated, whether an orthosis was prescribed for both limbs, or if the more severe limb was the one prescribed the orthosis. In both RCTs the orthosis was called a "wrist cock-up" orthosis with no reference or adherence to standard nomenclature as presented in hand therapy association orthosis schedules (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992).

2.10 Chapter Conclusion

The findings of the systematic review identified that the reason for orthosis prescription is not well defined in most of the included studies. Regardless of methodology and sample size, a lack of connection was evident between the reason for orthosis prescription, the outcome measures used and the stated outcome of the studies. There was little consideration of participants' manual ability and wrist and finger movement and how this guided orthosis prescription. Even studies presenting higher level evidence (RCTs) with high quality were limited in describing the reasons for, and decision making regarding, prescription of upper limb orthoses for children with CP.

Following findings from the review, it was recommended that continued orthosis prescription in clinical practice include: clear documentation of the reason for the orthosis; consideration of wrist and finger movement in orthosis prescription; outcome measurement linking effect with intended reason for the orthosis; and consistent terminology to describe the orthosis. Future clinical research is required to establish the effect of the intervention based on these practice recommendations. The following chapter (Chapter 3) considers the findings of the systematic review in more detail and identifies gaps in the literature that require further investigation.

Chapter Three

Upper limb orthosis prescription

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3.1 Introduction

Upper limb orthoses for people with neurologically-based impairment have been prescribed to: manage the effect of hypertonicity; prevent deformity and contracture; manage pain; maintain tissue and joint integrity; and improve function and participation in activity (Boscheinen-Morrin & Conolly, 2001; Colditz, 1996; Copley & Kuipers, 2014b; Fess, 2011; Wilton, 2013a). These varied reasons for upper limb orthosis prescription, as reported in the literature, provide some guidance for clinical practice. However, most published studies that explore the effect of upper limb orthoses have a low level of research evidence. This provides little direction to inform clinical practice (Novak et al., 2020). Currently, most upper limb orthotic prescription is based on practice tradition, biomechanical and neurophysiological theoretical assumptions and emerging but limited empirical evidence (Lannin et al., 2017).

The systematic review by Garbellini et al. (2017) (Study 1 of this doctoral program) identified a lack of clarity of the reasons for orthosis prescription, the orthoses' intended effect and the outcome measures used to determine that effect. Garbellini et al. (2017) proposed that this lack of clarity decreases the usefulness of the research literature to inform clinical practice about orthosis prescription. The lack of research evidence may be due, in part, to the complexity of researching a multi-faceted intervention. The question remains as to what information is currently available to guide clinician's decision making regarding upper limb orthosis prescription.

The first aim of this chapter is to further explore the reasons for orthosis prescription as identified from the systematic review (Garbellini et al., 2017) (Study 1). The reasons for orthosis prescription are detailed and discussed within the ICF, biomechanical and professional reasoning frameworks. The domains or components of these frameworks that relatied to the reasons given for orthosis prescription, identified within the studies included in Study 1 (16 studies) and the systematic review update (2 studies) were considered by the author. For example, no author presented a reason for orthosis prescription within the participation domain of the ICF, therefore the participation domain is not discussed. The second aim of this chapter is to identify and review existing resources intended to guide upper limb orthosis prescription. An algorithm, classification tool and model that organise this complex or unfamiliar information and guide practice are presented and discussed. The third, and final, aim of this chapter is to identify potential gaps that exist in the literature. The dynamic inter-relation of the ICF domains (World Health Organisation, 2001) is used to diagrammatically present these gaps.

3.2 Exploring the reasons for upper limb orthosis prescription

3.2.1 International Classification of Functioning, Disability and Health

The ICF provides a description of human functioning and its restrictions and organises this information into two parts. Part one deals with functioning and disability, which are considered outcomes of a dynamic interaction among body functions, body structures, activity and participation. Part two deals with contextual factors comprised of environmental and personal factors (World Health Organisation, 2001). The reasons for orthosis prescription identified in the literature can be linked to the domains of body structure, body function, and activity.

Body Structure

Body structures are defined as anatomical parts of the body (World Health Organisation, 2001) and those related to movement are the focus of this section. The systematic review by Garbellini et al. (2017) identified that eight out of 18 studies reported reasons for orthosis prescription aimed at addressing impairment of body structure, namely muscles and joints. These authors reported that prescription aimed to improve: (1) the structural position or support of joints (Barroso et al., 2011; Carmick, 1997; Currie & Mendiola, 1987; Jackman et al., 2018; 2019; Louwers et al., 2011); or (2) management of the effect of muscle tone, spasticity and/or contracture (Burtner et al., 2008; Kanellopoulos et al., 2009; Yasukawa et al., 2008; 2003).

Orthoses prescribed to support the thumb aimed to position the thumb in abduction and extension (Barroso et al., 2011; Currie & Mendiola, 1987; Louwers et al., 2011). Orthoses prescribed to address the position of the wrist or manage the effects of tone and spasticity in the wrist and/or finger musculature reported varying positions of the wrist, with the wrist in a neutral position (Kanellopoulos et al., 2009; Louwers et al., 2011; Yasukawa et al., 2003); 10 degrees of wrist extension (Burtner et al., 2008; Carmick, 1997); 20 degrees of wrist extension (Barroso et al., 2011); and 20–30 degrees of wrist extension (Jackman et al., 2018, 2019). There was little discussion in the publications regarding the rationale for the desired wrist position in the studies.

These studies provided some information to guide clinicians' decision making of orthosis prescription based on impaired body structure. For example, positioning of the thumb in abduction and extension to improve hand function was reported, but the rationale to clearly direct practice was lacking. Of concern were the eight studies that did not report any reason for orthosis prescription. In those eight studies it appeared that orthoses were prescribed because the child had CP, without any further justification or rationale. Omission of a clear rationale for orthosis prescription may increase the potential for an orthosis to be prescribed that does not address the

individual's movement impairment. Three studies (Jackman et al., 2018; 2019; Louwers et al., 2011) considered both the body structure in orthosis prescription (i.e. the wrist in either 20–30 degrees of extension or in a neutral position respectively) and also the body function of how the wrist and fingers moved in relation to each other.

Body Function

Body functions are defined as the physiological functions of body systems (World Health Organisation, 2001) with neuromusculoskeletal and movement related functions the focus of this section. Analysis of patterns of wrist and finger movement during function provide a basis for treatment decisions (Wilton, 2003). A classification of wrist and hand movement (Zancolli, 2003) was used in the study by Louwers et al. (2011) in relation to participant selection. Jackman et al. (2018; 2019) discussed customising the orthosis prescribed if the participant was not able to extend their fingers with the wrist positioned in 20–30 degrees of wrist extension. These studies documented neuromusculoskeletal and movement related function but didn't explicitly connect the reason for the specific orthosis prescription used in the study to the dynamic movement observed. These studies' respective use of movement analysis is discussed further.

Louwers et al. (2011) classified participants' hand deformity using the Zancolli (2003) classification. Classification was based on volitional release of the fingers through active wrist flexion and the ability to actively extend the wrist with the fingers flexed. Almost perfect agreement has been established for inter-rater reliability (n=23) for the Zancolli classification (Klingels et al., 2010). Louwers et al. (2011) used this reliable classification to determine severity of impairment of hand function. They excluded those who were classified with no ability to extend the fingers even with maximal passive wrist flexion from their study. In applying this criteria, Louwers et al. (2011) chose to exclude those children whose movement limitation was severe. It is the current author's interpretation, based on clinical experience, that this may be because children with this classification of movement do not usually improve with the use of a wrist orthosis worn during functional tasks. Louwers et al. (2011) discussed the difficulty some children have closing the hand around an object and holding it with the wrist in a flexed position. It is assumed therefore, that this was the rationale behind the decision to position the wrist in a neutral position in the orthosis. Louwers et al. (2011) reported that some children classified at Zancolli level IIB (finger extension only with wrist flexion) (Zancolli, 2003) needed more time to extend their fingers, with their wrist in a neutral position, as it took more effort to achieve this movement against tight or shortened finger flexor muscles. This observation highlights the importance of considering not only how orthosis prescription impacts movement, but how it may also affect the speed and quality of movement of the hand in bimanual activity.

Jackman et al. (2018; 2019) described analysing the movement of the fingers in relation to the child's ability to actively extend their fingers. If the child was unable to extend their fingers with their wrist in 20–30 degrees of wrist extension, the orthosis was customised during fabrication. There was no detail reported as to how the orthosis for the individual child was customised, only the assumption, made by the author of this thesis, that the amount of wrist extension was reduced until the child could extend their fingers. The connection between the movement analysis and respective outcome measures (Canadian Occupational Performance Measure (COPM) (Law et al., 1990); Goal Attainment Scaling (GAS) (Kiresuk et al., 2014) and Box and Blocks Test of Manual Dexterity (Mathiowetz et al., 1985) used in the Jackman et al. (2018; 2019) studies is not presented or discussed in detail. Practice-based knowledge and experience directs that consideration be given not only to analysis of movement pattern and how this guides orthosis prescription, but also to how impaired movement affects the activity itself.

Activity

Activity is the execution of a task or action by an individual (World Health Organisation, 2001). Mobility or movements required to carry, move and handle objects form part of the ICF activity and participation domains and are the focus of this section. Six studies included in the systematic review (Garbellini et al., 2017) used measures of upper extremity function. The studies, the outcome measures used, and the corresponding ICF domains as described by Wallen and Stewart (2016) are presented in Table 3.1.

Table 3.1 Measures of upper extremity function used in included studies

Study	Outcome measure	ICF domain
(Barroso et al., 2011)	Jebsen Taylor Hand Function Test* (Tipton-Burton, 2011)	Activity ± Body Function
(Burtner et al., 2008)	Nine Hole Peg Test (finger dexterity)* (Smith et al., 2000)	Activity ± Body Function
(Jackman et al., 2019)	Box and Blocks Test of Manual Dexterity (Mathiowetz et al., 1985)	Activity ± Body Function
(Jackman et al., 2018)	Canadian Occupational Performance Measure (COPM) (Law et al., 1990)	Activity
	Goal Attainment Scaling (GAS) (Kiresuk et al., 2014)	Activity
(Kanellopoulos et al., 2009)	Quality of Upper Extremity Skills Test (QUEST) (DeMatteo et al., 1993)	Activity ± Body Function
Louwers et al. (2011)	Assisting Hand Assessment (AHA) (Krumlinde-Sundholm et al., 2007)	Activity

Note: ± — plus or minus; * The Jebsen Taylor Hand Function Test and the Nine Hole Peg Test were not described in the paper by Wallen and Stewart (2016) and were classified by the author of this thesis. These tests have predominantly been used with the adult population post stroke and not with children.

In Table 3.1, all but one study, Kanellopoulos et al. (2009), prescribed an orthosis for wear during activity. The studies by Barroso et al. (2011), Burtner et al. (2008) and Jackman et al. (2019) used tests of manual dexterity which focus on unilateral movement to measure changes in activity level outcomes. Wallen and Stewart (2016) classified the Box and Blocks Test of Manual Dexterity (Mathiowetz et al., 1985) within the activity, plus or minus the body function domain of the ICF. It is assumed by the author of this thesis that the classification of the Box and Blocks Test of Manual Dexterity within the body function domain is related to the neuromusculoskeletal and movement related functions required to complete the test. Given the similarity of the other tests of manual dexterity it seems reasonable to classify them within the same ICF domains. The orthoses used in these studies were worn only during the immediate assessment period. In clinical practice, the speed of a child's ability to grasp, release or manipulate an object does not indicate if he/she is unable to execute a task. The link between these actions and the activity that orthosis prescription was aimed at improving is not clearly reported in these studies, apart from the movement of grasp and release.

Jackman et al. (2018) used goal achievement outcome measures — the COPM (Law et al., 1990) and GAS (Kiresuk et al., 2014) — to explore if there was a change in activity performance using a three parallel group RCT. The first group was prescribed an orthosis only and wore it for one hour per day when practicing set goals. The second group received only task-specific training intervention, Cognitive Orientation to daily Occupational Performance (CO-OP) (Cameron et al., 2017), daily for one hour for a period of ten consecutive working days. The third group received a combination of the CO-OP intervention with orthosis wear and an hour of orthosis wear when practicing set goals at home. There were no reported differences in goal achievement with orthosis wear compared to the other groups (Jackman et al., 2018). There was no discussion about the individual participant's activity limitations and if the goals set were due to impaired movement, the lack of use of the hand in specific activities, or other reason (e.g. cognitive strategy use). Again, the link between the activity and orthosis prescription was not clearly articulated.

It is unlikely that upper limb orthotic intervention applies to all children with CP. In practice, no orthosis exists that applies to all people, even those with the same diagnosis (Lannin et al., 2017). This raises the question as to whether an experienced clinician in the field would have prescribed the provided orthoses to all (or any) of the children who received one in the included studies. Detailing and discussing the reported reasons for orthosis prescription using the ICF (World Health Organisation, 2001) framework provided no further clarity to inform clinical practice. This was due to the variability in positioning of muscles and joints (particularly the wrist) to manage impairment at the body structure level; minimal consideration of movement

analysis to guide prescription; and a lack of explicit connection between orthosis prescription and how activity limitations might be addressed. The result of this analysis raised further issues that need consideration when determining rationale for orthosis prescription.

3.2.2 Biomechanical framework

At the body structure and function level, a biomechanical approach, focussing on anatomy, kinetics and kinematics, provides a way to analyse impairment and consider corrective options, including the use of upper limb orthoses (Copley & Kuipers, 2014b; Lannin et al., 2017; Wilton, 2013a). Parallels between the body structure and function domains of the ICF (World Health Organisation, 2001) as discussed above and a biomechanical approach can be made with the studies by Louwers et al. (2011) and Jackman et al. (2018; 2019). In these three studies, consideration was given as to how the body structures of the wrist and fingers moved in relation to each other. These authors appeared to consider the movement limitations observed within their rationales for orthosis prescription. None of the studies directly linked or described the reason for orthosis prescription relative to the dynamic movement observed or the forces and movement created by the anatomical structures of the wrist and hand.

An imbalance between joint, muscle and soft tissue structures of the hand and upper limb (body structure issue) can result in activity limitations (Lannin et al., 2017; Wilton, 2013a). At an activity level, a biomechanical frame of reference in occupational therapy provides an approach to: analyse how a person moves; the effect that movement has on occupational performance; and what intervention to implement to remediate impairment (McMillan, 2011; Rybski, 2012; Wilby, 2007). This activity level approach, to remediate impairment with a prescribed upper limb orthosis, was not explicitly described in any of the studies in the systematic review by Garbellini et al. (2017) or systematic review update.

There is a long standing premise that the static and dynamic aspects of deformity present in the wrist, fingers and thumb during movement of the hand must be considered when prescribing orthoses for children with CP (Wilton, 1983). An understanding of the relationship between the body's anatomy and the forces and motion it causes (biomechanics) and the use of biomechanical principles — such as advantageous applications of force and use of mechanical characteristics of materials to counteract impaired movement — is essential to successful prescription of any upper limb orthosis (Fess, 2011; Wilton, 2013a). Biomechanical considerations for the reasons for upper limb orthosis prescription were not well reflected in the reviewed literature.

3.2.3 Professional reasoning

A clinician's professional reasoning is complex and multi-faceted: it is comprised of layers of professional knowledge based on empirical evidence, practical know-how or practice based knowledge, emotional intelligence and instinct (Unsworth & Baker, 2016). Clinical reasoning occurs during interactions between the client, clinician and environment (Schell, 2019). Common aspects of professional reasoning — including scientific, diagnostic, and procedural reasoning (Schell, 2019) — can be extrapolated to understand the rationale for upper limb orthosis prescription as identified in the systematic review by Garbellini et al. (2017). The aspects of reasoning and their application to orthosis prescription in the studies can be summarised as follows:

- Scientific reasoning focuses on a diagnosis or condition and uses theory-based decision making and statistical evidence to select a treatment approach (Hooper, 1997; Schell, 2019; Unsworth & Baker, 2016). A diagnosis of CP or impairment of body structure and function were the foci of all reasons for orthosis prescription in the studies in the systematic review and review update.
- Diagnostic reasoning uses a blend of science and client-based information to explain why a client is experiencing problems, and to plan and direct client care (Rogers & Holm, 1991; Schell, 2019). Consideration of the client's movement (Jackman et al., 2019; Louwers et al., 2011) and their goals (Jackman et al., 2018) directed reasons for orthosis prescription in these three studies.
- Procedural reasoning may reflect protocols or intervention routines for identified conditions within the intervention setting (Schell, 2019). All studies in the systematic review were based upon research protocols and this was the reason for orthosis prescription. In their inclusion and exclusion criteria, none of the studies discussed or detailed whether the reasoning for orthosis prescription would have formed part of the child's regular care or reflected the organisation's usual practice.

The aspects of professional reasoning described above relate to the interactions between the client and clinician. There was no discussion of the client's personal or environmental factors (World Health Organisation, 2001) such as family support and experience with previous orthotic intervention and how this might influence their choice to participate in orthotic intervention. There was no discussion of the clinical environment, resource demand of time, equipment and material and the therapist's expertise and experience and how this might influence orthosis prescription. It is acknowledged that this lack of discussion, within the studies included in the

review, might be due to several factors. These include: reporting limitations; the investigating organisation's protocols; funding allocation for the study; and the selection of experienced investigators and interventionists for the study. As identified by Garbellini et al. (2017), there is a paucity of information and studies investigating these factors that guide clinicians' decision making when prescribing orthoses for children with CP.

3.3 Resources to guide orthosis prescription for children with CP

Frameworks or systems to organise complex or unfamiliar clinical information such as algorithms, classification systems or decision-making aids may be useful resources for a clinician with little practice-based knowledge specific to orthotic intervention (Copley & Kuipers, 2014a). An algorithm for orthosis prescription to correct deformity and prevent contracture (Lannin et al., 2017), the Neurological Hand Deformity Classification (NHDC) (Wilton, 2013b) and the Hypertonicity Intervention Planning Model (HIPM) (Copley & Kuipers, 1999, 2014a) are available as guides for clinicians considering upper limb orthotic intervention for people with neurologically-based upper limb impairment. They were each developed using practice-based knowledge due to the paucity of empirical evidence in the literature.

The algorithm for correcting deformity and preventing contracture (Lannin et al., 2017), provides a decision making tree to guide orthosis prescription. Decisions are guided by whether the client has: loss of, or is at risk of losing, passive range of motion (plus or minus the presence of hypertonicity); hypermobile joints; a risk of developing long-term pain; or functional limitations. The algorithm provides clear steps to guide the clinical reason for potential orthotic intervention. The algorithm does not consider how an individual's dynamic movement might guide orthosis prescription.

Wilton (1983) published an orthotic assessment profile to guide orthosis prescription for the spastic hand in CP which included consideration of the dynamic and static aspects of hand deformity. Through extended clinical contact in assessing and treating upper limb impairment of children and adults with CP and other neurological conditions, versions of a wrist and hand deformity classification were developed and refined, eventually culminating in the NHDC (Wilton, 2013b). The NHDC is an impairment-based tool that provides a biomechanical approach to classifying hand deformity determined by wrist position and wrist and finger movement (Garbellini & Wilton, 2017). The NHDC differentiates between varied wrist and hand positioning during movement and has potential to aid identification of body function and structure level factors, allowing the clinician to discriminate between positive (e.g. spasticity) and negative (e.g. weakness) factors that can be either harnessed or reduced through intervention

(Copley & Kuipers, 2014a). The NHDC was developed with a focus on impairment. It was not designed to classify activity or participation limitations nor to provide a structure to consider a client's goals. Initial intra and inter-rater reliability of the NHDC has been explored (Georgiades et al., 2014) but validity of the tool has not yet been established.

The HIPM was developed by Copley and Kuipers (1999; 2014a) as an aid to structure clinical reasoning and decision making regarding intervention to maintain the long-term health and function of the upper limb affected by neurological impairment. The model: (i) aids the identification and classification of impairment underlying each client's movement and their adaptation and compensation for that movement presentation; and (ii) uses that information to select appropriate interventions individualised by the client's goals (Copley & Kuipers, 2014a). The HIPM details how to choose interventions based on the individual's upper limb features and movement patterns, while considering the client's needs holistically (Copley & Kuipers, 2014a). The assessment of body function and structure level factors within the HIPM comprises various measures of range of motion, hypertonicity and spasticity commonly used in clinical practice. It also comprises tools such as the Zancolli (2003) and the House Thumb (1981) classifications. These tools, used to classify upper limb characteristics of deformity, have some evidence of reliability but an absence of evidence for validity (McConnell et al., 2011).

The algorithm for correcting deformity and preventing contracture (Lannin et al., 2017), the NHDC (Garbellini & Wilton, 2017; Wilton, 2013b) and the HIPM (Copley & Kuipers, 2014a) are available resources to guide orthosis prescription for children with CP. Previous published versions of the HIPM and NHDC have been available since the years 1999 and 2003 respectively. However, eight studies identified in the systematic review (Garbellini et al., 2017), published after these dates, did not report use of the HIPM and/or NHDC. This may be due to the lack of established validity and/or, an absence of research on the application of these clinical resources.

3.4 Gaps in the literature

Terms found in the literature from the systematic review (Garbellini et al., 2017) were categorised into each corresponding domain of the ICF (World Health Organisation, 2001). The diagrammatic representation of these terms within the ICF framework (World Health Organisation, 2001) is presented in Figure 3.1. The use of dashed lines, instead of solid, visually highlights potential gaps in the rationale for orthosis prescription as identified in the literature.

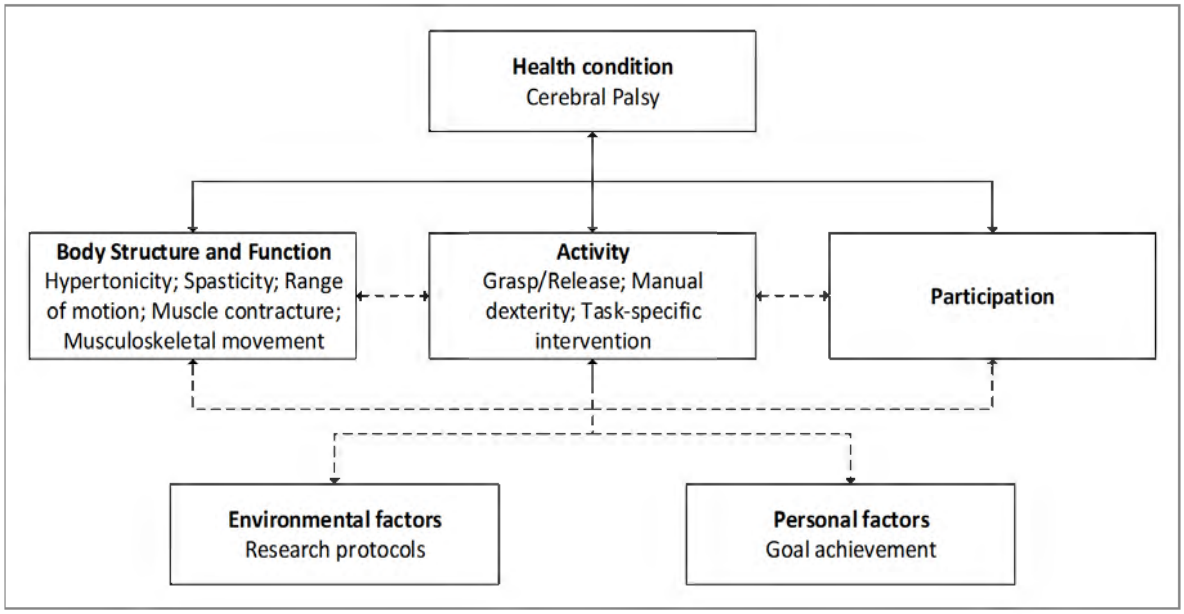

Figure 3.1 Gaps in the orthotic intervention literature as framed within the ICF.

Visual inspection of the ICF framework (World Health Organisation, 2001) (see Figure 3.1) shows the interaction among the ICF domains and the complex factors within each domain, identified in the literature, that a clinician should consider when prescribing an upper limb orthosis for a child with CP. These factors that guide a clinician's decision making were not well represented or explored in the literature. An exploration of what guides an occupational therapist's decision making when prescribing orthoses for children with CP has the potential to unlock important information about when orthoses might be useful.

Commonly used terms identified in the literature were typically categorised within the body structure and function or activity domains of the ICF framework (World Health Organisation, 2001), however little connection was made between these domains and the reasoning for orthosis prescription. This was evidenced by a lack of description of:

- the dynamic interplay of the wrist and finger musculature
- how movement impairment limited activity
- how orthosis prescription could be used to manage this impairment.

An absence of use of the HIPM and NHDC, resources to guide assessment and intervention for neurologically-based upper limb impairment, was also evident in the literature. This gap, in addition to the identified lack of description of the dynamic interplay of the wrist and finger musculature, or link of orthosis prescription to a child's hand deformity, led to a focus on the NHDC.

3.5 Chapter Conclusion

Two recurring themes arose from exploring and discussing the available research literature in more detail. Firstly, the identification of many factors, as framed within the ICF (World Health Organisation, 2001), that may guide clinicians' decision making of upper limb orthosis prescription for children with CP, but a paucity of studies investigating these factors. Secondly, that biomechanical considerations, particularly analysis of dynamic movement guiding orthosis prescription or the use of available clinical resources, were not well reflected or described in the literature.

To address the first theme, the focus of the next study (Study 2, Chapter 4) of this doctoral program was to explore occupational therapists' viewpoints about what guides their clinical practice related to upper limb orthosis prescription for children with CP using qualitative methodology. To address the second theme of a lack of biomechanical consideration for the reason for upper limb orthosis prescription or use of clinical resources, a hand deformity classification system, the NHDC, is considered in more detail. Subsequent chapters of this doctoral program present the NHDC (Chapter 5), explore its psychometric properties (Study 3, Chapter 6) and describe how it may be used in clinical practice (Study 4, Chapter 7).

Chapter Four

4

Factors guiding the prescription of upper limb orthoses for children with cerebral palsy

Preamble 62

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Preamble

Upper limb orthotic intervention for children with CP is common in clinical practice despite the low level of evidence to support this intervention (Lannin & Ada, 2011; Novak et al., 2020). In the previous chapter, reflection and detailed discussion on the systematic review by Garbellini et al. (2017) identified a paucity of information and studies on the factors that guide a clinician's decision making for upper limb orthosis prescription. Given this lack of information, a qualitative cross sectional (Q methodology) study was conducted to identify how occupational therapists viewed the factors that guided their practice of upper limb orthosis prescription for children with CP (published, Study 2 – see Appendix B).

The study was approved by the Human Research Ethics Committee of Australian Catholic University (Appendix D). The viewpoints identified from this qualitative study might inform the development of guidelines for orthosis prescription. This accepted manuscript is presented in the first part of this chapter.

The second part of this chapter presents a critical reflection (accepted manuscript) on the authors' experience using Q methodology (Garbellini et al., 2020) (see Appendix B for the published version). Frank and Polkinghorne (2010) stated that new researchers seek information about qualitative methodologies and acquire standards from published examples. This critical reflection paper was the result of an identified lack of published information in the literature regarding the factor analysis stage of Q methodology. The aim of the paper was to add experiential information for new researchers navigating Q methodology.

PUBLICATION 2 – PRESCRIBING UPPER LIMB ORTHOSES FOR CHILDREN WITH CEREBRAL PALSY: A Q METHODOLOGY STUDY OF OCCUPATIONAL THERAPISTS' DECISION MAKING

Publication details

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[2] See Appendix B, Section B.2 for the published edition of this paper

Abstract

Purpose: This study identified occupational therapists' viewpoints that guide their practice of upper limb orthosis prescription for children with cerebral palsy.

Methods: A qualitative study utilising Q methodology explored participants' viewpoints. Thirty-nine occupational therapists (38 females) were purposively recruited to rank statements generated from interviews of experienced clinicians and peer reviewed and published literature. Statements about reasons for orthoses prescription, were ranked according to what guides decision making the most to least. Data from ranked statements were analysed using by-person factor analysis to reveal the different ways statements were grouped. The resultant factors, based on the average arrangement of statements associated with each factor, were interpreted and named as viewpoints.

Results: Viewpoints identified: 1. Potential effect of the orthosis (n=12 sorts); 2. Biomechanical presentation (n=12 sorts); and 3. Client-Therapist relationship (n=10 sorts). The 'Client's goals' statement was ranked highest across all viewpoints.

Conclusion: Viewpoints identified may inform development of clinical guidelines. Further research is required to (i) identify valid and reliable classification and assessment tools to guide decision making; and (ii) establish the mechanism of the effect of orthotic intervention by considering the link between the biomechanical purpose of the orthosis (e.g., mobilise tissue) and aim of intervention (prevent contracture).

4.1 Introduction

In daily practice, clinicians make a range of clinical decisions using knowledge about factors internal and external to their clients based on their clinical reasoning. Clinical reasoning is defined as all aspects of thinking that guides clinical practice (Fleming, 1991a; Rogers, 1983; Unsworth, 2004). Rogers (1983) stated the goal of clinical reasoning is the development of a treatment recommendation in the interests of the particular client. Clinical reasoning is therefore shaped by the client and/or caregiver's concerns and treatment recommendations are individualised within the requirements of the client's particular situation (Cohn, 1991; Fleming, 1991a; Mattingly, 1991). Multiple approaches to reasoning are inherent in clinical practice (Schwartz, 1991) with reasoning varying according to the clinician's level of expertise (Harries & Harries, 2001).

Clinical decision making for clients with neurologically based upper limb impairment is complex, challenging and multifaceted (Copley & Kuipers, 1999, 2014a; Kuipers et al., 2009; Wilton, 2013b). In children with cerebral palsy (CP) decision making may be framed within the International Classification of Functioning, Disability and Health: Child and Youth (IFC-CY) (World Health Organisation, 2007). Consideration is given to internal factors within the body function and structure, activity and participation domains and to external factors in context of the child's family system and influence of their surrounding environment (World Health Organisation, 2007). Factors internal to the client include personal attributes, neurological musculoskeletal components, severity of condition and associated impairments. Factors external to the client include economic and service provision factors, and environmental factors such as interactions and opportunities provided by parents, caregivers or service providers, and clinician's knowledge and experience (Copley & Kuipers, 2014a; Kuipers et al., 2006; World Health Organisation, 2007).

A clinician's knowledge, experience and service environment guide their process of clinical reasoning (Fleming, 1991b; Mattingly & Fleming, 1994; Unsworth, 2004). Underpinning this reasoning is the clinician's world view, or outlook on life and the world, characterised by personal opinions, interpretations, biases, values, motivation and personal style (Barbosa et al., 1998; Hooper, 1997; Unsworth & Baker, 2016). Individuals do not show homogeneity or similarities in their world view, even following similar professional training (Wigger & Mrtek, 1994). Research from 15 years ago indicated that although therapists valued the application of research-based evidence to practice, they relied more heavily on clinical experience when making practice decisions (Bennett et al., 2003). Current practice aims to further integrate clinical evidence with clinical experience, although history suggests orthotic intervention

corrects deformity and prevents contracture, the evidence is limited and less convincing (Lannin et al., 2017). So understanding what guides current decision making is important.

Practice decisions regarding orthosis prescription require attention to three broad elements which include: (i) occupational goals of the individual; (ii) up-to-date information regarding efficacy, design, materials and wearing regimes; and (iii) relevance and practicality within the individual's life situation (Lannin et al., 2017). Currently there are no practice specific, research informed guidelines, regarding upper limb orthosis prescription for children with CP. The purpose of this study was to gain evidence on occupational therapists' viewpoints about orthosis prescription within current clinical practice in Australia. Increased knowledge regarding what therapists consider important (Wagman et al., 2012) when prescribing upper limb orthoses may inform the development of clinical guidelines.

The aim of this study was to identify occupational therapists' world views (or viewpoints) that guide their clinical practice related to orthosis prescription for children with CP. In the current absence of evidence regarding when orthotic intervention should be considered, what orthoses to use for which children, and timing and length of wear (Copley & Kuipers, 1999; Jackman et al., 2014b; Novak et al., 2013), it is expected that the viewpoints identified from those practising in this area can be used to inform the development of specific guidelines and future research to address some of these issues for this complex and varied clinical area.

4.2 Methods

This study was approved by the Human Research Ethics Committee of Australian Catholic University (2017-54E) in Victoria, Australia. The research activities conformed to the provisions of the Declaration of Helsinki. Informed written consent was obtained from each participant prior to data collection and data were de-identified to maintain confidentiality.

A qualitative cross-sectional study utilising Q methodology was conducted. Q methodology has an exploratory focus and is concerned with the individual viewpoint of participants (Brown, 1993; Dennis, 1986; Dziopa & Ahern, 2011; Van Exel & De Graaf, 2005). Q methodology maintains the subjectivity of participants using rigorous objective processes (Corr, 2006). Participants provide their perspective by sorting statements related to the topic in question (Newman & Ramlo, 2010). Consistent with the method, no definitive hypothesis or theory was proposed or tested for this study (Watts & Stenner, 2012). Q methodology involves the following stages: (1) concourse development, (2) development of the Q sample, (3) selection of the participant set (P set), (4) Q sorting, (5) factor analysis, and (6) interpretation of factors (Brown,

1970; Chee et al., 2015; Corr, 2006; Donner, 2001; Dziopa & Ahern, 2011; Van Exel & De Graaf, 2005; Watts & Stenner, 2012).

4.2.1 Concourse development

A body of knowledge, comprised of a collection of thoughts, ideas and information, referred to as the concourse in Q methodology, was generated from relevant sources related to upper limb orthosis prescription (Brown, 2008; Corr, 2001; Cross, 2005; McKenzie et al., 2011; Van Exel & De Graaf, 2005; Watts & Stenner, 2012). The systematic review by Garbellini et al. (2017) provided information regarding rationale for orthosis prescription and recommendations for orthoses use from a rigorous and systematic literature search. Published information, including textbooks, on upper limb orthosis prescription also yielded information included in the concourse. In addition, information sourced and transcribed from three open-ended telephone interviews with experienced occupational therapists was included in the concourse. Each occupational therapist had more than 15 years of experience in the prescription and fabrication of upper limb orthoses for children with CP. The development of the concourse laid the foundation from which the Q sample was drawn (Brown, 1993; Chee et al., 2015; Dziopa & Ahern, 2011).

4.2.2 Development of the Q sample

The Q sample is a set of statements representative of the major ideas and existing opinions on the topic (Barbosa et al., 1998; Van Exel & De Graaf, 2005; Watts & Stenner, 2012). Initial statements were drawn from the concourse and categorised utilising the domains of the ICF-CY (World Health Organisation, 2007) by the researchers. The statements (n=102) were reviewed within a focus group involving four occupational therapists, reflecting the sample of intended participants, independent to the research team and different from those previously interviewed. This group worked to remove duplicate ideas and combine similar statements. The statements were refined to 42 by the focus group. The researchers reviewed the 42 statements and removed two statements due to ambiguity. A senior speech and language pathologist independently reviewed the statements for readability and recommended changes that resulted in one statement becoming redundant. Once recommended changes were made, the statements were run through freely available readability software to ensure that the level of readability did not exceed a year 12 education level. As a result of these processes, the statements were further refined from 42 to a total of 39. The focus group was sent the 39 refined statements for comment and no additional changes were suggested. The final Q sample consisted of 39 statements.

To enhance the rigour of the study, the Q sorting process (utilising the 39 Q sample statements) was piloted with a purposively selected sample of three occupational therapists. This provided a further check in regard to whether the statements were comprehensive and that there were no irrelevant or obviously missing statements. The pilot also sought to identify if there was any perceived burden of sorting on the potential participants and that the sorting could be completed in an acceptable timeframe (i.e., no longer than 30 minutes).

4.2.3 Selection of the P set

The P set are the participants who sorted the Q sample statements (Van Exel & De Graaf, 2005). The P set was not random: participants were occupational therapists chosen based on their relevance to the study's aim, rather than their representativeness of a larger population (Van Exel & De Graaf, 2005). Large numbers of occupational therapists were not required as the aim of Q methodology was to identify the key opinions of the selected participant group (Thompson et al., 2016; Watts & Stenner, 2005). Disparity exists in the literature as to the number of participants for Q methodology (Dziopa & Ahern, 2011). Common sizes of participants in Q methodology studies are between 20 and 40 participants (Corr, 2006). Watts and Stenner (2005) recommend maintaining at least a 1:1 ratio of participants to Q sample statements and to avoid including many more participants than statements.

The number of participants determined for the study was 39, a 1:1 ratio of participants to Q sample statements. Occupational therapy and research managers from community service providers, not-for-profit organisations and tertiary health services around Australia were approached, inviting occupational therapists on staff to participate in the study. Networking and direct contact with colleagues of one of the researchers (SG) was also utilized for recruitment. Occupational therapists were included in the study provided they were currently active, or had worked, in treating children with CP, regardless of length of time, and they consented to participate. Demographic information describing the participants, including gender, years of clinical experience and description of work organisation was collected.

4.2.4 Q sorting

From October to December 2017 participants were recruited and ranked Q sample statements along a continuum of, "guides the least" to "guides the most" on a quasi-normal distribution grid. The grid accommodated the 39 statements with a distribution ranging from -5 to +5 (total 11). Figure 4.1 shows a participant sorting the statements and an example of the quasi-normal distribution grid. All sorts were done face to face with the same researcher (SG) at

the participant's place of work or a place convenient for the participant. The instructions set the conditions for how the participants approached and completed the sort (Barbosa et al., 1998; Brown, 1970; Watts & Stenner, 2012). In this study the instructions directed the participants to rank the statements according to what guides their decision making the most, to the least, in response to the question: What guides your decision making when prescribing upper limb orthoses (splints) for children with cerebral palsy?

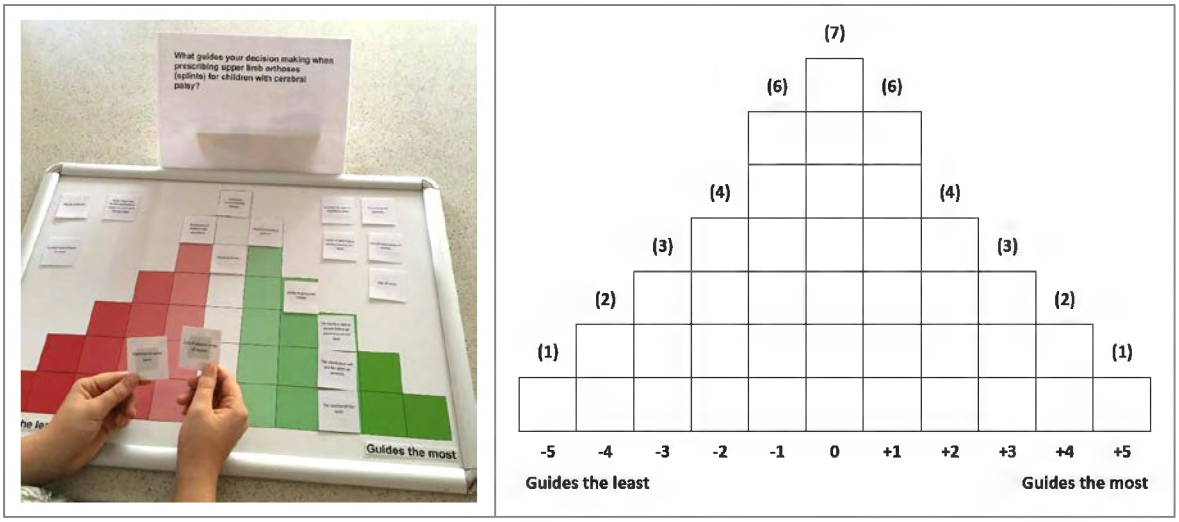

Figure 4.1 Participant sorting statements and an example of the quasi-normal distribution grid

Following completion of the Q sort the participants were asked: (i) Do you think any factors are missing that guide your decision making? (ii) If yes, please describe them; (iii) Are you satisfied with your final sort? and (iv) Do you have any comments? Data from participant comments provided additional information whether statements were missing and assisted in the interpretation and naming of the factors or viewpoints. To maintain the same set of conditions for all participants, regardless of identification of perceived missing items, the same number and wording of statements was provided to every participant.

4.2.5 Factor analysis

Factor analysis was employed to reduce the larger number of Q sorts to smaller groups of factors, usually two to five, representing a common perspective (Brown, 2008). Factor analysis was conducted utilising Ken-Q Analysis software, which was freely available online (Banasick, 2018). This involved entering the study parameters: (i) the name of the project; (ii) the Q sample (39 statements); (iii) the Q sort range (-5 to +5) on the quasi-normal distribution grid; (iv) the number of statements allocated for each column on the grid (see the numbers in brackets above each column in Figure 4.1); and (v) each participant's completed Q sort (statement rankings on the grid). A correlation matrix was then generated as an interim step that aimed to reveal factor

structure (Brown, 1993). A principle component analysis was run to examine the correlation matrix and extract underlying factors that revealed the number of qualitatively different ways the various Q sorts were organised by the participants (Barbosa et al., 1998; Brown, 2008; Corr, 2006; Wigger & Mrtek, 1994).

Factors were rotated using varimax rotation to identify how variables grouped together (Coakes, 2010) and to maximise how much the set of observations differed from each other (variance) (Stevens, 2002). Factor loadings provided the correlation between each of the Q sorts and the retained factors (Brown, 1993; McKeown & Thomas, 2013) and uncovered which Q sorts significantly loaded onto each factor. Factor loadings, with a value of 2.58 times the standard error of each Q sort (statistically significant at $p < 0.01$), were used to identify the Q sorts correlated with each factor (Brown, 1980, 1993; McKeown & Thomas, 2013; Newman & Ramlo, 2010). Standard error (SE) was calculated using the formula $SE=1/\sqrt{n}$, where n is the number of statements in the Q sample (Brown, 1993; McKeown & Thomas, 2013). Therefore, loadings of $> \pm 0.4131$ (calculated by $2.58(1/\sqrt{39})$) (McKeown & Thomas, 2013) identified the Q sorts correlated with each factor. Q sorts that significantly loaded onto more than one factor or did not significantly load onto any of the factors were excluded from further analysis. The weighted average statement score (factor score) of the Q sorts that defined each factor were calculated (Van Exel & De Graaf, 2005) and standardised to reflect a z-score (Atchley, 2007).

Representative Q sorts, demonstrating the average Q sort arrangement associated with each factor, were created by listing each statement in order of largest positive to largest negative z-score for each factor. The z-score determined where each statement was placed on the sorting grid of the representative Q sort (Brown, 1980; Donner, 2001; McKeown & Thomas, 2013; Newman & Ramlo, 2010; Wagman et al., 2012). Statements placed in statistically significantly locations between any pair of factors were identified as distinguishing statements. Statements that did not distinguish between any pair of factors were identified as consensus statements (Dziopa & Ahern, 2011). These final factors were then interpreted and named as viewpoints.

4.2.6 Interpretation of factors

The interpretation and naming of collective viewpoints was informed by the qualitative synthesis of three key outputs generated by the Ken Q analysis software: (i) the representative Q sorts; (ii) the distinguishing statements, positioned in the section of the grid that guided decision making the most in each of the representative Q sorts; and (iii) the consensus statements. Comparison of the position of statements on each representative Q sort, how the placement of statements distinguished the sorts from one another or demonstrated consensus, provided the framework for interpreting and naming each viewpoint.

4.3 Results

Analysis of the Q sorts completed by 39 of 41 participants approached (38 female; 1 male) resulted in three factors, each representing a viewpoint. The two participants approached who did not complete the Q sort were time limited when approached and subsequently, once 39 participants were reached, were no longer required to participate. The proportion of female to male participants is representative of 91.2% of females compared to 8.8% of males registered to practice occupational therapy in Australia in 2017 (Occupational Therapy Board of Australia, 2017). The years of the participants' practice, including years of practice with children with CP, their highest level of tertiary education, at the time of Q sort completion, and their self-reported level of experience with orthotic intervention (King et al., 2008) is displayed for each participant in Table 4.1 and arranged according to factor loadings associated with their Q sort. Thirty four Q sorts correlated with one of the three viewpoints (numbers in bold in the table). Five of the 39 Q sorts, not bolded, significantly loaded on more than one of the viewpoints and were therefore excluded from further analysis.

Table 4.2 displays the z-score of each statement, its rank (1-39) and represented Q sort value in parentheses (i.e. number on the grid) for each viewpoint. All statements are shown in descending order from distinguishing to consensus. Numbers in bold indicate a distinguishing statement for the individual viewpoints.

Table 4.1 **Participant demographics arranged according to factor loading**

	Participant demographics for each sort				Factor Loading		
Q sort	Years of clinical experience	Years of experience with CP	Highest level of education	Orthotic intervention experience	Factor 1	Factor 2	Factor 3
Q sort 11	19	16	Masters	Experienced	**0.8572**	0.0508	0.1479
Q sort 10	10	5	Bachelor	Intermediate	**0.7779**	0.0231	0.2894
Q sort 20	5	4	Bachelor	Novice	**0.6736**	0.5016	0.1322
Q sort 5	28	28	PhD	Experienced	**0.6494**	0.2301	0.2894
Q sort 31	23	3	Bachelor	Intermediate	**0.5879**	0.4430	0.3298
Q sort 38	30	26	Bachelor	Intermediate	**0.5743**	-0.0654	0.4140
Q sort 6	5	2	Honours	Intermediate	**0.5719**	0.3842	0.2897
Q sort 29	2	2	Honours	Novice	**0.5646**	0.3454	0.1460
Q sort 25	2	2	Bachelor	Novice	**0.5443**	0.1282	0.3032
Q sort 28	15	7	Masters	Novice	**0.5418**	0.0601	0.4561
Q sort 32	7	7	Bachelor	Intermediate	**0.4832**	0.0958	0.2579
Q sort 12	5	1	Bachelor	Novice	**0.4648**	0.3579	-0.0060
Q sort 2	14	7	Bachelor	Intermediate	0.2305	**0.7949**	0.2489
Q sort 33	7	5	Bachelor	Intermediate	-0.0279	**0.7359**	0.1006
Q sort 14	12	2	Honours	Experienced	0.3645	**0.6935**	0.2855
Q sort 34	5	5	Bachelor	Intermediate	0.5373	**0.6894**	-0.1090
Q sort 35	27	7	Post Grad	Experienced	-0.0301	**0.6475**	0.4825
Q sort 1	25	23	Bachelor	Intermediate	0.3516	**0.6077**	0.4125
Q sort 39	20	20	Honours	Intermediate	0.5532	**0.6051**	0.1122
Q sort 21	7	5	Honours	Intermediate	0.5336	**0.5847**	0.0623
Q sort 15	18	18	PhD	Experienced	-0.1797	**0.5756**	0.2888
Q sort 9	42	35	PhD	Experienced	0.1073	**0.5224**	0.4950
Q sort 30	10	8	Bachelor	Intermediate	0.3354	**0.5096**	0.3216
Q sort 3	25	25	Bachelor	Intermediate	0.3116	**0.4807**	0.2819
Q sort 16	25	9	Post Grad	Novice	0.1328	0.1796	**0.7947**
Q sort 17	25	1	Bachelor	Novice	0.3370	0.1223	**0.7353**
Q sort 22	18	15	Post Grad	Intermediate	0.2048	0.1732	**0.7263**
Q sort 18	6	6	Honours	Intermediate	0.3171	0.3301	**0.7112**
Q sort 7	12	7	Post Grad	Intermediate	0.3282	0.0767	**0.6774**
Q sort 19	17	13	Post Grad	Intermediate	0.2201	0.3141	**0.6553**
Q sort 13	11	11	Honours	Experienced	0.0860	0.3437	**0.6510**
Q sort 23	9	7	Honours	Intermediate	0.2680	0.4330	**0.6378**
Q sort 36	23	4	Honours	Intermediate	0.3083	0.4415	**0.6305**
Q sort 4	35	20	Masters	Novice	0.5573	0.0052	**0.5574**

	Participant demographics for each sort				Factor Loading		
Q sort	**Years of clinical experience**	**Years of experience with CP**	**Highest level of education**	**Orthotic intervention experience**	**Factor 1**	**Factor 2**	**Factor 3**
Q sort 8	25	25	PhD	Experienced	0.4226	0.4833	0.3896
Q sort 24	8	6	Bachelor	Novice	0.3615	0.5691	0.4572
Q sort 26	5	2	Bachelor	Novice	0.5222	0.4730	0.4003
Q sort 27	12	12	Bachelor	Intermediate	0.4296	0.3386	0.4627
Q sort 37	11	9	Masters	Intermediate	0.5219	0.3660	0.5154
				Percentage of explained variance	20	19	20
				Number of defining variables	12	12	10
				Factor score correlations	**Factor 1**	**Factor 2**	**Factor 3**
				Factor 1	1	0.6087	0.6576
				Factor 2	0.6087	1	0.6463
				Factor 3	0.6576	0.6463	1

Note: numbers in bold indicate Q sorts highly correlated with that factor ($p < 0.01$); CP cerebral palsy

Table 4.2 Q sample statements, statement z-scores, rank and Q sort value for each factor

Q sample statements	Factor 1		Factor 2		Factor 3		
	Z-score	Rank (Q sort value)	Z-score	Rank (Q sort value)	Z-score	Rank (Q sort value)	Z-score variance
25. Evidence in the literature to support the use of a splint	**1.12**[a]	**5 (3)**	**-0.46**[a]	**24 (-1)**	**-1.61**[a]	**37 (-4)**	1.256
29. The client/carer is able to follow the recommendations for wearing the splint	0.72	11 (1)	**-0.85**[a]	**32 (-2)**	0.81	9 (2)	0.584
2. The position of the thumb	**-0.61**[a]	**29 (-1)**	**1.13**	**6 (3)**	**0.64**	**10 (2)**	0.539
28. The client/carer will put the splint on correctly	**0.5**	**13**	**-0.72**[a]	**28 (-1)**	**0.95**	**8 (2)**	0.499
14. Potential to cause harm	**1.46**[a]	**3 (4)**	**-0.14**	**22 (0)**	**0.3**	**15 (1)**	0.456
33. The carers recognise and understand the need for the splint	**-0.04**[a]	**22 (0)**	**-0.59**[a]	**27 (-1)**	**0.99**[a]	**6 (3)**	0.43
22. Knowledge of pathophysiology of cerebral palsy	-0.47	27 (-1)	-0.35	23 (0)	**0.97**[a]	**7 (2)**	0.424
1. The position of the wrist	**-0.52**[a]	**28 (-1)**	**1.02**[a]	**7 (2)**	**0.41**[a]	**12 (1)**	0.404
21. Knowledge of anatomy and kinesiology	**-1.2**[a]	**35 (-3)**	-0.05	20 (0)	0.27	18 (0)	0.4
19. Ability to grasp and release	-0.38	25 (-1)	**0.94**[a]	**8 (2)**	-0.16	24 (-1)	0.337
7. Atypical movement patterns	-0.75	30 (-2)	**0.38**[a]	**17 (0)**	-0.86	31 (-2)	0.313
36. The client is willing to wear the splint	0.48	15 (1)	0.25	18 (0)	**1.45**[a]	**2 (4)**	0.271
15. Wrist, finger and thumb movement in relation to each other during a task	0.11	18 (0)	**1.23**[a]	**5 (3)**	0.14	20 (0)	0.271
12. Impact of splinting on sensory input to the hand	**0.35**	**17 (0)**	**-0.06**	**21 (0)**	**-0.9**[a]	**32 (-2)**	0.269
11. Skin hygiene	**1.1**[a]	**6 (3)**	**0.41**	**15 (1)**	**-0.12**	**23 (0)**	0.251
24. Success and failure with previous splint prescription	**0.39**	**16 (1)**	**-0.8**[a]	**31 (-2)**	**-0.12**	**22 (0)**	0.238
38. Client's goals	2.03	1 (5)	**1.25**[a]	**4 (3)**	2.4	1 (5)	0.227
5. Loss of active range of motion	0.48	14 (1)	**1.38**[a]	**3 (4)**	0.31	14 (1)	0.221
27. The family is able to ensure follow up appointments are kept	0.05	19 (0)	**-1.03**[a]	**35 (-3)**	-0.24	26 (-1)	0.207

Q sample statements	Factor 1		Factor 2		Factor 3		Z-score variance
	Z-score	Rank (Q sort value)	Z-score	Rank (Q sort value)	Z-score	Rank (Q sort value)	
31. Treatment such as Botulinum toxin and surgery	0.78	9 (2)	0.53	14 (1)	**-0.16**[a]	**25 (-1)**	0.158
3. The severity of spasticity	**-0.22**	**24 (-1)**	**0.75**	**11 (1)**	**0.28**	**16 (1)**	0.158
6. Loss of passive range of motion	**0.54**[a]	**12 (1)**	1.46	2 (4)	1.17	5 (3)	0.148
20. Access to resources for splint fabrication	-1.98	38 (-4)	-2.06	38 (-4)	**-1.29**[a]	**35 (-3)**	0.12
4. The severity of dystonic type movements	-0.02	21 (0)	0.18	19 (0)	**-0.63**[a]	**29 (-1)**	0.12
18. Potential for splint to impede function	1.01	7 (2)	0.83	10 (2)	**0.26**[a]	**19 (0)**	0.102
9. Upper limb neglect	-0.99	32 (-2)	**-0.46**[a]	**25 (-1)**	-1.23	34 (-3)	0.102
17. Potential for splint to improve function	1.39	4 (3)	**2.04**[a]	**1 (5)**	1.4	4 (3)	0.094
34. Age of client	**-1.3**[a]	**36 (-3)**	-0.74	29 (-1)	-0.61	28 (-1)	0.089
10. Presence of pain	1	8 (2)	0.7	12 (1)	**0.28**	**17 (0)**	0.088
16. Current level of hand function	1.48	2 (4)	**0.87**[a]	**9 (2)**	1.43	3 (4)	0.077
30. Ease of care and handling for caregivers	-0.11	23 (0)	**-0.56**	**26 (-1)**	0.1	21 (0)	0.076
13. Secondary musculoskeletal changes	0.04	20 (0)	0.64	13 (1)	0.38	13 (1)	0.061
35. The client's perceived success or failure with previous splint use	-0.41	26 (-1)	-0.95	34 (-3)	-0.59	27 (-1)	0.05
26. Health professional request for a splint [b]	-1.05	33 (-2)	-1.31	36 (-3)	-1.55	36 (-3)	0.042
23. Proficiency in splint fabrication [b]	-1.68	37 (-4)	-1.49	37 (-4)	-1.97	39 (-5)	0.039
37. Splint tolerance [b]	0.76	10 (2)	0.4	16 (1)	0.6	11 (1)	0.023
8. Muscle weakness [b]	-1.07	34 (-3)	-0.76	30 (-2)	-0.9	33 (-2)	0.016
32. Access to funding for the splint [b]	-2.11	39 (-5)	-2.12	39 (-5)	-1.86	38 (-4)	0.014
39. The client wants to change the look of their arm and hand [b]	-0.89	31 (-2)	-0.91	33 (-2)	-0.75	30 (-2)	0.005

Note: $p < 0.05$: ([a]) indicates Significance at $p < 0.01$. Numbers in bold indicate a distinguishing statement for the individual factor. ([b]) indicates a consensus statement. All statements are shown in descending order from distinguishing to consensus.

Three representative Q sorts were derived from the data and interpreted as representing the following collective viewpoints: *1. Potential effect of the orthosis; 2. Biomechanical presentation; and 3. Client therapist relationship.*

4.3.1 Viewpoint 1: Potential effect of the orthosis

Viewpoint 1 (derived from Factor 1) accounted for 20% of the explained variance with 12 Q sorts significantly associated with this factor (see Table 4.1). The representative Q sort for viewpoint 1 is shown in Figure 4.2. This viewpoint was characterised by the cluster of positively ranked statements on the right side of the grid, representing the potential effect of the orthosis in guiding decision making the most. Positive z-scores of the distinguishing statements within this cluster of statements (see Table 4.2) for this viewpoint included: *Potential to cause harm* (Q sort value 4); *Evidence in the literature to support the use of a splint* (Q sort value 3); and *Skin hygiene* (Q sort value 3).

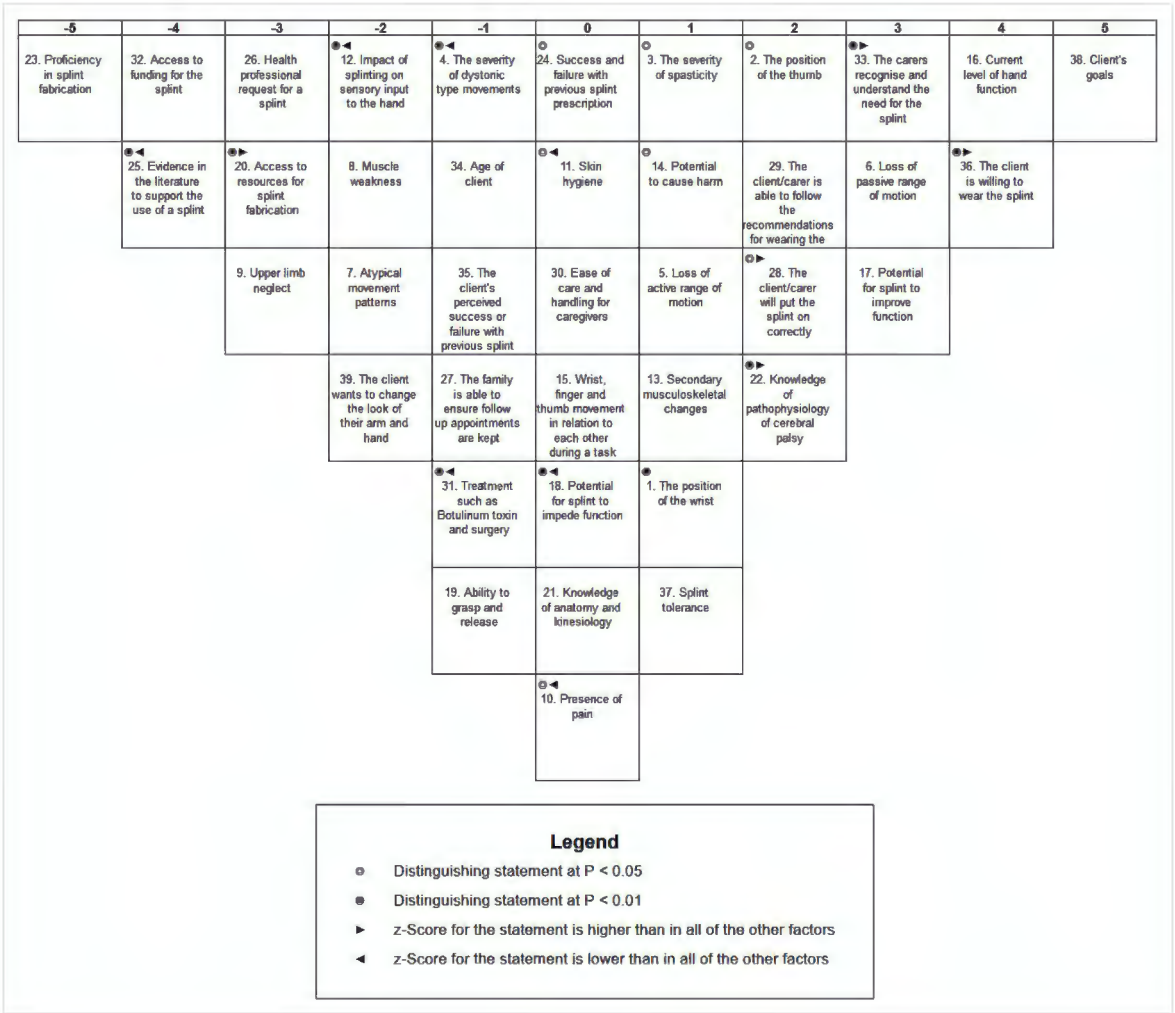

Figure 4.2 Representative Q sort for viewpoint 1

4.3.2 Viewpoint 2: Biomechanical presentation

Viewpoint 2 (derived from Factor 2) accounted for 19% of the explained variance with 12 Q sorts significantly associated with this factor (see Table 4.1). The representative Q sort for viewpoint 2 is shown in Figure 4.3. This viewpoint was characterised by the cluster of positively ranked statements on the right side of the grid, representing the perspective of biomechanical presentation guiding decision making the most. Positive z-scores of the distinguishing statements within this cluster of statements (see Table 4.2) for this viewpoint included: *Potential for splint to improve function* (Q sort value 5); *Loss of active range of motion* (Q sort value 4); *Client's goals* (Q sort value 3); *The position of thumb* (Q sort value 3); *Wrist, finger and thumb movement in relation to each other during a task* (Q sort value 3); *The position of the wrist* (Q sort value 2); *Ability to grasp and release* (Q sort value 2); *Current level of hand function* (Q sort value 2); and *Severity of spasticity* (Q sort value 1).

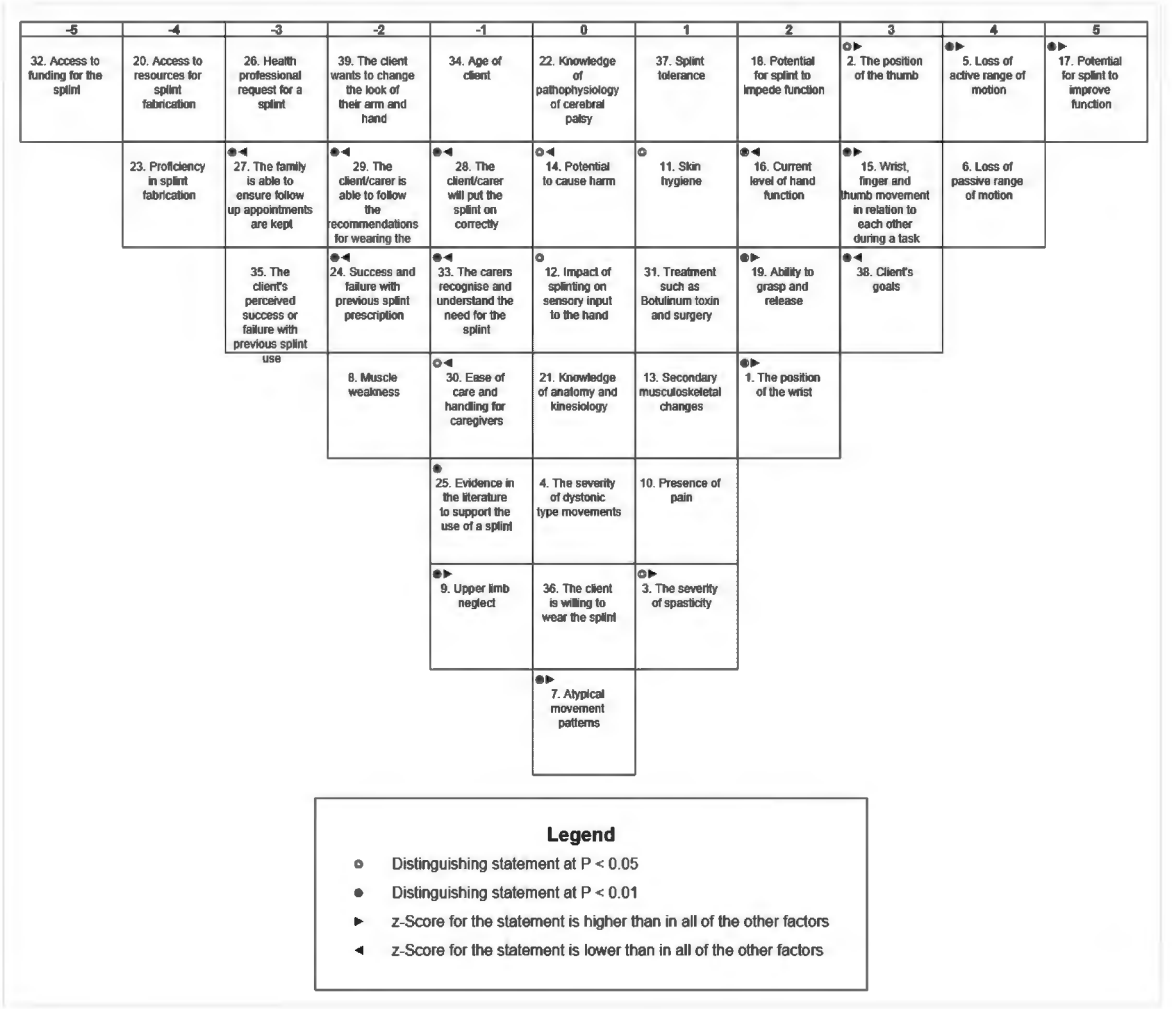

Figure 4.3 Representative Q sort for viewpoint 2

4.3.3 Viewpoint 3: Client-Therapist relationship

Viewpoint 3 (derived from Factor 3) accounted for 20% of the explained variance with 10 Q sorts significantly associated with this factor (see Table 4.1). The representative Q sort for viewpoint 3 is shown in Figure 4.4. This viewpoint was characterised by the cluster of positively ranked statements on the right side of the grid, representing the partnership between the child/carer(s) and therapist guiding decision making the most. Positive z-scores of the distinguishing statements within this cluster of statements (see Table 4.2) for this viewpoint included: *The client is willing to wear the splint* (Q sort value 4); *The carers recognise and understand the need for the splint* (Q sort value 3); *The position of thumb* (Q sort value 2); *The client/carer will put the splint on correctly* (Q sort value 2); and [The therapist's] *Knowledge of pathophysiology of cerebral palsy* (Q sort value 2).

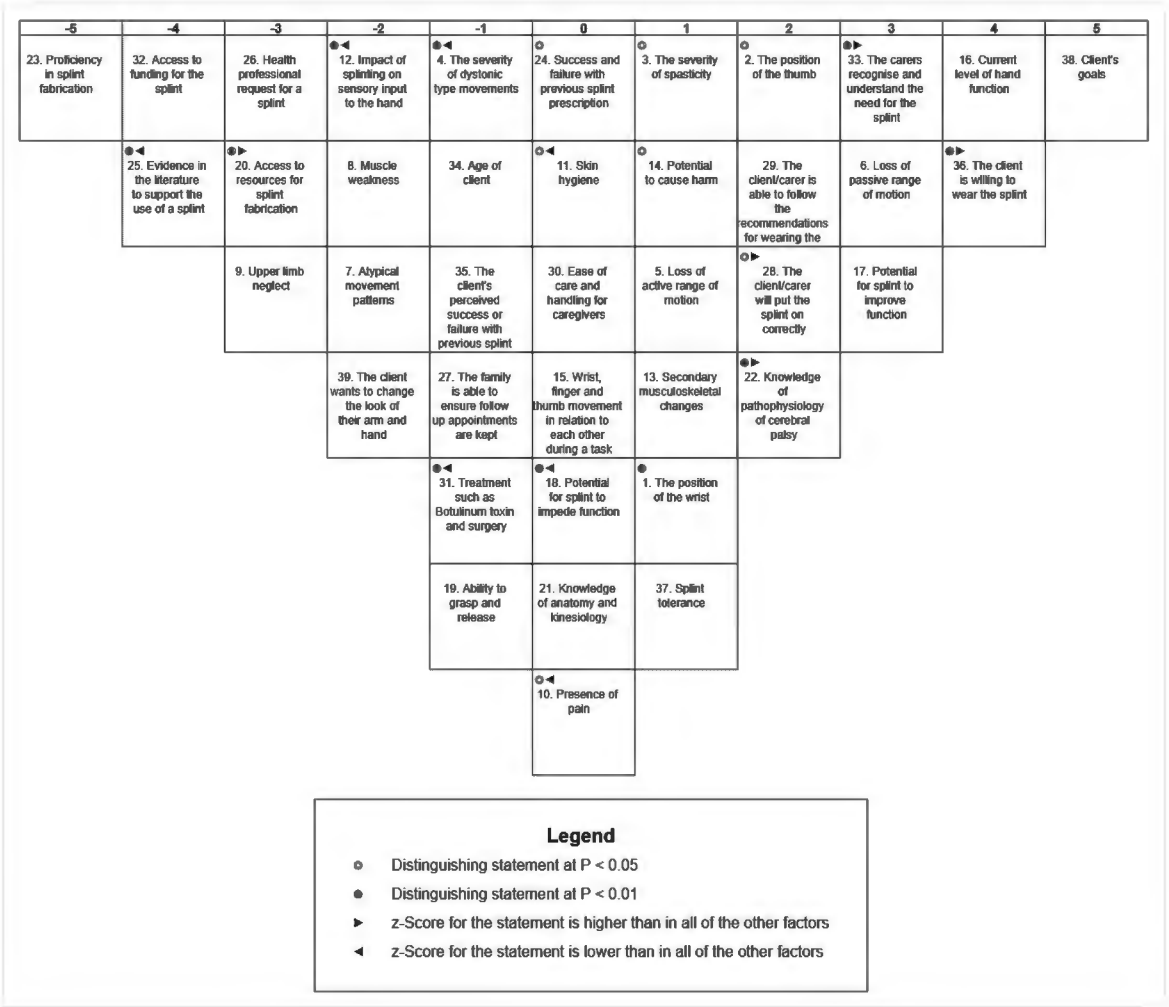

Figure 4.4 Representative Q sort for viewpoint 3

Six consensus statements were identified (see Table 4.2) with no statistically significant difference in Q sort value across all three viewpoints. The statements were ranked in a similar

position across all viewpoints on the Q sort grid indicating agreement on how much the statement guided decision making regarding orthosis prescription. Occupational therapists agreed that *splint tolerance* had a positive ranking (viewpoint 1: Q sort value 2, viewpoint 2: Q sort value 1, viewpoint 3: Q sort value 1). There was also agreement with the negative ranking of some statements: *the client wants to change the look of their hand* (viewpoint 1: Q sort value -2, viewpoint 2: Q sort value -2, viewpoint 3: Q sort value -2); *muscle weakness* (viewpoint 1: Q sort value -3, viewpoint 2: Q sort value -2, viewpoint 3: Q sort value -2); *health professional request for splint* (viewpoint 1: Q sort value -2, viewpoint 2: Q sort value -2, viewpoint 3: Q sort value -3); *proficiency in splint fabrication* (viewpoint 1: Q sort value -4, viewpoint 2: Q sort value -4, viewpoint 3: Q sort value -5); and *access to funding for splint* (viewpoint 1: Q sort value -5, viewpoint 2: Q sort value -5, viewpoint 3: Q sort value -4). These negatively ranked statements were consistently considered to guide decision making the least.

Twenty five participants responded they did not think there were any missing statements. Missing statements identified by 11 participants could generally be mapped to one of the existing statements. For example, "improving function for task specific activity" (Participant 38) could be mapped to statement 17, *potential for splint to improve function*. Some of the suggested statements were based on personal and environmental contextual factors (World Health Organisation, 2007). For example, "child's sleep and level of physical involvement" (Participant 5), "clinical anxiety in the child" (Participant 8), and "climate/environment" (Participant 39) were mapped to statement 35, *the client's perceived success or failure with previous splint use,* statement 36, *the client is willing to wear the splint*, and statement 37, *splint tolerance* respectively. When asked if they were satisfied with their final sort: 24 (62%) participants said yes; 11 (28%) said they were mostly satisfied; and 4 (10%) said they were not. The reasons for not being satisfied with the final sort included: the individualised nature of decision making for splinting, including goals and scientifically gained evidence; difficulty generalising decision making; and difficulty with the forced sort of the statements as many of the statements guided decision making.

4.4 Synthesis of results

The most highly ranked statement across the three viewpoints, based on z-score, rank (1-39) and Q sort value, was *client's goals*. This statement's z-score for viewpoints 1, 2 and 3 was 2.03 (rank 1), 1.25 (rank 4) and 2.03 (rank 1), respectively. The average Q sort value, in order, on the representative Q sort was five, three and five. Even though the *client's goals* statement was not a statistically significant consensus statement among all three viewpoints, its highly ranked position indicates the primary regard occupational therapists place on the client's goals

when deciding on upper limb orthosis prescription. This view, as presented in Figure 4.5, has been interpreted as an overarching perspective for the three identified viewpoints.

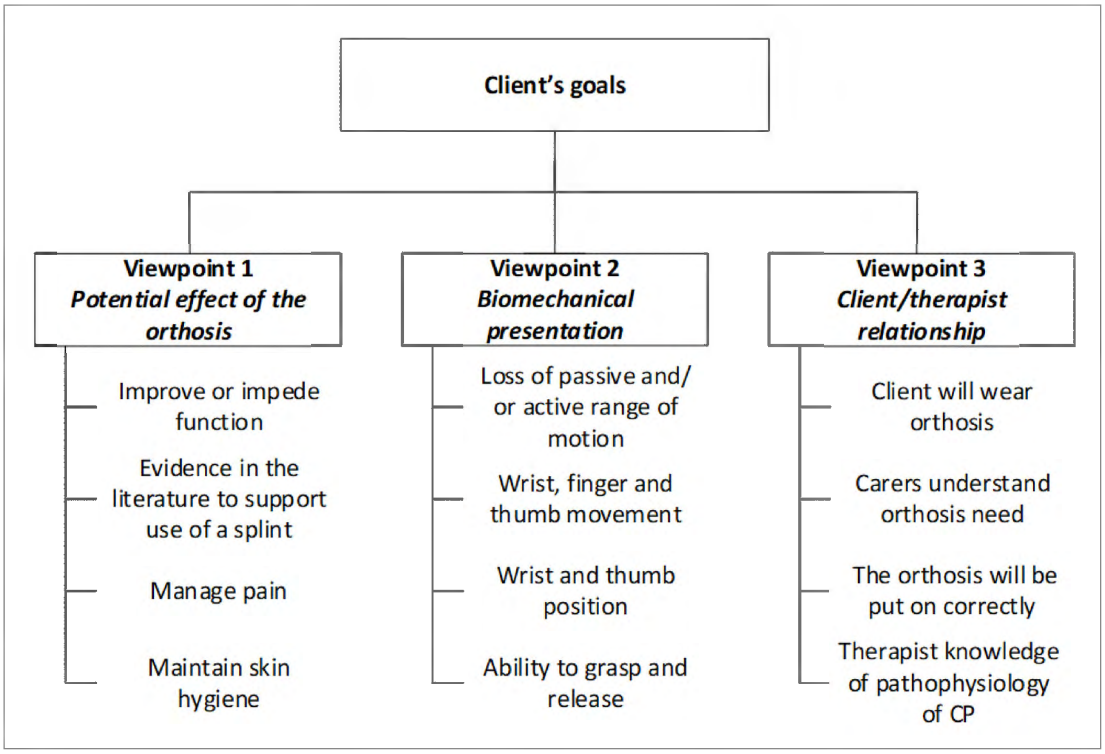

Figure 4.5 Occupational Therapist's viewpoints that guide orthosis prescription decision making

4.5 Discussion

Knowledge about how occupational therapists balance ideas that influence decision making in prescribing upper limb orthoses for children with CP provides a better understanding of decision making within this complex area of clinical practice. The process of by-person factor analysis within Q methodology reduced many statements that guide orthosis prescription decision making for children with CP to three viewpoints that may inform occupational therapy practice. Occupational therapists with varying levels of clinical experience, years of treating children with CP, education and self-determined experience with orthotic intervention were represented in each of the three viewpoints. This highlights the varied approaches to decision making regarding orthosis prescription and the complexity of the clinical area.

The first viewpoint, *potential effect of the orthosis*, revealed an eclectic clustering of statements that was not as obvious to interpret and name as the second and third viewpoints. The viewpoint was distinguished with statements that had an occupation focus, supported by evidence within the literature, regarding the outcome of the orthotic intervention itself. Decision making for this viewpoint was guided the most by upper limb orthotic intervention having the

potential to improve or impede function. This viewpoint did not appear to take a reductionist view of impairment, rather it considered promoting clients' engagement in occupation and meeting their occupational needs, utilising upper limb orthoses as the primary intervention strategy (Fisher & Bray Jones, 2017). This viewpoint is consistent with the conceptual framework of the Canadian Model of Occupational Performance where the focus is on the dynamic interaction between the person, environment and occupation that results in occupational performance (Boniface et al., 2008; Boniface & Seymour, 2013; Law & Laver-Fawcett, 2013).

The second viewpoint, *biomechanical presentation*, revealed a clustering of statements distinguished by range of motion, wrist and thumb joint positions, wrist, finger and thumb movement in relation to each other during a task, ability to grasp and release and current level of hand function. These statements align with biomechanical principles considered when making decisions about orthotic intervention. Biomechanical principles include assessment of joint structure and alignment, changes in muscle and soft tissue length, and muscle imbalance (Copley & Kuipers, 1999, 2014b). Biomechanical principles also use the principles of physiology and physics to explain successful application of orthoses to remediate changes in joint, muscle and soft tissue (Wilton, 2013a).

The third viewpoint, *client-therapist relationship*, revealed a clustering of statements that had a theoretical underpinning of client centred care. Client centred care has a focus on the dynamic relationship of the client and therapist (Parker, 2013). The therapist engages with the client to provide them with clinical information, including information based on the therapist's knowledge of the pathological processes and musculoskeletal changes that occur within cerebral palsy, to allow the client to make informed decisions about their needs. This collaboration establishes an understanding of the intervention and promotes participation in therapy (Parker, 2013; Sumsion & Law, 2006). This viewpoint demonstrates the importance of the client, carer and therapist working together to facilitate successful upper limb orthotic intervention.

Consensus statements, identified by consistent ranking of statements across all viewpoints, highlighted that environmental variables of access to funding, proficiency in orthosis fabrication and health professional requests for an orthosis, guided occupational therapists the least when making a decision regarding orthosis prescription. Caution in interpreting this is required to not diminish the importance of the individual statements. In the process of Q sorting, a forced sort on the quasi-normally distributed grid, statements had to be placed at the 'guides the least' value. This does not mean they do not guide decision making, only that they are less influential than the other statements.

The viewpoints identified in this study can be aligned with the recommendations for continued clinical use of upper limb orthotic intervention as presented by Garbellini et al. (2017). The *biomechanical presentation* viewpoint aligns with the first recommendation from the systematic review of describing the dynamic interplay of wrist and finger musculature as part of a comprehensive upper limb assessment. The therapist's decision making regarding orthosis prescription is guided by the structures of the upper limb that are potentially limiting movement. The *potential effect of the orthosis* and *client-therapist relationship* viewpoints align with the second recommendation of the systematic review to provide clear documentation of the use of the orthosis as an intervention including the aim or intended effect. Documenting the use and aim of the orthosis clarifies what the therapist is trying to remediate with the intervention, for example enhancing engagement in occupation during specific activity while wearing the orthosis. Having a clearly documented aim of orthosis use also allows transparency in decision making, collaboration and communication with the client, allowing them to make an informed decision regarding participation in the intervention. The *biomechanical presentation* viewpoint aligns with the third recommendation of the systematic review that orthoses should be described using consistent terminology. Utilising biomechanical principles, the therapist can describe the orthosis based on the anatomical focus and the purpose of the orthosis (immobilisation, mobilisation, restriction or torque transmission) on the upper limb (Australian Hand Therapy Association, 2012; Colditz, 1996; Fess et al., 2004; Garbellini et al., 2017; American Society of Hand Therapists, 1992). These viewpoints could be refined to inform the development of clinical guidelines.

The challenge in prescribing upper limb orthoses for children with CP may be in balancing the three viewpoints to guide decision making: 1. *The potential effect of the orthosis; 2. Biomechanical presentation; and 3. Client-therapist relationship*. Further exploration of the relationship between actual practice and these viewpoints is needed. As identified in this study, the elements of orthosis prescription decision making not only encompass individual occupational goals, evidence, relevance and practicality within the individual's life situation (Lannin et al., 2017), but also includes the biomechanical principles considered by occupational therapists (Copley & Kuipers, 1999, 2014b; Wilton, 2013a). Future clinical research is required to:

1. Identify valid and reliable classification and assessment tools to guide decision making regarding orthosis prescription (encompassing the viewpoints from this study).
2. Establish the effect of orthotic intervention through connecting the aim of orthotic intervention and the purpose of the orthosis utilising valid and reliable outcome measures.

A strength of this study was the robust process followed in the selection of Q sample statements, structured on the ICF-CY (World Health Organisation, 2007), refined by a focus group and piloted prior to commencement of Q sorting. The judgement of the researchers in interpreting and ascribing meaning to the viewpoints was guided by the data, but remains a complex process and may be seen as a limitation.

4.6 Conclusion

Upper limb orthotic prescription for children with CP is complex and the reasons to use orthoses may be influenced by many factors. Through the application of Q methodology, this study identified 'client goals' as a primary value that precedes three viewpoints for guiding decision making. The viewpoints were: *1. Potential effect of the orthosis; 2. Biomechanical presentation; and 3. Client-Therapist relationship*. The main elements contributing to each of these viewpoints were identified and discussed. These viewpoints may be used to provide structure in the identification of valid and reliable classification and assessment tools that may contribute to the development of domain specific guidelines when determining upper limb orthosis prescription for children with CP. The challenge for future clinical research is to connect the potential effect of the orthosis for a particular client, as decided by the therapist, with the purpose of the orthosis itself, informed by biomechanical presentation of the client, to establish the effect of the intervention.

4.7 Acknowledgements

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Declaration of interest

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

END OF PUBLICATION 2

PUBLICATION 3 – UNPACKING THE APPLICATION OF Q METHODOLOGY FOR USE IN OCCUPATIONAL THERAPY RESEARCH

Publication details

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[3] See Appendix B, Section B.3 for the published edition of this paper

Abstract

Background: Occupational therapy research has not fully utilised available research methods when exploring occupational therapists' views on specific interventions and service provision nor when exploring consumer priorities and the impact of occupational therapy services. Q methodology, a quantitative method for the systematic assessment of qualitative data, is an approach that can be used to examine viewpoints related to occupational therapy practice.

Purpose: This paper adds experiential knowledge to guide researchers new to navigating Q methodology and encourages occupational therapy researchers to consider the application of Q methodology when exploring viewpoints pertinent to practice and research.

Key issues: This paper provides more detailed reflection on each stage of Q methodology than is currently available in the literature, with a focus on the factor analysis stage, to support successful implementation of this method.

Implications: Sharing experience in implementing Q methodology may inform and encourage researchers in its use as one approach to combine qualitative methods and quantitative data analysis techniques. The rigor of the method's processes may add credibility to identified viewpoints and how they could inform occupational therapy practice.

4.8 Introduction

Health-based research is characterised by a broad range of qualitative methodologies. Occupational therapy researchers, however, have not fully utilised some of these methods to facilitate a better understanding of the consumer perspective regarding priorities for occupational therapy programs and the impact those programs have on the daily lives of the consumer (Borell et al., 2012). To assist with evaluating the effect of programs and implementing research findings into practice, Rapport et al. (2018) encourage the researcher to include consumer perspectives to inform health care research and service provision. Q methodology was identified by Corr (2001) as a potential approach to explore and determine subjective viewpoints and attitudes towards occupational therapy interventions, service provision, education and the occupational therapy profession. Despite Corr's encouragement, Q methodology is not commonly used in occupational therapy research.

Q methodology incorporates quantitative techniques for the systematic assessment of qualitative data (Brown, 1993; Dziopa & Ahern, 2011). Participants are asked to rank stimuli, usually statements, on a quasi-normally distributed grid, that provides one place for each statement. The statements are drawn from a larger body of information, representing the range of views on the topic, called the concourse (Kenward, 2019; Van Exel & De Graaf, 2005). The statements, defined as the Q sample, are ranked on a continuum, for example agree/disagree, most important/least important, according to the participant's perspective on a given topic (Chee et al., 2015; Corr, 2006; Dziopa & Ahern, 2011; Watts & Stenner, 2012). How the statements are placed on the grid are analysed with by-person factor analysis to identify the number of different ways the statements have been organised, which are then interpreted and labeled as viewpoints (Brown, 2008). This relatively straightforward description of Q methodology belies the rigorous, and somewhat complex, process of the method itself.

There is a misconception that Q methodology involves quantitative assessment (Dziopa & Ahern, 2011). However, Watts and Stenner (2005) describe it as a highly unusual qualitative research method featuring quantititive methods. The sorting of statements by the participant, from their point of view, is a dynamic way in which subjectivity can be expressed (Brown, 1993; Dziopa & Ahern, 2011). Q methodology employs by-person factor analysis to identify themes, determined by comparing how participants rank a set of statements (Dziopa & Ahern, 2011; Watts & Stenner, 2005). This interpretive approach to participant-driven subjectivity aligns with a qualitative paradigm of inquiry.

Q methodology has been described as a set of connected techniques within two key phases of data collection and data analysis (Baker, 2016). The process to collect and analyse data is comprised of the following stages: i) concourse development; ii) development of the Q sample; iii) selection of the participant set (P set); iv) Q sorting; v) factor analysis; and vi) interpretation of factors (which comprise the viewpoints) (Chee et al., 2015; Corr, 2006; Dziopa & Ahern, 2011; Watts & Stenner, 2012). The stages of the Q methodology process are presented in Figure **4.6**. Application of the stages provides the basis for systematic examination of the subjective viewpoints of study participants (Brown, 2008).

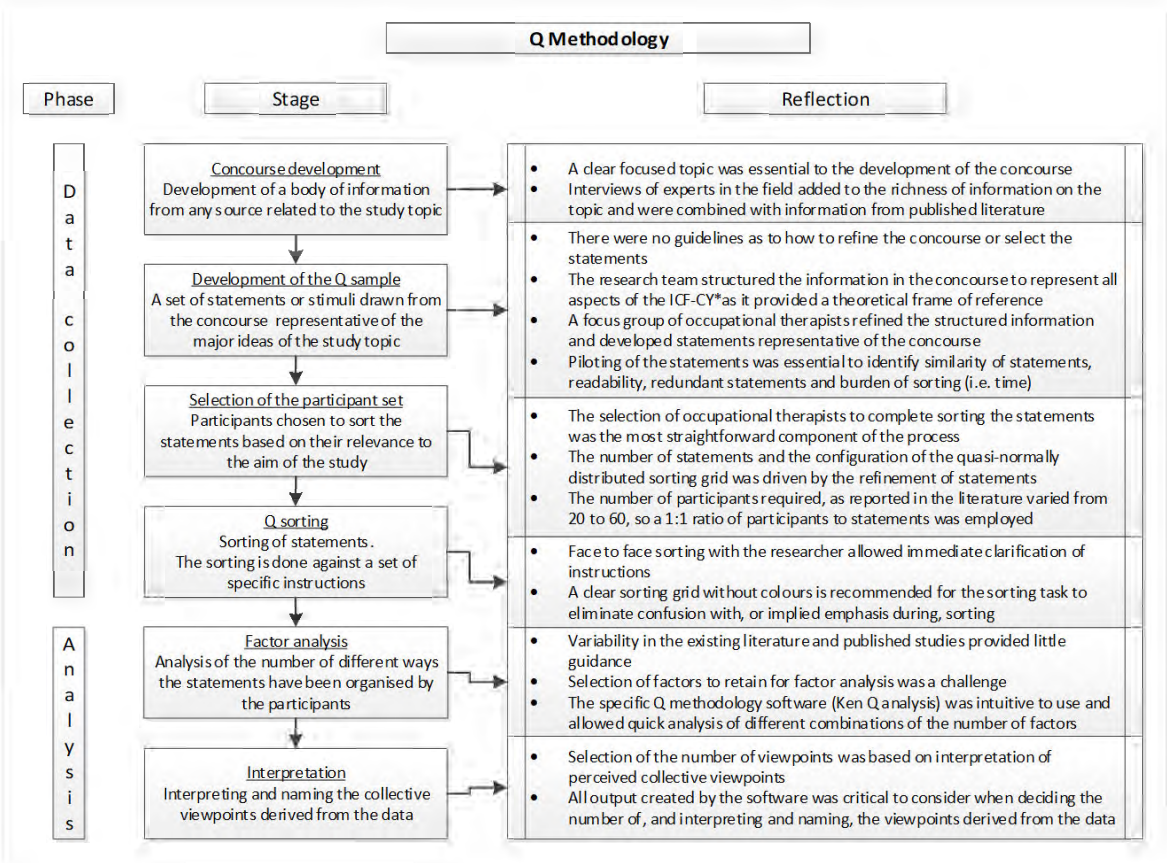

Figure 4.6 **The Q method phases and stages, and reflection on the knowledge needed to implement the method**

For more information and resources regarding Q methodology see www.qmethod.org. For a more detailed description on the stages of Q methodology, as presented in Figure **4.6**, readers are referred to Brown (1993); Corr (2001); Dziopa and Ahern (2011); Watts and Stenner (2012).

The qualitative approach of Q methodology aligns with the holistic nature of how people perceive themselves and view their daily activities in the world in which they live (Robertson, 1988). Q methodology provides a flexible approach combining qualitative methods of data collection and interpretation, with quantitative by-person factor analysis, to gather viewpoints

on the dynamic relationship between the person, environment and occupation (Boniface & Seymour, 2013; Robertson, 1988). Using Q methodology, the occupational therapy researcher can: (i) convey what it is like to have a condition; (ii) reflect on the features of a service or system; (iii) convey the perspective of a patient/client experience; (iv) convey the perspective on a hospital or community experience; and (v) summarise viewpoints from different groups, for example, occupational therapists (Robertson, 1988).

Q methodology has been applied in published occupational therapy and occupational science research to identify:

- Patients' perception of curative factors in occupational therapy groups (Webster & Schwartzberg, 1993);
- The comparison of an occupational therapy definition and consumers' experiences (Corr et al., 2005);
- What is considered important for life balance (Wagman et al., 2012);
- Viewpoints on community mobility and participation in older age (Fristedt et al., 2012);
- Food activities and maintenance of identity in later life (Plastow, 2014);
- Viewpoints on driving of individuals with and without autism spectrum disorder (Chee et al., 2015);
- Parents' viewpoints regarding the participation of their child with an acquired brain injury (Thompson et al., 2016);
- Identifying outcomes related to occupational therapy student community partnerships (Kramlinger et al., 2016);
- The role of Filipino occupational therapists in substance addiction and rehabilitation (Sy et al., 2018); and
- Prescribing upper limb orthoses for children with cerebral palsy: a Q methodology study of occupational therapists' decision making. (Garbellini et al., 2019).

These ten studies, identified from a comprehensive search, provide diverse examples of the application of Q methodology in occupational therapy research. Occupational therapists have also been active participants in published studies using Q methodology (Clarke & Holt, 2015) and probably in Q methodology studies either not submitted for publication, or not published. Dziopa and Ahern (2011), in a systematic review on the application of Q methodology, reflected that the topics Q methodology can examine are limited only by the researcher's imagination. So the question remains as to why Q methodology has not been utilised more in occupational therapy research.

In a paper on qualitative research in occupational therapy, Frank and Polkinghorne (2010) stated that new researchers acquire standards for qualitative research from published examples. The atypical nature of Q methodology and varying applications of the methodology within the literature has been described as a source of confusion (Dziopa & Ahern, 2011). For occupational therapy researchers seeking guidance from published examples, the stages of Q methodology are provided (Brown, 1993, 2008; Corr, 2001; Dziopa & Ahern, 2011), but detail about what is undertaken within each of the stages may appear contradictory across various publications. This lack of clarity is problematic for the researcher new to this method.

The purposes of this paper are to add experiential information to guide researchers new to Q methodology and to encourage occupational therapy researchers to consider the application of Q methodology to explore consumer, clinician and other stakeholder viewpoints that may inform service provision and further research. The study by Garbellini et al., (2019) provided the experiences that informed this discussion of Q methodology that aims to assist future researchers and people reading Q methodology papers with interpreting the evidence provided. A lack of clarity and variability in approach to choosing factors to retain and interpret within the data analysis phase was identified as a gap in the literature. This gap guided the critical reflection reported in this paper that describes our experience and provides direction for researchers new to Q methodology.

4.9 Critical reflection

Experience gained using Q methodology to explore occupational therapists decision making when prescribing upper limb orthoses for children with CP (Garbellini et al., 2019), formed the basis of this critical reflection. Reflections on each stage of the process of Q methodology are detailed in Figure **4.6**. Although the research team encountered challenges throughout the stages of the Q methodology process, most challenges were experienced during the data analysis phase, particularly within the stage of factor analysis. Watts and Stenner (2012) suggest that the interpretation of factors has less to do with aims of the investigators and more to do with the chosen number of factors to retain, method of rotation and the factors investigators decide to interpret. Understanding the steps to choose factors to retain and rotate, within the factor analysis stage, was critical to determining the number of collective viewpoints (factors) and naming them in the interpretation stage. Therefore, this paper provides information aimed at detailing these steps of the method. Figure 4.7 provides a summary of data management steps undertaken within the factor analysis stage.

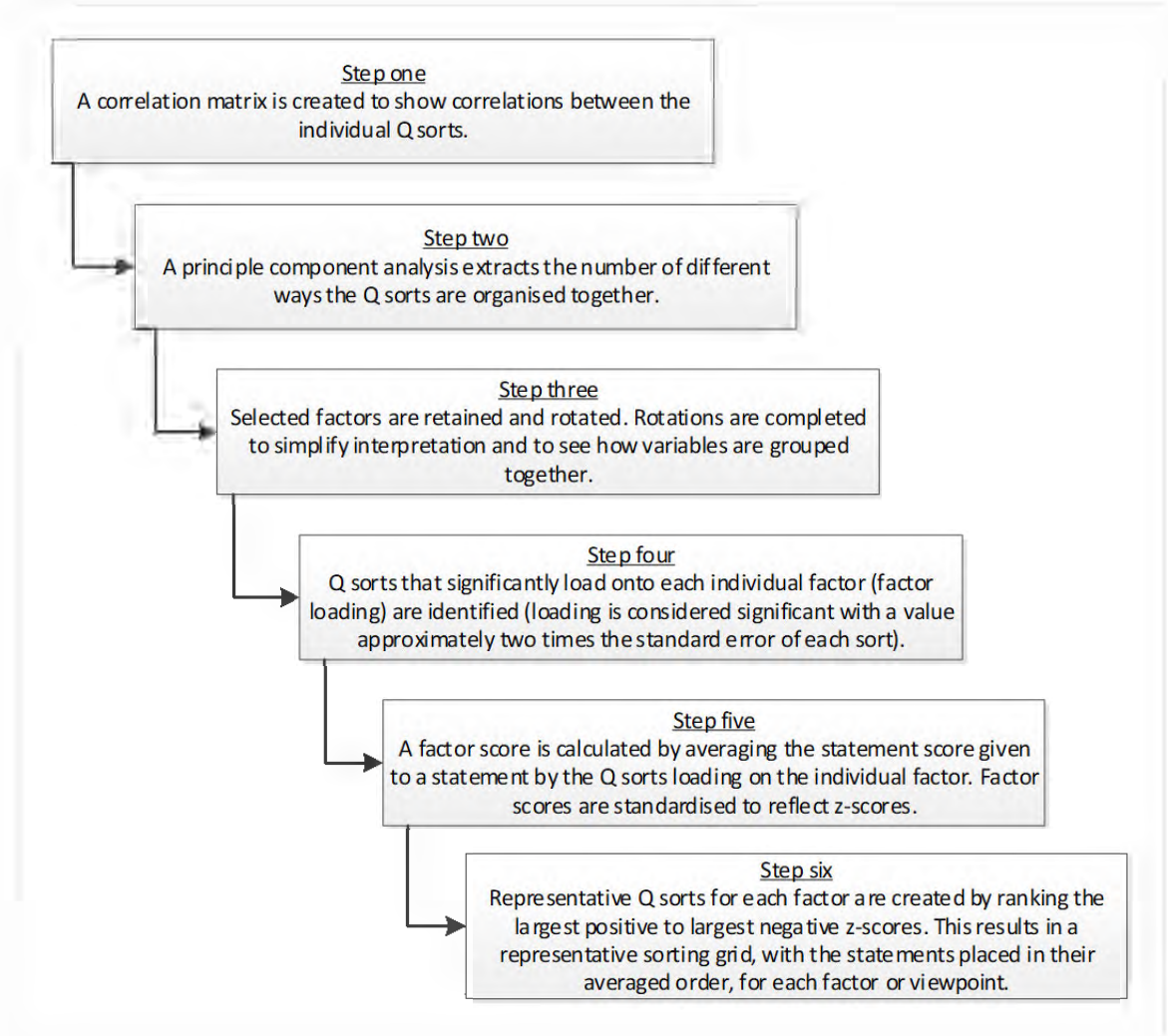

Figure 4.7 Q method stage of factor analysis

We experienced greatest challenge in selecting factors to retain and rotate (refer to Step three in Figure 4.7). Direction was taken from the studies by Chee et al. (2015) and Thompson et al. (2016) who applied the following five criteria to select factors:

1. Use of the default number for extraction within the software (PQ Method software (Schmolck, 2014))
2. Inclusion of factors with an eigenvalue of greater than one
3. At least two significant factor loadings required per retained factor
4. Multiplication of the highest two significantly loading factors should be greater than twice the standard error (Humphrey's rule)
5. All factors displayed prior to the scree plot levelling or plateauing should be retained

Application of these criteria to the data analysed within our study (Garbellini et al., 2019) is presented in Table 4.3. It was identified that strictly following the methods of these previous

studies would have resulted in only one factor, meeting all five criteria (see Factor 1 in Table 4.3), being retained factor for rotation. This decision would have limited the interpretation stage to the identification of one broad, and difficult to interpret, viewpoint versus the three viewpoints ultimately identified. Our experience suggests that the criteria should be applied to guide, not limit, selection of factors to retain for factor rotation. When these five criteria were used as a guide and applied without strict adherence, it allowed a more nuanced insight into the number of factors to retain for rotation. Our approach aimed to find the balance between identifying an amorphous single viewpoint and multiple individual perspectives.

Table 4.3 Application of selection criteria to retain factors

Selection criteria	Factor							
	1	2	3	4	5	6	7	8
1. Default number in software	√	√	√	√	√	√	√	√
2. Eigenvalue[a] > 1.0	√	√	√	√	√	√	√	√
3. Two significant factor loadings	√	√	√	X	X	X	X	X
4. Humphrey's rule	√	X	X	X	X	X	X	X
5. Scree plot test	√	?	?	?	X	X	X	X

Key: ✓ – Yes; X – No; ? – Dependent on where the levelling of the scree plot is determined.
[a]Eigenvalue is the sum of squared factor loadings for each factor (Brown, 1980).

The output generated by the Ken Q analysis software following factor rotation allowed relatively fast visual inspection of multiple combinations of retained factors. This confirmed the selection of factors to retain related to criteria one, two and five. A comparison of each different combination of retained factors with the number of Q sorts (participant's ranking of selected statements; refer to Figure **4.6**, Stage 4) that significantly loaded onto each factor (refer to step four in Figure 4.7) guided the selection of the final factors for interpretation. As the number of factors retained increased, it was gradually clearer as to when the number of Q sorts loading onto each factor were less related to a collective viewpoint and more to an individual perspective.

Application of criteria three and four, as in the studies by Chee et al. (2015) and Thompson et al. (2016) guided selection of final factors in our study, ensuring two significant factor loadings per retained factor and that the multiplication of these was greater than the standard error (refer to Step four in Figure 4.7 and Table 4.3). This approach, of iteratively comparing a combination of retained factors with consideration of the criteria applied by Chee et al. (2015) and (Thompson et al., 2016), provided a more nuanced insight in determining the final number of factors.

The interpretation and naming of viewpoints (refer to Figure **4.6**, Stage 6) by Garbellini et al. (2019) was informed by review of three key outputs from the Ken Q analysis software: (i) the representative Q sorts (the average Q sort arrangement for each viewpoint; refer to Step six of Figure 4.7); (ii) the distinguishing statements; and (iii) the consensus statements. Comparison of the position of statements on the grid of each representative Q sort, how the placement of statements distinguished the sorts from one another or demonstrated consensus, provided the framework for naming of each viewpoint. The challenge of this part of the process was determining a name for each viewpoint that was indicative of the qualitative category of thought it represented within the context of the issue under investigation (Brown, 1993).

The final outcome of the Q methodology study was a product of qualitatively generated statements, qualitatively sorted by participants, quantitatively analysed with by-person factor analysis, qualitatively interpreted and named. This incorporation of quantitative techniques within qualitative methodology is encouraged as one method to ensure that viewpoints of the consumer, and/or stakeholder, and/or community perspective is included in future occupational therapy research.

4.10 Summary

Q methodology is a rigorous research process, employing both qualitative and quantitative methods, that may be utilised in occupational therapy research to explore consumer and therapist viewpoints regarding interventions, service provision and the profession itself. This paper highlights issues to consider for future utilisation of this method. An understanding of each stage, in particular the factor analysis stage of the process, allows successful implementation of the method. Sharing of practical experience utilising Q methodology may inform and encourage occupational therapy researchers in its use and aid clinicians reading studies that use Q methodology in their interpretation of the evidence. Having a rigorous process to follow may add credibility to the viewpoints identified and how these viewpoints could inform occupational therapy practice and research.

Disclosure statement

The authors have stated that they had no interests that might be perceived as posing a conflict or bias. No funding support was received.

Key messages

- Q methodology can be used in occupational therapy research to explore consumer and therapist viewpoints regarding interventions, service provision, priorities and the profession itself.
- Q methodology employs a specific, repeatable process within each stage of the research process to ensure rigor.
- Q methodology provides an approach to combining qualitative research methods with quantitative analysis techniques to understand viewpoints of interest.

END OF PUBLICATION 3

4.11 Chapter Conclusion

Conclusions

Findings from the Q methodology study highlighted the complexity of upper limb orthosis prescription for children with CP, and clinicians' varied approaches to decision making. When considered together, the three viewpoints identified from this qualitative study could be used to inform upper limb orthosis prescription as aligned with the ICF framework (World Health Organisation, 2001).

The first viewpoint, the potential effect of the orthosis, aligns with activity and participation domains. The decision to prescribe an upper limb orthosis may be guided by whether an orthosis will meet the client's occupational needs and promote their engagement in occupation.

The second viewpoint, the biomechanical presentation of the client, aligns with the body structures and function domains. Assessment of changes in muscle length, joint structure and alignment and movement resulting from muscle imbalance of the wrist and hand (hand deformity) may guide decision making for orthosis prescription.

The third viewpoint, the client-therapist relationship, aligns with the personal and environmental contextual factors within the ICF framework (World Health Organisation, 2001). Orthosis prescription may be guided by the client's previous experience with the intervention, family/carer support of the intervention and the therapist's experience, expertise and the environmental constraints (i.e. time demand, available resources) within which they work.

Reflection on the systematic review by Garbellini et al. (2017) and the detailed discussion presented in Chapter 3 identified a lack of biomechanical considerations, as part of upper limb orthosis prescription, as a gap within the current literature. While biomechanical considerations

are not a feature of research studies, in practice at least some clinicians consider biomechanical information important when prescribing an upper limb orthosis. This Q methodology study identified that clinicians' decision making regarding upper limb orthosis prescription was not only guided by the potential effect of the orthosis and client-therapist relationship, but also by the client's biomechanical presentation of wrist and hand movement.

The systematic review by Garbellini et al. (2017) recommended further exploration and identification of valid and reliable tools to classify the dynamic interplay of wrist and finger muscle movement. To address the lack of movement analysis or use of available clinical resources identified in the literature, the introduction of the Neurological Hand Deformity Classification (NHDC) to classify hand deformity from the dynamic interplay of the client's wrist and finger movement (Chapter 5) and exploration of the psychometric properties of validity and reliability of the NHDC (Chapter 6) is the focus of the following chapters of this doctoral program.

Chapter Five

Background of The Neurological Hand Deformity Classification

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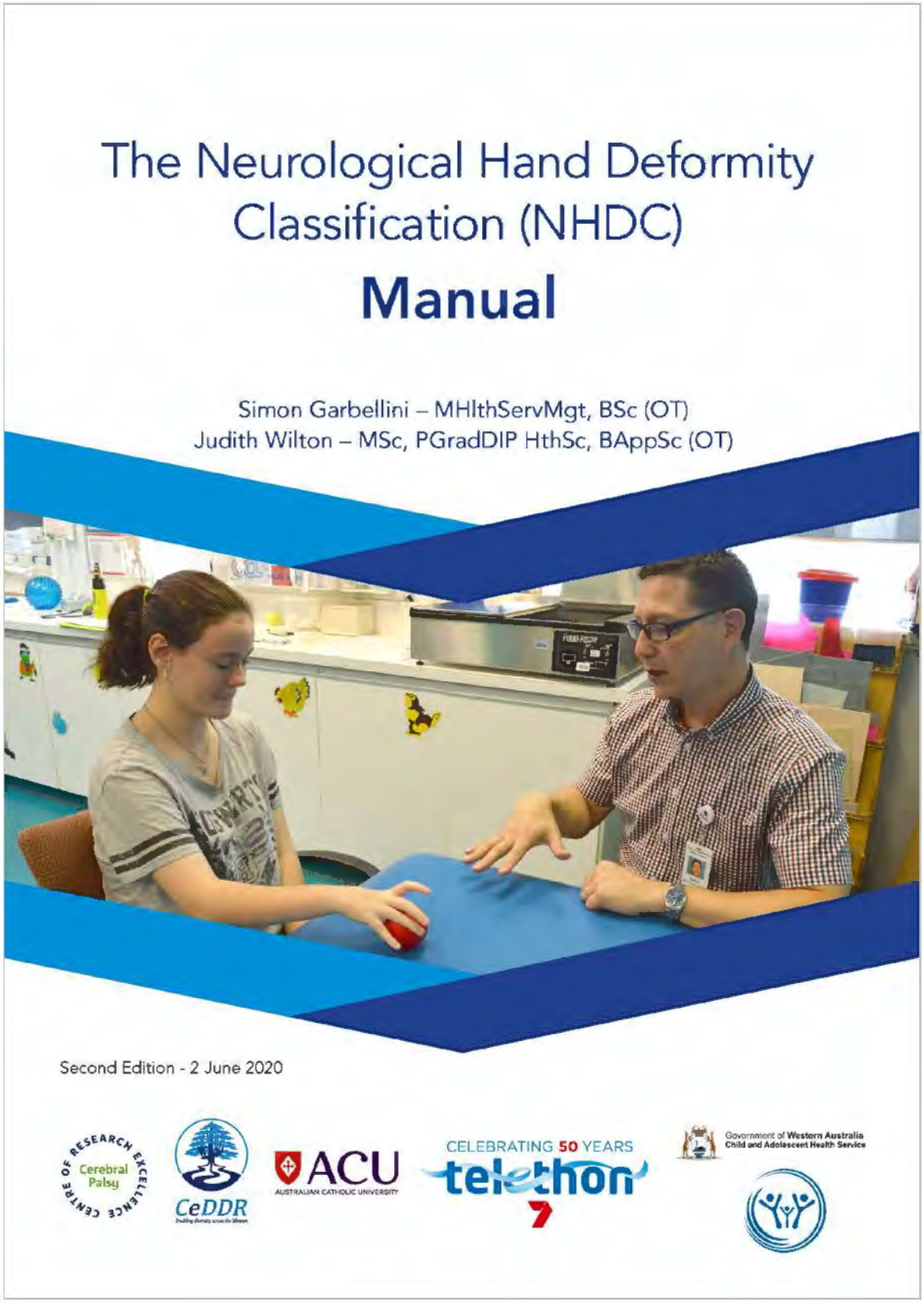
5.1 Introduction

Hand deformities can occur as a result of imbalance in wrist and finger muscle groups caused by positive and negative neurologically-based features (Chin et al., 2005; Copley & Kuipers, 2014b). Consideration must be given to how musculoskeletal and neural factors interact with each other and cause the dynamic presentation of hand deformity. Scales that discriminate between the characteristics influencing hand position or movement (Copley & Kuipers, 2014a) and consistent patterns of hand deformity observed in clinical practice have been developed (Wallen & Stewart, 2016).

The Neurological Hand Deformity Classification (NHDC) is one such tool developed to facilitate analysis of the biomechanical components of hand deformity. Information from the use of the NHDC may guide the selection of interventions to prevent or remediate impairment. Despite its availability, the NHDC has not been applied in research studies. This might be due to its limited psychometric properties or the need to have more detailed information on its application. Initial reliability of the NHDC has been explored (Georgiades et al., 2014). No evidence of construct validity or test-retest reliability was established. Following this initial reliability study, a lack of procedural guidelines in the application of the NHDC was identified by the author of this doctoral program. To ensure consistency in observation and recording of hand deformity using the NHDC a standardised procedure was developed and completed as part of this doctoral work. Background information on the NHDC, its aims and standard procedure, presented as a manual and website (Garbellini & Wilton, 2017) are provided in this chapter.

5.2 Neurological Hand Deformity Classification Manual



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1. The Neurological Hand Deformity Classification

1.1 Introduction

Active movement of the wrist and hand involves a complex interaction of intrinsic and extrinsic musculature. For people with neurologically-based impairment, an imbalance of muscle activity drives the pattern of wrist and hand deformity (Wilton, 2013). Even though impairment is unique to each individual, hand deformity tends to follow characteristic patterns (Wilton, 2003, 2013).

The primary purpose of any classification tool in health care is to discriminate between variations in the presentation of a health condition at a particular point in time (Kirshner & Guyatt, 1985; McConnell, Johnston, & Kerr, 2011; Rosenbaum, Eliasson, Hidecker, & Palisano, 2014; Rosenbaum et al., 1990). The Neurological Hand Deformity Classification (NHDC) is framed within the body functions and structure domain of the International Classification of Functioning Disability and Health (ICF) (World Health Organization, 2001). Thus, the NHDC is an impairment-based classification designed to categorise hand deformity. The NHDC was developed to classify deviation or loss of wrist and hand movement attributed to changes in body structure of the affected upper limb(s). It is designed for use with clients of any age across all neurologically-based upper limb impairments. The application of the NHDC is not limited to a specific diagnosis, but has been predominantly used by the authors in clinical practice with children and adults with cerebral palsy.

Note that the NHDC is not an assessment of the details of hand function or even of impairment. Classification is made according to observed wrist and hand movement. The NHDC consists of two extension categories (E1 and E2) and five flexion categories (F1 to F5). Differentiation between the categories is determined by wrist position and wrist and finger movement.

1.2 Purposes

The purposes of classifying hand deformity using the NHDC are:

- To facilitate observation and analysis of the anatomical and biomechanical components of neurologically-based wrist and hand deformity.
- To identify the primary factors causing the dynamic presentation of the deformity during active wrist and hand movement.
- To provide a framework for therapists to consider intervention options based upon the dynamic pattern of movement observed.

The NHDC is intended to categorise wrist and hand movement in action; assist identification of structures driving hand deformity; guide clinical assessment; and enhance clinical decision-making regarding the use of upper limb orthoses as part of the overall management of upper limb impairment.

1.3 Background

Classifications of patterns of wrist, finger and thumb deformities were originally developed by surgeons to select the most appropriate surgical intervention for individual patients (House, Gwathmey, & Fidler, 1981; Tonkin, Freitas, Koman, Leclercq, & Van Heest, 2008; Tonkin, Hatrick, Eckersley, & Couzens, 2001; Zancolli, 2003; Zancolli & Boyes, 1979; Zancolli, Goldner, & Swanson, 1983; Zancolli & Zancolli, 1981, 1984, 1987). Zancolli (2003) classified a child's 'spastic hand', into one of three groups, based upon observation of degree of wrist flexion required to produce voluntary release of the fingers. Flexion contracture of the wrist and or fingers is corrected through surgical techniques based on the classification level observed. House et al. (1981) developed a thumb deformity classification, with different static and dynamic components of thumb deformity described in four levels. Surgeries to address thumb deformities in different levels of the House classification included release of contractures, augmentation of weak muscles and skeletal stabilisation (House et al.,1981). However, since the function of the thumb is related to the position and stability of the wrist, the thumb deformity may be worsened when the wrist is placed in a more functional position (House et al.,1981). Therefore correction of thumb deformity must take into account deformity in the proximal joints, particularly the wrist (Tonkin et al.,2008).

Establishment of the validity and reliability of the Zancolli and House classifications was not undertaken by the authors of the classifications. Klingels et al. (2010) established inter-rater (=0.95) and test-retest reliability (=1) of the Zancolli classification, and inter-rater (= 0.73) and test-retest reliability (= 0.74) of the House classification, as part of a clinical test battery to assess upper limb impairment in children with hemiplegia.

Limitations were identified in the use of these surgical classifications to facilitate appropriate therapy treatment planning (Georgiades et al., 2014). The Zancolli classification is limited to patterns of wrist flexion deformity and does not include extension deformities, nor does it encourage the observer to consider the contributions of extrinsic and intrinsic finger and thumb musculature when describing deformity patterns. The House classification describes thumb deformity without reference to wrist position, whereas in surgical practice the wrist position and its impact on thumb deformity is considered prior to surgical intervention (House et al., 1981; Tonkin et al., 2008). A thorough search of the literature failed to yield instructions or guidelines for use of these classifications.

The Neurological Hand Deformity Classification (NHDC) was developed by Judith Wilton, occupational therapist, in recognition of limitations in the use of surgical classifications. The NHDC differs from these other classifications by observing dynamic movement of the whole hand and thumb, including flexion and extension deformities with concurrent thumb deformity. Clinical observations of consistent patterns of wrist and hand deformity during function in clinical practice (Georgiades et al., 2014; Wilton, 2003, 2013), the surgical classifications of spastic hand deformities in the wrist and fingers (Zancolli, 2003; Zancolli & Boyes, 1979; Zancolli et al., 1983; Zancolli & Zancolli, 1981, 1984, 1987) and patterns of thumb deformity (House et al., 1981) were utilised in the development of the NHDC.

1.4 Psychometric properties

The usefulness of measurement tools in decision making and clinical research depends on whether clinicians can rely on the generated information as accurate and meaningful (Portney & Watkins, 2015). Both reliability (the extent to which a tool is consistent and free from error) and validity (assuring the tool measures what it is intended to measure) are essential considerations when selecting a tool to use (Portney & Watkins, 2015). An initial reliability study of the NHDC has been published (Georgiades et al., 2014), and demonstrated high levels of both inter-observer and intra-observer reliability (=0.87 and =0.91 respectively). Further testing of the psychometric properties of the NHDC is currently being undertaken to provide additional evidence about construct validity, test-retest and inter-rater reliability.

Figure 1: Live classification of a client's right hand deformity using the NHDC.

2. Administration of the Neurological Hand Deformity Classification

2.1 General instruction

Classification of hand deformity is determined by **observing the client's wrist and hand in action** during movement towards and attempted grasp and release of objects as described in Section 4. The classification is based upon the **analysis of active wrist and finger movement in relation to the extended or flexed position of the wrist.** The extended or flexed wrist position is determined from a neutral (0°) wrist position as seen in Figure 1.

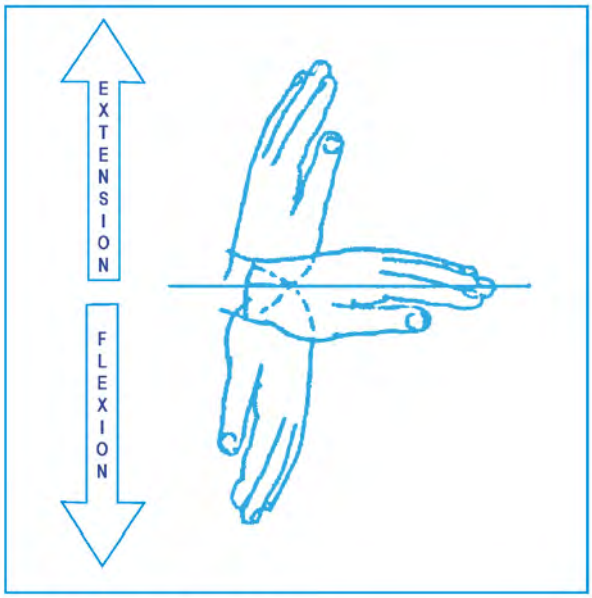


Figure 1: Guideline for wrist position

The pattern of wrist and hand movement is the focus of the classification regardless of success or the effectiveness of grasp and release. The procedure to classify hand deformity using the NHDC is the same in either a clinical or a research context.

2.2 Real time classification or video recording

Classification of hand deformity can be made either at the time of live clinical observation or completed at a later time from recorded footage. Regardless of the timing of classification, it is recommended that the client's wrist and hand movement be recorded. Footage of the individual client's wrist and hand movement allows comparison of the details of their movement over time.

2.3 Number of attempts

Observation of three attempts of wrist and hand movement in action may be required to make a classification. If there is variation in the different attempts of movement, classification is based upon the most consistent movement described during the three attempts.

2.4 Classification for young children

For young children observation of the wrist and hand movement in action will be elicited through play. If the young child is not cooperating, consider slowly demonstrating the action that you want the child to complete, engage the child in a game or ask the parent to demonstrate the action. The non-involved upper limb may need to be gently restrained to encourage movement of the limb for classification.

2.5 Clients with limited or no active movement

If the client is unable to approach, grasp or release the object on the table, the therapist must create a situation where attempted action of wrist and hand movement can be elicited and observed. This may be achieved by asking a parent or carer to present the object to the child. Observe any attempts of active movement noting the position of the wrist to determine classification level. If there is no active wrist or finger movement, the client will be classified as an E2 or F5 depending on whether the wrist is in an extended or flexed position.

3. Set up

3.1 Position of client

Figure 2: Position of client sitting at table for classification of right side

Figure 3: Position of client in supported seating system for classification of left side

The client is seated in a chair at a table (see Figure 2). The size of the chair must be such as to allow the client to have their feet on the floor and their bottom at the back of the chair. The table top should be approximately at waist height. If the client is not able to sit independently without support they can remain in a supported seating system (see Figure 3); sit with support from a therapist or carer; or where appropriate sit on the lap of their parent or carer.

3.2 Objects to elicit wrist and hand movement

It is essential that objects of an appropriate shape and size are used to elicit movement of the wrist, fingers and thumb. The object should be spherical in shape and approximately the size of the client's fist. Examples of objects (an apple and a ball) used to elicit wrist and hand movement are pictured in Figure 4 below.

Figure 4: The use of spherical objects (apple and ball) to elicit wrist and hand movement for classification using the NHDC.

3.3 Position of object

The object is placed at a forearm's distance from the edge of the table in the midline in front of the client. A "forearm's distance" is the distance from the client's elbow to wrist crease. The position of the object is important to avoid excessive reach. If the object is too far away, movement of the shoulder, elbow and forearm can compensate for a lack of wrist and finger motion.

If the client is in a supported seating system with a tray, the object can be placed on the tray (see Figure 3). If the client is sitting with support from a therapist or carer the object should be placed on a stable surface in front of them.

3.4 Position of camera

If the client's wrist and hand movement is to be videorecorded, the camera is placed one metre from the non-classified side of the client. The camera height should be level with table surface height. The camera zoom is used to focus on the wrist and hand. The wrist and thumb must be visible to capture the wrist and hand movement in action (see Figure 5).

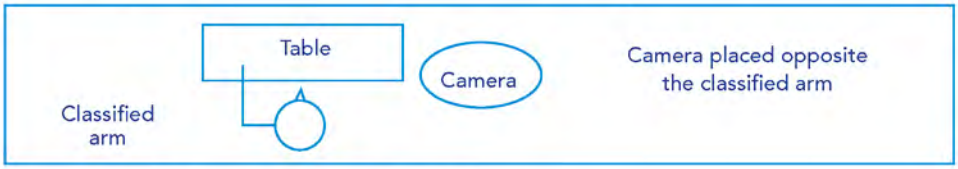


Figure 5: Position of camera (classifying left hand deformity)

3.5 Instructions

Once the client and camera are correctly positioned, place the object in front of the client. Ask the client to start with their hand on the edge of the table/tray in front of them before asking them to pick up and put the object down. This action may be repeated three times so that a consistent classification of hand deformity can be made. It is appropriate to explain to the client that you are only observing how the client's wrist and hand muscles work together, not how successful they are in grasping and releasing the object.

3.6 Important points to remember

- It is the observation of wrist and hand movement in action that is important.
- Objects are used to elicit movement.
- Objects, as pictured in Figure 4, may be substituted with similar objects provided the objects used do elicit the movement required to classify hand deformity.
- An extended or flexed wrist position is taken from the neutral wrist position of 0°, as pictured in Figure 1.
- The success or type of grip observed is not relevant for classification.

4. Determining classification level

The following observations of the client's wrist and hand movement guide classification and help differentiate between levels:

4.1 Wrist movement

- Is active wrist movement present?
- Is passive wrist movement present?
  - Is movement into wrist extension achieved through force of the hand on the object?
  - Is movement into wrist extension driven by tight finger flexors when grasping/attempting to grasp the object?

4.2 Wrist position

- Does the wrist flex past neutral (0°)?
- What is the wrist position during finger movement?
- Is the maximal degree of wrist flexion greater than 20°?
- Does the wrist remain in a static position of flexion or extension?

4.3 Finger movement

- Is there active finger flexion and extension?

Once the movements have been observed the hand deformity classification can be determined and documented. Figure 6, 'Wrist and finger movement considerations of the NHDC' decision tree in Section 5 is designed to guide hand deformity classification. Figure 7, 'Distinction between classification levels' in Section 6 is designed to help observers understand how to differentiate between the categories of the classification.

If there is a variation of wrist position during observation of the client's movement, ***the maximum degree of wrist flexion observed at any stage of the movement determines the classification.***

5. Wrist and finger movement considerations of the NHDC

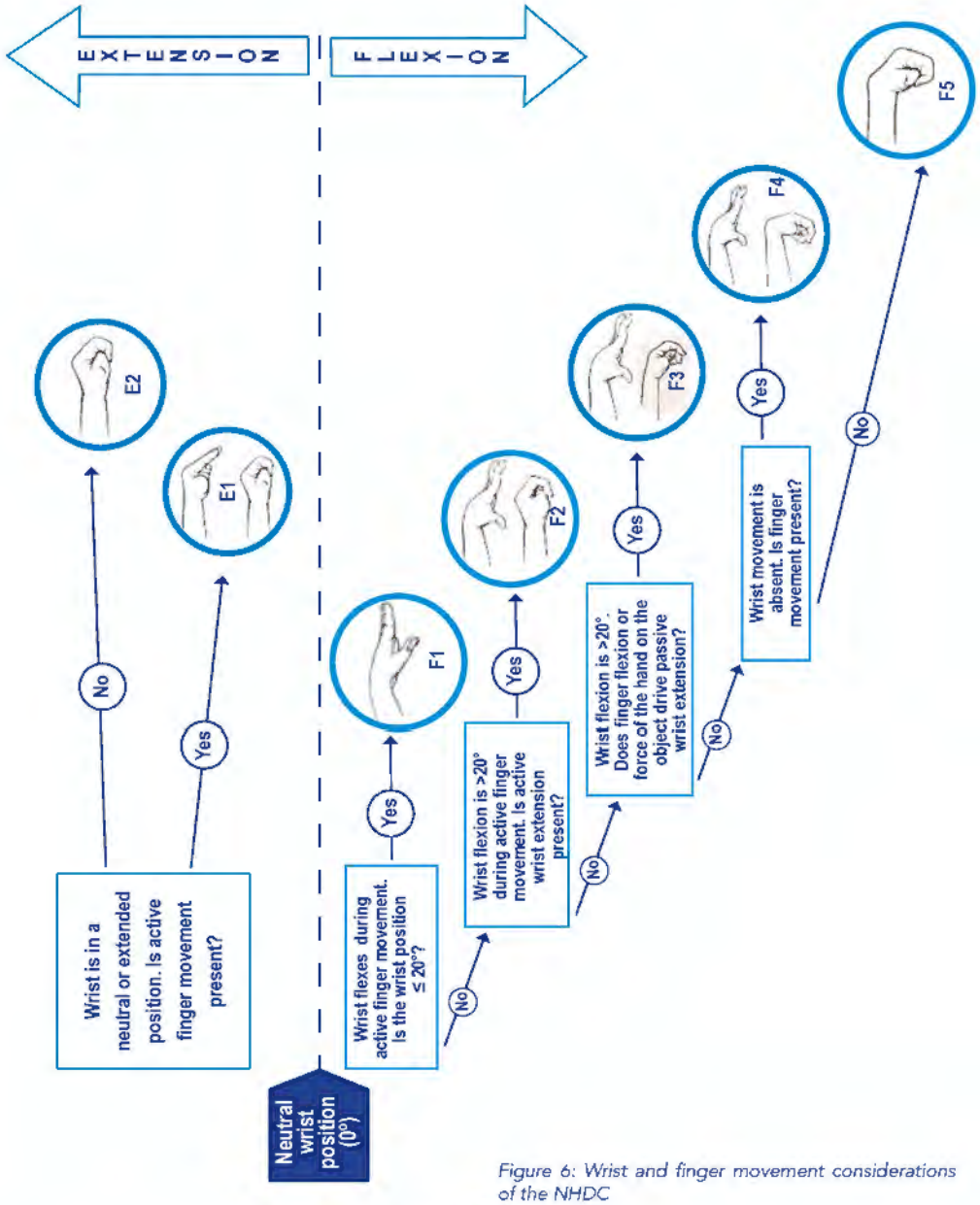
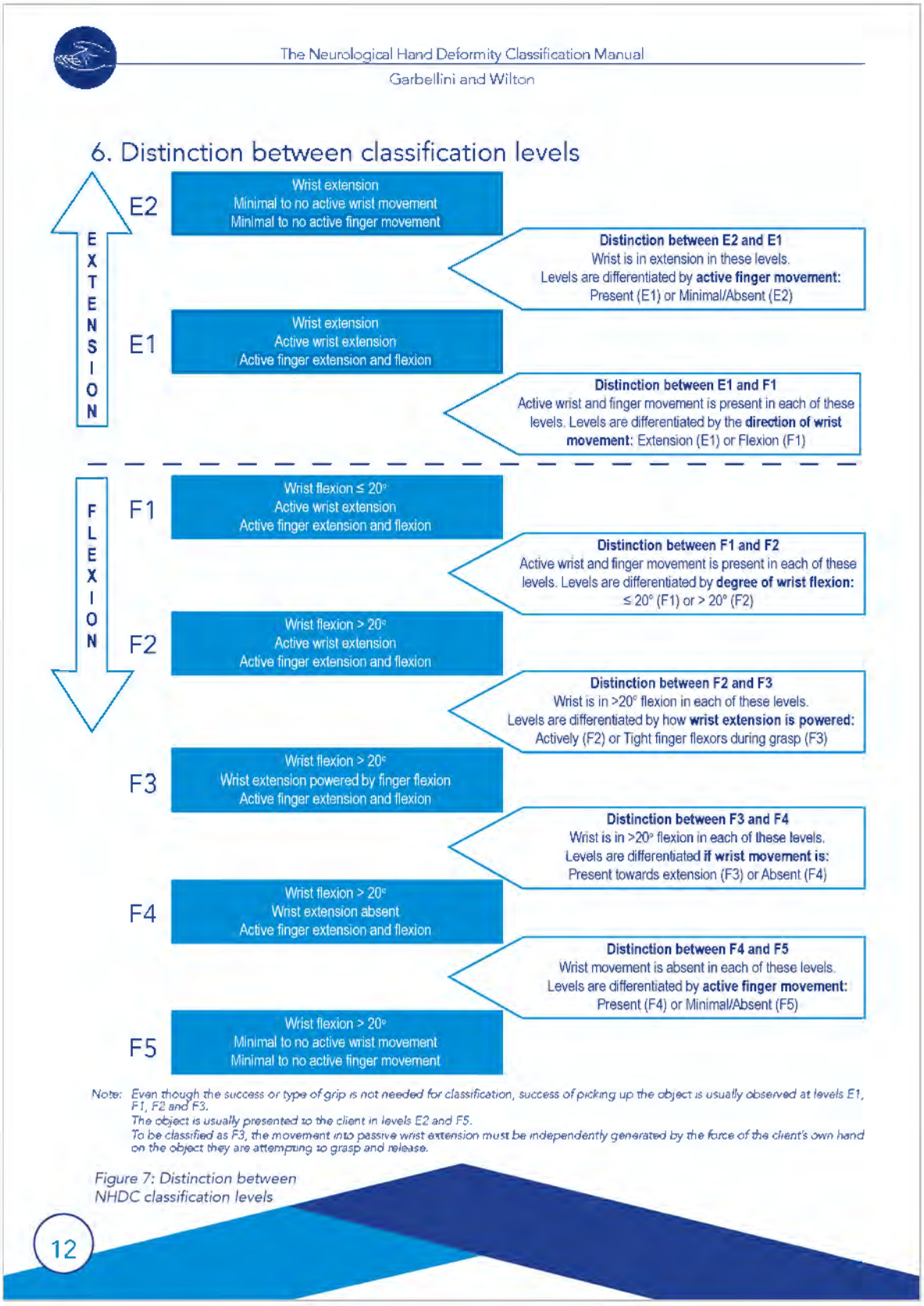


Figure 6: Wrist and finger movement considerations of the NHDC


The Neurological Hand Deformity Classification Manual
Garbellini and Wilton
6. Distinction between classification levels
EXTENSION
E2
Wrist extension
Minimal to no active wrist movement
Minimal to no active finger movement
Distinction between E2 and E1
Wrist is in extension in these levels.
Levels are differentiated by active finger movement:
Present (E1) or Minimal/Absent (E2)
E1
Wrist extension
Active wrist extension
Active finger extension and flexion
Distinction between E1 and F1
Active wrist and finger movement is present in each of these levels. Levels are differentiated by the direction of wrist movement: Extension (E1) or Flexion (F1)
FLEXION
F1
Wrist flexion ≤ 20°
Active wrist extension
Active finger extension and flexion
Distinction between F1 and F2
Active wrist and finger movement is present in each of these levels. Levels are differentiated by degree of wrist flexion:
≤ 20° (F1) or > 20° (F2)
F2
Wrist flexion > 20°
Active wrist extension
Active finger extension and flexion
Distinction between F2 and F3
Wrist is in >20° flexion in each of these levels.
Levels are differentiated by how wrist extension is powered:
Actively (F2) or Tight finger flexors during grasp (F3)
F3
Wrist flexion > 20°
Wrist extension powered by finger flexion
Active finger extension and flexion
Distinction between F3 and F4
Wrist is in >20° flexion in each of these levels.
Levels are differentiated if wrist movement is:
Present towards extension (F3) or Absent (F4)
F4
Wrist flexion > 20°
Wrist extension absent
Active finger extension and flexion
Distinction between F4 and F5
Wrist movement is absent in each of these levels.
Levels are differentiated by active finger movement:
Present (F4) or Minimal/Absent (F5)
F5
Wrist flexion > 20°
Minimal to no active wrist movement
Minimal to no active finger movement
Note: Even though the success or type of grip is not needed for classification, success of picking up the object is usually observed at levels E1, F1, F2 and F3.
The object is usually presented to the client in levels E2 and F5.
To be classified as F3, the movement into passive wrist extension must be independently generated by the force of the client's own hand on the object they are attempting to grasp and release.
Figure 7: Distinction between NHDC classification levels
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7. Clinical reporting

Having observed the client's movement, a classification level (e.g. F2, F4 or E1) is recorded. The following are **examples** of how this information may be documented as part of a comprehensive report or communicated to relevant people involved in the client's care.

F2 classification:

The NHDC was used to classify (client's name) left hand deformity at an F2 level. (Client's name) remained in wrist flexion during the approach, grasp and release of objects and released objects to the table top with his/her wrist position flexed at 45°. During the observed movement (client's name) demonstrated active wrist and finger extension on approach to the object.

F4 classification:

The NHDC was used to classify (client's name) right hand deformity at an F4 level. Objects were presented to (client's name) while seated in their supported seating system, which included a tray. On presentation of each object (client's name) extended his/her fingers in an attempt to grasp the object. His/her wrist remained at 70° of wrist flexion. No active wrist extension or passive movement driving wrist extension was observed. See the clinical example of a client classified with an NHDC level of F4 in section 11.1. In this case the information was used to prescribe an orthotic intervention as illustrated.

E1 classification:

The NHDC was used to classify (client's name) right hand deformity at an E1 level. (Client's name) approached the object with their wrist in extension. Active finger flexion and extension was observed but the wrist remained in extension during grasp and release of the object.

This structure can be followed for other levels of the classification.

This information will be useful to:

- Report the client's wrist and hand movement in action during a task;
- Maintain a record of the client's wrist and hand movement in action over time in a consistent format;
- Use the reported information for comparison with information gained from future use of the NHDC; and
- Prompt the clinician to consider intervention strategies to match the client's current level of active movement, for example if the client is classified with an NHDC F1 level, bimanual therapy or goal directed training may be an appropriate intervention to consider.

8. Clinical application

The NHDC is designed to guide consideration of the structures involved and enhance clinical decision making in the management of hand deformity. **The NHDC should not be used in isolation to determine intervention for a client with neurologically-based upper limb impairment.** Table 1, "The Neurological Hand Deformity Classification" (Section 9) outlines the structural and functional movement impairments associated with hand deformity. Identification of the dominant muscles and forces driving the pattern of deformity are considered and can be used to guide further clinical assessment and clinical intervention decision making. A comprehensive assessment is essential when considering any upper limb intervention. Assessment of upper limb impairment should include observation of posture and movement patterns and measurement of passive and active range of motion (Wilton, 2013).

9. The Neurological Hand Deformity Classification (Wilton, 2013)

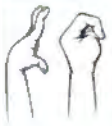
Type	F1. Wrist flexion ≤ 20°, thumb adduction	F2. Wrist flexion > 20°, active wrist & finger extension	F3. Wrist flexion > 20°, wrist extension powered by finger flexors and extensors	F4. Wrist flexion ≤ 20°, active finger flexion & extension, wrist extension absent	F5. Wrist flexion finger flexion, minimal active movement	E1. Wrist extension, finger movement powered by intrinsic muscle action	E2. Wrist extension, finger flexion, minimal active movement
							
Associated thumb deformity	Not always present CMC Adduction	CMC adduction MCP extension IP hyper extension	CMC adduction MCP & IP vary	CMC adduction MCP & IP vary	CMC adduction MCP & IP flexion	CMC adduction MCP flexion IP neutral	CMC adduction, MCP & IP flexion
Associated finger patterns	Hyper extension of PIP joints	Hyper extension of PIP joints	Hyper extension of PIP joints	Hyper extension of PIP joints	Flexion of IP joints	MCP flexion IP extension	Flexion, adduction at MCP joints, flexion of IPs associated with wrist extension posture
Primary location of spasticity	FCU AP	FDP & FDS AP, 1st DI	FCU, FCR, PL FDP & FDS AP	FCU, FCR, PL FDP & FDS AP	Combined spasticity extrinsic & intrinsic musculature of the fingers and thumb	ECRL & ECRB, ECU contributes to ulnar deviation, interossei, AP, FPB	Combined spasticity extrinsic & intrinsic musculature of the fingers and thumb
Muscles not effected by spasticity	Wrist extensors Extrinsic & intrinsic finger flexors & extensors	Wrist & thumb extensors Intrinsic finger musculature	Intrinsic finger musculature	Intrinsic finger musculature	Wrist musculature opposite to wrist position	FDP & FDS	Wrist flexor musculature opposite to wrist position
Contracture	Thumb web space	FDP & FDS end range extension Thumb web space	FCU, FCR limiting end range wrist extension combined with loss of end range FDS & FDP. Thumb web space.	FCU, FCR, PL, FDP, FDS limiting end range extension, Thumb web space	Severe deformity in wrist, fingers & thumb muscles with deficits in volar skin & soft tissue	Intrinsic finger flexors, thumb, palmar skin & fascial shortening. Hand hygiene critical	Wrist extensors & dorsal wrist capsule. Palmar skin & fascial contracture potential severe deformity of wrist, fingers MCP joints & thumb.
Functional deficit	Nil Limited thumb abduction compromising thumb span to clear object for grasp	Palm orientation in grasp, wrist control during finger flexion Thumb disadvantaged effective opposition	Reach and grasp compromised by wrist extension powered by active finger extension or reversed tenodesis action	Approach & grasp compromised by wrist position	No Function	Opening fingers & thumb for grasp disadvantaged by wrist extension - finger flexion/extension possible if wrist in neutral & thumb abducted	No Function

Table 1: The Neurological Hand Deformity Classification

10. Orthotic intervention considering hand deformity classification

Once the primary structures causing hand deformity have been identified, therapists need to determine the course of intervention/s, with orthotic intervention being only one potential option. It is essential that orthotic intervention includes clear understanding and description of the primary purpose of the orthosis (Garbellini, Robert, Randall, Elliott, & Imms, 2017).

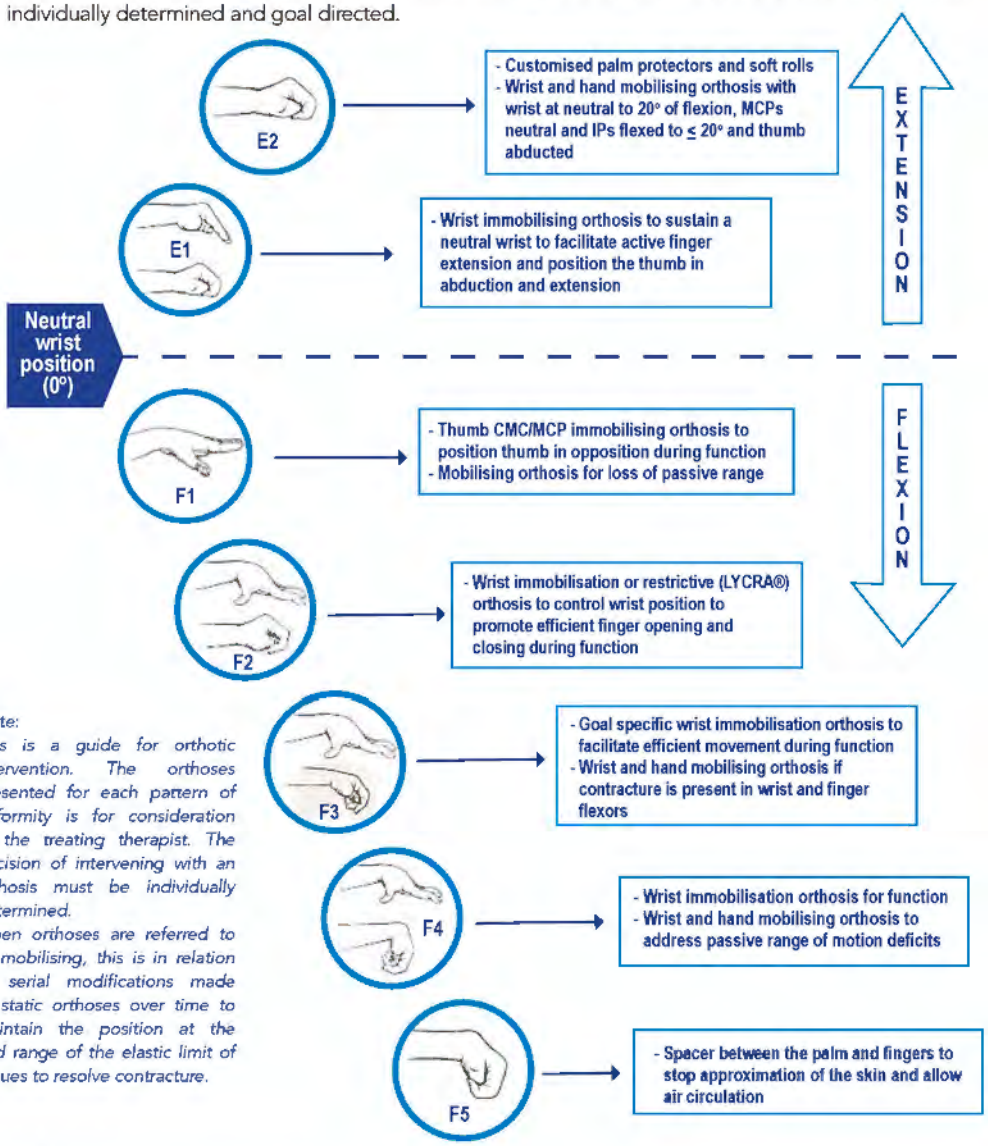
The primary purpose of orthoses are outlined in Table 2 below.

Primary purpose of orthosis	Definition
Immobilisation	Stop motion at a specific joint
Mobilisation	Apply forces to gain motion, either passively or as a dynamic movement assist
Restrictive	Prevent motion in one direction at a specific joint, while allowing motion in another direction
Torque transmission	Transfer movement torque created in one joint to another proximal or distal

Table 2: Primary purpose of orthoses, as derived from Australian Hand Therapy Association (2012); Colditz (1996); Fess (2011); American Society of Hand Therapists (1992).

11. Orthosis considerations utilising the NHDC

Figure 8 identifies orthoses that may be considered for each pattern of hand deformity and is provided as a tool to facilitate therapists' clinical decision making. It is essential that the choice of orthosis is individually determined and goal directed.



Note:
This is a guide for orthotic intervention. The orthoses presented for each pattern of deformity is for consideration of the treating therapist. The decision of intervening with an orthosis must be individually determined.
When orthoses are referred to as mobilising, this is in relation to serial modifications made to static orthoses over time to maintain the position at the end range of the elastic limit of tissues to resolve contracture.

Figure 8: Orthosis considerations using the NHDC

11.1 Clinical example

A client with an NHDC of F4 is portrayed in Figure 9 below. The client presented with a flexed wrist posture greater than 20° and active finger flexion. Active wrist and finger extension were absent. Comparison of previous clinical examination identified increasing loss of the client's composite passive wrist and finger extension.

Figure 9: F4 classification

Figure 10: Wrist and hand orthosis

The dorsal/volar wrist and hand mobilising orthosis, as pictured for the same client in Figure 10, is designed to address passive range of motion deficit and is worn at night to provide a low load, long duration stretch to flexor compartment musculature. The orthosis can be serially adjusted over time to accommodate any changes in range of motion. After consistent night wear, for at least 6-8 hours in duration a day and a period of three months, it would be anticipated that the client's passive range of motion would be greater than when orthosis wear commenced.

12. Conclusion

The NHDC is intended to:

- Categorise wrist and hand movement in action;
- Assist identification of structures driving hand deformity;
- Guide clinical assessment; and
- Enhance clinical decision making regarding the use of upper limb orthoses as part of the overall management of upper limb impairment.

13. Authors

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Simon has been working as an occupational therapist for more than 20 years. He is currently a Senior Occupational Therapist for the Cerebral Palsy Mobility Service within the Department of Paediatric Rehabilitation at Perth Children's Hospital. He runs a private practice with a focus on management of neurological upper limb impairment.

Simon is an associate investigator within two multicentre randomised controlled trials, at Australian Catholic University (ACU), investigating the medium to long-term use of rigid wrist and hand orthoses for children with cerebral palsy. Simon is completing a PhD at ACU that is embedded within the trials to investigate clinical decision-making about upper limb orthosis prescription for children with cerebral palsy, and use of the Neurological Hand Deformity Classification (NHDC) as a tool to guide orthotic prescription. Simon is a member of the Australian Hand Therapy Association (AHTA) and Australasian Academy of Cerebral Palsy and Developmental Medicine (AusACPDM).

Judith Wilton MSc, PGradDipHthSc, BAppSc (OT)

Judith has worked in upper limb rehabilitation as a clinical practitioner and academic since graduating as an occupational therapist over 35 years ago. She has worked in hand therapy private practice, and consulted to numerous organisations providing services to clients with neurological impairment.

Judith was a member of faculty of Curtin University's School of Occupational Therapy for over 20 years teaching undergraduate and postgraduate courses. She established the first Australian university-based hand and upper limb rehabilitation graduate programme for occupational therapists and physiotherapists. She has published numerous journal articles and presented courses and workshops on hand therapy and orthotic fabrication across Australia and internationally.

Judith is an active member of the AHTA, and has held numerous executive committee positions including National President. She is responsible for the development and continued presentation of courses on Hand and Upper Limb Orthotic Fabrication conducted by the AHTA across Australia. She was awarded life membership of the AHTA in 2002.

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16. References

American Society of Hand Therapists. (1992). Splint Classification System. Online: American Society of Hand Therapists.

Australian Hand Therapy Association. (2012). Australian Hand Therapy Association Hand and Upper Limb Orthosis/Splint schedule. Online.

Colditz, J. C. (1996). Principles of splinting and splint prescription. In C. A. Peimer (Ed.), Surgery of the Hand and Upper Extremity (pp. 2389-2410). New York: McGraw-Hill, Health Professions Division.

Fess, E. E. (2011). Orthoses for mobilization of joints: principles and methods. In T. M. Skirven, A. L. Osterman, J. Fedorczyk, & P. C. Amadio (Eds.), Rehabilitation of the hand and upper extremity : expert consult (pp. 1588-1598). Philadelphia: Elsevier Health Sciences.

Garbellini, S., Robert, Y., Randall, M., Elliott, C., & Imms, C. (2017). Rationale for prescription, and effectiveness of, upper limb orthotic intervention for children with cerebral palsy: a systematic review. Disability and Rehabilitation, 1-11. doi:10.1080/09638288.2017.1297498

Georgiades, M., Elliott, C., Wilton, J., Blair, E., Blackmore, M., & Garbellini, S. (2014). The Neurological Hand Deformity Classification for children with cerebral palsy. Australian Occupational Therapy Journal, 61, 394-402. doi:10.1111/1440-1630.12150

House, J., Gwathmey, F., & Fidler, M. (1981). A dynamic approach to the thumb-in palm deformity in cerebral palsy. J Bone Joint Surg Am, 63(2), 216-225.

Kirshner, B., & Guyatt, G. (1985). A methodological framework for assessing health indices. Journal of chronic diseases, 38(1), 27-36.

Klingels, K., De Cock, P., Molenaers, G., Desloovere, K., Huenaerts, C., Jaspers, E., & Feys, H. (2010). Upper limb motor and sensory impairments in children with hemiplegic cerebral palsy. Can they be measured reliably? Disability & Rehabilitation, 32(5), 409-416.

McConnell, K., Johnston, L., & Kerr, C. (2011). Upper limb function and deformity in cerebral palsy: a review of classification systems. Developmental Medicine & Child Neurology, 53(9), 799-805. doi:10.1111/j.1469-8749.2011.03953.x

Portney, L., & Watkins, M. (2015). Foundations of clinical research : applications to practice (3rd Edition. ed.): Philadelphia, PA : F.A. Davis Company.

Rosenbaum, P., Eliasson, A.-C., Hidecker, M. J. C., & Palisano, R. J. (2014). Classification in Childhood Disability: Focusing on Function in the 21st Century. Journal of Child Neurology, 29(8), 1036-1045. doi:10.1177/0883073814533008

Rosenbaum, P., Russell, D. J., Cadman, D. T., Gowland, C., Jarvis, S., & Hardy, S. (1990). Issues in Measuring Change in Motor Function in Children with Cerebral Palsy: A Special Communication. Physical Therapy, 70(2), 125-131.

Tonkin, M. A., Freitas, A., Koman, A., Leclercq, C., & Van Heest, A. (2008). The surgical management of thumb deformity in cerebral palsy. J Hand Surg Eur Vol, 33(1), 77-80. doi:10.1177/1753193407087891

Tonkin, M. A., Hatrick, N. C., Eckersley, J. R., & Couzens, G. (2001). Surgery for cerebral palsy part 3: classification and operative procedures for thumb deformity. J Hand Surg Br, 26(5), 465-470. doi:10.1054/jhsb.2001.0601

Wilton, J. (2003). Casting, splinting, and physical and occupational therapy of hand deformity and dysfunction in cerebral palsy. Hand clinics, 19(4), 573-584.

Wilton, J. (2013). Orthotic intervention and casting in the presence of neurological dysfunction Hand Splinting Orthotic Intervention: Prinicples of design and fabrication (2nd ed., pp. 195-235). Western Australia: Vivid Publishing.

World Health Organization. (2001). International Classification of Functioning, Disability and Health. Geneva. World Health Organization, 1-303.Zancolli, E. A. (2003). Surgical management of the hand in infantile spastic hemiplegia. Hand clinics, 19(4), 609-629.

Zancolli, E. A., & Boyes, J. H. (1979). Structural and dynamic bases of hand surgery. Philadelphia: J.B. Lippincott.

Zancolli, E. A., Goldner, L., & Swanson, A. (1983). Surgery of the spastic hand in cerebral palsy: Report of the Committee on Spastic Hand Evaluation. J Hand Surg, 8(5.2), 776 - 772.

Zancolli, E. A., & Zancolli, E. R. (1981). Surgical management of the hemiplegic spastic hand in cerebral palsy. The Surgical clinics of North America, 61(2), 395-406.

Zancolli, E. A., & Zancolli, E. R. (1984). The infantile spastic hand surgical indications and management. Annales de Chirurgie de la Main, 3(1), 66-75. doi:http://dx.doi.org/10.1016/S0753-9053(84)80064-2

Zancolli, E. A., & Zancolli, E. R. (1987). Surgical rehabilitation of the spastic upper limb in cerebral palsy. In D. W. Lamb (Ed.), The Paralysed Hand. Edinburgh [Lothian]; New York: Churchill Livingstone.ww

5.3 Chapter Conclusion

This chapter has provided the procedural information that was developed, in the form of a manual and a website, for the use of the NHDC. The focus of the next chapters of this doctoral program were to: explore the psychometric properties of construct validity and test–retest and inter-rater reliability of the NHDC (Study 3, Chapter 6); and present the clinical application of the NHDC in the form of case studies (Study 4, Chapter 7).

Chapter Six

6

Validity and reliability of the Neurological Hand Deformity Classification

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6.1 Introduction

The Neurological Hand Deformity Classification (NHDC), outlined in the previous chapter, was developed to classify hand deformity attributed to neurologically-based upper limb impairment (Garbellini & Wilton, 2017). The usefulness of the NHDC for clinical decision making depends on the extent to which clinicians can rely on the accuracy and relevance of the level of hand deformity classified (Portney & Watkins, 2015). Although classifications may be less complex than other assessments and measurement tools, it is important that they are evaluated for psychometric rigor. This includes evaluation of validity and reliability, to ensure consistency in reporting clinical and functional presentations (McConnell et al., 2011; Randall, Harvey, et al., 2013). Validity and test–retest reliability of the NHDC were not explored in the initial reliability study by Georgiades et al. (2014) and are explored further in this study. In addition, as part of the multi-centre randomised controlled trials (RCT) investigating the use of rigid wrist and hand orthoses – iWHOT and MIT (Imms et al., 2016) – it was possible to assess repeat inter-rater reliability with a larger sample size. The purpose of this study and the extended methodology are presented in the first part of this chapter. The second part of this chapter presents the manuscript prepared and submitted for consideration for publication in the *Journal of Hand Therapy*.

6.2 Study purpose

The purpose of this study was to further evaluate the psychometric properties of the NHDC. The study aimed to determine if the NHDC measured the construct of hand deformity to establish validity (Portney & Watkins, 2015). It also aimed to establish reliability by determining if the NHDC was consistent in discriminating between severity of hand deformity and, if the categorisation was reproducible, to establish reliability (Portney & Watkins, 2015).

6.3 Extended methodology

6.3.1 Design

A measurement study design (Portney & Watkins, 2015) was used to explore construct validity, test–retest and inter-rater reliability of the NHDC. This study was embedded within, and sourced data from, two multi-centre RCTs. The trials investigated the use of rigid wrist and hand orthoses for children (aged 0–3 years and 5–15 years) with CP (Imms et al., 2016). The trials were multi-centred and sponsored by Australian Catholic University.

6.3.2 Ethics

Ethical approval was sought and received from all relevant Health Research and Ethics Committees (HREC) of participating sites (Appendix D). Sites of the trials included: Australian Catholic University; Cerebral Palsy Alliance in NSW; Monash Children's Hospital and The Royal Children's Hospital in Victoria; and Perth Children's Hospital in Western Australia (Imms et al., 2016). The initial study site in Western Australia was Princess Margaret Hospital (PMH), until services relocated to the new Perth Children's Hospital in June 2018. As a result, the Child and Adolescent Health Services Human Research Ethics Committee migrated all PMH approved projects to the Research Governance Service and allocated new approval numbers. Approval numbers for all sites are included in the manuscript presented in the second part of this chapter. Informed written consent was obtained from parents or guardians who agreed to their child's participation in the study.

6.3.3 Participants

To be eligible for inclusion participants were diagnosed at risk of, or with, CP, and were aged between zero and three years or five and 15 years at the time of recruitment. Participants presented with abnormal or persistent flexion postures of the wrist and/or fingers/thumb during active use of the hand. The parents understood spoken and written English. Participants with a predominant dystonic motor type, allergy or sensitivity to thermoplastic material or inability to access one of the study sites were excluded (Imms et al., 2016).

6.3.4 Recruitment

Study information was provided to potential participants identified by the treating clinical teams at each site. Informed consent was sought from the parent(s)/guardian of the child. Eligible participants were confirmed by an assessor blind to group allocation at the participant's baseline assessment (Imms et al., 2016).

6.3.5 Data collection

All assessment data were collected by an occupational therapist/physiotherapist, trained in reliable administration of all measures. The assessors were blinded to the participant's allocation to orthosis or non-orthosis group. Test–retest data were collected at the Western Australian site by the investigator for this study (SG). All data were collected using a customised data collection form. Each data collection form is kept securely at each study site. Assessment data forms were scanned and electronically submitted, via a secure and ethically approved method, to the Project

Manager at ACU for central data management (Imms et al., 2016). Data were extracted from datasets of the two RCTs from August 2015 until September 2018.

6.3.6 Measures

The key dependent variable of this study was hand deformity as classified with the NHDC. Independent variables were selected to tap constructs from across the ICF-CY components. Measures of the independent variables were used to explore their relationships with the NHDC according to pre-specified hypotheses. The selected measures and the related hypotheses are presented in Table 6.1.

Table 6.1 Independent variables and hypothesised relationships with the NHDC

Measure [Variable name]	Construct	Redcap Database (Value labels) [Variable name]	Clinical use	Research use	Hypothesised NHDC correlation/ relationship	Analysis
Personal characteristics						
Age [Age]	The length of time a person has lived [a]	Date of Birth (DD-MM-YYYY) [demo_dob] Assessment date [ass_date] *Used to calculate age*	Expected developmental milestones Descriptor	Longitudinal data may show change over time with age Descriptor	As age increases (measured in years) NHDC increases in severity (F1 to F5 or E1 to E2) Fair positive correlation as deformity is predicted to occur over time	Spearman correlation coefficient
Gender [Gender]	Either of the two sexes (male or female) [a]	Male (1) Female (2) [demo-sex]	Descriptor	Descriptor	No relationship between gender and the NHDC	Chi squared test of independence
Manual Ability Classification System [MACS]	How children with CP handle objects in daily activities [b]	Level I (1) Level II (2) Level III (3) Level IV (4) Level V (5) [final_macs]	Classifies use of both hands together Child's ability to handle objects Classification	Describes population Level I least impaired Level V most impaired Classification	As the NHDC increases in severity (F1 to F5 or E1 to E2), the level of manual ability, as classified on the MACS (levels I to V) will increase At least a moderate to good positive correlation.	Spearman correlation coefficient

Measure [Variable name]	Construct	Redcap Database (Value labels) [Variable name]	Clinical use	Research use	Hypothesised NHDC correlation/ relationship	Analysis
Body structure and function domain of the ICF-CY (World Health Organisation, 2007)						
Passive Range of Motion [PROM]	Maximum joint motion achieved by application of an external force [c]	+ Extension range - Flexion range [wefe_prom_r_r2] [wefe_prom_l_r2]	Impaired range affects position of arm and hand May develop into contractures [d]	Measurement to track change over time	As the NHDC increases in severity (F1 to F5) the amount of passive wrist range of motion with fingers extended will decrease At least a moderate to good negative correlation	Spearman correlation coefficient
Active Range of Motion [AROM]	Maximum joint motion achieved by voluntary movement[c]	+ Extension range - Flexion range [bafa_ass_t1r] [bafa_ass_t1l] *Three trials 1, 2 and 3*	Limited active movement is related to limitations in hand activity [e]	Measurement to track change over time	As the NHDC increases in severity (F1 to F5) the amount of active range of motion (wrist extension with fingers extended, measured in degrees) will decrease. At least a fair negative correlation	Spearman correlation coefficient
Modified Ashworth Scale [MAS]	Assesses muscle tone by moving a joint and recording resistance to the movement [f]	1 (1) 1+ (2) 2 (3) 3 (4) 4 (5) [wefe_mas_r] [wefe_mas_l]	Impact of tone on movement [g]	Measurement to track change over time Tone increases as numbers on the scale increase	As the NHDC increases in severity (F1 to F5) the score on the Modified Ashworth Scale will increase in severity (5 items as recorded in RCT) At least a fair positive correlation	Spearman correlation coefficient

Measure [Variable name]	Construct	Redcap Database (Value labels) [Variable name]	Clinical use	Research use	Hypothesised NHDC correlation/ relationship	Analysis
Modified Tardieu Scale [MTS]	Assesses spasticity by the angle of initial catch on rapid passive movement (R1) to end of joint range (R2) [f]	+ Extension range - Flexion range [wefe_prom_r_r1] [wefe_prom_l_r1]	Impact of spasticity on movement [g]	Quantifies intensity of muscle reaction to quick movement [g]	As the NHDC increases in severity (F1 to F5) the R1 of the participant will occur earlier in the range of rapid passive wrist extension with fingers extended. Fair negative correlation	Spearman correlation coefficient
Activity domain of the ICF-CY (World Health Organisation, 2007)						
Box and block test of manual dexterity [B&B]	Tests manual ability [h]	Number of blocks moved in one minute Raw score ≤0 [bbt_r] [bbt_l]	Measures level of dexterity	Level of manual dexterity can be compared to normative data Can be used to assess effect of treatment [h]	On average, participants classified as E1, F1 and F2 on the NHDC will move more blocks in one minute compared to those participants classified as E2, F3-F5 on the NHDC	T test

Measure [Variable name]	Construct	Redcap Database (Value labels) [Variable name]	Clinical use	Research use	Hypothesised NHDC correlation/ relationship	Analysis
The Pediatric Evaluation of Disability – Computer Aided Test (PEDI-CAT) [PEDICAT]	Measure of function in daily life across four domains [i] Only the self-care domain was used.	Daily activity score – represents the child's status of function in self-care domain [i]	Measure of functional ability	Functional outcome measure for research [i] Response scores are on four-point scale ranging from 'unable' to 'easy'. Items summed to create the standard score will be used in analysis	As the NHDC increases in severity (F1 to F5) the score on the PEDI-CAT will tend to decrease. Hand deformity is hypothesised to have little negative correlation as limitations of function in self-care may differ depending on involvement of one or both upper limbs	Spearman correlation coefficient
ABILHAND-Kids questionnaire [ABILHAND]	Manual ability measured by completion of a questionnaire [j]	Total score (addition of ratings from impossible (0), difficult (1) to easy (2) with a range of 0–42) [j]	Scale measuring manual ability Provides guideline for treatment planning [j]	Measure of manual ability status over time Rasch analysis converts raw scores into a linear measure [j]	As the NHDC increases in severity (F1 to F5) the ABILHANDS-Kids score will decrease At least a fair negative correlation	Spearman correlation coefficient

Note: The strength of the relationship between variables (effect size) is verbally described as: 0.00 to 0.25 Little or no relationship; 0.25 to 0.50 Fair relationship; 0.50 to 0.75 Moderate to good relationship; and above 0.75 Good to excellent relationship (Portney & Watkins, 2015).
References: [a]*Oxford Dictionary* (2018); [b]Eliasson et al. (2006); [c]Horger (1990); [d]de Bruin et al. (2013); [e]Braendvik et al. (2010); [f]Numanoğlu and Günel (2012); [g]Morris (2002); [h]Mathiowetz et al. (1985); [i]Haley et al. (2011); [j]Arnould et al. (2004).

6.3.7 Procedures

Procedure for validity testing

Construct validity was assessed by calculating correlations between baseline assessment data of the NHDC and other measures used (Terwee et al., 2007; Westen & Rosenthal, 2003) in the RCT (Imms et al., 2016), for example, active and passive range of motion. If baseline data required for analysis were not complete, the next assessment timepoint containing all outcome measures required was used. Hypotheses set in advance allowed comparison of correlations to determine whether the correlations between the NHDC and other outcome measures had a similar association as predicted by the researchers. In other words, determining if the levels on the NHDC behaved as expected given the constructs under consideration (Mokkink et al., 2009; Westen & Rosenthal, 2003). The construct of hand deformity was expected to correlate in varying ways depending on the different independent variables as presented in Table 6.1. Correlation coefficients were interpreted as follows:

- 0.00 to 0.25 Little or no relationship;
- 0.25 to 0.50 Fair relationship;
- 0.50 to 0.75 Moderate to good relationship; and
- above 0.75 Good to excellent relationship (Portney & Watkins, 2015).

Procedure for test–retest reliability

A repeated measure component was implemented to establish test–retest reliability of the NHDC (Landis & Koch, 1977; Mokkink et al., 2018; Mokkink et al., 2009). Hand deformity of a sample of the Western Australian participants was classified on two occasions with the NHDC (Mokkink et al., 2009). Retest classification was completed within a two-week period. This was to test the hypothesis that the classification can be reliably made on repeat occasions and establish stability of the measure (Polit, 2014). To establish test–retest, timing of data collection for the second occasion varied across the period of the RCT. Most of the retesting was done following baseline or 6 monthly follow up assessment.

The NHDC is a classification of hand deformity according to observation of the dynamic interplay of wrist and hand muscles in action. There was no risk of participant recall or learning bias for this classification. The repeated classification within the two-week period was chosen to avoid assessment fatigue, yet be recent enough to avoid genuine clinical changes over time (Mokkink et al., 2018; Mokkink et al., 2009; Portney & Watkins, 2015; Terwee et al., 2007).

Testing conditions regarding set-up, instructions and objects used to observe the wrist and hand muscles in action followed the standard procedure as outlined in the Neurological Hand Deformity Classification manual and website. The manual and website are available online (Garbellini & Wilton, 2017) and are described in the previous chapter of this thesis (Chapter 5).

In this study, all NHDC assessments were video-recorded for later scoring. A freely available online tool, Research Randomizer (Urbaniak & Plous, 2018), was used to generate a random order for both the initial test and retest video recordings of the participant's NHDC. Each participant's randomised NHDC video recording was classified. To minimise the chance of the investigator's (SG) recall, or classification of the same participant's data in sequence, the randomised retest NHDC video recordings were classified after a wait period of three weeks.

Procedure for assessing inter-rater reliability

Data for the inter-rater reliability component of the study was sourced from NHDC video recordings of the children participating in the RCT at all sites. Video footage of each participant's wrist and hand movement allowed two raters, the RCT assessor blinded to the participant's group allocation and the current researcher (SG), to observe the same performance and classify hand deformity with the NHDC. Baseline assessment data of the NHDC was preferentially used. If baseline data for NHDC was missing the next assessment timepoint containing the required data for analysis was used. Prior to the researcher (SG) rating participant's NHDC, the Research Randomizer (Urbaniak & Plous, 2018) was again used to generate a random list of the participant's NHDC video recording to ensure a mix of the participants' data from each site.

6.3.8 Data analyses

Validity

Construct validity of the NHDC was assessed by comparing pre-determined hypothesised relationships to actual relationships (see Table 6.1). This was calculated with Spearman correlation coefficients, chi-squared test of independence or a dependent samples t-test depending on the data type (see Table 6.1). Comparisons were tabulated to consider the extent to which the NHDC scores performed as hypothesised. A positive rating for construct validity was determined if at least 75% of the pre-determined hypotheses corresponded to the results (Terwee et al., 2007).

Reliability

Test–retest reliability of the NHDC was assessed through percentage of absolute agreement between the repeated measures. Inter-rater reliability of the NHDC was assessed through percentage of absolute agreement between the two raters. In addition, Cohen's Kappa with 95% confidence intervals were produced, which has been chosen due to the nominal nature of the assessment tool's scale. Interpretation of Kappa coefficients will be as follows:

- < 0 Less than chance agreement;
- 0.01–0.20 Slight agreement;
- 0.21–0.40 Fair agreement;
- 0.41–0.60 Moderate agreement;
- 0.61–0.80 Substantial agreement;
- 0.81–0.99 Almost perfect agreement (Landis & Koch, 1977).

Terwee et al. (2007) and Streiner and Norman (2008) recommend an acceptable positive rating for reliability with an interpreted value of the kappa of at least 0.70 for a useful instrument. This guideline was used in interpreting whether the NHDC is a stable and reliable classification.

UNPUBLISHED MANUSCRIPT – THE NEUROLOGICAL HAND DEFORMITY CLASSIFICATION: CONSTRUCT VALIDITY, TEST–RETEST AND INTER-RATER RELIABILITY

Manuscript details

Title: The Neurological Hand Deformity Classification: construct validity, test–retest and inter-rater reliability

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Abstract

Background: The Neurological Hand Deformity Classification (NHDC) is an impairment-based classification that classifies hand deformity into one of two ordinal scales: flexion deformities (five levels) or extension deformities (two levels). Differentiation between the levels is determined by wrist position and wrist and finger movement.

Purpose: To examine aspects of validity and reliability of the NHDC.

Study design: A measurement design study guided by COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN).

Methods: Data from a convenience sample of 127 children with cerebral palsy (66 males: 61 females; age range 8mo to 14y 11mo; Manual Ability Classification System I (8.3%), II (47.2%), III (22.2%), IV (13.0%), V (9.3%)), participating in two randomized controlled trials investigating upper limb orthotic intervention were analysed. Construct validity was assessed by testing pre-determined hypotheses of relationships between the NHDC and measures of body function (for example, range of motion) and activity measures (for example, Box and Block Test of Manual Dexterity) with observed relationships. Reliability was assessed by calculating agreement between repeated measures (test–retest) and paired raters (inter-rater). Weighted kappa and Cohen's kappa with 95% confidence intervals were produced.

Results Predicted hypotheses for the NHDC were met in nine of 10 correlations with body structure measures and in two of seven correlations with activity measures. Test–retest for flexion deformities: κw=0.84; 95%CI 0.70 to 0.98; and extension deformities: κ=1.0; 95%CI 1.0 to 1.0 was good to excellent; inter-rater reliability for flexion deformities: κw=0.76; 95%CI 0.67 to 0.85; and extension deformities κ=0.75; 95%CI 0.43 to 1.0 was moderate to excellent.

Conclusion Expected relationships between the NHDC and other measures, stability between repeated measures and acceptable between-rater agreement supports confidence classifying hand deformity with the NHDC.

6.4 Introduction

Due to the heterogeneous nature of the cerebral palsy (CP) population, meaningful and reliable classification systems are important to ensure consistency in reporting clinical and functional presentations (Randall, Harvey, et al., 2013). Classifications are needed across a range of clinical domains, including hand function and deformity where impairment is common in children with CP (Eliasson, 2005). For example, a consecutively sampled cross-section of 100 children with CP from a children's hospital, identified 83% with upper limb involvement of which 36% had contracture, with limitations in the wrist and hand most frequent (Makki et al., 2014). In a population of 771 children with CP (aged 1–18 years, mean: 11 years 8 months), 19.4% had significantly decreased passive wrist extension with extended fingers at age four years compared to the youngest children (aged one to four years) (Hedberg-Graff et al., 2019). As severity of spasticity and hand deformity increases there is a higher level of impairment and functional deficits (Law et al., 2008; Russo et al., 2009).

Hand deformity classifications are a means to understand and analyse patterns of dynamic movement and assist in planning intervention (Wallen & Stewart, 2016). The Neurological Hand Deformity Classification (NHDC) (Garbellini & Wilton, 2017) (SI Figure 1, online supporting information) and Zancolli classification (McConnell et al., 2011; Zancolli, 2003) classify hand deformity. The Dynamic Positional Analysis (DPA) component of the Shriner's Hospital Upper Extremity Evaluation (SHUEE) (Davids et al., 2006) examines segmental positioning of the upper limb during activity. A comparison of these tools is presented in Table 6.2.

Table 6.2 Comparison of hand deformity classification tools

	Neurological Hand Deformity Classification	**Zancolli Classification**	**Dynamic Positional Analysis (SHUEE)**
Construct	Classification of the dynamic movement of wrist and hand muscles causing deformity.	Classification of volitional release of the fingers through active wrist flexion.	Documentation of the dynamic, segmental alignment of the upper extremity during activity.
Purpose	To assist identification of drivers for deformity and enhance clinical decision making regarding intervention planning.	To assist the selection of clients for surgical reconstruction.	To determine potential for improved function by identifying the dynamic position of the upper limb.
Population	Neurological impairment — Unilateral or bilateral upper limb presentation; any age.	CP — Unilateral or bilateral upper limb presentation; reported use from 4 years of age.	CP — Unilateral impairment (hemiplegia). 3–18 years of age.
Administration	Observation of the wrist and hand during movement towards, and attempted grasp and release of objects.	Observation of the degree of wrist flexion necessary to open the fingers and the ability to extend the wrist with fingers flexed during volitional movement.	Observation of the segmental alignment of the affected extremity during activity.
Basis of classification	The degree of extension or flexion of the wrist in relation to finger movement.	The degree of wrist flexion during volitional finger extension; active wrist extension with fingers flexed.	The alignment of five upper extremity segments (thumb, fingers, wrist, forearm and elbow).
Classification/Score	Wrist flexion classification levels: F1 to F5 Wrist extension classification levels: E1 to E2 Levels are graded and differentiated by the presence or absence of active wrist and finger movement relative to the flexed or extended wrist position.	I: Finger extension with <20–30^{o} of wrist flexion. II: Finger extension with >20–30^{o} of wrist flexion A – active wrist extension fingers flexed B – no active wrist extension fingers flexed. III: No active finger extension with active or passive wrist flexion.	For each segment available planes of movement are scored as follows: zero (pathological alignment) to three (normal or optimal alignment). Total score possible for each segment =12.

	Neurological Hand Deformity Classification	**Zancolli Classification**	**Dynamic Positional Analysis (SHUEE)**
Clinical application	To identify structures causing dynamic deformity to inform intervention such as use of orthoses.	To guide surgical intervention.	To track upper extremity function; select surgical interventions.
Reported evidence for validity	Explored in this study.	None.	Construct validity (wrist) was determined by a change in the mean wrist score out of 12, from zero (pre-surgery) to 6.4 (post-surgery), n=18 (Davids et al., 2006).
Reported evidence for reliability	Intra-rater: Almost perfect agreement κ=0.91 Inter-rater: Almost perfect agreement κ=0.87 n=26 (Georgiades et al., 2014). Further explored in this study.	Test–retest: Perfect agreement *κw*=1.00 Inter-rater: Almost perfect agreement *κw*=0.95 n=30 (Klingels et al., 2010).	ICC for wrist dynamic positional analysis: Intra-observer: excellent r=0.98 Inter-observer: excellent r=0.89 n=11 (Davids et al., 2006). Intra-observer: excellent r=0.83 Inter-observer: excellent r=0.77 n=20 (Heaver et al., 2015).

Note: ICC — Intraclass Correlation Coefficient

The NHDC was developed to address limitations noted with other hand deformity classifications (Georgiades et al., 2014). The NHDC includes wrist extension deformities and considers thumb posture, neither of which are included in the Zancolli classification (Georgiades et al., 2014). The NHDC also considers the movement of the fingers and thumb in relation to the wrist position while the DPA component of the SHUEE scores the segmental alignment of individual joints of the upper extremity (Davids et al., 2006). Thus, the NHDC offers a distinct approach to classifying hand deformity in CP.

The NHDC is framed within the body function and structure domain of the International Classification of Functioning, Disability and Health (ICF) (World Health Organisation, 2001), and does not classify or assess activity level performance. The NHDC provides a structured approach to identifying components of hand deformity through observation of the dynamic interplay of wrist and hand musculature (Garbellini & Wilton, 2017; Georgiades et al., 2014; Wilton, 2013b). Identifying these components may guide interventions to lessen the impact of those leading to progressive musculoskeletal change. High levels of inter-rater and intra-rater reliability (κ=0.87 and κ=0.91 respectively) were established in an initial reliability study involving 26 children and three observers (Georgiades et al., 2014). Evidence of validity has not been reported. The purpose of this study was to further examine the psychometric properties of the NHDC to establish confidence in its use.

The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN), study design checklist (Mokkink et al., 2019) guided the design and reporting of the psychometric properties of construct validity, and test–retest and inter-rater reliability. The construct validity of a measure is based on evidence supporting the measure's adequacy and usefulness of score interpretation in relationship with scores of other measures (Messick, 1995). Reliability is the extent to which a measure is consistent and free from error (Portney & Watkins, 2015). Test–retest and inter-rater reliability were investigated in this study. To build evidence about validity and reliability, this study explored three research questions.

When used with children with CP, does the NHDC demonstrate:

1. Construct validity as evaluated using hypothesis testing against specified criteria?
2. Test–retest stability between occasions of assessment when no change is expected?
3. Inter-rater reliability when different raters classify the same participants?

6.5 Methods

6.5.1 Design

A measurement study design was used and reported following the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines (von Elm et al., 2007). Data for this study were collected within two Australian multi-centre randomised controlled trials investigating the use of rigid wrist hand orthoses for children with CP: (i) the Infant Wrist Hand Orthosis Trial (iWHOT); and (ii) the Minimising Impairment Trial (MIT) (Imms et al., 2016). The trials were registered with the Australian New Zealand Clinical Trials Registry: iWHOT 12614001275651 and MIT 12614001276640. Eligibility for participation in the trials included children: (i) at risk for or diagnosed with CP; (ii) aged between zero and three years (iWHOT) or five and 15 years (MIT); and (iii) with abnormal or persistent flexion postures of the wrist/fingers and or thumb. Exclusion criteria included (i) upper limb dystonia without the presence of spasticity; and (ii) allergy or sensitivity to materials used to fabricate the orthoses (Imms et al., 2016).

6.5.2 Ethics

Ethical approval, for each trial site, was received from Australian Catholic University (HREC: 2014 317V and 2014 318V), Cerebral Palsy Alliance in NSW (HREC 2014-08-02 and 2014-08-03), Monash Children's Hospital (HREC: 14199B and 14201B), The Royal Children's Hospital (HREC: 34280A and 34279) in Victoria, and Perth Children's Hospital in Western Australia (HREC: RGS2456 and RGS2457). Informed written consent to participate was collected from parents or guardians of children on enrolment.

6.5.3 Data collection

NHDC data from the iWHOT and MIT, collected between August 2015 and September 2018, were extracted for this study. NHDC data were collected at baseline assessment and at six-month intervals, over a period of up to three years by assessors blinded to the participant's allocation to the orthosis or non-orthosis group. The NHDC can be scored live or from a video record of the person's available dynamic movement of the wrist and fingers during approach, grasp and release of an object. A website and manual (Garbellini & Wilton, 2017) have been developed which describe the standardised procedure for using the NHDC to ensure consistency in observation and/or recording. This was available to the assessors within the iWHOT and MIT. Observation of three attempts of wrist and hand movement may be required to make a classification, based upon the most consistent movement observed during the three attempts

(Garbellini & Wilton, 2017). A video record of the participant's NHDC was taken at each assessment timepoint in the iWHOT and MIT. The first occasion of iWHOT or MIT assessment (usually baseline) where NHDC level was recorded was used for analysis in this study.

6.5.4 Sample size

Sample sizes for the validity and reliability components of this study were guided by the COSMIN study design checklist, with an aim to recruit at least 100 participants (Mokkink et al., 2019).

6.5.5 Procedures

Validity: Construct validity was established by comparing whether pre-determined hypotheses of correlations between the NHDC and other outcome measures had similar associations with observed correlations, as predicted by the researchers (Mokkink et al., 2019; Westen & Rosenthal, 2003). Table 6.3 displays the independent variables, their construct, hypothesised relationship with the NHDC and analysis methods.

Table 6.3 Independent variables and their hypothesised relationship with the NHDC

Measure	Construct	Data source/s	Hypothesized NHDC correlation/relationship	Analysis method
Gender	Either of the two sexes (male or female) (Oxford Dictionary, 2018)	iWHOT MIT	No relationship between gender and the NHDC	Chi-squared test of independence
Passive Range of Motion (PROM)	Maximum joint motion achieved by application of an external force (Horger, 1990)	iWHOT MIT	As the NHDC flexion level increases (F1 to F5) the amount of passive wrist range of motion with fingers extended (measured in degrees) will decrease. At least a moderate to good negative correlation.	Spearman correlation coefficient
Active Range of Motion (AROM)	Maximum joint motion achieved by voluntary movement (Horger, 1990)	MIT	As the NHDC flexion level increases (F1 to F5), the amount of active range of motion (wrist extension with fingers extended, measured in degrees) will decrease. At least a moderate to good negative correlation.	Spearman correlation coefficient
Modified Ashworth Scale (MAS)	Assesses muscle tone by applying an external force to move a joint and recording resistance to the movement (Numanoğlu & Günel, 2012)	iWHOT MIT	As the NHDC flexion level increases (F1 to F5) the score on the Modified Ashworth Scale will increase (5 items as recorded in RCT). At least a fair positive correlation.	Spearman correlation coefficient
Modified Tardieu Scale (MTS)	Assesses spasticity by the angle of initial catch on rapid passive movement (R1) to end of joint range (R2) (Numanoğlu & Günel, 2012)	iWHOT MIT	As the NHDC flexion level increases (F1 to F5) the R1 will occur earlier in the range of rapid passive wrist extension with fingers extended. Fair negative correlation.	Spearman correlation coefficient
Box and block test of manual dexterity	Tests manual ability (Mathiowetz et al., 1985)	MIT	As NHDC flexion level increases (F1 to F5), participants will move fewer blocks in one minute. At least a fair negative correlation.	Spearman correlation coefficient
Age	The length of time a person has lived (Oxford Dictionary, 2018)	iWHOT MIT	As age increases (measured in years) hand deformity (NHDC level) may increase in some children. A fair positive correlation is predicted.	Spearman correlation coefficient

Measure	Construct	Data source/s	Hypothesized NHDC correlation/relationship	Analysis method
Manual Ability Classification System (MACS)	How children with CP handle objects in daily activities (Eliasson et al., 2006)	iWHOT MIT	As the NHDC levels increase (F1 to F5 or E1 to E2), the level of manual ability, as classified on the MACS (levels I to V) will increase. At least a fair positive correlation.	Spearman correlation coefficient
The Pediatric Evaluation of Disability – Computer Aided Test (PEDI-CAT): Self-care domain	Measure of level of function when performing self-care skills (Haley et al., 2011)	iWHOT MIT	As the NHDC levels increase (F1 to F5 or E1 to E2), the score on the PEDI-CAT will tend to decrease. No or little negative correlation.	Spearman correlation coefficient
ABILHAND-Kids questionnaire (ABILHAND)	Manual ability measured by completion of a questionnaire (Arnould et al., 2004)	MIT	As the NHDC levels increase (F1 to F5 or E1 to E2), the ABILHAND-Kids score will decrease. At least a fair negative correlation.	Spearman correlation coefficient

Reliability: Test–retest reliability was studied using a convenience sample from one iWHOT/MIT study site. Children were re-assessed using the NHDC within a two-week period following a scheduled assessment timepoint. As the NHDC is a classification of hand deformity according to observation of available dynamic hand movement, repeated classification within a two-week period was chosen to avoid fatigue from multiple attempts of movement, yet was recent enough to avoid clinical change occurring over time (Mokkink et al., 2018; Mokkink et al., 2009; Portney & Watkins, 2015).

Inter-rater reliability was studied by establishing the extent of agreement in classifications made by paired raters (Portney & Watkins, 2015). Participants were from multiple trial sites. A video record of participants' wrist and fingers in action allowed two independent raters to observe and classify the same performance using the NHDC. One rater (SG) rated all participants. The second rater was one of 12 blinded assessors employed and trained for iWHOT/MIT data collection, who contributed between 1 and 31 (mean 9.5 ratings per pair, total ratings=114) of the paired ratings. Where a difference in rating occurred between raters, the individual participant's video record was reviewed to explore possible causes of difference.

6.5.6 Statistical Analyses

The NHDC classifies hand deformity from a neutral wrist position into one of seven levels of deformity: flexion (F1 to F5) and extension (E1 to E2) (Garbellini & Wilton, 2017). To maintain the ordinal nature of the data, flexion and extension classifications were explored separately. Data were initially summarised and displayed graphically to explore relationships.

To investigate validity, the relationship of NHDC level for each limb (identified as correlation by limb) with: passive range of motion (PROM); peak active range of motion (AROM) (from up to three trials (Imms et al., 2016)); Modified Ashworth Scale (MAS) (Numanoğlu & Günel, 2012); Modified Tardieu Scale (MTS) (Numanoğlu & Günel, 2012); and Box and Block test of manual dexterity (Box and Blocks) (Mathiowetz et al., 1985) were explored (see Table 2). In the trials, only wrist extension, and not wrist flexion measures were taken for PROM, AROM, MAS and MTS, so it was not possible to correlate the extension deformity levels of the NHDC with these measures. The sample size is the total number of limbs for each analysis, regardless of the participant's unilateral or bilateral presentation.

The relationship of the NHDC (identified as correlation by child) with: gender; age; Manual Ability Classification System (MACS) (Eliasson et al., 2006); Pediatric Evaluation of Disability – Computer Aided Test (PEDI-CAT) (Haley et al., 2011); and the ABILHAND-Kids questionnaire (ABILHAND) (Arnould et al., 2004) was explored. For children with bilateral

presentation only one NHDC classification was used. The sample size is the number of participants for each relationship. If NHDC was the same for both limbs, the right limb measure was chosen for analysis. If the NHDC differed for both limbs, the more severely classified limb was chosen. The criteria for interpreting the strength of observed relationships was: 0.00 to 0.25 Little or no; 0.25 to 0.50 Fair; 0.50 to 0.75 Moderate to good; and above 0.75 Good to excellent relationship (Landis & Koch, 1977).

Test–retest reliability was assessed by calculating chance-corrected and absolute agreement between repeated measures. Weighted kappa and Cohen's kappa with 95% confidence intervals were produced for flexion and extension classifications respectively. Inter-rater reliability was assessed by calculating the percentage of agreement between two raters. Weighted kappa and Cohen's kappa with 95% confidence intervals were produced for flexion and extension classifications respectively. Interpretation of agreement using kappa was: 0 Less than chance; 0.01–0.20 Slight; 0.21–0.40 Fair; 0.41–0.60 Moderate; 0.61–0.80 Substantial; and 0.81–0.99 Almost perfect agreement (Streiner & Norman, 2008). For one data point, rater discrepancy was found between an F1 level (most active movement) and an F5 level (minimal to no active movement). Following review of the video record this data point was excluded as rater error was obvious.

6.6 Results

6.6.1 Participant characteristics

There were 127 participants aged 8 months to 14 years 11 months. The number and percentage of participant's topographical distribution, MACS and NHDC levels are presented in Table 6.4(a). Table 6.4(b) includes the number of participants (n) assessed with the measures used in the orthosis trials and was labelled accordingly.

Table 6.4(a) Participant characteristics (n=127)

Characteristic	n (%)	Range	Mean (SD)
Gender			
Male	66 (52%)	-	-
Female	61 (48%)		
Age at baseline assessment	-	8mo to 14y 11mo	6y 7mo (4y 7mo)
Topographical distribution			
Unilateral	96 (76%)	-	-
Bilateral	31a (24%)		
MACS [b]			
I	7 (5.9%)	-	-
II	56 (47.1%)		
III	25 (21.0%)		
IV	15 (12.6%)		
V	16 (13.4%)		
NHDC			
F1	34 (22.7%)		
F2	32 (21.3%)	-	-
F3	22 (14.7%)		
F4	14 (9.3%)		
F5	15 (10.0%)		
E1	27 (18.0%)		
E2	6 (4.0%)		

Table 6.4(b) Measures

Measure	n	Range	Mean (SD)
*PROM [c]	161	-92° to 97°	66.98° (36.76°)
†AROM [c] (MIT only)	72	-75° to 80°	2.69° (44.57°)
MAS [c]			
0	8	-	-
1	44		
1+	38		
2	51		
3	18		
4	0		
MTS [c]	114	-101° to 74°	-7.59° (39.54°)
Box and blocks [c] (MIT only)	79	0 to 40	10.92 (10.85)
PEDI-CAT [b]	120	30 to 61	47.27 (8.37)
ABILHAND [b] (MIT only)	69	14 to 21	20.55 (1.19)
Participants in analyses			
Validity	127	-	-
Test–retest	36		
Inter-rater	113		

Note: mo=months; y=years; [a] Of the 31 participants with bilateral involvement, 18 had the same NHDC level for both limbs and 13 had different NHDC levels for each limb. [b] by child; [c] all limbs included. *PROM denotes the end of passive range of wrist extension with fingers extended. †AROM denotes maximal active range of motion achieved of wrist extension with fingers extended.

6.6.2 Evidence for construct validity of the NHDC

As expected, there was no association between gender and NHDC for either flexion (X^2 (4)=1.898, p=0.755) or extension (X^2 (1)=0.160, p=0.689) deformities. Contrary to our hypothesis, there was little or no relationship between NHDC and age. The youngest participant classified at each level of the NHDC was: E2 – 1 year and 4 months; E1 – 8 months; F1 – 11 months; F2 – 13 months; F3 – 1 year 10 months; F4 – 2 years and 8 months; and F5 – 2 years 3 months (SI Figure 2, online supporting information). As expected, nine of the ten predicted relationships between NHDC and body structures measures, correlation by limb, were met. Only two of the seven predicted relationships between NHDC and activity measure, correlation by child, were met. Correlation coefficients between the NHDC and other measures by limb, and by child are displayed in Table 6.5(a) and 6.5(b) respectively.

Table 6.5(a) Spearman's rho correlation coefficients between NHDC and other measures
Correlation by limb

	NHDC Flexion						
	Right			Left			
	r_s	n	p	r_s	n	p	Hypotheses met
PROM	-0.442	62	0.000	-0.546	54	0.000	1 out of 2
AROM (peak)	-0.607	37	0.000	-0.632	28	0.000	2 out of 2
MAS	0.408	60	0.001	0.452	55	0.001	2 out of 2
MTS	-0.451	49	0.001	-0.381	43	0.012	2 out of 2
Box & Blocks	-0.564	41	0.000	-0.562	32	0.001	2 out of 2
						Total:	**9 out of 10**

Table 6.5(b) Spearman's rho correlation coefficients between NHDC and other measures
Correlation by child

	NHDC Flexion			NHDC Extension			
	r_s	n	p	r_s	n	p	Hypotheses met
Age	0.008	94	0.939	-0.244	25	0.240	0 out of 2
MACS	0.562	91	0.000	0.108	21	0.640	1 out of 2
PEDI-CAT	-0.327	90	0.002	-0.183	24	0.391	1 out of 2
ABILHAND	0.181	60	0.167	*Cannot be computed*			0 out of 1
						Total:	**2 out of 7**

Note: r_s =correlation; n = number; p = level of significance

6.6.3 Evidence for reliability of the NHDC

Absolute agreement for test–retest classifications (n=36 participants) for flexion deformities was 81% and extension deformities 100%. Test–retest reliability was almost perfect for flexion (κw=0.84; 95%CI 0.70 to 0.98) and extension (κ=1.0; 95%CI 1.0 to 1.0) deformities.

The absolute agreement between raters for flexion deformities (n=80 paired ratings) was 70% and 92% for extension deformities (n=26 paired ratings), and weighted kappa showed substantial agreement for flexion (κw=0.76; 95%CI 0.67 to 0.85) and extension deformity (κ=0.75; 95%CI 0.43 to 1.0) ratings.

Table 6.6 shows level of agreement for flexion deformity for test–retest and between raters.

Table 6.6 Cross-tabulation of NHDC flexion levels for (a) test–retest classifications, and (b) inter-rater classifications

(a)

Initial NHDC \ NHDC retest	F1	F2	F3	F4	F5	Total
F1	**2**	2	0	0	0	4
F2	0	**8**	0	1	0	9
F3	0	1	**2**	0	0	3
F4	0	0	1	**7**	0	8
F5	0	0	0	0	**3**	3
Total	2	11	3	8	3	27

(b)

NHDC Rater 1 \ NHDC Rater 2	F1	F2	F3	F4	F5	Total
F1	**22**	0	0	0	0	22
F2	6	**16**	3	0	0	25
F3	0	4	**5**	1	0	10
F4	0	0	6	**10**	0	16
F5	0	0	3	1	**3**	7
Total	28	20	17	12	3	80

Note: Numbers in ***bold*** indicate absolute agreement of NHDC at test–retest (a) and between raters (b).

Review of participants' video records where a difference in NHDC classification occurred identified possible causes of difference including: (i) objects being too big for the participant's hand; (ii) objects being placed too far away; (iii) the wrist position of younger children being obscured by wearable sensors (used as an outcome measure of passive and active movement in the iWHOT/MIT); and (iv) the passive extension movement of the participant's wrist being recorded as an F3 level, where minimal 'active' movement should have been classified as an F5 level.

6.7 Discussion

Construct validity of the NHDC was explored through testing relationships with other measures. For correlations by limb, nine out of ten predicted hypotheses were supported by the relationships found. As the NHDC is framed within the body function and structure domain of the ICF (World Health Organisation, 2001) the relationships found with other measures within the same domain support the NHDC as a valid tool to classify the construct of hand deformity.

For correlations by child, two of the predicted hypotheses for correlations with activity level measures of MACS (Eliasson et al., 2006) and PEDICAT (Haley et al., 2011) were met for NHDC flexion deformities, but not for extension deformities. The correlations demonstrated between increasing dynamic flexion deformity and activity measures may be due to the focus of those measures on ability to handle objects. This becomes increasingly difficult as flexion of the wrist and lack of finger movement (as classified on the NHDC) increases. These findings support clinical experience that as spasticity and hand deformity increase so does the level of impairment and functional deficits (Law et al., 2008; Russo et al., 2009). The lack of a relationship between measures with NHDC extension deformities could be due to: the restricted range of scores (only

two extension levels in the NHDC); small sample sizes, as only a small proportion of participants presented with extension deformities (25 out of 127); and very low prevalence, with only five participants classified with an E2 deformity. The lack of relationship between levels on the NHDC and scores on the ABILHAND (Arnould et al., 2004) may be the result of a restricted range of participants' ABILHAND scores (14–21 of possible 42—refer to Table 3). The restricted range provided little variation with which to explore the relationship.

Contrary to prediction that as age increases hand deformity may increase in some children, there was no relationship between NHDC and age. Our findings suggest that hand deformity may not show a steady increase with age as it may already be present at a young age: the youngest participant had a hand deformity classification of E1 at 8 months of age. Also, participants younger than three years were classified into each level of the NHDC. A child's NHDC level may provide an early indicator to identify those at risk of losing passive wrist extension, with extended fingers, by the age of four years, as found by Hedberg-Graff et al. (2019). NHDC level may not have a relationship with age as was predicted in this cross-sectional analysis, but longitudinal changes in an individual's NHDC level requires further exploration.

The NHDC demonstrated stability between occasions of classification as evidenced by almost perfect agreement in classifications given at two timepoints for the same cohort. Absolute agreement of 80% for flexion deformities and 100% for extension deformities provides strong support that the NHDC can achieve the same result with repeated administrations. Substantial agreement between raters using the NHDC was also established when rating the same observed movement of the wrist and fingers. Observed agreements of 70% for flexion deformities and 92% for extension deformities supports inter-rater reliability of the NHDC. The NHDC met an acceptable (and recommended) rating for reliability with a kappa of at least 0.7 (Streiner & Norman, 2008).

Reflection on the observed differences between raters (refer to Table 5B) provides an opportunity to consider what actions could be implemented to improve NHDC rater agreement. Most variation between raters occurred in classification of the F3 level. Review of participants' video records revealed differences in administration of objects to elicit the movement to classify hand deformity by some assessors, and the presence of atypical devices (wearable sensors) possibly obscuring accurate observation of wrist position. Reviewing the NHDC user manual and website identified a potential lack of clarity of instruction, and a need for additional description of level differentiation, particularly for F3 deformity. For example, the position of the wrist is key to classification of hand deformity using the NHDC. For a participant's hand to be classified at F3, the movement into passive wrist extension must be driven by the participant's

force of their hand on the object they are attempting to approach, grasp and release (Garbellini & Wilton, 2017). This point of difference for the F3 level, compared to F4 (where there is no active or passive wrist movement) or F5 (where the object needs to be presented to the child), is crucial for accurate classification. To increase accuracy in the clinical use of the NHDC administration instructions need to be updated to include emphasis on the differences between classification levels in the NHDC user manual and website (Garbellini & Wilton, 2017). Development of an online training module including video records to demonstrate NHDC administration and provide examples of each level of classification may increase reliable classification using the NHDC and build confidence in interpreting the clinical impact of NHDC levels (Portney & Watkins, 2015; Randall, Harvey, et al., 2013).

A strength of this study was it being situated within two larger national randomised trials, enabling data to be collected beyond one site, service provider, assessor and clinician. This extended both the volume of data and number of measures available against which to explore hypothesised relationships for the NHDC. However, participants with predominant dystonia were excluded from these trials and therefore this study. As the NHDC can be used for unilateral and bilateral neurologically-based upper limb presentations with any motor type the exclusion of those with dystonia, in addition to families declining to participate in the larger trials, are potential limitations to sample representativeness.

6.8 Conclusion

Evidence to support the validity of the NHDC as a tool to classify hand deformity was provided by the relationships confirmed with other measures of body function and structure. The relationships explored with activity level measures were less supportive. Each participant's level of hand deformity could be classified in one of the NHDC levels, regardless of age, manual ability, or topographical presentation. The NHDC demonstrated stability between occasions of classification and an acceptable level of rater agreement. These findings support the NHDC as a valid and reliable classification of hand deformity. Clarification of administration instructions to distinguish between classification levels, and development of an online training module is recommended to further enhance reliability.

6.9 Acknowledgements

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6.10 Supporting information

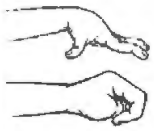
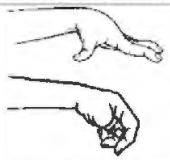
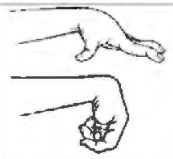
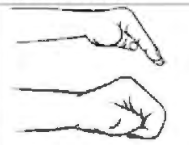
	Flexion Type					Extension Type	
	F1. Wrist flexion ≤ 20°, thumb adduction	**F2. Wrist flexion > 20°, active wrist & finger extension**	**F3. Wrist flexion > 20°, wrist extension powered by finger flexors and extensors**	**F4. Wrist flexion > 20°, active finger flexion & extension, wrist extension absent**	**F5. Wrist flexion finger flexion, minimal active movement**	**E1. Wrist extension, finger movement powered by intrinsic muscle action**	**E2. Wrist extension, finger flexion, minimal active movement**
							
Associated thumb deformity	Not always present CMC Adduction	CMC adduction MCP extension IP hyperextension	CMC adduction MCP & IP vary	CMC adduction MCP & IP vary	CMC adduction, MCP & IP flexion	CMC adduction MCP flexion IP neutral	CMC adduction, MCP & IP flexion
Associated finger patterns	Hyper extension of PIP joints	Hyper extension of PIP joints	Hyper extension of PIP joints	Hyper extension of PIP joints	Flexion of IP joints	MCP flexion IP extension	Flexion, adduction at MCP joints, flexion of IPs associated with wrist extension posture
Primary location of spasticity	FCU AP	FDP & FDS AP, 1st DI	FCU, FCR, PL FDP & FDS AP	FCU, FCR, PL FDP & FDS AP	Combined spasticity extrinsic & intrinsic musculature of the fingers and thumb	ECRL & ECRB, ECU contributes to ulnar deviation. Interossei, AP FPB	Combined spasticity extrinsic & intrinsic musculature of the fingers and thumb
Muscles not effected by spasticity	Wrist extensors Extrinsic & intrinsic finger flexors & extensors	Wrist & thumb extensors Intrinsic finger musculature	Intrinsic finger musculature	Intrinsic finger musculature	Wrist musculature opposite to wrist position	FDP & FDS	Wrist flexor musculature opposite to wrist position
Contracture	Thumb web space	FDP & FDS end range extension Thumb web space	FCU, FCR limiting end range wrist extension combined with loss of end range FDS & FDP. Thumb web space	FCU, FCR, PL, FDP, FDS limiting end range extension. Thumb web space	Severe deformity in wrist, fingers & thumb muscles with deficits in volar skin & soft tissue	Intrinsic finger flexors, thumb, palmar skin & fascial shortening. Hand hygiene critical.	Wrist extensors & dorsal wrist capsule. Palmar skin & fascial contracture potential severe deformity of wrist, fingers MCP joints & thumb.
Functional deficit	Nil Limited thumb abduction compromising thumb span to clear object for grasp	Palm orientation in grasp, wrist control during finger flexion Thumb disadvantaged effective opposition	Reach and grasp compromised by wrist extension powered by active finger extension or reversed tenodesis action	Approach & grasp compromised by wrist position	No Function	Opening fingers & thumb for grasp disadvantaged by wrist extension - finger flexion/extension possible if wrist in neutral & thumb abducted.	No Function

Figure 6.1 Neurological Hand Deformity Classification.

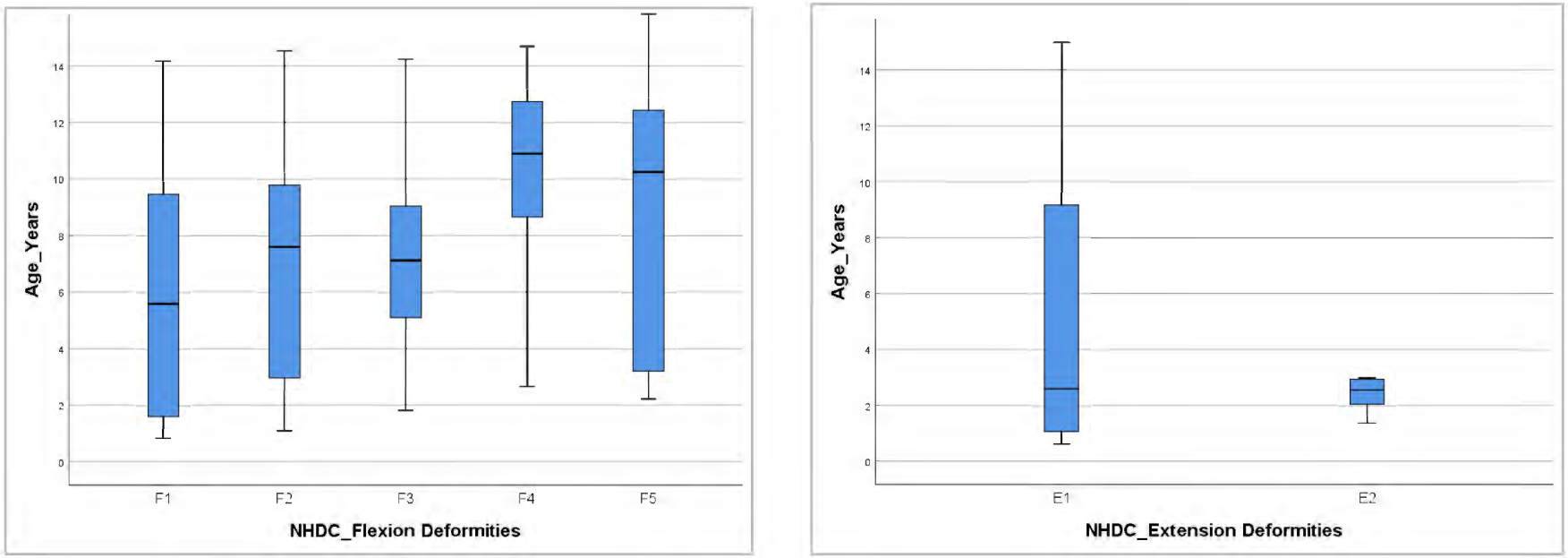


Figure 6.2 Relationship between NHDC and age

6.11 Chapter Conclusion

The aim of the measurement study in this chapter was to explore the construct validity and test–retest and inter-rater reliability of the NHDC. An extended methodology for the study was described in detail in the first part of the chapter. The results of the validity and reliability study demonstrate that the NHDC has relevance when measuring the construct of hand deformity and, that its use is accurate and reproducible. The analyses of construct validity and test–retest and inter-rater reliability suggests that the NHDC has the potential to guide decision making for upper limb interventions based on the biomechanical components of an individual's observed hand deformity. The clinical application of the NHDC is presented in the form of case studies in the next chapter (Chapter 7).

Chapter Seven

How can the Neurological Hand Deformity Classification be used in clinical practice? A case study approach

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7.1 Introduction

A case study is a practical form of enquiry that investigates a current phenomenon within its real-life context (Yin, 2014). Case study analysis should draw on the researcher's prior knowledge of the phenomenon while making use of relevant evidence (Rowley, 2002).

Accurate and clear case reports present information that may guide clinical research and inform clinical practice guidelines (Riley et al., 2017). In this chapter, case studies are used to illustrate how classifying hand deformity, with the Neurological Hand Deformity Classification (NHDC), can guide clinical decision making for choosing interventions. The CAse REport (CARE) guidelines (Riley et al., 2017) were used to structure this chapter to address the question: *How can the NHDC be used in clinical practice for children and youth with cerebral palsy?*

7.2 Method

7.2.1 Design

Descriptive case study methods were used to demonstrate how the NHDC can be embedded in decision making. The CARE guidelines (Riley et al., 2017) were adapted to provide a framework for these case studies. Adaptation of the guidelines was necessary to describe how to apply the NHDC in clinical practice, rather than describe the effect of a specific intervention. The body functions and structure domain of the ICF (World Health Organisation, 2001) was used as a framework to present clinical information of the individual cases. The outcome measures rating form (CanChild, 2004; Law, 1987) was used to structure the information about the clinical utility of the NHDC.

7.2.2 Participants

Children were eligible for selection as a case for this study if they were participants of one of two Australian multi-centre randomised controlled trials. The trials were investigating the use of rigid wrist hand orthoses for children with CP: (i) the Infant Wrist Hand Orthosis Trial (iWHOT); and (ii) the Minimising Impairment Trial (MIT) (Imms et al., 2016).

A total of 55 children participated in the iWHOT (n=21) and MIT (n=34) trials at PCH between August 2015 and September 2018. Participants were randomly allocated to a wrist hand orthosis group or a non-orthosis group following baseline assessment (Imms et al., 2016). Children were selected from the wrist hand orthosis group as detailed information of upper limb orthotic intervention was collected for participants in this group as part of the trials. This information included details of orthosis fabrication and photographs. Eleven children, aged

between zero and three years of age, were randomly allocated to the wrist hand orthosis intervention group in the iWHOT. Eighteen children, aged between five and 15 years of age, were allocated to the wrist hand orthosis intervention group in the MIT.

Two children were chosen as cases for this study as they represented the two different trials (participant 1 was in the iWHOT and participant 2 in the MIT) and had different severity of hand deformity. Participant 1 was chosen because their level of unilateral hand deformity represented one of the largest groups (21%; 32 out of a total of 127 children) of the children classified with the NHDC from Study 3. Participant 2 was chosen because their bilateral hand deformity represented the most severe wrist extension deformity of one hand, and wrist flexion deformity, as classified with the NHDC, on the other hand. A total of three limbs were classified in the two case studies. The participants' individual characteristics are presented in Table 7.1.

Table 7.1 Participant's characteristics

Characteristic	Participant 1	Participant 2
Gender	Male	Female
Age at baseline assessment	2 years 11 months	10 years 11 months
Topographical distribution	Unilateral	Bilateral
(mini) MACS	Level II	Level V
GMFCS	Level I	Level V
NHDC at baseline	Left: F2	Right: F5; Left E2

Note: MACS – Manual Ability Classification System (Eliasson et al., 2006); GMFCS – Gross Motor Function Classification System (Palisano et al., 2008); NHDC – Neurological Hand Deformity Classification (Garbellini & Wilton, 2017).

7.2.3 Classification tool

Appropriate tools must be selected by clinicians to attain the information they require to guide intervention in clinical practice (Ketalaar et al., 1998). The NHDC is an impairment-based classification system that provides a biomechanical approach to classify hand deformity (Garbellini & Wilton, 2017). It is a discriminatory classification (Kirshner & Guyatt, 1985) used to distinguish the differing levels of hand deformity between individuals with neurologically-based upper limb impairment. Classification of hand deformity is made according to the individual's observed wrist and hand movement. The NHDC was the tool of focus in the case studies presented. The purpose, psychometric properties and aspects of clinical utility of the NHDC are discussed further.

The purpose of the NHDC as described by Garbellini and Wilton (2017) is to:

- facilitate observation and analysis of the anatomical and biomechanical components of wrist and hand deformity in people with neurologically-based impairment.
- identify the primary factors causing the dynamic presentation of the deformity during active wrist and hand movement.
- provide a framework for therapists to consider intervention options based upon the dynamic pattern of movement observed.

The NHDC provides a structure to discriminate between different levels or severity of movement to classify varying presentations of hand deformity. Once the classification is made, the NHDC, presented in table form (Garbellini & Wilton, 2017; Wilton, 2013b), is designed to help identify the primary structures responsible for the deformity (Georgiades et al., 2014). Once hand deformity is classified and the structures causing the deformity are identified, individual intervention options can be considered.

The selection of a measurement tool should depend on evidence of the tool's validity and reliability specific to its purpose (Ketalaar et al., 1998). Construct validity and test–retest and inter-rater reliability of the NHDC has been established (Chapter 6 of this doctoral program). The NHDC has demonstrated capacity to discriminate the individual's presentation of hand deformity and classification decisions can be reliably reproduced. The NHDC also has a freely available website and manual online (Garbellini & Wilton, 2017) which provides a standard procedure for administration and classification.

The clinical utility of any classification or measurement tool must consider: format; clarity of instruction; qualification required to use the tool; time taken to complete measurement; and cost (CanChild, 2004; Law, 1987).

The clinical utility of the NHDC considering these elements is established as follows:

- Format – Observational classification requiring participation of the client.
- Clarity of instructions – Comprehensive written and diagrammatic instructions are outlined in the NHDC manual (Garbellini & Wilton, 2017).
- Qualifications – For use by clinicians working with clients with neurologically-based upper limb impairment, no formal training or certification required.
- Completion time – Administration 5–10 minutes, classification 5–10 minutes.
- Cost – Nil, freely available.

7.2.4 Administration and set up of the NHDC

The standard procedure to administer and set up the NHDC, as outlined in the NHDC manual and website (Garbellini & Wilton, 2017), was incorporated into the assessment protocol for the orthosis trials (Imms et al., 2016). The NHDC procedure included observation of three attempts of wrist and hand movement, with the most consistent movement used to classify the level of hand deformity. The maximum degree of wrist flexion observed at any stage of the movement determined the classification (Garbellini & Wilton, 2017). Video recording is recommended when classifying hand deformity with the NHDC. The video camera is placed a metre from the non-classified side of the participant (Garbellini & Wilton, 2017). Video recorded footage was available from each participants' NHDC. The footage was used to describe the administration and set up. Further details are provided for each participant in the following sections.

Participant 1

Participant 1 was a two year and 11-month-old boy who presented with a left hemiplegia. He had active reach and hand placement with his affected left upper limb. He was independently mobile and attending day-care at the time of baseline assessment. The participant sat on his parent's lap at an adjustable table set to the recommended height as described in the NHDC manual (Garbellini & Wilton, 2017). A soft toy, approximately the size of the participant's fist, was placed on the table in front of him. The participant's parent restrained his unaffected right upper limb and the participant was encouraged to approach, grasp and release the toy with his left hand. The NHDC was scored from a video recording of the participant's dynamic left wrist and hand movement.

Participant 2

Participant 2 was a ten year and 11-month-old girl who presented with involvement of all four limbs and a history of refractory epilepsy and cortical visual impairment. She had minimal to no active upper limb movement and was dependent for all her care needs. She was in year five at a supported education school at the time of baseline assessment. The participant was seated in a supported seating system on a manual wheelchair. Both hands of the participant were classified, so the video camera position was moved according to the side being classified. Due to the participant's inability to move towards the object, a ball, approximately the size of the participant's fist, was presented to the hand being classified. The ball was used to stimulate the dorsal and volar surfaces of the fingers to elicit any active movement. The NHDC was scored from the video recordings of the participant's wrist position and minimal amount of finger movement.

7.3 Clinical findings

The participants' NHDC levels constituted the clinical findings for each individual case presented. The classification of hand deformity using the NHDC was completed at the participant's baseline assessment for the orthosis trials (Imms et al., 2016). The level of hand deformity classified using the NHDC, along with a description of the level of deformity, and identification of the primary factors causing hand deformity for each participant is presented in the following sections.

7.3.1 Participant 1

Classification type: Participant 1 was classified with an NHDC level of ***F2*** for his left hand (see Figure 7.1). The participant had wrist flexion of greater than 20 degrees during active finger extension, had active finger flexion, and active wrist extension present during the observed movement.

Figure 7.1 F2 NHDC level for the left hand of Participant 1.

Primary structures causing deformity: Active wrist flexion of greater than 20 degrees, and wrist ulnar deviation during the observed movement were caused by increased muscle tone in the wrist flexors, predominantly Flexor Carpi Ulnaris (FCU). The movement into wrist flexion was also due to weakness of the wrist extensors, Extensor Carpi Radialis Longus (ECRL) and Extensor Carpi Radialis Brevis (ECRB). During the observed movement wrist flexion, metacarpophalangeal (MCP) hyperextension and interphalangeal (IP) joint flexion was caused by increased muscle tone or insufficient length of the finger flexors, Flexor Digitorum Superficialis (FDS) and Flexor Digitorum Profundus (FDP) when extending the fingers.

Associated thumb deformity: Left carpometacarpal (CMC) joint adduction and, MCP and IP joint hyperextension were present during the observed movement. The associated thumb deformity was driven by increased tone in the thumb adductor, Adductor Pollicis (AP). The flexed wrist position increased tension through the long thumb extensor, Extensor Pollicis Longus (EPL), causing hyperextension of the IP joint.

7.3.2 Participant 2

Classification type: Participant 2 was classified with an NHDC level of ***E2*** for her left hand (see Figure 7.2a) and an ***F5*** level for her right hand (see Figure 7.2b). On the left, the participant's wrist remained in wrist extension with no active wrist or finger movement. On the right, the participant's wrist remained in wrist flexion with no active wrist or finger movement.

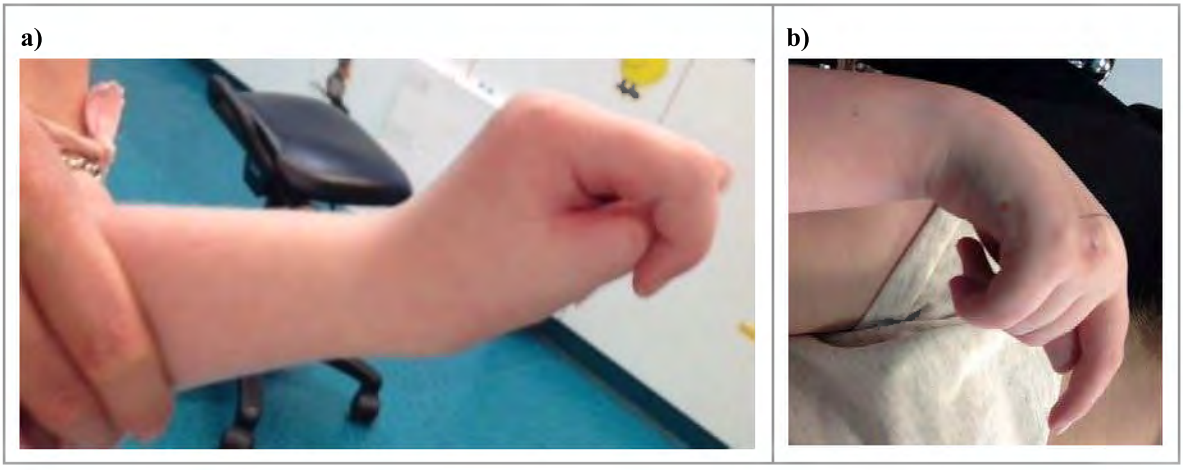

Figure 7.2 **NHDC levels for left and right hand of participant 2. (a) level E2 for the left hand, (b) level F5 for the right hand**

Primary structures causing deformity: On the left, the extended wrist position was driven by increased muscle tone or shortening of the extrinsic wrist extensors, ECRL, ECRB and Extensor Carpi Ulnaris (ECU). The participant's left forearm remained in pronation. Consideration of the observed forearm posture eliminated the reinforcing effect gravity would have added to the wrist extension deformity, if the forearm was in supination. The MCP joints were in 60 degrees of flexion, with the proximal interphalangeal (PIP) joints flexed to 70 degrees, and the distal interphalangeal (DIP) in 40 degrees of flexion. This observed posture was caused by increased muscle tone or shortening of the intrinsic muscles of the hand (causing MCP flexion) and shortening of the extrinsic finger flexors, FDS and FDP.

On the right, the flexed wrist posture was driven by increased muscle tone or shortening of the wrist and finger flexors Flexor Carpi Radialis (FCR), Palmaris Longus (PL), FCU, FDS and FDP. Consideration of the observed forearm position indicated the potential effect of gravity in reinforcing the wrist flexed posture of 50 degrees of flexion. An absence of active wrist extension was observed.

Associated thumb deformity: On the left, CMC joint adduction and MCP joint flexion with the thumb positioned between the index and middle fingers was observed. The left thumb posture was driven by increased muscle tone or shortening of the Flexor Pollicis Brevis (FPB) and AP intrinsic muscles of the thumb. On the right, CMC joint adduction with IP joint hyperextension was observed. The associated right thumb deformity was driven by increased tone in the thumb adductor, AP. The flexed wrist position increased tension through the long thumb extensor, EPL, causing hyperextension of the IP joint.

7.4 Diagnostic assessment

The NHDC provided structure to observe and classify the hand deformity of both cases presented above. Observation of the dynamic interplay of the participants' wrist and finger movement supported identification of the structures causing hand deformity which were further assessed.

The primary body structures causing hand deformity, as identified by use of the NHDC, were assessed for:

- Passive range of motion (PROM) using a goniometer
  - Assessment of end PROM of the wrist flexors was assessed with the fingers flexed
  - Assessment of end PROM of the finger flexors was assessed with the fingers extended
  - Assessment of end PROM of the wrist extensors was assessed with the fingers flexed
- Spasticity using the Modified Tardieu Scale (MTS), the initial resistance on rapid passive motion (R1) (Numanoğlu & Günel, 2012)
- Hypertonicity using the Modified Ashworth Scale (MAS) (Numanoğlu & Günel, 2012)

The outcome measures of PROM, MTS and MAS were part of the protocol within the orthosis trials. The range of wrist movement was referenced from -70 degrees (full flexion) to +80 degrees (full wrist extension), where 0 degrees indicated a neutral wrist position (Imms et al., 2016). The participants' assessment details are tabulated below.

7.4.1 Participant 1

Table 7.2 Assessment of the left hand body structures for Participant 1

	Diagnostic assessment		
Primary body structure causing deformity	**PROM (˚)**	**MTS (˚)**	**MAS**
Wrist flexors – FCU	80	-2	1+
Finger flexors – FDS/FDP	67	-60	3
Thumb adductor – AP	70	Not assessed	Not assessed

Note: PROM – End of passive wrist extension was measured with fingers flexed for wrist flexors and with fingers extended for finger flexors; MTS – Modified Tardieu Scale reports joint angle at initial resistance on rapid passive motion (R1); MAS – Modified Ashworth Scale; FCU – Flexor Carpi Ulnaris; FDS – Flexor Digitorum Superficialis; FDP – Flexor Digitorum Profundus; and AP – Adductor Pollicis.

A loss of passive motion of the finger flexors was indicated by the difference between end of passive motion for wrist extension with fingers flexed and with fingers extended. Spasticity was assessed in the wrist and finger flexors with the MTS. A combination of spasticity and loss

of passive motion of the finger flexors caused greater than 20 degrees of wrist flexion during active finger extension. This supported the NHDC classification of F2.

7.4.2 Participant 2

Table 7.3 Assessment of the left hand body structures for Participant 2

	Diagnostic assessment		
Primary body structure causing deformity	**PROM (°)**	**MTS (°)**	**MAS**
Wrist extensors – ECRL/ECRB/ECU	-10	No R1	2
Finger flexors – FDS/FDP	59	28	2
Intrinsic muscles – Lumbricales	0	Not assessed	Not assessed
Intrinsic muscles – FPB	0	Not assessed	Not assessed

Note: PROM – End of passive wrist flexion was measured with fingers flexed for wrist extensors and with fingers extended for finger flexors; MTS – Modified Tardieu Scale reports joint angle at initial resistance on rapid passive motion (R1); MAS – Modified Ashworth Scale; ECRL – Extensor Carpi Radialis Longus; ECRB – Extensor Carpi Radialis Brevis; ECU – Extensor Carpi Ulnaris; FDS – Flexor Digitorum Superficialis; FDP – Flexor Digitorum Profundus; and FPB – Flexor Pollicis Brevis.

Loss of passive wrist flexion supported the NHDC classification of E2, as the participant's left wrist remained in extension with minimal passive motion into wrist flexion available. The MCP joints of the fingers and thumb had no loss of passive motion of extension as there was no limitation in extension of these joints.

Table 7.4 Assessment of the right hand body structures for Participant 2

	Diagnostic assessment		
Primary body structure causing deformity	**PROM (°)**	**MTS (°)**	**MAS**
Wrist flexors – FCR/FCU/PL	66	34	1+
Finger flexors – FDS/FDP	-28	-40	2

Note: PROM – End of passive wrist extension was recorded with fingers flexed for wrist flexors and with fingers extended for finger flexors; MTS – Modified Tardieu Scale reports joint angle at initial resistance on rapid passive motion (R1); MAS – Modified Ashworth Scale; FCR – Flexor Carpi Radialis; FCU – Flexor Carpi Ulnaris; PL – Palmaris Longus; FDS – Flexor Digitorum Superficialis; and FDP – Flexor Digitorum Profundus.

Contracture in the finger flexors of the right hand was observed. There was a difference of 94 degrees between end of passive wrist extension with fingers flexed and with fingers extended. The persistent flexed wrist position and shortening of the finger flexors supported the NHDC F5 classification.

7.4.3 Additional diagnostic assessment

The NHDC was used to classify hand deformity and identify the body structures causing the deformity for both cases presented. Subsequently, those body structures were assessed and information provided about severity of impairment. This information forms one part of a more comprehensive upper limb assessment that quantifies the client's upper limb function and can be used to guide intervention. An example of the complexity of information required to assess hand movement and function, as included in the iWHOT and MIT orthosis studies (Imms et al., 2016), is represented within the ICF framework (World Health Organisation, 2001) in Figure 7.3.

In addition to the participants' body structure and function information, assessment of their capacity and performance for activity is also required to quantify upper extremity function and guide the selection of intervention options (Wallen & Stewart, 2016). In the context of upper extremity function: capacity describes the person's ability to reach, grasp, manipulate and release objects; and performance describes how they use their upper limb in everyday activity in their environment (World Health Organisation, 2001).

Most upper limb assessments provide a clinician with the opportunity to observe active range of motion and motor control during voluntary movement (Wallen & Stewart, 2016). The NHDC provides a system to observe, classify and consider structures causing hand deformity during available active movement and therefore formalises observational opportunities. Once assessment of a child's upper limb biomechanical presentation, capacity of upper limb function and actual functional performance has been completed, clinicians can then decide on how to best intervene.

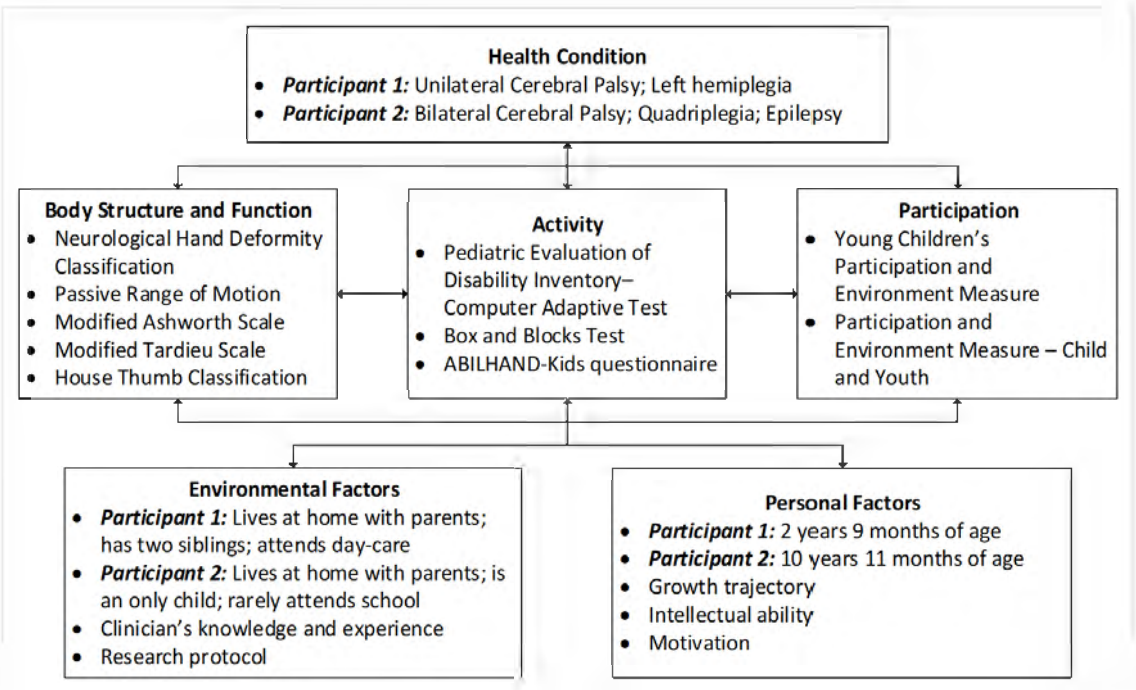

Figure 7.3 ICF representation of factors and assessments that could guide intervention

7.5 Therapeutic intervention

A clinician's decision making on how to intervene for children with CP is informed by the best available evidence (Novak et al., 2020), practice based knowledge, emotional intelligence and instinct (Unsworth & Baker, 2016). The focus of current upper limb intervention is to maximise the efficiency of the child's response to the environment and demands of the task, leading to changes in upper limb movement capabilities and task performance (Hoare & Greaves, 2017). As biomechanical presentation, capacity for upper limb function, functional performance and goals differ between individuals, so too does the choice of appropriate therapeutic intervention.

The level of hand deformity, classified with the NHDC, may guide the choice of upper limb intervention. For example, for children with a unilateral presentation, the level of hand deformity and movement impairment may inform the selection of constraint-induced movement therapy versus bimanual occupational therapy (Hoare & Greaves, 2017). Also, the level of hand deformity, capacity for movement and the child's performance of daily tasks should be considered when selecting goal-oriented training (Eliasson & Burtner, 2008). The level of hand deformity and identified structures causing the deformity, in combination with range of motion measures and spasticity measures, may influence the muscle selection for the use of Botulinum Toxin Type A (Fehlings et al., 2010) and inform the biomechanical application of an upper limb orthosis (Wilton, 2013b).

Study 1 of this doctoral program identified that biomechanical influences were not well reported in the literature as being considered in the rationale for upper limb orthosis prescription. The literature lacks sufficient description of the dynamic interplay of the wrist and fingers in action, and how orthosis prescription was guided by this. Prior to prescribing an upper limb orthosis it is essential to analyse the deforming forces generated by the wrist and finger musculature (Garbellini et al., 2017). Information on the direction, location and intensity of forces required to minimise or resolve the deformity with an orthosis can be determined from this analysis.

Garbellini et al. (2017) proposed recommendations for the continued use of upper limb orthotic intervention. These included: observation and analysis of hand deformity; clear documentation of the reason for orthosis prescription; and using consistent terminology to describe the purpose of the orthosis. The use of classification of hand deformity, the reason for orthosis prescription, purpose and description of the orthosis for each participant will now be presented in further detail.

7.5.1 Participant 1 – Left upper limb orthotic intervention

Analysis of left hand deformity

Participant 1 received an F2 classification for his left hand using the NHDC. The participant's left wrist and finger flexors were identified as the primary structures (Wilton, 2013b) causing the deforming forces (red arrows, Figure 7.4). The positional deformity was corrected with the application of reciprocal forces (Van Lede & Van Veldhoven, 1998) (green arrows), around the axis of the left wrist joint, with the application of a volar wrist orthosis (Figure 7.4). It is of note that the wrist and finger positions are greater than the angle of initial catch assessed with the MTS (Numanoğlu & Günel, 2012), potentially providing stretch to the wrist and finger flexors (refer to Table 7.2).

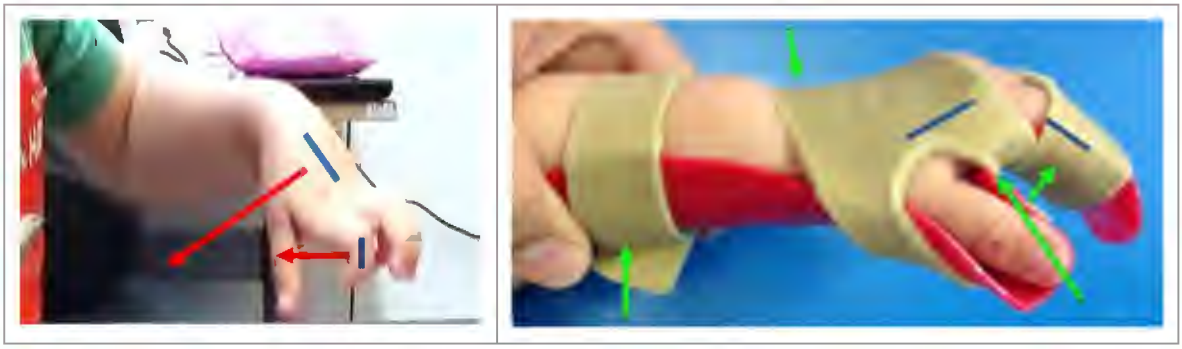

Figure 7.4 **Hand deforming forces without orthosis and reciprocal resolving forces with orthosis for the left hand**

Reason for left upper limb orthosis prescription

An orthosis was prescribed to maintain optimal postural alignment of the wrist and fingers to:

i. prevent complications associated with muscle shortenting due to abnormal posturing, and
ii. maximise potential for functional use of the hand (Imms et al., 2016).

Purpose of the orthosis

The primary purpose of the orthosis was to apply forces to extend the wrist and fingers in a position opposite to the deforming forces idenitified using the NHDC.

Description of the orthosis

The orthosis prescribed was a mobilising wrist and hand orthosis (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992) of a volar design. A two millimetre thick thermoplastic material was used (micro-perforated Orfit). The orthosis was held in place by VELCRO® straps (Velfoam).

Timing and dosage of wear

Orthosis prescription was for night wear for a period of at least six to eight hours during sleep. Participant 1 wore the left orthosis at night. This allowed the participant to have his left hand free to actively use as an assisting hand during the day. The participant wore the orthosis for the three-year period of the iWHOT. The orthosis was serially adjusted or re-made during this period to sustain mobilising forces on the deforming structures. Since completing the iWHOT the participant's parents have chosen to continue orthotic intervention with a left mobilising wrist and hand orthosis worn at night. The duration of orthosis wear at night currently totals four years.

7.5.2 Participant 2 – Bilateral upper limb orthotic intervention

Analysis of left hand deformity

Participant 2 received an E2 classification for her left hand using the NHDC. The participant's left wrist extensors, extrinsic finger flexors and intrinsic hand muscles were identified as the primary structures (Wilton, 2013b) causing the deforming forces (red arrows, Figure 7.5) of wrist extension, MCP flexion and finger flexion. The positional deformity was corrected through the application of forces (green arrows, Figure 7.5) with a wrist and hand orthosis. It is noted that the wrist is positioned in a neutral position, only 10 degrees less than the amount of available passive wrist flexion.

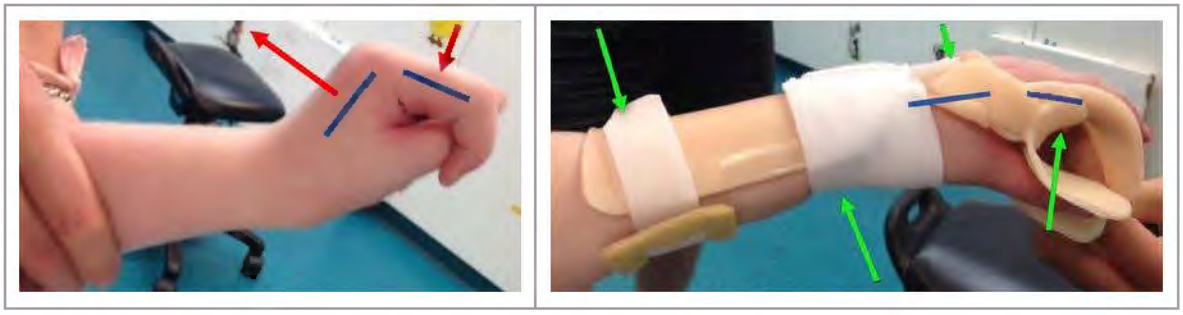

Figure 7.5 Hand deforming forces without orthosis and resolving forces with orthosis for the left hand.

Analysis of right hand deformity

Participant 2 received an F5 classification for her right hand using the NHDC. The participant's right wrist and finger flexors were identified as the primary structures (Wilton, 2013b) causing the deforming forces (red arrows, Figure 7.6) of wrist and finger flexion. The positional deformity was corrected through the application of forces (green arrows, Figure 7.6) with a wrist and hand orthosis.

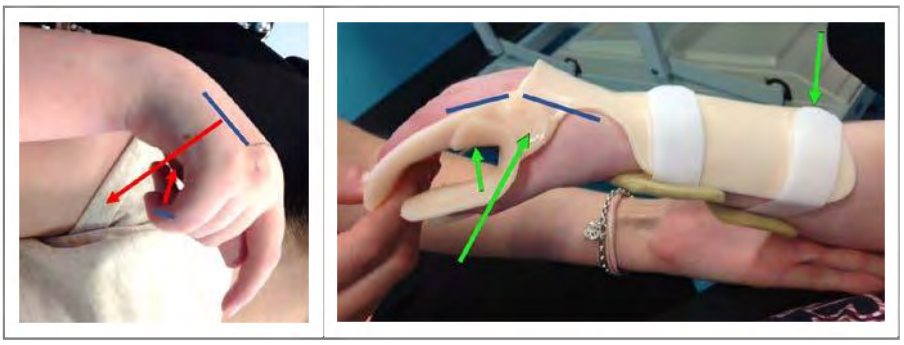

Figure 7.6 **Hand deforming forces without orthosis and resolving forces with orthosis for the right hand.**

Reason for upper limb orthosis prescription

Orthoses were prescribed to:

i. prevent further loss of passive range of motion at the wrist and fingers;
ii. prevent complications of pain and poor hygiene associated with muscle shortenting (Imms et al., 2016).

Purpose of the orthoses for left and right hands

The primary purpose of the orthosis for the left hand was to apply forces to flex the wrist and extend the MCP joints of the fingers in a position opposite to the deforming forces identified using the NHDC. The primary purpose of the orthosis for the right hand was to apply forces to extend the wrist and fingers in a position opposite to the deforming forces idenitified using the NHDC.

Description of the orthoses

The orthoses prescribed were mobilising wrist and hand orthoses (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992) of a dorsal/volar design. A 3.2 millimetre thick thermoplastic material was used (classic soft non-perforated Orfit). The orthosis was held in place by VELCRO® straps (Velcro and Velstretch).

Timing and dosage of wear

Orthosis prescription was for night wear for a period of at least six to eight hours during sleep. Participant 2 wore both the left and right orthoses at night. Given the participant had minimal active movement of her hands, sometimes her parents chose to don the orthoses for periods during the day. They did this if they felt her hands were particularly stiff, or that her muscle tone had increased due to seizure activity. The participant wore the orthoses during the

18-month period of the MIT study, until the study's cessation. The orthosis was serially adjusted or re-made during this period to sustain mobilising forces on the deforming structures. The participant's parents have chosen continued left and right mobilising wrist and hand orthoses wear at night. The duration of bilateral orthosis wear at night currently totals three and a half years. The participant did not have orthoses prior to her participation in the MIT.

Additionally, identification of the participant's left extension hand deformity was used to educate the parents and caregivers regarding the best way to open the participant's hand for cleaning and hygiene. Application of passive wrist flexion forces counteracted the extension forces. A more neutral wrist position decreased the amount of flexion force through the MCP joints which allowed the parents and carers to extend the MCP joints to clean the palmar surface of the hand.

7.6 Discussion

Assessment of neurologically-based upper limb impairment and choosing appropriate interventions is challenging. Understanding movement limitations of the wrist and finger musculature of people with neurologically-based upper limb impairment is critical to inform the reason for, and choice of, upper limb intervention (Garbellini et al., 2017). In the case studies presented, use of the NHDC provided structure to: observe and classify the dynamic movement of the wrist and hand musculature; identify the dynamic forces causing deformity; and guide decision making for intervention strategies. For clinicians new to working in the clinical area of CP, or managing neurologically-based upper limb conditions, the NHDC could provide a starting point to guide information-gathering and decision making.

The aim of the case studies was to provide a clinical context for the use of the NHDC, not to present all possible upper limb assessments and interventions. It is not proposed the NHDC be used in isolation as it is an impairment-based classification tool. A more complete clinical picture of the child through assessment of their unimanual capacity and bimanual performance would also guide the selection of upper limb intervention. For example, unilateral capacity of the upper limb can be assessed with the Melbourne Assessment 2 (Randall, Johnson, et al., 2013), or the Quality of Upper Extremity Skills Test (QUEST) (DeMatteo et al., 1993). Bimanual performance of the upper limbs can be assessed with the Assisting Hand Assessment (Krumlinde-Sundholm et al., 2007) or the Both Hands Assessment (Elvrum et al., 2018). Across the ICF domains that guide decision making, other relevant upper limb measures and environmental and personal factors must also be considered.

In a clinical setting, where time is limited, it is not possible for clinicians to use every upper limb measurement tool to assess the person's upper limb. Time efficient measures with established validity, reliability and clinical utility are required. A clinician's experience and work setting also influences the choice of measurement tool. With the recent clinical focus on activity and participation, tools to measure upper limb capacity and performance have been integrated into clinical practice. For example, the AHA is now regularly used to assess bimanual performance of children with unilateral presentation. Biomechanical measures of body structure and function are still used in practice, but what is measured can vary depending on the experience of the clinician. For example, assessment of range of motion for the upper limb may be completed for the whole upper limb or targeted in more detail for one or two joints. The NHDC can guide which body structures to assess in more detail based on the classification of hand deformity, and therefore potentially save time.

This chapter has presented two case studies to illustrate how the NHDC was used to guide the prescription of upper limb orthotic intervention in two multicentre trials. Analysis of the forces causing hand deformity allowed identification of the forces required to resolve the deformity. The resolution of forces was achieved with the application of a wrist and hand orthosis. It is noted that deforming forces are dynamic, even if the child's active range of motion at the joints impacted has decreased over time. If the deformity is fixed, then there is no passive range of motion available at the affected joints. This supports the recommendation that the NHDC not be used in isolation. It also highlights the importance of clinicians using standard terminology when describing the orthosis prescribed, and including whether the orthosis aims to immobilise or mobilise tissues (Australian Hand Therapy Association, 2012; Garbellini et al., 2017; American Society of Hand Therapists, 1992; Wilton, 2013a).

In the cases presented the orthoses were prescribed to mobilise tissues to maintain or gain passive range of motion. They were not prescribed to rest the joints. There was no pathology, for either case study, to suggest the joints needed to be immobilised or rested. A static orthosis was prescribed to create mobilising forces to correct the dynamic deformity. The static orthosis was serially adjusted to sustain the mobilising forces over time. The orthoses were worn at night to allow the participants to have their hands free for use during the day. Participant 1 was able to use his left hand to hold objects and stabilise objects with grip. Participant 2 was able to have her hands massaged and touched for care and cleaning.

Both participants wore the orthoses for the time and duration prescribed. Adherence and tolerance of the upper limb orthotic intervention was not a concern for either participant. The parents of both participants understood the reasoning for and were supportive of the intervention.

Neither participant reported sleep disturbance while wearing the orthoses. Any concerns of orthosis fit, or discomfort were addressed with modification of the orthosis. Modification was required to reduce pressure and shearing forces and maintain advantageous application of the resolving forces (opposite to the deforming forces) of the orthosis due to growth. Application of the NHDC enabled identification and understanding of the dynamic forces essential to the process of orthosis modification.

A strength of using a case study design is it allows the structured presentation of information within a clinical context. The application of the NHDC followed the same format and yielded similar information to guide decision making, even though the age of the participants and their severity of impairment differed. Conversely, the selection of cases may be seen as a potential limitation. The two participants were purposively selected out of a possible 29 children. It is unknown whether randomly selecting participants would have provided a different focus for the information presented in the case studies.

7.7 Chapter Conclusion

These case studies highlight that managing neurologically-based upper limb impairment is complex. Classification of hand deformity using the NHDC can guide further assessment of the primary structures causing deformity and connect analysis of body structure and function directly to intervention decisions, in this case orthosis prescription. Identification of the forces causing deformity from observation and classification of the wrist and hand muscles in action can guide how to counteract those forces with an appropriate intervention. Many factors must be considered to appropriately prescribe and fabricate upper limb orthoses.

In this chapter, a description of how the NHDC can be used in clinical practice has been presented in the form of two case studies. In each case study the NHDC was used to guide assessment at the body structure and function level of the ICF. The NHDC should form part of a comprehensive upper limb assessment that includes activity and participation measures, in the context of personal and environmental factors to inform best practice intervention.

Chapter Eight

Extended discussion and conclusion

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8.1 Introduction

The available research evidence to support upper limb orthosis prescription for children with CP has been graded as weak, and resulted in the recommendation that clinicians should 'probably do it' (Novak et al., 2020). This lack of research evidence has led to uncertainty when considering upper limb orthotic intervention. Uncertainty also comes from studies reporting that less than half of the children would wear an upper limb orthosis as prescribed (Andersson et al., 2019; Jackman et al., 2018; Russo et al., 2009). Even with the current state of the literature, some experienced clinicians are yet to relinquish orthosis prescription as an intervention option for children with CP. This may be due to the improvement clinicians observe with the use of orthotic intervention for individual clients, or their belief that the published evidence does not address the complexity of the intervention (Garbellini et al., 2017). The questions remain as to: why some children benefit from upper limb orthotic intervention; why some choose to wear prescribed orthoses and others do not; and whether clinicians are prescribing the right orthosis, for the right child, at the right time.

This final chapter synthesises the evidence from this doctoral program. The lack of upper limb orthotic intervention practice guidelines for this complex intervention are discussed in greater detail. Recommendations for clinical practice are made based on these findings. In addition, implications for future research and the limitations and strengths of this doctoral program are presented.

8.2 Summary of doctoral program

This doctoral program comprises four studies:

1. A systematic review (published, Study 1) following the PRISMA guidelines (Moher et al., 2009). Study 1 explored the questions:
    - What are the reasons for upper limb orthosis prescription for children with CP?
    - Is there a documented link between the reason for an upper limb orthosis according to its intended effect and outcome measures used to determine effect?
    - Is standard nomenclature used in the literature to describe upper limb orthoses?

2. A qualitative cross-sectional study using Q methodology (published, Study 2), which established an answer to:
    - What guides the decision making of occupational therapists' in Australia when prescribing upper limb orthoses?

3. A measurement study using quantitative methods (Study 3), which addressed the question:
    - What are the psychometric properties of the NHDC, and can it be used with confidence in clinical practice to classify hand deformity?
4. Case studies (Study 4) following the CARE guidelines (Riley et al., 2017). Study 4 formed part of the doctoral work to answer the question:
    - How can the NHDC be applied in clinical practice?

The completed work and findings of the doctoral program, including answers to the questions above, are outlined in Figure 8.1.

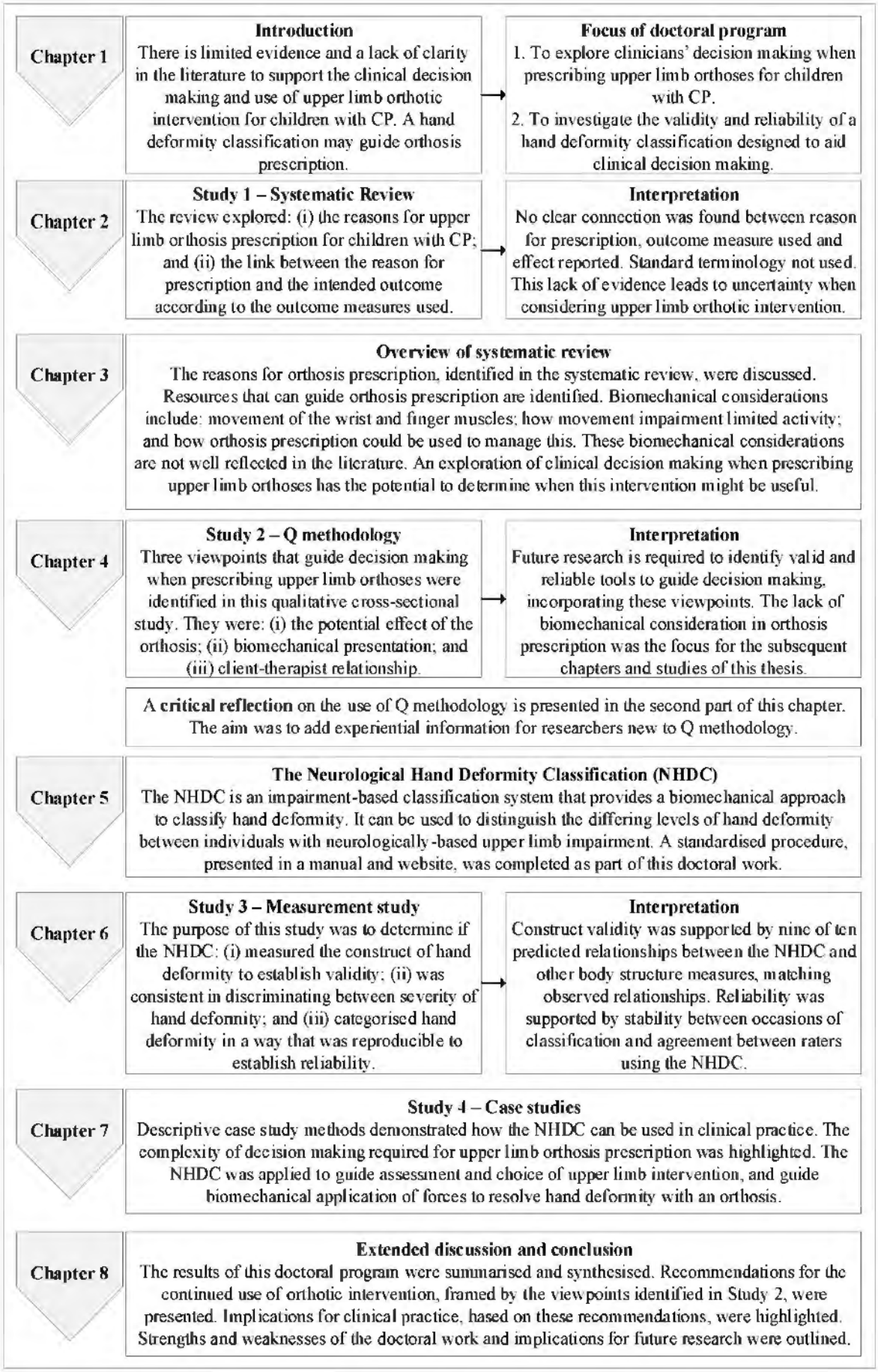


Figure 8.1 A summary of this doctoral program

8.3 The lack of upper limb orthotic intervention practice guidelines

Three viewpoints emerged from the Q methodology study (Study 2) when Australian occupational therapists were asked, "What guides your decision making when prescribing upper limb orthoses for children with CP?" The viewpoints included: client-therapist relationship; biomechanical presentation of the client; and potential effect of the orthosis (Garbellini et al., 2019). These viewpoints reflect clinical practice, and they are here used to frame further discussion of the lack of guidelines for upper limb orthosis prescription found throughout this doctoral work. Recommendations for clinical practice are presented following the discussion of each viewpoint.

8.3.1 Client-Therapist relationship

Upper limb orthotic intervention must be considered in the context of the client and his/her environment. This approach was not well reported in the literature; the goals of orthotic intervention for the children were not detailed (Garbellini et al., 2017). This is concerning given that the goals of the client was the primary regard of occupational therapists when rating what guides their decision making for orthosis prescription (Garbellini et al., 2019). This raises the question of whether the goals of orthotic intervention are not detailed in clinical practice and therefore poorly reflected in the research literature.

The challenge for clinicians, particularly with orthosis prescription, is to determine if orthotic intervention is the best intervention for the individual child. The relationship between the client and clinician is vital to ensure that realistic goals are set and that they are achievable (Copley et al.,2008). To determine if orthotic intervention is an appropriate intervention, clinicians must: consider the child's level of impairment and unimanual capacity; integrate how this affects bimanual or activity performance; understand what the child wants to do; and account for and accommodate influencing factors in the child's environment.

Recommendations for clinical practice

It is recommended that upper limb orthosis prescription be based on assessment findings and the occupational needs of the individual child, within the context of his/her environment.

Practice recommendations include:

- Clear documentation of the goal of orthotic intervention
- Application of an upper limb orthosis based on the occupational needs of the individual child
    - What the child wants to do
    - How important it is to the child
    - What the child can do (capacity)
    - What the child does (performance)
- Consideration of whether prescription of an orthosis will meet the goal of the upper limb intervention

8.3.2 Biomechanical presentation

Biomechanical considerations of the child's presentation, and the application of orthoses were not well reflected in the literature (Garbellini et al., 2017). Some of the included studies assessed range of motion, muscle tone and spasticity, however did not link these findings with the impact on movement or hand deformity. This disconnect was evident by the lack of description of the child's wrist and hand movement and severity of impairment. It is difficult to resolve impaired movement, or correct deformity without understanding the forces that create them, or the forces required to resolve them. There has also been a shift in clinical practice from a focus on body functions and structures to activity and participation (Lannin et al., 2017). Even with this shift in practice, attention on body functions and structures is still required to consider how they can potentially impact the other domains of activity and participation.

Recommendations for clinical practice

It is recommended that upper limb orthosis prescription be based on the biomechanical presentation of the individual child and the biomechanical effect of the prescribed orthosis. Practice recommendations include:

- Observation and classification of hand deformity using the NHDC
- Identification of the dynamic forces causing the hand deformity using the NHDC
- Consideration of the biomechanical forces required by an orthosis to resolve the deformity

8.3.3 Potential effect of the orthosis

The limited research evidence on the effect of upper limb orthotic intervention (Jackman et al., 2014b; Novak et al., 2020) provides little guidance for clinical practice. Closer inspection of the research evidence in the systematic review by Garbellini et al. (2017) found a lack of description, or absence of reasons behind orthosis prescription. The purpose of the orthoses was not clearly described. Additionally, there was often no clear link between the stated reason for the orthosis, the way the results were measured or the stated outcome of the intervention (Garbellini et al., 2017). Without clarity about the reason for orthosis prescription and the purpose of the orthosis, or link with the outcome measure used, the ability to use the research evidence to inform practice guidelines is limited (Garbellini et al., 2017).

Variability in the terminology used in clinical practice might explain some of the difficulty in researching, and the resultant lack of evidence of the effect of the intervention. The effect of prescribed upper limb orthoses is determined by: the reason for prescription; the purpose of the orthosis; and the orthosis and its wear. The development of practice guidelines is challenging without clear and common terminology to detail and describe these determinants of the effect of orthosis prescription. The lack of clarity and variability of terminology for these determinants is discussed further in the following section.

Reason for orthosis prescription

The reason for orthosis prescription has been termed a ‘clinical aim’ by Copley and Kuipers (2014b) and ‘clinical indication’ by Wilton (2003, 2013). The reasons for upper limb orthosis prescription have been described as: managing the positive or negative features of upper motor neuron syndrome; minimising the potential for secondary musculoskeletal change; resolving contracture; maintaining integrity of tissues; managing pain; and improving function (Copley & Kuipers, 2014b; Wilton, 2003, 2013). There was a lack of specificity in the reasons for orthosis prescription in the research literature (Garbellini et al., 2017). This lack of specificity provides little information for the development of practice guidelines and causes concern for clinical practice, as the reasons for orthosis prescription may apply to almost all children with CP. If orthosis prescription proceeds without a clear description of the purpose of the orthosis and when it is to be used, some children may be prescribed an orthosis that does not meet their clinical needs.

Purpose of the orthosis

The purpose of an orthosis should be based on its key function (Lannin & Ada, 2011). The Australian Hand Therapy Association (2012) and American Society of Hand Therapists (1992)

classify the purpose of orthoses as immobilising, mobilising, restricting or torque transmitting. There was little differentiation between the reason for orthosis prescription and the purpose of the orthosis in the CP research literature. No studies referred to the orthosis purpose based on its key function (Garbellini et al., 2017). Terminology used in textbooks and in the research literature to describe the reason for the orthosis, its purpose and description seem to be interchangeable. These terms include functional and non-functional orthoses and resting orthoses (Copley & Kuipers, 2014b; Jackman et al., 2019). Even though clinicians may understand the use of these terms, using them in clinical practice and in research may cause uncertainty. For example, a serially adjusted static orthosis can be used to mobilise tissues to gain passive range of motion. Using common terms, this orthosis would be referred to as a resting orthosis, a term that does not provide a clear description of the purpose of the orthosis which may not be 'to rest'. Communication between clinicians is difficult if inconsistent terminology is used to describe the same thing.

The orthosis and its wear

Orthoses have been described in many ways. In the studies included in the systematic review by Garbellini et al. (2017) orthoses were named (see Table 2.5 in Chapter 2):

- Cortical thumb orthosis
- Ultraflex orthosis
- Wrist extension thumb abduction (WETA) orthosis
- Volar wrist splint
- Dynamic spiral wrist hand splint
- Dorsal wrist splint
- Children's wrist and thumb support
- Static night splint
- Wrist cock-up orthosis

The names of some these orthoses give an idea as to the joints on which they were applied, others do not. Standard nomenclature to describe upper limb orthoses is not evident in the literature (Garbellini et al., 2017).

The timing and dosage of orthosis wear is also variable in the research literature (Garbellini et al., 2017). Some studies investigated the immediate effect of a wrist orthosis (Jackman et al., 2019; Louwers et al., 2011). The orthoses were only worn during the assessment. No effect from length of orthosis wear was determined. Other studies did not report any timing or duration of orthosis wear (Barroso et al., 2011; Burtner et al., 2008; Kanellopoulos et al., 2009). The lack of evidence of timing and duration of orthosis wear provides little information to guide clinical practice.

In clinical practice the timing and duration of orthosis wear is critical to achieve the desired effect of the intervention. Children are unlikely to wear an orthosis if it: is uncomfortable; restricts too much movement during function; makes things more difficult for them to do; stops them from using their hand in a way that is more efficient for them; or interferes with sleep. The biomechanical presentation of the child and considered action of the forces generated from an orthosis are critical to minimise the impact of these hindrances to orthosis acceptance and wear. Clinicians should consider if a lack of wear is attributable to: inappropriate prescription; a lack of clarity in the reason or purpose of the orthosis; or the lack of connection between the biomechanical forces applied by an orthosis and the biomechanical presentation of the child.

Recommendations for clinical practice

It is recommended that upper limb orthosis prescription include:

- Clear documentation of the reason for orthosis prescription
- A description of the purpose of the orthosis using the terminology of an orthosis classification schedule (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992)
- Use of an anatomical focus when describing the orthosis (Australian Hand Therapy Association, 2012; American Society of Hand Therapists, 1992), for example ‘wrist and hand orthosis’
- Application of outcome measures that are consistent with the reason for the orthosis when measuring the effect of the intervention
- Clear documentation of when and for how long the orthosis should be worn

8.4 Implications for clinical practice

The complexity of upper limb orthosis prescription can be seen in the many factors that require consideration for the use of this intervention. The clinical practice of orthosis prescription should be guided by the collective consideration of:

- Client-therapist factors
- Biomechanical factors
- Orthosis factors

Recommendations for clinical practice within each of these factors has been outlined above. These factors are not mutually exclusive. For example, the decision to prescribe an orthosis should not be determined from the child’s biomechanical presentation alone. A summary of the

practice recommendations above and the considerations to guide upper limb orthosis prescription are presented in Figure 8.2.

One of the main gaps identified throughout this doctoral work, was a lack of connection between the child's biomechanical presentation and the reason and purpose of upper limb orthosis prescription. Establishment of construct validity and test–retest and inter-rater reliability of the NHDC (Study 3), and the development of an NHDC procedural manual and website (Garbellini & Wilton, 2017), provide a means to address this gap. As part of the development process, resources were made to disseminate information for the implementation of the NHDC in clinical practice (Graham et al., 2006). In addition to the NHDC manual (Chapter 5), the resources include: NHDC quick guide and NHDC orthosis considerations (Appendix E).

Embedding the recommendations generated from the findings of this doctoral work and use of the NHDC in clinical practice will be challenging and take time. The challenge will be engaging clinicians to adapt clinical practice. Even though engagement with clinicians as the primary stakeholders is paramount (Graham et al., 2006), there needs to be recognition of the need for change, and acceptance that change is required. There is potential to engage with clinicians through the development of clinical workshops on upper limb orthotic intervention and to disseminate information through presentations at relevant scientific conferences. The development of practice guidelines and knowledge creation needs to be a partnership between consumers, clinicians and researchers (Graham et al., 2006, McKenzie & Hanley, 2012).

Figure 8.2 **Considerations to guide upper limb orthosis prescription**

8.5 Implications for research

The primary challenge for future research into upper limb orthotic intervention is to determine the effect of the intervention. A multi-layered approach including collaboration between clients, clinicians and researchers is required to develop research that will explore all aspects of the intervention (Graham et al., 2006; Grimshaw et al., 2012; McKenzie & Hanley, 2012). Elements to consider for future research, based on the findings of this doctoral work, are presented within the research cycle as described in the Framework on Consumer and Community Participation in Health and Medical Research (McKenzie & Hanley, 2012). The stages of the research cycle include: (i) deciding what to research; (ii) deciding how to do it; (iii) doing it; (iv) letting people know the results; and (v) knowing what to research next (McKenzie & Hanley, 2012).

8.5.1 Deciding what to research

The findings of this doctoral work have shown the variability of use of upper limb orthotic intervention (Garbellini et al., 2017). This variability in use of terminology, implementation and outcome measurement has resulted in difficulty determining the effect of the intervention. Collaboration between consumers, clinicians and researchers is required to develop research priorities for exploration of the efficacy of upper limb orthotic intervention. A multi-level approach to developing research questions is required, considering: the client's previous experience with orthotic intervention; the reasons clients have chosen not to wear a prescribed upper limb orthosis; current use of upper limb orthoses in clinical practice; use of practice guidelines (as per the considerations for orthosis prescription in Figure 8.2); and previous research methodology used. Clear research questions are required to enable selection of appropriate study methods.

8.5.2 Deciding how to do the research

A lack of specificity in what guided the prescription of upper limb orthoses for children with CP was evident in the research literature (Garbellini et al., 2017). Previous studies created homogenous groups based on their topographical presentation, for example unilateral spastic CP (Barroso et al., 2011; Louwers et al., 2011; Ozer et al., 2006). These studies did not describe the variability of each child's clinical presentation or movement limitation and how this guided orthosis prescription.

Deciding on how to explore the effect of upper limb orthotic intervention will need collaborative consideration of:

- *Who to involve in the research team* – It is recommended that consumers are involved at all stages of the research cycle, especially this stage of deciding how to do the research. The consumer's experience with orthotic intervention could improve the choice of study design and study population (the right orthosis for the right person at the right time). Engaging clinicians who work with children with CP and service provider managers will be essential to understand current clinical practice, and to implement change (if required) based on research findings.
- *Who to include as participants in the study* – Participants need to be chosen on the clinical indication or occupational need for orthotic intervention. Stratification based on NHDC level, presence or absence of active wrist extension, or Bimanual Fine Motor Function Classification level might direct selection of participants.
- *Study design* – Experimental methods such as single subject designs may be considered as an alternative to RCTs. Singe subject designs account for the heterogeneity or variability seen in children with CP and the complexity of clinical practice (Copley & Kuipers, 2014a; Portney & Watkins, 2015).
- *The intervention and its purpose* – Clarity is required in describing the reason for the intervention and purpose of the orthosis (Garbellini et al., 2017). An orthosis designed to immobilise the thumb in opposition and improve its position for grasp, may only be beneficial during targeted activities. The benefit of wearing the orthosis for other activities may be diminished if the position in which the orthosis places the thumb limits capacity to complete the activity. An orthosis worn at night to maintain muscle length may not improve the child's movement or functional ability during the day.
- *Timing and duration of the intervention* – The timing and duration of orthosis wear will impact the outcome of the intervention. Wearing instructions must be clear and match the intended purpose of the orthosis. Consideration of longitudinal research to explore the long-term effect of orthotic intervention is required. This is particularly pertinent when a main reason for orthosis use is the maintenance of muscle length and prevention of contractures.
- *What outcome measures to use* – The outcome measures used must match the intended reason(s) for orthosis prescription.

8.5.3 Doing the research

The practical aspects of doing upper limb orthotic research requires:

- Consideration of consumer engagement to promote and support recruitment
- A clear study protocol
- Training of assessors to ensure reliability in assessment
- Training of clinicians to ensure consistency in orthosis prescription
- Consideration of the child's biomechanical presentation in prescribing the orthosis (Garbellini et al., 2019)
- Experienced clinicians to fabricate the upper limb orthoses and monitor the intervention
- Training clinicians with less experience in orthosis fabrication to ensure fidelity of orthotic intervention
- Adequate time, expertise and resources to provide the intervention and collect data
- Data monitoring to check the reliability of the data
- Communicating the aim of the study, the clinical reason for an upper limb orthosis and what to expect during the study to the participants

8.5.4 Letting people know the results

Grimshaw et al. (2012) reported that considering what knowledge to share, to whom and by whom is essential to translate research into practice and policy. McKenzie and Hanley (2012) recommend keeping consumers and research participants involved throughout the research process. Collaborative consideration by consumers, clinicians, researchers and service provider managers to determine how the results are disseminated and implemented is required before the data and results are interpreted.

8.5.5 Knowing what to research next

An ongoing relationship between consumers, clinicians and researchers, using an integrated knowledge translation approach, can support the direction of future research (Gagliardi et al., 2016; Kothari & Wathen, 2012). Collaboration of these main stakeholders could ensure that research reflects clinical practice and that the research knowledge can be embedded in clinical practice (Graham et al., 2006; McKenzie & Hanley, 2012).

8.6 Limitations and strengths

The specific limitations and strengths of each study have been discussed within each respective chapter. The selection of Australian occupational therapists for the Q methodology study (Study 2) may reflect the bias of clinicians selected from a developed country. Generalisation of the results from this study should be made with caution. Future studies on what guides clinicians in the prescription of upper limb orthoses for children with CP could include a broader cross-section of clinicians from a range of cultural, economic and health-service backgrounds.

An additional limitation was seen with the sample size achieved for the NHDC test–retest component of Study 3. The sample achieved (n=36) was approximately one-third of the sample recommended by the COSMIN guidelines (Mokkink et al., 2019) to achieve a 'very good' rating. Access to participants to collect test–retest data was limited to the Western Australian site. Time-constraints and costs were too prohibitive for this data to be collected at other sites. The collection of the test–retest data was onerous as participants and their families needed to attend a second data collection point within a two-week period. If further testing of the psychometric properties of the NHDC occur, consideration of the sample size required and how the data will be collected is needed.

This doctoral work was embedded in two multi-centre national trials (Imms et al., 2016). This connection to the larger trials is seen as both a limitation and strength. Adherence to the study protocols (Imms et al., 2016), dictated the nature and timing of data collection. If the child was distressed, the focus of the assessment was collection of the primary outcome measure (end of passive range of wrist extension with fingers extended) rather than completion of other measures of the trial. A focus of this doctoral program was hand deformity classification with the NHDC. As a result, the NHDC data were not collected at all conducted assessments, limiting sample size.

Conversely, given the longitudinal nature of the orthosis trials and the part-time nature of this doctoral program, sequential knowledge gained at each stage of work was used to drive the next stage of research. Awareness of the trials within the therapy community across Australia potentially increased the participation of occupational therapists in Study 2. Access to data from multiple sites, rather than one site alone, increased the potential sample sizes for testing the psychometric properties of the NHDC (Study 3). The larger investigatory group responsible for the trials provided encouragement and support for the completion of this doctoral work.

Additional strengths to the findings of this doctoral program include the rigor of the studies by following: the PRISMA guidelines (Moher et al., 2009) for the systematic review (Study 1); Q methodology procedure (Watts & Stenner, 2012) for the qualitative cross-sectional study (Study 2); the COSMIN guidelines (Mokkink et al., 2019) for testing the psychometric properties of the NHDC (Study 3); and the CARE guidelines (Riley et al., 2017) for reporting of the case studies (Study 4).

8.7 Thesis Conclusions

The following conclusions may be drawn from the studies reported in this doctoral program:

1. Upper limb orthotic intervention for children with CP is complex.
2. Consistency in clinical practice is required. This includes consideration of the: goals of the client; client-therapist relationship; biomechanical presentation of the client and the effect of the orthosis.
3. The NHDC can be used with confidence to classify hand deformity, has stability between occasions of classification, and established agreement between raters. Case studies have shown how the NHDC can be used in clinical practice to guide upper limb assessment and choice of upper limb intervention.
4. Orthosis prescription must be tailored to the individual. The prescription of an orthosis is unlikely to be suitable for all children with neurologically-based upper limb impairment.
5. Collaboration between clients, clinicians, researchers and service provider managers is required to develop future research and practice guidelines.
6. The effect of the intervention and which children would most benefit from intervention needs to be determined.

References

NOTE: This reference list contains the citations for all chapters except for Chapter 5 where the references are contained within.

American Society of Hand Therapists. (1992). *Splint Classification System*. American Society of Hand Therapists.

Andersson, G., Renström, B., Blaszczyk, I., & Domellöf, E. (2019). Upper-extremity Spasticity-reducing Treatment in Adjunct to Movement Training and Orthoses in Children with Cerebral Palsy at Gross Motor Function-and Manual Ability Classification System Levels IV-V: A Descriptive Study. *Developmental Neurorehabilitation*, 1-10. https://doi.org/10.1080/17518423.2019.1655677

Atchley, W. R. (2007). *Introduction to principal components analysis*. Retrieved 18/02/2018 from ftp://statgen.ncsu.edu/pub/thorne/molevoclass/AtchleyOct19.pdf

Arner, M., Eliasson, A., Nicklasson, S., Sommerstein, K., & Hagglund, G. (2008). Hand Function in Cerebral Palsy. Report of 367 Children in a Population-Based Longitudinal Health Care Program. *The Journal of hand surgery, 33*(8), 1337 - 1347. https://doi.org/10.1016/j.jhsa.2008.02.032

Arnould, C., Bleyenheuft, Y., & Thonnard, J. (2014). Hand Functioning in Children with Cerebral Palsy. *Frontiers in Neurology, 5*, 48. https://doi.org/10.3389/fneur.2014.00048

Arnould, C., Penta, M., Renders, A., & Thonnard, J. (2004). ABILHAND-Kids: A measure of manual ability in children with cerebral palsy. *Neurology, 63*(6), 1045-1052. https://doi.org/10.1212/01.WNL.0000138423.77640.37

Australian Cerebral Palsy Register. (2013). *Report of the Australian Cerebral Palsy Register, Birth Years 1993-2006, February 2013*. www.cerebralpalsy.org.au

Australian Hand Therapy Association. (2012). Australian Hand Therapy Association Hand and Upper Limb Orthosis/Splint schedule. www.ahta.com.au

Autti-Ramo, I. M. D. P., Suoranta, J. M., Anttila, H. M., Malmivaara, A. M. D. P., & Makela, M. M. D. P. (2006). Effectiveness of Upper and Lower Limb Casting and Orthoses in Children with Cerebral Palsy: An Overview of Review Articles. *American Journal of Physical Medicine & Rehabilitation, 85*(1), 89-103. https://doi.org/10.1097/01.phm.0000179442.59847.27

Baker, R. (2016). *Rachel Baker introduces Q methodology*. https://qmethod.org/type/video/

Banasick, S. (2018). *Ken-Q Analysis* In *A web application for Q methodology* (Version 0.11.1) Free Software Foundation. https://shawnbanasick.github.io/ken-q-analysis/#section1

Barbosa, J. C., Willoughby, P., Rosenberg, C. A., & Mrtek, R. G. (1998). Statistical Methodology: VII. Q-Methodology, a Structural Analytic Approach to Medical Subjectivity. *Academic Emergency Medicine, 5*(10), 1032-1040. https://doi.org/10.1111/j.1553-2712.1998.tb02786.x

Barroso, P. N., Vecchio, S. D., Xavier, Y. R., Sesselmann, M., Araújo, P. A., & Pinotti, M. (2011). Improvement of hand function in children with cerebral palsy via an orthosis that provides wrist extension and thumb abduction. *Clinical Biomechanics, 26*(9), 937-943. https://doi.org/10.1016/j.clinbiomech.2011.05.006

Bartlett, D. J., & Palisano, R. J. (2000). A Multivariate Model of Determinants of Motor Change for Children With Cerebral Palsy. *Physical Therapy, 80*(6), 598-614. https://doi.org/10.1093/ptj/80.6.598

Basu, A. P., Pearse, J., Kelly, S., Wisher, V., & Kisler, J. (2014). Early intervention to improve hand function in hemiplegic cerebral palsy. *Frontiers in Neurology, 5*, 281. https://doi.org/10.3389/fneur.2014.00281

Bennett, S., Hoffmann, T., McCluskey, A., McKenna, K., Strong, J., & Tooth, L. (2003). Introducing OTseeker (Occupational Therapy Systematic Evaluation of Evidence): A New Evidence Database for Occupational Therapists. *American Journal of Occupational Therapy, 57*(6), 635-638. https://doi.org/10.5014/ajot.57.6.635

Blair, E. (2010). Epidemiology of the cerebral palsies. *Orthopedic Clinics of North America, 41*(4), 441-455. https://doi.org/10.1016/j.ocl.2010.06.004

Blair, E., Ballantyne, J., Horsman, S., & Chauvel, P. (1995). A study of a dynamic proximal stability splint in the management of children with cerebral palsy. *Developmental Medicine & Child Neurology, 37*(6), 544-554. https://doi.org/10.1111/j.1469-8749.1995.tb12041.x

Boniface, G., Fedden, T., Hurst, H., Mason, M., Phelps, C., Reagon, C., & Waygood, S. (2008). Using Theory to Underpin an Integrated Occupational Therapy Service through the Canadian Model of Occupational Performance. *British Journal of Occupational Therapy, 71*(12), 531-539. https://doi.org/10.1177/030802260807101206

Boniface, G., & Seymour, A. (2013). *Using Occupational Therapy Theory in Practice*. John Wiley & Sons, Incorporated.

Borell, L., Nygård, L., Asaba, E., Gustavsson, A., & Hemmingsson, H. (2012). Qualitative approaches in occupational therapy research. *Scandinavian Journal of Occupational Therapy, 19*(6), 521-529. https://doi.org/10.3109/11038128.2011.649782

Boscheinen-Morrin, J., & Conolly, W. B. (2001). *The hand: fundamentals of therapy* (3rd ed.). Butterworth-Heinemann.

Boyd, R. N., Morris, M. E., & Graham, H. K. (2001). Management of upper limb dysfunction in children with cerebral palsy: a systematic review. *European Journal of Neurology, 8*, 150-166. https://doi.org/10.1046/j.1468-1331.2001.00048.x

Braendvik, S. M., Elvrum, A. K., Vereijken, B., & Roeleveld, K. (2010, Feb). Relationship between neuromuscular body functions and upper extremity activity in children with cerebral palsy. *Developmental Medicine & Child Neurology, 52*(2), e29-34. https://doi.org/10.1111/j.1469-8749.2009.03490.x

Brand, P. W., & Hollister, A. M. (1999). *Clinical Mechanics of the Hand* (3rd ed.). Mosby.

Brown, S. R. (1970). On the use of variance designs in Q methodology. *The Psychological Record, 20*, 179-189.

Brown, S. R. (1980). *Political subjectivity: Applications of Q methodology in political science*. Yale University Press.

Brown, S. R. (1993). A primer on Q methodology. *Operant subjectivity, 16*(3/4), 91-138.

Brown, S. R. (2008). Q Methodology. In. SAGE Publications, Inc. https://doi.org/10.4135/9781412963909

Burgess, T., Rassafiani, M., & Copley, J. (2008). Factors associated with the acquisition of expertise in upper limb hypertonicity management. *British Journal of Occupational Therapy, 71*(8), 329-338. https://doi.org/10.1177/030802260807100806

Burtner, P. A., Poole, J. L., Torres, T., Medora, A. M., Abeyta, R., Keene, J., & Qualls, C. (2008). Effect of Wrist Hand Splints on Grip, Pinch, Manual Dexterity, and Muscle Activation in Children with Spastic Hemiplegia: A Preliminary Study. *Journal of Hand Therapy, 21*(1), 36-43. https://doi.org/10.1197/j.jht.2007.08.018

Cameron, D., Craig, T., Edwards, B., Missiuna, C., Schwellnus, H., & Polatajko, H. J. (2017) Cognitive Orientation to daily Occupational Performance (CO-OP): A New Approach for Children with Cerebral Palsy. *Physical & Occupational Therapy In Pediatrics, 37*(2), 183-198. https://doi.org/10.1080/01942638.2016.1185500

CanChild. (2004). *Outcome measures rating form*. McMaster University. https://www.canchild.ca

Cans, C. (2000). Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. *Developmental Medicine & Child Neurology, 42*(12), 816-824. https://doi.org/10.1111/j.1469-8749.2000.tb00695.x

Carmick, J. (1997). Use of Neuromuscular Electrical Stimulation and a Dorsal Wrist Splint to Improve the Hand Function of a Child With Spastic Hemiparesis. *Physical Therapy, 77*(6), 661-671. https://doi.org/10.1093/ptj/77.6.661

Casey, C. A., & Kratz, E. J. (1988). Soft Splinting With Neoprene: The Thumb Abduction Supinator Splint. *American Journal of Occupational Therapy, 42*(6), 395-398. https://doi.org/10.5014/ajot.42.6.395

Centre for Reviews and Dissemination. (2009). *Systematic reviews: CRD's guidance for undertaking reviews in health care* (Third ed.). CRD, University of York.

Chee, D. Y., Lee, H. C., Falkmer, M., Barnett, T., Falkmer, O., Siljehav, J., & Falkmer, T. (2015, Feb). Viewpoints on driving of individuals with and without autism spectrum disorder. *Developmental Neurorehabilitation, 18*(1), 26-36. https://doi.org/10.3109/17518423.2014.964377

Chhawchhria, Y. (2014). Comparison between above elbow wrist hand orthosis and combination of elbow gutter with below elbow wrist hand orthosis in children with spastic Cerebral Palsy. *Indian Journal of Occupational Therapy, 46*(3), 83-89.

Chin, T. Y. P., Duncan, J. A., Johnstone, B. R., & Graham, H. K. (2005). Management of the upper limb in cerebral palsy. *Journal of Pediatric Orthopaedics B, 14*(6), 389-404. https://doi.org/10.1097/01202412-200511000-00001

Clarke, D. J., & Holt, J. (2015). Understanding nursing practice in stroke units: a Q-methodological study. *Disability and Rehabilitation, 37*(20), 1870-1880. https://doi.org/10.3109/09638288.2014.986588

Coakes, S. J. (2010). *SPSS : analysis without anguish : version 17.0 for windows / Sheridan J. Coakes, Lyndall Steed, Clara Ong*. Wiley.

Cohn, E. S. (1991). Clinical Reasoning: Explicating Complexity. *American Journal of Occupational Therapy, 45*(11), 969-971. https://doi.org/10.5014/ajot.45.11.969

Colditz, J. C. (1996). Principles of splinting and splint prescription. In C. A. Peimer (Ed.), Surgery of the Hand and Upper Extremity (pp. 2389-2410). McGraw-Hill, Health Professions Division. https://books.google.com.au/books?id=5gsdgz_oWDMC

Copley, J., & Kuipers, K. (1999). *Management of upper limb hypertonicity*. Therapy Skill Builders.

Copley, J., & Kuipers, K. (2014a). The Hypertonicity Intervention Planning Model for Upper Limb Neurorehabilitation. In *Neurorehabilitation of the upper limb across the lifespan: Managing hypertonicity for optimal function* (pp. 54-79). John Wiley & Sons. https://doi.org/10.1002/9781118874875.ch3

Copley, J., & Kuipers, K. (2014b). *Neurorehabilitation of the upper limb across the lifespan: Managing hypertonicity for optimal function*. John Wiley & Sons.

Copley, J., Kuipers, K., Fleming, J., & Rassafiani, M. (2013). Individualised resting hand splints for adults with acquired brain injury: A randomized, single blinded, single case design. *NeuroRehabilitation, 32*(4), 885-898. https://doi.org/10.3233/NRE-130913

Copley, J., Turpin, M., Brosnan, J., & Nelson, A. (2008). Understanding and negotiating: Reasoning processes used by an occupational therapist to individualize intervention decisions for people with upper limb hypertonicity. *Disability and Rehabilitation, 30*(19), 1486-1498. https://doi.org/10.1080/09638280701654799

Corr, S. (2001). An Introduction to Q Methodology, a Research Technique. *British Journal of Occupational Therapy, 64*(6), 293-297. https://doi.org/10.1177/030802260106400605

Corr, S. (2006). Explrong perceptions about services using Q-sort methodology. In G. Kielhofner (Ed.), *Research in occupational therapy: Methods of inquiry for enhancing practice* (pp. 389-400). FA Davis.

Corr, S., Neill, G., & Turner, A. (2005). Comparing an Occupational Therapy Definition and Consumers' Experiences: A Q Methodology Study. *British Journal of Occupational Therapy, 68*(8), 338-346. https://doi.org/10.1177/030802260506800802

Cross, R. M. (2005). Exploring attitudes: the case for Q methodology. *Health Education Research, 20*(2), 206-213. https://doi.org/10.1093/her/cyg121

Currie, D. M., & Mendiola, A. (1987). Cortical thumb orthosis for children with spastic hemiplegic cerebral palsy. Archives of Physical Medicine and Rehabilitation, 68(4), 214-216. http://europepmc.org/abstract/MED/3566512

Davids, J. R., Peace, L. C., Wagner, L. V., Gidewall, M. A., Blackhurst, D. W., & Roberson, W. M. (2006). Validation of the Shriners Hospital for Children Upper Extremity Evaluation (SHUEE) for Children with Hemiplegic Cerebral Palsy. *The Journal of Bone and Joint Surgery, 88*(2), 326-333. https://doi.org/10.2106/jbjs.e.00298

de Bruin, M., Smeulders, M. J., & Kreulen, M. (2013). Why is joint range of motion limited in patients with cerebral palsy? *Journal of Hand Surgery European Volume, 38*(1), 8-13. https://doi.org/10.1177/1753193412444401

DeMatteo, C., Law, M., Russell, D., Pollock, N., Rosenbaum, P., & Walter, S. (1993). The reliability and validity of Quality of Upper Extremity Skills Test. *Physical and Occupational Therapy in Pediatrics, 13*(2), 1 - 18.

Dennis, K. E. (1986). Q methodology: relevance and application to nursing research. *Advances in Nursing Science, 8*(3), 6-17. http://journals.lww.com/advancesinnursingscience/Fulltext/1986/04000/Q_methodology__relevance_and_application_to.3.aspx

Donner, J. C. (2001). Using Q-sorts in participatory processes: an introduction to the methodology. *Social Development Papers, 36*, 24-49.

Dziopa, F., & Ahern, K. (2011). A systematic literature review of the applications of Q-technique and its methodology. *Methodology: European Journal of Research Methods for the Behavioral and Social Sciences, 7*(2), 39. https://doi.org/10.1027/1614-2241/a000021

Eliasson, A.-C. (2005). Improving the Use of Hands in Daily Activities. *Physical & Occupational Therapy in Pediatrics, 25*(3), 37-60. https://doi.org/doi:10.1080/J006v25n03_04

Eliasson, A.-C., & Burtner, P. A. (2008). Goal-oriented training of daily activities - A model for intervention. In *Improving hand function in cerebral palsy theory, evidence and intervention* (pp. 286-297). London : Mac Keith Press.

Eliasson, A.-C., Ekholm, C., & Carlstedt, T. (1998). Hand function in children with cerebral palsy after upper-limb tendon transfer and muscle release. *Developmental Medicine & Child Neurology, 40*(9), 612-621. https://doi.org/10.1111/j.1469-8749.1998.tb15427.x

Eliasson, A.-C., Krumlinde-Sundholm, L., Rösblad, B., Beckung, E., Arner, M., Öhrvall, A.-M., & Rosenbaum, P. (2006). The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine & Child Neurology, 48*(07), 549-554. https://doi.org/10.1111/j.1469-8749.2006.tb01313.x

Elvrum, A.-K., Andersen, G., Himmelmann, K., Beckung, E., Öhrvall, A.-M., Lydersen, S., & Vik, T. (2016). Bimanual Fine Motor Function (BFMF) Classification in Children with Cerebral Palsy: Aspects of Construct and Content Validity. *Physical & Occupational Therapy in Pediatrics, 36*(1), 1-16. https://doi.org/10.3109/01942638.2014.975314

Elvrum, A.-K. G., Zethræus, B.-M., Vik, T., & Krumlinde-Sundholm, L. (2018). Development and Validation of the Both Hands Assessment for Children With Bilateral Cerebral Palsy. *Physical & Occupational Therapy in Pediatrics, 38*(2), 113-126. https://doi.org/10.1080/01942638.2017.1318431

Esquenazi, A., Albanese, A., Chancellor, M. B., Elovic, E., Segal, K. R., Simpson, D. M., Smith, C. P., & Ward, A. B. (2013). Evidence-based review and assessment of botulinum neurotoxin for the treatment of adult spasticity in the upper motor neuron syndrome. *Toxicon, 67*, 115-128. https://doi.org/10.1016/j.toxicon.2012.11.025

Exner, C. E., & Bonder, B. R. (1983). Comparative Effects of Three Hand Splints on Bilateral Hand Use, Grasp, and Arm-Hand Posture in Hemiplegic Children: A Pilot Study. *OTJR: Occupation, Participation and Health, 3*(2), 75-92. https://doi.org/10.1177/153944928300300202

Farlex Medical Dictionary. (2012). https://medical-dictionary.thefreedictionary.com/

Fedrizzi, E., Pagliano, E., & Andreucci, E. (2003). Hand function in children with hemiplegic cerebral palsy: prospective follow-up and functional outcome in adolescence. *Developmental Medicine & Child Neurology, 45*(2), 85-91. https://doi.org/10.1111/j.1469-8749.2003.tb00910.x

Fehlings, D., Novak, I., Berweck, S., Hoare, B., Stott, N. S., & Russo, R. N. (2010). Botulinum toxin assessment, intervention and follow-up for paediatric upper limb hypertonicity: international consensus statement. *European Journal of Neurology, 17*(s2), 38-56. https://doi.org/10.1111/j.1468-1331.2010.03127.x

Fess, E. E. (2011). Orthoses for mobilization of joints: principles and methods. In T. M. Skirven, A. L. Osterman, J. Fedorczyk, & P. C. Amadio (Eds.), *Rehabilitation of the hand and upper extremity : expert consult* (pp. 1588-1598). Elsevier Health Sciences.

Fess, E. E., Gettle, K. S., Philips, C. A., & Janson, J. R. (2004). *Hand and upper extremity splinting: principles and methods*. Mosby Inc.

Fisher, A., & Bray Jones, K. (2017). Occupational Therapy Intervention Process Model. In J. Hinojoa, P. Kramer, & C. Royeen (Eds.), *Perspectives on Human Occupation: Theories Underlying Practice* (pp. 237-286). F.A. Davis.

Flegle, J. H., & Leibowitz, J. M. (1988). Improvement in grasp skill in children with hemiplegia with the MacKinnon splint. Research in developmental disabilities, 9(2), 145-151. https://dx.doi.org/10.1016/0891-4222(88)90048-0

Fleming, M. H. (1991a). Clinical reasoning in medicine compared with clinical reasoning in occupational therapy. *American Journal of Occupational Therapy, 45*(11), 988-996. https://doi.org/10.5014/ajot.45.11.988

Fleming, M. H. (1991b). The therapist with the three-track mind. *American Journal of Occupational Therapy, 45*(11), 1007-1014. https://doi.org/10.5014/ajot.45.11.1007

Foran, J. R. H., Steinman, S., Barash, I., Chambers, H. G., & Lieber, R. L. (2005). Structural and mechanical alterations in spastic skeletal muscle. Developmental Medicine & Child Neurology, 47(10), 713-717. https://doi.org/10.1111/j.1469-8749.2005.tb01063.x

Frank, G., & Polkinghorne, D. (2010). Qualitative Research in Occupational Therapy: From the First to the Second Generation. *OTJR: Occupation, Participation and Health, 30*(2), 51-57. https://doi.org/10.3928/15394492-20100325-02

Fristedt, S., Wretstrand, A., Björklund, A., Corr, S., & Falkmer, T. (2012). Viewpoints on community mobility and participation in older age. *Journal of Human Subjectivity, 10*(1), 103-123.

Fulford, F. E., & Brown, J. K. (1976, Jun). Position as a cause of deformity in children with cerebral palsy. *Dev Med Child Neurol, 18*(3), 305-314. https://doi.org/10.1111/j.1469-8749.1976.tb03652.x

Gagliardi, A. R., Berta, W., Kothari, A., Boyko, J., & Urquhart, R. (2016). Integrated knowledge translation (IKT) in health care: a scoping review. *Implementation Science, 11*(1), 38. https://doi.org/10.1186/s13012-016-0399-1

Gainsborough, M., Surman, G., Maestri, G., Colver, A., Cans, C. (2008). Validity and reliability of the guidelines of the Surveillance of Cerebral Palsy in Europe for the classification of cerebral palsy. *Developmental Medicine & Child Neurology, 50*(11), 828-831. https://doi.org/10.1111/j.1469-8749.2008.03141.x

Garbellini, S., Randall, M., Steele, M., Elliott, C., & Imms, C. (2019). Prescribing upper limb orthoses for children with cerebral palsy: a Q methodology study of occupational therapists' decision making. *Disability and Rehabilitation*, 1-11. https://doi.org/10.1080/09638288.2019.1573931

Garbellini, S., Randall, M., Steele, M., Elliott, C., & Imms, C. (2020). Unpacking the application of Q methodology for use in occupational therapy research. *Scandinavian Journal of Occupational Therapy*, 1–6. https://doi.org/10.1080/11038128.2019.1709542

Garbellini, S., Robert, Y., Randall, M., Elliott, C., & Imms, C. (2017). Rationale for prescription, and effectiveness of, upper limb orthotic intervention for children with cerebral palsy: a systematic review. *Disability and Rehabilitation*, 1-11. https://doi.org/10.1080/09638288.2017.1297498

Garbellini, S., & Wilton, J. (2017). *Neurological Hand Deformity Classification*. https://www.neurohanddeformity.com/

Georgiades, M., Elliott, C., Wilton, J., Blair, E., Blackmore, M., & Garbellini, S. (2014). The Neurological Hand Deformity Classification for children with cerebral palsy. *Australian Occupational Therapy Journal, 61*, 394-402. https://doi.org/10.1111/1440-1630.12150

Goodman, G., & Bazyk, S. (1991). The effects of a short thumb opponens splint on hand function in cerebral palsy: a single-subject study. *American Journal of Occupational Therapy, 45*(8), 726-731. https://doi.org/10.5014/ajot.45.8.726

Gormley Jr, M. E. (2001). Treatment of neuromuscular and musculoskeletal problems in cerebral palsy. *Developmental Neurorehabilitation, 4*(1), 5-16. https://doi.org/doi:10.1080/13638490151068393

Graham, H. K. (2004). Mechanisms of deformity. In D. Scrutton, D. Damiano, & M. Mayston (Eds.), *Management of the motor disorders of children with cerebral palsy* (2nd edition ed., pp. 105-129). Mac Keith Press.

Graham, H. K. (2005). Classifying cerebral palsy. *Journal of Pediatric Orthopaedics, 25*(1), 127-128.

Graham, H. K., & Selber, P. (2003). Musculoskeletal aspects of cerebral palsy. *Journal of Bone & Joint Surgery, British Volume, 85-B*(2), 157-166. https://doi.org/10.1302/0301-620x.85b2.14066

Graham, I., Logan, J., Harrison, M., Straus, S., Tetroe, J., Caswell, W., & Robinson, N. (2006). Lost in knowledge translation: time for a map? *Journal of Continuing Education in the Health Professions, 26*. https://doi.org/10.1002/chp.47

Grimshaw, J. M., Eccles, M. P., Lavis, J. N., Hill, S. J., & Squires, J. E. (2012). Knowledge translation of research findings. *Implementation Science, 7*. https://doi.org/10.1186/1748-5908-7-50

Haley, S. M., Coster, W. J., Dumas, H. M., Fragala-Pinkham, M. A., Kramer, J., Ni, P., Tian, F., Kao, Y.-C., Moed, R., & Ludlow, L. H. (2011). Accuracy and precision of the Pediatric Evaluation of Disability Inventory computer-adaptive tests (PEDI-CAT). *Developmental Medicine & Child Neurology, 53*(12), 1100-1106. https://doi.org/doi:10.1111/j.1469-8749.2011.04107.x

Hanna, S. E., Law, M. C., Rosenbaum, P. L., King, G. A., Walter, S. D., Pollock, N., & Russell, D. J. (2003). Development of hand function among children with cerebral palsy: growth curve analysis for ages 16 to 70 months. *Developmental Medicine & Child Neurology, 45*(7), 448-455. https://doi.org/10.1111/j.1469-8749.2003.tb00939.x

Harries, P. A., & Harries, C. (2001). Studying clinical reasoning, part 1: Have we been taking the wrong 'track'? *British Journal of Occupational Therapy, 64*(4), 164-168. https://doi.org/10.1177/030802260106400402

Heaver C, Jarvis S, Johnson B, Kuiper J, Freeman R. (2015). Shriners Hospital for Children Upper Extremity Evaluation (SHUEE): A reliability study. *Gait & Posture, 42*:S27-S28. https://doi.org/https://doi.org/10.1016/j.gaitpost.2015.03.056

Hedberg-Graff, J., Granstrom, F., Arner, M., & Krumlinde-Sundholm, L. (2019). Upper-limb contracture development in children with cerebral palsy: a population-based study. *Developmental Medicine & Child Neurology, 61*(2), 204-211. https://doi.org/10.1111/dmcn.14006

Hidecker, M. J. C., Paneth, N., Rosenbaum, P., Kent, R. D., Lillie, J., Eulenberg, J. B., Chester, J. K., Johnson, B., Michalsen, L., Evatt, M., & Taylor, K. (2011). Developing and validating the Communication Function Classification System for individuals with cerebral palsy. *Developmental Medicine & Child Neurology, 53*(8), 704-710. https://doi.org/10.1111/j.1469-8749.2011.03996.x

Hoare, B., & Greaves, S. (2017). Unimanual versus bimanual therapy in children with unilateral cerebral palsy: Same, same, but different. *Journal of Pediatric Rehabilitation Medicine, 10*(1), 47-59. https://doi.org/10.3233/PRM-170410

Hoare, B., Imms, C., Randall, M., & Carey, L. (2011). Linking cerebral palsy upper limb measures to the International Classification of Functioning, Disability and Health. *Journal of Rehabilitation Medicine, 43*(11), 987-996. https://doi.org/10.2340/16501977-0886

Hooper, B. (1997). The Relationship Between Pretheoretical Assumptions and Clinical Reasoning. *American Journal of Occupational Therapy, 51*(5), 328-338. https://doi.org/10.5014/ajot.51.5.328

Horger, M. M. (1990). The Reliability of Goniometric Measurements of Active and Passive Wrist Motions. *American Journal of Occupational Therapy, 44*(4), 342-348. https://doi.org/10.5014/ajot.44.4.342

House, J., Gwathmey, F., & Fidler, M. (1981). A dynamic approach to the thumb-in palm deformity in cerebral palsy. *Journal of Bone and Joint Surgery, American Volume, 63*(2), 216-225.

Howard, J., Soo, B., Graham, H. K., Boyd, R. N., Reid, S., Lanigan, A., Wolfe, R., & Reddihough, D. S. (2005). Cerebral palsy in Victoria: Motor types, topography and gross motor function. *Journal of Paediatrics and Child Health, 41*(9-10), 479-483. https://doi.org/10.1111/j.1440-1754.2005.00687.x

Hughes, A., Franzsen, D., & Freeme, J. (2017). The effect of neoprene thumb abduction splints on upper limb function in children with cerebral palsy. *The South African Journal of Occupational Therapy, 47*(3), 3-10.https://doi.org/10.17159/2310-3833/2017/v47n3a2

Imms, C., Wallen, M., Elliott, C., Hoare, B., Randall, M., Greaves, S., Adair, B., Bradshaw, E., Carter, R., Orsini, F., Shih, S. T. F., & Reddihough, D. (2016). Minimising impairment: Protocol for a multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy. *BMC Pediatrics, 16*(1), 70. https://doi.org/10.1186/s12887-016-0608-8

International Organization for Standardization. (1989). Prosthetics and Orthotics_(ISO/TC168). Retreived from https://www.iso.org/committee/53630.html

Ivanhoe, C., & Reistetter, T. (2004,). Spasticity: The Misunderstood Part of the Upper Motor Neuron Syndrome. *American Journal of Physical Medicine & Rehabilitation, 83*(10), S3-S9. https://doi.org/10.1097/01.phm.0000141125.28611.3e

Jackman, M., Novak, I., & Lannin, N. (2014a). Effectiveness of functional hand splinting and the cognitive orientation to occupational performance (CO-OP) approach in children with cerebral palsy and brain injury: two randomised controlled trial protocols. *BMC Neurology*, 14(1), 144. http://www.biomedcentral.com/1471-2377/14/144

Jackman, M., Novak, I., & Lannin, N. (2014b). Effectiveness of hand splints in children with cerebral palsy: a systematic review with meta-analysis. *Developmental Medicine & Child Neurology, 56*(2), 138-147. https://doi.org/10.1111/dmcn.12205

Jackman, M., Novak, I., Lannin, N., Froude, E., Miller, L., & Galea, C. (2018). Effectiveness of Cognitive Orientation to daily Occupational Performance over and above functional hand splints for children with cerebral palsy or brain injury: a randomized controlled trial. *BMC Pediatrics, 18*(1), 248. https://doi.org/10.1186/s12887-018-1213-9

Jackman, M., Novak, I., Lannin, N., & Galea, C. (2019). Immediate effect of a functional wrist orthosis for children with cerebral palsy or brain injury: A randomized controlled trial. *Journal of Hand Therapy*, 32(1), 10-16. https://doi.org/10.1016/j.jht.2017.09.006

Kanellopoulos, A. D., Mavrogenis, A. F., Mitsiokapa, E. A., Panagopoulos, D., Skouteli, H., Vrettos, S. G., Tzanos, G., & Papagelopoulos, P. J. (2009). Long lasting benefits following the combination of static night upper extremity splinting with botulinum toxin A injections in cerebral palsy children. *European Journal of Physical and Rehabilitation Medicine, 45*(4), 501-506.

Kenward, L. (2019). A literature review to guide novice researchers using Q methodology in the development of a framework for concourse management. *Nurse Researcher, 27*(1), 17-21. https://doi.org/10.7748/nr.2019.e1616

Ketalaar, M., Dahmen, A., Vermeer, J., & Helders, P. J. (1998). Functional motor abilities of children with cerebral palsy: a systematic literature review of assessment measures. *Clinical Rehabilitation, 12*(5), 369-380. http://dx.doi.org/10.1191/026921598673571117

King, G., Currie, M., Bartlett, D. J., Strachan, D., Tucker, M. A., & Willoughby, C. (2008). The development of expertise in paediatric rehabilitation therapists: The roles of motivation, openness to experience, and types of caseload experience. *Australian Occupational Therapy Journal, 55*(2), 108-122. https://doi.org/10.1111/j.1440-1630.2007.00681.x

Kinghorn, J., & Roberts, G. (1996). The effect of an inhibitive weight-bearing splint on tone and function: a single-case study. *American Journal of Occupational Therapy, 50*(10), 807-815. https://doi.org/10.5014/ajot.50.10.807

Kiresuk, T. J., Cardillo, J. E., & Lougaris, I. A. (2014). *Goal attainment scaling : applications, theory, and measurement*. London : Psychology Press, Taylor and Francis Group.

Kirshner, B., & Guyatt, G. (1985). A methodological framework for assessing health indices. *Journal of chronic diseases, 38*(1), 27-36. https://doi.org/10.1016/0021-9681(85)90005-0

Klingels, K., De Cock, P., Molenaers, G., Desloovere, K., Huenaerts, C., Jaspers, E., & Feys, H. (2010). Upper limb motor and sensory impairments in children with hemiplegic cerebral palsy. Can they be measured reliably? *Disability & Rehabilitation, 32*(5), 409-416. https://doi.org/10.3109/09638280903171469

Klingels, K., Meyer, S., Mailleux, L., Simon-Martinez, C., Hoskens, J., Monbaliu, E., Verheyden, G., Verbeke, G., Molenaers, G., Ortibus, E., & Feys, H. (2018). Time Course of Upper Limb Function in Children with Unilateral Cerebral Palsy: A Five-Year Follow-Up Study. *Neural Plasticity, 2018,* 2831342. https://doi.org/10.1155/2018/2831342

Kothari, A., & Wathen, C. (2012). A critical second look at integrated knowledge translation. *Health policy, 109*. https://doi.org/10.1016/j.healthpol.2012.11.004

Koman, L. A., Williams, R. M., Evans, P. J., Richardson, R., Naughton, M. J., Passmore, L., & Smith, B. P. (2008). Quantification of upper extremity function and range of motion in children with cerebral palsy. *Developmental Medicine & Child Neurology, 50*(12), 910-917. 10.1111/j.1469-8749.2008.03098.x

Kramlinger, A., Strecker Neufeld, P., & Berg, C. (2016). Creating a Community Capacity Assessment to Identify Agency Outcomes Related to Occupational Therapy Student Community Partnerships. *Occupational Therapy In Health Care, 30*(3), 255-271. https://doi.org/10.3109/07380577.2016.1160464

Kreulen, M., Smeulders, M. J. C., Veeger, H. E. J., & Hage, J. J. (2006). Movement patterns of the upper extremity and trunk before and after corrective surgery of impaired forearm rotation in patients with cerebral palsy. *Developmental Medicine & Child Neurology, 48*(6), 436-441. https://doi.org/10.1111/j.1469-8749.2006.tb01292.x

Krumlinde-Sundholm, L., Holmefur, M., Kottorp, A., & Eliasson, A.-C. (2007). The Assisting Hand Assessment: current evidence of validity, reliability, and responsiveness to change. *Developmental Medicine & Child Neurology, 49*(4), 259-264. https://doi.org/10.1111/j.1469-8749.2007.00259.x

Kuipers, K., & Grice, J. W. (2009). The structure of novice and expert occupational therapists' clinical reasoning before and after exposure to a domain-specific protocol. *Australian Occupational Therapy Journal, 56*(6), 418-427. https://doi.org/10.1111/j.1440-1630.2009.00793.x

Kuipers, K., McKenna, K., & Carlson, G. (2006). Factors Influencing Occupational Therapists' Clinical Decision Making for Clients with Upper Limb Performance Dysfunction following Brain Injury. *The British Journal of Occupational Therapy, 69*(3), 106-114. https://doi.org/10.1177/030802260606900303

Kuipers, K., Rassafiani, M., Ashburner, J., Griffin, J., Worley, L., Moes, L., Fleming, J., & Copley, J. (2009). Do clients with acquired brain injury use the splints prescribed by occupational therapists? A descriptive study. *NeuroRehabilitation, 24*(4), 365-375. https://doi.org/10.3233/NRE-2009-0491

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

Langlois, S., MacKinnon, J., & Pederson, L. (1989). Hand Splints and Cerebral Spasticity: A Review of the Literature. *Canadian Journal of Occupational Therapy, 56*(3), 113-119. https://doi.org/10.1177/000841748905600304

Lannin, N., & Ada, L. (2011). Neurorehabilitation splinting: Theory and principles of clinical use. *NeuroRehabilitation, 28*(1), 21-28. https://doi.org/10.3233/NRE-2011-0628

Lannin, N., Novak, I., & Jackman, M. (2017). Orthotics. In M. Curtin, M. Egan, & J. Adams (Eds.), *Occupational therapy for people experiencing illness, injury or impairment : promoting occupation and participation* (Seventh ed., pp. 541-558). Elsevier.

Law, K., Lee, E., Fung, B., Yan, L., Gudushauri, P., Wang, K., Ip, J., & Chow, S. (2008). Evaluation of deformity and hand function in cerebral palsy patients. *Journal of Orthopaedic Surgery and Research, 3*(1), 52. http://www.josr-online.com/content/3/1/52

Law, M. (1987). Measurement in occupational therapy: Scientific criteria for evaluation. *Canadian Journal of Occupational Therapy, 54*(3), 133-138. https://doi.org/10.1177/000841748705400308

Law, M., Baptiste, S., McColl, M., Opzoomer, A., Polatajko, H., & Pollock, N. (1990). The Canadian occupational performance measure: an outcome measure for occupational therapy. *Canadian Journal of Occupational Therapy, 57*(2), 82-87. https://doi.org/10.1177/000841749005700207

Law, M., & Laver-Fawcett, A. (2013). Canadian Model of Occupational Performance: 30 Years of Impact! *British Journal of Occupational Therapy, 76*(12), 519-519. https://doi.org/10.4276/030802213x13861576675123

Lidman, G., Nachemson, A., Peny-Dahlstrand, M., & Himmelmann, K. (2015). Botulinum toxin A injections and occupational therapy in children with unilateral spastic cerebral palsy: a randomized controlled trial. *Developmental Medicine & Child Neurology*, *57*(8), 754-761. https://doi.org/10.1111/dmcn.12739

Louwers, A., Meester-Delver, A., Folmer, K., Nollet, F., & Beelen, A. (2011). Immediate effect of a wrist and thumb brace on bimanual activities in children with hemiplegic cerebral palsy. *Developmental Medicine & Child Neurology, 53*(4), 321-326. https://doi.org/10.1111/j.1469-8749.2010.03849.x

Maher, C. G., Sherrington, C., Herbert, R. D., Moseley, A. M., & Elkins, M. (2003). Reliability of the PEDro Scale for Rating Quality of Randomized Controlled Trials. *Physical Therapy, 83*(8), 713-721.

Makki, D., Duodu, J., & Nixon, M. (2014). Prevalence and pattern of upper limb involvement in cerebral palsy. *Journal of children's orthopaedics, 8*(3), 215-219. https://doi.org/10.1007/s11832-014-0593-0

Mathiowetz, V., Federman, S., & Wiemer, D. (1985). Box and Block Test of Manual Dexterity: Norms for 6–19 Year Olds. *Canadian Journal of Occupational Therapy, 52*(5), 241-245. https://doi.org/10.1177/000841748505200505

Mattingly, C. (1991). What is Clinical Reasoning? *American Journal of Occupational Therapy, 45*(11), 979-986. https://doi.org/10.5014/ajot.45.11.979

Mattingly, C., & Fleming, M. H. (1994). *Clinical Reasoning: Forms of Inquiry in a Therapeutic Practice*. F.A. Davis. https://books.google.com.au/books?id=Vc91PwAACAAJ

Mayer, N. H., Esquenazi, A., & Childers, M. K. (1997). Common patterns of clinical motor dysfunction. *Muscle & Nerve, 20*(S6), 21-35.

McConnell, K., Johnston, L., & Kerr, C. (2011). Upper limb function and deformity in cerebral palsy: a review of classification systems. *Developmental Medicine & Child Neurology, 53*(9), 799-805. https://doi.org/10.1111/j.1469-8749.2011.03953.x

McKenzie, J., Braswell, B., Jelsma, J., & Naidoo, N. (2011). A case for the use of Q-methodology in disability research: lessons learned from a training workshop. *Disability and Rehabilitation, 33*(21-22), 2134-2141. https://doi.org/10.3109/09638288.2011.560327

McKenzie, A., & Hanley, B. (2012). *Consumer and Community Participation in Health and Medical Research: A practical guide for health and medical research organisations*. Consumer and community health research network. https://www.involvingpeopleinresearch.org.au/find-out-more/our-resources/

McKeown, B., & Thomas, D. (2013). Q Methodology (2 ed.) https://doi.org/10.4135/9781483384412

McMillan, I. R. (2011). The biomechanical frame of reference in occupational therapy. In E. A. Duncan (Ed.), *Foundations for Practice in Occupational Therapy* (5 ed., pp. 175-194). Elsevier Health Sciences.

McPherson, J. J. (1981). Objective Evaluation of a Splint Designed to Reduce Hypertonicity. *American Journal of Occupational Therapy, 35*(3), 189-194 186p. https://doi.org/10.5014/ajot.35.3.189

McPherson, J. J., Kreimeyer, D., Aalderks, M., & Gallagher, T. (1982). A comparison of dorsal and volar resting hand splints in the reduction of hypertonus. *American Journal of Occupational Therapy, 36*(10), 664-670. https://doi.org/10.5014/ajot.36.10.664

Messick, S. (1995). Standards of Validity and the Validity of Standards in Performance Asessment. *Educational Measurement: Issues and Practice, 14*(4), 5-8. https://doi.org/10.1111/j.1745-3992.1995.tb00881.x

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of internal medicine, 151*(4), 264-269. https://doi.org/10.1371/journal.pmed.1000097

Mokkink, L. B., de Vet, H. C. W., Prinsen, C. A. C., Patrick, D. L., Alonso, J., Bouter, L. M., & Terwee, C. B. (2018). COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. *Quality of Life Research, 27*(5), 1171-1179. https://doi.org/10.1007/s11136-017-1765-4

Mokkink, L. B., Prinsen, C. A. C., Patrick, D. L., Alonso, J., Bouter, L. M., De Vet, H. C. W., & Terwee, C. B. (2019). *COSMIN Study Design checklist for Patient-reported outcome measurement instruments*. https://www.cosmin.nl/wp-content/uploads/COSMIN-study-designing-checklist_final.pdf

Mokkink, L. B., Terwee, C. B., Patrick, D. L., Alonso, J., Stratford, P. W., Knol, D. L., Bouter, L. M., & De Vet, H. C. W. (2009). The COSMIN checklist manual. *Amsterdam: VU University Medical Centre*.

Morris, S. (2002). Ashworth and Tardieu Scales: their clinical relevance for measuring spasticity in adult and paediatric neurological populations. *Physical Therapy Reviews, 7*(1), 53-62. https://doi.org/10.1179/108331902125001770

Newman, I., & Ramlo, S. (2010). SAGE Handbook of Mixed Methods in Social & Behavioral Research. In A. Tashakkori & C. Teddlie (Eds.), (2 ed.). SAGE Publications, Inc. https://doi.org/10.4135/9781506335193

Novak, I., McIntyre, S., Morgan, C., Campbell, L., Dark, L., Morton, N., Stumbles, E., Wilson, S.-A., & Goldsmith, S. (2013). A systematic review of interventions for children with cerebral palsy: state of the evidence. *Developmental Medicine & Child Neurology, 55*(10), 885-910. https://doi.org/10.1111/dmcn.12246

Novak, I., Morgan, C., Fahey, M., Finch-Edmondson, M., Galea, C., Hines, A., Langdon, K., Namara, M. M., Paton, M. C. B., Popat, H., Shore, B., Khamis, A., Stanton, E., Finemore, O. P., Tricks, A., te Velde, A., Dark, L., Morton, N., & Badawi, N. (2020). State of the Evidence Traffic Lights 2019: Systematic Review of Interventions for Preventing and Treating Children with Cerebral Palsy. *Current Neurology and Neuroscience Reports, 20*(2), 3. https://doi.org/10.1007/s11910-020-1022-z

Numanoğlu, A., & Günel, M. K. (2012). Intraobserver reliability of modified Ashworth scale and modified Tardieu scale in the assessment of spasticity in children with cerebral palsy. *Acta orthopaedica et traumatologica turcica, 46*(3), 196. https://doi.org/10.3944/AOTT.2012.2697

Occupational Therapy Board of Ausrtalia. (2017). Registrant Data. Retreived from http://www.occupationaltherapyboard.gov.au/About/Statistics.aspx

Ohrvall, A. M., Krumlinde-Sundholm, L., & Eliasson, A. C. (2013). Exploration of the relationship between the Manual Ability Classification System and hand-function measures of capacity and performance. *Disability and Rehabilitation, 35*(11), 913-918. https://doi.org/10.3109/09638288.2012.714051

Oxford Dictionary. (2018). Oxford University Press. Retrieved 01/11/2019 from https://en.oxforddictionaries.com/

Ozer, K., Chesher, S. P., & Scheker, L. R. (2006). Neuromuscular electrical stimulation and dynamic bracing for the management of upper-extremity spasticity in children with cerebral palsy. *Developmental Medicine & Child Neurology, 48*(7), 559-563. https://doi.org/10.1111/j.1469-8749.2006.tb01315.x

Palisano, R., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology, 39*(4), 214-223. https://doi.org/10.1111/j.1469-8749.1997.tb07414.x

Park, E. S., Sim, E. G., & Rha, D.-w. (2011). Effect of upper limb deformities on gross motor and upper limb functions in children with spastic cerebral palsy. *Research in developmental disabilities, 32*(6), 2389-2397. https://doi.org/10.1016/j.ridd.2011.07.021

Parker, D. (2013). *An exploration of client-centred practice in occupational therapy: perspectives and impact* [University of Birmingham]. http://etheses.bham.ac.uk/4432/

Plastow, N. (2014). *Food Activities and the Maintenance of Identity in Later Life* [Brunel University]. Brunel University West London. https://bura.brunel.ac.uk/bitstream/2438/10893/1/FulltextThesis.pdf

Polit, D. F. (2014). Getting serious about test-retest reliability: a critique of retest research and some recommendations. *Quality Of Life Research: An International Journal Of Quality Of Life Aspects Of Treatment, Care And Rehabilitation, 23*(6), 1713-1720. https://doi.org/10.1007/s11136-014-0632-9

Pontén, E., Fridén, J., Thornell, L.-E., & Lieber, R. L. (2005). Spastic wrist flexors are more severely affected than wrist extensors in children with cerebral palsy. *Developmental Medicine & Child Neurology, 47*(6), 384-389. https://doi.org/10.1017/s0012162205000757

Portney, L., & Watkins, M. (2015). Foundations of clinical research : applications to practice (3rd Edition. ed.). Philadelphia, PA : F.A. Davis Company.

Postans, N., Wright, P., Bromwich, W., Wilkinson, I., Farmer, S. E., & Swain, I. (2010). The Combined Effect of Dynamic Splinting and Neuromuscular Electrical Stimulation in Reducing Wrist and Elbow Contractures in Six Children with Cerebral Palsy. *Prosthetics and Orthotics International, 34*(1), 10-19. https://doi.org/10.3109/03093640903051808

Randall, M., Harvey, A., Imms, C., Reid, S., Lee, K. J., & Reddihough, D. (2013). Reliable Classification of Functional Profiles and Movement Disorders of Children with Cerebral Palsy. *Physical & Occupational Therapy in Pediatrics, 33*(3), 342-352. https://doi.org/10.3109/01942638.2012.747584

Randall, M., Johnson, L., & Reddihough, D. S. (2013). The Melbourne Assessment 2: a test of unilateral upper limb function. Occupational Therapy Deptartment, Royal Children's Hospital, Melbourne.

Rapport, F., Clay-Williams, R., Churruca, K., Shih, P., Hogden, A., & Braithwaite, J. (2018). The struggle of translating science into action: Foundational concepts of implementation science. *Journal of Evaluation in Clinical Practice, 24*(1), 117-126. https://doi.org/doi:10.1111/jep.12741

Reddihough, D., & Collins, K. J. (2003). The epidemiology and causes of cerebral palsy. *Australian Journal of Physiotherapy, 49*(1), 7-12. https://doi.org/10.1016/s0004-9514(14)60183-5

Reid, D. T., & Sochaniwskyj, A. (1992). Influences of a hand positioning device on upper-extremity control of children with cerebral palsy. International Journal of Rehabilitation Research, 15(1), 15-30. https://doi.org/10.1097/00004356-199203000-00002

Riley, D. S., Barber, M. S., Kienle, G. S., Aronson, J. K., von Schoen-Angerer, T., Tugwell, P., Kiene, H., Helfand, M., Altman, D. G., Sox, H., Werthmann, P. G., Moher, D., Rison, R. A., Shamseer, L., Koch, C. A., Sun, G. H., Hanaway, P., Sudak, N. L., Kaszkin-Bettag, M., Carpenter, J. E., & Gagnier, J. J. (2017). CARE guidelines for case reports: explanation and elaboration document. *Journal of Clinical Epidemiology, 89*, 218-235. https://doi.org/10.1016/j.jclinepi.2017.04.026

Robertson, L. (1988). Qualitative Research Methods in Occupational Therapy. *British Journal of Occupational Therapy, 51*(10), 344-346. https://doi.org/10.1177/030802268805101004

Rogers, J. C. (1983). Eleanor Clarke Slagle Lectureship—1983; Clinical Reasoning: The Ethics, Science, and Art. *American Journal of Occupational Therapy, 37*(9), 601-616. https://doi.org/10.5014/ajot.37.9.601

Rogers, J. C., & Holm, M. B. (1991). Occupational Therapy Diagnostic Reasoning: A Component of Clinical Reasoning. *American Journal of Occupational Therapy, 45*(11), 1045-1053. https://doi.org/10.5014/ajot.45.11.1045

Rosenbaum, P., Eliasson, A.-C., Hidecker, M. J. C., & Palisano, R. J. (2014). Classification in Childhood Disability: Focusing on Function in the 21st Century. *Journal of Child Neurology, 29*(8), 1036-1045. https://doi.org/10.1177/0883073814533008

Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., Dan, B., & Jacobsson, B. (2007). A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine & Child Neurology Supplement, 109*, 8-14.

Rosenbaum, P., Russell, D. J., Cadman, D. T., Gowland, C., Jarvis, S., & Hardy, S. (1990). Issues in Measuring Change in Motor Function in Children with Cerebral Palsy: A Special Communication. *Physical Therapy, 70*(2), 125-131. http://ptjournal.apta.org/content/70/2/125.abstract

Rosenbaum, P., & Stewart, D. (2004). The World Health Organization International Classification of Functioning, Disability, and Health: a model to guide clinical thinking, practice and research in the field of cerebral palsy. *Seminars in pediatric neurology*, 11(1), 5-10. https://doi.org/10.1016/j.spen.2004.01.002

Rowley, J. (2002). Using case studies in research. *Management research news*.

Russo, R. (2015). Repeat botulinum toxin A injections: a step forward in the challenge to evaluate current clinical practice. *Developmental Medicine & Child Neurology, 57*(8), 702-703. https://doi.org/10.1111/dmcn.12764

Russo, R. N., Atkins, R., Haan, E., & Crotty, M. (2009). Upper limb orthoses and assistive technology utilization in children with hemiplegic cerebral palsy recruited from a population register. *Developmental Neurorehabilitation, 12*(2), 92-99. https://doi.org/10.1080/17518420902783223

Rybski, M. (2012). *Kinesiology for occupational therapy* (2nd ed.). Thorofare, N.J. : SLACK.

Sanger, T. D., Chen, D., Delgado, M. R., Gaebler-Spira, D., Hallett, M., Mink, J. W., & the taskforce on Childhood Motor Disorders. (2006). Definition and Classification of Negative Motor Signs in Childhood. *Pediatrics, 118*(5), 2159-2167. https://doi.org/10.1542/peds.2005-3016

Scheker, L. R., Chesher, S. P., & Ramirez, S. (1999). Neuromuscular electrical stimulation and dynamic bracing as a treatment for upper-extremity spasticity in children with cerebral palsy. *Journal of Hand Surgery, European Volume, 24*(2), 226-232. https://doi.org/10.1054/jhsb.1998.0002

Schell, B. (2019). Professional reasoning in practice. In B. Schell & G. Gillen (Eds.), *Willard & Spackman's Occupational Therapy* (13 ed.). Lippincott, Williams, & Wilkins.

Schell, B., & Cervero, R. M. (1993). Clinical Reasoning in Occupational Therapy: An Integrative Review. *American Journal of Occupational Therapy, 47*(7), 605-610. https://doi.org/10.5014/ajot.47.7.605

Schmolck, P. (2014). *PQMethod Software.* In (Version Release 2.35) http://schmolck.userweb.mwn.de/qmethod/

Scholtes, V. A. B., Becher, J. G., Beelen, A., & Lankhorst, G. J. (2006). Clinical assessment of spasticity in children with cerebral palsy: a critical review of available instruments. *Developmental Medicine & Child Neurology, 48*(1), 64-73. https://doi.org/10.1017/S0012162206000132

Schwartz, K. B. (1991). Clinical Reasoning and New Ideas on Intelligence: Implications for Teaching and Learning. *American Journal of Occupational Therapy, 45*(11), 1033-1037. https://doi.org/10.5014/ajot.45.11.1033

Shierk, A., Lake, A., & Haas, T. (2016). Review of Therapeutic Interventions for the Upper Limb Classified by Manual Ability in Children with Cerebral Palsy. *Seminars in Plastic Surgery, 30*(1), 14-23. https://doi.org/10.1055/s-0035-1571256

Smith, Y. A., Hong, E., & Presson, C. (2000). Normative and Validation Studies of the Nine-Hole Peg Test with Children. *Perceptual and Motor Skills, 90*(3), 823-843. https://doi.org/10.2466/pms.2000.90.3.823

Smithers-Sheedy, H., Badawi, N., Blair, E., Cans, C., Himmelmann, K., Krägeloh-Mann, I., McIntyre, S., Slee, J., Uldall, P., Watson, L., & Wilson, M. (2014). What constitutes cerebral palsy in the twenty-first century? *Developmental Medicine & Child Neurology, 56*(4), 323-328. https://doi.org/10.1111/dmcn.12262

Stanger, M., & Oresic, S. (2003). Rehabilitation Approaches for Children With Cerebral Palsy: Overview. *Journal of Child Neurology, 18*(1 suppl), S79-S88. https://doi.org/10.1177/08830738030180010S0201

Sterne, J., Higgins, J., & Reeves, B. (2014). *A Cochrane Risk Of Bias Assessment Tool: for Non-Randomized Studies of Interventions (ACROBAT-NRSI)*. Retrieved 17 January 2016 from http://www.riskofbias.info

Stevens, J. (2002). *Applied multivariate statistics for the social sciences* (4th ed.). Erlbaum.

Stewart, K., & Harvey, A. (2018). *The identification and measurement of dyskinesia in children with cerebral palsy. A toolkit for clinicians*. https://doi.org/10.25374/MCRI.6131264.v1

Streiner, D., & Norman, G. (2008). *Health Measurement Scales A practical guide to their development and use* (4th ed.). Oxford : Oxford University Press.

Sumsion, T., & Law, M. (2006). A Review of Evidence on the Conceptual Elements Informing Client-Centred Practice. *Canadian Journal of Occupational Therapy, 73*(3), 153-162. https://doi.org/10.1177/000841740607300303

Sy, M. P., Nobuo Ohshima, M., & Roraldo, P. N. R. (2018). The Role of Filipino Occupational Therapists in Substance Addiction and Rehabilitation: A Q-Methodology. *Occupational Therapy in Mental Health 34*(4), 367-388. https://doi.org/10.1080/0164212X.2018.1446206

Tardieu, G., & Tardieu, C. (1987). Cerebral palsy. Mechanical evaluation and conservative correction of limb joint contractures. *Clinical Orthopaedics and Related Research* (219), 63-69.

Tate, R. L., Rosenkoetter, U., Wakim, D., Sigmundsdottir, L., Doubleday, J., Togher, L., Mcdonald, S., & Perdices, M. (2015). *The Risk of Bias in N-of-1 Trials (RoBiNT) Scale : An expanded manual for the critical appraisal of single-case reports*.

Teplicky, R., Law, M., & Russell, D. (2002). The effectiveness of casts, orthoses, and splints for children with neurological disorders. *Infants & Young Children: An Interdisciplinary Journal of Special Care Practices, 15*(1), 42-50.

Terwee, C. B., Bot, S. D. M., de Boer, M. R., van der Windt, D. A. W. M., Knol, D. L., Dekker, J., Bouter, L. M., & de Vet, H. C. W. (2007). Quality criteria were proposed for measurement properties of health status questionnaires. *Journal of Clinical Epidemiology, 60*(1), 34-42. https://doi.org/10.1016/j.jclinepi.2006.03.012

Thompson, M., Elliott, C., Willis, C., Ward, R., Falkmer, M., Falkmer, T. r., Gubbay, A., & Girdler, S. (2016). Can, Want and Try: Parents' Viewpoints Regarding the Participation of Their Child with an Acquired Brain Injury [Report]. *PLoS ONE, 11*. https://doi.org/10.1371/journal.pone.0157951

Tipton-Burton, M. (2011). Jebsen–Taylor Hand Function Test. In J. S. Kreutzer, J. DeLuca, & B. Caplan (Eds.), *Encyclopedia of Clinical Neuropsychology* (pp. 1365-1365). Springer New York. https://doi.org/10.1007/978-0-387-79948-3_1946

Unsworth, C. (2004). Clinical reasoning: How do pragmatic reasoning, worldview and client-centredness fit? *British Journal of Occupational Therapy, 67*(1), 10-19. https://doi.org/10.1177/030802260406700103

Unsworth, C., & Baker, A. (2016). A systematic review of professional reasoning literature in occupational therapy. *British Journal of Occupational Therapy, 79*(1), 5-16. https://doi.org/doi:10.1177/0308022615599994

Urbaniak, G., & Plous, S. (2018). *Research Randomzer.* In https://www.randomizer.org/

Van Exel, J., & De Graaf, G. (2005). Q methodology: A sneak preview. https://www.researchgate.net/publication/228574836_Q_Methodology_A_Sneak_Preview

Van Lede, P., & Van Veldhoven, G. (1998). *Therapeutic hand splints: a rational approach. 1. Mechanical and biomechanical considerations*. Provan.

Vargus-Adams, J. (2009). Understanding function and other outcomes in cerebral palsy. *Physical medicine and rehabilitation clinics of North America, 20*(3), 567-575. https://doi.org/10.1016/j.pmr.2009.04.002

Vaz, D. V., Cotta, M., Fonseca, S. T., & De Melo Pertence, A. E. (2006). Muscle stiffness and strength and their relation to hand function in children with hemiplegic cerebral palsy. *Developmental Medicine & Child Neurology, 48*(9), 728-733. https://doi.org/10.1111/j.1469-8749.2006.tb01357.x

Veritas Health Innovation. (2013). *Covidence systematic review software*. www.covidence.org

von Elm, E., Altman, D.G., Egger, M., Pocock, S.J., Gotzsche, P.C., & Vandenbroucke, J.P. (2007). The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *Annals of Internal Medicine. 147*(8), 573-577. https://doi.org/10.1136/bmj.39335.541782.AD

Wagman, P., Hakansson, C., Jacobsson, C., Falkmer, T., & Bjorklund, A. (2012). What is considered important for life balance? Similarities and differences among some working adults. *Scandinavian Journal of Occupational Therapy, 19*(4), 377-384. https://doi.org/10.3109/11038128.2011.645552

Wallen, M., & Stewart, K. (2016). Grading and Quantification of Upper Extremity Function in Children with Spasticity. *Seminars in Plastic Surgery, 30*(1), 5-13. https://doi.org/10.1055/s-0035-1571257

Watts, S., & Stenner, P. (2005). Doing Q methodology: theory, method and interpretation. *Qualitative research in psychology, 2*(1), 67-91. https://doi.org/10.1191/1478088705qp022oa

Watts, S., & Stenner, P. (2012). *Doing Q methodological research: theory, method and interpretation*. SAGE Publications Ltd.

Webster, D., & Schwartzberg, S. L. (1993). Patients' Perception of Curative Factors in Occupational Therapy Groups. *Occupational Therapy in Mental Health, 12*(1), 3-24. https://doi.org/10.1300/J004v12n01_02

Westen, D., & Rosenthal, R. (2003). Quantifying construct validity: Two simple measures. *Journal of Personality and Social Psychology, 84*(3), 608-618. https://doi.org/10.1037//0022-3514.84.3.608

Wigger, U., & Mrtek, R. G. (1994). Use of Q-Technique to Examine Attitudes of Entering Pharmacy Students Toward Their Profession. *American Journal of Pharmaceutical Education, 58*(1), 8-15.

Wilby, H. J. (2007). The Importance of Maintaining a Focus on Performance Components in Occupational Therapy Practice. *British Journal of Occupational Therapy, 70*(3), 129-132. https://doi.org/10.1177/030802260707000306

Wilton, J. (1983). Prescription of Functional Orthosis for the Spastic Hand in Cerebral Palsy: An Assessment Profile. *Australian Occupational Therapy Journal, 30*(4), 137-147. https://doi.org/10.1111/j.1440-1630.1983.tb01439.x

Wilton, J. (2003). Casting, splinting, and physical and occupational therapy of hand deformity and dysfunction in cerebral palsy. *Hand Clinics, 19*(4), 573-584. https://doi.org/10.1016/S0749-0712(03)00044-1

Wilton, J. (2013a). *Hand splinting orthotic intervention: Principles of design and fabrication* (2nd ed.). Vivid Publishing.

Wilton, J. (2013b). Orthotic intervention and casting in the presence of neurological dysfunction. In *Hand Splinting Orthotic Intervention: Prinicples of design and fabrication* (2nd ed., pp. 195-235). Vivid Publishing.

World Health Organisation. (2001). *International Classification of Functioning, Disability and Health*. World Health Organization.

World Health Organisation. (2007). *International Classification of Functioning, Disability, and Health: Children & Youth Version: ICF-CY*. World Health Organization.

Yasukawa, A., & Cassar, M. (2009). Children With Elbow Extension Forearm Rotation Limitation: Functional Outcomes Using the Forearm Rotation Elbow Orthosis. *JPO: Journal of Prosthetics and Orthotics, 21*(3), 160-166. https://doi.org/10.1097/JPO.0b013e3181b16baf

Yasukawa, A., Lulinski, J., Thornton, L., & Jaudes, P. (2008). Improving elbow and wrist range of motion using a dynamic and static combination orthosis. *JPO: Journal of Prosthetics and Orthotics, 20*(2), 41-48. https://doi.org/10.1097/JPO.0b013e31816b1904

Yasukawa, A., Malas, B. S., & Gaebler-Spira, D. J. (2003). Efficacy for maintenance of elbow range of motion of two types of orthotic devices: a case series. *JPO: Journal of Prosthetics and Orthotics, 15*(2), 72-77. https://doi.org/10.1097/00008526-200304000-00007

Yin, R. K. (2014). *Case study research : design and methods* (5 edition.. ed.). Thousand Oaks, California : SAGE Publications.

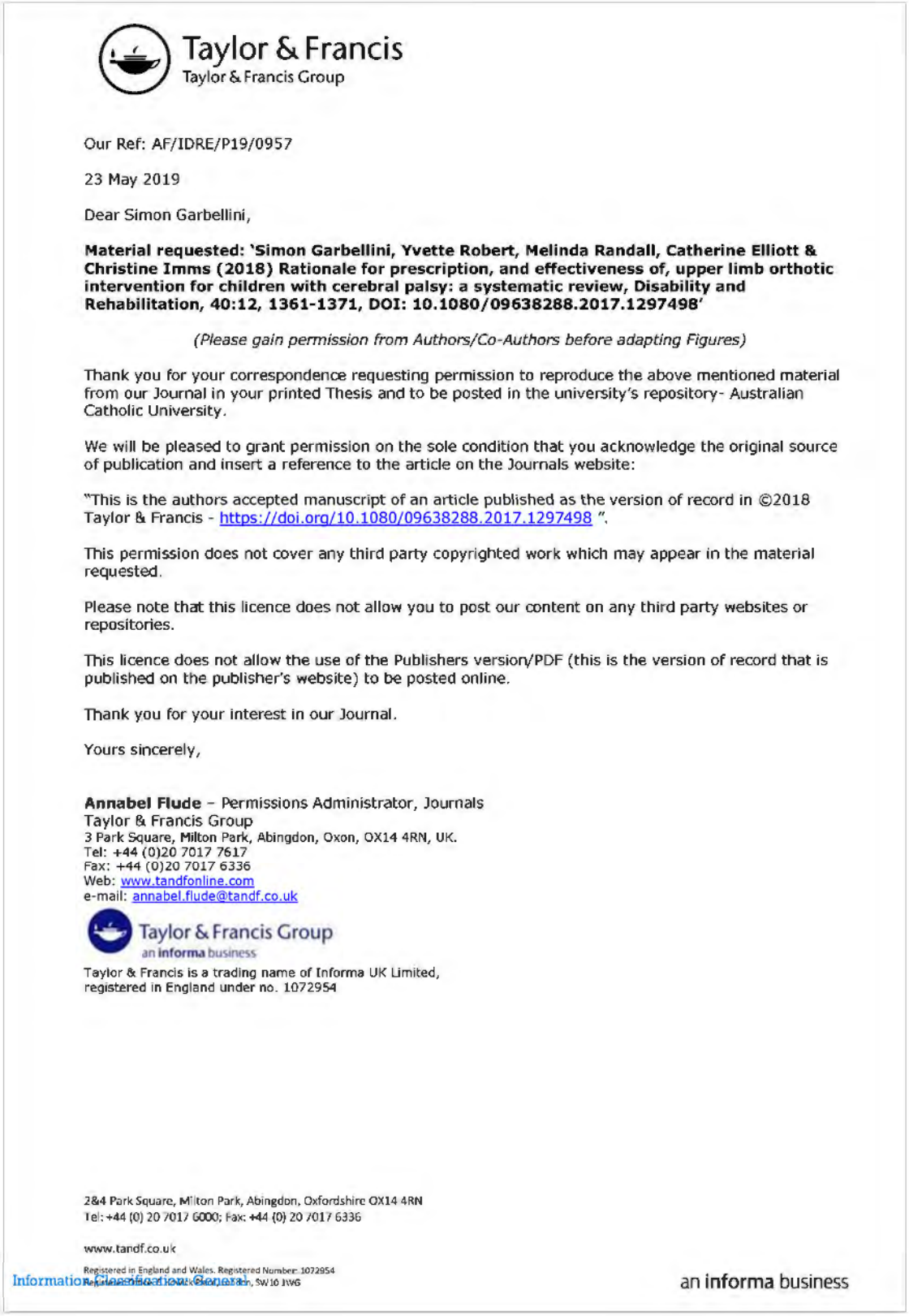
Zancolli, E. A. (2003). Surgical management of the hand in infantile spastic hemiplegia. *Hand clinics, 19*(4), 609-629. https://doi.org/10.1016/s0749-0712(03)00034-9

Zancolli, E. A., Goldner, L., & Swanson, A. (1983). Surgery of the spastic hand in cerebral palsy: Report of the Committee on Spastic Hand Evaluation. *Journal of Hand Surgery, 8*(5.2), 776 - 772. https://doi.org/10.1016/s0363-5023(83)80267-6

APPENDICES

Appendix A Approval to reproduce publications

Publication 1



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Publication 2

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Our Ref: AF/IDRE/P19/0959

23 May 2019

Dear Simon Garbellini,

Material requested: 'Simon Garbellini, Melinda Randall, Michael Steele, Catherine Elliott & Christine Imms (2019) Prescribing upper limb orthoses for children with cerebral palsy: a Q methodology study of occupational therapists' decision making, Disability and Rehabilitation, DOI: 10.1080/09638288.2019.1573931'

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Yours sincerely,

Annabel Flude – Permissions Administrator, Journals
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Publication 3

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Article (the "Article") entitled:	Unpacking the application of Q methodology for use in occupational therapy research
Article DOI:	10.1080/11038128.2019.1709542
Author(s):	Simon Garbellini, Melinda Randall, Michael Steele, Catherine Elliott, Christine Imms
To publish in the Journal:	Scandinavian Journal of Occupational Therapy
Journal ISSN:	1651-2014

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Signed and dated: **Simon Garbellini, 02 January 2020**

Publisher, 02 January 2020

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Appendix B Publications

B.1 Publication 1

DISABILITY AND REHABILITATION, 2017
http://dx.doi.org/10.1080/09638288.2017.1297498

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REVIEW ARTICLE

Rationale for prescription, and effectiveness of, upper limb orthotic intervention for children with cerebral palsy: a systematic review

Simon Garbellini[a,b], Yvette Robert[b], Melinda Randall[a], Catherine Elliott[b,c] and Christine Imms[a]

[a]Centre for Disability and Development Research, Australian Catholic University, Melbourne, VIC, Australia; [b]Department of Pediatric Rehabilitation, Princess Margaret Hospital, Perth, WA, Australia; [c]School of Occupational Therapy and Social Work, Curtin University, Perth, WA, Australia

ABSTRACT

Purpose: To explore (i) reasons for upper limb orthosis prescription for children with cerebral palsy (CP), (ii) the link between reason and effect according to intended outcome and outcome measure utilized and (iii) to classify the prescribed orthoses using standard terminology.

Method: A prospectively registered (center for reviews and dissemination: 42015022067) systematic review searched for experimental and observational studies investigating rigid/thermoplastic upper limb orthotic intervention for children aged 0–18 with CP. The Cochrane central register, MEDLINE, CINAHL, Embase, SCOPUS and Web of Science databases were searched. Included studies were assessed for risk of bias.

Results: Sixteen studies met selection criteria. Two studies described a specific reason for orthosis prescription, six prescribed orthoses to manage a clinical symptom and eight did not describe a reason. Eight studies were analyzed for effect according to intended outcome with no clear connection found between reasons for prescription, outcome measures utilized and effect reported.

Interpretation: The lack of evidence for upper limb orthotic intervention for children with CP leads to uncertainty when considering this treatment modality. Future research is needed to evaluate the effect of orthosis wear in relation to intended outcome utilizing robust methods and valid and reliable outcome measures.

ARTICLE HISTORY
Received 4 November 2016
Revised 6 February 2017
Accepted 16 February 2017

KEYWORDS
Upper limb; Orthoses; cerebral palsy; prescription; children

➤ IMPLICATIONS FOR REHABILITATION:

- Insufficient evidence exists about the reason for prescription of upper limb orthoses.
- The connection between reason for orthosis prescription, intended outcome, outcome measure utilized and observed effect is unclear.
- Recommend orthosis prescription to be accompanied by clear documentation of the aim of the orthosis and description using orthosis classification system terminology.
- Outcome measures consistent with the reason for orthosis prescription and intended outcome of the intervention are essential to measure effectiveness of the intervention.

Appendix C Prospero register for the systematic review (Publication 1)

UNIVERSITY of York
Centre for Reviews and Dissemination

NHS
National Institute for Health Research

PROSPERO International prospective register of systematic reviews

Prescription rationale and effectiveness of upper limb orthoses for children with cerebral palsy: a systematic review

Simon Garbellini, Christine Imms, Catherine Elliott, Melinda Randall, Yvette Robert

Citation

Simon Garbellini, Christine Imms, Catherine Elliott, Melinda Randall, Yvette Robert. Prescription rationale and effectiveness of upper limb orthoses for children with cerebral palsy: a systematic review. PROSPERO 2015:CRD42015022067 Available from http://www.crd.york.ac.uk/PROSPERO_REBRANDING/display_record.asp?ID=CRD42015022067

Review question(s)

What are the reported rationales for prescription of upper limb orthoses for children with cerebral palsy aged 0-18 years?

To what extent do the prescribed orthoses show evidence of effect according to their purpose?

Searches

Databases searched will include the Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE, CINAHL, EMBASE and Web of Science. Hand searches of reference lists of included studies will be conducted to ensure additional relevant references are identified.

There will be no limit placed on the publication dates of the included studies. Papers written in all languages will be assessed for inclusion if the abstract is in English. Full papers that require translation may be excluded due to a lack of resources. Where multiple publications reporting on the same study exist, data from the study will be extracted and reviewed only once.

Types of study to be included

Inclusion criteria:

1. The research design was a randomised controlled, quasi experimental study, or an observational study using a cohort or case-control or single case experimental study design.

Exclusion criteria:

1. The study design was qualitative or a systematic review. Although systematic reviews will be excluded, reference lists will be checked to ensure all primary research is located for inclusion.

Condition or domain being studied

Cerebral palsy

Participants/ population

Inclusion criteria:

1. Included participants are children aged 0 -18 years. In instances where a paper also includes participants older than 18 years, more than 50% of the participants must be between 0-18 years to be included;

2. Participants have a diagnosis of cerebral palsy. In instances where a paper also includes participants with other neurological diagnoses, such as acquired brain injury, more than 50% of the participants must have a diagnosis of CP to be included.

Exclusion criteria:

1. The participants had neuro-degenerative conditions.

Intervention(s), exposure(s)
Inclusion criteria:

1. The intervention included a rigid/thermoplastic orthosis applied to the upper limb (regardless of duration or dosage).

Exclusion criteria:

1. The orthotic intervention was designed to constrain the unaffected upper limb for the purpose of improving movement of the affected upper limb;

2. The orthotic intervention was a cast applied to the upper limb;

3. The orthosis used in the intervention was fully constructed of soft, non-rigid materials (e.g. neoprene, lycra);

Comparator(s)/ control
The intervention will be compared to any or no control/comparison groups.

Context
No limitations applied to context.

Outcome(s)
Primary outcomes
Outcome measures will not be used to determine inclusion of studies in this review. All outcomes are of interest including those measuring change in body function and structure, for example goniometric measurement of range of motion. Studies will not be excluded by non-inclusion of a particular outcome.

Both immediate and long term effects are of interest.

Secondary outcomes
None

Data extraction, (selection and coding)
Studies that meet the selection criteria will be reviewed for quality and data extraction. Two authors will independently extract data using a standardised data extraction form developed for the purpose of this review. Where differences of opinion exist between reviewers on data extracted, a third author will be consulted. In the case where data are not presented in the published study, raw data will be requested from the authors. Data extracted for each study will be tabulated.

Risk of bias (quality) assessment
All intervention studies will be assessed independently by two authors (SG, YR) using the Cochrane Collaboration's tool for assessing risk of bias (Higgins, Altman, & Sterne, 2011). The tool covers selection, performance, detection, attrition and reporting bias. It also allows for comment on other sources of bias not covered by the domains of the tool. Emphasis will be on assessing the risk of bias of the design and conduct of the studies not the quality of reporting.

The PEDro scale is an 11-item scale that will be utilised to assessment of methodological quality of randomised control trials. A study of reliability of the PEDro scale for rating quality of randomized

controlled trials was "fair" to "good" for the total PEDro score (Maher, Sherrington, Herbert, Moseley, & Elkins, 2003). Assessment of methodological quality of single-subject designs will be completed utilising the Single-Case Experimental Design (SCED) scale. The scale is a reliable and valid measure that delineates a minimum core set of criteria representing sound single-subject methodology. The SCED scale was designed to facilitate critical appraisal and reporting of single-subject methodology (Tate et al., 2008).

Strategy for data synthesis
A narrative synthesis is planned. The tabulated data will be analysed as sub-groups: purpose of the orthoses as related to the International Classification of Functioning, Disability and Health (ICF); Australian Hand Therapy Association (AHTA) orthosis schedule classification; clinical reasons of orthosis prescription; effect of orthosis and subtype of cerebral palsy. A comparison of the data will be discussed as it relates to orthosis prescription and its effectiveness in the management of upper limb impairment of children with cerebral palsy.

Analysis of subgroups or subsets
None planned

Dissemination plans
It is planned that the findings of this systematic review will be submitted to a peer reviewed journal for publication and presented at professional conferences.

Contact details for further information
Mr Garbellini

Department of Paediatric Rehabilitation

Princess Margaret Hospital

Level 2, 37 - 39 Hay Street

Subiaco, Western Australia, 6008

simon.garbellini@health.wa.gov.au

Organisational affiliation of the review
Australian Catholic University, Faculty of Health Sciences and Princess Margaret Hospital for Children Western Australia, Department of Paediatric Rehabilitation

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Review team
Mr Simon Garbellini, Australian Catholic University, Faculty of Health Sciences and Princess Margaret Hospital, Department of Paediatric Rehabilitation
Professor Christine Imms, Australian Catholic University, Faculty of Health Sciences
Professor Catherine Elliott, Child and Adolescent Health Services Western Australia and Curtin University, Faculty of Health Sciences
Dr Melinda Randall, Australian Catholic University, Faculty of Health Sciences
Ms Yvette Robert, Princess Margaret Hospital, Department of Paediatric Rehabilitation

Anticipated or actual start date
01 June 2015

Anticipated completion date
01 June 2016

Funding sources/sponsors
None

Conflicts of interest
None known

Language

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NHS
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English

Country
Australia

Subject index terms status
Subject indexing assigned by CRD

Subject index terms
Cerebral Palsy; Child; Humans; Orthotic Devices

Stage of review
Ongoing

Date of registration in PROSPERO
15 July 2015

Date of publication of this revision
15 July 2015

Stage of review at time of this submission	Started	Completed
Preliminary searches	Yes	No
Piloting of the study selection process	Yes	No
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

Appendix D Ethics Approvals

D.1 Australian Catholic University approvals (Study 2)

Fw: 2017-54E Ethics application approved!

Simon Garbellini <simon.garbellini@myacu.edu.au>
Thu 4/06/2020 8:59 AM
To: Garbellini, Simon <Simon.Garbellini@health.wa.gov.au>

From: Pratigya Pozniak <Pratigya.Pozniak@acu.edu.au> on behalf of Res Ethics <Res.Ethics@acu.edu.au>
Sent: Wednesday, 3 May 2017 8:25 AM
To: Christine Imms; Simon Garbellini
Cc: Res Ethics
Subject: 2017-54E Ethics application approved!

Dear Applicant,

Principal Investigator: Prof Christine Imms
Co-Investigator: Catherine Elliott, Dr Melinda Randall
Student Researcher: Simon Garbellini (HDR Student)
Ethics Register Number: 2017-54E
Project Title: Clinician's viewpoints on upper extremity orthosis prescription for children with cerebral palsy.
Date Approved: 03/05/2017
Ethics Clearance End Date: 31/12/2017

This is to certify that the above application has been reviewed by the Australian Catholic University Human Research Ethics Committee (ACU HREC). The application has been approved for the period given above.

Researchers are responsible for ensuring that all conditions of approval are adhered to, that they seek prior approval for any modifications and that they notify the HREC of any incidents or unexpected issues impacting on participants that arise in the course of their research. Researchers are also responsible for ensuring that they adhere to the requirements of the National Statement on Ethical Conduct in Human Research, the Australian Code for the Responsible Conduct of Research and the University¿s Code of Conduct.

Any queries relating to this application should be directed to the Ethics Secretariat (res.ethics@acu.edu.au). It is helpful if quote your ethics approval number in all communications with us.

If you require a formal approval certificate in addition to this email, please respond via reply email and one will be issued.

We wish you every success with your research.

Kind regards,

Kylie Pashley
on behalf of ACU HREC Chair, Dr Nadia Crittenden

Senior Research Ethics Officer | Research Services Office of the Deputy Vice Chancellor (Research) Australian Catholic University

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Fw: 2017-54E Extension approved

Simon Garbellini <simon.garbellini@myacu.edu.au>
Thu 4/06/2020 9:00 AM
To: Garbellini, Simon <Simon.Garbellini@health.wa.gov.au>

From: Ms Pratigya Pozniak <pratigya.pozniak@acu.edu.au>
Sent: Friday, 23 February 2018 12:07 PM
To: Professor Christine Imms; Simon Garbellini
Cc: Ms Pratigya Pozniak
Subject: 2017-54E Extension approved

Dear Christine

Ethics Register Number : 2017-54E
Project Title : Clinician's viewpoints on upper extremity orthosis prescription for children with cerebral palsy.
Data Collection Date Extended : 31/12/2018

Thank you for returning the Ethics Progress Report for your project.

The Deputy Chair of the Human Research Ethics Committee has approved your request to extend the project. The new expiry date for the project is the 31/12/2018 .

We wish you well in this ongoing project.

Kind regards,
Ms Pratigya Pozniak

Research Ethics Officer | Office of the Deputy Vice-Chancellor (Research)
Australian Catholic University
T: 02 9739 2646 E: res.ethics@acu.edu.au

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D.2 iWHOT approvals (Study 3)

Melinda Randall

Subject: FW: 2014 318V Registration of External Ethics Approval

-----Original Message-----
From: Kylie Pashley On Behalf Of Res Ethics
Sent: Monday, November 24, 2014 1:20 PM
To: Christine Imms
Cc: Res Ethics
Subject: 2014 318V Registration of External Ethics Approval

Dear Christine,

Principal Investigator: Prof Christine Imms
Co-Investigators: Dr Melinda Randall, Margaret Wallen, Catherine Elliot, Brian Hoare, Simon Garbellini, Francesca Orsini, Katherine Lee, Prof Rob Carter, Dr Brooke Adair, Dr Liz Bradshaw, Prof Dinah Reddihough, Susan Greaves
Ethics Register Number: 2014 318V Project Title: iWHOTrial: A multicentre randomised controlled trial of rigid wrist hand orthoses for young children with cerebral palsy.
Risk Level: Multi Site
Date Approved: 24/11/2014
Ethics Clearance End Date: 31/12/2018

The Australian Catholic University Human Research Ethics Committee has considered your application for registration of an externally approved ethics protocol and notes that this application has received ethics approval from Monash Health [SERP Reference: HREC/14/SHD/18, SSA Reference: SSA/14/SHB/21 and Monash Health HREC Ref: 14201B] and Cerebral Palsy Alliance [Reference 2014-08-02] with permissions to be provided from Princess Margaret Hospital (WA) and The Royal Children's Hospital (VIC).

The ACU HREC accepts the ethics approval with no additional requirements, save that ACU HREC is informed of any modifications of the research proposal and that copies of all progress reports and any other documents be forwarded to it. Any complaints involving ACU staff must also be notified to ACU HREC (National Statement 5.3.3)

We wish you well in this research project.

Regards,

Kylie Pashley
on behalf of ACU HREC Chair, Dr Nadia Crittenden Ethics Officer | Research Services Office of the Deputy Vice Chancellor (Research) res.ethics@acu.edu.au

1

Cerebral Palsy ALLIANCE

Professor Christine Imms
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy VIC 3067

4 September 2014

Dear Professor Imms

RE: "iWHOTrial: A Multicentre Randomised Controlled Trial of Rigid Wrist Hand Orthoses for Young Children with Cerebral Palsy"

Thank you for submitting the amended documents which we received 19 August 2014 following the conditional approval of your application of the above project. These amendments were tabled for consideration by Cerebral Palsy Alliance's Human Research Ethics Committee (HREC) at its meeting held on 3 September 2014.

Our HREC is constituted and operates in accordance with the *National Health and Medical Research Council's (NHMRC) National Statement on Ethical Conduct in Human Research (2007).*

I am pleased to inform you that your project meets the requirements of the *National Statement on Ethical Conduct In Human Research* and our Committee has granted final approval for this project

Details of the approval are as follows:

- **Project approval number:** 2014-08-02. Please use this number in all subsequent correspondence to the Committee.
- **Approval period:** September 2014 to September 2017
- **Authorised research personnel**:
  - Professor Christine Imms
  - Dr Margaret Wallen
  - Professor Catherine Elliott
  - Dr Brian Hoare
  - Dr Susan Greaves
  - Professor Rob Carter
  - Dr Melinda Randall
  - Dr Brooke Adair
  - Dr Elizabeth Bradshaw
  - Professor Dinah Reddihough
  - Dr Katherine Lee
  - Mr Simon Garbellini
  - Ms Francesca Orsini

187 Allambie Road, Allambie Heights, NSW 2100 | PO Box 184 Brookvale NSW 2100 | **ABN** 45 000 062 288
T 61 2 9975 8000 | **F** 61 2 9451 5209 | **E** info@cerebralpalsy.org.au | **W** cerebralpalsy.org.au

- **Approved documentation:**
  - Please attach a footer "This study has been approved by the Cerebral Palsy Alliance Human Research Ethics Committee. If you have any complaints or reservations about the ethical conduct of this research you may contact the Ethics Committee on (02) 9975 8000 or ethics@cerebralpalsy.org.au" to the information statements and consent forms, labelling this version 1. Please send a copy of the final updated documents to the Ethics Committee. If you wish to change these in the future please send a copy to the Ethics Committee for review.

Cerebral Palsy Alliance's Human Research Ethics Committee (HREC) is a fully constituted Ethics Committee in accordance with the National Statement on Ethical Conduct in Research Involving Humans 2007. The approval of this project is conditional upon your continuing compliance with the National Statement.

Accordingly, it is the responsibility of the chief investigator/s to:

- **Provide a summary of your progress on a yearly basis to the Committee commencing September 2015. A final report on completion and notification of any publications from this project is also requested. Failure to submit required reports will result in a suspension of consent for the project to continue.**
- Advise the HREC immediately in writing of any serious adverse events occurring during the course of the research.
- Advise the HREC immediately of all unforeseen events that might affect continued ethical acceptability of the project.
- Advise the HREC of any proposed changes to the research protocol, research personnel, information statement or consent form. All proposed amendments must be addressed in writing to the HREC and must be approved by the HREC before continuation of the project.
- Advise the HREC immediately, providing reasons, if the research is discontinued prior to its completion.
- Request an extension of ethics approval should the project not be completed within the time period specified above.
- Ensure that copies of all signed consent forms are retained and made available to the HREC on request.
- Provide a copy of this letter to any internal/external granting agencies if requested.

The Ethics Committee and Board of Directors wish you well with this important project.

Yours sincerely

[redacted]

Deborah Hoffman, on behalf of
Dr Neroli Best
Chair, Ethics Committee
Corebral Palsy Alliance Ethics Committee is a NHMRC HREC: EC00402

Human Research Ethics

Deakin Research Integrity
Burwood Campus
Postal: 221 Burwood Highway
Burwood Victoria 3125 Australia
Telephone 03 9251 7123
research-ethics@deakin.edu.au

Memorandum

To: Prof Rob Carter
School of Health & Social Development

B

cc: Dr Sophy Ting-Fang Shih

From: Deakin University Human Research Ethics Committee (DUHREC)

Date: 21 March, 2016

Subject: 2016-071

iWHOTrial: A Multicentre Randomised Controlled Trial of Rigid Wrist Hand Orthoses for Young Children with Cerebral Palsy

Please quote this project number in all future communications

Approval granted by Monash Health HREC for this project will be noted at the DUHREC meeting to be held on 18 April 2016.

It will be noted that approval has been granted for Prof Rob Carter, School of Health & Social Development, to undertake this project as stipulated in Monash Health HREC approval documentation.

The approval noted by the Deakin University Human Research Ethics Committee is given only for the project and for the period as stated in the memo. It is your responsibility to contact the Human Research Ethics Unit immediately should any of the following occur:

- Serious or unexpected adverse effects on the participants
- Any proposed changes in the protocol, including extensions of time.
- Any events which might affect the continuing ethical acceptability of the project.
- The project is discontinued before the expected date of completion.
- Modifications are requested by other HRECs.

In addition you will be required to report on the progress of your project at least once every year and at the conclusion of the project. Failure to report as required will result in suspension of your approval to proceed with the project.

DUHREC may need to audit this project as part of the requirements for monitoring set out in the National Statement on Ethical Conduct in Human Research (2007).

Human Research Ethics Unit
research-ethics@deakin.edu.au
Telephone: 03 9251 7123

MonashHealth

Research Directorate
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168
Australia

Postal address:
Locked Bag 29
Clayton South Vic 3169
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

9 October 2014

Prof Christine Imms
School of Allied Health
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy Vic 3067

Dear Prof Imms

Study title: iWHOTrial (Infant Wrist Hand Orthoses Trial): A Multicentre Randomised Controlled Trial of Rigid Wrist Hand Orthoses for Young Children with Cerebral Palsy

SERP Reference Number: HREC/14/SHB/18
SSA Reference Number: SSA/14/SHB/21
Monash Health HREC Ref: 14201B
Protocol number: iWHOTrial

Thank you for submitting a Site Specific Assessment Form for authorisation of the above project at Monash Health.

I am pleased to inform you that authorisation has been granted for this project to be conducted at the Monash Medical Centre Clayton campus of Monash Health.

The following conditions apply to this research project at your site. These conditions are additional to those imposed by the Human Research Ethics Committee that granted ethical approval:

The Principal Investigator is required to notify the Research Directorate, Monash Health of the following:

1. Any change in protocol and the reason for that change together with an indication of ethical implications (if any)
2. Serious or unexpected adverse effects of project on subjects and steps taken to deal with them
3. Any unforeseen events that might affect continued ethical acceptability of the project
4. Any expiry of the insurance coverage provided in respect of sponsored trials
5. Discontinuation of the project before the expected date of completion, giving reasons
6. Any change in personnel involved in the research project including any study member resigning from Monash Health &/or the study team.

At the conclusion of the project or every twelve months if the project continues, the Principal Investigator is required to complete and forward an annual report to the Committee.

List of Approved Documents:

Document	*Version*	*Date*
Monash Health Parent/Guardian Information and Consent Form	4.0	11 September 2014
Advertisement	2.0	18 July 2014

If you should have any queries about your project please contact Deborah Dell or Julie Gephart by email deborah.dell@monashhealth.org /julie.gephart@monashhealth.org

The HREC wishes you and your colleagues every success in your research.

Yours sincerely

[redacted]

Dr James Doery
Acting Chair, HREC A

Attachments: CTRA (x3)

Cc: Dr Brian Hoare Victorian Paediatric Rehabilitation Service Monash Children's Monash Medical Centre Clayton Vic 3168

Checklist: Post-ethics approval requirements that must be met before a research project can commence at a study site.

Please ensure that as a PI (including the CPI) the following are completed at each study site.

Requirements	**Yes/No/NA**
CTN Acknowledgement The PI must forward a copy of the CTN Acknowledgement to the Research Directorate	N/A
Clinical Trial Research Agreement The PI must forward an original fully executed copy of the CTRA to the Research Directorate.	Yes
Indemnity The PI must forward an original fully executed copy of the Indemnity to the Research Directorate.	N/A
Radiation If applicable, the RGO must contact the Medical Physicist to notify DHS, Radiation Safety Section to list the project on the Institute's licence.	N/A
Other Commonwealth statutory requirements Ensure compliance with the following e.g. Office of the Gene Technology Regulator, NHMRC Licensing Committee, NHMRC Cellular Therapies Advisory Committee.	N/A

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168
Australia

Postal address:
Locked Bag 29
Clayton South Vic 3169
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

19 September 2014

Prof Christine Imms
School of Allied Health
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy Vic 3067

Dear Prof Imms

Study title: iWHOTrial (Infant Wrist Hand Orthoses Trial): A Multicentre Randomised Controlled Trial of Rigid Wrist Hand Orthoses for Young Children with Cerebral Palsy
SERP Reference Number: HREC/14/SHB/18
Monash Health HREC Ref: 14201B
Protocol number: iWHOTrial

The Monash Health HREC B reviewed the above application at the meeting held on 19 June 2014 In addition, the HREC is satisfied that the responses to our correspondence of 25 June 2014 have been sufficiently addressed.

The HREC approved the above application on the basis of the information provided in the application form, protocol and supporting documentation.

This reviewing HREC is accredited by the Consultative Council for Clinical Trial Research under the single ethical review system.

Approval

The HREC approval is from 19 September 2014.

Approval is given in accordance with the research conforming to the *National Health and Medical Research Council Act 1992* and the *National Statement on Ethical Conduct in Human Research (2007)*. The HREC has ethically approved this research according to the Memorandum of Understanding between the Consultative Council and the participating organisations conducting the research.

Approval is given for this research project to be conducted at the following sites and campuses:
- Monash Medical Centre Clayton, Monash Health
- Royal Children's Hospital Melbourne

You must comply with the following conditions:

The Chief Principal Investigator is required to notify the Manager, Human Research Ethics Committees, Monash Health of:

1. Any change in protocol and the reason for that change together with an indication of ethical implications (if any)
2. Serious or unexpected adverse effects of project on subjects and steps taken to deal with them
3. Any unforeseen events that might affect continued ethical acceptability of the project

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168
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Tel (03) 9594 4611
Fax (03) 9594 6306

4. Any expiry of the insurance coverage provided in respect of sponsored trials
5. Discontinuation of the project before the expected date of completion, giving reasons
6. Any change in personnel involved in the research project including any study member resigning from Monash Health &/or the study team.

At the conclusion of the project or every twelve months if the project continues, the Principal Investigator is required to complete and forward an annual progress report to the Committee.

Reminders to submit annual progress report forms will be forwarded to the researcher.

The Coordinating Principal Investigator is responsible for notifying Principal Investigators. The Coordinating Principal Investigator and Principal Investigators should forward a copy of this letter to their site's Research Governance Officer.

Approved documents

Documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Protocol	3.0	13 August 2014
National Ethics Application Form	2.2	2014
Victorian Specific Module	2	25 August 2014
Advertisement	2.0	7 July 2014
Recruitment Letter	2.0	18 July 2014
Therapist Information Sheet	3.0	26 August 2014
Parent/Guardian Information and Consent Form	3.0	18 July 2014

Site-Specific Assessment (SSA)

SSA authorisation is required at all sites participating in the study. SSA must be authorised at a site before the research project can commence.

The completed Site-Specific Assessment Form and a copy of this ethics approval letter must be submitted to the Research Governance Officer for authorisation by the Chief Executive or delegate. This applies to each site participating in the research.

If you should have any queries about your project please contact Deborah Dell or Julie Gephart by email deborah.dell@monashhealth.org /julie.gephart@monashhealth.org

The HREC wishes you and your colleagues every success in your research.

Yours sincerely

[redacted]

Dr Simon Bower
Chair, HREC B

Cc: Dr Brian Hoare

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168
Australia

Postal address:
Locked Bag 29
Clayton South Vic 3169
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

Checklist: Post-ethics approval requirements that must be met before a research project can commence at a study site.

Please ensure that as a PI (including the CPI) the following are completed at each study site.

Requirements	**Yes/No/NA**
Ethics approval notification The PI must send a copy to the RGO at that study site.	Yes
HREC Review Only Indemnity The PI must forward a copy of the signed HREC Review Only Indemnity to the RGO at that study site.	N/A
CTN notification The PI must sign the CTN and forward to the RGO so the authority approving the conduct of the trial, at that site, can complete and sign.	N/A
SSA authorisation notification The PI must forward the SSA form and attached documents (e.g. CTRA) to the RGO so the authority approving the conduct of the trial, at that site, can complete and sign.	Yes
Radiation If applicable, the RGO must contact the Medical Physicist to notify DHS, Radiation Safety Section to list the project on the Institute's licence.	N/A
Other Commonwealth statutory requirements Ensure compliance with the following e.g. Office of the Gene Technology Regulator, NHMRC Licensing Committee, NHMRC Cellular Therapies Advisory Committee.	N/A

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC REF **2014061EP**
STUDY TITLE **iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist and Orthoses for Young Children with CereorarPalsy.**

The ethics application for the project referenced above was reviewed by the PMH Human Research Ethics Committee (HREC) at its meeting on 21/08/2014. It has been approved and the following documents have been approved for use in this project.

National Ethics Application Form
Scientific Protocol Form 4B
Project Protocol Version 3 dated 13 August 2014
Parent/Guardian Information Sheet Version 4 Dated 11 September 2014
Study Specific Questionnaire: Baseline Version 3 dated 19 August 2014
Study Specific Questionnaire: 12/24 Month Assessment Version 3 dated 19 August 2014
Study Specific Questionnaire: 6/18/30 Month Assessment Version 3 dated 19 August 2014
Study Specific Questionnaire: Study Completion Version 3 dated 19 August 2014
Orthosis questionnaire for children in the treatment group Version 3 Dated 19 August 2014
Confidentiality Agreements
Conflict of Interest Forms

Approval of this project from PMH HREC is valid to 21/08/2017 and on the basis of compliance with the 'Conditions of HREC Approval for a Research Project' (attached).

Note: If additional sites are recruited prior to the commencement of, or during the research project, the Coordinating Principal Investigator is required to notify the HREC. Notification of withdrawn sites should also be provided to the HREC in a timely fashion.

A copy of this ethical approval letter must be submitted by all site Principal Investigators to the Research Governance Office or equivalent body or individual at each participating institution in a timely manner to enable the institution to authorise the commencement of the project at its site/s.

This letter constitutes ethical approval only.

This project cannot proceed at any site until separate site authorisation has been obtained from the CE, or delegate, of the site under whose auspices the research will be conducted at that site.

The PMH HREC is registered with the Australian Health Ethics Committee and operates according to the NHMRC National Statement on Ethical Conduct in Human Research and International Conference on Harmonisation – Good Clinical Practice.

The HREC's Terms of Reference, Standard Operating Procedures, membership and standard forms are available from http://www.pmh.health.wa.gov.au/development/resources/ethics.htm or from the Ethics Office. Should you have any queries about the HREC's consideration of your project, please contact Ethics Office.

Please quote the above trial number 2014061EP on all correspondence associated with this trial.

Yours sincerely

Dr Mark Salmon
Executive Director
Medical Services

24/10/2014

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service
Research Governance Office

Our Ref: 2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HREC REF **2014061EP**
STUDY TITLE **iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist and Orthoses for Young Children with CereorarPalsy.**

On behalf of the Child and Adolescent Health Service, I give authorisation for your research project to be conducted at the following site(s):

Princess Margaret Hospital for Children - CAHS

This authorisation is based on the approval from PMH HREC and the review from the Research Governance Office. This authorisation is valid subject to the ongoing approval from the HREC.

This authorisation is based on the ethical approval from the HREC, and on the basis of compliance with the 'Conditions of Authorisation to Conduct a Research Project at Site' (attached) and with the compliance of all reports as required by the Research Governance Office and approving HREC. Non compliance with these requirements could result in the authorisation being withdrawn.

The responsibility for the conduct of this project remains with you as the Principal Investigator at the site.

Yours sincerely

[redacted]

Dr Mark Salmon
Executive Director
Medical Services

27/11/2014

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC REF **2014061EP**
STUDY TITLE **iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist and Orthoses for Young Children with CereorarPalsy.**

The ethics application for the project referenced above was reviewed by the PMH Human Research Ethics Committee (HREC) at its meeting on 21/08/2014. It has been approved and the following documents have been approved for use in this project.

National Ethics Application Form
Scientific Protocol Form 4B
Project Protocol Version 3 dated 13 August 2014
Parent/Guardian Information Sheet Version 4 Dated 11 September 2014
Study Specific Questionnaire: Baseline Version 3 dated 19 August 2014
Study Specific Questionnaire: 12/24 Month Assessment Version 3 dated 19 August 2014
Study Specific Questionnaire: 6/18/30 Month Assessment Version 3 dated 19 August 2014
Study Specific Questionnaire: Study Completion Version 3 dated 19 August 2014
Orthosis questionnaire for children in the treatment group Version 3 Dated 19 August 2014
Confidentiality Agreements
Conflict of Interest Forms

Approval of this project from PMH HREC is valid to 21/08/2017 and on the basis of compliance with the 'Conditions of HREC Approval for a Research Project' (attached).

Note: If additional sites are recruited prior to the commencement of, or during the research project, the Coordinating Principal Investigator is required to notify the HREC. Notification of withdrawn sites should also be provided to the HREC in a timely fashion.

A copy of this ethical approval letter must be submitted by all site Principal Investigators to the Research Governance Office or equivalent body or individual at each participating institution in a timely manner to enable the institution to authorise the commencement of the project at its site/s.

This letter constitutes ethical approval only.

This project cannot proceed at any site until separate site authorisation has been obtained from the CE, or delegate, of the site under whose auspices the research will be conducted at that site.

The PMH HREC is registered with the Australian Health Ethics Committee and operates according to the NHMRC National Statement on Ethical Conduct in Human Research and International Conference on Harmonisation – Good Clinical Practice.

The HREC's Terms of Reference, Standard Operating Procedures, membership and standard forms are available from http://www.pmh.health.wa.gov.au/development/resources/ethics.htm or from the Ethics Office. Should you have any queries about the HREC's consideration of your project, please contact Ethics Office.

Please quote the above trial number 2014061EP on all correspondence associated with this trial.

Yours sincerely

[redacted]

Dr Mark Salmon
Executive Director
Medical Services

24/10/2014

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

Government of **Western Australia**
Child and Adolescent Health Service

Our Ref: 2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HREC Ref: 2014061EP
Project Title: iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist hand Orthoses for Young Children with Cerebral Palsy

On behalf of the Child and Adolescent Health Service (CAHS), I give conditional authorisation for the above research project to be conducted at Perth Children's Hospital following final move day from Princess Margaret Hospital.

This approval is conditional upon finalisation of the relevant deed of variation to the research agreement for this study after the inclusion of the final move date (once announced).

At the time of issuing this letter the following documents are approved for use at PCH:

- Recruitment letter PCH v1 dated 1 August 2016
- PICF Parent Addendum PCH v1 dated 22 July 2016
- PICF Therapist Addendum PCH v1 dated
- PICF Parent PCH v1 dated 22 July 2016
- PICF Therapist PCH v1 dated 8 August 2016

Once the final move day has been confirmed, please insert the date into the Addendum to Consent (if relevant). When available, please also update any information sheet/consent forms with relevant extension numbers at Perth Children's Hospital to enable direct contact with the study team. Prior to commencing your research at PCH, please confirm TGA acknowledgment of site change details for the Clinical Trial Notification (if applicable). It is the responsibility of the study sponsor to notify the TGA of the change of site.

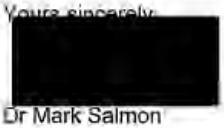
Any document changes required, other than those stated above, after the issuing of this letter (date of signature) will need to be submitted for review via the standard process for review of amendments.

This authorisation is based on the completion of the transition review conducted by the CAHS Research Governance Office and is valid subject to the ongoing approval from the approving Human Research Ethics Committee (HREC).

This authorisation is based on compliance with the 'Conditions of Authorisation to Conduct a Research Project at Site' (attached) and with the compliance of all reports as required by the CAHS Research Governance Office and approving HREC. Non-compliance with these requirements could result in the authorisation being withdrawn.

The responsibility for the conduct of this project remains with you as the Principal Investigator at the site.

Yours sincerely



Dr Mark Salmon
Director Clinical Services

pp

9 November 2017

CC: Dr Catherine Elliott
Roslyn Ward

The Royal Children's Hospital Melbourne
50 Flemington Road
Parkville Victoria 3052 Australia
TELEPHONE +61 3 9345 5522
www.rch.org.au

The Royal Children's Hospital Melbourne

24th April 2015

Dr Sue Greaves
Principal Investigator
Occupational Therapy
RCH

Dear Dr Greaves

Study title: iWHOTrial (Infant Wrist Hand Orthoses Trial): a multicentre randomised controlled trial of rigid wrist hand orthoses for young children with cerebral palsy

HREC Reference Number: HREC/14/SHB/18
RCH HREC Reference: 34279A
SSA Reference Number: SSA/14/RCHM/59

Thank you for submitting a Research Governance Only Application for authorisation of the above project at The Royal Children's Hospital, Parkville Victoria. I can confirm that the submission was received on 08 December 2014.

I am pleased to inform you that authorisation has been granted for this project to be conducted at The Royal Children's Hospital, Melbourne. **Please note this authorisation is only valid while approval from the reviewing HREC is current.**

Documents
Protocol v3 dated 13 Aug 2014
Parent/Guardian Information & Consent Form (RCH) v5.0 dated 25 Mar 2015
Therapist Information & Consent Form (RCH) v4.0 dated 25 Mar 2015
Recruitment Letter (RCH) v2.0 dated 18 Jul 2014
Advertising Flyer (RCH) v2.0 dated 18 Jul 2014

Please note the following conditions of approval:

1. Researchers must comply with the Investigator's Responsibilities in Research Procedure and Good Clinical Practice (ICH GCP)
2. Any proposed change in the protocol (or approved documents) or the addition of documents must be submitted to the Reviewing Human Research Ethics Committee (RHREC) for approval prior to implementation. The PI must also submit a copy of all documents relating to approved amendments to RCH Ethics Office for governance approval.
3. The Principal Investigator must notify the 1) CPI, 2) RCH Ethics Office and sponsor (if applicable) of:
   i. All internal (occurring in RCH participants) Serious Adverse Events (SAE) within 72 hours of occurrence.
   ii. Any other serious adverse effects to or complaints from RCH participants and steps taken to deal with them.
   iii. RCH Investigators withdrawing from or joining the project.
   NOTE: All SUSARs must be submitted to the TGA (as per the CTN guidelines).

The Royal Children's Hospital Melbourne
50 Flemington Road
Parkville Victoria 3052 Australia
TELEPHONE +61 3 9345 5522
www.rch.org.au

The Royal **Children's** Hospital Melbourne

4. A progress report must be submitted to RCH Ethics Office annually and at the conclusion of the project. A copy of this report should also be provided to the CPI.
5. RHREC approval must remain current for the entire duration of the project. Investigators undertaking projects without current RHREC approval risk their indemnity, funding and publication rights. Evidence of RHREC renewal approval will be required.
6. The PI must ensure that:
   - A fully executed CTN has been sent to the TGA prior to commencement and must place a copy of the TGA acknowledgment letter in the study file when received (2 weeks following submission).
   - The Clinical Trial Research Agreement and Indemnities are fully executed (i.e. signed by all parties) prior to commencement of the study.

If you have any matters that arise regarding conduct of the research at this site, please ensure you contact the Research Ethics and Governance office at RCH, Melbourne.

I wish you and your colleagues every success in your research.

Yours sincerely

[redacted]

Alexandra Robertson
Director
Research Ethics and Governance

Phone : (03) 9345 5044
Email : rch.ethics@rch.org.au
Web : www.rch.org.au

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 2522/2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

RE: 2014061EP - AMENDMENT OF TRIAL APPROVAL

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC Ref	**2014061EP**
Study Expiry Date	**24/10/2017**
Study Title	**iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist and Orthoses for Young Children with Cerebral Palsy**
Approval Date	**23/02/2016**

Thank you for your letter received by this office on 15/02/2016 enclosing the following amendment:

Protocol iWHO Trial Version 7.0 dated 21January2016
Parent/Guardian Information and Consent Form Version 4.3 dated 21January2016
Case reporting form Version 8 21January2016
Advertisement Version 3.0 Date: 21January2016
Recruitment Letter Version 2.2 dated 02 February 2016
Therapist Information and Consent Form Version 2.2 dated 02Februarv2016

The PMH HREC reviewed your request for the abovementioned amendment at its meeting and recommended the amendment for approval.

It should be noted that all other aspects of the approval remain unchanged. Particularly in relation to the progress reports required, as in National Statement S5.5 & S5.7.1, and any further amendments to the protocols.

Please quote the above trial number 2014061EP on all correspondence associated with this trial.

Yours sincerely

[redacted]

Dr Mark Salmon
Director Clinical Services

08/06/2016

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 1507/2014061EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

RE: 2014061EP - AMENDMENT OF TRIAL APPROVAL

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC Ref	**2014061EP**
Study Expiry Date	**24/10/2017**
Study Title	**iWHOTrial:A Multicentre Randomised Controlled Trial of Rigid Wrist and Orthoses for Young Children with Cerebral Palsy.**
Approval Date	**14/07/2015**

Thank you for your letter received by this office on 22/06/2015 enclosing the following amendment:

Protocol Version 6.0 dated 23 June 2015
Parent Guardian Information and Consent form Version 4.2 dated 23 June 2015
Parent Guardian Sensor Information Supplement Version 1.0 dated 23 June 2015

The PMH HREC reviewed your request for the abovementioned amendment at its meeting and recommended the amendment for approval.

It should be noted that all other aspects of the approval remain unchanged. Particularly in relation to the progress reports required, as in National Statement S5.5 & S5.7.1, and any further amendments to the protocols.

Please quote the above trial number 2014061EP on all correspondence associated with this trial.

Yours sincerely

[redacted]

Dr Mark Salmon
Executive Director
Medical Services

20/07/2015

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

D.3 MiT approvals (Study 3)

Melinda Randall

Subject: FW: 2014 317V Registration of External Ethics Approval

-----Original Message-----
From: Kylie Pashley On Behalf Of Res Ethics
Sent: Monday, November 24, 2014 1:34 PM
To: Christine Imms
Cc: Res Ethics
Subject: 2014 317V Registration of External Ethics Approval

Dear Christine,

Principal Investigator: Prof Christine Imms
Co-Investigators: Dr Melinda Randall, Prof Rob Carter, Dr Brooke Adair, Dr Liz Bradshaw, Prof Dinah Reddihough, Margaret Wallen, Catherine Elliot, Brian Hoare, Simon Garbellini, Francesca Orsini, Katherine Lee, Susan Greaves Ethics Register Number: 2014 317V Project Title: Minimising Impairment Trial: A multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy Risk Level: Multi Site Date Approved: 24/11/2014 Ethics Clearance End Date: 31/12/2018

The Australian Catholic University Human Research Ethics Committee has considered your application for registration of an externally approved ethics protocol and notes that this application has received ethics approval from Monash Health [Monash Health Reference: 14199B, SERP Reference Number: HREC/14/SHB/17, Protocol number MITrial] and Cerebral Palsy Alliance [Reference: 2014-08-03].

The ACU HREC accepts the ethics approval with no additional requirements, save that ACU HREC is informed of any modifications of the research proposal and that copies of all progress reports and any other documents be forwarded to it. Any complaints involving ACU staff must also be notified to ACU HREC (National Statement 5.3.3)

We wish you well in this research project.

Regards,

Kylie Pashley
on behalf of ACU HREC Chair, Dr Nadia Crittenden Ethics Officer | Research Services Office of the Deputy Vice Chancellor (Research) res.ethics@acu.edu.au

1

Cerebral Palsy ALLIANCE
Formerly known as The Spastic Centre

Professor Christine Imms
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy VIC 3067

4 September 2014

Dear Professor Imms

RE: "Minimising impairment: A multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy"

Thank you for submitting the amended documents which we received 19 August 2014 following the conditional approval of your application of the above project. These amendments were tabled for consideration by Cerebral Palsy Alliance's Human Research Ethics Committee (HREC) at its meeting held on 3 September 2014.

Our HREC is constituted and operates in accordance with the *National Health and Medical Research Council's (NHMRC) National Statement on Ethical Conduct in Human Research (2007).*

I am pleased to inform you that your project meets the requirements of the *National Statement on Ethical Conduct in Human Research* and our Committee has granted final approval for this project

Details of the approval are as follows:

- **Project approval number:** 2014-08-03. Please use this number in all subsequent correspondence to the Committee.
- **Approval period:** September 2014 to September 2017
- **Authorised research personnel**:
  - Professor Christine Imms
  - Dr Margaret Wallen
  - Professor Catherine Elliott
  - Dr Brian Hoare
  - Dr Susan Greaves
  - Professor Rob Carter
  - Dr Melinda Randall
  - Dr Brooke Adair
  - Dr Elizabeth Bradshaw
  - Professor Dinah Reddihough
  - Dr Katherine Lee
  - Mr Simon Garbellini
  - Ms Francesca Orsini

187 Allambie Road, Allambie Heights, NSW 2100 | PO Box 184 Brookvale NSW 2100 | **ABN** 45 000 062 288
T 61 2 9975 8000 | **F** 61 2 9451 5209 | **E** info@cerebralpalsy.org.au | **W** cerebralpalsy.org.au

- **Approved documentation:**
  - Please attach a footer "This study has been approved by the Cerebral Palsy Alliance Human Research Ethics Committee. If you have any complaints or reservations about the ethical conduct of this research you may contact the Ethics Committee on (02) 9975 8000 or ethics@cerebralpalsy.org.au" to the information statements and consent forms, labelling this version 1. Please send a copy of the final updated documents to the Ethics Committee. If you wish to change these in the future please send a copy to the Ethics Committee for review.

Cerebral Palsy Alliance's Human Research Ethics Committee (HREC) is a fully constituted Ethics Committee in accordance with the National Statement on Ethical Conduct in Research Involving Humans 2007. The approval of this project is conditional upon your continuing compliance with the National Statement.

Accordingly, it is the responsibility of the chief investigator/s to:

- **Provide a summary of your progress on a yearly basis to the Committee commencing September 2015. A final report on completion and notification of any publications from this project is also requested. Failure to submit required reports will result in a suspension of consent for the project to continue.**
- Advise the HREC immediately in writing of any serious adverse events occurring during the course of the research.
- Advise the HREC immediately of all unforeseen events that might affect continued ethical acceptability of the project.
- Advise the HREC of any proposed changes to the research protocol, research personnel, information statement or consent form. All proposed amendments must be addressed in writing to the HREC and must be approved by the HREC before continuation of the project.
- Advise the HREC immediately, providing reasons, if the research is discontinued prior to its completion.
- Request an extension of ethics approval should the project not be completed within the time period specified above.
- Ensure that copies of all signed consent forms are retained and made available to the HREC on request.
- Provide a copy of this letter to any internal/external granting agencies if requested.

The Ethics Committee and Board of Directors wish you well with this important project.

Yours sincerely

[redacted]

Deborah Hoffman, on behalf of
Dr Neroli Best
Chair, Ethics Committee
Cerebral Palsy Alliance Ethics Committee is a NHMRC HREC: EC00402

Human Research Ethics

Deakin Research Integrity
Burwood Campus Victoria
Postal: 221 Burwood Highway
Burwood Victoria 3125 Australia
Telephone 03 9251 7123
research-ethics@deakin.edu.au

Memorandum

To: Prof Rob Carter

School of Health & Social Development

B

cc: Ms Utsana Tonmukayakul

From: Deakin University Human Research Ethics Committee (DUHREC)

Date: 18 July, 2016

Subject: 2016-231

Minimising impairment: Multicentre Randomised Controlled Trial of Upper Limb Orthoses for Children with Cerebral Palsy

Please quote this project number in all future communications

Approval granted by Monash Health HREC for this project will be noted at the DUHREC meeting to be held on 25 July 2016.

It will be noted that approval has been granted for Prof Rob Carter, School of Health & Social Development, to undertake this project as stipulated in Monash Health HREC approval documentation.

The approval noted by the Deakin University Human Research Ethics Committee is given only for the project and for the period as stated in the memo. It is your responsibility to contact the Human Research Ethics Unit immediately should any of the following occur:

- Serious or unexpected adverse effects on the participants
- Any proposed changes in the protocol, including extensions of time.
- Any events which might affect the continuing ethical acceptability of the project.
- The project is discontinued before the expected date of completion.
- Modifications are requested by other HRECs.

In addition you will be required to report on the progress of your project at least once every year and at the conclusion of the project. Failure to report as required will result in suspension of your approval to proceed with the project.

DUHREC may need to audit this project as part of the requirements for monitoring set out in the National Statement on Ethical Conduct in Human Research (2007).

Human Research Ethics Unit
research-ethics@deakin.edu.au
Telephone: 03 9251 7123

MonashHealth

Research Directorate
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168
Australia

Postal address:
Locked Bag 29
Clayton South Vic 3169
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

9 October 2014

Prof Christine Imms
School of Allied Health
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy Vic 3067

Dear Prof Imms

Study title: Minimising Impairment: Multicentre Randomised Controlled Trial of Upper Limb Orthoses for Children with Cerebral Palsy
Monash Health HREC Ref: 14199B
SERP Reference Number: HREC/14/SHB/17
Monash Health HREC Ref: 14199B
SSA Reference Number: SSA/14/SHB/20
Protocol number: MITrial

Thank you for submitting a Site Specific Assessment Form for authorisation of the above project at Monash Health.

I am pleased to inform you that authorisation has been granted for this project to be conducted at the Monash Medical Centre Clayton campus of Monash Health.

The following conditions apply to this research project at your site. These conditions are additional to those imposed by the Human Research Ethics Committee that granted ethical approval:

The Principal Investigator is required to notify the Research Directorate, Monash Health of the following:

1. Any change in protocol and the reason for that change together with an indication of ethical implications (if any)
2. Serious or unexpected adverse effects of project on subjects and steps taken to deal with them
3. Any unforeseen events that might affect continued ethical acceptability of the project
4. Any expiry of the insurance coverage provided in respect of sponsored trials
5. Discontinuation of the project before the expected date of completion, giving reasons
6. Any change in personnel involved in the research project including any study member resigning from Monash Health &/or the study team.

At the conclusion of the project or every twelve months if the project continues, the Principal Investigator is required to complete and forward an annual report to the Committee.

List of Approved Documents:

Document	*Version*	*Date*
Monash Health Parent/Guardian Information and Consent Form	4	11 September 2014

If you should have any queries about your project please contact Deborah Dell or Julie Gephart by email deborah.dell@monashhealth.org /julie.gephart@monashhealth.org

The HREC wishes you and your colleagues every success in your research.

Yours sincerely

[redacted]

Dr James Doery
Acting Chair, HREC A

Cc: Dr Brian Hoare

Attachments: CTRA (x3)

Checklist: Post-ethics approval requirements that must be met before a research project can commence at a study site.

Please ensure that as a PI (including the CPI) the following are completed at each study site.

Requirements	**Yes/No/NA**
CTN Acknowledgement The PI must forward a copy of the CTN Acknowledgement to the Research Directorate	N/A
Clinical Trial Research Agreement The PI must forward an original fully executed copy of the CTRA to the Research Directorate.	Yes
Indemnity The PI must forward an original fully executed copy of the Indemnity to the Research Directorate.	N/A
Radiation If applicable, the RGO must contact the Medical Physicist to notify DHS, Radiation Safety Section to list the project on the Institute's licence.	N/A
Other Commonwealth statutory requirements Ensure compliance with the following e.g. Office of the Gene Technology Regulator, NHMRC Licensing Committee, NHMRC Cellular Therapies Advisory Committee.	N/A

[redacted]

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168

Postal address:
Locked Bag 29
Clayton South Vic 3169
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

19 September 2014

Prof Christine Imms
School of Allied Health
Level 2 Daniel Mannix Building
17 Young Street
Fitzroy Vic 3067

Dear Prof Imms

Study title: Minimising Impairment: Multicentre Randomised Controlled Trial of Upper Limb Orthoses for Children with Cerebral Palsy

SERP Reference Number: HREC/14/SHB/17
Monash Health HREC Ref: 14199B
Protocol number: MITrial

The Monash Health HREC B reviewed the above application at the meeting held on 19 June 2014 In addition, the HREC is satisfied that the responses to our correspondence of 25 June 2014 have been sufficiently addressed.

The HREC approved the above application on the basis of the information provided in the application form, protocol and supporting documentation.

This reviewing HREC is accredited by the Consultative Council for Clinical Trial Research under the single ethical review system.

Approval

The HREC approval is from 19 September 2014.

Approval is given in accordance with the research conforming to the *National Health and Medical Research Council Act 1992* and the *National Statement on Ethical Conduct in Human Research (2007)*. The HREC has ethically approved this research according to the Memorandum of Understanding between the Consultative Council and the participating organisations conducting the research.

Approval is given for this research project to be conducted at the following sites and campuses:

- Monash Medical Centre Clayton, Monash Health
- Royal Children's Hospital Melbourne

You must comply with the following conditions:

The Chief Principal Investigator is required to notify the Manager, Human Research Ethics Committees, Monash Health of:

1. Any change in protocol and the reason for that change together with an indication of ethical implications (if any)
2. Serious or unexpected adverse effects of project on subjects and steps taken to deal with them
3. Any unforeseen events that might affect continued ethical acceptability of the project
4. Any expiry of the insurance coverage provided in respect of sponsored trials
5. Discontinuation of the project before the expected date of completion, giving reasons

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6. Any change in personnel involved in the research project including any study member resigning from Monash Health &/or the study team.

At the conclusion of the project or every twelve months if the project continues, the Principal Investigator is required to complete and forward an annual progress report to the Committee.

Reminders to submit annual progress report forms will be forwarded to the researcher.

The Coordinating Principal Investigator is responsible for notifying Principal Investigators. The Coordinating Principal Investigator and Principal Investigators should forward a copy of this letter to their site's Research Governance Officer.

Approved documents

Documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Protocol	2.0	07 July 2014
National Ethics Application Form	2.2	2014
Victorian Specific Module	01	02 June 2014
Advertisement	2.0	18 July 2014
Recruitment Letter	2.0	18 July 2014
Information Statement Child	2.0	18 July 2014
Information Statement Adolescent	2.0	18 July 2014
Therapist Information Sheet	2.0	18 July 2014
Parent/Guardian Information and Consent Form	3.0	18 July 2014

Site-Specific Assessment (SSA)

SSA authorisation is required at all sites participating in the study. SSA must be authorised at a site before the research project can commence.

The completed Site-Specific Assessment Form and a copy of this ethics approval letter must be submitted to the Research Governance Officer for authorisation by the Chief Executive or delegate. This applies to each site participating in the research.

If you should have any queries about your project please contact Deborah Dell or Julie Gephart by email deborah.dell@monashhealth.org /julie.gephart@monashhealth.org

The HREC wishes you and your colleagues every success in your research.

Yours sincerely

Dr Simon Bower
Chair, HREC B

Cc: Dr Brian Hoare

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168

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Tel (03) 9594 4611
Fax (03) 9594 6306

Checklist: Post-ethics approval requirements that must be met before a research project can commence at a study site.

Please ensure that as a PI (including the CPI) the following are completed at each study site.

Requirements	**Yes/No/NA**
Ethics approval notification The PI must send a copy to the RGO at that study site.	Yes
HREC Review Only Indemnity The PI must forward a copy of the signed HREC Review Only Indemnity to the RGO at that study site.	Yes
CTN notification The PI must sign the CTN and forward to the RGO so the authority approving the conduct of the trial, at that site, can complete and sign.	N/A
SSA authorisation notification The PI must forward the SSA form and attached documents (e.g. CTRA) to the RGO so the authority approving the conduct of the trial, at that site, can complete and sign.	Yes
Radiation If applicable, the RGO must contact the Medical Physicist to notify DHS, Radiation Safety Section to list the project on the Institute's licence.	N/A
Other Commonwealth statutory requirements Ensure compliance with the following e.g. Office of the Gene Technology Regulator, NHMRC Licensing Committee, NHMRC Cellular Therapies Advisory Committee.	N/A

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 2014060EP

Professor Christine Imms
Level 2 Daniel Mannix Bldg
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC REF **2014060EP**
STUDY TITLE **Minimising impairment: Multicentre randomised controlled trials of upper limb orthoses for children with cerebral palsy**

The ethics application for the project referenced above was reviewed by the PMH Human Research Ethics Committee (HREC) at its meeting on 21/08/2014. It has been approved and the following documents have been approved for use in this project.

Project Protocol, version 2 dated 07 July 2014
Scientific Protocol Form 4B, Version 2 dated 07 July 2014
Case Report Form (CRF), Version 1 dated 30 April 2014
Data Collection Sheet, Study 1, Version 1 dated 30 April 2014
Data Collection Sheet, Study 2, Version 1 dated 30 April 2014
Therapist Information Sheet
Child Study questionnaire: Study completion, Version 3 dated 19 August 2014
Orthosis review questionnaire for children in the treatment group, Version 3 dated 19 August 2014
Child Study questionnaire: Baseline Version 3 dated 19 August 2014
Child Study questionnaire: 6/18/30 month assessments, Version 3 dated 19 August 2014
Child Study questionnaire: 12/24 month assessments, Version 3 19 August 2014
Brochure Orthoses for Older Children

Approval of this project from PMH HREC is valid to 21/08/2017 and on the basis of compliance with the 'Conditions of HREC Approval for a Research Project' (attached).

Note: If additional sites are recruited prior to the commencement of, or during the research project, the Coordinating Principal Investigator is required to notify the HREC. Notification of withdrawn sites should also be provided to the HREC in a timely fashion.

A copy of this ethical approval letter must be submitted by all site Principal Investigators to the Research Governance Office or equivalent body or individual at each participating institution in a timely manner to enable the institution to authorise the commencement of the project at its site/s.

This letter constitutes ethical approval only.

This project cannot proceed at any site until separate site authorisation has been obtained from the CE, or delegate, of the site under whose auspices the research will be conducted at that site.

The PMH HREC is registered with the Australian Health Ethics Committee and operates according to the NHMRC National Statement on Ethical Conduct in Human Research and International Conference on Harmonisation – Good Clinical Practice.

The HREC's Terms of Reference, Standard Operating Procedures, membership and standard forms are available from http://www.pmh.health.wa.gov.au/development/resources/ethics.htm or from the Ethics Office. Should you have any queries about the HREC's consideration of your project, please contact Ethics Office.

Please quote the above trial number 2014060EP on all correspondence associated with this trial.

Yours sincerely

Dr Mark Salmon
Executive Director
Medical Services

24/10/2014

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

Government of Western Australia
Department of Health
Child and Adolescent Health Service

CONDITIONS OF HREC APPROVAL FOR A RESEARCH PROJECT

The following general conditions apply to the research project approved by the Human Research Ethics Committee (HREC) and acceptance of the approval will be deemed to be an acceptance of these conditions by all investigators involved in the research project:

1. The responsibility for the conduct of projects lies with the Coordinating Principal Investigator (CPI), all correspondence should be signed by CPI.
2. Projects that do not commence within 12 months of the approval date may have their approval withdrawn and the project closed. The CPI must outline why the project approval should stand.
3. The submission of an application for HREC approval will be deemed to indicate that the investigator/s and any sponsor recognises the approving HREC is registered with the National Health and Medical Research Council (NHMRC) and that it complies in all respects with the National Statement on Ethical Conduct in Human Research and all other national and international ethical requirements. **The HREC will not enter into further correspondence on this point.**
4. A list of attendance at a specific meeting is available on request, but no voting records will be provided.
5. The CPI will notify the HREC of his or her inability to continue as CPI and will provide the name and contact information of their replacement. Failure to notify the HREC can result in the project being suspended or approval withdrawn.
6. The CPI will notify the HREC of any departures of named investigators. The CPI will also notify the HREC if any new investigators and/or sites join the project that will utilise the HREC's approval.
7. The CPI will inform the HREC about any changes to the project. The CPI is responsible for submitting any amendments to the approved documents listed on the approval letter, or any new documentation to be used in the project. Any new or amended documentation should be submitted in a timely manner and cannot be implemented at any participating site until they have received HREC approval.
8. The CPI is responsible for reporting adverse events, indicating whether or not the project should continue. Reporting requirements are as per the WA Health Research Governance and Single Ethical Review Standard Operating Procedures. Additional reports other than those outlined that are submitted to the HREC will be returned without acknowledgement. The HREC can request additional reporting requirements as a special condition of a research project.
9. Where a project requires a Data Safety Monitoring Board (DSMB) it is the CPI's responsibility to ensure this is in place before the commencement of the project and the HREC notified of this. All relevant reports from the DSMB should be submitted to HREC.
10. For projects where the site is acting as the sponsor (ie. investigator initiated project) it is the responsibility of the CPI to report serious and unexpected drug/device reactions, as well as other reactions/events to the Therapeutic Goods Administration (TGA). Please refer to TGA website for further information and the relevant forms (see http://www.tga.gov.au/pdf/clinical-trials-guidelines.pdf p71 for medications or p77 for devices).
11. If this project involves the use of an implantable device a properly monitored and up to date system for tracking participants is to be maintained for the life of the device in accordance with the National Statement section 3.3.22 (g).

12. The investigator is responsible for notifying the Therapeutic Drugs Administration of a device incident in accordance with the National Statement section 3.3.22 (g).
13. An annual report on an approved research project will be required on the anniversary date of the project's approval. HREC approvals are subject to the submission of these reports and approval may be suspended if the report is not submitted.
14. The HREC has the authority to audit the conduct of any project without notice. Exercise of this authority will only be considered if there are grounds to believe that some irregularity has occurred, if a complaint is received from a third party or the HREC decides to undertake an audit for Quality Improvement purposes.
15. The HREC can conduct random monitoring of any project. The CPI will be notified if their project has been selected. The CPI will be given a copy of the monitor's report along with the HREC and Research Governance Office (RGO) at each site.
16. Complaints relating to the conduct of a project should be directed to the HREC Chair and will be promptly investigated according to the Committee's complaints procedures.
17. CPI are reminded that records of consent or authorisation for participation in a project form part of the Acute Hospital Patient Record and should be stored with that record in accordance with the WA *Health Patient Information Retention and Disposal Schedule (Version 2) 2000.* A copy of the 'Participant Information Sheet' should also be included in the medical records as part of informed consent documentation.
18. The duration of HREC approval for a project is 3 year (with the option of 5 years) from the date of approval. The date of approval expiry is stipulated in the HREC approval letter.
19. If the project is to continue beyond the stipulated approval expiry date a request for an extension should be submitted prior to that expiry date. One extension of 3 years can be granted but approval beyond this time period may necessitate further review by the HREC.
20. Once the approval period has expired, the CPI is required to submit a final report. If the report is not received within 30 days the project will be closed and archived. An outstanding final report could impact on the CPI's ability to apply for approval for future projects.
21. If a project is suspended or terminated by the CPI, or a project sponsor, the CPI must immediately inform the HREC and the RGO at each site of this and the circumstances necessitating the suspension or termination of the project. Such notification should include information as to what procedures are in place to safeguard participants.
22. If a project fails to meet these conditions the HREC will contact the investigator(s) to request they rectify the identified issues. If, after being contacted by the HREC, the issues are not addressed the HREC approval will be withdrawn. The HREC will notify the RGO at each site within WA Health that work may no longer be conducted in relation to the project other than that concerning the participants safety.

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service
Research Governance Office

Our Ref: 2014060EP

Professor Christine Imms
Level 2 Daniel Mannix Building
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HREC REF **2014060EP**
STUDY TITLE **Minimising impairment: Multicentre randomised controlled trials of upper limb orthoses for children with cerebral palsy**

On behalf of the Child and Adolescent Health Service, I give authorisation for your research project to be conducted at the following site(s):

Princess Margaret Hospital for Children - CAHS

This authorisation is based on the approval from PMH HREC and the review from the Research Governance Office. This authorisation is valid subject to the ongoing approval from the HREC.

This authorisation is based on the ethical approval from the HREC, and on the basis of compliance with the 'Conditions of Authorisation to Conduct a Research Project at Site' (attached) and with the compliance of all reports as required by the Research Governance Office and approving HREC. Non compliance with these requirements could result in the authorisation being withdrawn.

The responsibility for the conduct of this project remains with you as the Principal Investigator at the site.

Yours sincerely

[redacted]

Dr Mark Salmon
Executive Director
Medical Services

27/11/2014

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service
Research Governance Office

CONDITIONS OF SITE AUTHORISATION TO CONDUCT A RESEARCH PROJECT

The following general conditions apply to the research project authorised to be conducted at the site(s) nominated in the accompanying letter. The acceptance of the site authorisation will be deemed to be an acceptance of these conditions by all investigators involved in the research project at the nominated site(s).

1. The responsibility for the conduct of project at a site lies with the nominated Principal Investigator (PI) at that site, all correspondence should be signed by PI.

2. The PI will inform the Research Governance Office (RGO) about any changes to the project. The PI is responsible for submitting any amendments to the approved documents listed on the approval letter, or any new documentation to be used in the project. Any new or amended documentation should be submitted in a timely manner and cannot be implemented at this site until they have received HREC approval for use at site(s).

3. The PI will notify the RGO of their inability to continue as PI at the site(s) and will provide the name and contact information of their replacement.

4. The PI will notify the RGO of any departures of named site investigators. The PI will also notify the RGO if any new site investigators join the project.

5. The PI is responsible for reporting site adverse events, using the standard forms available from the website. Reporting requirements are as per the WA Health Research Governance and Single Ethical Review Standard Operating Procedures. Additional reports, other than those outlined, that are submitted will be returned without acknowledgement.

6. The annual report that is submitted to the HREC should also be submitted to the RGO. This should include the site specific information which should be completed by the site PI.

7. The site has the authority to audit the conduct of any project without notice. Exercise of this authority will only be considered if there are grounds to believe that some irregularity has occurred, if a complaint is received from a third party or the site decides to undertake an audit for Quality Improvement purposes.

8. The site can conduct random monitoring of any project. The PI will be notified if their project has been selected. The PI will be given a copy

of the monitor's report along with the HREC and RGO.

9. Complaints relating to the conduct of a project should be directed to the RGO and will be promptly investigated according to the site Standard Operating Procedures.

10. The PI is reminded that records of consent or authorisation for participation in a project form part of the Acute Hospital Patient Record and should be stored with that record in accordance with the WA *Health Patient Information Retention and Disposal Schedule (Version 2) 2000.* A copy of the 'Participant Information Sheet' should also be included in the medical records as part of informed consent documentation.

11. Once the project has been closed at site, the PI is required to submit to the RGO a copy of the final report that is submitted to the HREC. This should include the site specific information which should be completed by the site PI. If the report is not received within 30 days the project will be closed and archived. An outstanding final report could impact on the PI's ability to apply for approval for future projects.

12. If a project is suspended or terminated the PI must ensure that the RGO at site is informed of this and the circumstances necessitating the suspension or termination of the project. Such notification should include information as to what procedures are in place to safeguard participants.

13. If a project fails to meet these conditions the RGO will contact the investigator(s) to request they rectify the identified issues. If, after being contacted by the RGO, the issues are not addressed the site authorisation will be withdrawn.

Government of **Western Australia**
Child and Adolescent Health Service

Our Ref: 2014060EP

Professor Christine Imms
Level 2 Daniel Mannix Building
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

HREC Ref: 2014060EP
Project Title: Minimising impairment: Multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy

On behalf of the Child and Adolescent Health Service (CAHS), I give conditional authorisation for the above research project to be conducted at Perth Children's Hospital following final move day from Princess Margaret Hospital.

This approval is conditional upon finalisation of the relevant deed of variation to the research agreement for this study after the inclusion of the final move date (once announced).

At the time of issuing this letter the following documents are approved for use at PCH:

- PICF Addendum PCH v1 dated 22 July 2016
- PICF Addendum Therapist PCH v1 dated 22 July 2016
- PICF Parent PCH v1 dated 22 July 2016
- PICF Therapist PCH v1 dated 22 July 2016
- Information sheet Adolescent PCH v1 dated 22 July 2016

Once the final move day has been confirmed, please insert the date into the Addendum to Consent (if relevant). When available, please also update any information sheet/consent forms with relevant extension numbers at Perth Children's Hospital to enable direct contact with the study team. Prior to commencing your research at PCH, please confirm TGA acknowledgment of site change details for the Clinical Trial Notification (if applicable). It is the responsibility of the study sponsor to notify the TGA of the change of site.

Any document changes required, other than those stated above, after the issuing of this letter (date of signature) will need to be submitted for review via the standard process for review of amendments.

This authorisation is based on the completion of the transition review conducted by the CAHS Research Governance Office and is valid subject to the ongoing approval from the approving Human Research Ethics Committee (HREC).

This authorisation is based on compliance with the 'Conditions of Authorisation to Conduct a Research Project at Site' (attached) and with the compliance of all reports as required by the CAHS Research Governance Office and approving HREC. Non-compliance with these requirements could result in the authorisation being withdrawn.

The responsibility for the conduct of this project remains with you as the Principal Investigator at the site.

Yours sincerely

[redacted]

Dr Mark Salmon
Director Clinical Services

pp

9 November 2017

CC: Dr Catherine Elliot
Roslyn Ward

Government of **Western Australia**
Child and Adolescent Health Service

Research Governance Office

CONDITIONS OF SITE AUTHORISATION TO CONDUCT A RESEARCH PROJECT

The following general conditions apply to the research project authorised to be conducted at the site(s) nominated in the accompanying letter. The acceptance of the site authorisation will be deemed to be an acceptance of these conditions by all investigators involved in the research project at the nominated site(s).

1. The responsibility for the conduct of project at a site lies with the nominated Principal Investigator (PI) at that site, all correspondence should be signed by PI.

2. The PI will inform the Research Governance Office (RGO) about any changes to the project. The PI is responsible for submitting any amendments to the approved documents listed on the approval letter, or any new documentation to be used in the project. Any new or amended documentation should be submitted in a timely manner and cannot be implemented at this site until they have received HREC approval for use at site(s).

3. The PI will notify the RGO of their inability to continue as PI at the site(s) and will provide the name and contact information of their replacement.

4. The PI will notify the RGO of any departures of named site investigators. The PI will also notify the RGO if any new site investigators join the project.

5. The PI is responsible for reporting site adverse events, using the standard forms available from the website. Reporting requirements are as per the WA Health Research Governance and Single Ethical Review Standard Operating Procedures. Additional reports, other than those outlined, that are submitted will be returned without acknowledgement.

6. The annual report that is submitted to the HREC should also be submitted to the RGO. This should include the site specific information which should be completed by the site PI.

7. The site has the authority to audit the conduct of any project without notice. Exercise of this authority will only be considered if there are grounds to believe that some irregularity has occurred, if a complaint is received from a third party or the site decides to undertake an audit for Quality Improvement purposes.

8. The site can conduct random monitoring of any project. The PI will be notified if their project has been selected. The PI will be given a copy

of the monitor's report along with the HREC and RGO.

9. Complaints relating to the conduct of a project should be directed to the RGO and will be promptly investigated according to the site Standard Operating Procedures.

10. The PI is reminded that records of consent or authorisation for participation in a project form part of the Acute Hospital Patient Record and should be stored with that record in accordance with the WA *Health Patient Information Retention and Disposal Schedule (Version 2) 2000.* A copy of the 'Participant Information Sheet' should also be included in the medical records as part of informed consent documentation.

11. Once the project has been closed at site, the PI is required to submit to the RGO a copy of the final report that is submitted to the HREC. This should include the site specific information which should be completed by the site PI. If the report is not received within 30 days the project will be closed and archived. An outstanding final report could impact on the PI's ability to apply for approval for future projects.

12. If a project is suspended or terminated the PI must ensure that the RGO at site is informed of this and the circumstances necessitating the suspension or termination of the project. Such notification should include information as to what procedures are in place to safeguard participants.

13. If a project fails to meet these conditions the RGO will contact the investigator(s) to request they rectify the identified issues. If, after being contacted by the RGO, the issues are not addressed the site authorisation will be withdrawn.

The Royal Children's Hospital Melbourne
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Parkville Victoria 3052 Australia
TELEPHONE +61 3 9345 5522
www.rch.org.au

The Royal Children's Hospital Melbourne

11 May 2015

Dr Sue Greaves
Occupational Therapy
The Royal Children's Hospital
Parkville, 3052

Dear Dr Greaves

Study title: Minimising impairment: a multicentre, randomised controlled trial of upper limb orthoses for children with cerebral palsy

HREC Reference Number: HREC/14/SHB/17
RCH HREC Reference: 34280A
SSA Reference Number: SSA/14/RCHM/60

Thank you for submitting a Site Specific Assessment Form for authorisation of the above project at The Royal Children's Hospital, Melbourne Victoria. I can confirm that the submission was received on 08 Dec 2014.

I am pleased to inform you that authorisation has been granted for this project to be conducted at The Royal Children's Hospital, Melbourne. **Please note this authorisation is only valid while approval from the reviewing HREC is current.**

The documents reviewed and authorised that are relevant to this authorisation are listed below.

Documents	Version	Date
SSA Application - AU/5/1E0E115		01 Apr 2015
RCH PICF (Parent/Guardian) (based on Master Version 4 dated 11 Sep 2014)	5.1	07 May 2015
RCH PICF (Adolescent) (based on Master Version 2 Date 18 Jul 2014)	3.0	31 Mar 2015
RCH PICF (Child) (based on Master Version 2 Date 18 Jul 2014)	3.0	7 Apr 2015
RCH PICF (Therapist) (based on Master Version 2 Date 18 Jul 2014)	3.0	7 Apr 2015
Recruitment Letter (RCH) (based on Master Version 2 Date 18 Jul 2014)	3.0	31 Mar 2015
Advertising Flyer (RCH) (based on Master Version 2 Date 18 Jul 2014)	2.0	18 Jul 2014

HREC approved documents are listed in reviewing HREC approval letter (dated: 19 Sep 2014).

Please note the following conditions of approval:

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1. Researchers must comply with the Investigator's Responsibilities in Research Procedure and Good Clinical Practice (ICH GCP)
2. Any proposed change in the protocol (or approved documents) or the addition of documents must be submitted to the Reviewing Human Research Ethics Committee (RHREC) for approval prior to implementation. The PI must also submit a copy of all documents relating to approved amendments to RCH Ethics Office for governance authorisation.
3. The Principal Investigator must notify the 1) CPI, 2) RCH Ethics Office and 3) Sponsor (if applicable) of:
   i. All internal (occurring in RCH participants) Serious Adverse Events (SAE) within 72 hours of occurrence.
   ii. Any other serious adverse effects to or complaints from RCH participants and steps taken to deal with them.
   iii. RCH Investigators withdrawing from or joining the project.
   NOTE: All SUSARs must be submitted to the TGA (as per the CTN guidelines).
4. A progress report must be submitted to RCH Ethics Office annually and at the conclusion of the project. A copy of this report should also be provided to the CPI.
5. RHREC approval must remain current for the entire duration of the project. Investigators undertaking projects without current RHREC approval risk their indemnity, funding and publication rights. Evidence of RHREC renewal approval will be required.
6. The PI must ensure that:
   - A fully executed CTN has been sent to the TGA prior to commencement and must place a copy of the TGA acknowledgment letter in the study file when received (2 weeks following submission).
   - The Clinical Trial Research Agreement and Indemnities are fully executed (i.e. signed by all parties) prior to commencement of the study.

If you have any matters that arise regarding conduct of the research at this site, please ensure you contact the Research Ethics & Governance office at RCH, Melbourne.

I wish you and your colleagues every success in your research.

Yours sincerely

Alexandra Robertson
Research Ethics & Governance

Phone : (03) 9345 5044
Email : rch.ethics@rch.org.au
Web : www.rch.org.au

Government of Western Australia
Department of Health
Child and Adolescent Health Service

Our Ref: 1506/2014060EP

Professor Christine Imms
Level 2 Daniel Mannix Building
Australian Catholic University
17 Young Street
Fitzroy VIC 3067

Dear Professor Imms

RE: 2014060EP - AMENDMENT OF TRIAL APPROVAL

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

HREC Ref	**2014060EP**
Study Expiry Date	**24/10/2017**
Study Title	**Minimising impairment: Multicentre randomised controlled trials of upper limb orthoses for children with cerebral palsy**

The review of the administrative amendment for the above project has been completed and it is now approved. This amendment was for :

Protocol Version 4.0 dated 23 June 2015

The responsibility for the conduct of this project remains with you as the Principal Investigator at the site/s.

I have been designated the authority to approve administrative amendments on behalf of the Child and Adolescent Health Service in accordance with the Standard Operating Procedures.

Yours sincerely

Dr Catherine Choong
Chair, Scientific Advisory Sub-Committee
20/07/2015

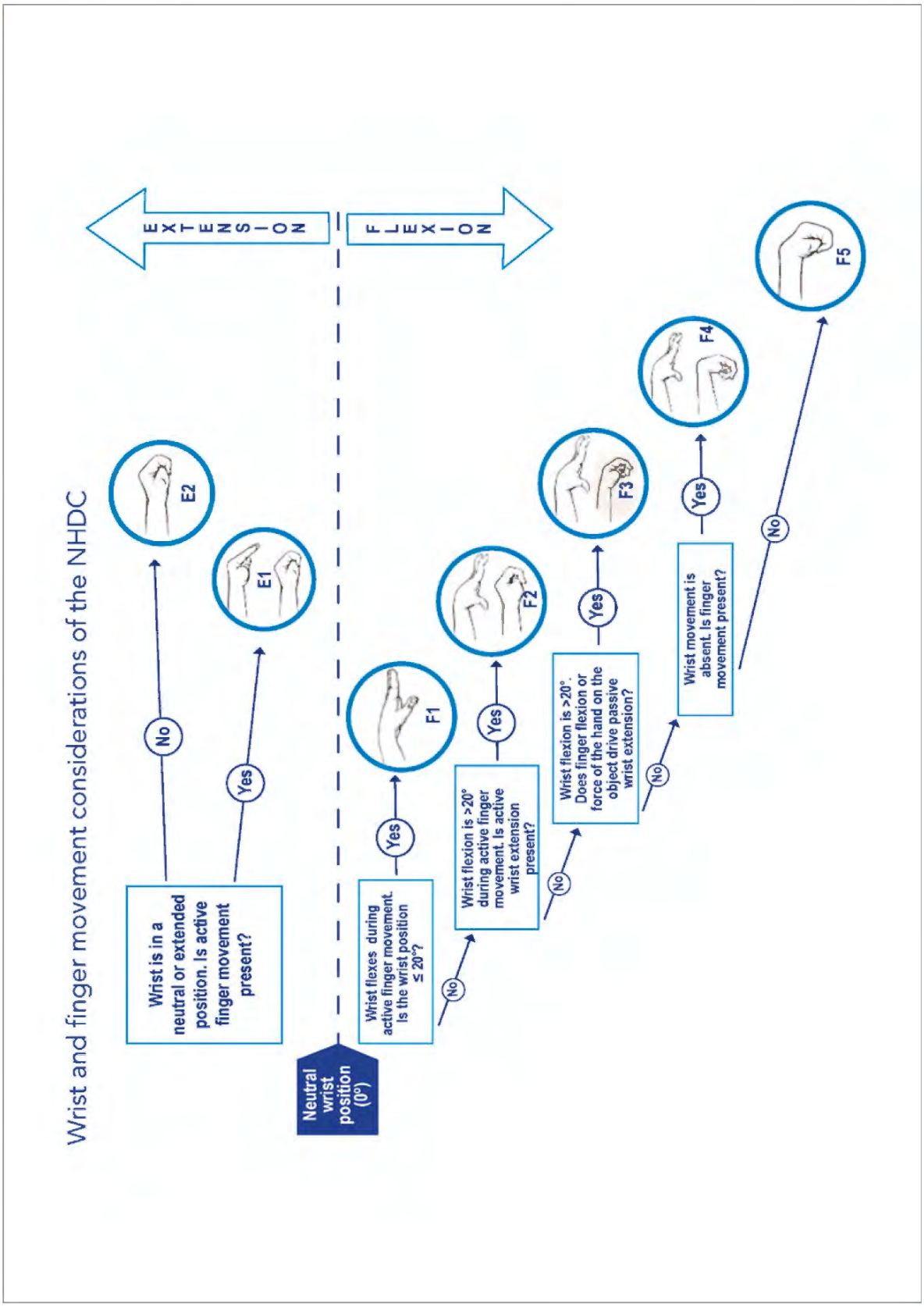
✓ I have reviewed this administrative amendment and there are no governance implications for CAHS.

☐ I have reviewed this administrative amendment and the governance implications have been addressed by the investigator to the satisfaction of the CAHS Research Governance Office.

Name: J. Westgarth-Taylor Signature: J. Westgarth-Taylor Date: 22-7-15

Appendix E Supplementary information for the NHDC

E.1 NHDC Quick Guide


Wrist and finger movement considerations of the NHDC
EXTENSION
FLEXION
Neutral wrist position (0°)
Wrist is in a neutral or extended position. Is active finger movement present?
No
Yes
E2
E1
Wrist flexes during active finger movement. Is the wrist position ≤ 20°?
Yes
No
F1
Wrist flexion is >20° during active finger movement. Is active wrist extension present?
Yes
No
F2
Wrist flexion is >20°. Does finger flexion or force of the hand on the object drive passive wrist extension?
Yes
No
F3
Wrist movement is absent. Is finger movement present?
Yes
No
F4
F5

E.2 Orthosis considerations utilising the NHDC