

**School of Occupational Therapy and Social Work**

**Evidence Based Adaptation and Evaluation  
of a Somatosensory Assessment for use  
with Children with Cerebral Palsy**

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**This thesis is presented for the Degree of  
Doctor of Philosophy  
of  
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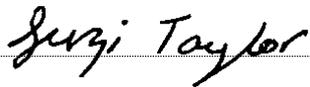


## **Declaration**

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

## **Human Ethics**

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research study received human research ethics approval from the Princess Margaret Hospital Human Research Ethics Committee (#2052) and reciprocal ethics from Curtin University Human Research Ethics Committee (#87), (#159).

A handwritten signature in black ink, reading "Suzi Taylor", is written over a horizontal dotted line.

**Susan Taylor**

1st September 2017



## Abstract

Children with cerebral palsy (CP) commonly experience deficits in somatosensation of the upper limb including touch, proprioception and haptic object recognition. Identifying and describing somatosensory deficits is important in understanding the impact of impairment on hand function, informing evidence-based practice, establishing frameworks for treatment, and in evaluating treatment outcomes. Currently, there are limited comprehensive assessment tools measuring somatosensory impairment in children with neurological conditions such as CP. Thus, this thesis described a four-phase research project, using multiple methods to review and adapt an adult assessment tool, the *sense\_assess*©, and determine its psychometric properties and applicability for use with a clinical population of children with CP aged 6 to 15 years (Figure 1). The adapted measure, the *sense\_assess*© *kids*, contained a measure of tactile registration, Protective Touch Test; a measure of tactile perception, Tactile Discrimination Test; a measure of limb position sense at the wrist, Wrist Position Sense Test; and a measure of haptic object recognition, functional Tactile Object Recognition Test. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) was used to develop a conceptual framework linking the four papers and appendices contained within this thesis. Specific research objectives were to:

1. Evaluate existing measures of somatosensation currently used for children and adults and discuss the benefit of adapting the existing adult *sense\_assess*© for children.
2. Examine the clinical acceptability of the *sense\_assess*© *kids* for use with children and adolescents with cerebral palsy.
3. Describe the measurement properties of the *sense\_assess*© *kids* subtests for use with children and adolescents with cerebral palsy.
4. Describe and compare the haptic exploratory procedures of typically developing children and children with cerebral palsy.

Paper 1 was a cross-sectional questionnaire-based study that described the perspectives of children and adolescents with CP following exposure to the *sense\_assess*© *kids* at one time-point. Paper 1 revealed that the majority, 21 of 26 children, indicated they were ‘very happy’ or ‘happy’ with the administration process

of the *sense\_assess© kids*. Content analysis revealed the majority of participants liked the sensation they felt in the hand when tested. This paper established that the *sense\_assess© kids* was acceptable to the population for whom it was intended. Papers 2 and 3 were instrument development and validation studies with experimental designs for responsiveness testing. These papers were underpinned by the COSMIN framework and focused on testing the psychometric properties of the adapted paediatric Wrist Position Sense Test (WPST) and functional Tactile Object Recognition Test (fTORT) subtests of the *sense\_assess© kids*. The WPST demonstrated construct validity, association with an activity measure, intrarater reliability and responsiveness to change. The fTORT demonstrated construct validity and association with an activity measure but further research is needed to develop its responsiveness, and also test its reliability.

Paper 4 was a cross-sectional exploratory study that used video data to describe and compare the hand movements used by young children with and without CP during administration of the fTORT. Rater reliability was determined by having a second coder evaluate 20% of the videos. Paper 4 established that children with and without CP performed similar patterns of haptic exploratory procedures (EPs) to identify objects however children with CP took more time and were less accurate. These findings indicated that somatosensation plays a pivotal role in haptic object recognition despite the development and presence of motor patterns to perform the optimal haptic EPs. There was also substantial agreement between the two raters' observations of expected EPs therefore providing evidence for a reliable method of recording the use of haptic EPs during the standardised fTORT administration.

Overall, this thesis provided evidence to support the *sense\_assess© kids* as a feasible measure of somatosensation for paediatric use however, further psychometric development is required before implementation into clinical practice. This thesis also provided a phased approach with evidence based frameworks that can be used to adapt or develop other paediatric somatosensory assessments. These findings add substantial knowledge to the field of paediatric rehabilitation because they advance evidence based clinical assessment and provide direction for clinical practice guidelines for the measurement of somatosensation.

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## Abbreviations and Acronyms

<b>2PD</b>	Two-point discrimination
<b>BBT</b>	Box and Block Test of Manual Dexterity
<b>CAHS</b>	Child and Adolescent Health Service
<b>COPM</b>	Canadian Occupational Performance Measure
<b>COSMIN</b>	COnsensus-based Standards for the selection of health Measurement INstruments
<b>CP</b>	Cerebral palsy
<b>Em-NSA</b>	Eramus MC modification of the (revised) Nottingham Sensory Assessment
<b>EMs</b>	Exploratory movements
<b>EPs</b>	Exploratory procedures
<b>FL</b>	Flicking
<b>FMA-S</b>	Fugl-Meyer Assessment sensory subscale
<b>FMT</b>	Fabric Matching Test
<b>fTORT</b>	functional Tactile Object Recognition Test
<b>GAS</b>	Goal Attainment Scale
<b>GEE</b>	General Estimating Equation
<b>GOT</b>	Grating Orientation Task
<b>HASTe</b>	Hand Active Sensation Test
<b>ICC</b>	Intraclass Correlation Coefficient
<b>ICF</b>	International Classification of Functioning, Disability and Health
<b>ICF-CY</b>	International Classification of Functioning, Disability and Health Child and Youth Version
<b>LI</b>	Lifting with enclosure
<b>LIDr</b>	Lifting and intentional dropping
<b>LITI</b>	Lifting and tilting
<b>MACS</b>	Manual Ability Classification System
<b>MTP</b>	Moving touch-pressure
<b>n</b>	Portion of total sample size
<b>N</b>	Total sample size
<b>NSMDA</b>	Neurosensory Motor Development Assessment
<b>OMRF</b>	Outcome Measures Rating Form
<b>PMH</b>	Princess Margaret Hospital for Children
<b>PTT</b>	Protective Touch Test
<b>RASP</b>	Rivermead Assessment of Somatosensory Performance
<b>rNSA</b>	Revised Nottingham Sensory Assessment
<b>RO</b>	Rolling
<b>SCSIT</b>	Southern California Sensory Integration Test

<b>SH</b>	Shaking
<b>SIPT</b>	Sensory Integration and Praxis Test
<b>STP</b>	Sustained touch-pressure
<b>SW</b>	Swiping
<b>TD</b>	Typically developing
<b>TDT</b>	Tactile Discrimination Test
<b>TI</b>	Tipping
<b>TO</b>	Tapping on object
<b>TOO</b>	Tapping object on surface
<b>TU</b>	Turning object over
<b>UHTI</b>	Unsupported holding and tilting
<b>WPST</b>	Wrist Position Sense Test

## List of Manuscripts

### Chapter Four

Taylor, S., Mclean, B., Blair, E., Valentine, J., Carey, L., Girdler, S., & Elliott, C. (2017d). Clinical acceptability of the sense\_assess© *kids*: children and youth perspectives. Manuscript accepted for publication in the Australian Occupational Therapy Journal.

### Chapter Five

Taylor, S., Mclean, B., Parsons, R., Carey, L., Girdler, S., & Elliott, C. (2017a). Wrist Position Sense Test for children with cerebral palsy. Manuscript submitted for publication to Disability and Rehabilitation.

### Chapter Six

Taylor, S., Mclean, B., Parsons, R., Carey, L., Girdler, S., & Elliott, C. (2017b). Construct validity and responsiveness of the functional Tactile Object Recognition Test. Manuscript submitted for publication to a special issue of the Australian Occupational Therapy Journal.

### Chapter Seven

Taylor, S., Girdler, S., McCutcheon, S., Mclean, B., Parsons, R., Jacoby, P., Carey, L., & Elliott, C. (2017e). Haptic exploratory procedures of children and youth with and without cerebral palsy. Manuscript submitted for publication to Physical & Occupational Therapy in Pediatrics.

### Appendix A

Taylor, S., Mclean, B., Parsons, R., Carey, L., Girdler, S., & Elliott, C. (2017c). Assessment of body sensations in children: age related effects and reliability. Manuscript submitted for publication to the British Journal of Occupational Therapy.

### Appendix B

Taylor, S., McLean, B., Falkmer, T., Carey, L., Girdler, S., Elliott, C., & Blair, E. (2016). Does somatosensation change with age in children and adolescents? A systematic review. *Child: Care, Health and Development*, 42(6), 809-824. doi: 10.1111/cch.12375



## Conference Presentations

**Taylor, Susan,** Girdler, Sonya, McCutcheon, Sara, Mclean, Belinda, Parsons, Richard, Jacoby, Peter, Carey, Leeanne, & Elliott, Catherine (2017, 19<sup>th</sup> – 21<sup>st</sup> July). *Haptic exploratory procedures of children and youth with and without cerebral palsy*. Abstract accepted for a poster session at the Occupational Therapy Australia 2017 National Conference and Exhibition, Perth. Australia.

**Taylor, Susan,** Mclean, Belinda, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2016, 10<sup>th</sup> – 11<sup>th</sup> June). *Clinical acceptability of the SENSE Assess Kids; children and youth perspectives*. Paper presented at the Occupational Therapy Australia, Perth 2016 Conference, Perth, Australia.

**Taylor, Susan,** Mclean, Belinda, Parsons, Richard, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2016, 1<sup>st</sup> – 4<sup>th</sup> June 2016). *Does sensation change with age in children and adolescents? A systematic review*. Poster presented at the International Conference on Cerebral Palsy and other Childhood-onset Disabilities, Stockholm, Sweden.

**Taylor, Susan,** Mclean, Belinda, Parsons, Richard, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2016, 18<sup>th</sup> April). *Does sensation change with age in children and adolescents? A systematic review*. Paper presented at the 2016 Symposium of WA Neuroscience (SWAN), Perth, Australia.

**Taylor, Susan,** Mclean, Belinda, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2016, 30<sup>th</sup> – 2<sup>nd</sup> April). *Clinical acceptability of the SENSE Assess Kids; children and youth perspectives*. Poster presented at the Australasian Academy of Cerebral Palsy and Developmental Medicine 8th Biennial Scientific Conference, Adelaide, Australia.

**Taylor, Susan,** Mclean, Belinda, Russo, Remo, Hobbs, David, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2016, 30<sup>th</sup> – 2<sup>nd</sup> April). *Sensory dysfunction in children with cerebral palsy: evaluation, functional impact and potential treatments*. Poster presented at the Australasian Academy of Cerebral Palsy and Developmental Medicine 8th Biennial Scientific Conference, Adelaide, Australia.

**Taylor, Susan,** Mclean, Belinda, Parsons, Richard, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2015, 21<sup>st</sup>-24<sup>th</sup> October). *Construct validity of the Wrist Position Sense Test and Functional Tactile Object Recognition Test*. Proceedings of the 69<sup>th</sup> Annual Meeting of American Academy of Cerebral Palsy and Developmental Medicine, Austin, Texas.

**Taylor, Susan,** Mclean, Belinda, Parsons, Richard, Blair, Eve, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2014, 30<sup>th</sup> September). *The Wrist Position Sense Test for use with children and adolescents*. Paper presented at the 2014 Symposium of WA Neuroscience (SWAN), Perth, Australia.

**Taylor, Susan,** Mclean, Belinda, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2014, 11<sup>th</sup>-14<sup>th</sup> March). *The SenScreen Kids – a tool to screen for upper limb somatosensation capacity in children*. Poster presented at the Australasian Academy of Cerebral Palsy and Developmental Medicine 7th Biennial Scientific Conference, Hunter Valley, Australia.

**Taylor, Susan,** Mclean, Belinda, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2014, 22<sup>nd</sup>-27<sup>th</sup> October). *The SENSE Assess Kids*. Poster presented at the Child and Adolescent Health Research Symposium, Perth, Australia.

**Taylor, Susan,** Mclean, Belinda, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine. *The SenScreen Kids, somatosensation and typical development. Proceedings of the 2012 (October) WA Occupational Therapy Association Conference*, Perth, Australia.

**Taylor, Susan,** Mclean, Belinda, Valentine, Jane, Carey, Leeanne, Girdler, Sonya, & Elliott, Catherine (2012, 20<sup>th</sup>-25<sup>th</sup> October). *The SenScreen Kids*. Paper presented at the Child and Adolescent Health Research Symposium, Perth, Australia.

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would also like to thank Princess Margaret Hospital for their ongoing logistical support and provision of resources throughout this research including the team at Medical Illustrations for producing exceptional professional photos to be included in the *sense\_assess© kids* Administration manual and the design and production of scientific posters for dissemination of our research findings.

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To my mother and father who have provided me with the utmost love, support and inspiration to achieve whatever I desire, I am forever grateful. They have always provided unwavering support especially in my most difficult of times throughout his PhD and they have celebrated every joyous moment too. Their intelligence and creativity when discussing my research ideas and later my thesis has been invaluable. To my brother who is not with us now, thank-you for showing me in my younger years that if you are persistent and you try your hardest, no matter how hard the task, you can make it. It is to my mum, dad and brother that I dedicate this thesis.

## Preface

Evidence based practice in somatosensory measurement is only possible when robust assessment tools with which to assess somatosensory ability have been standardised and tested within designated populations (Gilmore, Sakzewski, & Boyd, 2010). This is important to ensure accurate understanding of impairment to plan treatment, and to provide meaningful information about rehabilitation outcomes to families and healthcare professionals (Connell & Tyson, 2011). The routine use of valid and reliable somatosensory assessment tools in paediatric allied health is infrequent and there is a need to develop standardised assessment tools for paediatric use to evaluate somatosensory deficits in children (O'Connor, Kerr, Shields, & Imms, 2016; Auld, Boyd, Moseley, & Johnston, 2011). Paediatric somatosensory measures have been employed in research over the past 50 years however therapists still do not have a comprehensive measure of somatosensation that is validated for use in children and youth with neurological disorders (Auld et al., 2012b).

The research presented in this thesis was conducted in collaboration with La Trobe University and the Florey Institute of Neuroscience and Mental Health in Melbourne, Australia where a sense© program was developed including an assessment component, the SenScreen© Sensory Screening Tool and sense\_assess©, and a training component, sense© training (Carey, Matyas, & Macdonell, 2011). The sense© program was designed to measure and rehabilitate somatosensation of the affected upper limb in adults following stroke (Carey et al., 2011). The SenScreen© screening tool and sense\_assess© assessment contain subtests that measure the somatosensory capacity of the hand, without vision. The overall aim of this thesis was to develop a paediatric version, the sense\_assess© *kids*, for use in conjunction with sense© training, providing a comprehensive assessment and training program for children with neurological disorders as has been done for adults. Because the sense© program is designed to be an assessment and training package, sense© training was also adapted for children with cerebral palsy (CP) via an associated PhD at Princess Margaret Hospital for Children (PMH) titled “Discovering the sense of touch: Somatosensory discrimination in children with cerebral palsy”. The PhD candidate Susan Taylor is the lead developer of the sense\_assess© *kids* however, full

development and testing of all elements of psychometric measurement properties was beyond the scope of this PhD.

The SenScreen© screening tool was first adapted for children in partial fulfilment of Susan Taylor's Bachelor of Applied Science (Honours degree) in 2012 (Appendix A) and was titled; The SenScreen© Kids, somatosensation and typical development. The study established the intrarater reliability of the SenScreen© Kids and collected initial data on typical performance. The current thesis is an extension of this work and presents the adaptation of the *sense\_assess© kids* from the adult version, and examines its suitability for use with a clinical population of children and adolescents. As the subtests of the SenScreen© Kids were maintained, and given that they achieved reliability with typically developing (TD) children and reliability and validity with adults, it was expected that the *sense\_assess© kids* would achieve reliability and validity within a clinical population of children.

The current thesis was underpinned by three published evidence based frameworks related to assessment tool development and knowledge translation. The Outcome Measures Rating Form (OMRF) and OMRF guidelines (CanChild, 2004) were used to support pragmatic tool adaptation and development of the adult-to-paediatric adaptation protocol proposed in this thesis. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010) guided research study design and psychometric testing of the newly adapted tool. The Knowledge to Action Cycle (Graham et al., 2006) provided an overall plan for research design and dissemination of research findings into clinical practice and supported development of the Evidence Based Tool Development framework proposed in this thesis. Classification of impairment was based on The International Classification of Functioning, Disability and Health – Child and Youth Version (ICF-CY) and the Comprehensive ICF Core Set for children & youth with cerebral palsy from birth to 18 years of age and provided a systematic coding scheme to classify the somatosensory impairments measured by the assessment tool under examination in this thesis. (Schariti, Selb, Cieza, & O'Donnell, 2015; World Health Organization, 2007). There were no alternative frameworks appropriate that satisfied all aspects of the research questions posed in this thesis. The COSMIN was selected because it was the only comprehensive,

contemporary, evidence based framework widely used by similar allied health disciplines to examine the psychometric properties of clinical assessments (Beattie, Murphy, Atherton, & Lauder, 2015). Even so, the COSMIN still needed to be synthesised and extended to cover the areas of tool adaptation and clinical utility. Adding to this, the strength of the Knowledge to Action cycle as a translation framework to guide the clinical applicability of the work and therefore its ultimate influence and impact on a significant clinical area. The ICF is a well adopted framework that provides the allied health field with a common, international language facilitating communication amongst health professionals to improve clinical care (Jette, 2006).

The strengths of using evidence based frameworks such as those selected for this thesis lie in their clear and concise parameters for clinical investigation. These particular frameworks also highlight the benefit of using best practice models in clinical research.

Clinical research conducted at the doctoral level is expected to produce clinically meaningful insights and disseminate findings to peers, and other end users such as patients, their families and also clinicians. The research program associated with the current thesis is integrated into clinical care. The strategic knowledge partnerships generated as part of this thesis included: university departments, honours students, local paediatric hospital and rehabilitation centres, experienced clinicians, interstate medical institutions and patients and their families. These partnerships will accelerate knowledge translation and implementation of the *sense\_assess© kids* into clinical practice. Figure 1 illustrates the Phases of the thesis including acknowledgement of knowledge partnerships and supplementary documents that have supported the PhD research project (Table 2). Diagrams and infographics feature throughout this thesis, linking the conceptual frameworks and the knowledge translation plan (Chapter ten).

The Phases of thesis diagram (Figure 1) has been based on the Knowledge to Action Cycle model (Graham et al., 2006). The funnel shape represents initial knowledge inquiry and synthesis which begins broad in Phase one and the

application of an adult-to-paediatric adaptation protocol occurring in Phase two. As the funnel tapers, in Phase three, knowledge is tailored based on exploratory research outcomes. Only the most refined and accurate knowledge is translated into clinical practice at the end of the cycle in Phase four. Informing the Knowledge to Action Cycle is the COSMIN psychometric framework, and the Evidence Based Tool Development (EBTD) framework developed as part of this thesis. The EBTD framework, based on the COSMIN, was adapted with permission from the Journal of Clinical Epidemiology and reflects work completed, and work that needs to be completed prior to implementation of the *sense\_assess© kids*. Illustrations from the EBTD framework appear in Chapter one and also at the beginning of each thesis chapter to highlight which Phase was undertaken.

The credentials of the current clinical supervision team, academic supervisors, the PhD candidate's prior knowledge of the SenScreen© Kids and *sense\_assess©* and experience in tool development validated the undertaking of the current PhD research project. This thesis contains published and unpublished work and work prepared for publication, some of which has been co-authored. The statements of contribution to intellectual property are presented in the introduction to each paper and the start of each individual appendix.

Figure 1 Phases of the thesis based on the Knowledge to Action Cycle

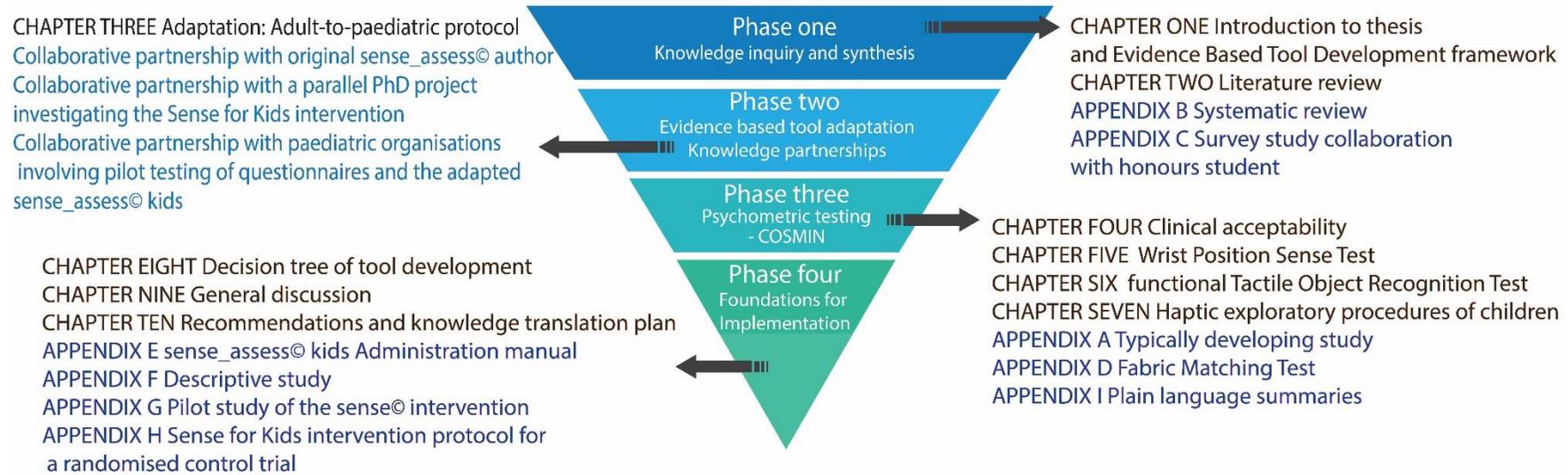


Figure 1. Phases of the current thesis represented in the form of a ‘Knowledge to Action’ funnel. Adapted from “Lost in knowledge translation: time for a map?” by I. D. Graham, J. Logan, M. B. Harrison, S. E. Straus, J. Tetroe, W. Caswell and N. Robinson, 2006, Journal of Continuing Education in the Health Professions, 26(1), p.19. Permission statement obtained for reuse (Appendix J).



# **CHAPTER 1 Introduction to Thesis**

## **1.1 Statement of the problem**

‘Is the sense\_assess© a suitable tool with which to assess somatosensory ability in children and youth with cerebral palsy?’

The overall aim of this thesis was to develop a robust tool for use by practising clinicians that objectively measures somatosensory capacity in children with cerebral palsy (CP).

## **1.2 Definition of somatosensation**

Somatosensory function is the ability to detect, recognise and discriminate between body (somato) sensations. The somatosensory system obtains information from limb movement (kinaesthesia) and limb position sense (proprioception), touch, pressure and perception of temperature and pain (Barker, 2008; Wingert et al., 2009). Somatosensory inputs are also involved in more complex central nervous system processes such as haptic object recognition and emotional responses to pain or itch (Carey, 2012). The term somatosensation does not include information received through the special senses such as vision, hearing, smell and taste (Carey, 2012). Much of the current literature uses the term ‘sensation’ to broadly describe many of the body sensations under examination in the current thesis however, for clarity and to be specific, this thesis uses the term somatosensation.

Somatosensation facilitates perception of the environment such that when somatosensory information is received the active process of selecting, organising, and interpreting information can occur (Dijkerman, & de Haan, 2007). Intact somatosensation enables skilful manipulation, precision grip, automatic movement corrections, anticipatory control and effective fine movement of the hand (Carey, 2012; Cooper et al., 1995; Eliasson & Burtner, 2008; Lundy-Ekman, 2002). If the mechanisms underlying capacity to achieve precise motor control are damaged (such as inefficient somatosensation or damaged afferent pathways) a child may not be able to gauge the calibration, adjustment and accuracy needed for fine hand use (Carey, 2012; Eliasson & Burtner, 2008). Motor control and somatosensory function are both important because they are mutually dependant (Gandevia, Macefield, Burke, &

McKenzie, 1990; Kalagher & Jones, 2011). Loss of somatic sensation negatively impacts the tactile exploration needed to perform daily activities in adults and children with neurological disorders such as CP (Carey, 1995; Carey et al., 2011; Eliasson, 2005).

Touch is used to describe the processes of tactile registration and tactile perception (Auld et al., 2011). Tactile registration is the basic initial processing of stimuli and can include terms such as touch sensitivity, tactile sensitivity, and touch threshold or threshold detection (Weinstein, 1993; Bell-Krotoski, Fess, Figarola, & Hiltz, 1995; Riquelme, Cifre, & Montoya, 2011). Tactile perception allows interpretation of stimuli including location, timing and identification (i.e. what the stimuli is) (Auld et al., 2011) and has been described by terms such as two-point discrimination, tactile spatial acuity or tactile spatial resolution (Bleyenheuft, Cols, Arnould, & Thonnard, 2006; Peters & Goldreich, 2013); tactile localisation (Yoshioka, Dillon, Beck, Rapp, & Landau, 2013). The functional aspects of tactile perception can be described as tactile or texture discrimination (Carey, Matyas, & Oke, 2002; Dunn et al., 2013).

Proprioception is the sense of the movement and position of the body and limbs in space (Stedman's medical dictionary, 2008). The term proprioception includes the static component of limb or joint position sense (Bremner et al., 2013) and the dynamic component of kinaesthesia (World Health Organization, 2007). Haptic object recognition relies on a complex integration of touch, proprioception and in-hand manipulation skills (Carey, 2012). Haptic ability is often referred to as haptic perception (Ballesteros, Bardisa, Miller, & Reales, 2005; Kalagher & Jones, 2011a, 2011b; Gori, Giuliana, Sandini, & Burr, 2012) or stereognosis<sup>1</sup> (Auld et al., 2012b), and terms such as dynamic touch (Kloos & Amazeen, 2002; Fitzpatrick & Flynn, 2010), gnostic hand function (Van Grunsven, Njikiktjien, Vranken, & Vuylsteke-Wauters, 2003) or haptic and tactile object recognition (Carey, 2012) are often used interchangeably or defined as part of haptic ability.

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<sup>1</sup> Although termed differently, stereognosis measures reviewed in this thesis are measures of haptic object recognition and do not investigate more complex cognitive perceptual impairments as the term gnosis suggests. It is for that reason that measures termed stereognosis have been examined in the same category as measures of haptic object recognition within the current thesis.

### **1.3 Cerebral palsy and somatosensory deficits**

CP is a broad term used to describe a group of disorders that impact the development, production and voluntary control of movement (Rosenbaum, 2003; Stanton, 2012). People with CP may also experience functional difficulties related to sensory, developmental, behavioural and learning impairments (Rosenbaum, 2003). The extent of impairment is dependent on the site and magnitude of the damage within the developing brain and the nature of the lesions, with deficits being highly individualised (McIntyre, 2011; van Haastert et al., 2011). Analysis of the data from the Western Australian, Victorian and South Australian population registers estimated that CP affects 2.1 individuals per 1,000 live births (ACPR Group, 2013) and almost 40,000 Australians are living with this condition (Access Economics, 2007).

Although movement disorder defines CP, it is well documented that somatosensory problems exist for many individuals (Bax, Goldstein, Rosenbaum, Leviton, & Paneth, 2005; Rosenbaum, 2003; Stanton, 2012). In a recent study examining 52 children with hemiplegic CP, Auld et al. (2012a) reported that 40% of the sample had upper limb sensory related registration and perception deficits. In other studies, tactile deficits were reported for between 42% to 90% of children with CP (Clayton, Fleming, & Copley, 2003; Cooper et al., 1995; McLean, Taylor, Valentine, Carey, & Elliott, 2017b), with the most commonly reported areas of somatosensory deficit being in tactile object recognition, two-point discrimination and limb position sense (proprioception) (Auld et al., 2012a; Auld et al., 2012b; Curry & Exner, 1988).

Void of injury ascending somatosensory neural pathways provide tactile and proprioceptive information (Marieb & Hoehn, 2007). By accessing these forms of information, the central nervous system can alter signals sent to descending motor pathways during grasp and associated manipulation of objects (Majnemer, Bourbonnais & Frak, 2008). In the upper limbs, both fine motor movements and tool use rely heavily on such feedback, as well as interhemispheric connections functional and structural, and serial and parallel processing within the system (Gibson & Pic, 2000; Majnemer, Bourbonnais & Frak, 2008; Dijkerman & De Haan, 2007). The central processing of somatosensation is complex because each part has individual

roles that cannot function in isolation and is essential to the function of the system as a whole (McLean et al., 2017c). Damage to corticomotor tracts and thalamocortical sensory pathways both contribute to upper limb motor impairment in hemiplegia (Hoon Jr et al., 2009). However, the corticospinal system has a marked capacity for reorganisation after lesions and there is evidence of compensatory recruitment of the unaffected hemisphere in children with hemiplegia (Eyre et al., 2001; Eyre et al., 2007). The type of corticospinal reorganisation depends on the extent of the brain lesion (Staudt et al., 2002; McLean et al., 2017c). Interhemispheric dissociation between somatosensory inputs and motor outputs may be a significant contributing factor to the impaired integration of sensorimotor function in a subset of children with hemiplegia (Jaspers et al., 2016; McLean et al., 2017c). Therapy can result in cortical reorganisation (Sutcliffe et al., 2007). Rehabilitation may be used to facilitate plasticity to achieve meaningful outcomes for an individual (Carey, 2012). But first we must be able to measure somatosensation accurately in order to comment on changes due to rehabilitation.

#### **1.4 Identified gaps: Current assessments and best practice methods**

Currently there are limited published standardised methods that have been tested to comprehensively measure somatosensory function in children with neurological disorders such as CP (Auld et al., 2012b). Without adequate clinically proven and standardised assessment tools there is no reliable method of evaluating treatment outcomes or providing objective information to allied health professionals and families in relation to the sensory functioning of children with neurological conditions (Cooper et al., 1995). Interventions involving somatosensory training (e.g. Carey, Macdonell, & Matyas, 2011) have been adapted for use with children with CP (McLean et al., 2017a). Therapists require robust assessment tools to describe patterns of somatosensory impairment, and to determine the efficacy of new treatment methods by accurately measuring outcomes (Carey et al., 2011; Fess, 2002; Randall, Imms, & Carey, 2008).

The use of standardised and valid measures in occupational therapy is stated as a core principal in clinical and professional guidelines (Brayman et al., 2005).

However, ‘gold standard’ comprehensive outcome measures of somatosensation are yet to be developed (Auld et al., 2012b; Connell & Tyson, 2012; Cooper et al., 1995; Krumlinde-Sundholm & Eliasson, 2002). Measures of somatosensory impairment have been used in research but few have established psychometric properties, can demonstrate robust validity or reliability or have available normative data for children and adolescents (Connell & Tyson, 2012; Carey et al., 2011; Fess, 2002; Novak, Mackinnon, Williams, & Kelly, 1993; Tassler & Dellon, 1995).

## **1.5 Evidence Based Tool Development framework**

The design of this research and subsequent reporting was informed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010). The COSMIN provides: 1) Explicit criteria for psychometric measurement properties to improve the design of studies examining measurement instruments; 2) A framework of evaluation to improve evidence based selection of health measurement instruments; 3) A quality assessment tool for systematic reviews investigating existing outcome measures and the methodological quality of study design (Miller, Ziviani, & Boyd, 2014; Mitchell, Ziviani, Oftedal, & Boyd, 2013).

A specific interest of this thesis was to describe the steps of evidence based tool development: as a four-phase process: First, to synthesise current knowledge and gaps in the literature second; to formalise the process of adapting the sense\_assess© third; to measure the psychometric properties of the assessment and the perspectives of end users such as children with somatosensory deficits and lastly; to develop a plan for implementation. In line with these phases, this research considered what needed to happen before psychometric testing (Knowledge inquiry and synthesis, and Adaptation) and what needed to happen after psychometric testing and before implementation (Psychometric testing, Clinical utility and Implementation) (Figure 2).

Figure 2 Evidence Based Tool Development framework based on the COSMIN

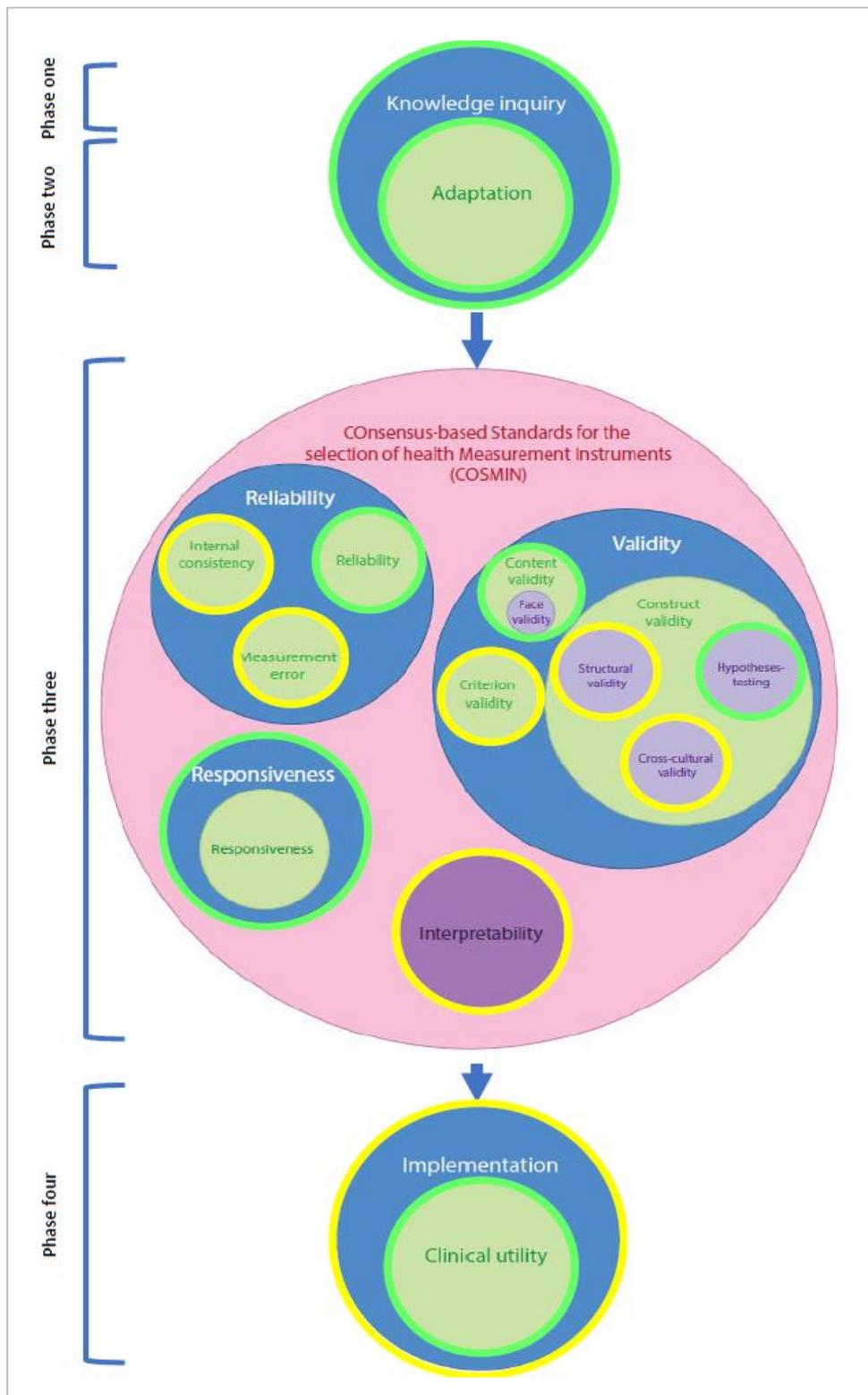


Figure 2. Graphic representation of the tool development and psychometric testing achieved and yet to be achieved. Domains completed within this thesis are highlighted in green, and the domains still requiring completion are highlighted in yellow. Adapted from “International consensus on taxonomy, terminology, and definitions of measurement properties: results of the COSMIN study,” by L. B. Mokkink, C. B. Terwee, D. L. Patrick, J. Alonso, P. W. Stratford, D. L. Knol, L. M. Bouter, H. C. W. de Vet, 2010, Journal of Clinical Epidemiology, 63, p.741. Permission statement obtained for reuse (Appendix K).

Clinical utility was defined as how the assessment would be applied and accepted within clinical practice and by a clinical population. Clinical utility determines the speed of future implementation of assessment tools in clinical practice (Metzler & Metz, 2010). In Figure 2, the Evidence Based Tool Development (EBTD) framework is presented and is used throughout the chapters in this thesis. The domains of the EBTD framework covered by this thesis are highlighted in green, and the domains still requiring completion are highlighted in yellow. Elements of the EBTD framework were also informed by the work of Randall, Imms and Carey (2008), Law (1987) and CanChild (2004) as described in Chapter three.

## **1.6 Study setting**

This PhD research project was conducted in partnership between Curtin University and the Child and Adolescent Health Service (CAHS) at Princess Margaret Hospital for Children (PMH) in Perth, Western Australia (WA). PMH provides tertiary and emergency care to paediatric patients in the state of WA with approximately 250,000 patient visits (inpatient and outpatient) each year (Metropolitan Health Service Annual Report 2014–15). The health service is committed to programs that promote lifelong health in children and adolescents. Recently there has been an increased awareness of the prevalence of somatosensory deficits in children with neurological disorders receiving rehabilitation at PMH. In 2011 CAHS and the PMH Foundation provided research funds to support the current investigation into assessment methodologies, and also clinical trials to improve somatosensation in patients on the Cerebral Palsy Mobility Register at PMH.

Test equipment and materials for the *sense\_assess© kids* were purchased by PMH and stored within the research facility of the Paediatric Rehabilitation Department. The School of Occupational Therapy and Social Work at Curtin University, Bentley provided an annual income in the form of an Australian Postgraduate Award and Curtin Research Scholarship, the school also provided logistical and infrastructure support and funding for technical equipment and consumable materials throughout the project. The Princess Margaret Hospital Foundation also provided income support in the form of a PhD Top-Up Scholarship.

The findings reported in this thesis support an organisational change to advance measurement and rehabilitation of somatosensory capacity in children with neurological conditions at PMH.

As described in the preface of this thesis the current doctoral research was closely linked to a parallel PhD testing the feasibility of a novel somatosensory intervention, Sense© for Kids. The parallel PhD involved a clinical population of 28 children and adolescents with hemiplegic CP and used data collected with the *sense\_assess© kids* as a primary outcome measure to quantify change as a result of the intervention. The Sense© for Kids intervention is referred to throughout this thesis therefore, to enhance the understanding of subsequent sections a brief outline of the intervention is described here. Sense© for Kids is designed to improve three aspects of somatosensory discrimination: haptic object recognition, tactile discrimination, and limb position sense (Carey et al., 2011; McLean et al., 2017a). Haptic object recognition is trained using graded object sets. The sense© training principles are: ‘active exploration with vision occluded; feedback on performance (accuracy and method of exploration); calibration with vision and with the less affected hand; anticipation trials in which the participant knows what to expect to feel (vision occluded); and repetition and progression from easy to more complex tasks to distinguish differences across a wide variety of stimuli’ (McLean et al., 2017a, p. 3).

## **1.7 Thesis outline and phases of research**

The aim of this thesis was to adapt and validate an existing adult measure of upper limb somatosensation for paediatric use by way of an evidence based process. Specifically, the research occurred across four phases: 1) Knowledge inquiry and synthesis; 2) Evidence based tool adaptation; 3) Psychometric testing using the COSMIN; and 3) Foundations for implementation (Figure 3, Table 1 and Table 2).

Figure 3 Structure of thesis with relation to the Phases of research

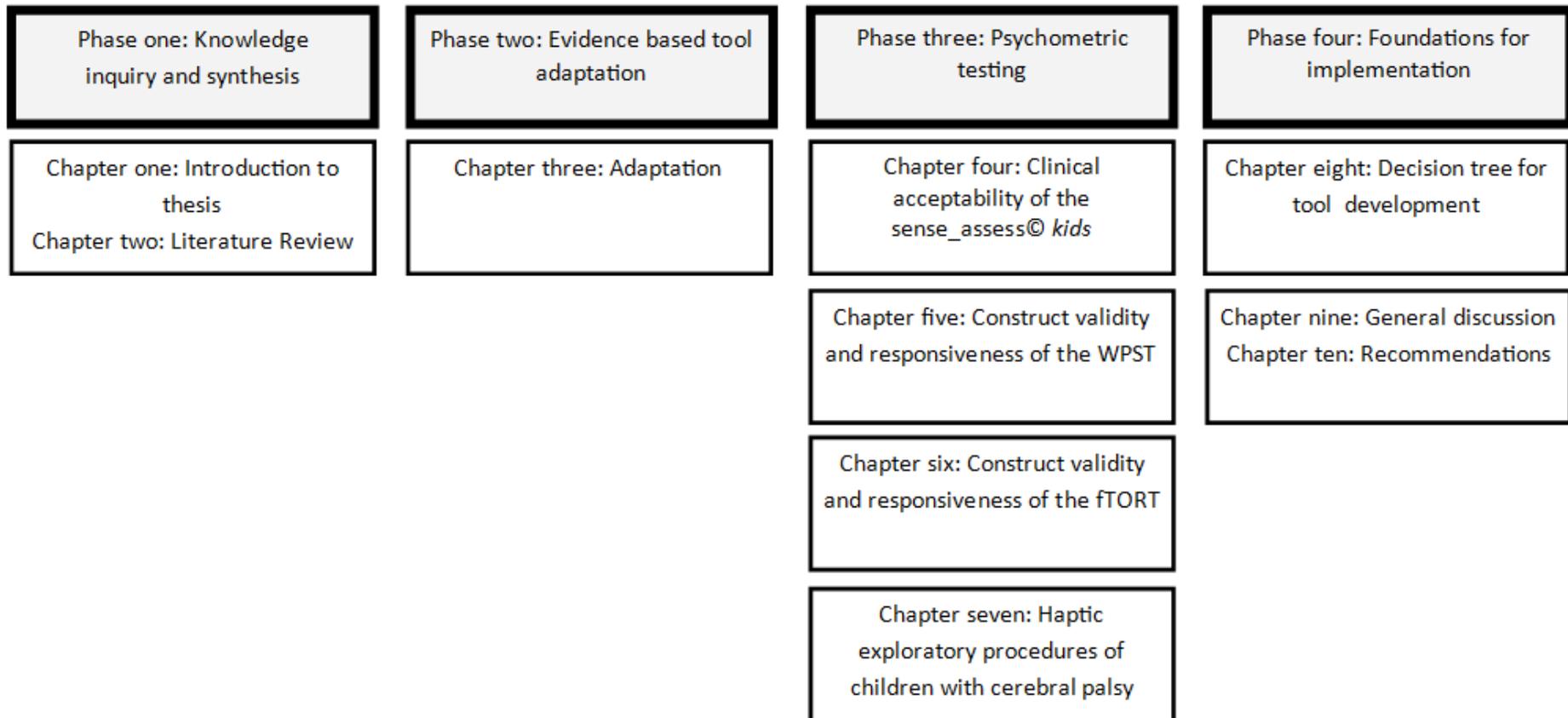


Figure 3. Graphic representation of the structure of the current thesis. The four shaded boxes at the top indicate Phases one to four. Boxes appearing underneath the shaded boxes outline the corresponding thesis sections.

### **1.7.1 Phase one – Knowledge inquiry and synthesis**

The primary objectives of Phase one of this research were to describe somatosensation and the importance of somatosensation for hand function and to review the literature related to measures of somatosensation (Chapter two). This phase underpinned the development of subsequent phases and was informed by examining (a) maturation of somatosensation throughout development (Appendix B), and (b) how therapists in Australia currently measure somatosensation in children (Appendix C).

### **1.7.2 Phase two – Evidence based tool adaptation**

Phase one identified an absence of suitable paediatric measures of somatosensation therefore, Phase two was used to devise an adult-to-paediatric adaptation protocol (Chapter three) to modify an existing adult measure with extensive psychometric evidence. In this phase, knowledge partnerships were created with end users (e.g. clinicians, children, families and manufacturers), research colleagues and experts in the field of somatosensation. The purpose of this process was twofold; to enhance the quality of the research by engaging multiple working partners; and to promote implementation of the research findings directly into clinical practice in the future.

### **1.7.3 Phase three – Psychometric testing based on the COSMIN**

The objective of Phase three was to trial the adapted paediatric version of the *sense\_assess*© and develop its psychometric properties in a population of TD children and children with CP. This phase involved examining test re-test reliability of a brief version of the assessment in TD children (Appendix A) and an alternative measure of tactile discrimination, the Fabric Matching Test (Appendix D) and resulted in development of a standardised administration manual containing psychometric data and normative standards for children aged 6 to 15 years (Appendix E). The *sense\_assess*© *kids* administration manual contains content developed and adapted as part of this PhD however, it drew on the original adult *sense\_assess*© manual content and therefore is included as an appendix in this thesis and not a chapter. Three experimental studies and one descriptive study were completed in Phase three as part of this thesis (Papers one to four). An overview of the aims and hypotheses of each of the papers is provided below.



## **Paper one**

Clinical acceptability of the sense\_assess© *kids*: children and youth perspectives

### ***Aim***

1. To determine if use of the sense\_assess© *kids* is acceptable to children and adolescents with CP using a short questionnaire.

### ***Hypothesis***

1. Children and adolescents with CP who have been tested using the sense\_assess© *kids* will respond neutrally to positively on a short questionnaire to all aspects of the assessment procedure.

## **Paper two**

Wrist Position Sense Test for children with cerebral palsy

### ***Aim***

1. To determine the construct validity of the Wrist Position Sense Test (WPST) subtest of the sense\_assess© *kids* using known groups, test re-test reliability, and associations with scores on an activity measure, the Box and Block test of Manual Dexterity (BBT) (Mathiowetz, Federman, & Wiemer, 1985a). Responsiveness of the WPST was examined during a somatosensory intervention trial for children with CP.

### ***Hypotheses***

#### ***Construct validity***

1. The WPST will reflect known differences in somatosensory performance between groups at risk of a somatosensory deficit and those who are TD.

#### ***Association with an activity measure***

2. Performance on the WPST will have a positive association with scores on an established upper limb measure of activity.

#### ***Test re-test reliability***

3. The WPST will demonstrate good intrarater agreement when administered to the same TD children at two different time-points three weeks apart.

### *Responsiveness*

4. The WPST will detect statistically significant differences between children with CP who have been exposed to a novel somatosensory intervention and those who received standard care.

## **Paper three**

Construct validity and responsiveness of the functional Tactile Object Recognition Test for children with cerebral palsy

### *Aim*

1. To determine the construct validity of the functional Tactile Object Recognition Test (fTORT) subtest of the *sense\_assess© kids* using known groups, and association with scores on an activity measure, the BBT (Mathiowetz et al., 1985a). Responsiveness of the fTORT was examined during a somatosensory intervention trial for children with CP.

### *Hypotheses*

#### *Construct validity*

1. The fTORT will reflect known differences in somatosensory performance between groups at risk of a somatosensory deficit and those who are TD.

#### *Association with an activity measure*

2. Performance on the fTORT will have a positive association with scores on an established upper limb measure of activity.

### *Responsiveness*

3. The fTORT will detect statistically significant differences between children with CP who have been exposed to a novel somatosensory intervention and those who received standard care.

Phase three also produced new knowledge about the hand movements of children with CP by comparing them to their TD peers during a subtest of the *sense\_assess© kids*, the fTORT (Paper four). Paper four also provided preliminary evidence to support a method used to describe the hand movements of young children with CP during the standardised fTORT test procedure. This

understanding will be significant to therapists who want to intervene and measure change in children's selection of haptic exploratory procedures after periods of somatosensory intervention aimed at improving haptic ability (see Appendix G and H for a description of such an intervention).

## **Paper four**

Haptic exploratory procedures of children and youth with and without cerebral palsy

### ***Aim***

1. To examine differences in haptic exploratory procedures (EPs) for TD children and children with CP during haptic object exploration as part of the fTORT.

### ***Hypotheses***

1. TD children aged 6 to 15 years will perform the same haptic EPs as previously classified in adults when exploring the sensory attributes of common objects.
2. TD children will perform more expected EPs, less additional EPs and exploratory movement (EMs), take less time to identify each object, and correctly identify more objects than children with CP.
3. Age, gender, hand dominance for the TD children or Manual Ability Classification System level and presence of somatosensory deficit for children with CP will be associated with presence of expected EPs, additional EPs and EMs, time taken and accuracy in identifying objects.
4. Interrater reliability of identification of EPs using the Cohen's kappa will be strong (0.8 - 0.9) between two raters coding video data for children with and without CP.

### **1.7.4 Phase four – Foundations for implementation**

The primary aim of Phase four was to describe the foundations for implementation of new knowledge into clinical practice. For example, a decision tree for tool development is provided that illustrates what has been accomplished for the sense\_assess© *kids* (Chapter eight), and an outline of the tasks required before integration of the tool into clinical care. Chapter nine, provides an overview and

discussion of findings, contributions to the field and limitations of the research conducted as part of this thesis. Chapter ten presents an outline of recommendations for the current use of the *sense\_assess© kids* in clinical practice and research, a plan for future psychometric testing that can be used to design similar studies, and suggestions for clinical practice guidelines for the assessment of somatosensory capacity in children with CP. The discussion and recommendations contribute to the overall thesis by synthesising the new findings and proposing a direction for knowledge translation to end users such as patients and their families, health organisations, clinicians, educators, and researchers.

Table 1 A summary of the methods used in each of the studies in the thesis

Study and short title	Paper one – Clinical acceptability	Paper two – Wrist Position Sense Test	Paper three – functional Tactile Object Recognition Test	Paper four - Haptic exploratory procedures
Study design	Descriptive design: Cross-sectional, questionnaire-based study following exposure to the sense_assess© kids.	Exploratory and experimental design: Instrument development and validation study, with experimental design for responsiveness.	Exploratory and experimental design: Instrument development and validation study, with experimental design for responsiveness.	Exploratory design: Cross-sectional, video data analysis study. Rater reliability was determined by having a second coder evaluate 20% of the videos.
Data type	Text from survey responses.	Nominal, scale, and ordinal.	Nominal, scale, and ordinal.	Nominal, scale, ordinal and video data.
Analysis design	Content analysis and descriptive statistics.	Within group and between group design comparing data from baseline and follow up time-points.	Within group and between group design comparing data from baseline and follow up time-points.	Comparison of two independent groups using video data.
Statistical methods	Summative content analysis and descriptive statistics.	General Estimating Equation and random-effects regression modelling.	General Estimating Equation and random-effects regression modelling.	Logistic, linear and Poisson regression modelling General Estimating Equation and Cohen's $\kappa$ .
Sample CP/TD	N=28 / N/A	N=28 / 39	N=28 / 39	N=24 /31
Key findings	21 of 26 children indicated they were 'very happy' or 'happy' with the administration process of the sense_assess© kids.	The WPST demonstrated construct validity, association with an activity measure, intrarater reliability and responsiveness to change.	The fTORT demonstrated construct validity and association with an activity measure. Further research is needed to develop its responsiveness.	Children with and without CP performed similar patterns of haptic EPs however children with CP took more time and were less accurate. Rater reliability was confirmed.

*Note.* WPST = Wrist Position Sense Test; fTORT = functional Tactile Object Recognition Test; EPs = Exploratory procedures; CP = cerebral palsy; TD = typically developing.

Table 2 A summary of the methods used in each of the appendices involving a research study

Study and short title	Appendix A Reliability study	Appendix B Systematic review	Appendix C Survey study	Appendix D FMT	Appendix F Descriptive study	Appendix G Intervention pilot study	Appendix H Intervention protocol
Study design	Exploratory design: Instrument development and validation, and observation study.	Descriptive design: Systematic literature review.	Descriptive design: Cross-sectional, questionnaire-based study.	Exploratory design: Instrument development and validation study.	Descriptive design: Cross-sectional.	Experimental design: Intervention trial.	Protocol for a randomised control trial.
Data type	Ordinal, scale, and nominal.	N/A	Text, survey responses.	Ordinal, scale, and nominal.	Ordinal, scale, and nominal.	Ordinal, scale, and nominal.	Ordinal, scale, and nominal.
Analysis design	Test re-test design, and within group design.	Systematic review of studies examining age-related changes in somatosensation.	Content analysis and descriptive statistics.	Test re-test design, and within group design.	Descriptive and correlations.	Within group and between group design comparing data from baseline and follow up time-points.	Within group and between group design comparing data from baseline and follow up time-points.
Statistical methods	Bland Altman plots, ANOVA.	N/A	Descriptive statistics.	Regression modelling and descriptive statistics.	Pearson product-moment correlation coefficients.	Mann–Whitney U rank-sum test, Friedman $X^2$ repeated-measures and Wilcoxon signed-rank test.	Mixed Model approach.
Sample CP/TD	N/A / N=88	N/A / 2,418	N/A / N/A	N/A / 30	N=28 / N/A	N=17 / N/A	N=50 intended / N/A
Key findings	The sense_assess© <i>kids</i> demonstrated test-retest reliability. Somatosensation increases with age	Somatosensation increases with age for TD children.	Therapists indicated low satisfaction and confidence with current somatosensory measures.	The FMT demonstrated inter and intrarater reliability. No differences were found for age,	Presence of somatosensory impairment in the upper limb of children with hemiplegic CP is	The sense© intervention is feasible in a clinical paediatric setting.	Protocol paper is predicted to outline the methods and treatment fidelity of the Sense© for Kids intervention.

Study and short title	Appendix A Reliability study	Appendix B Systematic review	Appendix C Survey study	Appendix D FMT	Appendix F Descriptive study	Appendix G Intervention pilot study	Appendix H Intervention protocol
	for TD children.			handedness or discrimination capacity.	high; with 82% having impairment.		

*Note.* ANOVA = Analysis of variance; CP = cerebral palsy; TD = typically developing.

## 1.8 Significance of the thesis

Impaired somatosensory functions in touch sensation, limb position sense and haptic object recognition can compromise upper limb and hand function. Children with CP are at a high risk of experiencing somatosensory deficits. Currently there is no gold standard assessment tool to evaluate somatosensory loss in children with CP and very few studies have investigated the suitability of existing somatosensory measures for use with children with CP. This thesis adapts an existing measure and proposes new knowledge by adapting and testing the comprehensive and standardised upper limb assessment of somatosensation with a target population of children and adolescents with and without neurological pathology. Development of a comprehensive somatosensory measure is intended to enable clinicians to measure somatosensation, and has the potential to allow for evidence based health policy by providing a foundation for clinical practice guidelines for the measurement of somatosensation in children with CP.

The *sense\_assess© kids* has been adapted for children as a measure of protective touch, tactile discrimination, wrist position sense and haptic object recognition, all of which have a direct influence on hand function. Given the gaps in research in the field of somatosensory measurement for children this thesis proposes new knowledge in relation to exploration and development of a somatosensory assessment, providing a potential catalyst to our understanding, and intervention development. This research aimed to generate evidence based frameworks for adaptation and psychometric development of paediatric assessment tools, and has the potential to inform preliminary clinical practice guidelines for the management of somatosensory deficits in children. The COSMIN (Mokkink et al., 2016) was selected as the basis for the Evidence Based Tool Development framework underpinning this research. The COSMIN describes the measurement properties that constitute a quality assessment tool in health care and provided guidelines for measuring the psychometric properties of the assessment tool.

In the past, variability in somatosensory assessment methodology has resulted in research findings that are difficult to compare, compromising the interpretation and implementation of new evidence (Taylor et al., 2016). This thesis seeks to highlight the use of systematic frameworks such as, the COSMIN when developing paediatric

outcome measures with a view to prevent such variability in the future. Research using robust somatosensory measurement in paediatrics has the potential to assist in determining the prevalence of somatosensory deficits and enable evaluation of interventions designed to ameliorate such deficits. This thesis thereby has the potential to contribute to research methods and knowledge translation in the fields of somatosensory measurement and instrument development.

Summary of potential contributions to the field:

1. Adaptation of a novel paediatric assessment tool including development of its standardised administration manual, test materials and equipment, normative data and testing of psychometric measurement properties. Future researchers could use, and continue to develop this new tool to inform future interventions and test their effectiveness.
2. Potential contribution to the understanding of somatosensory function and hand movements in TD children and children with CP.
3. Provision of evidence to support robust methods of measurement of somatosensory function.
4. Description of the application of a unique research method framework (including adaptation, psychometric development, clinical utility and implementation) that could inform the development of future clinical assessment tools and instrument development studies.

## CHAPTER 2 Scoping Review



Phase one – Knowledge inquiry and synthesis

### 2.1 Introduction to chapter

This review focused on touch, proprioception and haptic ability (please see Appendix B for a full description of terminology) and the structure of this review includes a description of the assessments' strengths and limitations, identification of the common issues associated with current somatosensory measures, a list of assessments with an original reference and a description of adult somatosensory measures potentially suitable for paediatric use. A scoping review methodology was selected to answer the exploratory research question and aim to map key concepts, types of evidence, and gaps in research related to somatosensory assessment tools (Colquhoun et al., 2014). The result was a systematic search, selection and synthesis of existing knowledge with the outcome of providing a list of recommendations.

### 2.2 Currently available measures of somatosensory capacity: A scoping review

Individuals with cerebral palsy (CP) often experience somatosensation impairment in one or more of the somatosensory domains as a result of primary neonatal or antenatal brain injury (van Haastert et al., 2011; McLean et al., 2017b). The most commonly reported areas of somatosensation affected are tactile object recognition, two-point discrimination and limb position sense (Auld et al., 2012a; Auld et al., 2012b; Curry & Exner, 1988; McLean et al., 2017b). Without a robust assessment

tool, it is difficult to evaluate treatment outcomes and the sensory functioning of children with neurological conditions (Cooper et al., 1995).

A recent clinimetric review evaluated measures of tactile registration, tactile perception and stereognosis suitable for use with children with unilateral CP and presented the currently available best practice methods (Auld et al., 2011). The clinimetric review used the following Population, Interest, and Context (PICO) (Moher et al., 2009) elements to guide study selection: Participants – Children with CP; Interest – Assessments that provided discriminative, predictive, or evaluative assessment of tactile function or perception with published clinimetric data; and Context - All assessments published in English between 1950 and November 2016. The scoping review of this thesis aimed to expand the scope of the previous clinimetric review by reporting on all current assessment methods used across the life span to measure the domains of touch and haptic ability and, in addition, proprioception. Examining all available assessment methods for children, adolescents and adults informed the development and adaptation of a suitable battery to be used with children with CP. A scoping review methodology was chosen rather than a systematic review because of the broad nature of the topic and the scope of inquiry. The scope of inquiry elements are defined below in the methods section.

### **2.3 Methods**

The methodology of this review follows the framework stages for the conduct of scoping reviews outlined in Colquhoun et al. (2014). This review was designed to include all available and relevant peer reviewed journal articles that described measures of somatosensation across the lifespan, including assessments with and without psychometric properties for their intended population. The aim was to summarise the suitability of these assessments for paediatric use. A comprehensive search strategy was developed and a search of five electronic databases was undertaken (MEDLINE, PsycINFO, PsycTESTS, Cumulative Index of Nursing and Allied Health Literature – CINAHL and SPORTDiscus) to identify relevant studies related to the scope of inquiry below. Reference lists of relevant studies were also manually screened for original citations. The pre-defined scope of inquiry headings were as follows: Concept – What are the most suitable assessment tools that could be used to measure somatosensation in children; Target population – Typically

developing individuals, or individuals with a neurological disorder causing somatosensory impairments such as CP and stroke aged from birth to 100 years; Health outcome of interest – Upper limb somatosensory assessment tools designed to measure one or more of the following domains; tactile registration, tactile discrimination, tactile perception, proprioception or haptic object recognition, with .. Exclusion criteria were studies exclusively examining somatosensory assessments of the lower limbs or trunk, sensory seeking, sensory processing or integration, visceral, vestibular, oral, auditory, olfactory or visual sensations, or assessments designed for individuals with spinal cord injury, peripheral nerve injury or chronic pain.

Published assessments as well as assessments designed for research were included in order to review all measures suitable for a comprehensive paediatric battery. The search term combination included assessment\* or outcome assessment\* or test\* or clinical assessment tool or validity or reliability or psychometric\* AND sensation or somato\* propriocept\* or limb position sense or haptic\* or haptic perception or haptic object recognition or touch\* or tactile\* or tactual or hand sensibility AND typical\* develop\* or child\* or adolescen\* or young adult\* or teen\* or young people or youth or adult\* AND cerebral palsy or stroke or neurolog\*.

## **2.4 Results**

The initial search yielded 465 articles, 14 duplicates were removed, 124 were selected by title and abstract, of these 77 articles were included for review (Figure 4). The review identified 22 different measures of touch, 19 of proprioception, 25 of haptic object recognition and 11 mixed modality assessments (i.e. test batteries with multiple somatosensory subtests).

Many assessments were common to several included articles therefore, a manual database search was conducted to identify the original author and citation of 71 relevant assessments. A summary of the assessment title, measurement focus and the original citation is provided in Table 3. Of the total 71 assessments summarised, seven had published standardised administration manuals.

Figure 4 Flowchart of study selection

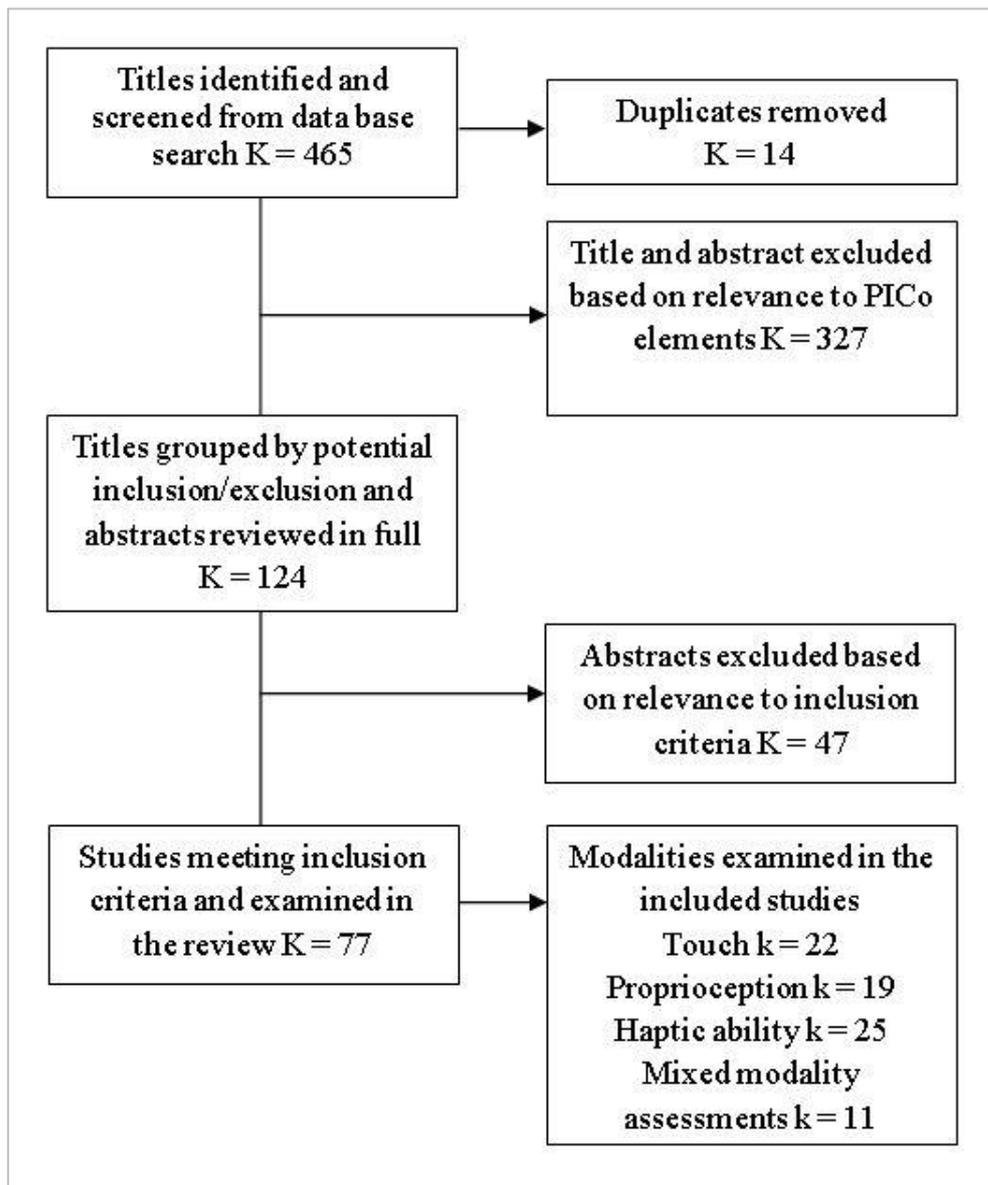


Figure 4. Flowchart to illustrate decisions made for study selection. K = total number of peer-reviewed journal articles; k = subgroup of peer-reviewed journal articles specific to different somatosensory domains.

Table 3 Summary of somatosensory measures identified in the scoping review

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
Domain of touch			
• AsTex®	Miller et al. (2009)	Texture perception	Children with CP and adults post-stroke
• Tactile Discrimination Test	Carey, Oke, & Matyas (1997)	Tactile discrimination	TD children and adults post-stroke
• Touch Test Evaluator	Stoelting (2001)	Touch sensation	Children with CP and adults post-stroke
• Semmes-Weinstein monofilaments	Weinstein (1993)	Touch sensitivity	Children with CP and adults post-stroke
• Grating Orientation Task (GOT)	Johnson & Phillips (1981)	Tactile spatial resolution	Children with CP and adults post-stroke
• von Frey filaments	von Frey (1894; 1896); Pearce (2006); Somedic Sales AB, Sweden	Touch threshold	Children with CP and adults post-stroke
• Tactile sensation of fingers	Blumenstein et al. (2015)	Tactile sensation of fingers	Children with CP
• Exteroception	Klingels et al. (2010)	Tactile sense	Children with CP
• Disk-Criminator®	MacKinnon & Dellon (1985)	Static or moving two-point discrimination	Children with CP and adults post-stroke
• Paperclip method	Moberg (1990)	Static or moving two-point discrimination	Children with CP
• Touch Perception Threshold test	Eek & Engardt (2003)	Perceptual threshold of touch, light touch sensation	Adults post-stroke
• Single Point Localization and Double Simultaneous subtests of the Neurosensory Motor Developmental Assessment (NSMDA)	Burns (1992)	Single Point Localization and Double Simultaneous	Children with CP

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
• Tactile Discrimination Test, modified from the method developed by Carey, Matyas, & Oke. (1993)	Kim & Choi-Kwon (1996)	Tactile discrimination	Adults post-stroke
• Light touch produced by a calibrated aesthesiometer	Halligan, Marshall, Hunt, & Wade (1997)	Tactile sensation	Adults post-stroke
• Touch awareness using a cotton ball or swab, fingertip or pencil eraser	Adams (1997)	General awareness of touch input	Children with CP and adults post-stroke
• Quantitative Sensory Testing using von Frey filaments	Rolke et al. (2006)	Mechanical detection threshold	Children with CP and adults post-stroke
• 30-g probe with round rubber tip 7 mm in diameter	Yoshioka, Dillon, Beck, Rapp, & Landau (2013)	Tactile localisation	TD children and children with Williams syndrome
• Moving touch-pressure (MTP) test and the sustained touch-pressure (STP) test	Dannenbaum, Michaelson, Desrosiers, & Levin (2002)	Moving and sustained touch pressure	Children with CP and adults post-stroke
• Fabric Matching Test (FMT)	Carey, Matyas, & Macdonell (2011)	Touch discrimination	Adults post-stroke
• Child Neuropsychological Assessment tactile object identification subtest	Ardila, Rosselli, Matute, & Inozemtseva (2011)	Tactile perception	TD children
Domain of proprioception			
• Wrist Position Sense Test (WPST)	Carey, Oke & Matyas (1996)	Wrist position sense	TD children and adults post-stroke
• Accurate identification of the direction of a joint motion	Hoon Jr et al. (2009)	Proprioception	Children with CP
• Ipsilateral remembered (same arm used for reference and matching targets), contralateral concurrent (reference arm moved and held at target position while opposite arm matched reference position), and contralateral remembered (reference arm moved to target position and then returned to start position before opposite arm matching position).	Langan, Kern, Hurvitz, & Brown (2014)	Limb position sense	Adults with CP

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
• The ‘Up or Down?’ test	DeGowin & DeGowin (1987)	Proprioception	Children with CP
• Automated measurement of proprioception	Leibowitz (2008)	Proprioception	Adults post-stroke
• Contralateral matching task required participants to match the position of one limb (reference limb), which was moved to the predetermined target position passively, by actively moving the other limb (matching limb) to the (mirror symmetric) position at the same distance as the reference arm.	Smorenburg, Ledebt, Deconinck, & Savelsbergh (2012)	Proprioception	Children with CP
• Recognition of the existence of movement, and (b) final position in imposed movement of a joint reported verbally or performance of the same movement with the contralateral limb.	Riquelme, Cifre, & Montoya (2011)	Proprioception	Children with CP
• Brief Kinesthesia Test	Ayres (1972; 1980; 1989)	Kinesthesia	TD children and children with sensory integration deficits
• Clinical test of wrist position	Carey et al. (1996)	Wrist position	TD children and adults post-stroke
• Proprioceptive Discrimination Test (also later known as the WPST)	Carey, Matyas, & Oke (1993)	Proprioceptive discrimination	Adults post-stroke
• KINARM robotic device	Scott (1999)	Position sense	Children with CP
• Kin Com 125 AP isokinetic dynamometer Configuration Chattanooga. Isokinetic instrument.	Chrysagis, Skordilis, Koutsouki, & Evans (2007)	Kinesthetic ability	Children with CP
• Joint-position sense and kinesthesia using a custom-built device	Wingert, Burton, Sinclair, Brunstrom, & Damiano (2009)	Joint-position sense and kinesthesia	Children with CP
• An active matching task of target positions void of visual feedback	Goble, Lewis, Hurvitz, & Brown (2005)	Proprioceptive accuracy	TD children
• A tendon vibration technique and serial pointing task	Hay, Bard, Ferrel, Olivier, & Fleury (2005)	Proprioception	TD children
• Computerized test and equipment comprised a tactile screen and a sensory stylus (for hand drawing) to measure line length	Liutsko, Muiños, & Tous-Ral (2014)	Proprioception	TD children

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
accuracy.			
<ul style="list-style-type: none"> <li>Passive mobilization of the metacarpophalangeal joints</li> </ul>	Cooper, Majnemer, Rosenblatt, & Birnbaum (1995)	Proprioception	Children with CP
	Domain of haptic ability		
<ul style="list-style-type: none"> <li>Stereognosis of 6 familiar objects</li> </ul>	Krumlinde-sundholm & Eliasson (2002)	Stereognosis	Children with CP
<ul style="list-style-type: none"> <li>Familiar (dinosaur) and unfamiliar (sea creature) models.</li> </ul>	Alexander, Johnson, & Schreiber (2002)	Haptic perception	TD children
<ul style="list-style-type: none"> <li>Sequence of differently sized plastic spheres</li> </ul>	Gori, Giuliana, Sandini, & Burr (2012)	Haptic size perception	TD children and adults
<ul style="list-style-type: none"> <li>Identification of weight via the hand</li> </ul>	Kloos & Amazeen (2002)	Dynamic touch	TD children
<ul style="list-style-type: none"> <li>The Dellon-modified Moberg pick-up test</li> </ul>	Dellon (1981)	Hand sensibility	TD children
<ul style="list-style-type: none"> <li>Perception of rod length</li> </ul>	Fitzpatrick, Carello, & Turvey (1994)	Dynamic touch	TD children
<ul style="list-style-type: none"> <li>Friction Discrimination Test</li> </ul>	Blennerhassett, Matyas, & Carey (2007)	Surface friction	Adults post-stroke
<ul style="list-style-type: none"> <li>Weight Matching Test</li> </ul>	Blennerhassett, Matyas, & Carey (2007)	Object weight discrimination	Adults post-stroke
<ul style="list-style-type: none"> <li>Hand Active Sensation Test (HASTe)</li> </ul>	Williams, Basso, Case-Smith, & Nichols-Larsen (2006)	Haptic perception	Children with CP
<ul style="list-style-type: none"> <li>Investigation of a wooden object through passive touch</li> </ul>	Van Grunsven, Njiokiktjien, Vranken, & Vuylsteke-Wauters (2003)	Manual haptic gnostic function	TD children
<ul style="list-style-type: none"> <li>Tactile object recognition using five common objects, four embossed geometric shapes, and eight embossed capital letters, and roughness discrimination</li> </ul>	Darian-Smith, & Oke (1980)	Roughness and object discrimination	Adults post-stroke

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
• Identification of common multidimensional objects	Lederman and Klatzky (1990)	Haptic object recognition	TD children
• Stereognosis using five shapes and five objects of daily use	Cooper et al. (1995)	Stereognosis	Children with CP
• functional Tactile Object Recognition Test (fTORT)	Carey, Nankervis, LeBlanc, & Harvey (2006)	Haptic object recognition	Adults post-stroke
• Brief Manual Form Perception Test modified from Ayres	Dunn (2015) adapted from Ayres (1972; 1980; 1989)	Haptic object recognition	Children with CP
• The Klingels method using 12 common objects	Klingels et al. (2010)	Stereognosis	Children with CP
• Motor enhanced tactile perception using 9 common objects	Auld, Ware, Boyd, Moseley, & Johnston (2012b); Feys et al. (2005)	Stereognosis	Children with CP
• Stereognosis using 3mm-thick plastic shapes	Bolanos, Bleck, Firestone, & Young (1989)	Stereognosis	Children with CP
• Object recognition using 11 common objects and material recognition using 10 textures	Robertson & Jones (1994)	Object and material recognition	Children with CP
• Luria-Nebraska test of Stereognosis	Golden, Purisch, & Hammeke (1979)	Stereognosis	TD children
• Manual Form Perception subtest of the Sensory Integration and Praxis Test (SIPT)	Ayres (1989)	Manual form perception	Children with sensory integration deficits
Mixed domain assessments			
• Manual Form Perception Test and graphaesthesia and kinesthesia subtests of the Sensory Integration and Praxis Test (SIPT)	Ayres (1972; 1989)	Graphaesthesia, kinesthesia, manual form perception	Children with sensory integration deficits
• Nottingham Sensory Assessment	Lincoln (1991)	Light touch, deep pressure, tactile localisation, temperature discrimination, proprioception, stereognosis and two-point discrimination	Healthy adults and adults post-stroke

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
• Revised Nottingham Sensory Assessment (rNSA)	Lincoln, Jackson, & Adams (1998)	Tactile sensation, proprioception, stereognosis, two-point discrimination	Healthy adults and adults post-stroke
• Fugl-Meyer Assessment sensory subscale (FMA-S)	Fugl-Meyer, Jaasko, Leyman, Olsson, & Steglind (1975)	Light touch and proprioception	Healthy adults and adults post-stroke
• Erasmus MC modification of the (revised) Nottingham Sensory Assessment (Em-NSA),	Stolk-Hornsveld et al. (2006)	Sharp/blunt discrimination, light touch, pressure, pinprick, proprioception and two-point discrimination	Healthy adults and adults post-stroke
• Rivermead Assessment of Somatosensory Performance (RASP)	Winward, Halligan, & Wade (2002)	Sharp/dull discrimination, surface pressure touch, surface localization, temperature discrimination, movement and direction proprioception discrimination, sensory extinction and two-point discrimination	Children with CP and adults post-stroke
• Robotic Sensory Trainer	Lambercy, Robles, Kim, & Gassert (2011)	Localization and proprioception	Healthy adults
• Upper Limb Training and Assessment program	Langan et al. (2014)	Home-based sensorimotor training program for tactile discrimination and stereognosis	Children with CP
• Tactile discrimination, joint position modified from Carey et al. (1993), pressure sensation, weight discrimination, letters tactile recognition	Smania, Montagnana, Faccioli, Fiaschi, & Aglioti (2003)	Tactile discrimination, joint position, pressure sensation, weight discrimination, letters tactile recognition	Adults post-stroke
• Sensory Assessment Battery	Cooper, Majnemer, Rosenblatt, & Birnbaum (1993)	Pressure sensitivity, two-point discrimination, stereognosis, proprioception, and directionality	Children with CP

Title of assessment or description of assessment method	Original citation	Measurement focus	Population examined in included studies
• Cotton wool and Thumb Localising Test	Welmer, Holmqvist, & Sommerfeld (2008); Hirayama, Fukutake, & Kawamura, (1999)	Light touch and proprioception	Adults post-stroke
• Stereognosis using 10 familiar objects and location of touch	Yekutieli, Jariwala, & Stretch (1994)	Stereognosis and location of touch	Children with CP
• A kinesthetic-to-visual matching task void of visual feedback to assess hand localisation	Contreras-Vidal (2006)	Kinesthesia, hand localisation	TD children

*Note.* CP = cerebral palsy; TD = typically developing

### **2.4.1 Measures of touch**

From Table 3, 20 methods of touch measurement were identified for use with children and adults, and of these 13 methods were identified for use with children with CP. The clinimetric review by Auld et al. (2011) recommended the use of the full set of 20 Semmes-Weinstein monofilaments, both static and moving two-point discrimination, and single-point localization to measure touch for children with CP because of the psychometric evidence to support their use. However, a comprehensive battery for functional sensibility was lacking in current literature.

Whether designed for clinical use or used in research with adults or children, limitations of the identified touch measures were similar. Measures of touch that had psychometric evidence related to validity or reliability were designed to measure tactile registration (initial awareness of stimuli) rather than the functional aspect of tactile discriminative capacity. Measures of simple tactile perception often involved a passive observer with stimuli applied to a passive hand such as the Disk-Criminator (MacKinnon & Dellon, 1985; Moberg, 1990) limiting the ability to ascertain functional tactile sensibility (i.e. touch perception using an active moving hand) (Carey, 1993). Other measures of touch perception such as, the moving touch-pressure or sustained touch-pressure test or double simultaneous method relied on examiner applied stimuli with no standardised methods to calibrate the pressure of multiple presentations (Dannenbaum, Michaelsen, Desrosiers & Levin, 2002; Burns, 1992; Blumenstein et al., 2015). Other measures tested domains of somatosensation within a broader assessment battery but lacked a comprehensive evaluation of touch perception and tactile discrimination therefore limiting the somatosensory profile of individuals (Fugl-Meyer, Jaasko, Leyman, Olsson, & Steglind, 1975; Lincoln, 1991; Lincoln, Jackson & Adams, 1998; Winward, Halligan, & Wade, 2002).

Measures that assessed a functional aspect of touch commonly lacked comprehensive standardisation (Yekutiel, Jariwala, & Stretch, 1994), were designed for research purposes only such as the Grating Orientation Task (GOT) (Johnson & Phillips, 1981; Kim & Choi-Kwon, 1996; Van Boven & Johnson, 1994), or were not commercially available such as the AsTex®, Sensory Assessment Battery, or Upper Limb Training and Assessment program that involved a home-based sensorimotor training program (Cooper et al., 1993; Langan et al., 2014; Miller et al., 2009;

Smania, Montagnana, Faccioli, Fiaschi, & Aglioti, 2003). The only standardised and commercially available measures of functional tactile discrimination identified were the Tactile Discrimination Test (TDT) (Carey et al., 1997) and the Fabric Matching Test (FMT) (Carey et al., 2011), however these were originally designed for use with adult stroke survivors. The TDT has also been used more recently with children although with limited psychometric data to date (Dunn et al., 2015).

#### **2.4.2 Measures of proprioception**

Of the 17 measures of proprioception in Table 3, eight studies reported measures of upper limb proprioception suitable for use with children with CP with only two electronic devices reporting psychometric evidence for this population. They were, the Kin Com 125 AP isokinetic dynamometer (Chrysagis, Skordilis, Koutsouki, & Evans, 2007) and KINARM (Scott, 1999; Dukelow, Herter, Bagg, & Scott, 2012). Four measures were subjective and consisted of passive mobilisation, direction of joint motion, recognition of the presence of movement, and contralateral matching (Cooper et al., 1995; Hoon Jr et al., 2009; Riquelme et al., 2011; Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2012). Only three measures were standardised, they were joint-position sense and kinesthesia using a custom-built device (Wingert et al., 2009), the Southern California Sensory Integration Test (SCSIT) (Ayres, 1972; Bumin & Kavak, 2010) and Proprioceptive Discrimination Test (Carey et al., 1993). Finally, three measures had evidence only for use with TD children; Brief Kinesthesia Test (Ayres, 1972; 1980; 1989), Clinical test of wrist position and Wrist Position Sense Test (Carey et al., 1996; Dunn et al., 2015). The current best practice measures of limb position sense and kinesthesia for children reported in current literature were replication of therapist imposed movements of the upper limb without vision however this method lacks standardisation and psychometrics for use with children with CP (Bentzel, 2008).

Like the limitations seen for measures of touch most proprioceptive assessments were limited in terms of published standardised procedures for positioning the upper limb during testing, commercial availability, device portability or reported psychometric properties (Ayres, 1972; 1980; 1989; Carey et al., 1996; Cooper et al., 1993; DeGowin & DeGowin, 1987; Hirayama, Fukutake, & Kawamura, 1999). Electronic devices requiring a computer interface, specialised

software or camera and motion capture system such as the kinesthetic-to-visual matching task (Contreras-Vidal, 2006; Liutsko, Muiños, & Tous-Ral, 2014) were not available for commercial use at the time of this review, while other studies reported on self-designed methods for research purposes which were not commercially available (Goble, Lewis, Hurvitz & Brown, 2005; Hay, Bard, Ferrel, Olivier, & Fleury, 2005; Hoon Jr et al., 2009; Riquelme et al., 2011; Smania et al., 2003). Several studies developed bulky custom built devices or experimental set-ups requiring motor control of the affected limb, with many lacking evidence of clinical utility (Langan et al., 2014; Chrysagis et al., 2007; Wingert et al., 2009).

Three published sensory assessments measured proprioception within a broader assessment battery and lacked the ability to provide in-depth evaluation of proprioception. They were, the Fugl-Meyer Assessment sensory subscale (FMA-S), the Revised Nottingham Sensory Assessment (rNSA) and the Rivermead Assessment of Somatosensory Performance (RASP) (Fugl-Meyer et al., 1975; Lincoln, 1991; Lincoln, Jackson, & Adams 1991; Winward et al., 2002). Traditionally, clinical measures of upper limb proprioception have required replication or matching of therapist imposed movements with the affected limb, verbal response to the passive direction of movement, or goniometers and active and passive joint angle scaling procedures (DeGowin & DeGowin, 1987; Carey et al., 1993; Cooper et al., 1995; Klingels et al., 2010; Smorenburg et al., 2012). These measures are largely subjective in nature and prone to examiner bias, without standardised procedures, and with little or no demonstrated reliability or sensitivity to detect changes over time (Carey, 1995; Carey et al., 1996). Furthermore, the majority of these measures have only been validated in adults.

Current robotic technology such as the KINARM (BKIN Technologies Ltd, Kingston, Ontario, Canada; Scott, 1999), automated measurement of proprioception (Leibowitz, 2008) or the Robotic Sensory Trainer (Lambercy, Robles, Kim, & Gassert, 2011) can provide reliable quantitative assessment of deficits in limb position sense however, they are limited in portability and are yet to establish psychometric data for a clinical paediatric population. The most widely used quantitative measure of proprioception in this review was the Wrist Position Sense Test (Carey et al., 1996). This measure has been extensively refined and updated

with improvements in commercial availability, clinical application with adults and early application with TD children (Carey et al., 1996; Dunn et al., 2015). However, its major limitation is that it was designed for use with adults post-stroke with no psychometric data available for a clinical population of children.

### **2.4.3 Measures of haptic ability**

The current review identified 21 measures of haptic ability and eight were used with children with CP as summarised in Table 3. No studies reported extensive psychometric evidence for this population. Eight measures employed common everyday objects in the testing procedure. However, small numbers of familiar objects were subject to a ceiling effect; and there could be no standardisation if objects were to be sourced by the examiner. The other 13 measures of haptic object recognition employed less familiar forms such as geometric shapes, plastic spheres or custom built devices instead of common objects therefore limiting their measurement to recognition of discrete attributes such as shape, weight, surface texture, size, or rod length (Blennerhassett, Matyas, & Carey, 2007; Bolanos et al., 1989; Fitzpatrick et al., 1994; Gori et al., 2012; Kloos & Amazeen, 2002). Test stimuli designed exclusively for research purposes such as these may impact the ability to measure the outcomes of interest in clinical practice. Auld et al. (2011) suggested the current best practice measure for haptic object recognition with psychometric evidence of reliability with children with CP was the Klingels' stereognosis method employing 12 common objects, three matched pairs of similar items and six unmatched dissimilar items (Klingels et al., 2010).

Many measures of haptic object recognition lacked ordinal scales or pairs of similar objects to enhance sensitivity (Darian-Smith & Oke, 1980; Dellon, 1981; Dunn et al., 2015). Objects were often not commercially available such as the 11 common objects used as a stereognosis measure and material recognition using 10 textures (Krumlinde-Sundholm & Eliasson, 2002; Langan et al., 2014; Robertson & Jones, 1994; Smania et al., 2003). Other measures were brief, testing domains of haptic ability within a broader assessment battery such as the Luria-Nebraska subtest of stereognosis, Nottingham Sensory Assessment (NSA) or rNSA, SCSIT or the Sensory Integration and Praxis Test (SIPT) subtest of graphaesthesia or manual from perception, or the tactile object identification subtest of the Child

Neuropsychological Assessment (Ardila, Rosselli, Matute, & Inozemtseva, 2011; Ayres, 1972; 1989; Golden, Purisch, & Hammeke, 1979).

The functional Tactile Object Recognition Test (fTORT) (Carey et al., 2006) is a standardised ordinal scale measure of haptic object recognition with 42 common objects categorised into 14 sensory related object sets. An ordinal scale increases the test's sensitivity to detect changes over time which has been lacking in current measures of haptic object recognition (Auld et al., 2012b). In its application to paediatric populations its main limitation was that it was designed for use with adults post-stroke and had no normative data for children nor had its psychometric properties assessed in children.

## **2.5 Discussion**

This scoping review aimed to identify the most suitable assessment tools that could be used to measure somatosensation in children. What this scoping review found was that there were no comprehensive assessment tools suitable for use in their current form and that the best practice methods described by Auld et al. (2012b) should still be used. We discuss the findings as they relate to the study purpose but first we discuss implications related to clinical practice considering the obvious limitations that were present throughout the results of this review. We identified six common limitations of the identified somatosensory measures regardless of the domain being measured or the population they were designed for: i) a lack of standard procedures; ii) designed for research purposes with variability in assessment methods; iii) limited evaluation of clinical utility; iv) limited ability to measure somatosensory constructs comprehensively; v) limited as measures of functional hand sensibility; vi) lack of normative data or psychometric evidence for use with children.

### **2.5.1 Standardisation**

The most frequent limitation of assessments identified in this review was a lack of standard administration procedures, rendering measures subject to administrator and patient bias (Bentzel, 2008; Cooper et al., 1995). A standardised protocol ensures a consistent administration procedure with optimal environmental conditions, and enhances the reliability and validity of assessments (Corr & Siddons, 2005).

### **2.5.2 Measures designed for research purposes**

It has been recognised that the use of similar assessment methodologies in research aids the potential for comparison of findings and meta-analyses (Connell & Tyson, 2011). Many studies in this review used varying assessment methods often designed for research purposes only and were not commercially available. Treatment efficacy and appraisal of assessment tools cannot be based on a body of clinical-outcome research that is heterogeneous in terms of methodological quality and content (Robey & Schultz, 1998).

### **2.5.3 Clinical utility**

The clinical utility of a tool including special requirements for equipment, training costs, interpretation of data, assessment format and time required to complete the assessment are key factors that support translation of new assessment methods into clinical practice (Metzler & Metz, 2010; CanChild, 2004). Although robotic technology is emerging as a robust and accurate means to measure proprioceptive ability, a compact unit is not available yet (Rinderknecht, Popp, Lambercy, & Gassert, 2016). Many of the electronic measures requiring special equipment and software are not yet portable or commercially available and may be costly and/or require specialist training.

### **2.5.4 Comprehensive assessment**

Using a consistent and comprehensive somatosensory assessment methodology can provide more accurate information about somatosensory domains during rehabilitation, enhancing their ability to measure change due to treatment (Wu, Chuang, Ma, Lin, & Chen, 2016). Four levels of a comprehensive somatosensory measurement have been outlined: detection (noticing stimuli), discrimination (distinguishing among stimuli), scaling (grading stimuli), and object recognition (knowing what the object is by touch) (Borstad & Nichols-Larsen, 2014). Somatosensory subtests within a broader assessment battery identified in this review may lack the ability to measure each domain comprehensively. The only measures of somatosensation identified in the present review that included the four domains of somatosensory measurement were validated for adults post-stroke and TD children by Carey and colleagues (Carey et al., 2006; Carey et al., 2011; Carey et al., 1996; Carey et al., 1997; Dunn et al., 2015).

### **2.5.5 Non-functional somatosensory assessments**

The relationship between static touch detection measures and functional sensibility is often limited. Functional sensibility involves alternative processing and use of somatosensory information, often at the level of discrimination and object recognition (Carey, 1993). There are noted difficulties in determining a child's functional somatosensory ability from a measure of impairment (Richards & Malouin, 2013). This review found that many measures of tactile registration and perception, and of proprioception examined sensitivity to passive touch with no active movement of the hand.

### **2.5.6 Lack of normative standards and psychometric evidence**

Current somatosensory measures often lack normative data for paediatric populations with the majority having been designed for use with adults (Taylor et al., 2016), and for the same reason their psychometric properties are seldom available for children.

## **2.6 Recommendations**

This review identified no gold standard somatosensory measure for use with children. The current recommendation is to continue to use the best practice methods for touch and stereognosis described by Auld et al. (2012b), however there is a need to develop comprehensive functional somatosensory batteries inclusive of the functional aspects of touch, proprioception and haptic ability (Connell & Tyson, 2011; Cooper et al., 1993). It is proposed that a possible solution would be to adapt an adult measure with extensive published psychometric data. The most quantitative, comprehensive, commercially available, functional, standardised and psychometrically tested somatosensory measures used in research with children, and therefore the most suitable for adaptation, were the Semmes-Weinstein monofilaments, TDT (Carey et al., 1997), fTORT (Carey et al., 2006), WPST (Carey et al., 1996) and the FMT (Carey et al., 2011) a measure of texture discrimination. The 4.56 protective Semmes-Weinstein monofilament of the Touch-Test® Sensory Evaluators (Touch-Test® Sensory Evaluators, 2001), TDT, fTORT and WPST make up four components of the sense\_assess© test battery. The sense\_assess© was designed to measure somatosensory capacity of the upper limb in adults post-stroke (Carey et al., 2011). The FMT is a valid measure of texture discrimination for adults,

but currently it is not part of the sense\_assess© test battery. The TDT was chosen as the tactile discrimination measure of the sense\_assess© over the FMT due to its durability and because plastic surfaces can be reproduced to exact specifications using a programmable laser cutting machine, whereas fabric materials cannot.

The Semmes-Weinstein monofilaments have excellent test re-test reliability for children with hemiplegic CP (ICC = 0.96 for the impaired hand; ICC = 0.90 for the unimpaired hand) (Auld et al., 2012b). The sense\_assess© uses the red monofilament (4.56 borderline protective sensation) of the Touch-Test® Sensory Evaluators to screen touch detection at 30 various body regions, including face, forearm, hand and lower limb prior to administering the other subtests (Weinstein, 1993; Stoelting, 2001).

The TDT provides a quantitative measure of discriminative touch sensation, measuring the ability of the index finger to discriminate between finely graded plastic textures using a three-forced choice design with vision occluded. The measure has high ( $r = .92$ ) retest reliability, good discriminative properties within a population of adults post-stroke, and normative standards for age-matched unimpaired adults (Carey, 1995; Carey et al., 1997).

The FMT (Carey et al., 2011) is a measure of texture discrimination measuring the ability of the index finger to identify differences in material textures. The FMT consists of two identical fabric disks, covered in ten standard cotton-based fabric surfaces ranked 1 to 10, smoothest to roughest. Participants were required to match two fabrics as closely as possible. This measure has high test-retest reliability ( $r = .85$  to  $.92$ ) and good discriminative validity in an adult stroke population (Carey & Matyas, 2005).

The fTORT is designed to test recognition of objects through the sense of touch without vision. The test objects have been chosen to measure discrimination of texture; hardness; temperature; weight; shape; size and properties related to the object's function (Carey et al., 2006; Lederman & Klatzky, 1987). The fTORT consists of forty-two numbered objects depicted on a poster, fourteen test objects for manipulation and five display objects to provide context for size. The objects are common, everyday objects and each three-object set consists of an exact object match,

a comparator object that differed in one sensory attribute e.g. weight, and a distractor object that differed in more than one sensory attribute e.g. weight and shape. The FTORT has age-adjusted normative standards, high reliability ( $r = .85$  to  $.92$ ) and good discriminative properties for adults aged 21 to 79 years (Carey et al., 2011).

The WPST is a quantitative measure of a person's capacity to indicate knowledge of wrist position following examiner imposed movements at the wrist (Carey et al., 1996). The assessment equipment consists of wrist and forearm splints, and protractor scales and a pointer system. The examiner moves the splinted wrist to one of 20 predetermined positions and asks the participant to move the visible pointer to the position of their splinted wrist which is occluded from view. The WPST has age-adjusted normative standards, high reliability ( $r = .85$  to  $.92$ ) and good discriminative properties for adults aged 21 to 79 years (Carey et al., 2011; Carey et al., 1996). The measure has also been used with TD children (Dunn et al., 2015).

Due to the current lack of somatosensory measures for children, the strong psychometric properties of these tests for use with adults, and stroke survivors, and the acceptability of the WPST and TDT with TD children (Dunn et al., 2015; Carey et al., 1996) the sense\_assess© was deemed the most appropriate candidate measure of somatosensation in a paediatric population aged 6 to 15 years<sup>2</sup>. Findings from this scoping review pointed to the need for a guiding overall research question; Is the sense\_assess© a suitable tool with which to assess somatosensory ability in children and youth with cerebral palsy? To address the limitations of current measures identified in this review the adaptation process needs to include the following steps: standardisation of the instructions and test procedure for a paediatric population; development of the assessment as a clinical tool with clinical acceptability for end users; confirmed financial sponsors to support commercial reproduction; maintained the comprehensive subtests of protective touch, tactile discrimination, wrist position sense and haptic object recognition; the functional aspects of the subtests were also maintained and adapted for use with children with CP; normative data was collected in an age-matched TD population and the psychometric properties were measured in a clinical population.

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<sup>2</sup> Please see Chapter three for a full description of the adaptation process.

## **Limitations of scoping review review**

While this scoping review provides a comprehensive summary of current somatosensory measures, findings must be interpreted in the context of its limitations. This scoping review was designed to provide a summary of the topic prior to a clearly defined clinical question so risk of bias was not addressed with more than one reviewer. Another limitation was the difficulty making a conclusive statement about existing psychometric evidence for children with CP. This review only considered the psychometric evidence reported in the included studies, therefore psychometric evidence may exist for some assessments elsewhere but it was beyond the scope of the current scoping review to report on this extensively.

## **2.7 Conclusion**

This review identified current assessment methods related to the somatosensory domains of touch, proprioception and haptic object recognition, identifying six common issues related to somatosensory measurement tools. It is recommended that the six limitations of current measures be considered in the planning stages of tool development to produce high quality study designs. By examining all assessment methods for children, adolescents and adults a battery potentially suitable for use with children with CP was identified. The results from this review can also be used to inform best practice guidelines for the measurement of somatosensation in children with CP.



## CHAPTER 3 Adaptation



Phase two – Evidence based tool adaptation

### 3.1 Introduction to chapter

The *sense\_assess*© is designed to measure body functions, not impairment, thus the focus of measurement is on the capacity for performance, not what caused the disability (World Health Organization, 2001). Using a conceptual framework based on the work of Law (1987) and Law, Baum and Dunn (2005), an outcome measure adaptation protocol was developed for the adult *sense\_assess*©. The work of Randall, Imms and Carey (2008) also closely informed the adult-to-paediatric adaptation protocol, who successfully modified and established evidence for the use of a modified version of the Melbourne Assessment of Unilateral Upper Limb Function for a younger age group than it was originally designed.

The aim of the approach outlined in this chapter was to: i) synthesise and extend the existing frameworks; ii) establish the appropriateness of the current adult-to-paediatric adaptation protocol; and iii) use this knowledge to modify the adult *sense\_assess*© for paediatric use i.e. *sense\_assess*© *kids*. Employing an evidence based framework early in tool development is crucial to implementing high quality research and achieving positive outcomes for an assessment tool under investigation (Law et al., 2005). Well-designed outcome measures can determine the efficacy of interventions and can detect clinically meaningful changes in function over time (Corr & Siddons, 2005). To do this accurately it is crucial that the measurement tools

used by health professionals are reliable, accurate and demonstrate utility in clinical practice (Corr & Siddons, 2005).

When adapting an existing adult tool for paediatric use, prior to psychometric testing outlined in the COSMIN, the Outcome Measures Rating Form (OMRF) and accompanying (OMRF) guidelines (CanChild, 2004) can be used as a guide. The OMRF resources were designed to critically review health related outcome measures and improve evidence based selection by determining the scientific rigor and utility of a measurement instrument (Law, 1987). The OMRF framework complements the COSMIN because it contains a similar taxonomy of psychometric properties, with the addition of clinical utility.

The OMRF was used in the current chapter to inform the development of the adaption protocol for the sense\_assess©. The OMRF framework consists of 1) *Primary purpose* of the assessment including a clear focus of the attribute to be measured using the International Classification of Functioning (ICF) (World Health Organization, 2001); 2) *Clinical utility* including clarity of instructions and format; 3) *Scale construction* including adequate selection of constructs, and level of measurement e.g. interval or ordinal; 4) *Standardisation* including the administration manual and test equipment; 5) *Normative data*; 6) evidence of *Reliability*; 7) *Validity*; 8) *Responsiveness* and 9) *Overall utility*. In this thesis, the elements of the OMRF framework were not intended to be applied in sequential order rather, they were interwoven through different stages of adaptation. Using the OMRF framework to guide adaptation ensured that there was sufficient evidence to support the tool for paediatric use, and to help determine the tools value and usefulness in clinical practice.

### **3.2 Previous adaptation by team**

To determine if the adult sense\_assess© could be successfully used with children aged 6 to 15 years early pilot work was undertaken with children without neurological impairment. This work was not part of the current thesis and occurred as part of an honours project completed by the PhD candidate prior to the current doctoral research (Appendix A). The adaptation was for the SenScreen© Sensory Screening Tool and involved removal of subtests that were related to the daily activities of adults; the Sensory Questionnaire (Carey et al., 2011) and Hand

Function Survey (Blennerhassett, Avery, & Carey, 2010) and removal of subtests that were not related to the somatosensory domains under examination for children such as the Hot and Cold Discrimination Test (Roylan® Hot Cold Discrimination Kit) and the Jebsen Taylor Hand Function Test (small objects) (Jebsen, Taylor, Trieschmann, Trotter, & Howard, 1969). The functional Tactile Object Recognition Test (fTORT) contains an accuracy component (score out of 42 correct) and a timed component (duration in seconds of haptic exploration before response is given). The timed component of the fTORT was removed as it was not deemed a robust indicator of haptic object recognition capacity (see Chapter eight).

Figure 5 Summary of adaptation

#### Honours degree: Previous adaptation by PhD candidate and team

- SenScreen© Kids adaptation
- Minor modification of instructions and subtests
- Early pilot testing
- Reliability testing
- Preliminary normative data

#### Current PhD: Adult-to-paediatric adaptation protocol

- *sense\_assess*© *kids* adaptation
- Comprehensive review of existing measure
- Measurement constructs described using the International Classification of Functioning
- Clinical expertise used to establish content validity
- Detailed modification of instructions, materials and equipment
- Normative data
- Psychometric development in a clinical population

Minor modifications of test instructions were made, the word ‘kids’ was included in the assessment title to differentiate from the adult version, intrarater reliability was measured and initial data on typical performance were collected (Appendix A). The *sense\_assess*© contains the same four subtests as the SenScreen©, but with extended test forms summarised as follows: 16 test stimuli specific to the hands for the Protective Touch Test (PTT) instead of the original stimuli for multiple body regions (Taylor et al., 2017f; Touch-Test® Sensory Evaluators, 2001); presentation of 20 textures instead of the brief version of 12 for the Tactile Discrimination Test (TDT) (Carey, Oke, & Matyas, 1997); identification of 14 test objects instead of the brief version of 7 for the fTORT (Carey, Nankervis, LeBlanc, &

Harvey, 2006); and 20 different test angles were presented instead of the brief version of 10 for the Wrist Position Sense Test (WPST) (Carey, Oke, & Matyas, 1996).

### 3.3 Adult-to-paediatric adaptation protocol

The adult-to-paediatric adaptation protocol (Figure 6) was developed to avoid the common limitations experienced in current somatosensory measurement identified by the scoping review in Chapter two. The adult-to-paediatric adaptation protocol was intended to be used in sequential order as follows; a) comprehensive review of the existing measure (sense\_assess©) and investigation of the relevant somatosensory constructs to be measured in children with CP described using the ICF (World Health Organization, 2001); b) use of clinical expertise to examine content validity; c) modification of equipment; d) modification of instructions and e) pilot testing to determine if further changes were required (Please see Paper three, Chapter six, for an example of adaptation to the fTORT instructions to a paediatric version).

Figure 6 Adult-to-paediatric adaptation protocol



Figure 6. Graphic representation of the stages of adaptation formalised as part of this thesis and informed by “Establishing validity of a modified Melbourne Assessment for children ages 2 to 4 years,” by M. Randall, C. Imms and L. Carey, 2008, *American Journal of Occupational Therapy*, 62, p. 376 and “Outcome Measures Rating Form Guidelines,” by CanChild Centre for Disability Research, 2004, Ontario, Canada.

### **3.3.1 Comprehensive review**

This stage was informed by the OMRF framework elements of Primary purpose, Scale construction, Reliability, Validity, and Normative data. All available published literature and grey literature were sourced from electronic databases related to the sense\_assess© and the component measures (Chapter 2). In addition, personal communication with the original authors provided raw data related to individual subtests and an opportunity to discuss tool development and adaptation. In this stage, the relevant somatosensory constructs to be measured in children were described using the International Classification of Functioning, Disability and Health – Child and Youth Version (ICF-CY) and the Comprehensive ICF Core Set for children & youth with cerebral palsy (CP) from birth to 18 years of age (Schiariti, Selb, Cieza, & O' Donnell, 2015; World Health Organization, 2007). In the domain of body functions individuals with somatosensory impairment may exhibit changes in b 156 perceptual functions (specific mental functions of recognising and interpreting sensory stimuli) including b 1564 tactile perception, and sensory functions including b 260 proprioceptive, b 265 touch function and b 270 sensory functions related to temperature, vibration, pressure and noxious stimuli.

### **3.3.2 Clinical expertise and peer review**

This stage was informed by the OMRF framework elements of Primary purpose, Clinical utility, and Overall utility. An initial draft of the modifications intended for the sense\_assess© were distributed to three expert clinicians each with more than 10 years of experience in paediatrics from the Paediatric Rehabilitation Department of PMH, Perth, Australia. They included two senior occupational therapists specialising in hand therapy and upper limb rehabilitation in CP and a senior speech pathologist. Feedback from the clinicians was incorporated and pilot testing of the adapted assessment was undertaken with five TD school-aged children aged 6 to 11 years, a parent and another senior occupational therapist.

### **3.3.3 Modification of equipment**

This stage was informed by the OMRF framework elements of Standardisation and Overall utility. The process involved design and fabrication of smaller test equipment in conjunction with the original author and test developer, which was piloted with five TD school-aged children aged 6 to 11 years. Paediatric specific instructional

pictures, to be included in the assessment manual, were taken as part of a professional photo shoot by the Medical Illustrations Department at CAHS.

### **3.3.4 Modification of instructions**

This stage was informed by the OMRF framework elements of Standardisation and Clinical utility. The language used in the sense\_assess© instructions was unlikely to be easily understood by young children without modification because of the influence of cognitive development on receptive language skills for children aged between 6 and 15 years old. Adaptation of instructions was based on consideration of semantics and perceptual concepts appropriate to developmental level (Berlin, Blank, & Rose, 1980; Bloom, 2001). This process included standardising test instructions and modifying the assessment manual procedure for a younger clinical population that often experience intellectual difficulties as a comorbidity of CP (Maenner et al., 2016). The instruction modification process incorporated feedback from parents and children and was overseen and reviewed by a senior speech pathologist and senior occupational therapist both experienced in working with children with CP. The senior speech pathologist provided feedback about the language used and level of instruction and demonstration needed during administration. Feedback was also obtained from the originator of the tests to ensure basic procedures and content were adequately represented.

### **3.3.5 Pilot testing**

This stage was informed by the OMRF framework elements of Clinical utility and Overall utility. Pilot testing of the adapted assessment occurred with five typically developing school-aged children (6 to 11 years old). The pilot phase involved full administration of the adapted sense\_assess© and information was recorded concerning administration duration, time needed to set up each subtest, and appropriateness of the age-related physical dimensions of the modified equipment. This stage established the content validity, and applicability of the adapted sense\_assess© to a paediatric population.

### 3.4 Description of sense\_assess© subtests and the adaptation process<sup>3</sup>

#### 3.4.1 Protective Touch Test description

The purpose of the PTT is to screen for the ability to detect touch, close to the normal threshold for protective sensation. The test uses the red monofilament (4.56 borderline protective sensation) of the Touch-Test® Sensory Evaluators (Touch-Test® Sensory Evaluators, 2001). The monofilament is applied to the skin at a 90° angle until it bows to form a ‘C’ shape and held for 1.5 seconds (Figure 7).

Figure 7 Monofilament 'C' shape

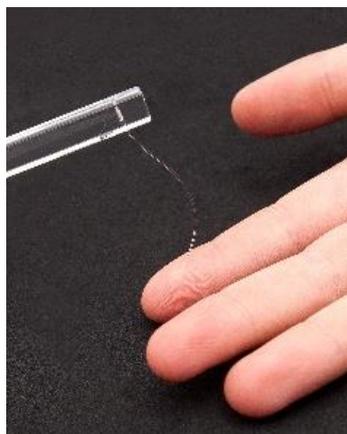


Figure 7. Photo depicting the 4.56 monofilament being pressed against the index finger to form a ‘C’ shape, the monofilament is pressed for 1.5 seconds.

#### 3.4.2 Protective Touch Test adaptation

The adult test version of the PTT involved testing at various body regions, including the face, forearm, hands and lower limb. The paediatric version was modified to screen touch detection at 16 regions on the hands. Eight stimuli are applied per hand with four stimuli on the palmer aspects and four on the dorsal aspects (Figure 8). Regions were based on the common testing sites recommended in the Touch-Test® administration manual and consideration of the digits used for functional grasp.

The procedure was modified by removing the prompt, *‘I am going to alert you to each trial by saying, “Now”’*. This prompt warned when the stimuli would be administered and provided an opportunity to give a false response or guess rather than identification. The prompt was replaced by, *‘Say, “Yes” when you feel the*

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<sup>3</sup> Please see Appendix E sense\_assess© kids Administration manual for a full description of subtests including scoring and interpretation.

*string touching you*' thereby also eliminating the need for the sham trial. The test form was modified by replacing the representative picture of the body regions with a picture of the areas of the hands to be tested.

Figure 8 Hand regions tested in the Protective Touch Test

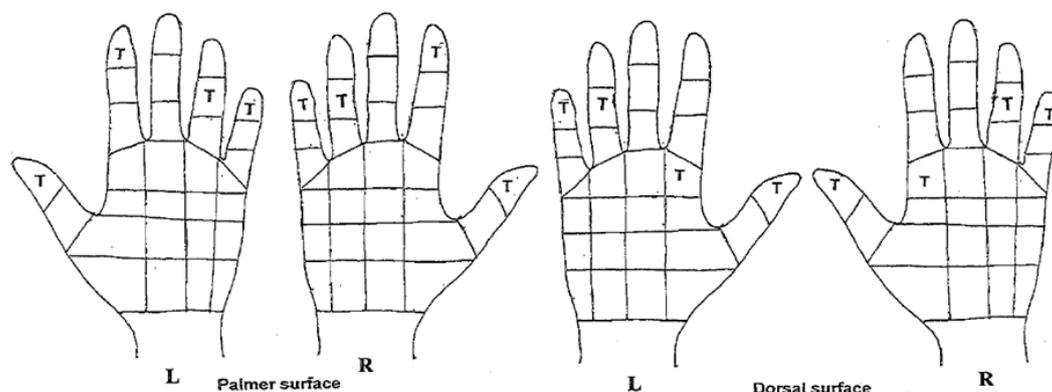


Figure 8. Illustration depicting the 16 sites on the dorsal and palmer aspects of the hands, symbolised by a 'T', that are tested within the Protective Touch Test.

### 3.4.3 Tactile Discrimination Test description

The TDT measures the ability to discriminate between finely graded plastic textures using a three-alternative forced choice design (Figure 9). The participant is asked to place their hand in a prone position through a frame and curtain so they cannot see their hand (Figure 10). The administrator guides the index finger of the tested hand across different textures one at a time at a constant speed and pressure. The texture grid has rows of three where two textures are the same and one is different. The task is to discriminate the texture that is different, i.e. the odd one out. The administrator verbalises *'first'*, *'second'* or *'third'* as the participant's index finger is guided over the textures. The level of tactile discrimination is determined by ability to verbalise or place their finger on the different texture in each row.

### 3.4.4 Tactile Discrimination Test adaptation

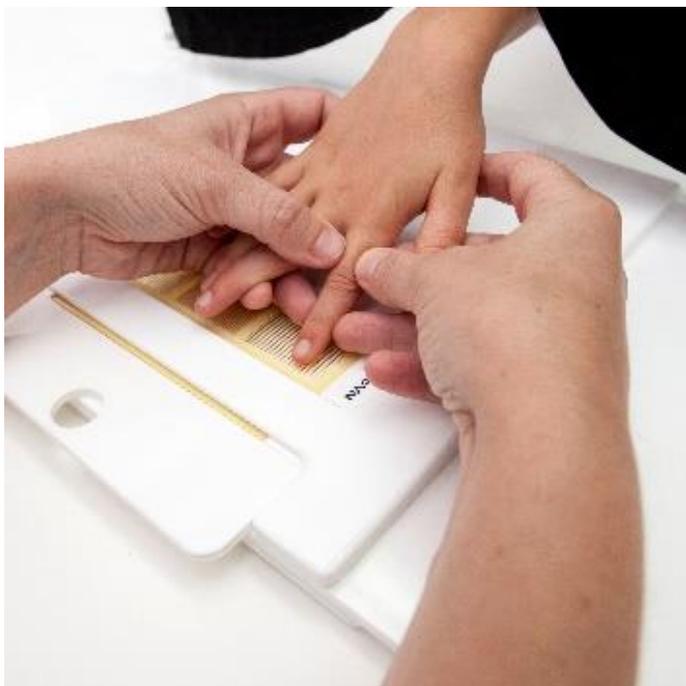
A paediatric version of the TDT equipment was already created by the original author (L. Carey) where the length of the base plate was shorter allowing the participant to reach the textures easily without stretching. As part of the current thesis the test instructions were simplified to include terminology familiar to developmental level such as explaining the word 'texture' as 'lines' and replacing 'finer' with 'smoother'. The plastic grating with the smallest difference compared to

the reference texture was removed as this difference was deemed too difficult for children to discriminate and had shown to be too difficult for adults post-stroke (L. Carey, personal communication, November 18, 2013).

Figure 9 Tactile Discrimination Test equipment



Figure 10 Standardised presentation of textures



### **3.4.5 Wrist Position Sense Test description**

The WPST assesses the capacity to indicate knowledge of wrist position following imposed movements at the wrist. The test involves matching of 20 predetermined imposed wrist positions in the flexion-extension range. The assessment box consists of vision occluded thermoplastic wrist and forearm splints attached to a visible pointer and protractor system (Figures 11, 12). The examiner secures the participant's hand firmly into the forearm and hand splints with the wrist in neutral position and elbow flexed at 90 degrees. The hand splint is attached to the lever in a position that ensures standardised movement at the wrist relative to the axis of movement (Figure 12).

### **3.4.6 Wrist Position Sense Test adaptation**

The physical dimensions of the adult test equipment were not suited to all paediatric participants in the pilot phase. Therefore, measurements of forearm and hand size were taken of the pilot participants aged 6 to 15 years and were used to fabricate a smaller version of the WPST apparatus and small, medium and large thermoplastic splints. The procedure was further modified to accommodate contracture or spasticity of the upper limb by including foam padding or instructions for physical assistance to help secure the wrist and forearm firmly but safely to ensure standardised positioning. The test positions were not adapted however, the test protocol now included a step of establishing the maximum flexion and extension capacity for movement at the wrist for children. This modification was used to establish the flexion-extension capacity of the participant's wrist before completing the pre-determined flexion-extension test positions.

The test instructions were modified to include, *'Can you feel me moving your hand? How comfortably can you bend your hand this way?'* The maximum range of movement was recorded from the pre-test trial and test angles were reduced if they were going to exceed the maximum range. Modified test angles were recorded on the test form and scoring and interpretation remained the same. The test instructions were modified to include terminology familiar to developmental level such as, *'Indicate the position that matches the position of your hand'* was modified to, *'Show me where you think your hand is pointing'*.

Figure 11 Wrist Position Sense Test equipment

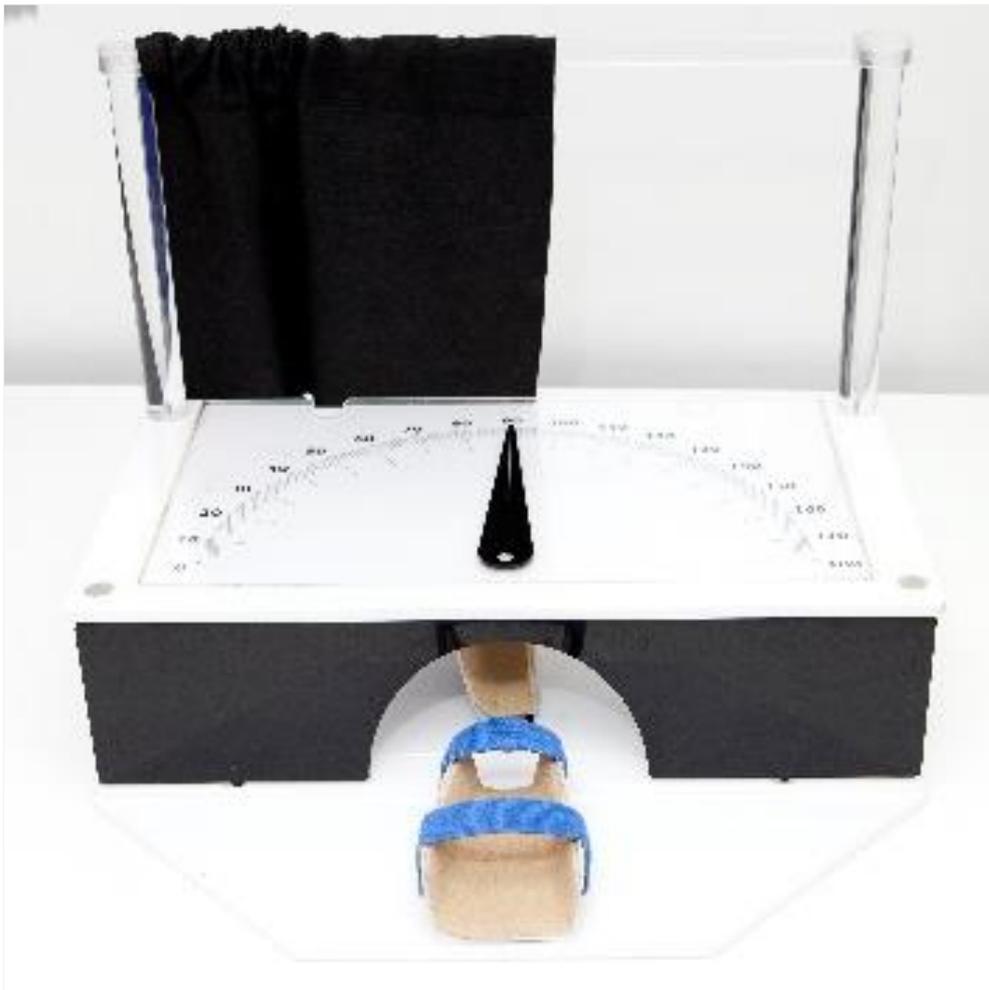


Figure 12 Administration of Wrist Position Sense Test



### 3.4.7 functional Tactile Object Recognition Test description

The fTORT was designed to test recognition of objects through the sense of touch. The kit contains a numbered response poster of 42 common everyday objects, 28 of these are potential manipulatives to be handled by the participant and only 14 are presented at any one time. There are four different test forms with 14 objects that are combinations of the 28 manipulatives with the aim of avoiding a learning effect if repeated assessment is required. The other 14 are distractor objects represented on the numbered response poster only. The 42 objects on the numbered response poster are ordered into 14 sets of three objects; an exact object match, a comparator object that differs in one sensory attribute e.g. weight, and a distractor object that differs in more than one sensory attribute e.g. weight and shape (Figure 13). Participants are tested by placing their hand in supine position through a frame with a curtain (Figure 14). The numbered response poster is placed in front of the participant and one of the 14 test objects is placed in the hand in a pseudo-random order specified on the test form. The participant is then asked to identify what they are holding by choosing the corresponding object on the numbered response poster. Fourteen test objects are provided for manipulation and responses and methods of haptic exploration are recorded on the test form.

Figure 13 functional Tactile Object Recognition Test poster of objects

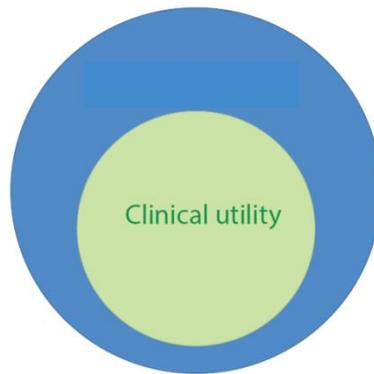


Figure 13. The functional Tactile Object Recognition Test poster contains common objects numbered from 1 to 42. Object sets appear in consecutive order in groups of three.





## CHAPTER 4 Clinical Acceptability



Phase three – Psychometric testing using the COSMIN

Phase four – Foundations for implementation

### 4.1 Introduction to chapter

Although clinical acceptability is not classified as a psychometric property of an assessment tool it is included in Phase three because testing clinical acceptability is crucial when determining a tool's suitability for specific clinical environments. The adaptation protocol of the previous chapter ensured that a systematic process was followed when modifying the *sense\_assess*© for paediatric use. In the present chapter the adapted test battery, the *sense\_assess*© *kids*, was empirically evaluated for use with the intended clinical population of children and adolescents. Evaluation of consumer acceptability cannot be neglected in the tool development process (Yuen & Austin, 2014). Involving consumers (patients and clinicians) and the community (public) in health and medical research, particularly in the planning and decision making phase is emerging as best practice for research involving human participants (McKenzie, Alpers, Heyworth, Phuong, & Hanley, 2016). Increasing awareness of consumers and community members in research, changing attitudes to future implementation of activities and methods of involvement are part of a science promoting the value of consumer participation in research (Tallon, Chard, & Dieppe, 2000; McKenzie et al., 2016).

In the current chapter steps were taken to evaluate consumers' perspectives after completing the *sense\_assess*© *kids*. Unfortunately, the consumers were not able to be involved in the planning of the research as the assessment tool had already been

established in the adult population, and psychometric testing adhered to a formalised process. The following paper investigated the clinical acceptability of the sense\_assess© *kids* following the adaptation process and prior to psychometric testing. Involving stakeholders in the process provided knowledge and personal experiences which helped to shape further modifications to the sense\_assess© *kids*' administration procedure advancing the sense\_assess© *kids* as a clinically useful assessment tool.

## **4.2 PAPER ONE: Clinical acceptability**

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### **Clinical acceptability of the sense\_assess© kids: children and youth perspectives**

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## **Abstract**

**Introduction:** The sense\_assess© *kids* is an evidence-based assessment designed to measure the functional somatosensory capacity of the upper limb of children with cerebral palsy (CP). The objective of the current study was to determine if the sense\_assess© *kids* was clinically acceptable to children and youth.

**Methods:** A questionnaire was completed by participants following administration of the sense\_assess© *kids* by a trained occupational therapist. Twenty-six children with spastic hemiplegic CP (aged 6y - 15y 6m; mean 10y 8m; 16 boys) were recruited. Participants responded to questions regarding the administration and level of difficulty of the sense\_assess© *kids* using a Q-Sort of 'like' and 'dislike', Likert scales and short answers. Content analysis was applied.

**Results:** Twenty-one of twenty-six children, indicated they were 'very happy' or 'happy' with the administration process of the sense\_assess© *kids*. Most participants indicated that they liked the sensation they felt in the hand when tested.

**Conclusion:** This study has demonstrated the acceptability of sense\_assess© *kids* for the population for whom it is intended.



## **Introduction**

Somatosensory function is the ability to detect, recognise and discriminate body (somato) sensations (Carey, 2012). The somatosensory system allows us to interpret sensory messages received from our body and consists of sensory receptors located in the skin, tissues, and joints; the nerve cell tracts in the body and spinal cord; and brain centres that process and modulate incoming sensory information (Puce & Carey, 2010). It contributes to our capacity to continually detect changes in the external and internal environment (Marieb, 2007). The somatosensory system receives information relating to limb movement and position (kinaesthesia and proprioception), touch (tactile registration and tactile discrimination), and perception of temperature, pain and itch (Barker, 2008; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2008). Somatosensory inputs are also involved in more complex central nervous system processes such as haptic object recognition (Carey, 2012).

Somatosensory capacity greatly influences the achievement of functional hand skills for children and youth (Auld, Boyd, Moseley, & Johnston, 2011; Dunn et al., 2015). Somatosensation enables skilful manipulation, precision grip, anticipatory control and effective fine movement of the hand (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Majnemer, Bourbonnais, & Frak, 2008; Lundy-Ekman, 2002). Haptic object recognition involves in-hand manipulation and tactile and proprioceptive feedback to identify sensory attributes such as texture, size, weight or the shape of objects (Overvliet, Smeets, & Brenner, 2008). Recognition of objects using tactile and proprioceptive feedback is particularly important when the hands are occluded from vision (Carey, 2012; American Occupational Therapy Association, 2002).

Recent studies document the maturation of somatosensory function across early childhood to adolescence (Dunn et al., 2015; Gori, Giuliana, Sandini, & Burr, 2012; Mallau, Vaugoyeau, & Assaiante, 2010). If the pathways for receiving afferent somatosensory feedback are damaged, maturation of the somatosensory system and subsequent upper limb function may not develop as expected (Sakzewski, Ziviani, & Boyd, 2010; van Haastert et al., 2011). The neural pathways subserving somatosensory function pass through areas of the brain that are commonly injured in the developing brain in children with cerebral palsy (CP) (Hoon Jr et al., 2009).

The revised definition of CP published in 2007 clarified that the movement disorder was frequently accompanied by a number of other disorders such as, disturbances in sensation, communication, cognition, perception, and/or behaviour, and/or by an accompanying seizure disorder (Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). Somatosensory deficits can impact self-care, such as eating and dressing, and leisure activities like team sports, resulting in restrictions in activity and participation for children with CP (Koldoff & Holtzclaw, 2015). In current literature, somatosensory deficits are reported between 31% and 97% of children with CP (Auld et al., 2011; Majinemer, Bourbonnais, & Frak, 2008; McLean, Taylor, Valentine, Carey, & Elliott, 2017a; Sakzewski et al., 2010).

The wide range of prevalence reported may be impacted by use of a variety of different assessment tools, particularly their sensitivity to deficits, and the domain of somatosensation under examination; as it is in other neurological conditions, such as stroke (Carey & Matyas, 2011). The most commonly reported areas of somatosensation affected are tactile discrimination, object recognition, and proprioception (Auld, Boyd, Moseley, Ware, & Johnston, 2012). Traditionally, assessments for children with CP have focussed on motor function and the evaluation of somatosensory capacity is underdeveloped (Auld, Boyd, Moseley, & Johnston, 2011). Due to the impact of sensation on function and the number of children with CP with somatosensory disturbances, a comprehensive somatosensory assessment to plan and evaluate therapeutic interventions is required (Rosenbaum et al., 1990; Sakzewski, Boyd, & Ziviani, 2007).

Despite the recognised need to measure somatosensory functioning, there is little use of standardised outcome measures in clinical practice (Auld et al., 2011; Pumpa, Cahill & Carey, 2015; Taylor et al., 2016; Walmsley et al., 2017). Clinical pressures, including time constraints, expanding caseloads, lack of assessment accreditation, and limited perceived utility may negatively influence the use of outcome measures by occupational therapists (Bowman & Llewellyn, 2002). Assessment of somatosensory capacity in children with CP requires evaluative measures with specific structural characteristics that are clinically useful (Rosenbaum et al., 1990). A recent clinimetric review (Auld, Ware, Boyd, Moseley, & Johnston, 2012b) recommended the use of the full 20-item Semmes Weinstein

Monofilament kit (Weinstein, 1993) to evaluate tactile registration and the Disk-Criminator (MacKinnon & Dellon, 1985) to evaluate domains of spatial perception in children with CP. Measures of double simultaneous testing (Burns, Ensby, & Norrie, 1989) and stereognosis (Klingels et al., 2010) have good discriminative properties among children with CP, but may lack responsiveness to change over time (Auld et al., 2012b). Furthermore, there is limited evidence of clinical utility of somatosensory assessments for paediatric use. Clinical utility is particularly important when assessing vulnerable populations such as children with CP so not to burden them with lengthy or complicated assessment procedures (Australian Government, 2007). However, clinical utility is a multifaceted construct incorporating clinical acceptability to both patients and therapists as well as pragmatic consideration of ease of use and interpretation, and the financial and temporal demands entailed (Smart, 2006).

Clinical acceptability is a fundamental aspect that is missing from several frameworks of clinical utility (Bourke-Taylor, 2003; Law, 1987; Rosenbaum et al., 1990). Clinical acceptability requires investigation of the barriers and facilitators perceived by therapists and clients when introducing novel outcome measures to clinical practice. A comprehensive framework of clinical utility is proposed that includes clinical acceptability as a fundamental construct. The framework is based on existing literature and includes examination of (1) clarity of instructions and validated purpose; (2) clarity about constructs to be measured and the intended population; (3) administration format; (4) time to complete administration, scoring and interpretation; (5) equipment cost and therapist accreditation and (6) clinical acceptability to clients (Bourke-Taylor, 2003; Law, 1987; Rosenbaum et al., 1990; Smart, 2006). Clinical utility is not classified as a psychometric property of an assessment tool therefore it stands outside of current taxonomies of measurement properties such as the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010). Evaluation of clinical utility occurs after the clinical feasibility of an intervention or validity of an assessment has been established. The framework proposed in this paper can be used to review validated assessment tools within the clinical setting to determine the tool's suitability for a specific environment.

Occupational therapists desire appropriate pediatric outcome measures that are easy to use with administration procedures acceptable to their clinical population (Law, Hurley, Hurley, King, & Hanna, 2003). If paediatric evaluative measures are not acceptable to children or youth they have limited utility in clinical practice. In the current paper we report on one aspect of the clinical utility of the *sense\_assess© kids* (Taylor et al., 2017f) namely, the clinical acceptability to the client, using self-report from children and youth with CP. Adopting a structured approach to tool development has several advantages when assessment tools are in an early phase of development (Robey & Schultz, 1998). Advancing novel assessment tools from one phase of development to the next provides clear understanding of (a) the level of existing psychometric evidence, and (b) the psychometric testing required for moving forward in the process (Robey & Schultz, 1998). A phased clinical-outcome research model can be adapted from the more familiar interventional model and applied to outcome measure development (Whyte, Gordon, & Gonzalez Rothi, 2009). The model includes Stage 1: Research hypotheses formulation and initial tool selection or development; Stage 2: Early efficacy (formulating and standardising assessment protocols and testing in optimal environments); Stage 3: Late efficacy (hypotheses related to psychometrics and assessment usefulness are tested with appropriate sample sizes and clinical populations in sub-optimal environments); Stage 4: Effectiveness (new assessments are introduced into routine clinical practice) and Stage 5: Post-marketing effectiveness (clinical utility is evaluated including cost, positive changes to clinical care pathways and consumer acceptability).

This research was undertaken as Stage 3 Late efficacy; testing the clinical acceptability of a newly adapted assessment tool. Evaluating clinical acceptability appears in this mid stage of hypothesis testing enabling feedback, and modifications that can be integrated prior to evaluating post-marketing effectiveness and clinical utility in Stage 5. Research relating to Stage 1 and Stage 2 for the *sense\_assess© kids* is described in Chapters 2 to 6. The objective was to determine if the *sense\_assess© kids* (Taylor et al., 2017f) was acceptable for use with children and youth with CP aged 6 to 15 years. We predicted that participants would respond neutrally or positively to all aspects of the assessment procedure.

## **The sense\_assess© kids: Test description and association with somatosensory domains**

The sense\_assess© kids (Taylor et al., 2017f) is an evidence based assessment designed to measure the functional somatosensory capacity of the upper limb of children with CP. The sense\_assess© kids test battery is comprised of four somatosensory tests that have been separately validated in children and adults, primarily by the authors and their colleagues. The battery includes a measure of tactile registration, the Protective Touch Test (Figure 15) (Taylor et al., 2017f; Touch-Test® Sensory Evaluators, 2001), tactile discrimination, the Tactile Discrimination Test (Figure 16) (Carey, Oke, & Matyas, 1997), wrist position sense, the Wrist Position Sense Test (Figure 17) (Carey, Oke, & Matyas, 1996), and haptic object recognition, the functional Tactile Object Recognition Test (Figure 18) (Carey, Nankervis, LeBlanc, & Harvey, 2006).

Figure 15 Protective Touch Test

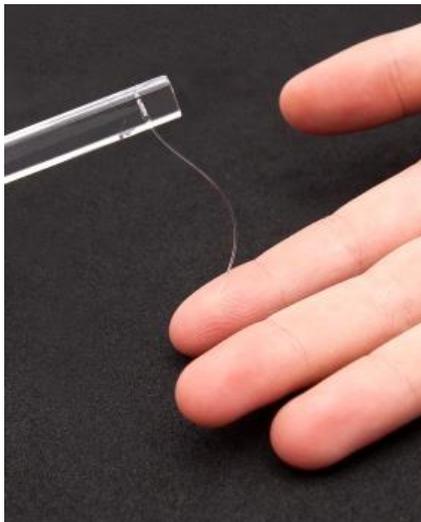
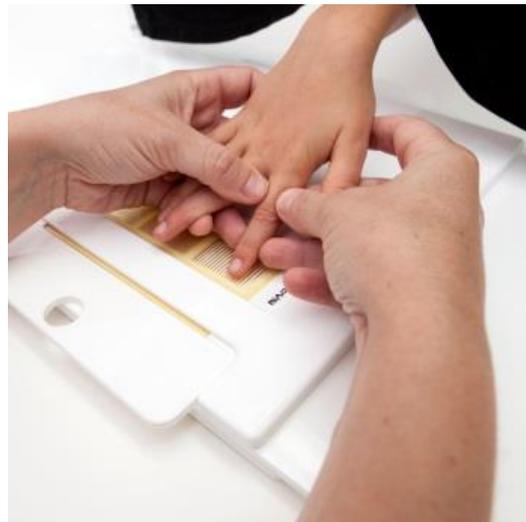


Figure 16 Tactile Discrimination Test



**Protective Touch Test (PTT)** (Taylor et al., 2017f; Touch-Test® Sensory Evaluators, 2001). Tactile registration is the basic initial processing of stimuli and/or sensing of surfaces (Auld, Boyd, Moseley, & Johnston, 2011). The first sense\_assess© kids subtest measures tactile registration, close to the threshold of protective touch, across regions of the dorsal and palmar aspects of the hands and uses the 4.56 monofilament of the Touch-Test® Sensory Evaluator kit (Taylor et al., 2017f; Touch-Test® Sensory Evaluators, 2001). The PTT has high interobserver agreement of 99%-100%

with typically developing (TD) children and youth aged 6 to 15 years (Taylor et al., 2017c).

***Tactile Discrimination Test (TDT)*** (Carey et al., 1997). Tactile discrimination is the ability to distinguish between different surface textures using information received through the sense of touch (Carey, Matyas, & Oke, 2002; Dunn et al., 2013). The TDT (Carey et al., 1997) measures the ability of the index finger to discriminate differences in finely graded textured surfaces using a three-alternative forced choice design. The TDT has high retest reliability and normative standards for adults and normative standards for children aged 6 to 15 years (Carey et al., 1997; Taylor et al., 2017c).

Figure 17 Wrist Position Sense Test



Figure 17. The examiner is positioned directly in front of the participant with a small curtain preventing the participant from seeing where their hand is positioned on the lower protractor and a large curtain separating the examiner and participant.

***Wrist Position Sense Test (WPST)*** (Carey et al., 1996). Wrist position sense is a component of proprioception that aids in detection of the position of the upper limbs in space (Bremner et al., 2013). The WPST (Carey et al., 1996) is a measure of proprioceptively-guided limb position sense and quantifies the ability to identify wrist angle in the flexion-extension range following therapist imposed movements. The box-like apparatus uses a protractor scale to measure degrees of error (difference between actual test position and participant response). The test form contains 20 predetermined test angles, between 25° and 152°, presented in pseudo random order. The WPST has normative standards for adults (~40 to 85 years) and children (6 to 15

years) and has high retest reliability for both populations (Carey et al., 1996; 2002; Taylor et al., 2017c). The test has good discriminative validity and has demonstrated construct validity and responsiveness for children with CP aged 6 to 15 years (Taylor et al., 2017a).

Figure 18 functional Tactile Object Recognition Test



Figure 18. Administration of the functional Tactile Object Recognition Test. The examiner is positioned to the right of the participant with testing completed on the affected side.

***functional Tactile Object Recognition Test (fTORT)*** (Carey et al., 2006). Haptic object recognition is a higher order cognitive process and involves a complex integration of touch, proprioception and fine motor skills. Haptic object recognition is the ability to use touch to identify the nature of the material (e.g. texture or temperature) or geometric properties of objects (Lederman & Klatzky, 1987; 2009). The fTORT (Carey et al., 2006) is designed to test haptic recognition of objects with vision occluded. Sets of common everyday objects are displayed on a numbered response poster and identical test objects are presented for manipulation in a standardised manner. The participant is asked to wear earmuffs so no accidental auditory clues are given during testing. The fTORT has age-adjusted normative standards, high reliability ( $r = .85$  to  $.92$ ) and good discriminative properties for adults aged 21 to 79 years (Carey et al., 2011; Nankervis, 2004). The test has also demonstrated construct validity for children with CP aged 6 to 15 years (Taylor et al., 2017b) and has normative standards for children aged 6 to 15 years (Taylor et al.,

2017c). Practice trials are conducted prior to each subtest to ensure comprehension of instructions.

The *sense\_assess© kids* satisfies the following components of clinical utility; (1) an administration manual that includes (i) a clear purpose and overview, (ii) standardised administration instructions and standardised script; (iii) descriptions of somatosensory constructs to be measured based on current literature, (iv) descriptions of the diagnostic groups for whom it is designed, (v) references to the empirical evidence of construct validity and intrarater reliability, and (vi) description of the format of administration (requires active participation of children verbally and physically); (2) for accredited and experienced assessors time to complete administration is approximately 45 minutes and an additional 20 minutes for scoring. Interpretation is simple based on pre-defined criteria from a normative sample; (3) current cost of the core *sense\_assess© kids* assessment kit is approximately \$3,000AUD, assessor accreditation and training is a two day course currently costing \$450AUD.

## **Methods**

### ***Study design***

The study design was cross-sectional. A questionnaire, with open ended questions, was provided to participants following exposure to the *sense\_assess© kids* to identify children and youths' perspectives of the assessment. Descriptive and thematic analysis of responses from a questionnaire is a useful means of exploring individual perspectives when little is known about the possible outcomes in advance (Greco, Lambert, & Park, 2016). As part of a larger study examining the psychometric properties of the *sense\_assess© kids* (Taylor et al., 2017a; 2017b) and feasibility of a novel intervention, *sense©* training for children with CP (McLean et al., 2017a), participants were administered a brief questionnaire immediately following completion of the *sense\_assess© kids*. While the larger study obtained data using various outcome measures to validate the *sense\_assess© kids*, the questionnaire was used for descriptive purposes and was not used as an outcome measure for the larger studies.

Recruitment occurred via the larger study *sense©* training study and the eligibility criteria were the same; Australian children with a paediatrician-confirmed

description of spastic hemiplegic CP, aged 6 to 15 years. Exclusion criteria included surgery of the affected upper limb in the previous 12 months or inability to understand or respond to simple instructions such as, *'In this activity you will be asked to tell me the everyday objects I put in your hand to feel, without looking'*. All participants eligible for the larger *sense*© training study were invited to complete the questionnaire. The study protocol was approved by the Human Research Ethics Committee of Princess Margaret Hospital for Children (#2052) and Curtin University (#87), Perth, Australia and all procedures were in accordance with the revised Helsinki Declaration (2000). All participants were informed of the study's risks and benefits, that their participation was voluntary and that their identity would remain confidential. Signed informed consent and/or assent were obtained for all participants.

#### ***Data collection instrument***

The six-item questionnaire was designed to be a client-centred interview and contained two semantic differential scales, a photographic Q-Sort, and three questions requiring short answers (Figure 19). The main outcome variables were emotional responses to the administration process (e.g., scale of happy to sad), perceived level of difficulty when completing assessment items, and overall opinion of the assessment procedure and equipment. Item selection was based on relevance to acceptability and current literature regarding qualitative measurement and self-reporting for children (Berdeaux, Hervié, Smajda, & Marquis, 1998; Portney & Watkins, 2009).

Figure 19 Data collection instrument

Participant code \_\_\_\_\_ D.O.B \_\_\_\_\_ Date of assessment \_\_\_\_\_

**Question 1.** How did you feel when doing this activity? Please mark on here with a pencil.

Very happy      Happy      Just ok      Unhappy      Very unhappy

**Question 2.** Please look at these picture cards of the activity that you just did. Sort them into two piles. Put the ones you liked on the left, put the ones you disliked on the right.

Record participant's response here with an L or a D

PTT \_\_\_\_\_ TDT \_\_\_\_\_ ftORT \_\_\_\_\_ WPST \_\_\_\_\_

From the 'Like' and 'Dislike' piles ask the participant the following questions for each card:

**Question 3.** What did you like about:

PTT \_\_\_\_\_  
TDT \_\_\_\_\_  
ftORT \_\_\_\_\_  
WPST \_\_\_\_\_

**Question 4.** What didn't you like about:

PTT \_\_\_\_\_  
TDT \_\_\_\_\_  
ftORT \_\_\_\_\_  
WPST \_\_\_\_\_

**Question 5.** What did you think about the time it took to do?  
i.e. Too long  Too short

Other response \_\_\_\_\_

**Question 6.** How difficult did you find the activity? Please mark on here with a pencil.

\_\_\_\_\_/\_\_\_\_\_/\_\_\_\_\_/\_\_\_\_\_/

Very difficult      Difficult      Easy      Too easy

**Thank-you**

Rationale and details of the questionnaire follow. *Questions 1 and 6.* A Semantic differential scale is based on a continuum that extends between two extreme opposites (Portney & Watkins, 2009). Question 1 aimed to measure participants' feelings about the administration procedure and question 6 was used to assess participants' perception of difficulty. Similar self-report scales have been used with children to assess perceptions of pain (Garra et al., 2010).

*Question 2.* Q-sort methodologies have been used as an effective and interactive way to characterise the attitudes, opinions or judgements of individuals with CP aged 5 to 12 years (Calley et al., 2012). For question 2 participants in the current study were asked to sort a photograph of each sense\_assess© kids subtests into a pile of 'like' or 'dislike'. The occupational therapist specified that the photographs needed to go in one of the two piles so that the participant was forced to make a choice (Portney & Watkins, 2009).

*Questions 3 to 5.* Open-ended questions requiring short answers are useful when the researcher is not sure of all possible responses to a question (Portney & Watkins, 2009). Questions 3, 4 and 5 aimed to record responses in the participants' own words about what aspects they liked and didn't like about each assessment subtest.

As no tool specific to assessing children's responses to assessment procedures could be found the questionnaire was designed for the current study so little is known about its reliability and validity or its use for children with CP. However, face validity of the questionnaire was considered acceptable by three senior occupational therapists at a tertiary care hospital and the format of the questionnaire allowed children to be active participants. Each age group was able to respond without assistance from parents.

### ***Data collection procedures***

Questionnaires were administered by an occupational therapist (first author) immediately after administration of the sense\_assess© kids. All participants sat and responded to the questionnaire, however not all questionnaire items were completed by all participants. Administrator error, children's motivation to complete the questionnaire and difficulties keeping young children on task impacted data

collection. Field notes often noted poor concentration and data were missing more frequently for the youngest children (6 to 7 years).

To address potential observation bias the questionnaire was designed with standardised instructions thus limiting the effect of personal interview technique. Interviewer bias was controlled for by utilising participant self-report in the questionnaire design. The occupational therapist recorded responses immediately and in the participant's own words. To control for recall bias the questionnaire was administered immediately following administration of the *sense\_assess© kids*.

### ***Data analysis***

A summative content analysis was used to count and compare keywords and sentences derived directly from the short answer text data. This process was performed by the occupational therapist who administered the questionnaires and the process repeated by an independent coder who was not involved in the data collection, as recommended by Neuendorf (2002). Participants' responses were brief and concise which limited the risk of misinterpretation e.g. 'Tickly, liked feeling on finger' or 'good challenge'. Statements such as these were reduced into concept categories. Consensus was reached for minor discrepancies via discussion between the two coders. Descriptive statistics were generated in Microsoft Excel 2010 to calculate responses to the semantic differential scales and photographic Q-Sort.

### **Results**

Twenty-eight children were potentially eligible and invited to participate in the acceptability study. Two children were unable to complete the questionnaire after the *sense\_assess© kids* administration due to poor concentration. Twenty-six children with spastic hemiplegic CP were confirmed eligible and included for analysis (age range 6y - 15y 6m; mean age 10y 8m; 16 boys, 10 girls; Manual Ability Classification System (MACS) (Eliasson et al., 2006) level I, n = 10; level II, n = 16; right hemiplegia n = 14; left hemiplegia n = 12). All participants attended main stream school and had at least one family member that was employed full time. The majority of participants, 21 of 26, indicated they were 'Very happy' or 'Happy' with the administration process, the remaining 5 indicated they felt 'Just ok'. Not all participants responded to all questions concerning specific tests: 17/25 liked the PTT,

17/24 liked the TDT, 23/25 liked the fTORT and 18/25 liked the WPST. None of the 20 participants found it 'Very difficult' to complete; 7 found it 'Difficult', 10 found it 'Easy' and 3 found it 'Very easy' though this was not reflected in their test scores. Of 20 responders regarding duration of the test: 7 found it 'Too long', 12 'Just right' and 1 found it 'Too short'. Content analysis revealed that the majority of participants liked 'guessing', i.e., trying to identify the sensory stimuli without vision and reported that the subtests were a good challenge. The majority of participants also liked the sensation felt at the hand when tested with different subtest equipment.

The most common short answers from the children who liked the PTT ( $n = 17$ ) were that they liked the sensation of the monofilament because it felt 'unusual' and 'tickled', that it was challenging and they reported enjoyment when identifying the presence or absence of stimuli. Children who reported not liking the PTT ( $n = 8$ ) said the monofilament 'hurt' or 'tickled' their finger and some didn't like the sensation of 'spikiness'. For the children who liked the TDT ( $n = 17$ ), the most common short answers revealed that they liked feeling the textures and enjoyed choosing which texture was different, and that it was challenging. The children who did not like the TDT ( $n = 7$ ) said they disliked how 'hard' it was and they disliked feeling the textures as they 'hurt' or were 'uncomfortable'. Children who liked the fTORT ( $n = 23$ ) said it was fun feeling the objects, and recognising the objects by touch, as it was challenging and they enjoyed wearing the ear muffs. There was only one common negative short answer for the children who disliked the fTORT ( $n = 3$ ); they reported it was hard to recognise objects in the hand. For the children who liked the WPST ( $n = 18$ ), they liked their hand being moved by the occupational therapist, they liked identifying their hand's position and that it was challenging, they liked the splint as it was comfortable and they liked using the pointer. The children who did not like the WPST ( $n = 7$ ) reported that the wrist and forearm splint 'hurt', they disliked how 'hard' it was and disliked having their wrist being moved by the occupational therapist as it produced confusion.

## **Discussion**

This study has established that the administration process and duration, subtest equipment and perceived level of difficulty of the sense\_assess© kids were generally acceptable to children and youth with spastic hemiplegic CP aged 6 to 15 years.

Obtaining the perspective of children and youth about the sense\_assess© *kids* has clearly demonstrated its clinical acceptability prior to further psychometric testing and knowledge translation. Across the different subtests, the majority (60%-92% depending on subtest) of participants responded positively to all aspects of the assessment procedure. The importance of an acceptable paediatric assessment is reflected in literature; functional, engaging and easy to use measures enhance the assessment experience and provide reliable data (Linder & Linus, 2009).

The current findings are consistent with other tool development studies that also found assessment procedures that are enjoyable, fun and simple to administer are more acceptable for clinical use (Jongbloed-Pereboom, Nijhuis-van Der Sanden, & Steenbergen, 2013; Krumlinde-Sundholm & Eliasson, 2003). Although participants generally responded positively, the areas where responses were not positive require further development. It is noteworthy that the two subtests (fTORT; WPST) requiring active participation of children and an opportunity for choice (selecting object from response poster; moving pointer on protractor) rated more favourably than the subtests where the therapist was in control of all stimulus administration (pressing monofilament on skin, moving child's finger across textures). Assessments involving autonomous choice, game-play or active participation of the child, where children are meaningfully involved, encourage self-motivation to complete tasks and may be more readily accepted into clinical practice (O'Grady & Dusing, 2015; Poulsen, Ziviani, & Cuskelly, 2013).

Negative responses relating to the TDT such as 'uncomfortable' might be because the therapist is required to move the index finger across the plastic graded textures multiple times using a standard pressure and speed. It is recommended that the amount of exposures is reduced and additional breaks are incorporated into the testing procedure. Standardised exposure to the textures supports the tool's psychometric properties however being able to adjust finger pressure and number of repetitions is surely part of successfully interpreting sensation for an individual. In future, it may be worth assessing whether children reliably identify the correct answer more often if they are allowed to choose their own finger pressure and number of repetitions.

A small number of children ( $n = 4$ ) reported that the WPST test apparatus was 'uncomfortable' or that it 'hurt'. This is a critical finding in this early phase of

tool adaptation. This issue may be resolved with a simple adjustment to the size of the wrist and forearm splints. It is recommended that interchangeable small, medium and large sized splints are included in test equipment. A further essential recommendation is to ask the child's level of comfort after the practice trial of the WPST and checking throughout the administration procedure. If a child's position on the chair, or alignment to the equipment has changed, the administration should be paused and the child's postural positioning should be corrected as per the administration manual. The examiner will also need to reset the child's hand position to 90 degrees once the child is repositioned to avoid complete loss of limb position awareness. Although some children perceived the TDT, fTORT and WPST as 'hard', this challenge is important from a psychometric testing perspective and avoids the risk of this population achieving maximum scores on the subtests, i.e., a ceiling effect (Auld et al., 2012b). With that in mind, extending the range of difficulty to also include 'easy' stimuli, to provide children with an opportunity to identify at least one difference correctly would not only make the children happier, but also provide greater precision at the lower end of achievement, i.e. avoiding a floor effect.

### **Limitations**

The brief clinical acceptability questionnaire was designed for the purposes of the current study only. Although authors tested face validity, and based it on current literature, the questionnaire lacked a formal evaluation of its measurement properties as is recommended (Ramaswami et al., 2012). Findings from content analysis within this study may be limited in that participants were familiar with the occupational therapist conducting the questionnaire which could have biased results. In the current study authors were not able to measure sensory integration or behaviour or emotional responses to somatosensory stimuli that may be triggered by imposed touch. It is recommended that this be considered in future research considering that children with CP may have comorbid medical conditions.

### **Future research**

Self-report by a child has been described as a simple and reliable measure of emotional responses in clinical practice (Voepel-Lewis et al., 2008). The translation of research into clinical practice is made easier by simple to use, time efficient and

cost-effective instruments with supporting psychometric data reported in a meaningful way (Radia-George, Imms, & Taylor, 2014). A perceived lack of clinical utility of outcome measures can inhibit their use by clinicians and department managers and this often persists because of the resulting lack of evidence to refute this perception (Radia-George et al., 2014).

The current study was conducted under research conditions and indicated the acceptability to participants of the *sense\_assess© kids* in clinical practice. The next phase of tool development (Stage 4) is recommended to involve evaluation of clinical acceptability to therapists (clarity of instructions, purpose, format, administration time, cost and overall acceptability). Outcomes could be used to identify challenges to adoption of the *sense\_assess© kids* into clinical practice (Voepel-Lewis et al., 2008). Current perspectives regarding knowledge translation recognise the need for active collaboration between researchers and therapists as end users, and researchers (Metzler & Metz, 2010). This approach of consumer involvement ensures thorough examination of the implications for therapists and clients prior to transfer of new research knowledge.

## **Conclusion**

The current study has demonstrated preliminary clinical acceptability of the *sense\_assess© kids* for children with hemiplegic CP aged 6 to 15 years. The *sense\_assess© kids* is the only comprehensive standardised assessment of somatosensory function for children with CP. The current study contributes to the further understanding of this novel assessment tool to measure somatosensory capacity, inform intervention outcome, and improve functional outcomes for children with neurological disorders.

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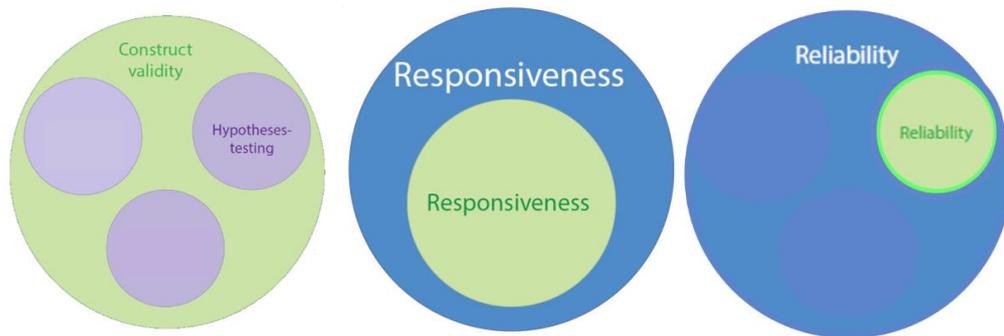
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## CHAPTER 5 Wrist Position Sense Test



Phase three – Psychometric testing using the COSMIN

### 5.1 Introduction to chapter

This chapter corresponds to Phase three and early development of psychometric measurement properties using the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010). The previous chapter established that the *sense\_assess© kids* is a clinically acceptable tool for use with children with cerebral palsy (CP) aged 6 to 15years. The next step was to determine the rigour of measurement properties for the *sense\_assess© kids*. Studies evaluating the psychometric properties of outcome measures should be of high methodological quality to ensure that appropriate inferences can be made about the assessment tool (Mokkink et al., 2010).

The measurement property of reliability is the degree to which an assessment tool produces stable and consistent results across different time-points (Portney & Watkins, 2009). Validity is the degree to which an assessment measures the construct it was designed to measure (Portney & Watkins, 2009) and responsiveness is the ability to detect change in the construct over time (Mokkink et al., 2010). Testing the validity and responsiveness of the Wrist Position Sense Test (WPST) was chosen as the first step before reliability testing in a clinical population to ensure that the WPST was measuring the intended construct. Although it is important for an assessment tool to demonstrate both validity and reliability, validity testing precedes reliability because a reliable test may not necessarily be measuring a valid construct. In the following paper the COSMIN (Mokkink et al., 2010) was used

as a framework to guide evaluation of the psychometric properties of the WPST, including construct validity and responsiveness via hypothesis testing and reliability using a test re-test design.

## **5.2 PAPER TWO: Wrist Position Sense Test**

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The PhD Candidate, Susan Taylor accounted for 85 per cent of the intellectual property associated with the final manuscript. Collectively, the remaining authors contributed 15 per cent.

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### **Construct validity, reliability and responsiveness of the Wrist Position Sense Test for use in children with cerebral palsy.**

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## **Abstract**

**Introduction:** The development and validation of assessment tools for children with neurological conditions is important to evaluate changes in outcomes following intervention. This study investigated the construct validity, test re-test reliability and responsiveness of the Wrist Position Sense Test (WPST) for children with cerebral palsy (CP).

**Methods:** A cross-sectional study of 28 children with spastic hemiplegic CP (mean age 10y 8m; SD 2y 4m; 16 male) and 39 typically developing (TD) children (mean age 11y; SD 2.9y; 19 male) was employed to investigate construct validity and association with an upper limb activity measure the Box and Block Test (BBT). Twenty-two TD children were tested at a second time-point to examine test re-test reliability. Seventeen children with CP were randomly allocated to either a treatment (n = 10) or control (n = 7) group and assessed at four time-points to determine test responsiveness.

**Results:** There was a significantly greater difference in mean error of indicated wrist position ( $p < .01$ ) in children with CP at baseline ( $M = 21.6^\circ$ ,  $SD = 21.6^\circ$ ) than in TD children ( $M = 12.8^\circ$ ,  $SD = 11.0^\circ$ ). Larger WPST errors were associated with poorer performance on the BBT ( $p < .01$ ) indicating a substantial association, and there was no consistent difference between time-points indicating good test re-test reliability. The WPST also demonstrated responsiveness to intervention with a statistically significant reduction in mean error following treatment ( $p < .001$ ) that was not seen in the control group ( $p = 0.28$ ). This WPST will be important to understand deficits, plan intervention and measure outcome effectiveness.

**Conclusion:** The WPST demonstrated construct validity, good test re-test reliability and responsiveness to intervention, and was highly correlated with measures of activity and impairment. The WPST has potential to diagnose deficits, plan intervention and measure outcome effectiveness in children with CP. This study contributes to the establishment of psychometric properties for a novel clinical assessment tool and advances clinical measurement of wrist position sense for children with neurological conditions such as CP.



## **Introduction**

Upper limb proprioception informs perceptual judgements of the physical position of the arms and hands in space and contributes to how efficiently the hands can execute daily motor tasks (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Dijkerman & de Haan, 2007; Krumlinde-Sundholm & Eliasson, 2002). Typical development of proprioceptive sensibility can be altered as a result of primary neonatal or antenatal brain injury (Rosenbaum, 2003). Cerebral palsy (CP) is a term used to describe a group of non-progressive, but often changing, disorders of the motor system that includes disturbances of sensation; such as proprioceptive deficits in 46% to 82% of individuals with CP (Bax et al., 2005; Cooper et al., 1995; McLean, Taylor, Valentine, Carey, & Elliott, 2017a; Van Heest, House, & Putnam, 1993). Motor impairment in CP may be caused by disruption to somatosensory pathways including those responsible for processing proprioceptive information (Hoon Jr et al., 2009). Accurate assessment of somatosensory impairment in children with CP would allow further understanding of this deficit, development of targeted interventions and also measurement of outcomes (Russo, 2011).

Currently there are limited valid and reliable assessments of proprioceptive ability for use with children with CP (Connell & Tyson, 2012; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009). Methods of measuring upper limb proprioception in research often employ electronic devices requiring a computer interface or camera, bulky custom built devices limited in portability, or experimental set-ups that require motor control of the affected limb (Dukelow et al., 2010; Wingert et al., 2009). Many lack standardised procedures, evidence of clinical utility or reported psychometric characteristics (Fuentes, Mostofsky, & Bastian, 2011; Klingels et al., 2010; Little et al., 2015; Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2012). Traditionally, clinical measures of upper limb proprioception have required replication or between-limb matching of therapist imposed movements of the affected limb, verbal response to the passive direction of movement, or goniometers and active and passive joint angle scaling procedures (Carey, 1993; Klingels et al., 2010; Bentzel, 2008). These measures are often subjective in nature with little or no demonstrated reliability or sensitivity to detect changes over time (Carey, Oke, & Matyas, 1996).

The Wrist Position Sense Test (WPST) (Carey et al., 1996) was designed as a clinical tool to measure limb position sense at the wrist in the flexion/extension range, in adults following a stroke. The outcome of interest is included in the International Classification of Functioning, Disability and Health (ICF) definition of body functions (B2: Additional Sensory Functions) of which proprioceptive function (B260) is one aspect (WHO, 2001). Because the WPST is a standardised and norm-referenced assessment, it differs from commonly used subjective measures. Due to the lack of measures of limb position sense for children with CP and the success of the test with adult stroke survivors (Carey et al., 1996), and preliminary evidence of the application of a brief version with typically developing (TD) children (Dunn et al., 2013; 2015; Taylor et al., 2017b), the WPST was adapted to measure the same construct for children with CP aged 6 to 15 years. A recent evaluation of one aspect of clinical utility demonstrated the adapted tool's clinical acceptability to a sample of children with CP aged 6 to 15 years (Taylor et al., 2017d).

There is emerging evidence to suggest that impairments in proprioception including limb position sense, impact an individual's ability to perform motor tasks (Cooper et al., 1995; Krumlinde-Sundholm & Eliasson, 2002). However, it is not well known if measures of proprioceptive function are associated with measures of upper limb activity such as grasp and release. James et al. (2015) recently reported that current impairment measures do not consistently reflect gains in functional outcomes such as bimanual hand use or manual dexterity for children with CP affecting the upper extremity. They also concluded that activity measures correlated with each other more consistently than impairment measures, and are likely to provide more appropriate indicators of upper extremity function after surgery (James et al., 2015).

The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010) describes the measurement properties that constitute a quality assessment tool in health care and provides guidelines for measuring the psychometric properties of an assessment tool. The study design and subsequent reporting within the current paper were based on the COSMIN with the aim of producing a clear, robust and reproducible research methodology. The psychometric properties evaluated in this paper adhere to the

COSMIN taxonomy including validity, reliability and responsiveness (Mokkink et al., 2010). The COSMIN's definition of measurement properties describes one aspect of construct validity as the degree to which the scores of the instrument are consistent with pre-determined hypotheses e.g. relationships to scores of other instruments, or differences between relevant groups. Because a standardised criterion measure of wrist position sense was not available at the time of data collection, we therefore examined the ability of the tool to discriminate differences in wrist position sense in children with and without expected impairment, and the relationship between the WPST and a measure of upper limb activity function as an outcome of interest in children with CP. Authors chose the Box and Block Test of Manual Dexterity (BBT) (Mathiowetz, Federman, & Wiemer, 1985a) as a test of association because it is a brief, validated and standardised upper limb activity measure for use with children with CP and could be used to compare scores with the WPST.

Specifically, the current study aimed to investigate the construct validity of the WPST by looking at the differences between typically developing (TD) children and children with CP at one time-point. We hypothesised that the WPST would reflect known differences in wrist position sense performance between groups. We also examined the association between scores on the WPST and the BBT (Mathiowetz et al., 1985a) hypothesising that there would be a positive association between the measures. Test re-test intrarater reliability of the WPST was examined using Bland Altman plots to investigate differences in performance between two time-points (Bland & Altman, 2010). Finally, for responsiveness testing it was hypothesised that the WPST would detect significant differences between children receiving standard care and those additionally exposed to a novel somatosensory discrimination intervention (McLean, Taylor, Valentine, Carey, & Elliott, 2017a).

## **Methods**

### ***Study design***

Four different studies involving psychometric testing were conducted as part of this paper and involved the WPST and BBT as the primary outcome measures. Children with CP and TD children were tested with the WPST, with TD children being tested twice as part of the test re-test reliability study. Children with CP were also tested multiple times, pre and post an intervention, with the WPST and the BBT. The

assessor was an occupational therapist who was trained in the use of the assessments. The assessor could not be blinded to groups (TD or CP) because of the clinical presentation of children with CP. For the responsiveness study, the assessor was blinded to allocation of the participants with CP. The study was conducted in Perth, Western Australia and the data were collected in participant's homes in metropolitan and semi-rural areas of Perth. Recruitment for the TD participants occurred via professional and personal contacts and data collection for both TD and CP groups occurred between March 2013 and December 2014. The study protocol was approved by the Human Research Ethics Committee of Curtin University (#87) and Princess Margaret Hospital for Children (#2052), Perth, Australia.

***Construct validity study: Comparison of WPST between CP and TD children***

A cross-sectional study was employed to investigate construct validity. WPST scores obtained from TD children (n = 39) were compared with those of children with hemiplegic CP (n = 28) who are at risk of a proprioceptive deficit. Scores from the WPST from both groups were taken at one time-point, which was the baseline for the CP group prior to an intervention.

***Association study: Comparison between WPST and BBT in children with CP***

For the group with CP, scores on the WPST were also compared with scores from the BBT to determine presence of an associative relationship. Scores were taken at one time-point (baseline) prior to an intervention.

***Reliability study: Test-retest reliability in TD children***

For reliability testing a convenience sample from the total TD population included in the construct validity study were assessed with the WPST at two time-points by the same rater approximately three-weeks apart.

***Responsiveness study: Ability to detect change in WPST pre-post somatosensory intervention***

A recent random allocation intervention trial investigated the efficacy of a novel somatosensory intervention based on perceptual-learning to improve somatosensory discrimination in children with CP (McLean, Taylor, Valentine, Carey, & Elliott, 2017b; Carey, Macdonell & Matyas, 2011). Australian New Zealand Clinical Trials Registry (ACTRN12614000314628). Children with hemiplegic CP were randomly

allocated to a treatment (n = 7) or control group (n = 10). The treatment group received somatosensory discrimination training three times a week for six weeks in addition to standard care while the control group received standard care only. The WPST (Carey et al., 1996) and the BBT (Mathiowetz et al., 1985a) were among the primary outcome measures administered at four time-points in the intervention trial.

### ***Participants***

Inclusion criteria for the construct validity and association study were children with spastic hemiplegic CP with a diagnosis confirmed by a paediatrician, aged 6 to 15 years. Exclusion criteria included surgery on the affected upper limb in the previous 12 months or inability to understand or respond to simple instructions. Children without CP who participated in the construct validity and reliability study were a convenience sample of individuals aged 6 to 15 years. Eligibility criteria for the responsiveness study were the same as for the construct validity and association study plus presence of a proprioceptive impairment as measured by the WPST with relation to normative standards of mean error provided in this paper.

### **Outcome measures**

#### ***The Wrist Position Sense Test***

The WPST administration manual and equipment was adapted from a version designed for adults (Carey et al., 1996). Adaptation of the standardised test instructions and testing procedure to a paediatric version considered the semantics and perceptual concepts appropriate to paediatric development (Berlin, Blank, & Rose, 1980; Bloom, 2001). Pilot testing of the adapted assessment was undertaken with five TD school-aged children (6 to 11 years old), a parent and a senior occupational therapist. The WPST has age-adjusted normative standards for adults in the age range 21 to 79 years, high reliability ( $r = .85$  to  $.92$ ) and good discriminative test properties (Carey et al., 2011; Carey et al., 1996). The paediatric WPST of the current study has demonstrated clinical acceptability for use with children with CP aged 6 to 15 years (Taylor et al., 2017d). A brief paediatric version of the WPST, validated as part of an earlier study by the authors demonstrated good agreement between two time-points using Bland Altman plots to investigate intrarater reliability (Bland & Altman, 2010; Taylor et al., 2017c).

*Apparatus.* The box-like apparatus (Figure 20) used two 180° protractor scales with markings at 1° intervals to measure degrees of wrist position sense error as a continuous measurement scale. Participants sat in a chair facing the examiner with the apparatus on a table between them. The participant's hand and wrist were occluded by the box during assessment. The forearm and hand were secured in separate thermoplastic splints on the lower protractor, with the wrist supported in the neutral position and elbow flexed at 90°. The hand splint was attached to a horizontal lever in a position that ensured repeatable and standardised movement of the wrist in the flexion-extension range. The axis of movement of the response pointer exactly matched that of the test lever (Carey et al., 1996).

Figure 20 The Wrist Position Sense Test apparatus



*Figure 20.* Note. Photograph of the set up of the paediatric WPST test procedure. The adult test procedure was originally published in “Impaired limb position sense after stroke: A quantitative test for clinical use,” by L. M. Carey, L. Oke, and T. Matyas, 1996, *Archives of Physical Medicine and Rehabilitation*, 77, pp. 1271-8.

*Task and procedure.* The test form contained 20 predetermined test angles, between 56° to 152° for the left hand and 25° to 125° for the right. The specific angle ranges were chosen to include near full range of flexion-extension movement at the wrist. The ranges differed between hands to provide a comparable and comfortable range of wrist flexion (~65 degrees) and extension (~35 degrees) movement. Movement to test positions was imposed by the therapist in pseudo random order. Participants used

their free hand to move a pointer on the top protractor to indicate their estimate of the position of the occluded hand.

*Scoring and interpretation.* The absolute difference in degrees between the test angle and the participant's responses for the 20 test positions was summed. The WPST score was the average of these. A higher average error score indicated a larger error and therefore poorer performance. Observations of the 39 TD participants within this study generated preliminary normative data for the WPST for children aged 6 to 15 years. Based on these observations, average degrees of error greater than the following values indicate impaired wrist position sense; 16.3° (right hand), 18.6° (left hand) for 6 to 8 year olds; 13° (right hand), 14.4° (left hand) for 9 to 11 year olds; and 13° (right hand), 14.3° (left hand) for 12 to 15 year olds. Administration took approximately 10 minutes per hand depending on the personal factors and characteristics of each child. Scoring took approximately 5 minutes.

### ***Box and Block Test of Manual Dexterity***

The BBT is a measure of unilateral gross manual dexterity (Mathiowetz et al., 1985a). The manual task required in the BBT is in line with the ICF definitions of activity (d440: Fine Hand Use) including picking up (d4400), grasping (d4401) and releasing (d4403) (WHO, 2001). The BBT has high test re-test reliability for TD children ( $r = .84$ ) and adults ( $r = .96$ ); high concurrent validity and high inter-rater reliability ( $r = .99$ ) in adults (Jongbloed-Pereboom, Nijhuis-van Der Sanden, & Steenbergen, 2013; Mathiowetz, Volland, Kashman, & Weber, 1985b); norms for TD children aged 6 to 19 years (Mathiowetz et al., 1985a); and is regularly used in research for children with CP (Golubović & Slavković, 2014; Holmström et al., 2010).

### ***Data analysis***

The response errors for each angle tested as part of the WPST were compared between participant groups using a (random effects) regression model (Portney & Watkins, 2009). This model took into account the association between the repeated tests performed by the same participant. The dependent variable for this analysis was the logarithm of absolute error, as this transformation removed some skewness so that it followed a normal distribution more closely than the raw data. For the children with CP, the relationships between scores on the BBT and the WPST were

examined by including the total score on the BBT as an independent variable in the regression model, and assessing the statistical significance of its association with the WPST.

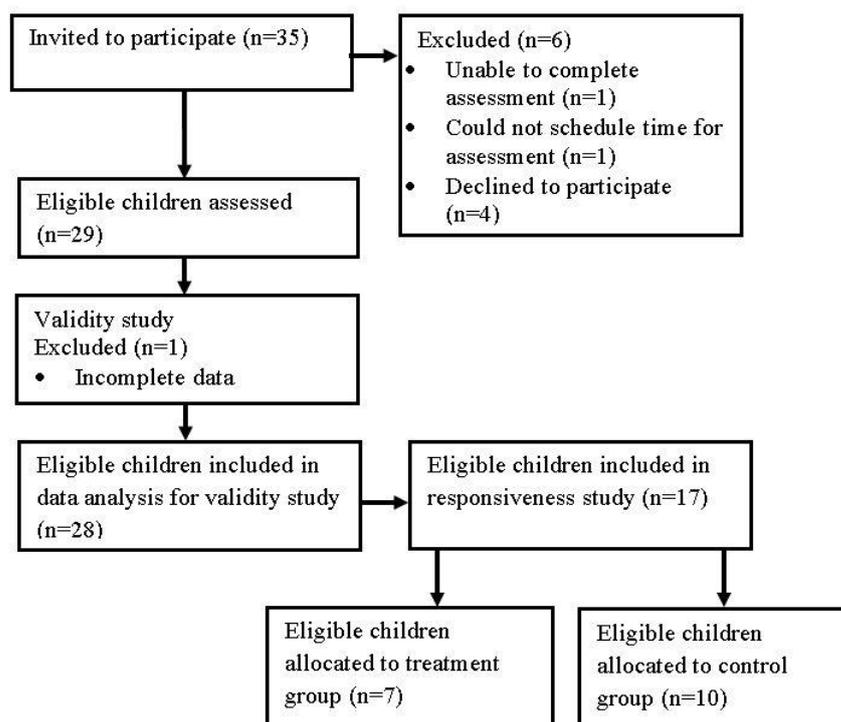
Statistical methods for test re-test reliability testing involved using Bland Altman plots to examine the consistency in scores between two time-points. For responsiveness, the baseline WPST score of each participant was subtracted from the WPST scores at each of three post baseline time-points, and the mean change, standard deviation (SD) and effect size (mean change/group SD) at each time-point calculated. These differences represented the changes from baseline in the WPST error scores. Effect size was considered small if between  $\geq 0.1$  and  $< 0.3$ , medium if between  $\geq 0.3$  and  $< 0.5$  and large if  $\geq 0.5$ . (Cohen, 1992). Analyses were conducted using the SAS version 9.2 software (SAS Institute, Cary, NC, USA 2008), and a p-value  $< 0.05$  was taken to indicate statistically significant associations.

## **Results**

### ***Participants***

For the construct validity and association study, 28 children with CP were confirmed eligible and included in the study (age range 6y - 15y 6m; mean age 10y 8m; SD 2y 4m; 16 boys; right hemiplegia n = 15, Manual Ability Classification System (MACS) (Eliasson et al., 2006) level I n = 10) (Figure 21). Thirty-nine TD children were recruited and included in the analysis for the construct validity study (age range 6y - 15y 5m; mean age 11y; SD 2y 9m; 19 boys; right hand dominance n = 37). The test-retest study involved a convenience sample of 22 of the 39 participants (mean 8y 8m; SD 2y 6m) to investigate intrarater reliability. For the responsiveness study, all 17 participants recruited for the intervention trial were included for analysis. Ten children were randomly allocated to the control group (mean age 10y 2m; SD 2y 9m; 5 boys, left hemiplegia n = 7, MACS level II n = 8) and seven children to the treatment group (mean age 10y 1m; SD 2y 6m; 4 boys, left hemiplegia n = 2, MACS level II n = 6).

Figure 21 Flowchart of dual study recruitment of children with cerebral palsy



### ***Construct validity of the WPST***

There was a significant difference between the TD and the CP groups for the WPST ( $F_{1,1246} = 6.34$ ;  $p = .01$ ) indicating greater accuracy in sensing wrist position for the TD group (Table 4). Throughout administration of the WPST for children with CP there was no significant increase in error over time suggesting that neither fatigue nor attention level affected performance ( $F_{1,491} = 1.04$ ;  $p = 0.31$ ).

Table 4 Wrist Position Sense Test average error scores for children with and without cerebral palsy

Group	Max. score*	Min. score.	Mean	SD	P value
Typically Developing ( $n = 39$ )	37.7°	5.2°	12.8°	11.0°	0.01
Cerebral Palsy ( $n = 28$ )	40.9°	7.1°	21.6°	21.6°	

Note. \*Scores are reported as average error scores across the 20 test positions. SD = standard deviation, Max = maximum, Min = minimum.

### ***Association between the WPST and BBT***

For the children with CP, average error on the WPST was inversely associated with the number of blocks being moved on the BBT; such that every additional block moved was associated with a decrease of 0.45 degrees of error on the WPST, ( $F_{1,491} = 8.53$ ;  $p < .005$ ).

### ***Reliability of the WPST***

The Bland Altman (Bland & Altman, 2010) determined there were no consistent difference between time-points indicating intrarater reliability of the WPST with TD children aged 6 to 15 years (Table 5).

Table 5 Wrist Position Sense Test reliability results for typically developing children

TD sample N=22	WPST Error in degrees
Mean difference RH	-0.87
LOA	4.65 - 2.91
95% CI	-1.73 - -0.01
Mean difference LH	-1.39
LOA	-4.78 - 1.99
95% CI	-2.16 - -0.63

Note. TD = typically developing; WPST = Wrist Position Sense Test; RH = right hand; LH = left hand; LOA = Limits of agreement; CI = confidence interval.

### ***Responsiveness of the WPST***

There were no significant differences between intervention and controls for the WPST ( $p = .31$ ) however, there were significant within group differences over time for the treatment group ( $p < .001$ ) (Table 6). There was a statistically significant decrease in mean error score on the WPST for the treatment group following the intervention with a reduction in mean error from time-point one ( $24.7^\circ$ ) to time-point two ( $16.9^\circ$ ) ( $t_{538} = 3.8$ ;  $p < .001$ ), which was maintained at time-points three ( $19.5^\circ$ ) ( $t_{538} = 3.28$ ;  $p < .01$ ) and four ( $16.8^\circ$ ) ( $t_{538} = 3.19$ ;  $p = .002$ ). The significant improvement in error score from time-point one to two was  $8^\circ$ , while changes between time-points two, three and four (all post intervention) were smaller (all within approximately  $3^\circ$ ). The effect size measuring the impact of the intervention was large between time-point one to time-point two ( $r = 0.632$   $p = 0.018$ ), and this was maintained at time-point four ( $r = 0.632$   $p = 0.018$ ). There were no statistically significant differences across time-points in the control group (Table 6).

Table 6 Wrist Position Sense Test mean error scores for the treatment and control group

Time-point	TP 1 M(SD)	TP 2 M(SD)	TP 3 M(SD)	TP 4 M(SD)	P value
Treatment ( $n = 7$ )	24.7° (20.8°)	16.9° (17.4°)	19.5° (19.8°)	16.8° (14.4°)	0.0005
Control ( $n = 10$ )	27.1° (24.7°)	21.6° (21.4°)	22.4° (21.8°)	21° (16°)	0.28

Note. TP = time-point, M = mean, SD = standard deviation

## **Discussion**

### ***Construct validity of the WPST***

This study established preliminary psychometrics of the WPST for children with CP. The WPST detected a larger error in sensing wrist position for children with CP than in TD children of comparable ages, indicating that it is able to discriminate presence of impairment in children with CP as hypothesised. The current findings are consistent with those for adults (Carey et al., 1996), and with similar studies examining differences in kinaesthetic and proprioceptive ability for children with CP and their TD peers (Chrysagis, Skordilis, Koutsouki, & Evans, 2007; Goble, Hurvitz, & Brown, 2009).

These positive findings are likely due to the strengths in the design of the WPST test protocol. Test protocols requiring children to memorise limb position prior to a response, complete cross-modal matching (proprioceptive to visual transfer), and remain in an extended physical position (limb distal to trunk or standing position) add a degree of difficulty that may confound scores of proprioceptive abilities (Smorenburg et al., 2012; Wann, 1991). Deficits such as impaired working memory, impulsive behaviour, attention deficit and sensorimotor impairment can impact the performance of children with CP (Dourado, Andrade, Ramos-Jorge, Moreira, & Oliveira-Ferreira, 2013). Therefore, the WPST test procedure was designed to minimise additional cognitive and sensorimotor demands and not confound proprioceptive ability. The WPST is performed seated with the upper limb in a neutral position supporting precise localisation of the limb in proximity to the body. The WPST test procedure also has the advantage of allowing subjects to indicate wrist position whilst the arm remains in that position, minimising memory demands (Carey, 1993). The WPST appears appropriate for children with CP, with the observer assisting achievement of the desired wrist position so that the participant is not hampered by any spasticity or muscle weakness that they may have.

### ***Association between WPST and BBT***

In the present study, the WPST demonstrated a statistically significant inverse association between mean error in wrist position and performance on the BBT. In the absence of a standardised measure of limb position sense to compare with, establishing a clear positive association with an upper limb activity measure supports

the WPST as an evaluative tool. The WPST indicated a positive relationship between wrist position sense and manual dexterity. Therefore, the WPST could provide future opportunities for improved reporting of proprioceptive deficits and their impact on hand function.

### ***Responsiveness of the WPST***

The WPST was responsive to change in the treatment group following an intervention involving somatosensory discrimination training, as was also seen in adult stroke survivors (Carey et al., 2011). A significant change in scores on the WPST was found in those that received the intervention, but not in the control group. The treatment and control groups were well matched for age, gender and MACS score: they were less well matched for laterality of hemiplegia, but the significance of this difference for this trial is unknown. A post hoc power calculation was completed to determine the optimal sample size to detect responsiveness changes for this study. G\* Power (Faul, Erdfelder, Buchner, & Lang, 2009) was used to compute power, given that alpha was set to .05, N was 17, and the effect size was 0.9052 (calculated from the parallel PhD, see Appendix H of this thesis). The power calculation identified that this study had sufficient participants and adequate power (0.95) to detect accurate findings with the inferential statistics used. The ability of the WPST to detect impairments and changes over time due to treatment may be due to the greater sensitivity of an interval scale in comparison to an ordinal or categorical measurement scale (Carey et al., 1996; Portney & Watkins, 2009).

The WPST identified a large effect size from baseline to final post-intervention measurement. Unfortunately, the current study was not designed to determine how much change was clinically important. Because of the positive association reported in this current study it is recommended that a minimal clinically important difference be established through research comparing individual change in wrist position sense with changes in unilateral or bimanual performance as assessed by activity measures such as the BBT, or Assisting Hand Assessment (AHA) (Krumlinde-Sundholm & Eliasson, 2003), or somatosensory based goals, as assessed by Goal Attainment Scaling (GAS) (Kiresuk, Smith, & Cardillo, 1994) or the Canadian Occupational Performance Measure (COPM) (Law et al., 1998).

Clinical evaluation and treatment of children with CP has traditionally focused on addressing motor impairment without considering the impact of somatosensory impairment such as limb position sense (Russo, 2011). The WPST has the potential to predict improvements in functional outcomes where other measures of impairment have failed to do this (James et al., 2015). Understanding the unique nature of CP and the impact of somatosensory deficits on functional outcomes will help guide further development of somatosensory assessments and more precisely tailored rehabilitation and treatment strategies (Nevalainen et al., 2012).

### **Limitations**

The TD population was not matched for gender or age due to the recruitment method being a convenience sample. This is a limitation of the study as the TD group ended up having a mean age of one year older than the group with CP. The authors acknowledge that the construction of the WPST apparatus as illustrated in Figure 20 measures only one dimension of proprioception, passive limb position and also might provide additional tactile input to the hand within the splint. However, even if there were any such effects, under standard conditions of testing it would remain constant and therefore any changes or differences in the values of measurements (over time or between groups) would be valid. The research design was intended to examine the psychometric properties of construct validity, reliability and responsiveness in as much detail as possible however, the small sample size prevented investigation of all possible confounders.

Engagement in the WPST testing procedure was potentially influenced by personal and environmental factors such as time of day, attention and arousal level, assessment setting and developmental level. In this research, sensory integration, and behavioural or emotional responses to somatosensory stimuli that may have been triggered by imposed touch was not addressed. In addition, a larger sample size would have allowed for investigation of correlations between the WPST and other outcome measures such as, the AHA (Krumlinde-Sundholm & Eliasson, 2003), GAS (Kiresuk, Smith, & Cardillo, 1994), or COPM (Law et al., 1998) thus enabling further investigation of construct validity.

## **Conclusion**

Construct validity, intrarater reliability, and responsiveness of the WPST have been demonstrated for children with CP, and the WPST has been shown to be associated with a measure of unilateral upper limb activity. This study contributes to the establishment of psychometric properties for a novel clinical assessment tool and advances clinical measurement of wrist position sense for children with neurological conditions such as CP.

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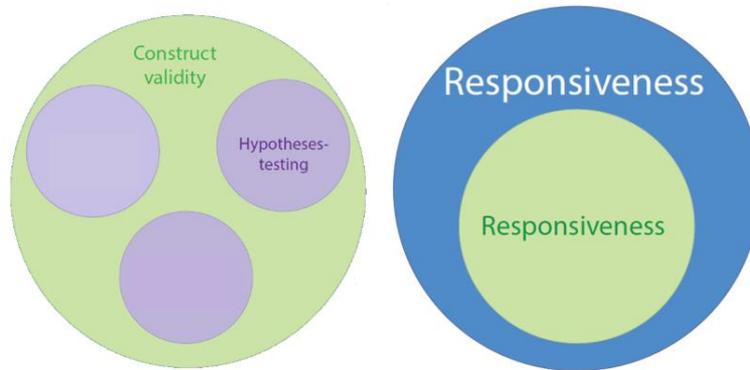
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## CHAPTER 6 functional Tactile Object Recognition Test



Phase three – Psychometric testing using the COSMIN

### 6.1 Introduction to chapter

The last chapter established preliminary psychometrics for a measure of wrist position sense for children with cerebral palsy (CP). This chapter seeks to do the same for a measure of haptic object recognition using a similar methodology. In the past, measures of haptic object recognition for children have proven difficult to refine and standardise (Bolanos et al., 1989; Cooper et al., 1995; Klingels et al., 2010). Confounding factors such as perceptual, and cognitive or cross modal transfer demands of tests, and appropriate selection of functional objects have been examined in the literature (Gori, 2015; Krumlinde-Sundholm, 2002). What is known is that: many objects representing a range of sensory properties increase a test's discriminative ability; objects that are standardised provide more reliable data; and enjoyable and engaging measures are more readily accepted particularly in younger populations (Carey, 1993; Nankervis, 2006; Ziviani, 2013). It is worth pointing out that Chapters five and six share the same paediatric sample however, one child could not complete the fTORT due to difficulties inattention. Therefore, there were six participants in the intervention group in this chapter and seven in the previous chapter.



## **6.2 PAPER THREE: functional Tactile Object Recognition Test**

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The PhD Candidate, Susan Taylor accounted for 85 per cent of the intellectual property associated with the final manuscript. Collectively, the remaining authors contributed 15 per cent.

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### **Construct validity and responsiveness of the functional Tactile Object Recognition Test for children with cerebral palsy**

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## **Abstract**

**Introduction:** The functional Tactile Object Recognition Test (fTORT) is a measure of haptic object recognition capacity recently adapted for use with children with neurological impairment. The current study aimed to investigate the construct validity and responsiveness of the fTORT and its association with a measure of upper limb activity.

**Methods:** A cross-sectional study of 28 children with spastic hemiplegic cerebral palsy (CP) (mean age 10y 8m; SD 2y 4m; 16 male) and 39 typically developing (TD) children (mean age 11y; SD 2y 9m; 19 male) was utilised to investigate construct validity and association between measures. Sixteen children with CP (mean age 10y 10m; SD 2y 8m; 9 male) were randomly allocated to either a treatment (n = 6) or control group (n = 10) and assessed at four time-points to assess test responsiveness.

**Results:** TD children scored significantly higher ( $p < .0001$ ) on the fTORT (M = 2.83, SD = 0.17) than those with CP (M = 2.19, SD = 0.78). fTORT scores demonstrated a significant association with scores on the activity measure ( $p < .01$ ). There were no significant between or within group differences in fTORT scores across the four time-points for the treatment or control group ( $p = .22$ ).

**Conclusion:** The fTORT demonstrated construct validity, and was positively associated with an upper limb activity measure but scores did not change significantly following somatosensory training. Further testing is currently underway to comprehensively investigate the psychometric properties of the tool.



## **Introduction**

Somatosensation is important for the development of hand function and determines how accurately and efficiently the hands are used to execute daily activities (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Dunn et al., 2015). Somatosensation includes submodalities of tactile registration and tactile perception, vibration, deep pressure, proprioception including limb position sense, temperature sensation, and emotional responses to pain and itch (Carey, 2012; Dunn et al., 2013). Haptic object recognition<sup>4</sup> is a complex somatosensory function relying on two haptic subsystems to extract sensory information about an object (Lederman & Klatzky, 2009). The sensory subsystem uses cutaneous, thermal, and kinesthetic receptors to obtain external information which informs the sensorimotor subsystem, resulting in active object manipulation (Lederman & Klatzky, 2009). An unimpaired individual uses their hands to capitalise on somatosensory input through the use of haptic exploratory procedures (Lederman & Klatzky, 1987; Morash, 2016). Haptic exploratory procedures are purposeful movements used to extract information about an object such as its material (e.g. texture or temperature) or geometric qualities (e.g. shape or size) (Lederman & Klatzky, 1987; 1993).

From infancy, children experience significant haptic development with the hands undergoing rapid and ongoing changes in haptic capacity well into adolescence (Fitzpatrick & Flynn, 2010; Gori, 2015). Although development and refinement of the sensory system is expected with increasing age, typical somatosensory development can be disrupted by neurological disturbances such as is seen in cerebral palsy (CP) (Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). CP is a collective term applied to a group of permanent motor disorders resulting from neurological injury occurring pre natally or in the early years of life (Rosenbaum, 2003). Deficits in haptic object recognition are common for children with CP, between 57% to 86%, and these deficits correlate with diminished dexterity of an affected hand (Cooper et al., 1995; McLean, Taylor, Valentine, Carey, & Elliott, 2017b; Sakzewski, Ziviani, & Boyd, 2010; Yekutieli, Jariwala, & Stretch, 1994). These deficits can present bilaterally or unilaterally, and often with more than one somatosensory domain involved (e.g.

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<sup>4</sup> Please note haptic object recognition is often called stereognosis in current literature.

proprioception and haptic object recognition) (Auld, Ware, Boyd, Moseley, & Johnston, 2012b; McLean et al., 2017b; Wingert, 2007).

### **Literature review**

While challenging, there is a clinical need to quantify haptic object recognition ability and its impact on hand dexterity in children with CP in order to tailor effective interventions (Klingels et al., 2010). At present, there are limited assessments with robust psychometric evidence or standard procedures to measure the haptic object recognition ability of children with CP (Connell & Tyson, 2012; Krumlindesundholm & Eliasson, 2002; Taylor et al., 2016). Auld, Boyd, Moseley, & Johnston (2011) recently appraised the clinimetric properties of tactile assessments for children with CP reporting that only three measures of stereognosis out of fourteen were considered to provide consistent assessment methodology and robust clinimetric properties. They were the Manual Form Perception (MFP) test, the Klingels method, and the Coopers method (Ayres, 1988; Cooper, Majnemer, Rosenblatt, & Birnbaum, 1993; Cooper et al., 1995; Klingels et al., 2010). However, only the Klingels method was recommended for use due to its superior clinical utility which was lacking in the other measures. The Klingels test of stereognosis consists of 12 familiar objects, with three matched pairs similar in shape and size (pencil/pen, coin/button, paperclip/safety pin) and six objects that differ markedly from each other (key, clothespin, marble, comb, spoon, ball). The child is provided with reference objects placed in the visually occluded hand, and is asked to identify the test object in their hand by providing a verbal or physical response (pointing). Standardisation is limited in that objects are sourced by the tester so the specific sensory attributes of the items may differ. The Klingels test has also demonstrated a ceiling effect for TD children (Intraclass Correlation Coefficient - ICC = 0) and lacked reliability for children with CP (% exact agreement = 47% to 50%) (Auld et al., 2012b).

Due to the limitations of existing measures, a newly adapted assessment for the evaluation of haptic object recognition for children has been developed. The functional Tactile Object Recognition Test (fTORT) (Carey, Matyas, & Macdonell, 2011; Nankervis, 2006) was designed to measure the haptic object recognition ability of adults following stroke. The fTORT was adapted by the current authors to detect

changes in this somatosensory ability for children with CP aged 6 to 15 years. The fTORT has demonstrated clinical acceptability for children with CP aged 6 to 15 years (Taylor et al., 2017d) and also has age-adjusted normative standards, high reliability ( $r = .85$  to  $.92$ ) and good discriminative test properties for adults aged 21 to 79 years (Carey et al., 2011).

Employing a structured approach to establish comprehensive psychometrics for the fTORT, the design of this study and subsequent reporting was based on the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010). The COSMIN provides explicit criteria for good measurement properties to improve the design of studies examining measurement instruments. Following the COSMIN taxonomy, this study examined the domains of construct validity (via differences between relevant groups) and responsiveness (detecting anticipated changes over time in haptic object recognition ability). In the absence of a criterion measure of haptic object recognition to conduct criterion related validity as specified in the COSMIN, we investigated the association of the fTORT scores with scores on an activity measure of unilateral upper limb performance.

It was hypothesised that the fTORT would reflect known differences in somatosensory performance between those who are typically developing (TD) and individuals with CP who are at risk of a somatosensory deficit. Second, it was hypothesised there would be a significant association between performance on the fTORT, and the Box and Block Test of Manual Dexterity (BBT) (Mathiowetz, Federman, & Wiemer, 1985a). For responsiveness testing it was hypothesised that: i) the fTORT would detect statistically significant differences over time between children receiving standard care and those additionally exposed to a somatosensory intervention; and ii) the fTORT would detect statistically significant differences over time in the treatment group.

## **Methods**

### ***Study design***

Three different study designs involving psychometric testing were conducted as part of this research and involved the fTORT and BBT as the primary outcome measures. Children with CP and TD children were tested with the fTORT. Children with CP

were also tested multiple times, pre and post an intervention, and with the BBT. The assessor was an occupational therapist trained in the use of the assessments who could not be blinded to groups because of the clinical presentation of children with CP, however for the responsiveness study the assessor was blinded to allocation of the participants with CP. The study was conducted in Perth, Western Australia and the data were collected in participant's homes in metropolitan and semi-rural areas of Perth. Recruitment for the TD participants occurred via professional and personal contacts and data collection for both TD and CP groups occurred between March Recruitment and data collection occurred between March 2013 and December 2014. The study protocol was approved by the Human Research Ethics Committee of Curtin University (#87) and Princess Margaret Hospital for Children (#2052), Perth, Australia.

**Construct validity study:** Comparison of fTORT between CP and TD children.

Employing a cross-sectional study design and known groups method, fTORT scores obtained from TD children ( $n = 39$ ) at one time-point were compared with the baseline measurement of children with spastic hemiplegic CP ( $n = 28$ ) prior to an intervention.

**Association study:** Comparison between fTORT and BBT in children with CP.

The BBT (Mathiowetz et al., 1985a) was administered to those children with CP ( $n = 28$ ) also at baseline for comparison with their baseline fTORT scores prior to an intervention.

**Responsiveness study:** Ability to detect change in fTORT pre-post somatosensory intervention.

Responsiveness of the fTORT was measured using data from a recent intervention trial with random allocation examining the efficacy of a novel somatosensory intervention based on perceptual-learning to improve somatosensory discrimination (McLean et al., 2017a; Carey et al., 2011). This trial is registered with the Australian New Zealand Clinical Trials Registry (ACTRN12614000314628) and has demonstrated preliminary efficacy in improving somatosensory capacity for children with hemiplegia CP aged 6 to 15 years (McLean et al., 2017a). Children with CP

were randomly allocated to treatment (n = 6) or control group (n = 10). The treatment group received standard care and also somatosensory discrimination training three times a week for six weeks and the control group received standard care only. The fTORT and BBT were among the primary outcome measures administered at four time-points in the intervention study.

### ***Participants***

Inclusion criteria for the construct validity and association study were children with spastic hemiplegic CP with a diagnosis confirmed by a paediatrician, aged 6 to 15 years. Exclusion criteria included surgery on the affected upper limb in the previous 12 months or inability to understand or respond to simple instructions. Children without CP who participated in the construct validity were a convenience sample of individuals aged 6 to 15 years. Eligibility criteria for the responsiveness study were the same as for the construct validity and association study plus presence of haptic object recognition impairment as measured by the fTORT with relation to the normative standards of object identification accuracy provided in this paper. Participants were assessed in their own homes at a time most suitable to the individual and family (usually after school or weekend mornings). All efforts were made to ensure the assessment environment was quiet and free from distraction.

### **Outcome measures**

#### ***The functional Tactile Object Recognition Test***

The fTORT administration manual and equipment (Figure 22) was originally developed for adults with stroke (Carey et al., 2011) and in this study, was adapted by the current authors for use with children and youth with neurological impairment. Evidence based adaptation of the fTORT instructions and testing procedure to a paediatric version was based on consideration of semantics and perceptual concepts appropriate to developmental level (Berlin, Blank, & Rose, 1980; Bloom, 2001). The following adaptations were made 1) standardised script instructions condensed to single step instructions; 2) use of language and concepts understood at a primary school level; 3) encourage children to focus attention on the examiner, or on the relevant aspects of the test procedure as required; 4) ensure adequate time is given for responses; 5) demonstrate what is required; 6) confirm comprehension of instructions by asking the child to repeat what they have been asked to do; 7) use



Table 7 Summary of the 14 object sets included on the fTORT response poster

Object set	Diagnostic sensory attribute	Relative size
Half full milk bottle (EO)	Weight	Large
Full milk bottle (CO)	Weight	
Empty soft drink bottle (DO)	Weight, shape & size	
Metal door knob (EO)	Temperature	Large
Wooden door knob (CO)	Temperature	
Glass hexagonal door knob (DO)	Temperature & texture	
Hardcover book (EO)	Hardness	Large
Paperback book (CO)	Hardness	
Small magazine (DO)	Hardness & size	
Metal bowl (EO)	Temperature	Large
Plastic bowl (CO)	Temperature	
Plastic plate (DO)	Temperature, size & shape	
Material and zipper (EO)	Function/motion	Small
Material and buttons(CO)	Function/motion	
Material and belt buckle (DO)	Function/motion, size & shape	
Click light switch (EO)	Function/motion	Small
Turn light switch (CO)	Function/motion	
Electric power point (DO)	Function/motion, size & shape	
Spoon (EO)	Shape	Large
Fork (CO)	Shape	
Chopstick (DO)	Shape, size & texture	
Large faced watch (EO)	Size	Small
Small faced watch (CO)	Size	
Stop watch (DO)	Size & shape	
Full glass mayonnaise jar (EO)	Weight	Large
Empty glass mayonnaise jar (CO)	Weight	
Glass spice grinder (DO)	Weight, size & shape	
Soft plastic cup (EO)	Hardness	Large
Hard plastic cup (CO)	Hardness	
Metal cup (DO)	Hardness, size & temperature	
Plastic credit card (EO)	Texture	Small
Small paper card (CO)	Texture	
Flat fridge magnet (DO)	Texture, size & shape	
House key (EO)	Size	Small
Filing cabinet key (CO)	Size	
Padlock (DO)	Size, weight & shape	
Wooden clothes peg (EO)	Texture	Small
Plastic clothes peg (CO)	Texture	
Plastic hook (DO)	Texture, size & shape	
Spiral pasta (EO)	Shape	Small
Cylinder pasta (CO)	Shape	
Raisin (DO)	Shape, texture & size	

*Note.* EO = exact object; CO = comparator object; DO = distractor object. Adapted with permission from “The SenScreen© Sensory Screen Tool Administration Manual” by L. M. Carey, Y. Mak, and A. M. Tan, 2011.

*Task and procedure.* The assessment began with the examiner reading through the script instructions followed by a familiarisation phase and practice trial. The participant's arm was placed with the palm facing up behind the curtain. Depending on size the objects were placed into the palm of the participant's hand, between the thumb and first two fingers, or the object was placed on the table and the participant's pronated hand and fingers were moved over it. Manipulation was unilateral and participants were assisted to perform the appropriate exploratory procedures by the examiner. This assistance ensured adequate exploration of the most salient features of the objects. Each child was presented with 14 test objects and verbalised their response or pointed to the object on the response poster. The participants were allowed to explore the items for a free period of time however if a participant had severely impaired sensation and exceeded 30 seconds of exploration the fTORT subtest was ceased. This value was based on calculating the average time 28 participants with cerebral palsy took to explore objects within the fTORT (average of 16 seconds of exploration  $\pm$  13 seconds).

*Scoring and interpretation.* The child's response was marked as being completely correct (exact object, score=3), selection of object differing in one sensory attribute (comparator object, score=2), object differing in more than one sensory attribute (distractor object, score=1) and completely incorrect (object other than object set, score=0). The measurement outcome, accuracy in recognising objects, was determined by calculating a score out of a possible 42. Observations of the 39 TD participants within this study generated preliminary normative data for the fTORT for children aged 6 to 15 years. Based on these observations a score  $>36$  for the right and left hand indicates intact haptic object recognition ability for 6 to 8 year olds;  $>38$  (right and left hand) for 9 to 11 year olds; and  $>39$  (right hand)  $>38$  (left hand) for 12 to 15 year olds. Scores below this range may indicate impaired haptic object recognition. The fTORT uses a rank order scale to produce ordinal data. Administration and scoring takes approximately 15 minutes per hand depending on the personal factors and characteristics of each child.

### ***Box and Block Test of Manual Dexterity***

The BBT is an activity measure of gross manual dexterity and has high test re-test reliability for TD children and adults ( $r = .84$ ;  $r = .96$ ), high concurrent validity, and

high inter-rater reliability (ICC 0.99) for adults (Jongbloed-Pereboom, Nijhuis-van Der Sanden, & Steenbergen, 2013; Mathiowetz, Volland, Kashman, & Weber, 1985b) and is commonly used in research with children with CP (Golubović & Slavković, 2014; Holmström et al., 2010). The number of blocks transferred unilaterally in 60 seconds is the recorded outcome of the test.

### ***Data analysis***

To determine construct validity individual scores on the fTORT were compared between participant groups using a Generalised Estimating Equation (GEE) (Portney & Watkins, 2009). This model classified the response to each challenge as correct or incorrect, and took into account any correlation between the multiple responses made by each participant. Analyses were performed with different definitions of ‘correctness’: 1) Exact object, 2) Exact or Comparator object, 3) Exact, comparator or distractor object. In addition, an overall score for each participant was calculated, and the mean scores were compared between the participant groups using a simple t-test. For individuals with CP, associations between scores on the fTORT and BBT were examined by including the BBT score as an independent variable in the GEE model. To examine responsiveness, fTORT scores of each participant in the intervention and control groups at each of three post baseline time-points were subtracted from the baseline score, and the mean change, SD and effect size (mean change/ group SD) at each time-point calculated. A random effects regression model was used to explore any changes in scores over time. Naming the person identifier as the random effect meant that correlations between scores for the same participant were considered. Effect size was considered small if between  $\geq 0.1$  and  $< 0.3$ , medium if between  $\geq 0.3$  and  $< 0.5$  and large if  $\geq 0.5$ . (Cohen, 1992). Statistical analyses were conducted using the SAS version 9.2 software (SAS Institute, Cary, NC, USA 2008), and  $\alpha$  was set at  $p < 0.05$ .

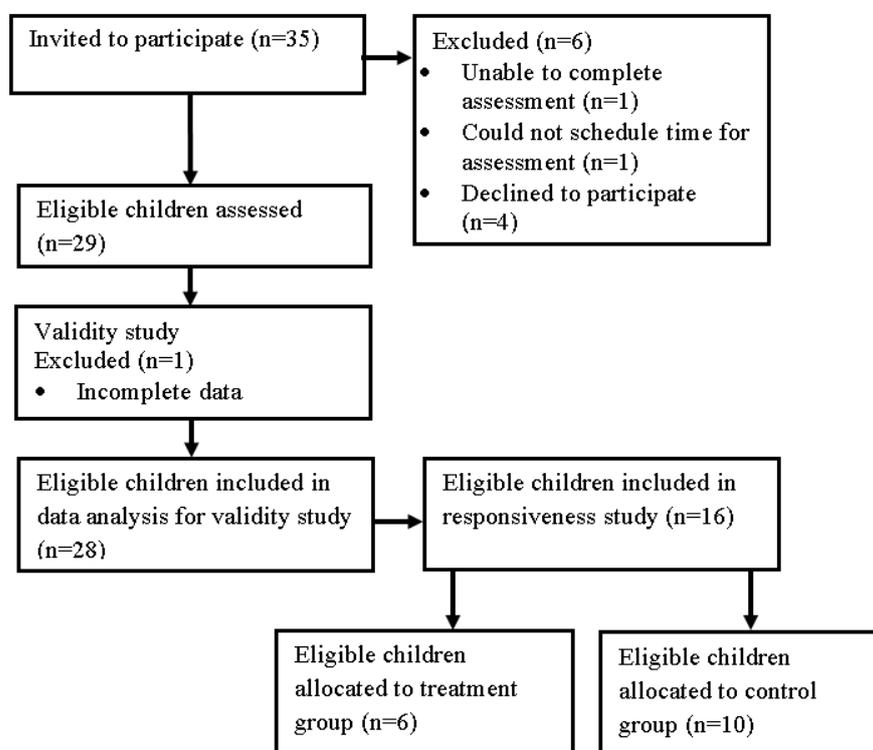
## **Results**

### ***Participants***

Recruitment occurred between March 2013 and December 2013 and 28 children with CP were confirmed eligible and included in the analysis (age range 6y - 15y 6m; mean age 10y 8m; SD 2y 4m; 16 male; MACS level I n = 10; right hemiplegia n = 15). A Manual Ability Classification System (MACS) of level I or II indicates only

minor difficulties in handling objects requiring fine motor control (Eliasson et al., 2006) therefore, participants with a MACS level of I or II were included in the study. Thirty-nine TD children were recruited and included for analysis (age range 6y - 15y 5m; mean age 11y; SD 2y 9m; 19 male; right hand dominance n = 37). For the responsiveness study 16 participants (treatment n = 6; control n = 10) recruited for the intervention study were included for analysis (age range 6y - 15y 5m; mean age 10y 10m; SD 2y 8m; 9 male; MACS level I n = 3; right hemiplegia n = 7) (Figure 23).

Figure 23 Flowchart of recruitment



### ***Construct validity of the fTORT***

A t-test to compare the total fTORT scores between children with and without CP showed a very significant difference ( $t_{65} = 4.9$ ;  $p$  value  $< .0001$ ) indicating greater haptic object recognition ability for the TD group ( $n = 39$ ;  $M = 2.83$ ,  $SD = 0.17$ ) than the group with CP ( $n = 28$ ;  $M = 2.19$ ,  $SD = 0.78$ ). Throughout administration of the fTORT for children with CP there was no systematic correlations between fTORT scores and order of test object presentations. This suggests that performance was not affected by attention span or a learning effect.

### ***Association between the fTORT and BBT***

There was a significant negative association between scores on the fTORT and BBT ( $p < .01$ ) such that increased manual dexterity (number of blocks moved) was correlated with fewer errors on the fTORT.

### ***Responsiveness of the fTORT***

There were no statistically significant differences in fTORT scores between time-points for the treatment or control group ( $F_{3,45} = 1.52$ ;  $p = 0.22$ ) There was also no significant increase in mean fTORT scores for either the treatment or control group between any time-points (Table 8; p-values obtained from the random effects regression model).

Table 8 functional Tactile Object Recognition Test mean scores for the treatment and control group

Timepoint	TP 1 M(SD)	TP 2 M(SD)	TP 3 M(SD)	TP 4 M(SD)	P value
Treatment ( $n = 6$ )	23.4(14.5)	25.7(14.2)	25.8(13.6)	24.1(14.3)	0.50
Control ( $n = 10$ )	27.3(8.4)	27.9(12)	31.2(10)	29.9(11.7)	0.34

*Note.* fTORT = functional Tactile Object Recognition Test; TD = typically developing; CP = cerebral palsy; M = mean; SD = standard deviation.

## **Discussion**

This research aimed to examine the preliminary psychometric properties of the fTORT for use with children with CP. The COSMIN (Mokkink et al., 2010) provided a framework by which to design the research methodology. The following is a synthesis of these early findings for the fTORT in children with consideration of future research directions.

### ***Construct validity of the fTORT***

The fTORT reflected statistically significant differences between children and adolescents diagnosed with CP at risk of somatosensory deficits, and TD children. The present findings of construct validity support a previous study with adults, where the fTORT demonstrated good discriminative validity by distinguishing impaired somatosensory performance of adult stroke survivors from healthy age-matched adults (Carey et al., 2011). The discriminative ability of the fTORT may be due to the theoretically driven selection of test objects (Carey et al., 2006). Previous literature advocates the use of familiar everyday objects in haptic object recognition

assessment methodologies for children (Auld et al., 2011; Yekutieli et al., 1994). However more recently studies have indicated a ceiling effect for measures of stereognosis that use common objects and suggest that novel or more complex objects are needed to reflect changes in performance over time (Auld et al., 2012b; Russo, 2011). None of the participants in the current study (TD or CP) achieved a maximum score on the fTORT, and variability in performance was seen across age groups. These results may indicate that the fTORT objects are familiar enough to be recognised by adults and children, but sufficiently novel to prevent a ceiling effect (Nankervis, 2006). Alternatively, the design of the tool which allows for graded item difficulty within and across item sets based on number of varied sensory attributes may be sufficiently challenging even with the use of common familiar objects (Carey et al., 2011).

#### ***Association between fTORT and BBT***

We found a statistically significant association between performance on the fTORT and the BBT, indicating that unilateral dexterity was associated with better haptic object recognition. Other studies suggest that stereognosis testing using common objects, is a more robust measure of sensory deficit than impairment measures (e.g. 2-point discrimination) when testing young children with CP (Yekutieli et al., 1993). In a recent study by James et al., (2015), the authors found that impairment measures, including range of motion, did not reliably predict the functional dynamic range of motion used to perform activities for children with upper extremity CP. Our study has the potential to address practice gaps such as these for haptic perception. Weemonstrated that a measure of haptic object recognition capacity was associated with an activity measure, supporting the use of measures of ability rather than measures evaluating the impairment level.

*Strengths of fTORT.* A potential problem associated with other haptic object recognition testing procedures, is they rely on independent in-hand manipulation techniques and a child's ability to perform haptic exploratory procedures (Auld et al., 2011). In-hand manipulation can be significantly compromised for children with neurological and musculoskeletal deficits due to CP (Van Heest et al., 1993). Movement of an object across the skin by an examiner has been found to achieve success equivalent to a child performing the exploration themselves (Cermak, 2006).

Recent studies have recommended removing the need for in-hand manipulation when testing children who cannot move the object themselves (Auld et al., 2011). This supports our use of standardised motor-assisted haptic exploration using matched exploratory procedures as a strategy to compensate for the neurological and/or musculoskeletal impairments experienced by children with CP that may restrict in-hand manipulation and expected exploratory procedures.

*Limitations of fTORT.* The fTORT requires mapping between visual and haptic information. This cross-modal matching increases the cognitive and visual perceptual demands of the testing procedure. There are many other performance components required for successful completion of this task not just somatosensory processing (Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2012; Wann, 1991). The fTORT requires a moderate level of cognition, cross-modal transfer, visual perceptual, and language and motor skills during test instructions and in the response mode. Although it is difficult to remove these elements when testing haptic object recognition, the possible confounding effects on test scores cannot be ignored.

The TD population was not matched for gender or age due to the recruitment method being a convenience sample. This is a limitation of the study as the TD group ended up having a mean age of one year older than the group with CP. Engagement in the fTORT testing procedure may be influenced by personal and environmental factors such as time of day, attention and arousal level, assessment setting and developmental level. Extensive work has been done on the importance of engagement and self-determination for children during therapy and the assessment process (Cuskelly & King, 2013; Poulsen, Ziviani, & Cuskelly, 2013). Assessments involving structured play or game based activities and active participation of the child encourage self-motivation to complete tasks and may have more utility in clinical practice (O'Grady & Dusing, 2015; Poulsen, Ziviani, & Cuskelly, 2013). However, play or game based assessments may not be suitable to measure haptic object recognition in children with CP because they further increase the number of performance components required to complete the task successfully. It is important to understand the potential demands that somatosensory assessment may place on other mental functions and to ensure that assessment occurs without consequence for deficits in these skills (Rosenbaum et al., 1990).

### ***Responsiveness of the fTORT***

In the current study, there were no significant differences in fTORT scores between the treatment and control group, and no significant changes in fTORT scores over time in either group. However, in a similar study involving adult participants, the fTORT was able to detect statistically significant differences between a treatment ( $n = 25$ ) and control group ( $n = 25$ ) (Carey et al., 2011). The treatment group received the same somatosensory discrimination intervention as this study. Between-group differences were statistically significant  $p = 0.004$  with greater improvement in somatosensory capacity reported for the treatment group. Differences in these results may be attributed to a larger sample size ( $N = 50$ ), the possibility that adult participants displayed a vested interest in therapy outcomes to achieve improvements, and had all their mental faculties intact pre-stroke. It is also possible that: 1) the fTORT is not responsive to change in children; 2) the children had no change as a result of the intervention; 3) the current study was insufficiently powered to detect a change. A post hoc power calculation was completed to determine the optimal sample size to detect responsiveness changes. G\* Power (Faul, Erdfelder, Buchner, & Lang, 2009) was used to compute power, given that alpha was set to .05,  $N$  was 16, and the effect size was 0.9 (calculated from the parallel PhD, see Appendix H of this thesis). This post-hoc power analysis identified that the fTORT study was adequately powered (0.94) and therefore is an unlikely contributing factor to the poor responsiveness findings.

Difficulties in testing responsiveness have been documented in paediatric literature for new clinical outcome measures (Jerosch-Herold, 2005). Extraneous information in these types of instruments can lead to decreased responsiveness (Law, 1987). The fTORT was designed as both a discriminative and evaluative measure, and as such requires further responsiveness testing. Future responsiveness testing is recommended to involve broader sub-groups of CP and evaluation of responsiveness with a sample size based on a *priori* power analysis (Portney & Watkins, 2009).

Reliability testing of the fTORT within the population of interest is also necessary to examine any influence of measurement error on estimates of responsiveness. Measures of somatosensation are routinely used in rehabilitation settings and as outcome measures for interventions targeting upper limb functioning,

despite the absence of known psychometric properties for children (Klingels et al., 2010). The fTORT is in an early phase of development. Future research testing reliability, responsiveness and clinical utility with a larger sample size, different sub-groups of CP and other neurological conditions is required. This study aimed to provide a standardised outcome measure of haptic object recognition with adequate sensitivity to reflect changes in performance during future somatosensory intervention trials. Describing the fTORT apparatus and administration procedure, its application in clinical practice and initial psychometric properties, underpins successful clinical implementation in the future (Fixsen & Ogden, 2014).

## **Conclusion**

There are currently limited best practice measures for the evaluation of haptic object recognition for children with CP. The fTORT shows promise as a quantitative, discriminative measure of haptic object recognition and further work is needed to test and improve its responsiveness. Until further psychometric testing, the fTORT may be used, with careful monitoring, in paediatric clinical research to measure haptic object recognition. Findings from this study contribute to the development of psychometric properties for a novel clinical assessment tool and advance clinical measurement of haptic object recognition for children. Future research is needed to further understand haptic object recognition ability in children with neurological conditions and standardise guidelines for clinical assessment procedures.

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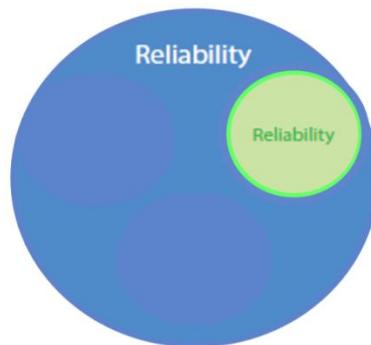
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## CHAPTER 7 Haptic Exploratory Procedures



Phase three – Psychometric testing using the COSMIN

### 7.1 Introduction to chapter

This chapter corresponds to Phase three and testing the preliminary psychometrics of the functional Tactile Object Recognition Test (fTORT) by examining the reliability of a video protocol to record and describe the haptic exploratory procedures (EPs) of children with cerebral palsy (CP). This chapter also aimed to describe the haptic EPs of children with and without CP of comparable age. Haptic object recognition is important for exploration of the immediate environment and is influenced by a child's use of haptic EPs. Haptic EPs are employed to extract a combination of cutaneous, thermal, and kinesthetic feedback and active in-hand manipulation to extract an object's sensory properties (Kalagher, 2015). Currently, there is no standardised method of recording the exploratory hand movements of children when manipulating and identifying objects. Knowledge of the relationship between haptic EPs and haptic object recognition ability has therefore been limited (Lederman & Klatzky, 1987; Nankervis, 2006).

The following paper describes the use of the standardised test procedure of the fTORT coupled with a digital recording to record the spontaneous use of haptic EPs used by children with and without CP when exploring the 14 common test objects of the fTORT. Comparing haptic EPs during object exploration will assist in developing paediatric specific recommendations for the fTORT test procedure and Sense© for Kids training protocol (Appendix H). Motor enhanced protocols for manipulation of objects during haptic object recognition testing has been recommended (Auld et al., 2012b). The information from this paper can inform protocols to enhance motor performance for

children with CP during the fTORT and Sense© for Kids training where motor function is compromised.

## **7.2 PAPER FOUR: Haptic exploratory procedures**

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The PhD Candidate, Susan Taylor accounted for 85 per cent of the intellectual property associated with the final manuscript. Collectively, the remaining authors contributed 15 per cent.

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### **Haptic exploratory procedures of children and youth with and without cerebral palsy**

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## **Abstract**

**Introduction:** The current study describes and compares the haptic exploratory procedures (EPs) and exploratory movements (EMs) of typically developing (TD) children and children with spastic hemiplegic cerebral palsy (CP). This study was also used to test the interrater reliability of a novel digital recording method.

**Methods:** Video data from a cross-sectional study were used to examine haptic EPs performed during the functional Tactile Object Recognition Test (fTORT) (Carey, Nankervis, LeBlanc, & Harvey, 2006). Fifty-four participants were recruited and consisted of 31 TD children (age range 6y 1m – 15y 9m; mean age 10y; SD 2y 9m; 14 male) and 23 children with spastic hemiplegic CP (aged 6y – 15y 5m; mean age 10y 2m (SD 2y 5m), 13 male; Manual Ability Classification System level I n = 11, level II n = 12; right hemiplegia n = 12).

**Results:** TD children and children with CP performed similar patterns of haptic EPs. There were no statistically significant differences between groups for expected EP ( $p = .146$ ), additional EPs ( $p = .778$ ), or EMs ( $p = .687$ ) but there was for mean duration of exploration ( $p < .001$ ) and accuracy ( $p < .001$ ). This suggests that although children with CP performed similar haptic EPs for each object as TD children, they took more time and were less accurate in their identification.

**Conclusion:** This study provides evidence for a reliable method of recording the use of haptic EPs during the standardised fTORT administration. This study also provides information to clinicians that will inform the development of evidence based assessments and treatments for haptic EPs for children with CP.



## Introduction

Identification of objects using the hands requires information from cutaneous, thermal, and kinesthetic receptors within the sensorimotor system to guide active object manipulation (Lederman & Klatzky, 2009). Typical patterns of object manipulation have been classified into eight typical hand movements, called haptic exploratory procedures (EPs) that are used to obtain specific object information (Lederman & Klatzky, 1987). Haptic EPs assist in identification of objects with and without vision (haptic object recognition), determine an object's function and guide motor planning (Klatzky, Lederman, & Mankinen, 2005). The hand movements differ depending on what type of sensory attribute is being explored. Lederman and Klatzky (1987) described seven common sensory attributes in their original work; texture, hardness, temperature, weight, global shape, exact shape, part motion and function. Lateral motion may be used to detect different textures, pressure might be employed to test the rigidity or hardness of an object and unsupported holding will help to detect an object's weight (Lederman & Klatzky, 1990). Some hand movements performed during haptic object exploration cannot be classified as EPs and they have been called 'actions' (Withagen, Kappers, Vervloed, Knoors, & Verhoeven, 2013), and in this current paper 'exploratory movements'. The term exploratory movements (EMs) was adopted in the current paper to align with consistent terminology used in the *sense\_assess© kids* Administration manual (e.g. exploratory procedures). Actions or exploratory movements can be described as additional patterns of hand movements beyond 'task maintenance' that often occur in conjunction with EPs and sometimes independently of them (Lederman & Klatzky, 1987, p. 351; Withagen et al., 2013). For example, a young child may use the EM of banging in place of, or in combination with the more efficient EP of pressure to test for rigidity (Lederman & Klatzky, 1987).

The development of haptic object recognition contributes directly to an infant's cognitive development and is an important developmental process underlying a child's ability to understand and control objects for occupational tasks. (Ballesteros, Bardisa, Millar, & Reales, 2005; Henderson & Pehoski, 2006; Yu, Hinojosa, Howe, & Voelbel, 2012). When the haptic system is disrupted by damage to the developing brain, as may be seen in CP, haptic exploration and haptic object

recognition are impaired (Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2008). Deficits in haptic object recognition are common for children with CP with a prevalence of between 57% to 86% (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Sakzewski, Ziviani, & Boyd, 2010; Van Heest, House, & Putnam, 1993; Yekutieli, Jariwala, & Stretch, 1994; McLean et al., 2017b). Many studies have investigated the haptic object recognition ability and development of haptic EPs for TD children (Kalagher, 2015; Molina et al., 2015) and children who are visually impaired (Bara, 2014; Withagen et al., 2013). However, to the authors' knowledge no studies have explored the haptic EPs or EMs of children with CP.

The functional Tactile Object Recognition Test (fTORT) (Carey et al., 2006) is designed to test the unilateral haptic recognition of objects without vision and record the use of haptic EPs during free object exploration. The original fTORT test forms (Carey et al., 2017) were used to record frequency and type of EPs by circling the observed EPs for each object on the test form. Listed on the test form were the expected EPs for each object based on evidence from Lederman and Klatzky (1987) and normative data from healthy adults (Carey et al., 2006). The expected EP was the most optimal and most common EP used for identification of fTORT objects by healthy adults (Carey et al., 2006; Lederman & Klatzky, 1987). Accuracy of object recognition was also recorded as a score out of 42, with 42 being the highest score achievable. The current study developed a new method of digitally recording the hand movements during the fTORT to accurately classify the observed haptic EPs and in addition, the presence of EMs. The child's hand was filmed from two different angles during testing as per the standardised video protocol described later.

This current study aimed to describe the haptic EPs of a sample of TD children and compare them to children with spastic hemiplegic CP with a Manual Ability Classification System (MACS) level of I or II (Eliasson et al., 2006) with the anticipation of developing recommendations for somatosensory intervention. It was hypothesised that: a) TD children aged 6 to 15 years would perform the same haptic EPs as previously classified in adults (i.e. expected EP per object) when exploring the sensory attributes of fTORT objects (Lederman & Klatzky, 1987); b) TD children would perform more expected EPs, less additional EPs and EMs, take less time to identify each object and correctly identify more objects than children with CP; c)

age, gender, hand dominance for the TD children or MACS level and presence of somatosensory deficit for children with CP would be associated with differences relative to TD; and d) interrater reliability of identification of EPs using the Cohen's kappa would be strong between two raters coding video data for children with and without CP (0.8 - 0.9).

## **Methods**

### ***Study design***

A cross-sectional study was employed to examine haptic object recognition performance and haptic EPs performed by children and youth during the fTORT. The study was conducted in Perth, Western Australia and the data were collected in participant's homes in metropolitan and semi-rural areas of Perth. Recruitment for the TD participants occurred via professional and personal contacts and data collection for both TD and CP groups occurred between March 2013 and December 2014. The study protocol was approved by the Human Research Ethics Committee of Princess Margaret Hospital for Children (#2052) and Curtin University (#87), Perth, Australia.

Using video data collected during the fTORT administration we measured the following variables:

1) Whether the expected EP was performed, the expected EP was the most optimal and most common EP used for identification of fTORT objects by healthy adults.

2) Additional EPs, which was a count of any additional EPs that were performed that were not most optimal for identification of the particular object but still fit within the classification of an EP. Additional EPs are often used in sequence with the expected EP to gather additional sensory information.

3) Frequency of EMs were recorded. EMs are movements that cannot be classified as EPs but are patterns that were observed frequently.

4) Time taken was the total time in minutes and seconds taken to identify an object, starting from when the object was placed in the hand until the participant selected the object from the response poster.

5) Accuracy in object recognition was measured as a categorical response of 'yes' correctly identified or 'no' incorrect.

### ***Participants***

Inclusion criteria were children aged 6 to 15 years with spastic hemiplegic CP with a confirmed diagnosis by a paediatrician. Exclusion criteria included surgery of the affected upper limb in the previous 12 months, or paralysis, spasticity or flaccidity of the upper limb that prevented voluntary motor control, or an inability to understand and respond to simple instructions such as, '*In this activity you will be asked to tell me the everyday objects I put in your hand to feel without looking*'. The TD study population was a convenience sample of school-aged children aged 6 to 15 years recruited through personal and professional contacts of the authors. Participants were excluded if they could not understand or respond to simple instructions.

### ***Outcome measure***

The fTORT was originally designed for use with adult stroke survivors but was adapted to create a paediatric version as part of this thesis (see Chapter three). The test contains 28 common everyday objects (Table 9) chosen to represent the sensory attributes of texture, hardness, temperature, weight global shape, exact shape, and function of an object, outlined in Lederman and Klatzky (1987). The fTORT also contains a response poster of 42 numbered objects, 28 of these objects are provided for manipulation during the fTORT and 14 objects are tested per hand. The numbered response poster is placed in front of the participant and one of the 14 test objects is placed in the participant's hand in sequence. The participant is then asked to identify what they are holding by choosing the corresponding object on the poster (Carey et al., 2017). Participants do not receive explicit or implicit instructions related to perceptual goals for the test objects. Accuracy of object recognition is recorded as a score out of 42. Based on normative data for the fTORT a score >36 for the left or right hand indicates intact haptic object recognition ability for 6 to 8 year olds; >38 (left and right hand) for 9 to 11 year olds; and >39 (right hand) >38 (left hand) for 12 to 15 year olds. Scores below this range may indicate impaired haptic object recognition.

Table 9 functional Tactile Object Recognition Test objects

Sensory attribute	Test Object	Expected EP
Temperature	Metal doorknob	Static contact
Temperature	Wood doorknob	Static contact
Temperature	Steel bowl	Static contact
Temperature	Plastic bowl	Static contact
Hardness	Hardcover book	Pressure
Hardness	Paperback book	Pressure
Hardness	Firm plastic cup	Pressure
Hardness	Crush plastic cup	Pressure
Function	Zipper	Part motion
Function	Buttons	Part motion
Function	Click switch	Part motion
Function	Turn light switch	Part motion
Shape	Cylinder pasta	Contour following
Shape	Spiral pasta	Contour following
Shape	Fork	Contour following
Shape	Spoon	Contour following
Size	Small watch	Enclosure
Size	Large watch	Enclosure
Size	House key	Enclosure
Size	Filing cabinet key	Enclosure
Weight	Empty jar	Unsupported holding
Weight	Full jar	Unsupported holding
Weight	½ full milk bottle	Unsupported holding
Weight	Full milk bottle	Unsupported holding
Texture	Paper card	Lateral motion
Texture	Plastic card	Lateral motion
Texture	Plastic peg	Lateral motion
Texture	Wooden peg	Lateral motion

*Note.* Adapted from L. Carey, Y. Mak, and A. M. Tan, 2011, SenScreen© Kids Administration manual.

Participants were assessed at one time-point in their own homes by an occupational therapist trained in the use of the fTORT. The test was administered in a standardised manner as per manual specifications with an approximate administration and scoring time of 15 minutes. The participant's hand movements were video-recorded using a tripod and camera positioned directly behind and above the examiners shoulder and tilted down at a 45° angle. The examiner was seated to the side of the participant (Figure 24). If the right arm was being tested, the examiner and camera was positioned on the right side and vice versa for the left. The video

data were coded to extract information about the occurrence of the variables under examination: 1) expected EP; 2) frequency of additional EPs; 3) frequency of EMs; 4) duration of response time and 5) accuracy.

Figure 24 Administration set up of the functional Tactile Object Recognition Test



### ***Data analysis***

General Estimating Equations (GEE) were used to compare between group differences (TD and CP) for the variables under examination. GEEs were also used to examine within group differences for age, gender, hand dominance or MACS level. Logistic regression modeling was used to analyse binary variables of expected EP and accuracy, linear regression was used for the continuous measure of time taken to respond and Poisson regression modeling was used for variables with multiple counts such as frequency of additional EPs and EMs. Rater reliability was determined by having a second coder evaluate 20% of the video data. Cohen's kappa was calculated to determine the agreement between raters' observations of the expected EP per object (Landis & Koch, 1977).

## **Results**

### ***Participants***

Fifty-nine participants were confirmed eligible and consisted of 31 TD children (aged 6y 1m – 15y 9m; mean age 10y; SD 2y 9m; 14 male) and 28 children with spastic hemiplegic CP (aged 6y – 15y 6m; mean age 10y 2m; SD 2y 4m); 16 male; MACS level I n = 11, level II n = 17; right hemiplegia n = 12). One child did not complete the fTORT test procedure as they were unable to attend to the task and

were therefore excluded from the data analysis. Four children with CP required hand over hand assistance from the examiner to explore the objects due to compromised motor control and the natural patterns of haptic EPs could not be examined so they were also excluded. Therefore, there were 23 children with CP included in the study and data analysis (age range 6 y - 15y 5m; mean age 10y 2m; SD 2y 5m; 13 male; MACS level I n = 11, level II n = 12; right hemiplegia n = 12). Of the 23 children confirmed eligible for this study 13 had a haptic object recognition deficit as identified by the fTORT.

## **Results**

### ***Reliability of method to record hand movements***

Cohen's  $\kappa$  estimated the frequency of non-chance agreement between the two raters' observations of video recordings of each child's hand movements. There was substantial agreement between the two raters' observations of expected EP,  $\kappa = .643$   $p < .0005$  (Landis & Koch, 1977).

### ***Normative performance***

TD children performed the same haptic EPs previously classified in adults (Carey et al., 2006; Lederman & Klatzky, 1987). Descriptive tables of type of EPs performed were computed for children with and without CP (Figures 25 and 26).

Figure 25 Count of haptic exploratory procedures performed by typically developing children

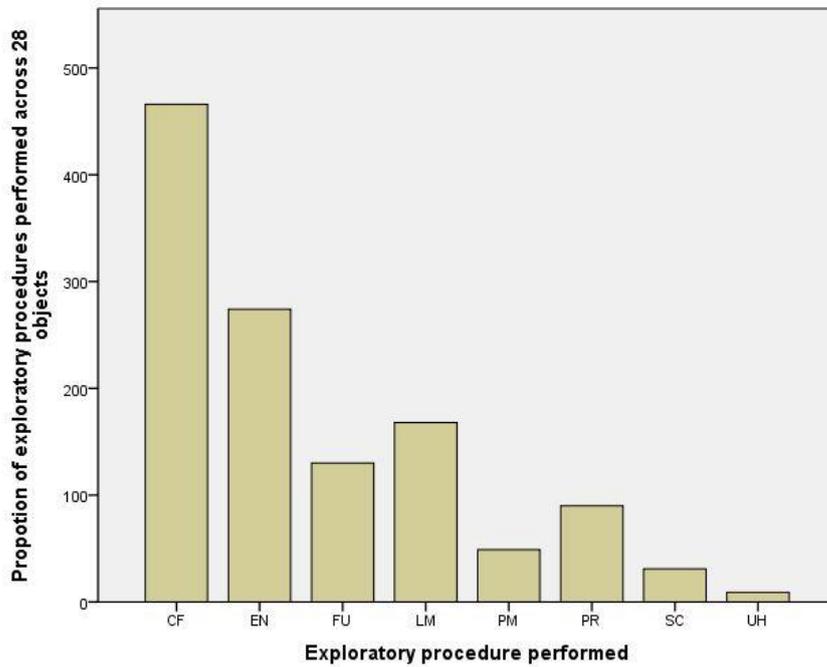


Figure 25. Graphic representation of exploratory procedures performed by 31 children without cerebral palsy for each functional Tactile Object Recognition Test object that was presented for manipulation in this study. CF = contour following; EN = enclosure; FU = function; LM = lateral motion; PM = part motion; PR = pressure, SC = static contact; UH = unsupported holding.

Figure 26 Count of haptic exploratory procedures performed by children with cerebral palsy

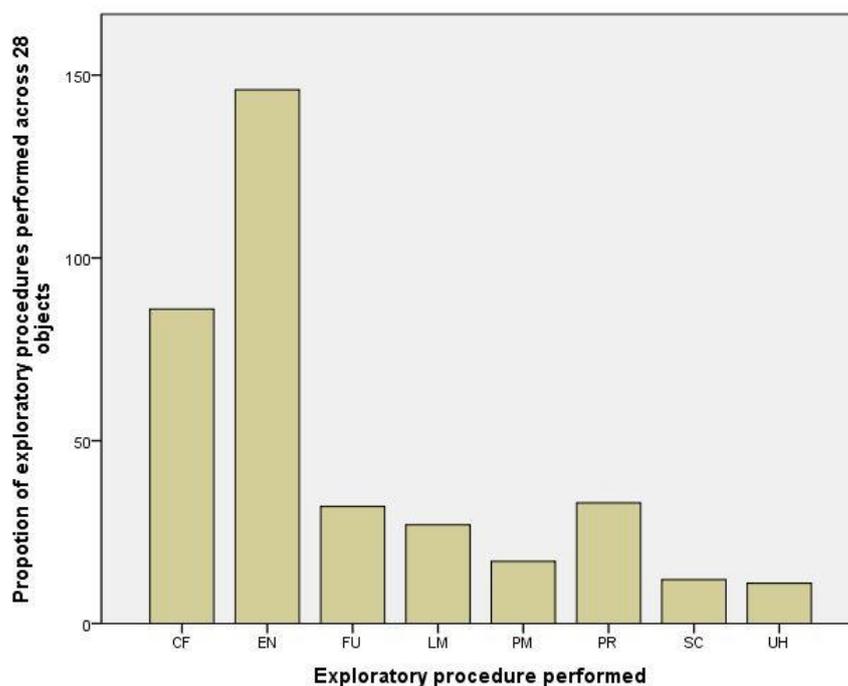


Figure 26. Graphic representation of exploratory procedures performed by 23 children with cerebral palsy for each functional Tactile Object Recognition Test object that was presented for manipulation in this study. CF = contour following; EN = enclosure; FU = function; LM = lateral motion; PM = part motion; PR = pressure, SC = static contact; UH = unsupported holding.

TD children performed all the expected EPs when identifying objects related to texture, temperature, global shape, exact shape, part motion and weight but not consistently for function or hardness. For function (buttons) the most commonly used EP was contour following and for hardness (hardcover book or paperback book) function testing was the most common. On average TD children performed five different EPs per object and even though children performed the expected EPs for six of the seven sensory attributes, the expected EP was not always the most common in the sequence of haptic exploration (Table 10).

Table 10 Haptic exploratory procedures of typically developing children related to the functional Tactile Object Recognition objects

Sensory attribute	Test object	Expected exploratory procedure	Most frequent exploratory procedure
Temperature	Metal doorknob	Static contact	*Contour following
Temperature	Wood doorknob	Static contact	Not tested
Temperature	Steel bowl	Static contact	*Enclosure
Temperature	Plastic bowl	Static contact	*Enclosure

Sensory attribute	Test object	Expected exploratory procedure	Most frequent exploratory procedure
Hardness	Hardcover book	Pressure	*Function
Hardness	Paperback book	Pressure	*Function
Hardness	Firm plastic cup	Pressure	Pressure
Hardness	Crush plastic cup	Pressure	Pressure
Function	Zipper	Part motion	*Contour following
Function	Buttons	Part motion	*Contour following
Function	Click switch	Part motion	Part motion
Function	Turn switch	Part motion	Not tested
Shape	Cylinder pasta	Contour following	Contour following
Shape	Spiral pasta	Contour following	Contour following
Shape	Fork	Contour following	Contour following
Shape	Spoon	Contour following	Contour following
Size	Small watch	Enclosure	Enclosure
Size	Large watch	Enclosure	Enclosure
Size	House key	Enclosure	*Contour following
Size	Filing cabinet key	Enclosure	*Contour following
Weight	Empty jar	Unsupported holding	*Enclosure
Weight	Full jar	Unsupported holding	*Enclosure
Weight	½ full milk bottle	Unsupported holding	*Contour following
Weight	Full milk bottle	Unsupported holding	*Contour following
Texture	Paper card	Lateral motion	Lateral motion
Texture	Plastic card	Lateral motion	Not tested
Texture	Plastic peg	Lateral motion	*Contour following
Texture	Wooden peg	Lateral motion	*Contour following

*Note.* \* Indicates that the expected exploratory procedure as described in the functional Tactile Object Recognition Test test form was not the most frequently used for the object

The present study also identified 12 EMs that could not strictly be classified as EPs (Figures 27 and 28) and there were five EMs that TD children performed most often. For temperature, hardness and part motion the most frequent EMs observed were tapping the object on a surface, tapping on the object and turning the object over on a surface repeatedly. Tapping the object on a surface and tapping on the object was used for exact shape and weight with the addition of shaking the object to identify weight. For texture (i.e. pegs, paper and plastic cards), the most frequent EM was tapping the object on the surface of a table. The EMs of lifting with enclosure, lifting and intentional dropping, lifting and tilting and unsupported holding and tilting are likely subsets of the unsupported holding EP originally described by Lederman and Klatzky (1987). In this study, they are listed separately to demonstrate the varied exploration children used to determine not only weight by

dropping the objects but volume of the objects by tilting and gaining information from rotational inertia.

Figure 27 Exploratory movements performed by typically developing children

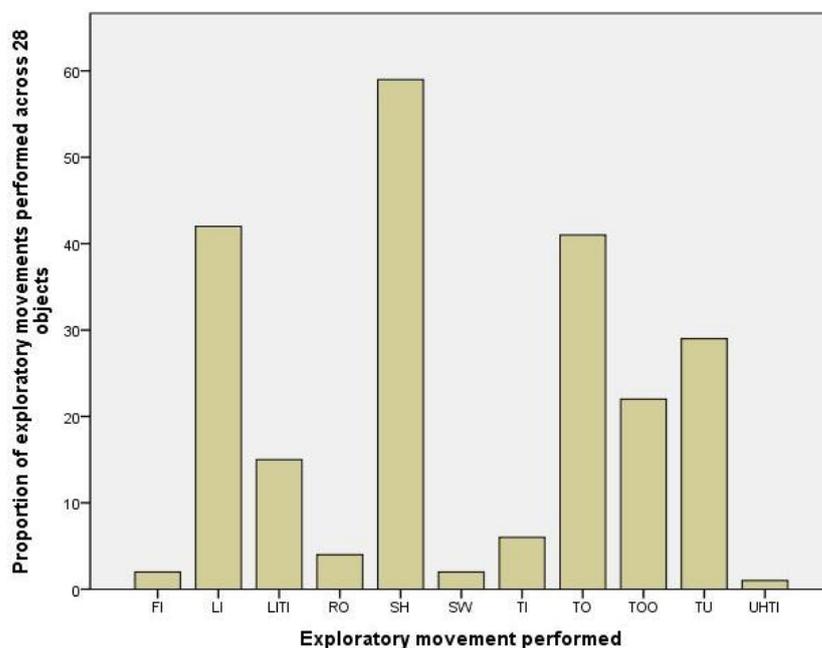


Figure 27. Graphic representation of exploratory movements performed by 31 children without cerebral palsy for each functional Tactile Object Recognition Test object that was presented for manipulation in this study. FL = flicking; LI = lifting with enclosure; LITI = lifting and tilting; RO = rolling; SH = shaking; SW = swiping; TI = tipping; TO = tapping on object; TOO = tapping object on surface; TU = turning object over; UHTI = unsupported holding and tilting.

Figure 28 Exploratory movements performed by children with cerebral palsy

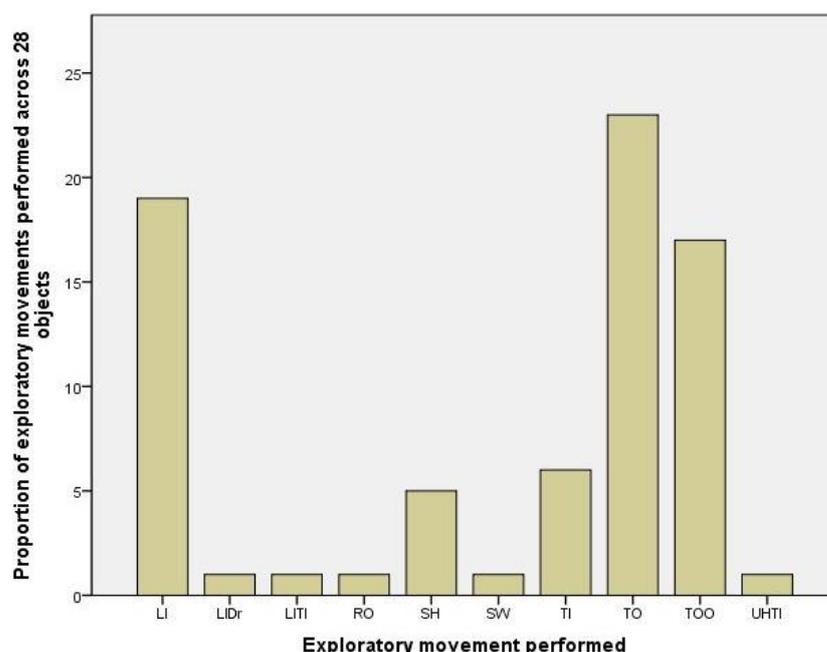


Figure 28. Graphic representation of exploratory movements performed by 23 children with cerebral palsy for each functional Tactile Object Recognition Test object that was presented for manipulation in this study. LI = lifting with enclosure; LIDr = Lifting and intentional dropping; LITI = lifting and tilting; RO = rolling; SH = shaking; SW = swiping; TI = tipping; TO = tapping on object; TOO = tapping object on surface; UHTI = unsupported holding and tilting.

### Comparison between children with and without cerebral palsy

TD children and children with CP performed similar haptic EPs and EMs but at different proportions for each fTORT object presented. There was no statistically significant difference between groups for number of expected EPs ( $p = .146$ ), number of additional EPs ( $p = .778$ ), or number of EMs ( $p = .687$ ) per object however differences in mean time ( $p < .001$ ) and accuracy ( $p < .001$ ) were significant. This suggests that although children with CP performed similar haptic EPs for each object as TD children, the children with CP took more time and were less accurate.

Even though between group analysis revealed no significant difference for proportion of expected EP, additional EPs or EMs, the children with CP consistently had a lower mean for each variable (Table 11). Greater standard error and variability in results for all variables also indicated that their overall performance on the fTORT, and execution of hand movements were impaired when compared to TD children

Table 11 Means and standard errors for each variable based on group

Variable	TD (N = 31)		CP (N = 23)		P value
	Mean	Std. Error (95% CI)	Mean	Std. Error (95% CI)	
Mean number of expected exploratory procedures per object	0.45	0.014 (.42-.48)	0.40	0.035 (.33-.47)	.146
Mean number of additional exploratory procedures per object	1.04	0.024 (.99-1.08)	1.01	0.073 (.88-1.17)	.778
Mean number of exploratory movements per object	0.27	0.032 (.21-.34)	0.29	0.046 (.21-.40)	.687
Time taken to explore each object in seconds	8.60	0.502 (7.63-9.6)	17.40	1.520 (14.4-20.4)	<.001*
Mean accuracy per object	0.84	0.018 (.80-.87)	0.61	0.057 (.50-.72)	<.001*

Note. \*Indicates statistical significance. TD = typically developing; CP = cerebral palsy; Std = standard; CI = confidence interval.

The same GEEs were used to examine each variable for the TD group however, the older age group included a greater proportion of females so age, gender and hand dominance were added to the model and an adjusted analysis was performed. The dominant hand was significantly better at selecting optimal EPs ( $p < .001$ ) and the older age group (9 - 15 yrs) was significantly more accurate than the younger age group (6 - 8 yrs) ( $p < .001$ ). Females took slightly longer to explore objects (mean 8.75 seconds) than males (mean 8.53 seconds) but were more accurate; although the difference in these results did not reach statistical significance ( $p = .80$  for time taken;  $p = .098$  for accuracy).

The same analysis was performed for children with CP adding a variable for MACS level instead of hand dominance and another for haptic object recognition deficit compared with no deficit as measured by the fTORT (e.g. a score  $>36$  for the right hand indicating intact haptic object recognition ability for 6 to 8 year olds). Children with MACS level II performed fewer expected EPs ( $p = <.0001$ ) and took more time to explore objects ( $p = .004$ ) than children with MACS level I however there were no statistically significant differences in accuracy ( $p = .732$ ). Children with MACS level II used fewer additional EPs and fewer EMs consistent with less overall movement. When comparing children with and without haptic object recognition deficits identified by the fTORT, statistically significant differences were observed for accuracy only ( $p = .017$ ) as is to be expected. Children classified as

having a haptic object recognition deficit took more time but this was of borderline statistical significance ( $p = .055$ ). Following the results of the initial GEEs a more detailed between groups analysis was performed comparing the dominant and non-dominant hand of TD children with the affected hand of children with CP for the same variables. There was a significant difference between the dominant hand of TD children and the affected hand of children with CP, but no statistically significant differences were seen for the non-dominant hand. This indicates that the performance of the affected hand of children with CP and the non-dominant hand of TD children were comparable when tested with the fTORT.

## **Discussion**

This study provides preliminary evidence that children with CP can and do perform some of the same optimal haptic EPs as previously published for TD children and adults (Lederman & Klatzky, 1987; 1993; Withagen et al., 2013). In the current study children with CP performed similar expected haptic EPs and additional haptic EPs as their TD peers. The observed similarities were surprising, especially the choice of optimal EPs, given that children with CP often experience significant deficits in haptic object recognition (Auld, Boyd, Moseley, Ware, & Johnston, 2012a). Despite the similarities in hand movements, children with CP took longer to explore each object and were less accurate in object identification than TD children. The same was true when children with CP with and without a somatosensory deficit were compared; children with a haptic object recognition deficit took more time and were less accurate. These findings indicate that somatosensation plays a pivotal role in haptic object recognition despite the development and presence of motor patterns to perform the optimal haptic EPs. The mutually dependent relationship between motor and sensory function is well known (Russo, 2011; Latash & Lestienne, 2006). Ecological frameworks describe exploratory activity as more than a mere motor process that is supported by sensory input from an external layout (Gibson, 2000). However, the similarity in EPs between CP and TD groups is surprising given the often restricted information available through their somatosensory system (Cooper et al., 1995).

MACS level significantly impacted selection of optimal EPs and the time taken to explore objects. Children with MACS level II used fewer additional EPs and

fewer EMs and were slower at identifying objects. However, unlike TD children this finding does not indicate an increased use of optimal EPs but rather reduced patterns of movement altogether due to decreased hand function. These results are not surprising considering that MACS level I is defined as ‘Objects handled easily and successfully’ and MACS Level II as ‘Handles most objects but with some reduced quality and/or speed’ (Eliasson et al., 2006). Children with MACS level I had slightly better accuracy than children with MACS level II although this did not reach statistical significance. This again indicates that a greater range of motor abilities was not the primary determinant of accuracy and that somatosensation is essential to haptic object recognition.

In our study, the types of haptic EPs chosen by children with and without CP were similar to adults however all children also performed extra hand movements not classifiable as EPs. This finding supports the recent work of Withagen et al. (2013) who found that children with and without a visual impairment also performed additional hand movements when identifying the sensory attributes of weight, size, exact shape and texture. Withagen et al. (2013) described the purpose of ‘actions’ as gathering extra information about an object. In their study, they found that most actions occurred during the tasks of exact shape and weight for blind and sighted children. In our study, the most EMs occurred during exploration of weight for children with and without CP.

These additional movements may suggest object recognition difficulties or the need for greater confirmation related to specific sensory attributes. The additional movements may provide extra information which children may need more than adults who are more familiar with a range of sensory attributes of everyday objects. For example, tapping on an object and tapping the object on a surface may be a primitive exploratory behaviour to test for hardness as is often seen in infants (Klatzky, 2005). Instead of using pressure infants perform the exploratory behaviour of banging for hard objects more often than for soft ones (Klatzky, 2005). Geometrically novel objects (sensory attribute of global shape) may elicit turning the object over and over to compensate for underdeveloped skills in contour following. With relation to other additional EPs, Lederman and Klatzky (1990) reported that a highly generalised enclose and lift sequence observed in their study extracted low

level gross information about common (test) objects. We also identified a primitive behaviour for extracting information about weight. Shaking was the most frequently used EM for TD children and lifting with enclosure was the most frequent for children with CP.

The optimal EP for weight, unsupported holding, was at a very low proportion for children with and without CP even for the small and light empty jar. Exact weight may be difficult to determine for children with and without CP aged 6 to 15 years who therefore use an increased amount of dynamic touch and haptic exploration. For children with CP their common EM of lifting with enclosure may be equivalent to shaking for TD children but due to minor motor impairment and reduced strength vigorous shaking may have been too difficult. Riley, Wagman, Santana, Carello, and Turvey (2002) highlighted that the more difficult the detection and discrimination of an object the more pronounced exploratory behaviours become. From a developmental perspective, somatosensation increases with age so it is possible that children need more input supplied to proprioceptors via shaking and lifting to detect weight than adults (Jansen, Tiest, & Kappers, 2015; Taylor et al., 2016).

For TD children the dominant hand was significantly better at selecting optimal EPs than the non-dominant hand and the older age group (9 - 15 yrs) was significantly more accurate than the younger age group (6 - 8 yrs). Interestingly, young children with CP (6 - 8 yrs) were slightly more accurate than older children (9 - 15 yrs). The clinical significance of this finding is uncertain but we speculate that due to the highly heterogeneous nature of neurological impairment, severity of somatosensory deficit may also vary despite age, gender or area of hemiparesis.

There was a significant difference between the dominant hand of TD children and the affected hand of children with CP that was not apparent when all data were grouped together in the initial analysis. This finding is important because studies often compare the impaired hand of children with CP to the non-dominant hand of TD children (Jaspers et al., 2011). The present data provides an indication that the affected hand of children with CP and the non-dominant hand of TD children are comparable. This finding offers further evidence to support an appropriate method of comparison between clinical and non-clinical groups for future research studies examining hand function.

This study demonstrated the importance of observing how children perform tasks, not just whether they complete the tasks (Pehoski, Henderson, & Tickle-Degnen, 1997). Even though our sample of children with CP performed the expected hand movements required to identify each fTORT object they couldn't consistently identify them. This information will be important to consider when planning rehabilitation related to selection of optimal haptic EPs, and deficits in haptic object recognition. It is becoming increasingly evident that the selection of haptic EPs helps to increase haptic perception. Morrongiello, Humphrey, Timney, Choi, & Rocca (1994) found that the haptic EPs used by children who were blind influenced the quality of haptic perception; they described the systematic way blind children explored objects to be positively correlated with their haptic object recognition scores.

Somatosensory interventions have the potential to enhance the mechanisms for extracting somatosensory information and therefore influence the quality of haptic perception (McLinden, 2012). Perceptually based somatosensory interventions such as, sense© training can provide haptic learning experiences that are well structured, graded appropriately and progressive (Carey, Macdonell, & Matyas, 2011). There is preliminary evidence to suggest that sense© improves haptic object recognition in children with CP aged 6 to 15 years (McLean et al., 2017a). Based on principles of perceptual learning and learning-dependent neural plasticity sense© is designed to improve three aspects of somatosensory discrimination: haptic object recognition, tactile discrimination, and limb position sense (Carey et al., 2011; McLean et al., 2017a). Haptic object recognition is trained using graded object sets that reflect each of the seven sensory attributes that were originally described by Lederman and Klatzky (1987) and were also examined in the current study.

What the current paper can add to sense© is the opportunity for a more in-depth examination of exploratory behaviours using the data from TD children and children with CP. During sense© training if the active exploration performed by a child is the optimal EP for the sensory attribute the child may progress to a more complex level. If additional methods of exploration or more time is required (using additional EPs, EMs or exceeding a reasonable amount of time of exploration; 16 seconds  $\pm$  13 seconds), more graded perceptual training can be provided. From the current data, we know that if a child is tapping on an object or tapping the object on a

surface, that has a sensory attribute of hardness, they require focused training for the EP of pressure. If a child is shaking an object in an attempt to identify weight then this would require training in unsupported holding, and turning the object over indicates a need for training in contour following. The data provided in the current paper can be used as performance guidelines for haptic EPs for children aged 6 to 15 years. Ultimately sense© aims to improve somatosensory discrimination capacity for children with CP but fine tuning the methods to achieve object recognition will give children the best chance for rehabilitation and improved haptic object recognition.

The normative data from this study have been included in the fTORT test administration manual for children (Carey et al., 2017). Knowledge of typical function can be used at the initial assessment stage to identify less optimal exploratory behaviour in children with CP and direct goals for sense© training. This study also provides a standardised video protocol (Appendix E) that is used to record and evaluate hand movements performed during the fTORT. Using video to record this extra data and two reviewers to code the hand movements adds a reliable and multi-dimensional method to measure EPs, EMs and time taken to identify each object.

## **Limitations**

Due to the test forms chosen in the administration phase of this study we did not provide the wooden doorknob, turn light switch or plastic card for manipulation therefore, we do not have data for these objects. There was also no object that represented function testing only part motion therefore we don't know if children would have selected function testing for objects that had function testing as their expected EP. Children performed free unilateral exploration and were not provided with explicit or implicit perceptual goals. It is unclear if more perceptually driven goals would have reduced the occurrence of additional EPs and EMs. It should be known that despite the absence of perceptual goals and the inclusion of novel and complex objects in the test procedure, children from both groups still spontaneously performed all eight of the haptic EPs previously published for adults (Lederman and Klatzky, 1987). Although we achieved substantial rater agreement, video coding is inherently subjective (Klatzky, Lederman, & Balakrishnan, 1991). Electronic sensors designed to record human manipulative behaviour from a robotic standpoint may provide superior accuracy for measuring EPs during haptic tasks due to their ability

to be more sensitive to small movements (Klatzky et al., 1991). However, until further development and commercial availability clinicians can rely on the standardised methods described in this paper.

## **Conclusion**

Understanding the haptic EPs of TD children is fundamental when developing an evidence based haptic somatosensory intervention. This paper provides further understanding of TD EPs as well as describing the EPs of children with CP. With this new knowledge clinicians are able to tailor haptic interventions for children.

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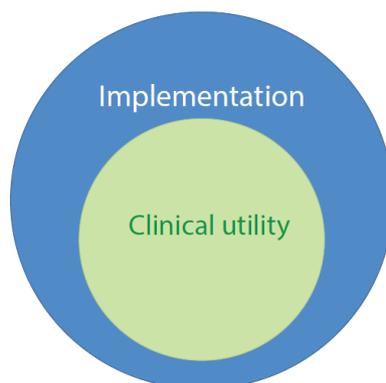
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## CHAPTER 8 Decision Tree for Tool Development



Phase four – Foundations for implementation

### 8.1 Introduction to chapter

The decision tree for tool development outlined in this chapter corresponds to Phase four, Foundations for implementation. This chapter was written as a comprehensive reflection on methodologies used throughout the honours work associated with this PhD and also decisions made throughout this doctoral research. This chapter aimed to outline the thought process made over five years of tool development so successful research methodologies could be expanded and the repeat of unsuccessful endeavors could be prevented. The limitation of this chapter is that methods and decisions often seem disconnected to the previous chapters, this is because background decisions around methodology prior to execution of research are not featured in scientific reporting. The purpose of this chapter is to state that development of the *sense\_assess© kids* has been directed by empirical evidence and such frameworks as the Evidence Based Tool Development framework underpinning this thesis. Using this evidence based approach minimised bias in the evaluation of the tool's suitability for use with children with cerebral palsy (CP). The current chapter outlines the decision tree developed from the findings of this thesis and describes the tool development that is still required before implementation of the *sense\_assess© kids* as a clinical outcome measure. A decision tree in health research can help examine all possible options when faced with a difficult decision such as choosing the best pathway of tool development when a subtest has not performed as expected (Weinstein et al., 2003). Each square of the decision tree was informed by the

research findings of Chapter three Adaptation, and Appendix A, a reliability study involving typically developing children.

Figure 29 Decision tree for tool development

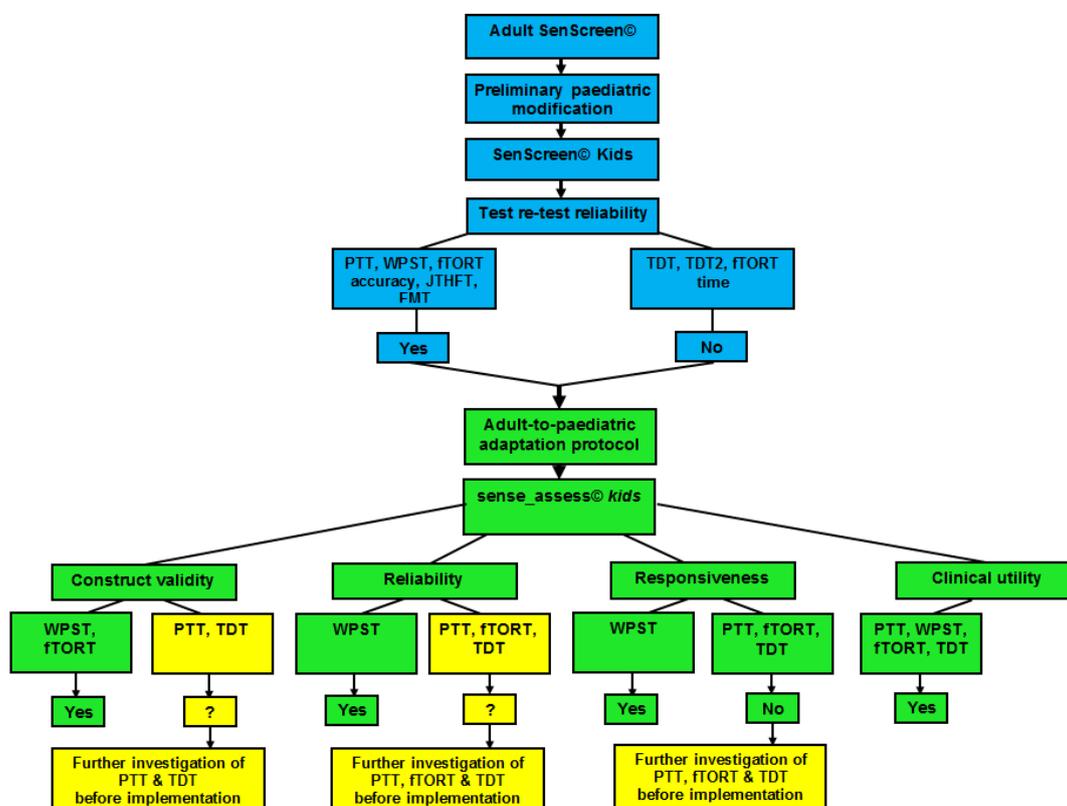


Figure 29. Graphic representing the decisions made before and during the current thesis: blue represents research undertaken as part of the PhD candidate’s honours research; green indicates what has been achieved as part of the current thesis; yellow represents further research required. PTT = Protective Touch Test; WPST = Wrist Position Sense Test; fTORT = functional Tactile Object Recognition Test; JTHFT = Jebsen Taylor Hand Function Test; FMT = Fabric Matching Test; TDT = Tactile Discrimination Test.

## 8.2 Description of decisions made for tool development of the sense\_assess© kids

The SenScreen© Kids is a screening tool containing brief versions of all subtests of the sense\_assess© kids. The blue squares in Figure 29 represent the preliminary paediatric modification, and reliability testing within a typical population of children aged 6 to 15 years for the SenScreen© Kids (Appendix A). This research was undertaken as an honours project completed by the PhD candidate prior to the current doctoral studies and prior to conceptualisation of the sense\_assess© kids. The following information in Table 12 is from Appendix A and refers to the brief subtests of the SenScreen© Kids: the Protective Touch Test (PTT) demonstrated high

interobserver agreement of 99% - 100%; the reliability of both the Wrist Position Sense Test (WPST) and the accuracy component of the functional Tactile Object Recognition Test (fTORT) demonstrated good intrarater agreement between two time-points using Bland Altman plots (Bland & Altman, 2010); the timed component of the fTORT and the Tactile Discrimination Test (TDT) demonstrated poor agreement between the two time-points.

Recording the time with a stopwatch and entering the seconds on the test form for each object of the fTORT also lengthened the test procedure which may have had a negative impact on clinical utility for the tool. Time also seemed to be impacted by motor capacity, potentially confounding its value as a score reflecting somatosensory contributions to haptic object recognition. Therefore, although the timed and accuracy components of the fTORT demonstrated good intrarater agreement within a typically developing (TD) population, fTORT accuracy was retained as the core measure of haptic object recognition.

Bland Altman plots also showed good intrarater agreement for the Jebsen Taylor Hand Function Test (JTHFT) (Table 12), as had previous literature for children (Reedman, Beagley, Sakzewski, & Boyd, 2016), however it is a measure of hand function not somatosensation therefore it was removed. Removal of the JTHFT also reduced administration time of the SecScreen© Kids test battery.

Table 12 Test re-test scores for the SenScreen© Kids subtests

Subtest Sample N=22	TDT Accuracy	fTORT Accuracy	fTORT Time	WPST Degrees	JTHFT Time
Mean difference RH	2.41	0.41	-0.14	-1.59	-0.87
LOA	-83.65 - 88.48	-1.90 - 2.72	-0.99 - 0.70	-12.98 - 9.81	4.65 - 2.91
95% CI	-9.35 - 14.17	-0.11 - 0.93	-0.33 - 0.05	-4.23 - 1.06	-1.73 - -0.01
Mean difference LH	2.80	0.73	-0.11	-3.02	-1.39
LOA	-78.90 - 84.50	-1.70 - 3.16	-0.77 - 0.55	-15.87 - 9.82	-4.78 - 1.99
95% CI	-8.37 - 13.96	0.18 - 1.28	-0.26 - 0.04	-5.93 to -0.12	-2.16 - -0.63

*Note.* TDT = Tactile Discrimination Test; fTORT = functional Tactile Object Recognition Test; WPST = Wrist Position Sense Test; JTHFT = Jebsen Taylor Hand Function Test; RH = right hand; LH = left hand; LOA = Limits of agreement; CI = confidence interval.

The green squares represent research completed as part of the current thesis. Adaptation involved extension of the test forms from the brief SenScreen© version to the more discriminative and comprehensive sense\_assess© test protocol. The adult-to-

paediatric adaptation protocol was applied and the adapted PTT, TDT, fTORT and WPST demonstrated acceptability to a population of children with CP aged 6 to 15 years (Chapter four). Minor modifications were made to the TDT based on this study also described in Chapter four (e.g., less repeated exposure to textures and more rest breaks).

Initially, in a previous investigation by the PhD candidate, the brief version of the TDT (a subtest of the SenScreen© Kids) demonstrated a weak relationship between two time-points (Intraclass Correlation Coefficient - ICC .425), indicating poor intrarater reliability within a TD population ( $n = 44$ ). Early preliminary data also showed children with CP ( $n = 8$ ) were achieving scores no better than chance for any of the textures across two time-points. It appeared that the children with CP were guessing their responses and it was suspected that either the textures were too similar for children with diminished sensation to identify their differences or that the test instructions were not understood. Therefore, changes were made to the TDT test protocol.

A simpler TDT protocol (TDT2) was developed still using the same textures of the TDT; 170 (smoothest), 210, 260 and 300 (roughest) but employing a two-alternative forced choice design where the children were asked to choose the 'rougher' texture out of two different textures. Previously in the original TDT version, children were asked to choose the 'different' texture out of three textures. For the TDT2 the reliability between time-points depended heavily on the texture being used. To examine reliability of the new TDT2 version a General Estimating Equation (GEE) model was used to assess the dependence of accuracy on the independent variable of time-point one and time-point two for 31 TD children. This model adjusted for the four different texture sets and accounted for the repeated attempts made by each child. The accuracy between time-points depended on the texture being used, scores for the 210 texture improved between time-points ( $p > .01$ ) indicating poor reliability or a learning effect, but scores were reliable for the 170 ( $p = .27$ ), 260 ( $p = .18$ ) and 300 ( $p = .67$ ) textures.

Because the TDT2 did not perform as well as expected, the extended version of the TDT (subtest of the sense\_assess© kids) was examined for intrarater reliability with 22 TD children aged 6 to 15 years (Appendix A). The TDT

demonstrated good intrarater agreement between two time-points for all textures using Bland Altman plots (Bland & Altman, 2010). The Fabric Matching Test (FMT) was examined at this time as an alternative measure of tactile discrimination while the TDT and TDT2 were under further investigation. The FMT was subjected to the same process as the *sense\_assess© kids* as part of this thesis to adapt it from an adult to paediatric version and a full description of the test and its psychometric properties is included in Appendix D and Appendix E. The FMT can be used in place of the TDT as part of the *sense\_assess© kids* until conclusive evidence supports the use or disuse of the TDT. It is important to note that the FMT only has psychometric evidence for a clinical population of adults post-stroke and TD children aged 6 to 15 years. Further psychometric testing continues for children with CP.

During this phase of testing the TDT2 and FMT more data were collected for children with spastic hemiplegic CP (n = 18) for the original TDT protocol and were examined using a GEE model. The success rate was significantly lower for the more difficult textures (170 and 210) than the easier textures (260 and 300) (Table 13). This performance pattern was similar to that seen in the TD population and provided hope for use of the original TDT version in future research. Further investigation needs to continue for the 210 texture, across all populations tested (CP and TD). The 210 texture appears to be the least reliable out of the four textures, but its discriminative properties may be valuable in terms of test sensitivity once properly understood. The new data suggested that the difficulty of the TDT may support its discriminative ability for small differences in tactile discrimination ability and it is recommended that the reliability of the TDT is tested within a larger sample of children with CP aged 6 to 15 years.

Table 13 General Estimating Equation examining patterns of tactile discrimination at one time-point for children with cerebral palsy

Texture	Successes	Odds ratio	95% CI	p-value
260 or 300	90/150 (60.0%)	1 (ref)		
210	25/75 (33.3%)	0.33	0.15 to 0.75	.0077
170	27/75 (36.%)	0.38	0.16 to 0.87	.0224

Note. CI = confidence interval

As part of this thesis a study was designed to examine the haptic exploratory procedures of TD children and children with CP during the fTORT (Chapter seven).

This study established statistically significant differences between groups for time and accuracy during the fTORT, indicating that children with CP took more time to explore objects and were less accurate than their TD peers. At this stage, time cannot be separated from motor capacity therefore, until further investigation recording of time required to explore objects should remain as part of the fTORT test procedure as per the original design. This study also indicated that with further tool development the fTORT can be used to describe haptic EPs for children with CP for comparison with typical performance standards collected as part of this thesis.

Based on the decision tree for tool development the current sense\_assess© *kids* test battery is as follows: Protective Touch Test, Tactile Discrimination Test, functional Tactile Object Recognition Test (accuracy and time) and Wrist Position Sense Test. Also included in the sense\_assess© *kids* Administration manual as appendices are the supplementary FMT, fTORT video protocol and JTHFT. The current format of the sense\_assess© *kids* test battery is suitable for use in research and further investigation is required before implementation of the full battery into clinical practice. The yellow squares of Figure 29 represent the subtests that require further tool development and a plan for this is explained in detail in Chapter ten.

Analysis completed as part of the decision tree phase of this thesis helped establish preliminary guidelines for three different levels of difficulty for the test stimuli of the TDT, fTORT and WPST (Table 14). The discriminative levels were based on an analysis of GEE parameter estimates to calculate standard error across duration of the test and test stimuli for the fTORT and TDT, and a random effects regression model was used for the WPST. The levels of difficulty are intended to be used as follows: Prior to knowing an individual's somatosensory ability, children are tested with the easiest level first (Level 1) and if they cannot complete this level then it is unlikely they will be able to detect the test stimulus in subsequent levels (Levels 2 and 3). The levels of difficulty aim to reduce the burden of assessment for the more impaired however, the estimates have not yet been tested. It is recommended that they are included in future clinical research using the sense\_assess© *kids* to test their validity and reliability.

Table 14 Levels of difficulty for the sense\_assess© *kids*

Test	Level 1 (Easy)	Level 2 (Moderate)	Level 3 (Difficult)
TDT	Texture 300	Texture 260	Texture 170
fTORT	Click switch	Firm plastic cup	Cylinder pasta
	Crush plastic cup	Half full milk bottle	Empty jar
	Metal doorknob	Hard cover book	House key
	Spoon	Plastic card	Small faced watch
	Stainless steel bowl	Paper card	
	Fork	Wooden clothes peg	
WPST	Close to 90°	Extension	Flexion
Left hand	Between 70° - 110°	Between 30° - 69°	Between 151° - 180°
Right hand	Between 70° - 110°	Between 111° - 150°	Between 0° - 29°

*Note.* TDT = Tactile Discrimination Test; fTORT = functional Tactile Object Recognition Test; WPST = Wrist Position Sense Test.

These guidelines have been included as an appendix in the sense\_assess © *kids* Administration manual (Appendix E) along with the FMT and JTHFT administration procedure and fTORT video protocol. Because the brief version of the PTT, TDT, fTORT and WPST demonstrated good agreement using Bland Altman plots it is expected that the extended test protocols will as well. It is recommended that further reliability and validity testing occurs for all measures within a clinical population (see Chapter 10).



## CHAPTER 9 General Discussion

Between 46% to 97% of children with cerebral palsy (CP) experience an upper limb somatosensory deficit (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; McLean et al., 2017b; Van Heest, House, & Putnam, 1993). There is currently no comprehensive and standardised assessment to describe or evaluate somatosensory deficits or predict the functional impact of somatosensory impairment for these children (Auld, Ware, Boyd, Moseley, & Johnston, 2012b; Connell & Tyson, 2012). Children with CP face additional challenges due to comorbid conditions and compromised motor function (Bax et al., 2005), therefore a somatosensory measure must be sensitive to the specific needs of this population (Auld et al., 2012b). This thesis satisfied a scientific curiosity to adapt and test a battery of somatosensory tools deemed the most suitable for paediatric use. This thesis has provided a framework for application in future research and acknowledges that other tools might also be useful to adapt or modify. Tools identified in Chapter one of this thesis that may benefit from further investigation are as follows: the Grating Orientation Task (Johnson & Phillips, 1981) for touch perception; computerised measures of proprioception and limb position sense (Chrysagis, Skordilis, Koutsouki, & Evans, 2007; Scott, 1999); and the Hand Active Sensation Test (Williams, Basso, Case-Smith, & Nichols-Larsen, 2006) for haptic perception. Adaptation, modification or further research with larger cohorts would be useful to strengthen sensitivity, clinical utility and paediatric acceptability of these measures. The battery investigated by Auld et al. (2011; 2012b) and colleagues showed promise however the issues of ceiling effects for the Klingels method (Klingels et al., 2010), learning effects and poor reliability for the AsTex® (Miller et al., 2009), and overall responsiveness of the battery requires remediation. Removal of these tools from the battery leaves only tactile registration and spatial perception measures. These measures examine extremely important domains, however as a comprehensive measure fitting the scope of inquiry for this thesis to address the shortfalls of other measures, they were not preferable to adapting the sense\_assess©. Having moved through the stages of tool development outlined in this thesis the following is a synthesis of the findings with relation to current literature.

## **9.1 Phase one – Knowledge inquiry and synthesis**

Phase one searched for currently available somatosensory measures for adults and children. The scoping review (Chapter two) identified that paediatric specific assessments were limited but there was scope to adapt an adult somatosensory assessment with published psychometric data, for paediatric use. Comprehensive review of literature prior to developing or adapting an assessment tool is a well-recognised strategy (Randall, Imms, & Carey, 2008). The scoping review identified the following six common limitations associated with somatosensory measures regardless of whether they were designed for adults or children: a lack of standard procedures; designed for research purposes; limited evidence of clinical utility; lack the ability to measure somatosensory constructs comprehensively; or functional hand sensibility; a lack of normative data, or evidence concerning psychometric properties for use with children.

Inconsistencies in tool design and test procedures can jeopardise the accurate assessment of a child's impairment level or function. Inconsistencies can also limit comparison of research findings due to variability in data collection methods (Dijkers, Murphy, & Krellman, 2012; Taylor et al., 2016). In healthcare, there is a need for rigorous research methodology for tool development studies to avoid similar issues and variability in outcome measurement results (Bourke-Taylor, 2014). It is recommended that the six issues above be considered alongside frameworks such as the Outcome Measures Rating Form (OMRF) (CanChild, 2004) and the COnsensus-based Standards for the selection of health status Measurement INstruments (COSMIN) (Mokkink et al., 2010) at the planning stage of tool design or tool adaptation as it is difficult to address issues after psychometric testing has already commenced.

The strategic knowledge partnerships created as part of Phase one helped to established the foundations for implementation of clinical somatosensory assessment and management in the future. Knowledge partnerships with clinicians, researchers, organisations and academics enhanced the tool development process. Initial partnership with the original author of the sense\_assess© granted full access to equipment, assessment administration training, design protocols and exchange of intellectual property. Collaboration through universities involved an honours

program that produced a publication about therapists' current perspectives of somatosensory measurement (Appendix C), and a parallel PhD examining the feasibility of a somatosensory intervention (Appendix F, G, H and I). This ensured that the most advanced research and knowledge for somatosensory assessment and management was utilised to support the research within the current thesis.

Research partnerships with the Paediatric Rehabilitation Department at the state children's hospital, and the state-wide Ability Centre for people living with a disability in Perth, Western Australia allowed access to suitable research participants and forged implementation pathways for the future. Clinicians from both organisations were involved in the early testing and evaluation of the *sense\_assess© kids* providing crucial information in the adaptation phase. Involving clinicians and managers from two of the largest rehabilitation centres in Western Australia informs them of advances in assessment and treatment strategies, and provides a clear pathway to implementation of new research into clinical practice. Finally, early collaborative partnerships with consumers such as children and their families provided knowledge about how to modify the assessment to enhance acceptability by those who may be exposed to it in the future. Collectively Phase one provides a summary of the literature related to somatosensory assessment and current challenges, and an outline of how to create knowledge partnerships that can be used to foster rapid uptake of research knowledge into clinical practice. Clinicians and researchers can refer to this new knowledge from Phase one prior to future tool development to enhance accurate diagnosis of children with a somatosensory deficit.

## **9.2 Phase two – Evidence based tool adaptation**

Chapter three produced an adult-to-paediatric adaptation protocol (Figure 4) that was used successfully to modify the adult *sense\_assess©*. The adult-to-paediatric adaptation chapter of the current thesis (Chapter three) extended this process to include details relevant to children with CP, somatosensation, adult to paediatric modification and elements from the OMRF and COSMIN. Employing an evidence based framework early in tool development is crucial to the design of high quality research and the development of a robust assessment tool (Law et al., 2005). Because of its success in the current doctoral research, the adult-to-paediatric adaptation protocol can assist in modifying an assessment for a broader range of age groups or

diagnostic groups, reduce time and cost of developing a new assessment tool and support the clinical benefits of modifying an already validated measure.

### **9.3 Phase three – Psychometric testing based on the COSMIN**

Phase three involved completion of four papers examining the psychometric properties and clinical acceptability of the *sense\_assess© kids* subtests.

#### **9.3.1 Clinical acceptability**

The clinical acceptability study of Chapter four established that the administration procedure and duration, subtest equipment and perceived level of difficulty of the *sense\_assess© kids* were acceptable to the intended population of children and adolescents with spastic hemiplegic CP aged 6 to 15 years. This study demonstrated that the *sense\_assess© kids* is clinically useful and supported further investigation of its psychometric properties. This paper also highlighted the importance of inviting end users to be involved in the initial stages of the tool development process. The gold standard for consumer engagement in research is when consumers identify the research need and set the appropriate research plan themselves (McKenzie, Alpers, Heyworth, Phuong, & Hanley, 2016). When that cannot be done because the research need has already been identified, the alternative is to engage end users as early as possible in the research process (McKenzie & Haines, 2014).

The comprehensive framework of clinical utility provided in Chapter four supports consumer engagement by including evaluation of clinical acceptability. The information provided by the consumers was incorporated into the *senses\_assess© kids* administration procedure and will accelerate the implementation of the tool in the future. This framework can be used by researchers and clinicians in the early phase of tool development to shape clinically useful tools for their targeted population. Asking the children and adolescents' perspectives about the *sense\_assess© kids* has meant that the assessment administration and equipment has been tailored to their needs. Ensuring the comfort and acceptability of an assessment tool to patients is equally important to the accurate assessment of the outcome of interest.

### **9.3.2 Wrist Position Sense Test**

Chapter five provided empirical evidence to support the construct validity and responsiveness of the Wrist Position Sense Test (WPST) for use with children with spastic hemiplegic CP aged 6 to 15 years. The WPST demonstrated a statistically significant association with performance on the Box and Block Test (BBT) and could detect statistically significant differences in wrist positions sense between children with and without CP. The WPST was also responsive to change in the treatment group of an intervention trial involving somatosensory discrimination training. These current findings had not previously been examined in children and can help advance clinical practice guidelines for measurement of wrist position sense as discussed in Chapter ten. This paper also highlights the current limitations in existing measures of proprioception including limb position sense, and informs researchers that objective measures with standardised procedures are paramount when developing new clinical assessment tools (Connell & Tyson, 2012).

### **9.3.3 functional Tactile Object Recognition Test**

Chapter six aimed to provide empirical evidence to support the construct validity and responsiveness of the functional Tactile Object Recognition Test (fTORT) for use with children with spastic hemiplegic CP aged 6 to 15 years. This paper established that the fTORT is a valid measure of haptic object recognition and further research is required to examine responsiveness. The fTORT demonstrated a statistically significant association with performance on the BBT and could detect statistically significant differences in haptic object recognition between children with and without CP. The fTORT was not responsive to change in the treatment group of a matched-pairs trial involving somatosensory discrimination training. This may be due to several reasons and the test requires further investigation in a clinical population. In this study, it may have been observed that the fTORT was not responsive to the intervention, but it is too soon to conclude that the fTORT is not a responsive evaluative tool. For that, alternative explanations need to be negated eg. (i) the intervention did not significantly improve haptic object recognition, (ii) the sample size ( $N = 16$ ) was not large enough to detect small changes in haptic object recognition. These findings provide a robust platform of which to build future research examining the measurement properties of the WPST and fTORT. It is recommended that improvements in haptic object recognition and wrist position

sense be compared with improvements in somatosensory based goals, as assessed by Goal Attainment Scaling (GAS) (Kiresuk, Smith, & Cardillo, 1994) or the Canadian Occupational Performance Measure (COPM) (Law et al., 1998).

### **9.3.4 Haptic exploratory procedures**

Chapter seven established that children with CP performed similar exploratory procedures (EPs) for each fTORT test object as typically developing (TD) children but took more time and were less accurate at identifying objects. These findings add to the body of evidence recognising the vital role of somatosensation for hand function (Gibson, 2000). It is known that somatosensation and motor function are mutually dependent (Russo, 2011), however this study suggested that somatosensation influenced object identification accuracy more than haptic EPs. The descriptive data from children with and without CP has been included in the fTORT section of the *sense\_assess© kids* Administration manual (Appendix E). This provides a new function for the fTORT; to document and describe the hand movements of children and youth with CP, and identify limitations in the selection and use of optimal haptic EPs.

## **9.4 Phase four – Foundations for Implementation**

### **9.4.1 Decision tree for tool development**

Chapter eight provided documentation of the decisions made during adaptation, pilot testing and psychometric testing for the *sense\_assess© kids*. The decision tree for tool development provided an opportunity to record the negative as well as the positive outcomes to guide future research. Throughout tool adaptation the decision tree helped to examine all possible options when faced with a difficult decision such as choosing the best pathway for the *sense\_assess© kids* when the TDT did not perform as expected. This chapter established that the SenScreen© Kids could be successfully adapted into the *sense\_assess© kids* by using the extended test forms but removing the timed component of the fTORT and the Jebsen Taylor Hand Function Test (Jebsen et al., 1969). The *sense\_assess© kids* benefitted from removing these items in terms of clinical acceptability but as further research findings from the thesis demonstrated these items might need to be considered when clinical reasoning judges them to be of importance to rehabilitation outcomes.

This chapter acknowledges the use of core somatosensory measures including the Protective Touch Test , Tactile Discrimination Test (or the Fabric Matching Test until further psychometric testing is complete), functional Tactile Object Recognition Test (with timed component added, and video component of haptic EPs), and the Wrist Position Sense Test. It was concluded in this chapter that the complete *sense\_assess© kids* test battery is not ready for implementation into clinical practice. It is recommended that future psychometric testing occurs as outlined in Chapter ten.

#### **9.4.2 Administration manual**

This thesis also produced the *sense\_assess© kids* Administration manual that expanded on the SenScreen© Kids Administration manual originally adapted as part of the honours program described in the preface of this thesis. Appendix E contains the adapted Administration manual of the *sense\_assess© kids*. The content of the administration manual is in adherence with the OMRF framework (Law, 1987) and includes detailed information about the *sense\_assess© kids* focus of measurement (somatosensation; protective touch, tactile discrimination, haptic object recognition and wrist position sense) purpose (discriminate and evaluate) format (active participation of child), administration set-up (standardised equipment, procedure and instructions), scale construction (nominal, interval and ordinal) and empirical evidence to support the tool's use (normative data, construct validity, responsiveness and clinical acceptability).

There is still limited use of standardised somatosensory outcome measures in clinical practice (Pumpa, Cahill, & Carey, 2015; Taylor et al., 2016; Walmsley et al., 2017). The main reasons for this are a lack of knowledge of how to assess, lack of confidence in conducting the assessment and a lack of access to equipment including test forms, assessment guidelines and test items (Auld & Johnston, 2016). Standardised assessment tools require a comprehensive administration manual and standardised test items. Test items should not be sourced by the therapist performing the assessment because the specific characteristics of the items cannot be standardised across different cohorts (Auld et al., 2011). Qualitative reports have suggested that available departmental assessment guidelines and test forms may

facilitate the implementation of somatosensory assessment in paediatrics (Auld & Johnson, 2016).

Appendix E provides the standardised administration manual for the sense\_assess© *kids*. Developing an accompanying manual to be sold commercially with the sense\_assess© *kids* test kit ensures that therapists are provided with guidelines and standardised equipment and test forms for administration. Amending the administration manual as part of this thesis has also meant that comprehensive psychometric testing can be continued as per the Recommendations in Chapter ten. In the context of a paucity of existing literature on suitable comprehensive measures of somatosensory capacity for children, this four-phased research project successfully met the overall aim of adapting an existing assessment of somatosensation for adults, to one for children (the sense\_assess© *kids*). The sense\_assess© *kids* was evaluated for preliminary psychometric properties, and suitability for use with children with CP was established.

## **9.5 Limitations**

Limitations are discussed within each paper of this thesis however, in this section limitations are discussed in relation to the thesis and overall research design, sample, and individual sense\_assess© *kids* subtests.

### **9.5.1 Research design**

The research design was intended to examine psychometric properties in as much detail as possible however, the individual study designs prevented investigation of all possible confounders. Engagement in the sense\_assess© *kids* testing procedure was potentially influenced by personal and environmental factors such as time of day, attention and arousal level, cognitive functioning, assessment setting and developmental level of the child. In this research, sensory integration and behavioural or emotional responses to somatosensory stimuli that may have been triggered by imposed touch was not addressed. Although the sample of children with CP was heterogenous in terms of laterality of spasticity and associated impairment, the impact of the nature and severity of the brain lesions on somatosensory function was not considered.

## 9.5.2 Sample

The parallel PhD, of which the current thesis relied on for its participants, was not able to recruit suitable matches for each participant for the matched-pair design before cessation of the preliminary investigation. Therefore, this affected the size of the participant sample within the current thesis. For this reason, a post hoc power calculation was completed for the responsiveness studies to determine the optimal sample size to detect responsiveness changes. For the fTORT study in Chapter six G\* Power (Faul, Erdfelder, Buchner, & Lang, 2009) was used to compute power, given that alpha was set to .05, N was 16, and the effect size was 0.9 (calculated from the parallel study, see Appendix H of the thesis). This post-hoc power analysis identified that the fTORT study was adequately powered (0.94) and therefore unlikely to be a contributing factor to the poor responsiveness findings. Using the same method in G\* Power for the WPST study of Chapter five we identified that the study also had sufficient participants and adequate power (0.96) to detect accurate findings with the inferential statistics used.

All studies included in this thesis would have benefitted from a larger sample size particularly for the association studies of Papers two and three ( $n = 17$ ) because it would have allowed for comparison between scores on the WPST and fTORT with the other outcome measures: COPM (Law et al., 1998), GAS (Kiresuk, Smith, & Cardillo, 1994) or the Assisting Hand Assessment (AHA) (Krumlinde-Sundholm et al., 2007). The current doctoral research was closely linked to a parallel PhD and when cessation of recruitment occurred in the parallel research it affected the size of the participant sample within the current thesis. An initial analysis using a chi-square comparing scores from the WPST to the PTT and AHA, and scores from the fTORT to the PTT and AHA revealed a very weak correlation ( $X^2(2, N = 17) = 3.52, p = .14$ ). However, this result is not conclusive because results could vary and may become significant with a larger sample size;  $N > 50$ . The current research only examined the most affected hand of children with spastic hemiplegic CP with Manual Ability Classification System (Eliasson et al., 2006) levels I and II and the findings should only be generalised to other body areas or to other children with different descriptions of CP with great caution.

### 9.5.3 Individual subtests

The PTT is a measure of tactile registration and was designed to be a prerequisite screen prior to completion of the other sense\_ assess© *kids* subtests. Therefore, it was not administered at each of the four time-points of the responsiveness study (Chapter five and six), also it was not an outcome of interest in the intervention study (Appendix G). This prevented comparison of changes in tactile registration associated with sense© training over time. Because of the anomalies that exist in somatosensory deficits for children with hemiplegic CP (McLean et al., 2017b) it is recommended that protective touch become an outcome of interest in future research for the sense\_ assess© *kids* and Sense© for Kids training.

The TDT proved to be the most difficult test to adapt for children with CP. A major shortfall of this research was ceasing data collection for the TDT. At the time, this was an ethical decision, the test was deemed too difficult for children with hemiplegic CP because the responses provided by children and youth ( $n = 8$ ) were distributed randomly. However, analysing data from ten more children with CP ( $n = 18$ ) using a Generalised Estimating Equation revealed expected performance patterns similar to TD children for the different textures. Please see Chapter eight for a full description of the changes made to the TDT. Further testing of the TDT in children with CP is indicated.

One of the major drawbacks of the WPST was the size of the device and time required to set the equipment up. Feedback provided from a therapist at Ability Centre said they would use the test because it provided valuable information but it was a long assessment and time consuming to set up the test equipment. The authors also acknowledge that the construction of the WPST apparatus as illustrated in Figure 11 might provide additional tactile input to the hand within the splint. However, even if there were any such effects, under standard conditions of testing it would remain constant and therefore any systematic changes or differences in the magnitude of errors (over time or between different children) would be valid.

For the fTORT limitations centre around the size of objects and the inclusion of a response poster that requires cross-modal transfer of haptic-to-visual information. Therefore, other skills are required for successful completion of the fTORT not just somatosensory processing and extraneous information in this type of

instrument could lead to decreased responsiveness (Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2012; Wann, 1991; Law, 1987).



## CHAPTER 10 Recommendations

### 10.1 Current use of sense\_assess© kids

The core sense\_assess© kids test battery is recommended in the current thesis to include the Protective Touch Test (PTT), Tactile Discrimination Test (TDT), functional Tactile Object Recognition Test (fTORT) accuracy and Wrist Position Sense Test (WPST) with supplementary assessment using the Fabric Matching Test (FMT) and the timed responses of the fTORT if required. Subtests of the sense\_assess© kids that have not been tested as part of the tool development of this thesis but are still included in the proposed sense\_assess© kids test battery are the Jebsen Taylor Hand Function Test (JTHFT), Hot Cold discrimination and paediatric surveys of hand function and somatosensation. There is no intervention to improve temperature discrimination, and it is not a tactile registration or perception construct (Auld & Johnston, 2016) therefore, it is recommended for use as an evaluation of safety only. The original sense\_assess© manual included questionnaires related to hand function and the ability to use the hand in daily activities, and also awareness of any changes in or loss of somatosensation and its impact on limb function and daily activities. Literature supports the use of questionnaires to evaluate awareness and impact of somatosensory deficits on daily function and there is evidence to support self-report surveys alongside quantitative measurement (Cunningham et al., 1996; Liddle & McKenna, 2000). The addition of paediatric appropriate questionnaires as non-core sense\_assess© kids subtests could be valuable. Objective measures can indicate disablement at the impairment and activity levels of the International classification of functioning, disability and health (ICF) but limitations of participation are primarily subjective (Liddle & McKenna, 2000; World Health Organization, 2001).

Individualised outcome measures such as, Goal Attainment Scaling (GAS) (Kiresuk, Smith, & Cardillo, 1994) or the Canadian Occupational Performance Measure (COPM) (Law et al., 1998) measure outcomes that are most relevant to the client or caregiver (Fleminger & Powell, 1999). Therefore, it is recommended that the sense\_assess© kids subtests PTT and WPST be used, with careful monitoring, and in combination with goal based measures (e.g. COPM, GAS) in the paediatric clinical setting to measure the somatosensory domains of tactile registration and wrist position

sense. It is recommended that the fTORT continue to be used in clinical research to evaluate the somatosensory domains of haptic object recognition and establish further psychometric properties. It is recommended that the TDT and FMT undergo further psychometric development in a clinical population prior to use in clinical research or practice.

The role of somatosensation in behaviour is well understood (Baranek, David, Poe, Stone, & Watson, 2006) however, there is little clinical evidence of its importance at the activity and participation level. Therefore, further research is also recommended to investigate the link between somatosensory impairment and limitations in activity and participation.

## **10.2 Future psychometric testing of the sense\_assess© kids**

It is recommended that testing the measurement properties outlined in the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010) that were not tested as part of this thesis continue for the sense\_assess© kids subtests (highlighted in yellow Figure 29) with a sample of at least 50 participants (Mokkink et al., 2010) with homogenous samples, different groups and clinical presentations. This includes structural and cross-cultural validity for all subtests, inter and intrarater reliability and internal consistency within a clinical population for the TDT, fTORT and WPST. Construct validity and responsiveness testing for the TDT and FMT, and continued evaluation of responsiveness for the fTORT. Criterion-related validity remains difficult to test when there is no gold standard somatosensory measure for comparison. It is intended that the plan for a correlation study described next, comparing scores of the sense\_assess© kids with impairment and activity measures will compensate for this limitation.

Appendix H describes a protocol for a randomised control trial (RCT) that uses the sense\_assess© kids as the primary outcome measure of somatosensation of the upper limb in children with hemiplegic CP (expected sample n = 50). Other outcome measures in the protocol include the COPM (Law et al., 1998), GAS (Kiresuk, Smith, & Cardillo, 1994) and the Assisting Hand Assessment (AHA) (Krumlinde-Sundholm et al., 2007). It is recommended that the correlation between results of the

sense\_assess© *kids* and measures of activity and participation be examined to investigate construct validity.

In addition to this, further scientific inquiry could establish if the SenScreen© Sensory Screening Tool is sufficient compared to the full test battery. Unpublished data in a population of adults post stroke suggests that it shows promise as a sensitive, responsive measure however more data is required before conclusive evidence is presented L. Carey (personal communication, July 30, 2017) . The SenScreen© would have enhanced clinical utility as it includes all sense\_assess© subtests with a shorter administration time.

### **10.3 Clinical practice guidelines**

The scoping review of Chapter two can be used to design a Delphi survey involving experts in the field, with the aim of developing consensus around the standards that should be met for somatosensory assessment in children with disabilities (Portney & Watkins, 2009). As a preliminary step, an outline of potential clinical practice guidelines for somatosensory measurement has commenced as part of the research findings provided through this thesis.

Clinical practice guidelines for the assessment of children with a neurological disorder aged 6 to 15 years with a suspected somatosensory deficit are proposed as follows:

- Multiple domains of somatosensation are assessed by a comprehensive battery because deficits occur in combination across somatosensory domains, with children often experiencing impairment in three or more somatosensory domains (McLean et al., 2017b).
- Both the least affected and most affected hand should be assessed. While the less affected hand of patients with hemiplegia does not usually demonstrate severe sensorimotor impairments, it is common for tactile deficits to extend to the “unimpaired” hand as well (Auld et al., 2012a; Verrel, Bekkering, & Steenbergen, 2008).
- Somatosensory assessments require standardisation to ensure accurate comparison between different groups (Cooper et al., 1995 and Appendix B).

- Somatosensory assessments are more accurate when interesting and engaging for young children (Poulsen et al., 2013 and Chapter six).
- Somatosensory assessments require sensitivity to the characteristics of children with neurological impairment: e.g. reducing the cognitive demands of the test, rest breaks incorporated into the test procedure and postural and positioning considerations (Chapter five and six).
- Somatosensory capacity should be screened on admission or assessed when it is clinically indicated (Walmsley et al., 2017).
- Somatosensory assessment should occur throughout treatment, and in the discharge planning phase (Walmsley et al., 2017).
- Those who can administer a somatosensory assessment need to hold a degree or license to practice in the healthcare or allied healthcare field and complete formal training in the ethical administration, scoring, and interpretation of the assessment (Pearson Education Inc, 2017).

### **10.3.1 Recommended somatosensory measures.**

Best practice somatosensory measures recommended by Auld et al., 2011 include:

- Semmes-Weinstein monofilaments (SWM) (Weinstein, 1993).
- Moving and static two-point discrimination (MacKinnon & Dellon, 1985).
- Single-point localization using the largest SWM (Burns, Ensby, & Norrie, 1989).

Best practice somatosensory measures as recommended in this thesis include:

- Wrist Position Sense Test (WPST) (Carey et al., 1996).
- Protective Touch Test (Taylor et al., 2017f; Touch-Test® Sensory Evaluators, 2001).

### **10.3.2 Concurrent functional measures to be used in future research**

Functional measures to be used in conjunction with somatosensory assessments as recommended in this thesis include:

- Assisting Hand Assessment (Krumlinde-Sundholm, 2003).
- Canadian Occupational Performance Measure (Law et al., 1990).

- Goal Attainment Scaling (Kiresuk, Smith, & Cardillo, 1994).
- Jebsen Taylor Hand Function Test (Jebsen, Taylor, Trieschmann, Trotter, & Howard, 1969).

#### **10.4 Knowledge translation plan**

Even after a tool is satisfactorily developed, clinical uptake is often very slow (Auld & Johnson, 2016). Through the knowledge partnerships outlined in Chapter one of this thesis, this research project was aligned with the clinical practice environment by being linked to a major paediatric hospital and rehabilitation centre in Western Australia. This involved collaborating with key stakeholders within the organisations to participate in the research project and therefore, direct dissemination of findings. One example of this direct dissemination of research findings to clinical practice was in the form of a mini-symposium organised by the manager of a state-wide rehabilitation centre, Ability Centre. The PhD candidate was asked to assist in the design of the symposium and present a summary of the current PhD thesis. The target audience was allied health therapists who will be end users of the *sense\_assess© kids* and knowledge users of other literature linked to this current thesis (Appendices A,B,C,D,F,G,H). The presentation aimed to present the key messages in a plain language summary appropriate to the target audience. An associated infographic depicting the key messages of the doctoral research was designed by the PhD candidate to support knowledge translation (KT) at this level (see Figure 30).

As part of this thesis a KT plan was developed (Table 15), it was informed by the barriers and facilitators identified by Auld and Johnston (2016), and by the KT training manual by Barwick and Heiden (2016). The purpose of the KT plan was to outline the possible channels available to deliver the key messages of this thesis related to clinical use of the *sense\_assess© kids*. Therefore, the specific KT plan is designed for: children and their families who can use the knowledge to inform their healthcare decisions, and identify relevant future research questions; therapists who will be administering the somatosensory assessments; managers who will purchase the assessments and promote their use; and other medical personnel who will refer children with a suspected deficit to allied health services. The PhD candidate has completed formal training via the Scientist Knowledge Translation Training

Australia Course (2016) and will access KT supports within research partner organisations to implement the KT plan if needed (e.g. University of Western Australia and Curtin University).

Figure 30 Infographic summarising key messages in plain language

# HOW WELL CAN WE MEASURE SENSATION IN THE HANDS OF CHILDREN WITH CEREBRAL PALSY?

FOR MORE INFO EMAIL:  
SUSAN.TAYLOR4@HEALTH.WA.GOV.AU

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 **SENSORY IMPAIRMENT**  
Children with cerebral palsy often experience sensory deficits in the hands and this affects the way they use them.

 **STATEMENT OF THE PROBLEM**  
Clinicians do not have a comprehensive way of measuring how well children feel things with their hands.

 **ASSESSMENT DEVELOPMENT**  
An assessment used for adult stroke survivors has been modified for children with cerebral palsy

 **CAN WE USE THE NEW ASSESSMENTS?**  
This assessment has been tested to see if we can use it for children with cerebral palsy. With careful monitoring we can use some of the new sensation measures in practice.

 **FUTURE RESEARCH**  
Further testing continues to strengthen the new assessment tools and support advances in sensory intervention to improve outcomes for children with cerebral palsy.

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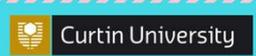
  



Table 15 Knowledge translation plan

Partners	Key messages	Strategies
Researchers	<p>Construct validity, reliability, responsiveness and clinical acceptability has been examined for sense_assess© <i>kid</i> subtests. There are now future research opportunities to continue psychometric testing of the sense_assess© <i>kids</i> and these are the measurement properties highlighted in yellow in Figure 2.</p> <p>Creating and maintaining relationships with collaborative knowledge partners is necessary for uptake of clinical research findings. Knowledge partnerships can help shape future research processes, define gaps in service delivery, generate clinical research questions and methods, and develop resources (including organisational assessment guidelines and templates).</p>	<p>Face-to-face meetings</p> <p>Local, national and international symposiums and conferences</p> <p>Peer review journal publications</p> <p>Collaborate with local universities to increase PhD and honours candidature, and student practicums related to evidence based practice in a clinical setting.</p>
Clinicians	<p>Measuring somatosensation is key to providing tailored somatosensory interventions.</p> <p>The sense_assess© <i>kids</i>' subtests; the PTT and WPST can be used, with careful monitoring, and in combination with goal based measures (e.g. COPM, GAS) in clinical practice.</p> <p>The fTORT, TDT and FMT can be used in clinical research to further test their psychometric properties prior to use in clinical practice.</p> <p>If you identify a potential issue related to sensory deficits for a patient, you can notify one of the researchers on our team and we can aim to involve them in the current research program at PMH.</p>	<p>Offer certification in the sense_assess© <i>kids</i> assessment and Sense© for Kids intervention.</p> <p>Provide somatosensory training workshops related to neurorehabilitation, and measurement and treatment of somatosensory deficits</p> <p>Engage clinicians in collaborative research projects</p>

Partners	Key messages	Strategies
Families and children	<p>Clinicians are now aware of somatosensory deficits and their impact on hand function for children.</p> <p>We have adapted an assessment and training program for management of these deficits, and it is being trialled with children and youth in Western Australia.</p> <p>If you or your child identify that they cannot feel things with their hands as well as other children you can discuss this with your paediatrician or allied health clinician with the aim of linking in with the current research occurring at PMH.</p> <p>If you have ideas for future research please contact one of our research team. We would like to engage you as consumer advocates to discuss what you and your child find important to research.</p>	<p>Consumer forums to summarise research findings and plans for future research.</p> <p>Inviting consumer engagement in this process through focus groups and sending emails to those who have already expressed interest.</p> <p>Participants are notified of the studies completion and outcomes via plain language summaries, brief reports, and certificates of participation.</p> <p>Involve participants in the dissemination process by inviting them to speak at forums, use their photos for scientific posters and presentations.</p> <p>Scientific posters displayed in the Paediatric Rehabilitation Department of the children's hospital frequented by many research participants and families.</p>
Opinion leader	<p>Most therapists are aware of the importance of measuring somatosensation but many are not confident in the currently available assessment tools.</p> <p>If assessments are purchased, promoted, and manuals and test forms are made available, and education is provided therapists are more likely to utilise assessments.</p> <p>The sense_assess© kids can be purchased and used for clinical research at PMH for those clinicians who are interested in research.</p> <p>A major research study related to somatosensory identification and management is about to commence at PMH so clinicians can suggest patients to be involved.</p>	<p>Face-to-face meetings to raise the profile of somatosensory assessment and management.</p> <p>Funded professional development opportunities will improve skills, confidence and awareness.</p> <p>Administration is allocated time to print test forms, and keep them available and updated.</p> <p>If the equipment is purchased, and the evidence is explained via an internal report outlining clinical utility (cost, time to administer etc).</p> <p>Continuous quality improvement and evaluation or clinical practice reminders could be introduced via department meetings.</p>

*Note.* PMH = Princess Margaret Hospital; PTT = Protective Touch Test; WPST = Wrist Position Sense Test; COPM = Canadian Occupational Performance Measure; GAS = Goal Attainment Scale; fTORT = functional Tactile Object Recognition Test; TDT = Tactile Discrimination Test; FMT = Fabric Matching Test.

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## APPENDICES

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## **APPENDIX A      Typically Developing Study**

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The PhD Candidate, Susan Taylor accounted for 85 per cent of the intellectual property associated with the final manuscript. Collectively, the remaining authors contributed 15 per cent.

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### **Assessing body sensations in children: reliability of assessment and effects of age**

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## **Abstract**

**Introduction:** Somatosensation is essential for all gross and fine motor skills. Impaired somatosensory functions such as touch sensation, limb position sense (proprioception) or haptic ability can delay learning new motor tasks and limit activity and participation. We aimed to provide normative data for the SenScreen© Sensory Screening Tool subtests of touch, wrist position sense and haptic object recognition in typically developing (TD) children and adolescents and investigate their intrarater reliability

**Methods:** A cross-sectional study of 88 TD children aged 6 to 15 years (mean age = 10.3yrs; SD = 2.6yrs) was used to determine the effects of age and gender on somatosensory capacity as measured by the SenScreen©. Intrarater reliability of SenScreen© subtests and the Small Objects component of the Jebsen Taylor Hand Function Test (JTHFT) was assessed in 22 of the 88 participants (mean age = 8.8yrs; SD = 2.6yrs). The SenScreen© test battery consists of the Protective Touch Test (PTT), Tactile Discrimination Test (TDT), functional Tactile Object Recognition Test (fTORT), and the Wrist Position Sense Test (WPST).

**Results:** Statistically significant differences were observed between age groups for tactile discrimination, wrist position sense and haptic object recognition, but not for touch registration for which all except one participant achieved a maximum score. There was no effect of gender. The PTT, fTORT, WPST and JTHFT demonstrated good intrarater agreement between time-points, but the TDT did not.

**Conclusions:** Somatosensory capacity increases with age for TD children aged 6 to 15 years. Three subtests of the SenScreen© demonstrated intrarater reliability with TD children. Further investigation of reliability of the TDT is required, and all subtests require psychometric testing with clinical populations.



## **Introduction**

Somatosensation refers to our ability to detect, recognise and discriminate body (somato) sensations. An intact somatosensory system maintains our capacity to detect changes in the external and internal environments (Marieb & Hoehn, 2007). Somatosensation includes the tactile and proprioceptive ability essential for skilled movements of the hand and finer movements of the fingers (Krumlind-Sundholm & Eliasson, 2002; Pehoski, 2006a; Stillman, 2002; Yu, 2012). Somatosensory inputs are also involved in more complex central nervous system processes such as haptic object recognition and emotional responses to pain or itch (Carey, 2012). The term ‘touch’ includes tactile registration and tactile perception, including light touch, touch sensitivity, tactile spatial resolution and tactile discrimination (Auld et al., 2012b; Bair et al., 2011; Bleyenheuft et al., 2006; Riquelme et al., 2011). Tactile sensation allows us to register threats such as pain, tell apart sharp and dull objects or different surface textures, process communicative touch from others and facilitate exploration of novel objects from a very young age (Dunn et al., 2013; Lederman and Klatzky, 2009).

Proprioception allows continuous adaptation to the environment (Stillman, 2002). It guides fine motor movements such as position of the wrist, precision grip and calibration of the pressure required for hand writing (Bumin & Kavak, 2010). Proprioception involves both static and dynamic components including limb position sense and movement sense and has a direct influence on coordinated movement (Goble et al., 2005). Haptic object recognition converts tactile stimuli into identifiable information such as object texture, hardness, temperature, weight and shape (Kalagher & Jones, 2011). The development of haptic object recognition using in-hand manipulation underlies a child’s ability to control objects necessary for educational activities, such as handwriting and has been shown to directly contribute to cognitive development in infancy (Case-Smith, 2010; Yu, 2012; Pehoski, 2006b).

Impairments in sensation can impede the development of new motor tasks (Krumlind-Sundholm & Eliasson, 2002). Proficient touch, limb position sense and haptic object recognition are important capacities of an intact somatosensory system, impacting on the functional use of the upper limb, particularly the hand (Bernstein et al., 2006; Carey et al., 1996). Information provided by the somatosensory system is

crucial for typical childhood development with children relying on cross validation between vision and the somatic senses when using their hands (Bremner et al., 2008; Carey, 2012). In adults, the loss of somatic sensation impacts on the tactile abilities needed to perform daily activities and for children deficits in sensation are strongly related to impaired dexterity (Carey et al., 2011a; Krumlind-Sundholm & Eliasson, 2002).

In order to appraise an individual's somatosensory capacity therapists must first be able to measure it (Fess, 2002). Quantifying somatosensory capacity is vital in understanding the impact of impairment on function, and provides information to guide treatment planning (Carey et al., 2011b; Fess, 2002). While assessments of fine motor ability are prevalent, there is a lack of comprehensive somatosensory assessments available for children (Auld et al., 2011; de Bruin, 2008; Stewart, 2010). Many assessments do not have documented validity or reliability in paediatrics, or lack standardised procedures increasing the risk of administrator and patient bias (Bentzel, 2008; Fess, 2002; Novak et al., 1993; Tassler & Dellon, 1995). Current best practice outcome measures for individual components of somatosensation are the Semmes-Weinstein monofilaments (Weinstein, 1993), and static and moving two-point discrimination (MacKinnon & Dellon, 1985) for touch registration and perception, haptic to visual object matching for haptic object recognition using the Klingels method (Klingels et al., 2010), and limb matching for proprioception of the upper limb (Auld et al., 2011; 2012b; Bentzel, 2008). However, the tactile assessments only measure one aspect of somatosensation and do not reflect functional ability. There is no standardised protocol for testing proprioception, and the lack of novel objects in the Klingels can produce a ceiling effect leaving no criterion standard against which to comprehensively measure somatosensory capacity for children (Bentzel, 2008; Boop, 2009; DeMatteo et al., 1993; Pei-Fang et al., 1998; Scheer et al., 1994; Wang et al., 2006).

This paper describes a new evidenced based, standardised measure of somatosensation capacity for use with children and youth. The SenScreen© Sensory Screening Tool has been developed for adults who experience somatosensory loss following stroke. The complete adult assessment includes the Sensory Questionnaire identifying awareness of any changes in or loss of body sensation and impact on limb

function and daily activities following stroke. The Protective Touch Test (PTT) which measures touch registration, close to the level of protective sensation across regions of the dorsal and palmar aspects of the hands, uses the 4.56 monofilament of the Touch-Test® Sensory Evaluator kit (Touch-Test® Sensory Evaluator). The Tactile Discrimination Test (TDT) (Carey et al., 1997) measures the ability to discriminate differences in finely graded textured surfaces using a three-alternative forced choice design.

The functional Tactile Object Recognition Test (fTORT) (Carey et al., 2006) is designed to test recognition of objects through touch without vision. Objects are displayed on a response poster and identical objects are provided for manipulation. Accuracy (correct identification of objects), time taken to explore objects and type of exploratory procedures are recorded as part of the test protocol. The Wrist Position Sense Test (WPST) (Carey et al., 1996) measures the capacity to identify wrist angle in the flexion-extension range following imposed movements by the therapist. The box-like apparatus uses a protractor scale to measure degrees of error. Fine motor ability is measured by recording the time taken to place six items into a can during the Small Objects component of The Jebsen Taylor Hand Function Test (JTHFT) (Jebsen, 1969). The Hand Function Survey - Brief Version (Blennerhassett et al., 2010) tests the ability to use the hand in daily activities and the Hot Cold Discrimination Test (Roylan® Hot Cold Discrimination Test Kit) assesses temperature discrimination.

A paediatric specific assessment tool of somatosensation would make it possible to understand the somatosensory functioning of children and adolescents. Therefore, our main aim was to adapt the relevant subtests of the SenScreen© and test their intrarater reliability for use with typically developing (TD) children aged 6 to 15 years. Secondly, we investigated if somatosensory capacity increased with age for TD children and adolescents including protective touch, tactile discrimination, haptic object recognition, wrist position sense and fine motor ability. Our first undertaking was to adapt the adult SenScreen© test battery for paediatric use. The self-report subsets; Sensory Questionnaire and Hand Function Survey were excluded from the battery because they required responses about daily activities that were not related to children and there was a need to reduce the administration duration for children.

The Hot Cold Discrimination Test was removed because there was a broader aim to develop a sensory training program involving somatosensory measurement and treatment and the intervention was not designed to ameliorate the somatosensory domain of temperature (McLean et al., 2017a). The remaining four SenScreen© subtests were as follows: Protective Touch Test (PTT), Tactile Discrimination Test (TDT), functional Tactile Object Recognition Test (fTORT), and Wrist Position Sense Test (WPST). Modification involved simplifying the standardised test instructions, modifying the assessment manual, test protocol and size of test equipment for a younger population. The adapted battery was renamed the SenScreen© Kids. The Jebsen Taylor Hand Function Test - Small Objects (JTHFT) was retained as an adjunct measure of uni-manual hand function to allow for future investigation of the relationship between somatosensation and motor function.

## **Methods**

With a convenience sample of 88 TD children and adolescents aged 6 to 15 years, a cross-sectional design was used to measure somatosensory capacity by gender and age in 3 strata: <8.9, 9-11.9 and >12 years. Twenty-two of these children were assessed again approximately 3 weeks later to investigate the intrarater reliability of the SenScreen© Kids. Ethical approval was granted by Edith Cowan University Ethics Committee, Perth, Western Australia (#8071), and each parent and/or guardian gave written informed consent, while all participants gave verbal assent. Recruitment occurred via professional and personal contacts between March 2012 and December 2013. Participants were assessed in their own homes at a time most convenient to the individual and family. Statistical methods included Bland Altman plots and exact agreement for the reliability study, and one way ANOVAs and Chi-Square tests for the developmental study.

## **Results**

The sample comprised 49 girls and 39 boys (mean age = 10.3yrs; SD = 2.6yrs). The majority of participants (n = 79) had right hand dominance. Reliability data collection involved 14 girls and 8 boys (mean age = 8.8; SD = 2.6). Not all participants were measured with all subtests.

**Developmental study:** Statistically significant differences were established between age groups for tactile discrimination, wrist position sense and haptic object recognition but not tactile registration (Table 1). Results are presented for the left and right hands, not by hand dominance. For tactile registration, children across all age groups for the right hand achieved 100% accuracy out of a possible six points (mean = 6; SD = .00). Only nine of the 88 participants in our sample had left-hand dominance therefore developmental differences in hand dominance was not analysed. Post hoc analysis revealed that the statistical significant differences existed between the <8.9 and 9 to 11.9 year age group and <8.9 and 12+ age group for tactile discrimination and haptic object recognition, and between the <8.9 and 9 to 11.9 year age group for wrist position sense. The relationship between gender and performance on the SenScreen© Kids subtests was not statistically significant: TDT right ( $X^2(30) = 32.175, p = .36$ ), left  $X^2(35) = 35.213, p = .46$ ); fTORT right ( $X^2(7) = 3.051, p = .88$ ), left ( $X^2(7) = 9.122, p = .24$ ), WPST right  $X^2(45) = 43.216, p = .55$ ), left  $X^2(44) = 45.919, p = .39$ ).

Table 1 SenScreen© Kids subtest scores and somatosensory domain measured

Subtest	Age group			P value
	<8.9 years M (SD)	9 - 11.9years M (SD)	12+ years M (SD)	
PTT Tactile registration				
N=56	n=21	n=18	n=17	
Right hand	6.0 (.00)	6.0 (.00)	6.0 (.00)	
Left hand	5.9 (.22)	6.0 (.00)	6.0 (.00)	.44
TDT Tactile discrimination				
N=45	n=14	n=14	n=17	
Right hand	47.94 (21.74)	64.12 (17.97)	69.08 (12.33)	.005
Left hand	38.45 (15.51)	60.87 (24.00)	72.16 (19.11)	.000
fTORT Haptic ability				
N=69	n=22	n=24	n=23	
Right hand	39 (2)	40 (2)	41 (1)	.000
Left hand	38 (3)	40 (2)	40 (1)	.006
WPST Wrist position sense				
N=52	n=18	n=17	n=17	
Right hand	13.8 (5.2)	11.7 (2.7)	10 (3.5)	.04
Left hand	15.6 (6.5)	12.3 (4.3)	11 (5.2)	.03

*Note.* TDT = Tactile Discrimination Test; fTORT = functional Tactile Object Recognition Test; WPST = Wrist Position Sense Test; JTHFT = Jebsen Taylor Hand Function Test; RH = right hand; LH = left hand; LOA = Limits of agreement; CI = confidence interval.

**Reliability study:** Three of the four SenScreen© subtests, and the JTHFT demonstrated good agreement between time-points (Table 2). Percent exact agreement was 99%-100% for the PTT, and the limits of agreement were small for the WPST right hand, fTORT and the JTHFT. The limits of agreement were wide for the TDT, and the WPST left hand; indicating poor intrarater reliability.

Table 2 Test re-test scores for the SenScreen© Kids subtests

Subtest Sample N=22	TDT Accuracy	fTORT Accuracy	fTORT Time	WPST Degrees	JTHFT Time
Mean difference RH	2.41	0.41	-0.14	-1.59	-0.87
LOA	-83.65 – 88.48	-1.90 – 2.72	-0.99 – 0.70	-12.98 – 9.81	4.65 – 2.91
95% CI	-9.35 – 14.17	-0.11 – 0.93	-0.33 – 0.05	-4.23 – 1.06	-1.73 – -0.01
Mean difference LH	2.80	0.73	-0.11	-3.02	-1.39
LOA	-78.90 – 84.50	-1.70 – 3.16	-0.77 – 0.55	-15.87 – 9.82	-4.78 – 1.99
95% CI	-8.37 – 13.96	0.18 – 1.28	-0.26 – 0.04	-5.93 – -0.12	-2.16 – -0.63

*Note.* TDT = Tactile Discrimination Test; fTORT = functional Tactile Object Recognition Test; WPST = Wrist Position Sense Test; JTHFT = Jebsen Taylor Hand Function Test; RH = right hand; LH = left hand; LOA = Limits of agreement; CI = confidence interval.

## Discussion

In the current study, good intrarater agreement was established for the PTT with children and adolescents similar to other studies involving the Semmes-Weinstein monofilaments and adults with median nerve injury (mean Intraclass Correlation Coefficient of 0.78 across test areas of the shoulder, wrist, hip and ankle) (Thibault et al, 1994). We also found that the TDT, fTORT and WPST demonstrated good intrarater agreement across two time-points. Similar to studies involving adults post-stroke where each measure achieved high reliability ( $r = .85$  to  $.92$ ) (Carey et al., 1996; 1997; Carey, Macdonell, & Matyas, 2011a). Jebsen, (1969) found the JTHFT to have moderate to high reliability ( $r = 0.60$  to  $0.99$ ) within a healthy population aged 20 to 94 years and in our sample of TD children the JTHFT also demonstrated good intrarater agreement.

The current study found that tactile discrimination increases with age. Our finding supports the work by Shiah et al. (2011) and Ardila et al. (2011) who

calculated a positive effect of age on touch sensation for children and adolescents aged 2 to 24 years. We also found that proprioceptive accuracy improves with age, with maximal error seen for children in the youngest age group (<8.9 years). This supports findings by Contreras-Vidal, (2006) and Goble et al. (2005) who reported that movement coordination and accuracy of limb position increases from ages 5 to 11 years. Children in our study demonstrated increased haptic object recognition ability with age which supports previous research by Kalagher and Jones. (2011) who reported the capacity to extract object properties and accurately identify objects through haptic exploration improved with age from childhood to young adulthood. It is expected that TD children and adolescents have intact protective touch sensation which was supported in the current study. The utility of the PTT will be in its ability to screen for deficits in children at risk of somatosensory impairment, such as those with cerebral palsy and will enhance the individual's somatosensory capacity profile when combined with other SenScreen© Kids subtests. Due to the limitations in our sample, the effects of hand dominance on the development of somatosensory capacity could not be explored. Future research is recommended to include a greater number of participants with an equal distribution of left and right hand dominance. Our main aim was to examine the suitability of an adult assessment tool for paediatric use. The tool now needs to be examined for test-retest reliability, validity, sensitivity and responsiveness in a clinical population.

Previously unavailable intrarater reliability data for the four subtests of the SenScreen© Kids has been provided for children and adolescents. The current study has determined that three of the SenScreen© Kids subtests are reliable for use with TD children and adolescents aged 6 to 15 years. Data from TD participants provides the SenScreen© Kids with objectively defined criteria of abnormality. These standards may be used as a point of reference in clinical practice where previously, normative data existed only for adults. This initial work will lead to providing clinicians with an evidenced based comprehensive assessment tool to measure somatosensory capacity, support frameworks for intervention protocols and enable comparison between clinical groups.

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## **APPENDIX B      Systematic Review**

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# Does somatosensation change with age in children and adolescents? A systematic review

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## Abstract

**Background** Somatosensory modalities, such as touch, proprioception and haptic ability, greatly influence the achievement of developmental milestones for children. Describing somatosensory impairment, natural variability and typical or expected developmental changes across age groups will help establish frameworks for intervention in clinical populations. This systematic review aimed to determine how different somatosensory modalities develop across childhood into adolescence to use as a point of reference for children at risk of somatosensory impairment.

**Methods** Searches of five electronic databases were undertaken through EBSCO-host (MEDLINE, CINAHL, PsycINFO, SPORTDiscus and ERIC) for studies measuring at least one somatosensory modality in typically developing individuals between birth and 18 years and analysed by age. Characteristics of studies were collected including country of origin, sample size, demographics and outcome measure used. Quality assessment and data extraction were performed by two independent reviewers.

**Results** Twenty three cross-sectional studies were included from a total of 188 articles retrieved: 8 examined aspects of touch, 5 proprioception and 10 haptic ability. Variability of study designs and variation in assessment tools precluded any formal meta-analysis.

**Conclusions** Somatosensation matures through childhood into adolescence; however, the present review found the pattern of somatosensory development varied depending on the assessment tool used and the aspect of somatosensation being measured, making it difficult to describe typical performance. There is a need for comprehensive assessment batteries to measure the somatosensation, including touch, proprioception and haptic ability, of children at risk of somatosensory impairment to aid in the development of effective interventions.

## Keywords

adolescent, assessment, child, haptic ability, proprioception, somatosensation, touch, typical development

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## Introduction

Everyday functioning is dependent on the ability to meet the sensory and motor demands of an ever changing environment (Lundy-Ekman, 2002; Ben-Sasson *et al.*, 2009). A child relies

on the sensation of touch for precision and dexterity, proprioception for hand control and gross motor function, and haptic ability to efficiently explore objects with their hands (Marieb & Hoehn, 2007; Kalagher & Jones, 2011b). The somatosensory system allows registration and characterisation

of touch and further tactile exploration provides perceptual information about external objects and surfaces (Dijkerman & De Haan, 2007). Touch is a multifaceted construct and we have employed a previously developed framework to organise our thinking about the various terms of reference for touch.

The framework includes two domains: tactile registration and tactile perception (Auld, Boyd, Moseley & Johnston, 2011). Tactile registration is the basic initial processing of stimuli and/or sensing of surfaces and can include terms such as touch sensitivity, tactile sensitivity, and touch threshold or threshold detection which is described as the minimal external stimulus that produces excitation of a cutaneous receptor (Weinstein, 1993; Bell-Krotoski *et al.*, 1995; Riquelme *et al.*, 2011). Tactile perception allows interpretation and understanding of stimulus such as location, timing and identification (i.e. what the stimulus is) (Auld, Boyd, Moseley & Johnston, 2011). Tactile perception may include terms such as tactile spatial acuity or tactile spatial resolution which is the ability to perceive the fine structure of a surface pressed against the finger (Bleyenheuft *et al.*, 2006; Peters & Goldreich, 2013); tactile localisation which is the ability to indicate the location of a stimulus on the skin (Yoshioka *et al.*, 2013) and tactile discrimination which is the ability to distinguish between different surface textures (Carey, Matyas & Oke, 2002; Dunn *et al.*, 2013).

All aspects of touch are important for calibrating grasp and preventing objects from slipping, discriminating between different textures and in locating the origin of sensations within the immediate environment (Eliasson, 2006; Bremner *et al.*, 2008). Disruptions in tactile registration and perception can impact dextral performance and everyday activities (Carey *et al.*, 2002; Krumlinde-Sundholm & Eliasson, 2002).

Proprioception is the sense of the movement and position of the body, and in particular the limbs (Stedman's medical dictionary, 2008). The term proprioception includes the static component of limb or joint position sense (Bremner *et al.*, 2013) and the dynamic component of sensing movement through kinaesthesia (World Health Organisation, 2001). The amalgamation of kinesthesia and limb position sense assists in proprioceptive localisation of the hand (Contreras-Vidal, 2006). Research consistently reports a clear link between proprioception and hand function, with proprioception affecting all gross motor and fine motor skills (Stillman, 2002; Bumin & Kavak, 2010). Proprioception is therefore important for education as it affects pencil grip and application of pressure (Bumin & Kavak, 2010), keyboard skills (Rao *et al.*, 2000) and is central to the execution of goal directed action through postural control (Viel *et al.*, 2009).

Haptic ability is often referred to as haptic perception (Ballesteros *et al.*, 2005; Kalagher & Jones, 2011a, 2011b; Gori *et al.*, 2012) or stereognosis (Auld *et al.*, 2012), and terms such as dynamic touch (Kloos & Amazeen, 2002; Fitzpatrick & Flynn, 2010), gnostic hand function (Van Grunsven *et al.*, 2003) or haptic object recognition (Carey, 2012) are often used interchangeably or defined as part of haptic ability. In this review we will principally use haptic ability to encompass the aforementioned subset terms for ease of reporting. It is important to note that although touch and proprioception influence haptic ability and are important for efficient exploration of objects (Dunn *et al.*, 2013) haptic ability relies on higher order cognitive processes and involves a complex integration of these somatosensory inputs as well as effective haptic exploratory procedures (Carey, 2012). Identification of objects using haptic ability requires touch and in-hand object manipulation to extract information such as texture, hardness, temperature, size, weight and shape of an object (Lederman & Klatzky, 2009). Haptic ability is critical to haptic object recognition, because the way an individual moves his or her hands, and their choice of exploratory procedures, determines the sensory information extracted and ultimately the meaning that is derived from an object (Jones & Lederman, 2006; Kalagher & Jones, 2011b). A child's learning and early development is influenced by their level of haptic ability (McLinden, 2004; Ballesteros *et al.*, 2005).

Typical development is underpinned by an intact somatosensory system with unimpaired functioning of touch, proprioception and haptic ability (Royeen & Lane, 1991; Pehoski, 2006). Typical functional somatosensation provides a benchmark against which to compare clinical populations who are known to experience somatosensory deficits in the hands (Cooper *et al.*, 1995; Clayton, 2003; Auld *et al.*, 2012). From current literature we know that between 31 and 97% of children with cerebral palsy assessed experienced deficits in touch registration, proprioception and/or touch perception and stereognosis (Cooper *et al.*, 1995; Wingert *et al.*, 2008; Auld *et al.*, 2012). Identifying and describing somatosensory deficits are crucial in understanding the impact of impairment on hand function and, as a result, establishing frameworks for intervention (Carey, 2012). Little is known about the developmental trajectory of somatosensation in typically developing children and adolescents despite the publication of sensory and motor developmental milestones (Case-Smith, 2010; Vroman, 2010). The objective of the current systematic review was to identify how somatosensory modalities mature in typically developing children from birth to 18 years to use as a point of reference for children at risk of somatosensory impairment.

## Methods

A search of five electronic databases was undertaken to identify studies that met *a priori* inclusion criteria. Databases were chosen based on their relevance to the Participants, Interventions, Comparisons, Outcome and Study design (PICOS) model that formulated the review objective (Moher *et al.*, 2009). The relevant PICOS elements used to determine the eligibility of studies were:

### Participants

Typically developing children from birth to 18 years with no history of neurological, sensory, sensorimotor, cognitive, psychiatric, psychological or physical impairments. *Comparisons*: Changes across age, birth to 18 years. *Outcomes*: Tactile, haptic and/or proprioceptive ability. *Study designs*: Any meeting inclusion criteria.

This review was conducted according to the Centre for Reviews and Dissemination Guidelines (Centre for Reviews and Dissemination, 2009) and is presented following the PRISMA statement for reporting systematic reviews and meta-analyses (Moher *et al.*, 2009). Electronic resources were screened from 2002 via EBSCO-host (MEDLINE, Cumulative Index of Nursing and Allied Health Literature – CINAHL, PsycINFO, SPORTDiscus with Full Text and ERIC) to April 2015. Reference lists of relevant studies were also manually screened. Only studies published between 2002 and 2015 were included as this review aimed to answer the objective with maximal appreciation of current research designs and contemporary knowledge.

Articles were selected according to the following inclusion criteria: (i) study describes somatosensation in typically developing children aged between birth and 18 years analysed by age (including those selected as a comparison group to a clinical population); (ii) comparison between two or more age groups; (iii) description of functional aspects of touch, proprioception or haptic ability for the hands and upper limb. Exclusion criteria were: (i) studies where confounding factors could not be eliminated, e.g. prior knowledge of outcome measure or influence of other sensory modalities, such as vision; (ii) studies examining somatosensation of the lower limbs or trunk, sensory seeking, sensory processing or integration, visceral, vestibular, oral, auditory, olfactory or visual sensations.

Search term combinations included a population term (i.e., *typic\* develop\* child\**) a sensory term (e.g. *propriocept\**) and a descriptor (*change\**). The selected search strategy was *sensation or propriocept\* or haptic or haptic perception or haptic*

*object recognition or touch or tactile or tactual or hand sensibility. fs AND child\* or adolescen\* or young adulthood or teenagers or young people or youth or typical\* develop\* AND Development\* change or change\* or maturation or age\* or longitudinal.fs or follow-up or lifespan*. Searches were guided by a librarian who assisted with the formulation of the search strategy through truncation and explosion of text terms and selection of Boolean operators. The search strategy located many articles related to drugs, smoking and sensory-seeking behaviours that were irrelevant to this review because of the terms *adolescent\**, *teenagers* and *youth*; however it was deemed appropriate to include these terms in order to maximise sensitivity.

Each paper was assessed for methodological quality and risk of bias by two independent reviewers using: (i) 10 item checklist for quality of reporting and (ii) critical review of applicable criteria for quality of evidence. These methods were based on the assessment tool for quantitative studies developed by Kmet *et al.* (2004). The checklist was modified by removing irrelevant items such as allocation of clinical comparison groups or items related to interventional studies. The 10 item checklist enabled each reviewer to allocate a summary score (total sum / total possible sum of 20) and rating of strong, good, adequate or limited to describe the methodological quality of reporting items most significant to this review (Lee *et al.*, 2008). In addition, two items were chosen from the Kmet checklist of methodological quality of evidence and summarised descriptively: Was the group appropriate for objectives? (e.g. socioeconomic status, demographic information or age); were the measurements of outcomes resistant to bias? (e.g. psychometric properties reported or available). Item selection was based on what the results of the studies relevant to this review would be most vulnerable to and enabled detection of systematic bias. If the studies did not meet the criteria for quality of reporting or evidence they were excluded (Table 1).

Data extraction forms were developed prior to the literature search. Two authors independently reviewed each study and extracted the following data: first author, country of origin of study and sample (often comparison group), study design, sample size, age range, gender, somatosensory modality examined, outcome measure used and associated psychometric information and summary of results. The variability of included studies with respect to age range, method of measurement of somatosensation and, hence, the aspect of the modality under investigation, and the method of reporting results precluded any formal meta-analysis therefore the findings are summarised descriptively.

**Table 1.** Description of four studies excluded for methodological reasons

First author, year of publication, sample origin	Somatosensory modality of interest	Design and sample	Typically developing criteria	Outcome measure	Exclusion criteria
Alexander (2002) USA	Haptic perception	Cross-sectional N = 35 (n = 1 dropout) Aged 4–9 years 31 males 4 females	None disclosed	Cross comparison task (identical or not), haptically explored pairs of familiar (dinosaur) and unfamiliar (sea creature) models.	Measurement of outcomes not resistant to bias: Confounding factor; there was some association between age (and IQ) and knowledge, and the fact that children did better on cross comparisons of dinosaurs than of sea creatures suggests that knowledge is exerting some effect.
Auld (2012)	Tactile registration and tactile perception	Cross-sectional N = 31 Aged 5–17 years 15 males 16 females	Typically developing children; no impairment in intellect (<70 on the Kaufman brief Intelligence test), upper limb performance (Jebsen), behavior (DSM-IV), or peripheral nerve lesions, upper limb fractures, or uncorrected visual impairment	Tactile registration: 20 item SWM kit. Spatial tactile perception: Single point localization; largest SWM. Two-point discrimination: Disk-Criminator; Double simultaneous; two bristles on a wooden rod. Texture tactile perception: AsTex. Motor enhanced tactile perception; stereognosis using 9 common objects.	Group not appropriate for objectives: Author unable to provide additional information for age comparisons because of small group sample size.
Bremner (2008)	Tactile spatial localisation	Cross-sectional Experiment 1 N = 12 Aged 10 months 9 males 3 females N = 24 Aged 6.5 months 11 males 13 females	Gestational age exceeding 37 weeks	Direction of ocular and/or manual responses to vibrostimulation (10 m group) to + or – vibrostimulation (6.5 m group).	Measurement of outcomes not resistant to bias: Because of evidence of habituation across trials the outcome measure may be unreliable and the experimental method differs between the 2 groups being compared.
Liutsko et al. (2014)	Proprioception	Cross-sectional N = 41 Aged 12–17 years	Self-described healthy, none receiving medication or had neurological problems. Normal or corrected-to-normal vision	Computerized test and equipment comprised a tactile screen and a sensory stylus (for hand drawing) to measure line length accuracy.	Group not appropriate for objectives: Author unable to provide additional information for age comparisons because of small group sample size

## Results

The electronic and manual searches detected 188 articles, 27 cross-sectional studies met the inclusion criteria.<sup>1</sup> Four were subsequently excluded on account of poor methodology, the inability to eliminate the impact of confounding factors, such as prior knowledge of the somatosensory task, or because the required data could not be extracted at the group level (Table 1). The included studies investigated aspects of touch  $n = 8$ ; proprioception  $n = 5$ ; and haptic ability  $n = 10$  (Fig. 1). No systematic reviews were identified. All 23 studies were cross sectional in design, with a combined total of 2418 typically developing participants in which 749 were assessed for touch (age range = 3–18 years), 256 participants for proprioception (age range 3–18 years) and 1413 participants for aspects of haptic ability (age range = 2.5–16 years). All participants met the criteria of typical development, 58% of the total participants were from the Americas (United States, Mexico/Columbia, Canada) and 38% were of European background (Belgium, France, Italy, Netherlands, Spain) and 4% from Australia. Methodological quality of reporting summary scores ranged from adequate (11/20) to strong (17/20) (Table 2).

### Touch

To measure touch sensitivity Riquelme *et al.* (2011) used standardised von Frey monofilaments (0.14–1.01 mm) which have documented psychometric properties (Somedic Sales AB, Sweden). To measure tactile perception Abu-Dahab *et al.* (2013) used the Reitan–Kolve test of Finger Agnosia and Dunn *et al.* (2013; 2015) used the standardised Tactile Discrimination Test to measure tactile discrimination. Both measures reported robust psychometric properties for adults (Reitan & Wolfson, 1985; Carey, Oke, & Matyas, 1997). Bleyenheuft *et al.* (2006; 2010) employed the Grating Orientation Task (GOT) to measure tactile spatial resolution and Peters and Goldreich (2013) used the GOT to measure tactile spatial acuity. Original literature reports the GOT as a measure of tactile spatial acuity (Craig, 1999; Gibson and Craig, 2002) and also tactile spatial resolution (Johnson & Phillips, 1981), and reports the terms interchangeably (Craig and Johnson 2000). The GOT has been validated in adults, but not children (Craig, 1999; Gibson and Craig, 2002). Yoshioka *et al.* (2013), measured tactile localisation using testing procedures and stimuli designed

<sup>1</sup> Please note three studies (Abu-Dahab *et al.*, 2013; Dunn *et al.*, 2013; Dunn *et al.*, 2015) examined aspects of touch, proprioception and/or haptic ability in the same paper using the same population so therefore were counted multiple times across somatosensory categories.

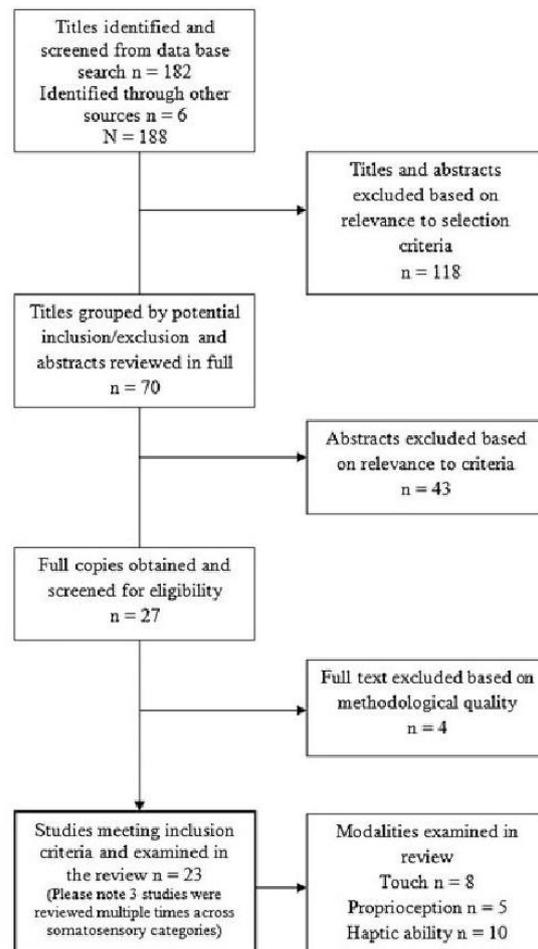


Figure 1. Flowchart of study selection.

specifically for their study, and did not report on psychometric validity.

The association with age varied with the aspect of touch measured. One study examined tactile registration and reported a positive effect of age however reported no statistically significant differences. Riquelme *et al.* (2011) showed touch sensitivity improved with age; touch thresholds in 6–10 years olds were greater (less sensitive) than adolescents (11–17 years).

Seven studies examined tactile perception and reported a positive effect of age with three studies reporting statistically significant differences. Bleyenheuft *et al.* (2010) reported scores for tactile spatial resolution were highly correlated with age. Calculated values for the GOT decreased (improved) with age

Table 2. Description of included studies

First author, year of publication, sample origin	Somatosensory modality of interest and subcategory	Outcome measure	Design and sample	Typically developing criteria	Results	Methodological quality (Total sum/total possible sum)
<b>Touch</b> Riquelme (2011) Spain	<b>Tactile registration</b> Touch sensitivity	von Frey monofilaments.	Cross-sectional N = 100 Aged 6–17 years 50 6–10 year olds 50 11–17 year olds	Cognitive level that allowed understanding of simple dichotomous questions.	<b>Improved with age.</b> There was an effect of age for touch sensitivity between age groups: touch thresholds in 6–10 year olds were greater (less sensitive) than adolescents (11–17 years).	Adequate (12/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Abu-Dahab (2013) USA	<b>Tactile perception</b> Finger recognition	Reitan-Kolve tests of Finger Agnosia.	Cross-sectional N = 38 Aged 5–12 years 12 5–<8 year olds 26 8–<12 year olds	Healthy volunteers, no history of learning disability, neuropsychiatric disorder, psychological disorder, family history of autism or heritable neuropsychiatric disorder.	<b>Improved with age.</b> There was an effect of age with errors in finger recognition decreasing from 80% (5–<8 years) to 44% (8–<12 years).	Adequate (11/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Dunn (2015) USA, Australia	<b>Tactile perception</b> Tactile discrimination	Tactile Discrimination Test using standard texture gratings for stimuli.	Cross-sectional N = 71 Aged 3–12 years 29 3–6 year olds 42 7–12 year olds 35 males/36 females Cross-sectional N = 64 Aged 3–<19 years 18 3–<7 year olds 46 7–<19 year olds	Community-dwelling children, able to follow instructions.	<b>Improved with age.</b> Tactile discrimination improved with age with lowest scores reported in the youngest age group (3–6 years).	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Psychometric data reported.
Dunn (2013) USA, Australia	<b>Tactile perception</b> Tactile discrimination	Tactile Discrimination Test.	Cross-sectional N = 18 Aged 3–<19 years 18 3–<7 year olds 46 7–<19 year olds	Community-dwelling children.	<b>Improved with age.</b> Tactile discrimination improved with age. A statistically significant difference ( $p < .0001$ ) was reported between 3–<7 year olds and 7–<19 year olds.	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Psychometric data reported.
Bleyenheuft (2006) Belgium	<b>Tactile perception</b> Tactile spatial resolution	Grating Orientation Task (GOT) using an enlarged set of JVP domes.	Cross-sectional N = 222 Aged 6–16 years 105 males/117 females	Free from disease or injury that could affect manual tactile sensitivity.	<b>Improved with age.</b> There was a statistically significant difference ( $p < .0001$ ) between 6–9 year olds and 10–16 year olds for tactile spatial resolution. Scores decreased (improved) with age until 10–11 years of age and then stabilised.	Adequate (14/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported for children.
Bleyenheuft (2010) Belgium	<b>Tactile perception</b> Tactile spatial resolution	Grating Orientation Task (GOT) using an enlarged set of JVP domes.	Cross-sectional N = 104 Aged 4–17 years 52 males/52 females	'Healthy', no hand disease or injury.	<b>Improved with age.</b> Tactile spatial resolution improved with age. There was a statistically significant difference in calculated GOT values; scores decreased (improved) with age from	Good (15/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported for children.

Continues

First author, year of publication, sample origin	Somatosensory modality of interest and subcategory	Outcome measure	Design and sample	Typically developing criteria	Results	Methodological quality (Total sum/total possible sum)
Peters (2013) Canada	<b>Tactile perception</b> Tactile spatial acuity	Grating orientation task (GOT).	Cross-sectional N = 100 Aged 6–16 years 50 males 50 females	Participants free from cuts, calluses or scars on their dominant index finger, as well as conditions that might affect their sense of touch such as diabetes, cognitive impairment, dyslexia or neurological conditions.	4 to 17 years ( $p < 0.001$ ) and stabilised at 10–16 years. Lowest tactile spatial resolution was reported for the youngest children (4–5 years).  <b>Did not improve with age.</b> No significant effect of age on tactile spatial acuity ( $p = 0.403$ ). Age alone does not impact tactile spatial acuity; other factors may include fingertip size.	Good (16/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Yoshioka (2013) USA	<b>Tactile perception</b> Tactile localisation	Stimuli delivered via a 30-g probe with round rubber tip 7 mm in diameter. Error was calculated as the distance between the stimulus and response location.	Cross-sectional N = 50 Aged 4–9 years 18.4-year olds 16 6 year olds 16 9 year olds	Normally developing children.	<b>Improved with age.</b> Mean errors in tactile localisation decreased with age from 4 to 9 years and was stable thereafter. The average error of stimulus-localisation was largest for children aged 4–6 years.	Good (15/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
<b>Proprioception</b> Contreras-Vidal (2006) USA	<b>Proprioception</b> Kinesthesia, hand localisation	A kinesthetic-to-visual matching task void of visual feedback to assess hand localisation (internal hand representation) and control of movement. Movement time (MT), terminal end-point error (EPE) and initial directional error (IDE) was measured.	Cross-sectional N = 15 Aged 5–10 years 5 5–6 year olds 5 7–8 year olds 5 9–10 year olds	None disclosed.	<b>Improved with age.</b> Statistically significant effect of age for all dependent variables (MT $p < .005$ ; EPE $p < .001$ ; IDE $p < .05$ ). Constant and variable errors decreased as age increased. End-point error variability of the hand was largest for the youngest children and did not differ within the older age groups.	Good (15/20) Comparison group was appropriate for objectives; however small sample size and subject group was not adequately described. Measurement of outcomes resistant to bias: No psychometric data reported.
Dunn (2013) USA, Australia	<b>Proprioception</b> Kinesthesia	Brief Kinesthesia Test asking participants to reproduce upper limb movements without vision after being guided by an examiner.	Cross-sectional N = 100 Aged 3–<19 years 37 3–<7 year olds 63 7–<19 year olds	Community-dwelling children.	<b>Improved with age.</b> Kinesthesia improved with age with statistically significant differences seen between the younger (3–<7) and older age groups ( $7-<19$ ) ( $p = 0.000$ ).	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Psychometric data reported.

Continues

Table 2. (Continued)

First author, year of publication, sample origin	Somatosensory modality of interest and subcategory	Outcome measure	Design and sample	Typically developing criteria	Results	Methodological quality (Total sum/total possible sum)
Goble (2005) USA	<b>Proprioception</b> Proprioceptive accuracy	An active matching task of target positions void of visual feedback to assess proprioceptively guided movements.	Cross-sectional N=18 Aged 8–18 years 9 8–10 year olds 9 16–18 year olds	Good general health and able to perform common upper limb activities.	<b>Improved with age.</b> Statistically significant effect of age for accuracy in matching movements ( $p < 0.001$ ). Matching error decreased as age increased.	Good (15/20) Comparison group was appropriate for objectives, however small sample size. Measurement of outcomes resistant to bias: No psychometric data reported.
Hay (2005) France	<b>Proprioception</b> Proprioceptive information	A tendon vibration technique and serial pointing task to assess the role of proprioceptive inputs for goal-directed movements. Constant error (CE), variable error (VE) and position error (PE) were measured.	Cross-sectional N=52 Aged 5–11 years	Average in terms of school achievement with no known sensorimotor impairments in their health records.	<b>Improved with age.</b> Statistically significant effect of age for CE ( $p < .02$ ), VE ( $p < .01$ ) and PE ( $p < .02$ ). Variable and position errors decreased with increasing age with the lowest accuracy at age 5.	Good (15/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Dunn (2015) USA, Australia	<b>Proprioception</b> Kinesthesia, wrist position sense	A battery of 3 proprioceptive tests to assess wrist position sense and kinesthesia. (Brief Kinesthesia Test, Wrist Position Sense Test and Clinical Test of Wrist Position).	Cross-sectional N=71 Aged 3–12 years 29 3–6 year olds 42 7–12 year olds 35 males/36 females	Community-dwelling children, able to follow instructions.	<b>Improved with age.</b> Kinesthesia and wrist position sense improved with age with a decrease in error reported from 3 to 12 years.	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Psychometric data reported.
Gori (2012) Italy	<b>Haptic ability</b> Haptic size perception	Participants were presented a sequence of differently sized plastic spheres in the hand (vision occluded) and were asked to report which sphere appeared larger.	Cross-sectional N=34 Aged 6–16 years 9 6 year olds 9 8 year olds 8 10 year olds 4 14 year olds 4 16 year olds	<b>Haptic ability</b> Children from elementary, intermediate and high schools in Prato (PO Italy).	<b>Improved with age.</b> More errors in perceived size for younger children (6–10 years) than older children (13–16 years).	Adequate (14/20) Comparison group was appropriate for objectives, however small numbers for 14–16 year olds. Measurement of outcomes resistant to bias: No psychometric data reported.
Kalagher (2011a) USA	<b>Haptic ability</b> Haptic perception, haptic exploration, haptic object recognition	Haptic exploration condition using 16 novel and 9 familiar objects to assess haptic-to-visual object matching skills.	Cross-sectional N=72 Aged 2.5–5 years 12 2.5 year olds 12 3 year olds 12 3.5 year olds 12 4 year olds 12 4.5 year olds 12 5 year olds 5–7 males and 5–7 females in each age group	Representative of local community in ethnicity, racial identity and social class.	<b>Improved with age.</b> Statistically significant effect of age ( $p < .01$ ). Older children (5 years) made more correct shape based matches than younger children (2.5–4.5 years). Children younger than 5 years matched 'randomly' and did not produce efficient exploratory	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.

Continues

First author, year of publication, sample origin	Somatosensory modality of interest and subcategory	Outcome measure	Design and sample	Typically developing criteria	Results	Methodological quality (Total sum/total possible sum)
Kalagher (2011b) USA	<b>Haptic ability</b> Haptic exploratory procedures	Haptic condition match-to-sample task, involving haptic inspection of 11 object sets to assess haptic exploratory behaviours.	Cross-sectional N = 36 Aged 3–5 years 12 3 year olds 12 4 year olds 12 5 year olds 5–7 males and 5–7 females in each age group	Representative of local community in ethnicity, racial identity and social class.	procedures when manipulating objects. <b>Improved with age.</b> Children aged 3–5 years can and do produce similar haptic exploratory procedures as adults when determining object properties.	Strong (17/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Kloos (2002) USA	<b>Haptic ability</b> Dynamic touch	A task involving identification of weight via the hand, void of visual feedback to assess perceptual ability through dynamic touch.	Cross-sectional N = 18 Aged 3–5 years 7 3 year olds 5 4-year olds 6 5 year olds 11 males 7 females	None disclosed.	<b>Improved with age.</b> There was a statistically significant effect of age for detection of changes in mass $p < .005$ . Children were more sensitive to changes in mass than younger children.	Adequate (12/20) Comparison group was appropriate for objectives, however small sample size and subject group was not adequately described. Measurement of outcomes resistant to bias: No psychometric data reported.
Abu-Dahab (2013) USA	<b>Haptic ability</b> Tactile perceptual skills (haptic object recognition)	Luria-Nebraska test of Stereognosis.	Cross-sectional N = 38 Aged 5–12 years 12 5–<8 year olds 26 8–<12 year olds	Healthy volunteers, no history of learning disability, neuropsychiatric disorder, psychological disorder, family history of autism or heritable neuropsychiatric disorder.	<b>Did not improve with age.</b> Indicated no significant decrease in error for stereognosis between the youngest and oldest age groups 24% Error (5–<8 years); 25% Error (8–<12 years).	Adequate (11/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Ardila (2011) Mexico/Columbia	<b>Haptic ability</b> Tactile perception (haptic object recognition)	Child Neuropsychological Assessment to assess cognitive development (subtest included tactile object identification).	Cross-sectional N = 788 Aged 5–16 years 350 males 438 females	No history of neurological, mental retardation, learning disabilities or psychiatric problems.	<b>Improved with age.</b> Performance in the tactile perception domain improved as age increased with the lowest scores reported for 5–6 year olds (13.8–14.9) when compared with 11–16 year olds (15.7–15.8).	Adequate (14/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Reference to psychometric data; high external validity.
Ballesteros (2005) Spain	<b>Haptic ability</b> Haptic perception	A haptic test battery to assess perceptual and cognitive abilities using active touch (20 subtests).	Cross-sectional N = 60 Aged 3–16 years	Children with no known psychological or physical impairments.	<b>Improved with age.</b> Effect of age apparent, improvement in 'Object naming' subtest as age increased however no statistical significance. Older children (14–<16 years) performed better than younger children (3–<5 years). <b>Improved with age.</b> Children's haptic object	Good (15/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Psychometric data reported; high internal consistency.
Dunn (2015) USA, Australia	<b>Haptic ability</b> Haptic object recognition	Cross-sectional N = 71 Aged 3–12 years				Strong (17/20) Comparison group was appropriate

Continues

Table 2. (Continued)

First author, year of publication, sample origin	Somatosensory modality of interest and subcategory	Outcome measure	Design and sample	Typically developing criteria	Results	Methodological quality (Total sum/total possible sum)
Fitzpatrick (2010) USA	<b>Haptic ability</b> Haptic perception	Brief Manual Form Perception Test.	29 3–6 year olds 42 7–12 year olds 35 males 36 females	Community-dwelling children, able to follow instructions.	recognition accuracy improved as they got older (from 3 to 12 years). Use of systematic exploration strategies accounted for most of the improvements in performance. <b>Improved with age.</b> Age was statistically significant with 3 year olds having lower accuracy than 4 and 5 year olds ( $p = .007$ ).	Adequate (14/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: No psychometric data reported.
Van Grunsven (2003) The Netherlands	<b>Haptic ability</b> Manual haptic gnosis/function	Investigation of a wooden object through passive touch without visual control to assess the development of gnosis hand function (shape and size detection).	Cross-sectional $N = 40$ Aged 3–5 years 12 3 year olds 15 4 year olds 13 5 year olds 25 males 20 females	None disclosed.	<b>Improved with age.</b> There is an effect of age on the development of morphognostic function; 48% of the 6 year olds correctly drew objects that their fingers had perceived compared with 61%–91% for the 11 year olds.	Good (16/20) Comparison group was appropriate for objectives. Measurement of outcomes resistant to bias: Acceptable test-retest reliability reported.

from 4 to 17 years ( $p < 0.001$ ) and stabilised at 10 to 16 years. Lowest tactile spatial resolution was reported for the youngest children (4–5 years). Similarly, in their 2006 study Bleyenheuft and colleagues reported a highly significant difference ( $p < 0.001$ ) between 6 to 9 year olds and 10 to 16 year olds for tactile spatial resolution. Dunn *et al.* (2013) reported that children 6 years and younger (3–<7) performed significantly less well ( $p < .0001$ ) than the older age group (7–<19) for tactile discrimination. Similarly, Dunn *et al.* (2015) described an increase in tactile discrimination as age increased with the older age group (7–12 years) outperforming the younger age group (3–6 years). Yoshioka *et al.* (2013) established that mean errors in tactile localisation decreased with age between 4 and 9 years and were stable thereafter. It was reported that 10 to 12 year olds reached an adult level of performance (0.02 of the hand length = average distance between the stimulus and response locations with relation to hand length) compared with 4 to 6 year olds who demonstrated a large average error (.05–.08 of hand length) (Yoshioka *et al.*, 2013). Abu-Dahab *et al.* (2013) reported an improvement in finger recognition as age increased with errors decreasing from 80% (5–<8 years) to 44% (8–<12 years). In contrast, Peters and Goldreich (2013) was the only study to report no significant effect ( $p = 0.403$ ) of age on tactile spatial acuity for their participant sample aged 6 to 16 years.

#### Proprioception

Reported outcome measures for proprioception involved either programmed stimulation or electrically controlled devices, or therapist imposed movements of the upper limb. A digitising tablet and pen (WACOM InTuos™) was used by Contreras-Vidal (2006) to detect errors in proprioceptive localisation of the hand. No psychometric information was reported. Goble *et al.* (2005) used custom software and forearm plates designed to measure elbow joint rotation, indicating proprioceptively guided movements through voltage output and Hay *et al.* (2005) used computer operated vibrators and targets during a serial pointing task. Goble *et al.* (2005) and Hay *et al.* (2005) reported that their testing procedure was based on previous research methodology; however, no psychometric data were reported. Dunn *et al.* (2013) used the Brief Kinesthesia Test (Ayres, 1972; 1980; 1989) and Dunn *et al.* (2015) used an assessment battery including the Brief Kinesthesia Test, Wrist Position Sense Test and the Clinical Test of Wrist Position (Carey, Oke, & Matyas, 1996). The Wrist Position Sense Test reported robust psychometric properties for adults.

All five studies reported that proprioception improved with age and four reported statistical significance. Contreras-Vidal (2006) found a statistically significant effect of age for all dependent variables (movement time  $p < .005$ ; end-point error  $p < .001$ ; initial directional error  $p < .05$ ). Constant and variable errors decreased as age increased from 5 to 10 years old. End-point error variability of the hand was largest for the youngest children (5–6 year old group) and did not differ within the older age groups (7–8 and 9–10 year olds). Goble *et al.* (2005) reported matching errors significantly decreased as age increased ( $p < .001$ ) with a mean absolute error of 6.2° for children (8–10 years) and 3.7° for adolescents (16–18 years). Errors made by children were also more variable and ranged from 0.1° to 20.8° compared with the adolescents where range of error was between 0.0° and 14.1°. Dunn *et al.* (2015) reported a decrease in error from 3 to 12 years for kinesthesia and also reported more variability in scores for 3 to 6 years olds compared with 7 to 12 year olds. Dunn *et al.* (2013) reported a statistically significant improvement ( $p = .000$ ) between 3 to <7 year olds and 7 to <19 year olds for kinesthesia. Hay *et al.* (2005) reported a statistically significant effect of age for constant error ( $p < .02$ ), variable error ( $p < .01$ ) and position error ( $p < .02$ ). Hay *et al.* (2005) also found highest variability in the youngest participants (5 years old) when performing the serial pointing task in extension, and poorest terminal accuracy at 5 years compared with the most accurate at ages 9 and 11 years. Variable and position errors decreased with increasing age with the lowest accuracy at age 5 years however in contrast the smallest constant error was made at age 5 and 11 years and the largest at age seven. Authors attributed the finding to young children's ballistic-type of movement control producing reasonably efficient end target accuracy. Goble *et al.* (2005) reported that although adolescents (16–18 years) had significantly higher proprioceptive control of movement than children (8–10 years) ( $p < 0.001$ ), the postural performance of 16 to 18 year olds did not match that of adults or individuals in late adolescence. Goble *et al.* (2005) also reported that pubertal growth and reorganisation of internal body schemas between ages 11 to 15 years in females and 13 to 17 years in males may temporarily decrease proprioceptive ability.

Dunn *et al.* (2015) was the only study to report on limb position sense at the wrist and reported a decrease in error from 3 to 12 years.

#### Haptic ability

Haptic ability was measured with a variety of different outcome measures. The 'Haptic Test Battery' used by Ballesteros *et al.*

(2005) demonstrated satisfactory subtest reliability (Cronbach's  $\alpha$  coefficient 0.54 to 0.88) and construct validity using factor analysis. Van Grunsven *et al.* (2003) reported acceptable agreement (weighted coefficient kappa .86 for left fingers, .78 for left thumb, .80 for right fingers and .79 for right thumb), for test–retest reliability for the 'Bottle test', specifically designed for their study. Ardila *et al.* (2011) measured identification of objects, using the left or right hand, with the Sensory Perception (identification of objects) subtest of the Child Neuropsychological Assessment (Matute *et al.*, 2007), which has documented external validity. Abu-Dahab used the Luria-Nebraska test of Stereognosis and did not report on the subtest's psychometric properties (Golden, Purisch & Hammeke, 1979). Three studies used instruments reported in previous literature; however, no psychometric data were reported. They included: novel category exemplars, the 'matching to sample' task (Kalagher & Jones, 2011a; 2011b) and Dunn *et al.* (2015) used The Brief Manual Form Perception Test adapted from Ayres (1972; 1980; 1989). Across the remaining studies outcome measures lacked definition in terms of reported reliability, validity, standardisation or available normative data such as 'plastic spheres 5 mm in diameter' (Gori *et al.*, 2012), '9 objects differing in mass and volume' (Kloos & Amazeen, 2002) and 'haptic matching task' (Fitzpatrick & Flynn, 2010).

Nine of the 10 studies examining haptic ability reported a positive association with age and three studies reported statistical significance. Fitzpatrick and Flynn (2010) reported significant improvements in haptic perception; 4 and 5 year olds demonstrated more accuracy than 3 year olds ( $p = .007$ ) for the haptic matching task. Kalagher and Jones (2011a) reported a statistically significant effect of age ( $p < .01$ ) for children matching novel category exemplars. Older children (5 years) made more shape based matches than younger children (2.5–4.5 years) in the haptic exploration condition. Children younger than 5 years matched 'randomly' and did not produce efficient exploratory procedures when manipulating objects. In a subsequent study Kalagher and Jones (2011b) reported that young children (3–5 years) did in fact produce similar haptic exploratory procedures as adults when determining object properties. Kalagher and Jones (2011b) attributed the contrast in their findings to differences in testing procedures. Their first study required intermodal transfer (haptic to visual matching) and their subsequent study did not. The second study also added perceptual goals where children were asked to find information about specific object properties. Dunn *et al.* (2015) reported improvements in haptic object recognition accuracy from 3 to 12 years and

the use of systematic exploration strategies accounted for most of the improvements in performance. Results showed that capacity to extract object properties and accurately identify objects through haptic exploration improved; however, performance did not progress linearly as age increased.

Ardila *et al.* (2011) reported an association between increasing age and object identification performance on the Sensory Perception subtest. Lowest scores were reported for 5 to 6 year olds (13.8–14.9) when compared with 11 to 16 year olds (15.7–15.8); however, the difference did not reach statistical significance. Ballesteros *et al.* (2005) reported that the ability to name objects increased with age; however, this progression was not linear across all age groups, although older children (14–<16 years) performed better than younger children (3–<5 years). Kloos and Amazeen (2002) reported that 5 year olds were more consistently proficient in detecting changes in mass of an object than 3 year olds. The interaction of mass, but not volume, and age was significant ( $p < .005$ ). For haptic judgments of size without vision, Gori *et al.* (2012) revealed more errors in perceived size for younger children (6–10 years) than older children (13–16 years) and only older children showed a capacity for multisensory integration between visual size perception and haptic calibration. Van Grunsven *et al.* (2003) showed clear developmental changes across two age groups of increasing haptic gnostic hand function with 48% of the 6 year olds correctly drawing the objects that fingers of both left and right hands had perceived, and 91% and 61% of the 11 year olds drawing correctly objects perceived by the right and left hands, respectively. In contrast, Abu-Dahab *et al.* (2013) was the only study to report no significant effect of age for stereognosis between the youngest and oldest age groups 24% Errors (5–<8 years); 25% Errors (8–<12 years).

## Discussion

Somatosensation matures through childhood into adolescence; however, the present review found the pattern of somatosensory development varied depending on the outcome measure used and the aspect of somatosensation being measured, making it difficult to describe expected performance. Almost all of the studies examining touch reported a positive correlation between increasing age and tactile registration and tactile perception. These findings support current evidence of improvements in tactile registration and perception between childhood and adulthood (Lundy-Ekman, 2002). In comparison, Bleyenheuft *et al.* (2006) and Peters and Goldreich (2013) used a similar outcome measure (Grating Orientation Task)

and reported contrasting results; however, the authors did agree that age alone does not determine tactile spatial acuity and resolution. Both authors suggested that skin hydration, receptor density or central maturation play a role as previous studies have highlighted (Stevens & Choo, 1996; Manning & Tremblay, 2006). The effects of age on touch were dependent on the outcome measure and the aspect of touch measured. Understanding the impact of touch on hand function requires measurement of multiple aspects of the tactile construct. Comprehensive tactile assessment should include both phases of tactile registration and perception, a tactile assessment framework described by Auld *et al.* (2011; 2012).

A significant improvement in proprioceptive ability was seen between 5 and 8 years and mature patterns of coordination were not well developed until after 9 to 10 years of age (Goble *et al.*, 2005; Hay *et al.*, 2005; Contreras-Vidal, 2006). Recent literature also states that refinements in proprioceptive input needed for motor output gradually occur throughout childhood from 6 years of age and continue well into adolescence (Visser & Geuze, 2000; Mallau *et al.*, 2010). Goble *et al.* (2005) considered the impact of recalibration of internal body schemas in response to pubertal growth between the ages of 11 to 15 years (females) and 13 to 17 years (males). Similarly, Viel *et al.* (2009) suggested that body scheme disturbances may lead to a transient period of proprioceptive neglect for adolescents aged 14 to 15 years. These findings may indicate a diversion from proprioceptive feedback in preference for more stable sensory systems, such as vision, in order to smoothly coordinate movement at this age (Viel *et al.*, 2009). Again, proprioceptive improvements are not linear or symmetric when considering the effect of age, with mixed results and variability seen between different testing conditions (Hay *et al.*, 2005). Objective and repeatable measures are required to quantify changes in proprioception across all ages.

All studies examining haptic ability reported an increase in haptic performance and/or use of haptic exploratory procedures with increasing age. Four of the studies reported variation at an individual level, indicating that improvements did not progress linearly as age increased; however, it was possible to identify trends in means when considering group data (Kloos & Amazeen, 2002; Ballesteros *et al.*, 2005; Fitzpatrick & Flynn, 2010; Abu-Dahab *et al.*, 2013). Kalagher and Jones (2011a; 2011b) reported a presence of haptic exploratory procedures similar to adults by age 5; however, current literature suggests that a child's ability to select the most efficient haptic exploratory procedure may develop at a later age (>7 years) (Alexander *et al.*, 2002). Perceptual processing of haptic input, haptic-to-visual intermodal transfer

and attentional capacity has been suggested as the means of improving haptic perception across childhood to adolescence and why children may develop manual perceptual proficiency at very different rates (Fitzpatrick & Flynn, 2010; Kalagher & Jones, 2011a; Cote, 2014). Gender, prior knowledge of test objects and ability to select efficient exploratory procedures also affects performance on haptic test batteries (Alexander *et al.*, 2002; Kalagher and Jones 2011a; 2011b).

While it is recognised that many factors influence the assessment of individuals in the developmental period, posing many challenges, amalgamating findings from this research is further complicated by the wide variability of methods and outcomes measures used (Dijkers *et al.*, 2012). In the same way, developing and evaluating the effectiveness of new interventions or other procedures become difficult without assessment tools standardised in the clinical populations for whom they are intended (Fess, 2002; Randall, 2008). Although measures of somatosensation impairment are used in research, few are normed, have robust psychometric properties or demonstrated validity or reliability in children and adolescents (Novak *et al.*, 1993; Tassler & Dellon, 1995; Fess, 2002; Connell & Tyson, 2012). Across the majority of included studies there was an absence of best practice methodology. Many assessment tools were designed specifically for research purposes and may have limited utility in clinical practice (Whyte *et al.*, 2009; Dijkers *et al.*, 2012).

Our ability to understand somatosensation in typically developing children, and best practice measures for clinical populations, is clearly linked to valid and reliable assessment (Cooper *et al.*, 1995). In a recent clinimetric review by Auld *et al.* (2011) the current best practice measures of touch for children with neurological disorders, such as cerebral palsy, were Semmes–Weinstein monofilaments, both static and moving two-point discrimination, and single-point localization (Burns, 1992; Weinstein, 1993; Tassler & Dellon, 1995). Best practice measures of limb position sense and kinesthesia for children is replication of therapist imposed movements of the upper limb without vision (Bentzel, 2008). Auld *et al.* (2011) suggest the best practice measure for haptic recognition for children with CP is the Klingels' stereognosis method requiring the use of 12 common objects, three matched pairs of similar items and six unmatched different items (Klingels *et al.*, 2010). Haptic test objects need to be easily manipulated by small hands and novel enough to avoid a ceiling effect (Auld *et al.*, 2012). While the current review recognises best practices measures in CP, overall they lack psychometric rigour, similar to the outcome measures utilised in selected studies. Outcome measures in the current review showed face validity but were inconsistent in terms of overall rigour, reliability, or other

types of validity, standard procedures or normative data. Further, varied units of measurement precluded any formal meta-analysis. Because of the lack of studies containing standardised measures and small sample sizes within the available evidence, interpretation of findings requires caution.

Somatosensation (touch, proprioception and haptic ability) increases with age from 2.5 years to 18 years; however, improvements do not progress linearly across time. The current review aimed to describe typical performance to use as a point of reference for children at risk of somatosensory impairment. Instead what we found were patterns of somatosensory development that varied depending on the outcome measure used and, hence, the aspect of somatosensation being measured, making it difficult to describe expected performance. Identifying and describing somatosensory deficits are crucial in understanding the impact of impairment on hand function and to inform evidence-based frameworks for intervention. Children and adolescents with neurological conditions, such as cerebral palsy, would benefit from individual somatosensory profiles gained from comprehensive measurement tools that comprise touch registration and perception, proprioception and haptic ability.

### Key messages

- Somatosensation (touch registration and perception, proprioception and haptic ability) increases with age from 2.5 years to 18 years.
- Somatosensory performance does not progress linearly as age increases and variability in somatosensory performance exists for individuals depending on the outcome measure used.
- Variability of outcome measures within research limits the comparison of results between studies and impacts the development of somatosensory interventions.
- Comprehensive assessment batteries of somatosensation are needed.

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## **APPENDIX C      Survey Study**

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Honours student Corrin Walmsley accounted for 85 per cent of the intellectual property associated with the final manuscript, collectively Susan Taylor and the other authors accounted for 15 per cent.

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**What is current standard practice of therapists in the measurement of somatosensation in children with cerebral palsy and other neurodevelopmental disorders?**

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## **Abstract**

**Introduction:** Somatosensation is the ability to detect and recognize external body sensations such as touch, vibration, pressure, pain, temperature and proprioception. Cerebral palsy is a neurological disorder that is often accompanied by impairments in somatosensation, however current assessments have limited psychometrics established for use with these children. The aim of this study was to identify therapists' current practice and perspectives related to the assessment of somatosensation in children with neurological disorders.

**Methods:** A cross-sectional questionnaire was used to identify somatosensory assessments currently used in clinical practice, time allocated to assessment, and therapists' satisfaction and confidence using the available assessments of somatosensation. The questionnaire was adapted from a previously utilised questionnaire that identified therapists' use of somatosensory assessments with adults' post stroke.

**Results:** A total of 135 therapists responded to the questionnaire. Seventy-nine (92%) occupational therapists and 44 (89.7%) physiotherapists indicated that they currently assessed or treated children with somatosensory deficits. Sixty-four (82.1%) occupational therapists and 38 (86.3%) physiotherapists regarded assessment of somatosensation in children with neurological disorders as important to very important. However, only 7 (8.8%) occupational therapists and 7 (15.9%) physiotherapists reported confidence in their ability to do so. The methods with which therapists detect and measure somatosensory impairment in children with neurological disorders are variable, with non-standardised and/or informal assessments most frequently used.

**Conclusion:** Therapists indicated low satisfaction and confidence with currently available assessments, which highlights the need for a comprehensive and standardised assessment of somatosensation for use in children with neurological disorders.



## **Introduction**

Sensation is the conscious or subconscious awareness of information from the external environment (Tortora & Grabowski, 2000). Without the function of sensation, it is challenging for humans to make sense of their surroundings (Parham & Mailloux, 2010). Somatosensation is the ability to interpret body sensations and incorporates the detection, discrimination and recognition of body senses such as touch, vibration, pressure, pain, temperature, proprioception and also more complex integrations of somatosensory inputs such as haptic object recognition (Carey, 2012). Somatosensation can help to enable exploration of the environment, has the potential to enhance performance and participation in functional tasks, and contributes to our learning and interaction as social beings (Auld, Boyd, Moseley, & Johnston, 2011; Bentzel, 2008; Parham & Mailloux, 2010).

Cerebral palsy (CP) is a common neurological disorder of childhood (Koman, Smith, & Shilt, 2004), with an increasing body of evidence to support the presence of somatosensory deficits in the upper limb (Auld, Boyd, Moseley, Ware, & Johnston, 2012b; Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Van Heest, House, & Putnam, 1993). A recent study reported that in a sample of 52 children with hemiplegic CP, 77% had tactile deficits and 40% were found to have both tactile registration and perception deficits of the hand (Auld, Boyd, Moseley, & Johnston., 2012a). In other studies involving children with CP, somatosensory deficits have been reported for 46% to 97% of these children (Yekutieli, Jariwala & Stretch, 1994; Van Heest et al., 1993).

Impairment in tactile sensation and proprioception have the potential to reduce fine motor control and manipulative ability and can make it difficult to identify the physical properties of objects as well as judge the amount of force required to complete a task (Bentzel, 2008; Cascio, 2011; Bumin & Kavak, 2010; Simões-Franklin, Whitaker, & Newell, 2011; Parham & Mailloux, 2001). The effect of these impairments on participation in everyday activities can be extensive (Parham & Mailloux, 2001) and a child's overall development and learning experiences can be compromised as a result of being unable to interpret tactile sensation in an accurate and proficient way (Arnould, Penta, & Thonnard, 2007; Cascio, 2011;

Parham & Mailloux, 2001). Despite this, very little is known about how therapists assess these impairments in children with neurological disorders.

Of the clinical measures available, most only have established psychometric properties for use with adults post stroke or peripheral nerve injury (Connell & Tyson, 2011; Krumlinde-Sundholm & Eliasson, 2002). Over recent years, the clinimetric properties of some of these assessments have been explored for use in children with CP (Auld, Russo, Moseley, & Johnston, 2014). In the impaired hand of 16 children with CP, Auld and colleagues (2012a) compared the reliability of the following assessments using the intraclass correlation coefficient (ICC) and agreement scores compared using the percentage of exact agreement (%EA). The Semmes-Weinstein monofilaments (SWM's), a measure of tactile registration, demonstrated excellent test-retest reliability (ICC = 0.96) and acceptable test retest agreement (%EA  $\pm$  1 = 75). Static and moving two-point discrimination are measures of spatial perception, with the static component demonstrated excellent test retest reliability (ICC = 0.96) and acceptable test retest agreement (%EA = 80), with moving two-point discrimination resulting in less agreement (%EA = 53). The AsTex® is a measure of texture discrimination which demonstrated fair test retest reliability (ICC = 0.42 - 0.48) (Auld et al., 2012a). These measures focus on touch registration and discrimination. Despite some of these assessments demonstrating adequate reliability, a comprehensive assessment tool of high quality should have all measurement properties developed and evaluated. This would include the validity, reliability and responsiveness within an intended clinical population as suggested by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (Mokkink et al., 2010).

Auld and colleagues (2011) concluded there was no one tool that can sufficiently address all areas of somatosensation, with recommendations directed towards combining the use of subtests from existing assessments. The SWM's (Weinstein, 1993), Disk-Criminator (MacKinnon & Dellon, 1985) as well as Single Point Localisation (SLP) and Double Simultaneous which are both subsets of the Neurological, Sensory, Motor, Developmental Assessment (NSMDA) (Burns, 1992), are recommended to provide a comprehensive evaluation of tactile function (Auld et al., 2011). Additionally, a comprehensive assessment of somatosensation should

evaluate multiple domains of somatosensation such as touch, proprioception and haptic object recognition (Carey, 1995; Connell & Tyson, 2011; Cooper et al., 1993). Given this knowledge, the aim of the current research was to identify therapists' current practice and their perspectives related to the assessment of domains of somatosensation for children with neurological disorders

## **Methods**

### ***Participants***

Occupational therapists and physiotherapists working with children with neurological disorders and somatosensory deficits were identified and invited to participate in the study. Therapists were required to have one year or more of clinical experience in the field of paediatrics and currently be working in Australia.

### ***Instrument***

A cross-sectional questionnaire was developed for this study based on previous research by Pumpa, Cahill and Carey (2015) who utilised a questionnaire to identify somatosensory assessments used by therapists for adults post stroke. The adapted questionnaire used in the current study included 34 questions and obtained information about therapists' current practice and perspectives on somatosensory assessment for their current paediatric neurological caseload. Paper and online versions of the questionnaire were disseminated. Questions sought information on demographics, somatosensory assessments currently used in clinical practice, time allocated to somatosensory assessment, and therapists' satisfaction and confidence using currently available somatosensory assessments. The questionnaire was piloted with five practicing occupational therapists prior to distribution. Minor changes were made to the questionnaire based on their recommendations. Dichotomised (e.g. yes/no, male/female), five point Likert-type scales (i.e. strongly disagree to strongly agree) and frequency ratings (always to never) were included in the questionnaire. Some question items allowed for multiple response options i.e. What sensory assessments do you normally use? Please tick all that apply.

### ***Procedure***

Participants were recruited in three separate streams. Stream one involved attending a one-day somatosensory workshop intended for paediatric occupational therapists and

physiotherapists at Princess Margaret Hospital for Children in Perth, Western Australia in November 2013. A brief outline of the study was presented, with hard copies of the participant information sheets, informed consent forms and questionnaires provided to all 33 eligible attendees of the workshop. Completed questionnaires were collected at the end of the workshop.

Stream two involved contacting the head therapists of occupational therapy and physiotherapy departments in 52 hospitals within Australia that were identified as having a paediatric department. In addition to this, ten organisations and services were also identified as having therapists that met the inclusion criteria. Hospitals (both public and private) and organisations in metropolitan, regional and rural areas of Australia were included.

Between March and June 2014, the head therapists of occupational therapy and physiotherapy departments or relevant therapists working in paediatric departments were contacted via phone and provided with a verbal outline of the study. If therapists expressed interest and deemed the study relevant to their department, additional information and the link to the online questionnaire was provided via email. A total of 179 emails were sent to paediatric occupational therapists and physiotherapists around Australia, most of whom circulated the questionnaire within their departments and to other relevant therapists.

Stream three involved a brief outline of the study presented to attendees at the monthly Developmental Occupational Therapy (DOT; WA) Inc. meeting. DOT; WA Inc. is affiliated with the Western Australia Occupational Therapy Association and facilitates the Paediatric Interest Group. Convenience sampling was utilised and participant information sheets and the link to the online questionnaire was circulated via email to their member base of approximately 300 paediatric occupational therapists within Western Australia.

### ***Data analysis***

The online responses of the questionnaire were extracted from the Qualtrics survey platform (Qualtrics, 2013). Both online and hard copy responses were manually entered into IBM SPSS Version 20 software (SPSS Statistics, 2010) for cleaning, coding and analysis. Frequencies were calculated for ordinal and nominal data.

Responses that appeared in questionnaire sections titled ‘other’ and qualitative responses were reviewed by the research team using content analysis for common themes and relevant information included in the results. Not all questionnaire items sum to 100% as participants were able to select more than one response to some questions.

### ***Ethical considerations***

Participants that completed the hard copy of the questionnaire gave informed written consent to participate. Consent to participate in the study was assumed by completion and submission of the online questionnaire. Ethical approval was obtained from the Human Ethics Research Committee at Curtin University (approval number OTSW-12-2013).

### **Results**

A total of 135 questionnaires were completed and returned. Stream one yielded 26 responses out of a possible 33 therapists who attended the workshop, while an additional 109 responses were obtained from stream two and three combined. Table 1 illustrates the demographic characteristics of the participants. Eighty-nine (63.7%) of the questionnaires were completed by occupational therapists and 49 (36.3%) completed by physiotherapists. The majority of the sample was female (97.8%). Most of the respondents were from Western Australia (n = 69, 51.5%), and New South Wales (n = 37, 27.4%). Participants from the two aforementioned states made up 78.5% of the sample. Most therapists worked in the metropolitan areas of Australia (n = 96, 72.2%), followed by regional (n = 28, 21.2), then rural (n = 6, 4.5%), and most worked in the private sector (n = 103, 76.3%). Therapists worked an average of 32 (SD = 9.5) hours per week.

Table 1 Demographic information of participants (n = 135)

Characteristic	Variable	
	N	%
	Gender	
Female	132	97.8
Male	3	2.2

Characteristic	Variable	
	N	%
Discipline		
Occupational Therapy	86	63.7
Physiotherapy	49	36.3
Highest degree level		
Bachelor degree	94	69.6
Entry level masters	9	6.7
Postgraduate diploma/certificate	18	13.3
Post graduate masters	12	8.9
Honours	2	1.5
Years of clinical experience working in paediatrics		
1-2	19	14.1
3-5	41	30.4
6-10	30	22.2
>10	45	33.3
Population area of practice		
Metropolitan	96	72.2
Regional	28	21.2
Rural	6	4.5
Remote	2	1.5
Sector of work		
Private	103	76.3
Public	29	21.5
Both	3	2.2
Setting		
Acute care hospital	25	18.5
Rehabilitation ward within acute hospital	6	4.4
Outpatient (hospital-based)	14	10.4
Rehabilitation in the home	2	1.5
Community health centre	36	26.7
Community rehabilitation centre	8	5.9
Private practice	12	8.9
Non-government organisation	14	10.4
Government organisation	2	1.5

Characteristic	Variable	
	N	%
Community not specified	11	8.1
Other	5	3.7

### ***Somatosensory assessment***

Most occupational therapists (n = 79, 91.8%) and physiotherapists (n = 44, 89.7%) reported that they currently assess for somatosensory loss in children with neurological disorders. Of these, a smaller proportion of occupational therapists (n = 25, 31.6%) and physiotherapists (n = 29, 65.9%) reported that they “routinely” assessed somatosensation. CP was the most common neurological disorder reported by therapists (n = 86, 69.9%), followed by acquired brain injury (n = 45, 36.6%).

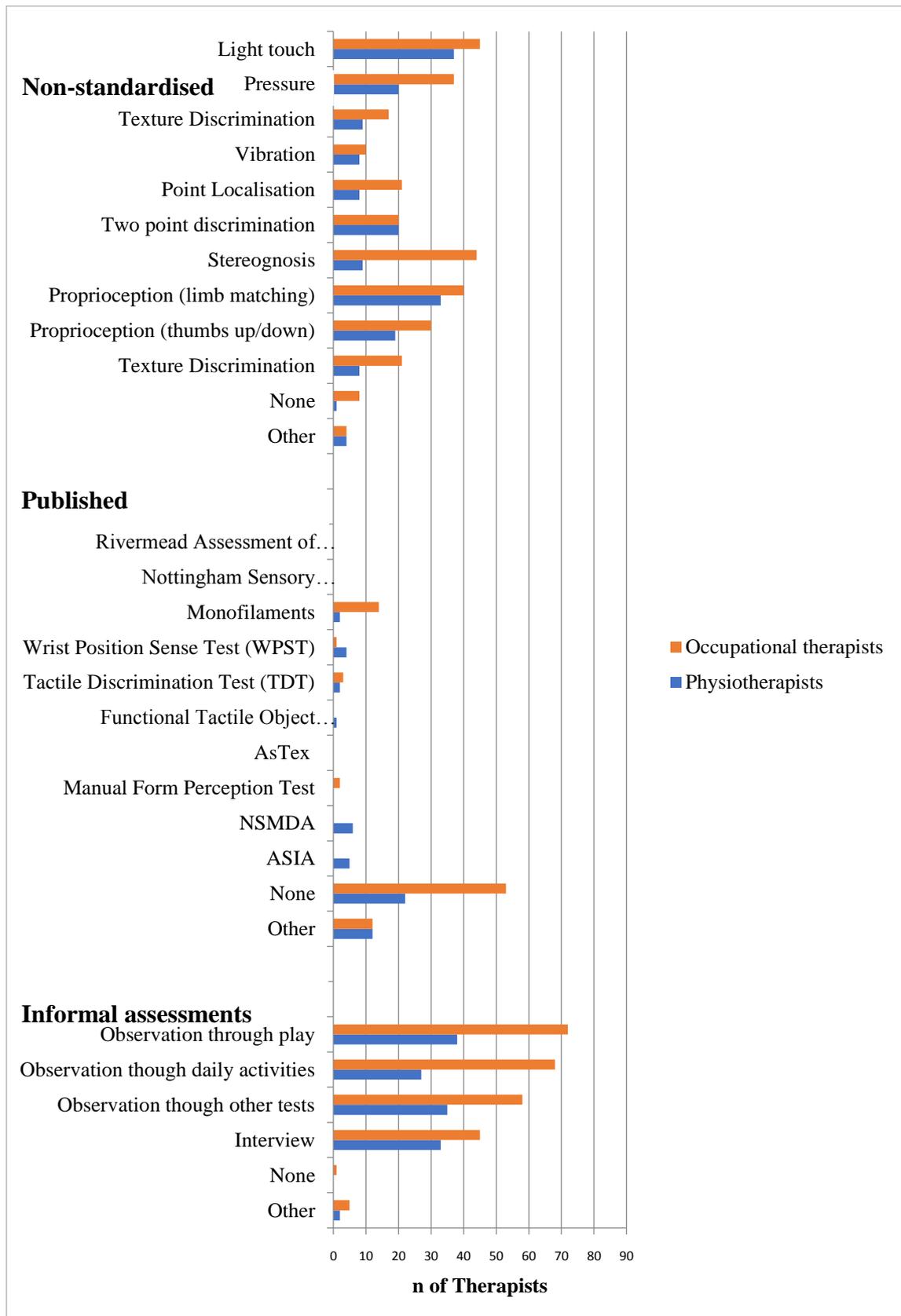
Therapists typically saw between one and two children a week with somatosensory impairments, and tended to assess for somatosensory capacity on admission (n = 75, 62.5%) or when it was clinically indicated (n = 34, 28%). The majority of therapists allocate five (n = 39, 32.0%) or ten (n = 44, 36.1%) minutes to somatosensory assessment, with a proportion of therapists indicating no time at all allocated to somatosensory assessment (n = 20, 16.4%). Sixty-four therapists (61.0%) indicated they felt their allocated time for somatosensory assessment was sufficient. The most common reasons for conducting a somatosensory assessment were to: assist with treatment planning (n = 113, 93.4%), diagnosis (n = 67, 55.4%), for educating the patient, family and other staff members (n = 64, 52.9%) and to review progress (n = 56, 46.3%). A smaller proportion of therapists conducted somatosensory assessment in the discharge planning phase (n = 19, 15.7%). Although both occupational therapists (n = 63, 88.7%) and physiotherapists (n = 32, 74.4%) conducted somatosensory assessments on the upper limb, physiotherapists most commonly assessed the lower limbs (n = 40, 93%).

### ***Main types of somatosensory assessments used in clinical practice***

Participants were asked to nominate the non-standardised, informal and published assessments they used to assess somatosensation. As shown in Figure 1, respondents more frequently used non-standardised and/or informal assessments than published assessments. The most commonly used non-standardised assessment was light touch (n = 82, 66.7%), followed by proprioception (limb matching) (n = 73, 59.3%). The other methods of informal assessment used by therapists included observation

through play (n = 110, 89.4%), observation through daily activities (n = 95, 77.2%) and observation in other tests (n = 93, 75.6%). The use of three or four methods of informal assessment was common (n = 90, 73.1%) across both disciplines. The SWMs (Weinstein, 1993), or Weinstein Enhanced Sensory Test (WEST) hand monofilaments (Weinstein, 1993), were the most commonly used published assessment by all therapists (n = 16, 13.8%) to measure tactile registration. In addition to the somatosensory assessments listed on the questionnaire, physiotherapists indicated the use of the NSMDA (Burns, 1992) (n = 6, 14.6%) and American Spinal Injury Assessment (ASIA) (Maynard et al., 1997) (n = 5, 12.2%). A large proportion of occupational therapists (n = 53, 70.1%) and physiotherapists (n = 28, 68.2%) reported not using any published assessments.

Figure 1 Type and frequency of somatosensory assessments used by therapists



### ***Perceptions of current somatosensory assessments***

The majority of occupational therapists (n = 64, 82.1%) and physiotherapists (n = 38, 86.3%) considered assessing somatosensation in children with neurological disorders to be 'important' to 'very important'. Despite acknowledging the importance of somatosensation assessment, many occupational therapists (n = 39, 52.0%) and physiotherapists (n = 16, 32.3%) indicated they were 'not confident at all' to 'somewhat confident' in their ability to detect somatosensory loss. Only a small proportion of occupational therapists (n = 7, 8.8%) and physiotherapists (n = 7, 15.9%) reported confidence in their ability to measure degree of somatosensory loss for this population. Overall, more than half of the occupational therapists (n = 51, 66.2%) and physiotherapists (n = 23, 58.1%) reported they were 'somewhat' to 'not at all' satisfied with the somatosensory assessments they currently use in clinical practice. Across both disciplines, 64.2% (n = 79) believe there is a need to change the current method of assessing somatosensation in children with neurological disorders.

### **Discussion**

This study identified therapists' current practice and perspectives within Australia related to the assessment of somatosensation in children with neurological disorders. Occupational therapists and physiotherapists acknowledged the importance of assessing somatosensation for these children. This acknowledgement is vital as research indicates that a large proportion of children with CP experience impairments in somatosensation (Auld et al, 2012b; Cooper et al., 1995; Van Heest et al., 1993), with the most commonly reported areas affected being tactile object recognition, two-point discrimination and limb position sense (Auld et al., 2012a; Auld et al., 2012b; Curry & Exner, 1988; McLean, Taylor, Blair, Valentine, Carey, & Elliott, 2017b).

The findings of this current study are consistent with the findings from Pumpa and colleagues (2015) who found more than 70% of therapists were not using standardized assessments with stroke survivors. They are also consistent with the recent findings of Auld and Johnston (2016), who examined the use of assessments for the somatosensory domain of tactile registration among paediatric therapists. They reported that paediatric occupational therapists and physiotherapists might not be conducting

tactile assessment methods according to evidence-based recommendations (Auld and Johnston, 2016).

Measures of tactile registration, i.e. the SWM's or WEST hand monofilaments, were the most frequently reported published assessments used by therapists in the current study. Auld and colleagues (2012b) explored the properties of the SWMs as an assessment of tactile registration in children with CP. They reported excellent test-retest reliability and clinical utility, and acceptable test-retest agreement in children with unilateral CP aged 8 to 18 (Auld et al., 2012b). However, the monofilaments alone only measure one aspect of tactile function (tactile registration), thereby providing limited information to inform treatment planning or predict rehabilitation outcomes if not used in conjunction with other tactile perception assessments (Auld et al., 2012b) or other measures of somatosensory function, such as proprioception and haptic objects recognition (Carey, 2015; McLean et al., 2017b).

Light touch and proprioception were the non-standardised assessments most commonly used by therapists in the current study. This is consistent with findings of non-standardised somatosensory assessment used for adults post stroke (Pumpa et al., 2015; Doyle, Bennett, & Gustafsson, 2013; Winward, Halligan, & Wade, 1999). The use of these subjective assessments however can result in a decrease in measurement accuracy as well as increase the risk examiner of bias relative to quantitative and standardized measures (Bentzel, 2008). Additionally, the use of informal assessment such as observation through play, daily activities and/or through observation of performance during other assessment tools was frequently reported in this current study. Although clinical observation is important, using that alone can make it difficult to monitor patient progress and change over time in response to intervention.

Therapists in the current study also reported they lacked confidence in their ability to detect and measure the degree of somatosensory loss in their usual caseload. Low levels of confidence are reflected despite the largest proportion of therapists having 10 or more years of experience in paediatrics. In contrast, 72.7% of therapists working with adults post stroke reported they were moderately to very confident in their ability to detect sensory loss using the available somatosensory

assessments (Pumpa et al., 2015). The difference in confidence levels among therapists could be reflective of a possible increase in awareness of the presence of somatosensory impairment in adults post stroke, in line with increased evidence base and the introduction of clinical guidelines, which are yet to be established for a paediatric population. Despite this, therapists across both the adult and paediatric populations indicated an overall lack of satisfaction with the available somatosensory assessments for use with their respective clinical populations.

Additionally, therapists in the current study reported satisfaction with the time they allocated for use of somatosensory assessments, despite the majority (68.1%) completing the assessment within 5 or 10 minutes. This warrants further investigation into the use of comprehensive somatosensory assessments for use with children with neurological disorders. Measures of registration such as the SWMs may only take 5 to 10 minutes for administration and interpretation, however a more comprehensive functional measure that generates more meaningful data may take a lot longer.

As with all survey methodologies, this study has its limitations. The number of occupational therapists and physiotherapists working in paediatrics in Australia is unknown and therefore the number of eligible therapists that could have participated in this study cannot be determined. As a consequence, the calculation of return rates or comparison between responder and non-responders could not be determined. In addition to this, the majority of respondents were female and therefore results may not be representative of the male therapist population. Despite known limitations, this study had a large response sample of 135 therapists representing all Australian states, apart from the Northern Territory. This included metropolitan, regional, rural and remote areas, and therapists working in private and public sectors. Variance existed in the years of clinical experience and degrees obtained among therapists. As a result of the diversity across both disciplines, the information obtained from this study is useful to help understand clinical practice and therapist's perceptions of somatosensory assessment for paediatric populations.

The overall aim of this study was to identify current practice and therapist's perspectives related to somatosensory assessment in children with neurological disorders. The results indicate therapists more often use non-standardised and/or

informal assessments than assessments with standardised published administration manuals or instructions. To our knowledge, there is currently no standardised somatosensory tool for use with children with neurological disorders that incorporates touch, limb position sense and haptic object recognition, which are the domains recommended for the assessment to be a comprehensive measure of somatosensation.

The study of interventions designed to promote neuroplastic changes in somatosensation is a growing field of research (Aisen et al., 2011; Anderson, Spencer-Smith, & Wood, 2011; Carey, Macdonell, & Matyas, 2011). The concept of neural plasticity has guided somatosensory intervention for adults post stroke, with specific interventions demonstrating the ability to increase cortical activation of the brain as measured by functional magnetic resonance imaging (fMRI) (Carey et al., 2016). Likewise, children with CP have the potential to respond in a similar way (Wittenberg, 2009). There is emerging evidence to support the efficacy of somatosensory discrimination interventions to improve body position sense for children with spastic hemiplegic CP aged 6 to 15 years (McLean et al., 2017a). Preliminary findings indicate improvements in goal performance, proprioception, and bimanual hand use (McLean et al., 2017a). A quality comprehensive assessment of somatosensation is crucial to the ongoing development of somatosensory discrimination interventions. Such an assessment could be used to determine an individual's baseline function and monitor changes over time in response to somatosensory discrimination intervention.

Future research should be directed towards the development of a comprehensive and quantitative tool to measure somatosensation in children, with consideration made for the various types, severity and associated impairments children with neurological disorders experience. Additionally, effort should be made to translate the evidence we do know about somatosensory impairment and assessment for these children into clinical practice. In 2011 Auld and colleagues made recommendations as to the most appropriate combination of measures to assess tactile registration and perception in children with CP. The outcomes of this clinimetric review recommended the use of the SWM, the Disk-Criminator as well as Single Point Localisation and Double Simultaneous, which are both subsets of the NSMDA. While the SWM was the most commonly used published assessment in this current study, the NSMDA was

under-utilised with only six physiotherapists indicating use of this assessment. This suggests this research has not yet been fully translated into clinical practice. The translation of knowledge from research into clinical practice is vital and occupational therapists and physiotherapists have a responsibility to seek and integrate this new knowledge to ensure the best possible care is provided to patients (Metzler & Metz, 2010).

Active collaboration between therapists as end users, and researchers is needed to ensure the transfer of new knowledge (Metzler & Metz, 2010). This collaboration can occur through researchers identifying knowledge users (i.e. therapists), offering educational outreach through interactive groups or in house clinical research symposiums (Barwick & Heiden, 2016). Departmental managers can assist in this process by ensuring clinical performance feedback for evidence-based practices, and continuing quality improvement activities within departments (Barwick & Heiden, 2016). The results of this current study, along with that recently completed by Auld & Johnston (2016), have established a baseline of what therapists working with children with neurological disorders currently know and are using clinically for the assessment of somatosensation. One of the long-term goals for somatosensory measurement and treatment should be the development of an assessment approach that can inclusively measure all domains of somatosensation and which takes into consideration the facilitators and barriers identified to aid successful clinical implementation (Auld & Johnson, 2016). This study highlights that effort needs to be directed towards effectively disseminating what we currently know and what is readily available for therapists to utilise in the meantime to ensure the assessment of somatosensation is not being overlooked for this population

## **Conclusion**

There is an emerging awareness of the important role that somatosensation plays in the functional outcomes of children with neurological disorders (Auld et al., 2011; McLean et al., 2017a). Deficits in somatosensation are known to exacerbate motor impairments, potentially impacting on a child's occupational performance and participation in the home, school and wider community (Boyd et al., 2010). Valid and reliable assessment tools are limited for children with neurological disorders and this study has highlighted that although therapists understand the importance of somatosensation they are not confident with current somatosensory measures.

Gathering knowledge of current practice related to the assessment of somatosensation in children with neurological disorders will help guide clinical knowledge towards best practice. This study has highlighted the need for more standardised, reliable, and valid methods of assessing somatosensation in children with CP and other neurological disorders. It also highlights the need to systematically translate evidence-based research on measurement of somatosensation in children with CP into clinical practice.

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## **APPENDIX D      Fabric Matching Test**

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Honours student Hannah Overheu accounted for 85 per cent of the intellectual property associated with the final manuscript, collectively Susan Taylor and the other authors accounted for 15 per cent.

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### **The Fabric Matching Test: reliability for use as a measure of texture discrimination in children**

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## **Abstract**

**Introduction:** Currently there are limited measures in the field of rehabilitation to evaluate a child's tactile discrimination. The primary aim of this research was to test the reliability of the Fabric Matching Test (FMT) in typically developing (TD) children. Secondly, we wanted to determine if there was a relationship between age or handedness, and texture discrimination capacity.

**Methods:** A test-retest design was employed to examine inter-rater and intrarater reliability. Thirty TD children were recruited (aged 6y 1m - 15y 9m; mean age 10y; 14 boys). The FMT is a measure of tactile discrimination and consists of two identical fabric disks, each with ten standardised fabrics. Participants are required to match fabrics using discriminative touch.

**Results:** Intrarater reliability; 81.2% of responses were either identical, or differed by 1 fabric or less. Inter-rater reliability; there was an 84.2% agreement with the difference being within 1 fabric. There was no significant association between age, handedness or discrimination capacity of TD children.

**Conclusion:** Inter and intrarater reliability of the FMT has been supported. This study contributes to the study of disability and rehabilitation by establishing the preliminary psychometric properties for a novel assessment tool for children with neurological conditions such as cerebral palsy.



## **Introduction**

Somatosensation and motor performance are integral to a child's successful development (Barker, 2008; Carey, Lamp, & Turville, 2016; Lundy-Ekman, 2013; Seeley, Vanputte, Regan, & Russo, 2011; Zaporozhets, 2002). Sensation is the process where internal and external sensory stimuli are registered and understood through one or more of the body's sensory systems (Dunn et al., 2013; Keith, Elliott, Anderson, & Novak, 2002; Seeley et al., 2011). This process allows us to understand our body in its environment (Auld, Boyd, Moseley, & Johnston, 2011). Somatosensations, which include our ability to sense pressure, pain, vibration, proprioception and touch, assist us to interpret and interact with our surroundings (Doherty & Hughes, 2009).

The process of interpreting and perceiving somatosensory information begins prenatally and extends into adolescence (Piek, 2012). As a child's nervous system develops, so too does their somatosensory system, and their ability to interpret and perceive somatosensory information (Lundy-Ekman, 2013; Riquelme & Montoya, 2010). The perception of somatosensory information, in turn impacts our motor system and motor function (Keizer, Fael, Wierda, & van Wijhe, 2008; Frey et al., 2011). As a child gains mastery over their upper limb movements, their capacity to interact with their environment increases, which further refines their ability to understand and utilise somatosensory information (Case-Smith, 2006). The interaction between these somatosensory and motor systems is a complex synergistic relationship. An impairment in any somatosensory modality can alter a child's sensory experiences, greatly impacting their development and ability to participate in meaningful activities (Lane & Bundy, 2012; Zaporozhets, 2002). Tactile discrimination (ability to differentiate textures by touch) has been linked to hand function and is necessary for precision grip and isolated finger movements during fine motor activities (Bleyenheuft & Gordon, 2013; Carey, Oke, & Matyas, 1997; Ebied, Kemp, & Frostick, 2004; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2008). The processing of tactile information can be altered as a result of primary neonatal or antenatal brain injury such as cerebral palsy (CP) (Krumlinde-Sundholm & Eliasson, 2002; Rosenbaum, 2003).

Tactile impairments of the upper limb affect between 42% to 97% of children with CP (Koman, Smith, & Shilt, 2004; Yekutieli, Jariwala, & Stretch, 2004). There is no gold standard method for assessing tactile discrimination impairments for children with CP (Auld et al., 2011; Connell & Tyson, 2012). Instead, studies use a variety of tests to measure tactile abilities (Overheu, Carey, Elliott, Girdler, & Taylor, 2014; Taylor et al., 2016). There appears to be no consensus regarding which tests are the most appropriate measures of tactile discrimination. Many of the existing tests were found to rely on experimental animal models or were limited in terms of standardised testing procedures or published clinimetric properties supporting use with children (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Wingert et al., 2008). Paediatric assessments with robust psychometric properties, are needed to assist in identifying tactile discrimination impairments and detect change during the rehabilitation process (Fess, 2002; Mattingly, 2008; Portney & Watkins, 2009).

Texture discrimination tests are thought to be functional tests of touch ability (Carey, 1995). The Fabric Matching Test (FMT) (Carey & Matyas, 2005) is one such test of texture discrimination and it is currently being used in the adult stroke population with robust and reliable results. According to the CONsensus-based Standards for the selection of health Measurement INSTRUMENTS (COSMIN) checklist (Mokkink et al., 2010), which was developed to bring consensus for criteria of good measurement properties of health related instruments. Test-retest reliability is included as a key component in the taxonomy of measurement properties (Figure 1). The taxonomy summarises the important factors to consider when selecting a health measurement and can also be used to design robust research testing the measurement properties of an instrument.

Figure 1. The COSMIN Taxonomy of Measurement properties



Figure 1. Originally published in “International consensus on taxonomy, terminology, and definitions of measurement properties: results of the COSMIN study,” by L. B. Mokkink, C. B. Terwee, D. L. Patrick, J. Alonso, P. W. Stratford, D. L. Knol, L. M. Bouter, H. C. W. de Vet, 2010, Journal of Clinical Epidemiology, 63, p.741.

Clinically, the most commonly used tactile assessments include calibrated aesthesiometers such as the Semmes Weinstein monofilaments (Weinstein, 1993) or the Von Frey filaments (Keizer et al., 2008). These are generally used to test for tactile registration (Auld et al., 2011; Cooper et al., 1995). Some studies have demonstrated the psychometric properties of the Semmes Weinstein monofilaments in paediatric populations (Krumlinde-Sundholm & Eliasson, 2002). Auld et al. (2012a) evaluated the test re-test reliability of the Semmes Weinstein monofilaments

for children with CP and reported excellent reliability (Intraclass Correlation Coefficient - ICC = 0.96 for the impaired hand; ICC = 0.90 for the unimpaired hand). However, testing procedures appeared to be flawed due to limitations in standardising the force applied by various testers (Carey, 1995; Riquelme & Montoya, 2010; Riquelme et al., 2011). Cooper et al. (1995) found children to be distractible during assessment, particularly when using the Semmes Weinstein monofilaments. This could be because the test doesn't require active participation from the child hindering engagement in the assessment process. Like adults, children are more likely to engage in an assessment if they can attribute meaning to the assessment (Ziviani, 2012).

Tactile discrimination measures such as the Disk-Criminator (MacKinnon & Dellon, 1985), are reliable and sensitive enough to discern tactile deficits in a paediatric population (Auld et al., 2012a; Krumlinde-Sundholm & Eliasson, 2002). However, the Semmes Weinstein monofilaments and two-point discrimination tests, do not necessarily correlate well with hand function (Carey, 1995). The AsTex® (Miller et al., 2009) and the Van Boven Phillips gratings (Vanboven & Johnson, 1994) have shown greater sensitivity in detecting spatial cues than two-point discrimination tests (Auld et al., 2012a; Sanger & Kukke, 2007). Though, both demonstrated poor sensitivity in detecting deficits in children with CP (Auld et al., 2012a; Wingert et al., 2008). The available tests of tactile sensibility do not satisfy the criteria of assessing functional tactile ability (touch perception using an active moving hand) (Carey, 1995). These tests lack a relationship to functionality, which is important when working with children in order to engage, motivate and maintain their attention in the assessment and intervention process (Ziviani, 2012).

Robust and reliable tactile discrimination tests, which measure the dynamic aspects of the tactile system are important for monitoring change in touch discrimination outcomes and potential impact on paediatric functional outcomes (Carey et al., 1997). The primary aim of this research was to test the inter and intrarater reliability of the Fabric Matching Test (FMT) (Carey & Matyas, 2005) in TD children aged 6 to 15 years living in Perth, Western Australia. We hypothesised that the FMT would have a greater than 75% inter and intrarater reliability. Our secondary aim was to determine if there was a relationship between age, handedness or texture

discrimination ability of TD children. We predicted scores on the FMT in children 6 to 8 years would be significantly lower, than scores for children aged 9 to 15 years. We also aimed to explore any associations between the different graded textures used in the FMT.

## **Methods**

### ***Study design***

A test-retest reliability design was selected based on the COSMIN (Mokkink et al., 2010) to investigate inter and intrarater reliability of the FMT. The FMT was administered by the same trained rater at two different time-points (2-3 weeks apart), to the same children to evaluate test-retest reliability (Vaz, Falkmer, Passmore, Parsons, & Andreou, 2013). Inter-rater reliability was determined through administration of the FMT by two separate raters at one time-point.

### ***Setting***

Children were recruited through convenience sampling from the Perth metropolitan area. Recruitment and data collection occurred between October 2014 and May 2015. Data collection occurred in the homes of each participant, with parents present and follow up assessment occurred two to three weeks later.

### ***Participants***

Children were included if they were typically developing, as self-reported by parents. Children with a traumatic physical injury, known physical, neurological or developmental disorders or who were unable to understand spoken English or follow simple instructions were excluded. The age distribution was selected based on tactile perception performance, which matures at approximately 6 years and plateaus at 15 years (Benton, Hamsher, Varney, & Spreen, 1983; Bleyenheuft, Cols, Arnould, & Thonnard, 2006; Viel, Vaugoyeau, & Assaiante, 2009).

### ***Study size***

The sample size of  $N = 30$  was deemed an adequate number for a preliminary test-retest design based on similar studies (Portney & Watkins, 2009).

## Main Outcome Measurement

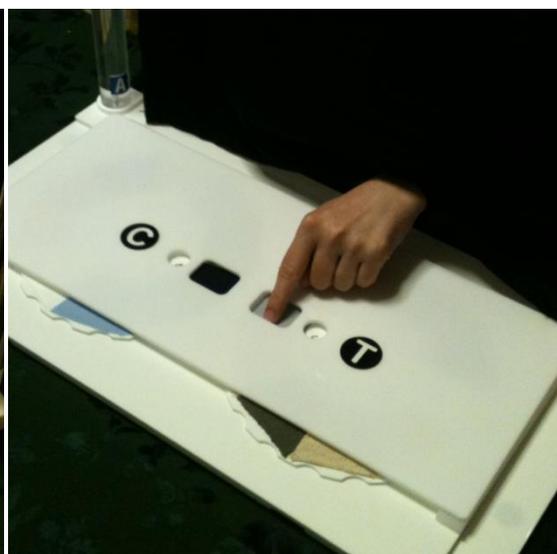
### *Fabric Matching Test*

Texture discrimination ability was measured using the FMT (Carey & Matyas, 2005). The FMT is a measure of tactile discrimination which has been developed by Carey and colleagues (2005) for an adult stroke population. This measure was found to have high test-retest reliability ( $r = .85$  to  $.92$ ) and good discriminative validity in an adult stroke population (Carey & Matyas, 2005). The FMT consists of two identical fabric disks, covered in ten standard cotton-based fabric surfaces (Figure 2 and Figure 3) ranked 1 to 10 smoothest to roughest (Macháčková, Vyskotová, Opavský, & Sochorová, 2010). Participants were required to match two fabrics as closely as possible using discriminative touch. The instructions of the FMT were modified as part of this study to suit a paediatric population and the changes were pilot tested with five TD school-aged children (6-11 years old) prior to data collection. The test is scored using the Spearman rank correlation to measure the individual's accuracy of matching for the rank ordered fabrics.

Figure 1 Administration set-up of the Fabric Matching Test



Figure 2 Image of an individual exploring a texture of the Fabric Matching test



### *Procedure.*

Data were collected by two raters at the participants' homes with their parent or guardian present. The children's age, gender and hand preference were recorded prior to commencing data collection. FMT data were collected in accordance with

the FMT administration protocol and paediatric modified instructions. The FMT required approximately 10-15 minutes to complete. Responses were recorded on a test form. The child was not given any indication of accuracy of responses.

### ***Data analysis***

A regression model was used to examine any possible association between multiple responses by the same participant (as a random effect) for intra and inter-rater reliability. Inter and intrarater agreement was deemed good to high if agreement of overall mean FMT scores obtained by the same rater across two time-points or between different raters at one time-point were greater than 75%.

Descriptive statistics were used to compare responses between younger and older children (6-8 and 9-15 years) and to summarise which fabrics were associated with the most error. The regression model also examined the influence on the error of the hand (left/right), and whether the dominant hand was used (Yes/No). Data were analysed using the SAS Package 9.2 (SAS Institute, 2008) and following convention, a  $p$ -value  $<0.05$  was taken to indicate a statistically significant association in all analyses.

### ***Ethical considerations***

This study was conducted in accordance with the National Health and Medical Research Council (NHMRC) guidelines (Council NHaMR, 2009). Parents were provided with information regarding the purpose of the study and confidentiality procedures and each parent and / or guardian gave written informed consent and all participants gave verbal assent prior to data collection. Ethics approval was obtained from Curtin University Human Research Ethics Committee.

## **Results**

Thirty TD children ( $N = 30$ ), 14 male and 16 female, aged 6 to 15 years (median age 10 years) were recruited. Background characteristics are shown in Table 1. Each participant completed the FMT with each hand on three separate occasions (time-point 1: rater 1, two weeks later, time-point 2: rater 1 and 2). There were 11 participants aged 6 to 7 years, and 19 who were between 8 to 15 years of age. Twenty-four of the participants had right hand dominance.

Table 1 Participant demographics

Variable	Score/Statistic	Total Group (N=30)
Age (years)	Mean	9.6
	SD	3.1
	6-7	11
	8-15	19
Gender	Female	16
	Male	14
Hand Dominance	Right	24
	Left	6

*Note.* SD = Standard Deviation; N = total participant sample.

### ***Reliability***

For intrarater reliability analysis found that 81.2% of responses between the first time-point and second time-point were either identical or incorrect within one fabric or less. The overall mean error in FMT scores for the 30 participants [95% CI] was found to be: -0.11 [-0.23, +0.01]. For inter-rater reliability, there was an 84.2% agreement with the difference being within 1 fabric for FMT results when administered by two different raters. The overall mean difference in FMT scores between raters was: 0.02 [-0.10, 0.14].

### ***Influence of test fabric, age and handedness.***

Time-point 1 data indicated that 86% of all responses were within 1 fabric of the correct answer. The random effects regression model found that the overall mean [95% CI] for the error at baseline was: -0.02 [-0.13, 0.10]. The regression model found no significant association between the change in response and the test fabric ( $p = 0.065$ ), age ( $p = 0.50$ ) or handedness ( $p = 0.42$ ) of the child. A comparison was made between the measurements observed by both raters to determine consistency. No association was found between the differences and roughness of the test fabric ( $p = 0.99$ ) or dominant hand ( $p = 0.57$ ).

### **Discussion**

As evidenced by our results, we confirmed our first hypothesis, that the FMT would demonstrate a greater than 75% inter and intrarater reliability. This is the first preliminary reliability study using the FMT with a paediatric population and thus the findings are important as it is the initial step in developing this tool for clinical use.

The present findings support a previous study, where the FMT demonstrated high test-retest reliability with adult stroke survivors (Carey & Matyas, 2005).

Our secondary objective was to establish if there might be a relationship between age or handedness, and the texture discrimination ability of TD children. Literature related to childhood development reports that somatosensation improves with age from infancy to adolescence (Case-Smith, 2010; Eliasson, 2006; Taylor et al., 2016; Zaporozhets, 2002). However, improved texture discrimination was not associated with age in our data. This was also the case for Sanger and Kukke (2007), who found no significant association between spatial tactile discrimination and age in both TD children and children with CP. In contrast, a study by Dunn et al. (2013), comparing tactile and kinaesthetic performance across five different age groups from 3 to 65 years, reported that somatosensory performance generally improved with age until late adulthood. Similarly, Bleyenheuft et al. (2006) found tactile spatial performance improved with age until 10 to 11 years, and then plateaued. In the current study, we also found no association between hand dominance and tactile discrimination ability, and this reflects results in similar recent studies (Bleyenheuft et al., 2006; Dunn et al., 2013). The current study provides preliminary normative data for the FMT and contributes to the understanding of TD children's tactile profile, which determines the benchmark for comparison to children with neurological disorders such as CP.

The FMT uses commonly encountered fabrics to determine tactile discrimination ability (Carey, 1995). The use of these common fabrics has ecological validity in relation to functional tasks, such as being able to judge the quality of a fabric with our fingers when dressing. Like the process of dressing, the FMT relies on the lateral movement between the fingertip and the textured fabric. Tactile performance has been found to be enhanced when there is tangential movement between the skin and a textured surface (Carey, 1995; Carey et al., 1997; Lederman, 1982). This movement provides a rich source of information to increase tactile performance and discriminative ability (Carey et al., 1997). Performance in age appropriate tasks such as dressing, is often identified by children with disabilities and their parents, as important in facilitating confidence and independence and is

therefore important in goal directed assessment (Henderson, 2006; Lane & Bundy, 2012; Ziviani, 2012).

In a recent clinimetric review Auld et al. (2011) recommended that two phases of tactile function be assessed tactile registration and tactile perception. Tactile perception is the more complex ability of being able to understand, interpret, or give meaning to stimuli (Auld et al., 2011). The FMT is focused on tactile perception by evaluating texture discrimination. While assessment of tactile registration, which is a basic precursor to tactile perception, is recognised as important, Carey (1995) describes the value of assessments that focus on how the tactile information is being perceived and interpreted in the context of functional sensibility. The FMT employs a standardised assessment protocol and standardised graded stimuli, in contrast to other commonly used tactile measures. These commonly used measures were found to have inconsistent administration protocols, limited psychometric evidence for use with a paediatric population or lacked an ecological validity to functional tasks (Auld et al., 2011; Carey, 1995; Connell & Tyson, 2012).

Specific sensory assessments are needed to quantify complex tactile functioning and the deficits that impact on hand function (Auld, Boyd, Moseley, Ware, & Johnston, 2012b). Health care professionals have identified sensory assessment as essential to clinical practice and useful for prognosis of functional ability and these measures need to be clinically useful (Connell & Tyson, 2012; Rosenbaum, 2003). The FMT takes approximately 10-15 minutes to administer, has easy to follow standardised procedures and has the potential for translation into clinical use. With further psychometric testing this quantitative clinical tool, could assist health professionals to understand performance patterns that may be markers of risk, assist in treatment planning, and monitor performance in texture discrimination.

Currently, there is limited available literature that examines impact of tactile deficits on motor function or engagement in daily activities, among children with neurological conditions such as CP (Cooper et al., 1995). Establishing the reliability of the FMT is a crucial step in tool development, with the end goal of being able to understand the impact of a somatosensory impairment on a child's daily life. Future research is needed to further develop the psychometric properties of the FMT

including validity and responsiveness, and to collect normative data from a larger sample of children with which to compare to clinical populations. Future research might also be used to explore the feasibility of the FMT within a clinical population such as CP. Dunn et al. (2015) recommended that touch perception be assessed in children aged 3 to 6 years. Testing the FMT with children younger than 6 years would also be an area for future research to inform early somatosensory intervention programs.

### ***Limitations***

The sample size and spread of ages across participants, though acceptable for a preliminary reliability study, limited further analysis of tactile discrimination abilities related to age. It was difficult to determine the impact of child engagement on results, and this was not taken into consideration when the data was analysed.

### **Conclusion**

Current tests of tactile discrimination lack a relationship to functionality, which is important when working with children to engage, motivate and maintain their attention in the assessment and intervention process. The FMT is a standardised, quantitative measure of a person's ability to discriminate textures. Data from this study contribute to understanding TD children's tactile profile, and thus helps to establish the benchmark for comparison to children with neurological disorders.

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## **APPENDIX E      sense\_assess© *kids* Administration Manual**

The PhD Candidate, Susan Taylor accounted for 75 per cent of the intellectual property associated with the paediatric version of the administration manual. Collectively, the remaining authors contributed 25 per cent.





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# **sense\_assess©kids**

For children and adolescents 6-15 years

## **Administration Manual**

### **1.1**

**In association with:**



National Stroke Research Institute



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# **sense\_assess© kids**

## **Administration Manual**

### **Introduction**

This manual provides a detailed guide to the set up and administration of the sense\_assess© kids for use with children and adolescents. At the back of this manual are appendices to support the use of the sense\_assess© kids.

#### **Purpose:**

The sense\_assess© kids has been developed to measure the sensory capacity of the upper limb in the clinical setting. Its purpose is to provide clinicians with an evidence based assessment tool that will aid accurate and early detection of sensory impairments across somatosensory modalities.

#### **Overview:**

The sense\_assess© kids uses standardised sensory assessment tools, including the 4.56 Semmes Weinstein Monofilament (Semmes, 1960), Tactile Discrimination Test (TDT) (Carey et al., 1997), functional Tactile Object Recognition Test (fTORT) (Carey et al., 2006) and Wrist Position Sense Test (WPST) (Carey et al., 1996) to test the various attributes of sensation, including protective touch sensibility, tactile/texture discrimination, tactile object recognition and proprioception (wrist position sense). Assessors need to be trained in the methods of assessment and accredited.

These tests have been shown empirically to be able to differentiate impaired performance relative to normal performance standards. The sense\_assess© kids provides clinicians with an evidence based tool to assess for sensory impairment of children in the clinical setting.

#### **Use:**

The four sensory tests included in this test battery assess for impairment in various aspects of sensation therefore should be conducted as a battery. However, they can be used separately when required. The sense\_assess© kids is designed to be used with children and adolescents aged 6 to 15 years with neurological impairment such as cerebral palsy or acquired brain injury.

#### **General Guidelines for Set Up prior to testing**

During test administration, the participant is to be seated in a chair with arms, usually opposite the examiner, and with the apparatus set up on a table waist-height to the participant. Care should be taken to support the affected arm of a participant on the table or arm of the chair (possibly with a pillow) if they have paralysis, spasticity or flaccidity of the upper limb. The curtain is used to occlude the participant's view of test stimuli during administration.

#### **Test sequence**

The following test sequence is suggested:

TEST	SET UP	ADMINISTRATION	SCORING	INTERPRETATION
1 Protective Touch Test	2-3min	~5min	~1min	~2min
2 Tactile Discrimination Test (TDT)	2-3min	~12min	~5min	~2min
3 functional Tactile Object Recognition Test (fTORT)	~10min	~12min	~5min	~5min
4 Wrist Position Sense Test (WPST)	~10min	~12min	~5min	~5min

Total administration time per hand: ~ 40 minutes

Please note: scoring times are based on electronic data entry forms completed after administration

## Protective Touch Test (Touch-Test® Sensory Evaluator)

### Description

The purpose of this test is to screen for the ability to detect touch registration in children and adolescents with neurological impairment such as cerebral palsy or acquired brain injury. The test uses the red monofilament (4.56 protective sensation) to screen touch detection at 16 regions on the palmer and dorsal aspects of the hands.

### Equipment

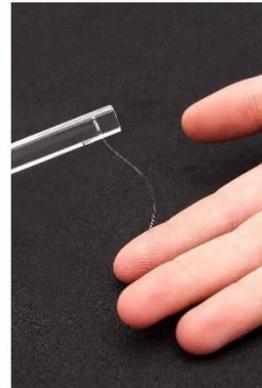
Red, 4.56 monofilament – protective sensation  
Assessment form and pen  
Test board with foam mat and curtain  
Waist height table for participant  
(Booster cushion provided by examiner as required)  
Two chairs (one with armrests for the participant as required)

### Set Up

Testing should be completed in a quiet area to help the participant fully attend to the testing procedure. During test administration, the participant is to be seated in a chair in front of the examiner with hands resting in supine on the foam surface of the test board. The examiner should have the red monofilament (4.56), assessment form and pen at hand.



The 4.56 red monofilament



Applying the monofilament at a 90° angle to form a 'C' shape

### Test Administration

Eight stimuli are applied per hand across palmer and dorsal regions. Regions are based on the common testing sites recommended in the Touch Test® administration manual and consideration of the digits used for functional grasp. The symbol T (Touch) on the test form indicates that a stimulus is provided to the tested hand region. Apply the monofilament at a 90° angle against the skin until it bows to form a 'C' shape, as shown in the figure above. Hold in place for 1.5s and then remove (Touch-Test® Sensory Evaluator, 2002). During the test administration, visual feedback of the stimuli is restricted as the participant's hands are placed on the foam test board using the curtain to occlude vision.



Child with hands in supine being tested during the Protective Touch Test.

### ***Verbal Instructions***

*(Explain the test using the standardised script as provided below)*

**I'm going to use this special touching string (*point to and bend monofilament on own fingertip*) to gently touch your skin and see how well you can feel it. This curtain will be pulled across so you won't see your hands (*briefly demonstrate on self and let child feel sensation on the back of their less affected hand*). I'm going to touch both sides of your hands like this (*demonstrate on self in sequence as per test form*). Your job will be to tell me if you can feel it. Say 'yes' when you feel the string touching you. Ready?**

### Test Form

The test form includes a representative picture of the hand regions tested. Place a circle around trials that have been correctly identified by the participant and a cross through those that are not. Note the number of correct responses to stimuli.

### Scoring

The number of correct responses indicates the participant's protective touch sensibility detection ability at each hand region.

### Interpretation

Based on normative data (Table 1) a score of 15 for the left hand and 16 for the right hand would indicate intact protective touch sensibility (Taylor et al., 2017a). Further testing with the full Touch-Test® 5 piece Hand Kit may be conducted if the participant scores below this.

Table 1. Normative data for children aged 6 to 15 years

Subtest	<8.9 years M (SD)	9 - 11.9years M (SD)	12+ years M (SD)
PTT Tactile registration N=56	n=21	n=18	n=17
Right hand number of correct responses out of a possible six	6(.00)	6(.00)	6(.00)
Left hand number of correct responses out of a possible six	5.9(.22)	6(.00)	6(.00)

Note. M=mean; SD=standard deviation; PTT=Protective Touch Test; N=total sample; n=portion of sample.

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**Protective Touch Test form**  
(Touch-Test® Sensory Evaluator)

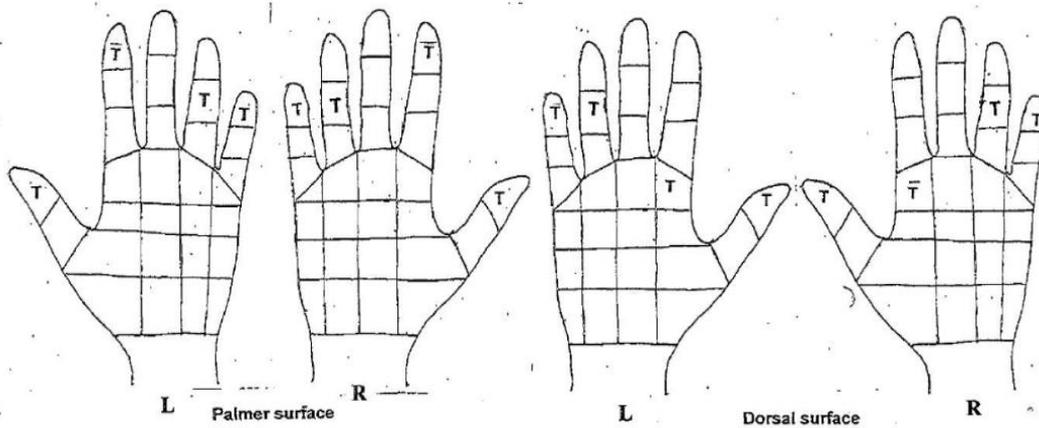
**Scoring** - O = Correct Response  
X = Incorrect Response

Participant name: \_\_\_\_\_

Date: \_\_\_\_\_

Therapist name: \_\_\_\_\_

Hand tested: Left / Right



O = Correct response  
X = Incorrect response

**Comments:**

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## Tactile Discrimination Test (TDT)

(Carey, Oke & Matyas, 1997)

### Description

The Tactile Discrimination Test (TDT) is a quantitative measure of ability to discriminate differences in finely graded textured surfaces using a three-alternative forced choice design. The participant is guided to explore the sets of texture grids using the sense of touch in a standard manner with their index finger and indicate the texture that is different. The test has high retest reliability and normative standards for adults, normative standards for children and adolescents aged 6 to 15 years (Carey et al., 1997; Taylor et al., 2017a) and clinical acceptability for children with cerebral palsy aged 6 to 15 years (Taylor et al., 2017b).

### Equipment

Texture grid plate  
Test board with curtain and guide for the texture grid plate  
Assessment form and pen  
Waist height table for participant  
(Booster cushion provided by examiner as required)  
Two chairs (one with armrests for the participant as required)



Test board, curtain and texture grid plate



Texture grid

### Caution:

Do NOT place the plastic gratings of the texture grid in contact with any moisture or water. Take care when handling the texture grid and test board and do not allow the participant to use their fingernails to feel the grids as it will damage them. The grids may be cleaned by lightly wiping them with an alcohol wipe or dry soft brush if required.

## Set Up

The participant is seated in a chair opposite the examiner. The test board with curtain and guide for the plate is set up on a waist-height table for the participant. The elbow should be supported in a position of approximately 90 degrees flexion. Care should be taken to support the assisting arm/hand of the patient on the table or arm of the chair (possibly with a pillow) if they have paralysis, spasticity or flaccidity of the upper limb. The forearm is aligned with the central lines of the testing board and the hand is placed centrally through the curtain. The examiner checks that the participant's hands are not cold. The patient is required to warm their hands to approximately room temperature before being tested if needed.

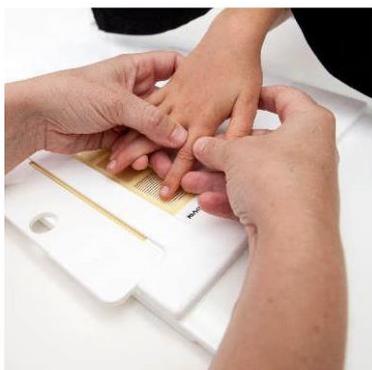
Surface sets are coded regarding size of difference (i.e. 170, 210, 260 or 300), position of surface that is different (i.e. 1, 2, or 3), and whether the *different* surface is larger (>) or less (<) than the comparison surface. Thus, the code 300>2 appearing vertically on the right side of the test grid plate from the examiners view depicts the '300' stimulus difference set, that the surface that is different is the larger/rougher surface ('>') and that it is in position '2' (grids in the first and third positions are the same).

Four surface sets are used and they are presented in pseudo-random order as shown on the test form.

## Test Administration

The examiner allows the participant to feel a set of texture grids, guiding the participant's index finger over the three textures. Three standardised 'swipes' are completed (i.e. left to right, right to left, left to right once more). Ensure that the texture grid is in place in the windowed opening. The examiner should help orient the participant to the task by saying 'first', 'second', and 'third' as he/she moves across each of the surfaces. The participant may revisit the textures before making their choice. Follow the order of presentation as indicated on the test form. Be careful to ensure that the arrow on the test plate matches the examiner's view as indicated on the test form.

The examiner should guide the fingertip across the surfaces in a standard manner (see below). Support the weight of the hand in your two hands and move the finger from side to side across the surfaces using the lateral border of the finger (to minimise extraneous touch). Move the participant's hand and finger as a unit at a constant speed and pressure. It helps to support your elbows on the table to give you stability and achieve a more constant pressure and controlled movement.



Examiner guiding the index finger in a standardised manner



Image portraying the standardised set up of the Tactile Discrimination Test

### Verbal Instructions

*(Explain the test using the standardised script as provided below)*

**The purpose of this activity is to see how well you can feel different textures without looking. The textures look like lines as you can see here** *(point to surface set 300 > 2 and touch each texture slowly)*. **Let's feel them now** *(demonstrate lateral motion over textures left to right for participant saying 'first, second, third')*.

**In each row of 3, two of these textures are the same and only one is different. Can you show me which two are the same? Now can you show me which one is different?**

**The one that is different could be rougher, such as here** *(point to 300 > 2)* **or smoother as in this row here** *(point to 210 < 2)*. **It could be in any position, such as in the middle** *(point to fine texture in 210 < 2)*, **on one side** *(point to rougher texture in 260 > 1)* **or the other** *(point to rough texture in 170 > 3)*. **Some are easy to pick the difference and some are hard.**

**Using this finger** *(point to your own index finger or theirs)* **I will ask you to feel each of these textures without looking. This curtain will be pulled across** *(demonstrate level of occlusion then lift curtain again)*. **I will guide your hand to feel them like this saying 'first, second and third'** *(demonstrate standardised lateral motion over each of the 3 textures in surface set 300>3)*. **Tell me which one you think is different by saying 'first, second or third'. If you're unsure I can help you to feel them again.**

**Do you understand? Let's have a practice first** *(pull curtain across, present 300 < 3 or 300 > 2)* **Which one is different? In subsequent trials examiner may prompt; Which one is different? It may be a rough or fine texture.**

*If child is hesitant to respond to the finer textures, say; What would be your best guess?*

### Test Form

Record the participant's response in the space provided for each trial, as below. The position on the form is determined according to how it is viewed (left to right) by the examiner (ie position 3 on the form matches the stimulus that is on the examiner's right). Note that the correct response is highlighted in the test form. You can use this to double check you are recording the response correctly.

Eg.

1 2 **3**

At the time of testing (or later) score the participant's response as 1= correct or 0= incorrect. The number of correct trials for each test texture is recorded in the score table.

### Scoring

Determine the score by summing the raw test scores from the test form.

Example:

Texture Set	Response	Number correct /3
170	2, 2, 3	1
210	2, 1, 3	1
260	2, 3, 1	2
300	3, 1, 3	3

Determine the area under the curve (AuC) using the 'ready reckoner' table below or the electronic test form. Determine the result by looking under the respective columns and rows for each of the texture sets.

TABLE FOR ESTIMATING AREAS UNDER THE PSYCHOMETRIC FUNCTION (Proportion of possible area)

		0, 1			2			3		
No. correct 1700 set	No. correct 2100 set	0, 1	2	3	0, 1	2	3	0, 1	2	3
		0, 1	0, 1	0.00	2.91	22.35	6.52	21.91	32.36	14.42
	2	0.00	14.32	24.92	8.54	22.31	50.36	19.29	69.36	68.81
	3	3.00	18.04	37.29	10.67	33.68	55.36	26.56	90.13	84.65
2	0, 1	0.00	2.19	20.24	7.09	22.56	30.91	15.73	49.76	49.09
	2	0.08	15.05	52.12	9.08	27.22	44.82	20.07	70.11	70.12
	3	0.11	17.95	64.08	11.21	35.31	98.67	29.45	98.65	100.00
3	0, 1	0.00	2.99	12.98	7.64	23.20	33.61	16.35	51.46	47.09
	2	0.06	15.66	24.68	9.56	27.97	41.71	22.83	73.45	93.04
	3	6.01	17.71	36.98	11.79	42.75	99.97	32.57	99.92	100.00

Note. In the ready reckoner table above the term: 1700 set, 2100 set, 2600 set and 300 set corresponds to the 170, 210, 260 and 300 textures on the texture grid plate.

**Interpretation**

**An Area under the curve (AuC) of 100 = all responses correct, no error**

**6-8 years: AuC <34.6 for the right hand and <31.3 for the left hand**

Impaired tactile discrimination is suggested if the AuC score is *less than* 34.6 (right hand) or 31.3 (left hand) for children and adolescents aged 6-8 years

**9-11 years: AuC <56.0 for the right hand and <51.7 for the left hand**

Impaired tactile discrimination is suggested if the AuC score is *less than* 56.0 (right hand) or 51.7 (left hand) for children and adolescents aged 9-11 years

**12-15 years: AuC <61.3 for the right hand and <61.5 for the left hand**

Impaired tactile discrimination is suggested if the AuC score is *less than* 61.3 (righthand) or 61.5 (left hand) for children and adolescents aged 12-15 years

**References:**

- Taylor, S., Mclean, B., Parsons, R., Carey, L. M., Girdler, S., & Elliott, C. (2017a). Assessment of body sensations in children: age related effects and reliability. Manuscript in preparation.
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**Tactile Discrimination Test form**  
(Carey, Oke & Matyas, 1997)

Participant name: \_\_\_\_\_ Date: \_\_\_\_\_ Therapist name: \_\_\_\_\_

Hand Tested: \_\_\_\_\_  
Left / Right

Trial: 300>2

↓	Response	Score	↑	Response	Score	↓	Response	Score	↑	Response	Score	↓	Response	Score
170>3	1 2 3		260>3	1 2 3		300>2	1 2 3		300>2	1 2 3		170>3	1 2 3	
210<2	1 2 3		300>2	1 2 3		210>2	1 2 3		210<2	1 2 3		210<2	1 2 3	
260>1	1 2 3		210<2	1 2 3		260>1	1 2 3		170>1	1 2 3		260>1	1 2 3	
300>2	1 2 3		170>1	1 2 3		170>3	1 2 3		260>3	1 2 3		300>2	1 2 3	

Comments: (e.g. need for multiple breaks due to short attention)

Score:

Percent spatial increase	Texture Set	Response (raw data in order) - (miss)   (correct)	Number correct/5
13.33	170		
40	210		
73.33	260		
100	300		

Area under the curve: \_\_\_\_\_ (%)

## functional Tactile Object Recognition Test (fTORT)

(Carey, Nankervis, LeBlanc, Harvey, 2006)

### Description

The functional Tactile Object Recognition Test (fTORT) (Carey et al., 2006) is designed to test recognition of objects through the sense of touch. The test contains 14 object sets – the objects are common, everyday objects. Each object set consists of two objects, that differ in one sensory attribute e.g. weight, and a distracter object that differs in more than one sensory attribute e.g. weight and shape. Object sets are displayed visually on the response poster and actual objects are provided to be manipulated. The fTORT has age-adjusted normative standards, high reliability ( $r = .85$  to  $.92$ ) and good discriminative test properties for adults aged 21 to 79 years (Carey et al., 2011; Nankervis, 2004). The test has also demonstrated construct validity and clinical acceptability for children with cerebral palsy aged 6 to 15 years (Taylor et al., 2017b; 2017c) and has normative standards for children aged 6 to 15 years (Taylor et al., 2017a).

### Equipment

Poster of objects  
14 actual test objects to be manipulated  
Test board with foam mat and curtain to occlude vision  
5 display objects – Metal Bowl, Spoon, Full Jar, Paper Card and House Key  
Assessment form and pen  
Waist height table for participant  
(Booster cushion provided by examiner as required)  
Two chairs (one with armrests for the participant as required)  
Stop Watch  
(If digitally recording, follow the instructions in Appendix A at the back of this manual)



Administration set up of the functional Tactile Object Recognition Test

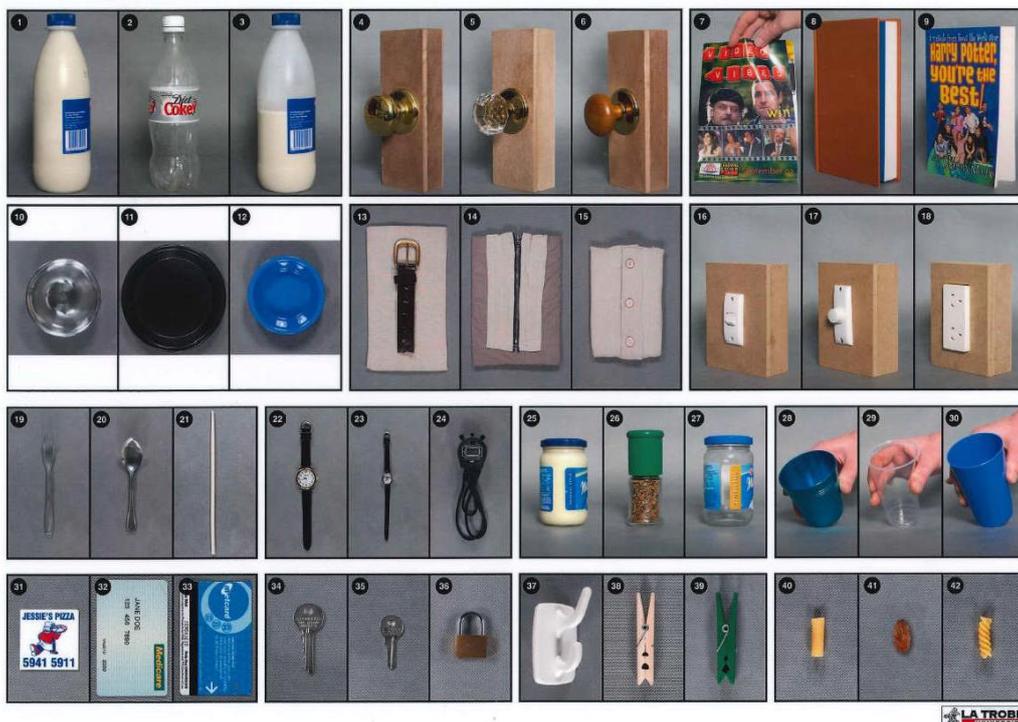


Participant view of standardised administration set up

Table 2. Objects used and corresponding poster number

Sensory attribute	Test Object	Item pair	Item distractor
Weight	1 Full milk bottle	3 Half full milk bottle	2 Empty coke bottle
Temperature	4 Metal doorknob	6 Wooden doorknob	5 Crystal doorknob
Hardness	8 Hardcover book	9 Paperback book	7 Magazine
Temperature	10 Steel bowl	12 Plastic bowl	11 Plastic plate
Part function	14 Zipper	15 Buttons	13 Watch strap
Part function	16 Click switch	17 Turn switch	18 Power point
Shape	20 Spoon	19 Fork	21 Chopstick
Size	23 Small watch	22 Large watch	24 Stop watch
Weight	27 Empty jar	25 Full jar	26 Pepper grinder
Hardness	30 Firm plastic cup	29 Crush plastic cup	28 Aluminum cup
Texture	32 Plastic card	33 Paper card	31 Fridge magnet
Size	34 House key	35 Filing cabinet key	36 Padlock
Texture	38 Wooden peg	39 Plastic peg	37 Plastic hook
Shape	40 Cylinder pasta	42 Spiral pasta	41 Sultana

### Object poster



### Set Up

The examiner sits opposite the participant or to the side of the participant, depending on which arm is being tested (i.e. if the right arm is to be tested, sit on the right side of the participant).

A curtain is placed beside the participant to occlude vision of the test objects. The poster of the test and distracter objects is placed on the table at comfortable viewing distance for the participant. Objects used to help with size calibration (metal bowl, spoon, full jar, paper card and house key) are positioned along the top of the poster, in the same orientation as the object in the poster.

The participant's hand to be tested is placed through the curtain out of view next to the examiner, with their palm facing up and their arm resting on the test board. A foam mat is placed under the participant's arm to minimise noise if the object is dropped. The participant is asked to put on the earmuffs (or use disposable ear plugs) to ensure that they receive no auditory clues from the test items.

During testing, the actual test objects should be kept out of the participant's vision. The subject may need to be encouraged to look at all the photographs before choosing. A stopwatch, test form and pen should be at hand.



Test object being manipulated



Five objects placed in view of the participant for size calibration

### Test Administration

Once the instructions below have been explained to the participant, instruct participant to put on earmuffs. During each trial, one object from each object set (14 in total) is presented. Place the object in the participant's hand, as outlined below. Participants are then asked to identify the object from the choices provided on the poster (they may state the object name, the object number or point to the object on the poster).

Record the item number identified, score and time taken on the test form. Take note also of the haptic exploratory procedures used by the participant and the level of assistance required (motor enhanced tactile exploration by therapist) and record these in the comments column of the test form.

### Haptic exploratory Procedures

The use of optimal haptic exploratory procedures (EPs) used by typically developing individuals contributes to their ability to identify objects through touch (Lederman & Klatzky, 1987). Observe the participant's ability to use the various optimal haptic EPs to identify the objects given to them during the fTORT. The optimal haptic EP for each object is listed across the bottom of the test form. Please see Appendix B at the back of this manual for normative data relating to haptic EPs used by children without disabilities aged 6 to 15 years for each of the test objects (Taylor et al., 2016a). Use of EPs that differ to the optimal EPs as shown below (Table 3.) or the normative data in Appendix B may indicate a need for training in the use of optimal haptic EPs.

Table 3. Optimal haptic exploratory procedures per sensory attribute

Associated Sensory Attribute	Exploratory Procedure	Action
Size	Enclosure (En) 	The hand and fingers wrap closely around the object surface
Temperature	Static Contact (SC) 	The skin is placed in contact with the object surface without movement.
Weight	Unsupported Holding (US) 	The hand supports the weight of the object without any other forms of external support. This involves moving the hand up and down in space.
Hardness	Pressure (Pr) 	Exerting force on the surface of the object using the hand or fingers.
Texture	Lateral Motion (LM) 	Movement of the hand or fingers laterally over the surface of the object
Shape	Contour Following (CF) 	Movement of the hand and fingers over the shape and edges of the object.
Function/ Motion	Function (Fu) 	Using the object to perform the function it is made to do.
	Part Motion (PM) 	Manipulating the part of the object to perform the action it was made to do.

### Presentation of objects

The participant's arm should be placed in supination with palm facing upwards behind the curtain. For larger objects, e.g., the jar, cup or bowl, present objects into the palm of the participant's hand, enclosing the participant's fingers around the object or around the most prominent features of the object, e.g., the rim of a bowl (Refer to figures A and B). For smaller objects, such as the keys or pasta, place the object between the thumb and first 2 fingers to allow a tripod grasp around the object, allowing the participant to explore the object freely (Refer to figure C). For objects that require function, e.g., zippers, buttons or switches, place the object on the table and run the participant's fingers over the most prominent feature of the object, e.g., the pull tab of the zipper. (Refer to figure D).

If the participant is unable to manipulate an object independently, use two hands to assist the participant to explore the most prominent attributes of the object using the optimal EPs relative to that object.

Once the participant has provided a response, remove the object from the participant's hand.

### Verbal Instructions

*(Explain the test using the standardised script as provided below)*

**In this activity, you will be asked to tell me the everyday objects I put in your hand to feel, without looking. This curtain will be pulled across.**

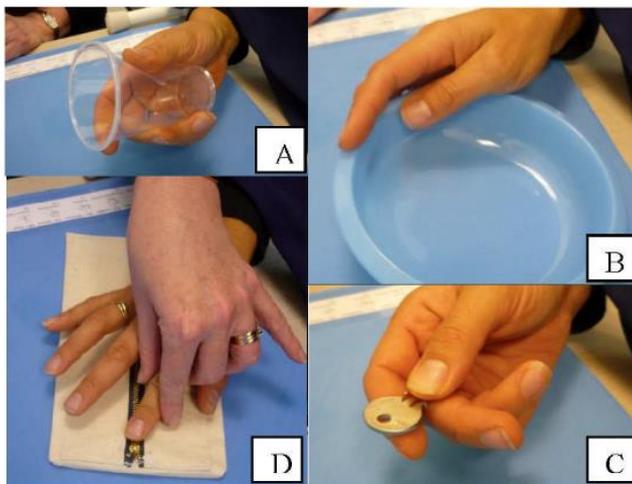
**You've got a poster of some objects. They are ordered from large to small (*the examiner points to the objects in sequence*). Some are more alike than others. Can you see them clearly? I'll give you an object to feel that is also on this poster and I'd like you to point to it on this poster or tell me what number it is. *Point to the size calibration objects*; These are to show you what the objects look like in real life. I have more than one of these objects so I may also put these objects in your hand.**

**Let's have a practice first, please place the earmuffs on and then put your hand through this curtain upturned like this (*demonstrate supine hand position. Next present the coke bottle practice item*).**

*When presenting smaller objects, say; I'm placing the object in your hand now.*

*When presenting larger or heavier objects, say; I'm placing your hand around the object now.*

**Motor enhanced protocol:** If the participant is unable to independently manipulate a test object due to impaired motor function the examiner should help facilitate exploration of the object through guiding the object through the participant's hand or moving the participant's hand to replicate the optimal haptic EP for the test object. Use Appendix A to select other haptic EPs related to the object to use in combination if the participant continues to have difficulty recognising the object.



## Test Form

Indicate the participant's response in the Response Number column. Award 3 points if the participant correctly identifies the object, 2 points if an item pair is identified and 1 point if an item distracter is identified. No points are awarded if an object from a different object set is chosen. Record comments including EP used, and if assistance was required. Four different test forms are provided each with a slightly different set of 14 test objects to avoid a learning affect if testing both hands at one time point.

## Scoring

Determine the score by summing all the responses to provide a score out of a possible 42.

## Interpretation

**A score of 42 = all responses correct, no error**

**6-8 years: <37 for the right hand and <37 for the left hand**

Impaired tactile object recognition is suggested if the summed score is *less than 37* (right and left hand) for children and adolescents aged 6-8 years

**9-11 years: <39 for the right hand and <39 for the left hand**

Impaired tactile object recognition is suggested if the summed score is *less than 39* (right and left hand) for children and adolescents aged 9-11 years

**12-15 years: <40 for the right hand and <39 for the left hand**

Impaired tactile object recognition is suggested if the summed score is *less than 40* (right hand) or 39 (left hand) for children and adolescents aged 12-15 years

## References:

- Taylor, S., Mclean, B., Jacoby, P., Parsons, R., McCutcheon, S., Carey, L. M., Girdler, S., & Elliott, C. (2017d). Haptic exploratory procedures performed by children and youth with and without cerebral palsy during the functional Tactile Object Recognition Test. Manuscript in preparation.
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- Nankervis, J. (2004). *Normative standards for the funtional Tactual Object Recognition Test (fTORT): A visual response mode*. (Unpublished honours thesis). Latrobe University, Bundoora, VIC.
- Lederman, S. J., & Klatzky, R. L. (1987). Hand movements: A window into haptic object recognition. *Cognitive Psychology*, *19*, 342–368.
- Lederman, S.J., & Klatzky, R.L. (2004). Haptic identification of common objects: Effects of constraining the manual exploration process. *Perception & Psychophysics*, *66*(4), 618-628.



**Test version 2: Scoring sheet - FUNCTIONAL TEST OF TACTUAL OBJECT RECOGNITION, Subject Code: \_\_\_\_\_ Date: \_\_\_\_\_**

Dominant hand: Right / Left		Hand tested: Right / Left		Assessor:		Test phase:							
Object No.	Test object (random selection of objects)	MDA	Response (object no. or name)	Score (Inter) 3 - Correct match / exact item 2 - Item pair 1 - Item distracter 0 - Incorrect	Time taken	Exploratory procedures used * (see definitions below)							
Trial	Coke Bottle					En	SC	UH	Pr	LM	CF	Fu	PM
Test 1	Spoon	SH				En	SC	UH	Pr	LM	CF	Fu	PM
Test 2	Metal doorknob	TM				En	SC	UH	Pr	LM	CF	Fu	PM
Test 3	Stainless steel bowl	TM				En	SC	UH	Pr	LM	CF	Fu	PM
Test 4	Plastic card	TX				En	SC	UH	Pr	LM	CF	Fu	PM
Test 5	Zipper	MO				En	SC	UH	Pr	LM	CF	Fu	PM
Test 6	Small faced watch	SI				En	SC	UH	Pr	LM	CF	Fu	PM
Test 7	Empty jar	WT				En	SC	UH	Pr	LM	CF	Fu	PM
Test 8	House key	SI				En	SC	UH	Pr	LM	CF	Fu	PM
Test 9	Wooden clothes peg	TX				En	SC	UH	Pr	LM	CF	Fu	PM
Test 10	Hardcover book	HA				En	SC	UH	Pr	LM	CF	Fu	PM
Test 11	Full milk bottle	WT				En	SC	UH	Pr	LM	CF	Fu	PM
Test 12	Firm plastic cup	HA				En	SC	UH	Pr	LM	CF	Fu	PM
Test 13	Click switch	MO				En	SC	UH	Pr	LM	CF	Fu	PM
Test 14	Cylinder pasta	SH				En	SC	UH	Pr	LM	CF	Fu	PM

**SI = size**  **TM = temperature**  **WT = weight**  **UH = unsupported holding**  **HA = hardness**  **Pr = pressure**  **TX = texture**  **SIH = shape**  **MO = function/motion** 

**En** = Enclosure  **SC** = Static contact  **UH** = Unsupported Holding  **Pr** = Pressure  **LM** = Lateral motion  **CF** = Contour Following  **Fu** = Function  **PM** = Part Motion 

**Test version 3: Scoring sheet - FUNCTIONAL TEST OF TACTUAL OBJECT RECOGNITION. Subject Code: \_\_\_\_\_ Date: \_\_\_\_\_**

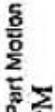
Dominant hand: Right / Left		Hand tested: Right / Left		Assessor:		Test phase:	
Object No.	Test object (random selection of objects)	MDA	Response (object no. or name)	Score (later) 3 - Correct match /exact item 2 - Item pair 1 - Item distracter 0 - Incorrect	Time taken	Explanatory procedures used * (see definitions below)	
Trial	Coke Bottle					En	SC UH Pr LM CF Fu PM
Test 1	Plastic bowl	TM				En	SC UH Pr LM CF Fu PM
Test 2	Paper card	TX				En	SC UH Pr LM CF Fu PM
Test 3	Zipper	MO				En	SC UH Pr LM CF Fu PM
Test 4	Half full milk bottle	WT				En	SC UH Pr LM CF Fu PM
Test 5	Wooden clothes peg	TX				En	SC UH Pr LM CF Fu PM
Test 6	Hardcover book	HA				En	SC UH Pr LM CF Fu PM
Test 7	Empty jar	WT				En	SC UH Pr LM CF Fu PM
Test 8	Filing cabinet key	SI				En	SC UH Pr LM CF Fu PM
Test 9	Fork	SH				En	SC UH Pr LM CF Fu PM
Test 10	Crush plastic cup	HA				En	SC UH Pr LM CF Fu PM
Test 11	Large faced watch	SI				En	SC UH Pr LM CF Fu PM
Test 12	Click switch	MO				En	SC UH Pr LM CF Fu PM
Test 13	Cylinder pasta	SH				En	SC UH Pr LM CF Fu PM
Test 14	Wooden doorknob	TM				En	SC UH Pr LM CF Fu PM

**SI = size**  **TM = temperature**  **WT = weight**  **HA = hardness**  **IA = hardness**  **TX = texture**  **SH = shape**  **MO = function/ motion** 

**En** Enclosure  **SC** Static contact  **UH** Unsupported Holding  **Pr** Pressure  **LM** Lateral Motion  **CF** Contour Following  **Fu** Function  **PM** Part Motion 

Test version 4: Scoring sheet - FUNCTIONAL TEST OF TACTUAL OBJECT RECOGNITION. Subject Code: \_\_\_\_\_ Date: \_\_\_\_\_  
 Dominant hand: Right / Left Hand tested: Right / Left Assessor: \_\_\_\_\_ Test phase: \_\_\_\_\_

Object No.	Test object (random selection of objects)	MDA	Response (object no or name)	Score (later) 3 - Correct match exact item 2 - Item pair 1 - Item distracter 0 - Incorrect	Time taken	Exploratory procedures used * (see definitions below)									
						En	SC	UH	Pr	LM	CF	Fu	PM		
Trial	Coke bottle					En	SC	UH	Pr	LM	CF	Fu	PM		
Test 1	Plastic clothes peg	TX				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 2	House key	SI				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 3	Paperback book	HA				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 4	Half full milk bottle	WT				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 5	Paper card	TX				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 6	Button	MO				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 7	Cylinder shaped pasta	SH				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 8	Crush plastic cup	HA				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 9	Empty jar	WT				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 10	Small faced watch	SI				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 11	Plastic bowl	TM				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 12	Click switch	MO				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 13	Spoon	SH				En	SC	UH	Pr	LM	CF	Fu	PM		
Test 14	Wooden doorknob	TM				En	SC	UH	Pr	LM	CF	Fu	PM		

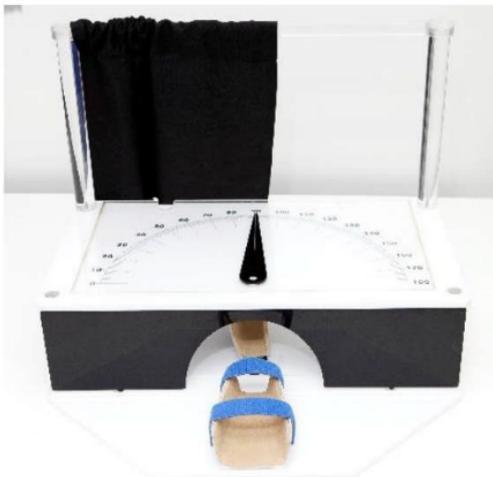
**SI = size**  **TM = temperature**  **WT = weight**  **HA = hardness**  **TX = texture**  **SH = shape**  **MO = function/motion**   
**En** Enclosure  **UH** Unsupported Holding  **Pr** Pressure  **LM** Lateral Motion  **CF** Contour Following  **Fu** Function  **PM** Part Motion 

## Wrist Position Sense Test (WPST)

(Carey, Oke & Matyas, 1996)

### Description

The Wrist Position Sense Test (WPST) (Carey et al., 1996) assesses a person's capacity to indicate knowledge of wrist position following imposed movements at the wrist. The test involves matching of 20 predetermined imposed wrist positions in the flexion-extension range using a pointer aligned with the axis of movement at the wrist joint and a protractor scale. The test is standardised, has normative standards for adults (~40 to 85 years) and children (6 to 15 years) and has high retest reliability for both populations ( $r = .88$  and  $.92$  for adults) (Carey et al., 1996; 2002; Taylor et al., 2017a). The test has good discriminative validity and has demonstrated clinical acceptability, construct validity and responsiveness for children with cerebral palsy aged 6 to 15 years (Carey et al., 1996; Taylor et al., 2017b; 2017d).



Wrist Position Sense Test assessment box

### Equipment

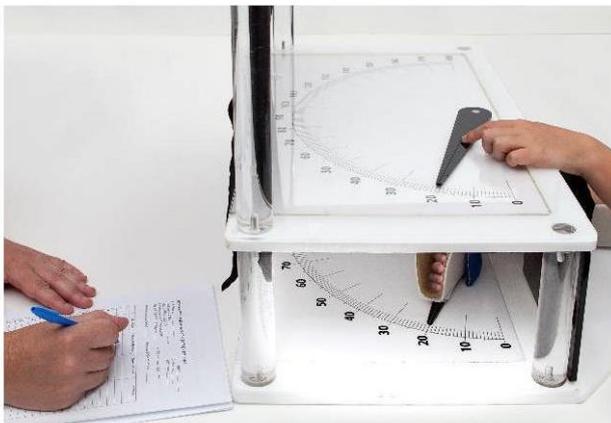
- WPST assessment box
- Universal forearm and hand splints
- Assessment form and pen
- Waist height table for participant  
(Booster cushion provided by examiner as required)
- Two chairs (one with armrests for the participant as required)

### Set up

The participant is seated in a chair opposite the examiner. The WPST assessment box is set up on a table between them. The forearm splint and the universal hand splint are in place. The top protractor scale is removed and placed at the side.

The examiner secures the participant's hand into the forearm and hand splints with the wrist in neutral position and elbow flexed at 90 degrees. The hand is placed through the opening and held firmly in the hand splint. The hand splint is then attached to the lever in a position that ensures the wrist axis of movement is directly aligned with the axis of movement of the lever. You may ensure correct alignment by placing your first two fingers together beneath the participant's wrist joint and feeling for the axis of movement of the wrist as you move the

participant's hand in the splint. The apparatus should be lined up directly in front of the participant. To accommodate for contracture or spasticity of the upper limb include foam padding or physical assistance to help secure the wrist and forearm more comfortably and ensure standardised positioning.



Participant responds using the pointer on the superior protractor



Standardised positioning of the wrist in the hand splint

### Test Administration

Move the participant's hand to the different angles that are indicated on the test form. Twenty positions are tested for each hand.

If the participant is unable to move the pointer with their hand, the examiner may move the pointer slowly for the participant until the participant indicates the position verbally by saying 'Stop'. Allow the participant to visually check their judgment once the pointer is in position. The participant may wish to alter their response. If the participant also has difficulty giving a verbal response, ask them to generally indicate the direction of the movement and slowly move the pointer in this direction until the participant indicates their response by asking you to 'Stop'.

If a participant is unable to move their wrist into the test positions stated on the test form due to spasticity or fixed deformity, alternative positions may be required, especially for maximum extension. These new positions may be  $10^{\circ}$  -  $25^{\circ}$  less than those on the test form and are chosen to be within comfortable range of movement for the participant. Altered test positions can be recorded on the test form in the column marked 'Alternative test positions'.

### Verbal Instructions

*(Explain the test using the standardised script as provided below)*

*(Fit wrist and forearm splints first). The purpose of this activity is to see how well you can feel where your hand is pointing without looking at it. These splints (point to the hand splint and forearm splint) keep your hand and arm straight and I will place this board in so you cannot see. (The top plate and protractor are removed from the WPST to position child's hand in WPST box).*

**I'm going to move your hand to different positions like this** (*examiner moves the hand to a position using the lever. Child views their hand being moved*). **Can you feel me moving your hand? How comfortably can you bend your hand this way** (mark down angle on test form) **and this way** (mark down angle on test form).

**Please move this pointer** (*examiner points to the response pointer positioned on the top of the box*) **to show me where you think your hand is pointing, like this** (*examiner moves the pointer to line up with the position of the hand*). **You can place the pointer on any number or mark in between the numbers** (*show angle numbers and non-numbered markers between angle numbers*).

**The pointer lines up with your index finger through here, don't worry about where your thumb is** (*examiner touches wrist at axis of movement - the 'snuffbox' and indicates a straight line through to the tip of the index finger.*)

**Let's have a practice first.** (*Examiner positions hand in a practice position*). **I can tell you how you are going in this practice, but I won't be able to give you any clues or tell you how you're going after the practice.** Commence test trial.

#### **Test Form**

Read the participant's response, to the closest degree, from the protractor scale. Record the degree indicated on the scoring sheet. Determine error of estimation by calculating the difference in degrees between the test angle and angle indicated. Calculate absolute error for all angles.

#### **Scoring**

Sum absolute error to give total error. Average error is then determined by dividing by the number of stimuli (i.e. by 20).

#### **Interpretation**

**An average error of 0° = all angles correct, no error**

**6-8 years: >16.3° for the right hand and >18.6° for the left hand**

Impaired wrist position sense is suggested if the average error is *greater than* 16.3° (right hand) or 18.6° (left hand) for children and adolescents aged 6-8 years

**9-11 years: >13° for the right hand and >14.3° for the left hand**

Impaired wrist position sense is suggested if the average error is *greater than* 13° (right hand) or 14.3° (left hand) for children and adolescents aged 9-11 years

**12-15 years: >11.7° for the right hand and >12.6° for the left hand**

Impaired wrist position sense is suggested if the average error is *greater than* 11.7° (right hand) or 12.6° (left hand) for children and adolescents aged 12-15 years

## References:

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**Wrist Position Sense Test forms**  
(Carey, Oke & Matyas, 1996)

**Wrist Position Sense Test (WPST) – LEFT Hand**

Participant ID: \_\_\_\_\_

Date: \_\_\_\_\_

Therapist name: \_\_\_\_\_

Hand Tested: **LEFT**

WPST kit size used    Lge    Sml

Maximum active flexion \_\_\_\_\_    Maximum active extension \_\_\_\_\_

	Test Position	Alternative Position	Degree Indicated	Error (Deg)
1	120			
2	71			
3	102			
4	97			
5	119			
6	79			
7	133			
8	88			
9	64			
10	87			
11	134			
12	107			
13	145			
14	121			
15	60			
16	152			
17	95			
18	56			
19	104			
20	143			

**CALCULATIONS:**

Total Error (degrees): \_\_\_\_\_

Average Error (degrees): \_\_\_\_\_

Standard Deviation of Error (degrees): \_\_\_\_\_

**Comments:**

\_\_\_\_\_  
\_\_\_\_\_  
\_\_\_\_\_

**Wrist Position Sense Test (WSPT)**  
(Carey, Oke & Matyas, 1996)

**Wrist Position Sense Test (WPST) – RIGHT Hand**

Participant ID: \_\_\_\_\_

Date: \_\_\_\_\_

Therapist name: \_\_\_\_\_

Hand Tested: **RIGHT**

WPST kit size used    Lge    Sml

Maximum active flexion \_\_\_\_\_    Maximum active extension \_\_\_\_\_

	Test Position	Alternative Position	Degree Indicated	Error (Deg)
1	104			
2	58			
3	41			
4	68			
5	28			
6	79			
7	121			
8	35			
9	64			
10	92			
11	57			
12	109			
13	76			
14	74			
15	125			
16	25			
17	77			
18	41			
19	119			
20	98			

**CALCULATIONS:**

Total Error (degrees): \_\_\_\_\_

Average Error (degrees): \_\_\_\_\_

Standard Deviation of Error (degrees): \_\_\_\_\_

**Comments:**

\_\_\_\_\_  
\_\_\_\_\_  
\_\_\_\_\_

## References:

- Blennerhassett, J. M., Carey, L. M. & Matyas, T. A. (2006). Grip force regulation during pinch grip lifts under somatosensory guidance: comparison between people with stroke and healthy controls. *Archives of Physical Medicine & Rehabilitation*, 87, 418-429.
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## Appendix A - Guidelines for filming the sense\_assess@kids assessment

The following are guidelines for filming the sense\_assess@kids to ensure all important aspects of hand movements are captured. The guidelines are based on The Melbourne Assessment of Unilateral Upper Limb Function: test administration manual (Randall, Johnson, & Reddihough, 1999). Children with poor postural control may obscure the viewing of hand movements during administration. In these situations the camera positions may be adjusted.

Standard focus refers to capturing the child's upper body and table surface. Zoom focus refers to close up focus of the child's affected hand

### Equipment required for filming

- Digital camera with battery
- Tripod
- Writable DVD
- Tape measure
- Adhesive markers

### Preparation

- Measure and mark the digital camera's position on the floor by allocating a dot for each tripod leg point to achieve consistent angles across administrations

### Child

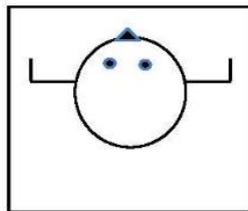
- Preferably dressed in a top with short sleeves to view the forearm and hand clearly
- Film a card with the date, time of day and participant's age and gender written on it prior to each participant administration

### Order of tasks

1. Protective Touch Test

Film between the sagittal and frontal planes with standard focus and zoomed out. The **X** marks the position for the video camera 3 metres away from and 1.5 metres to the side of the child's body so the therapist can move freely during administration.

**X**  
**Sagittal plane**  
**full body**  
**1.5 metres**

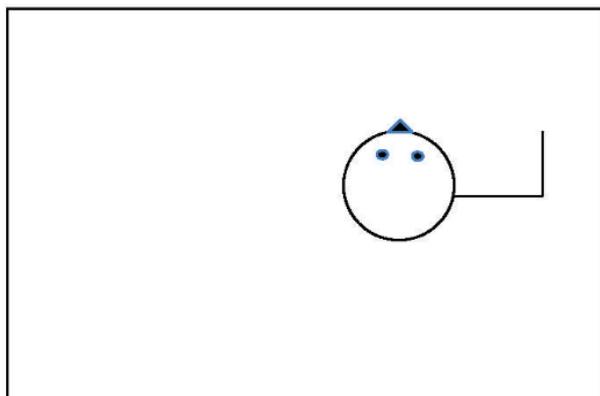


2. Tactile Discrimination Test
3. functional Tactile Object Recognition Test
4. Wrist Position Sense Test
5. Fabric Matching Test

Film in the sagittal plane with camera 1 positioned 3 metres in front of the child with standard focus. Camera 2 is to be positioned between the sagittal and frontal planes 1.5 metres from the affected arm being assessed with zoom focus both on the hand and sense\_assess© kids test items.

X  
Camera 1  
3 metres

X  
Camera 2  
1.5  
metres

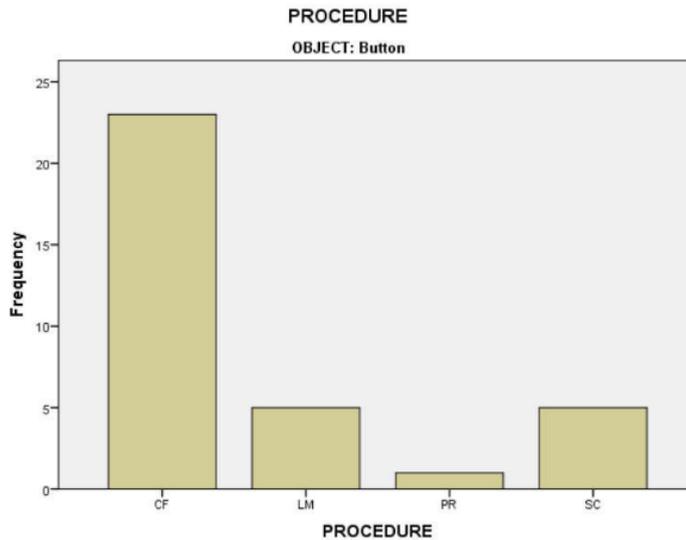


**References:**

Randall, M., Johnson, L., & Reddihough, D. (1999). The Melbourne Assessment of Unilateral Upper Limb Function: test administration manual. Melbourne: Royal Children's Hospital, Melbourne.

## Appendix B - Haptic exploratory procedures: normative standards

Note: Children should be performing, or guided to explore the functional Tactile Object Recognition Test objects using the expected haptic exploratory procedures (EPs) listed in bold. However, the use of additional EPs may also be common. EPs or exploratory movements performed that are not listed as additional EPs for each object should be documented as this may indicate inefficient exploratory behaviour. This evidence is based on preliminary normative data from thirty-one 6 to 15 year old children and youth.



**Expected haptic EP:**

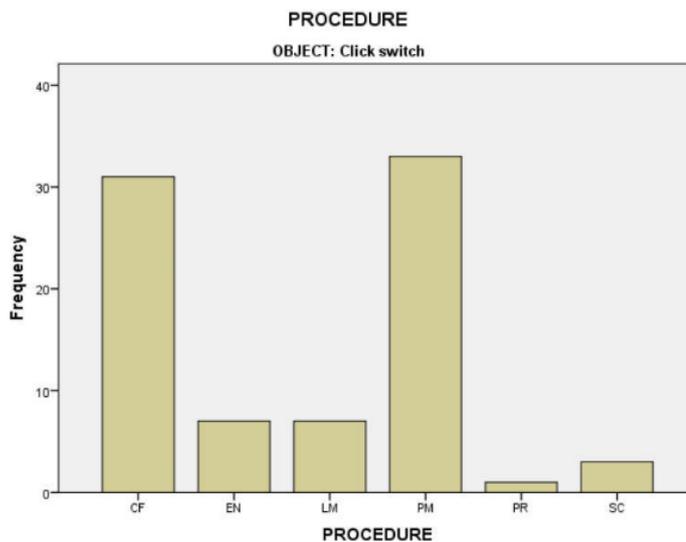
**Part motion (PM)**

Additional haptic EPs:

Contour Following (CF)

Lateral Motion (LM)

Static Contact (SC)



**Expected haptic EP:**

**Part Motion (PM)**

Additional haptic EPs:

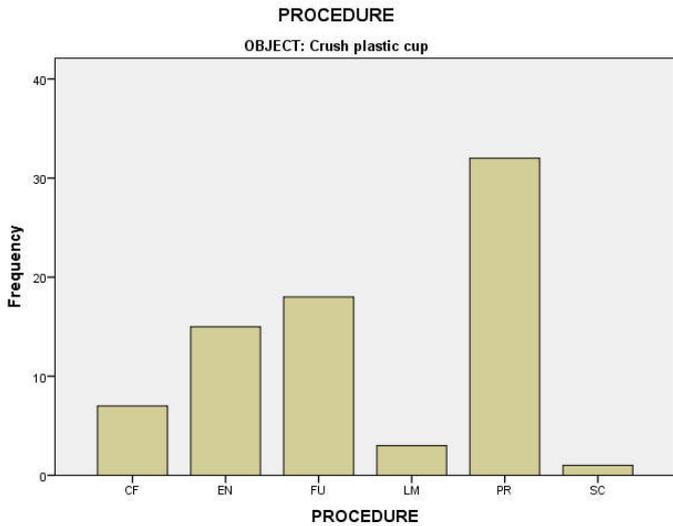
Contour Following (CF)

Enclosure (En)

Lateral Motion (LM)

Pressure (Pr)

Static Contact (SC)

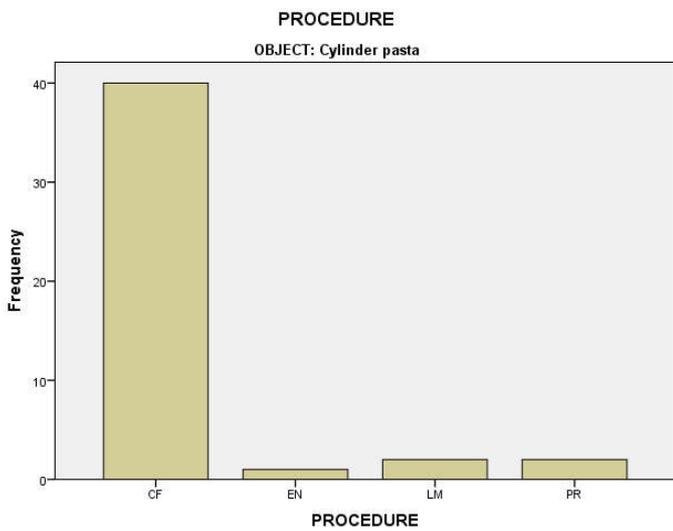


**Expected haptic EP:**

**Pressure (Pr)**

Additional haptic EPs:

Contour Following (CF)  
Enclosure (En)  
Function (Fu)  
Lateral Motion (LM)  
Static Contact (SC)

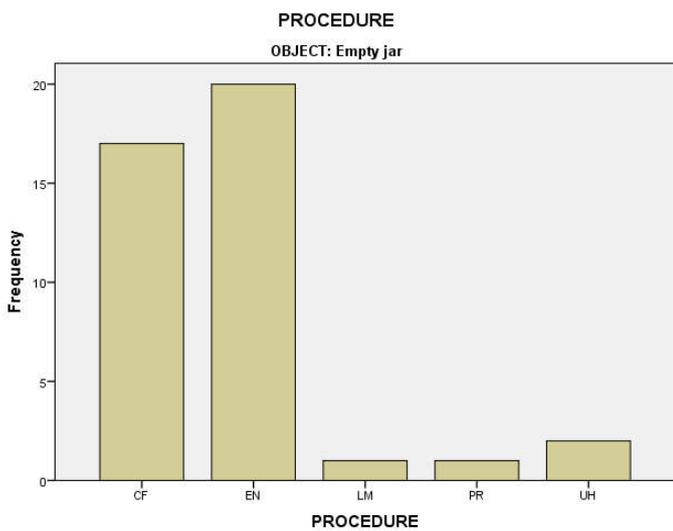


**Expected haptic EP:**

**Contour Following (CF)**

Additional haptic EPs:

Enclosure (En)  
Lateral Motion (LM)  
Pressure (Pr)

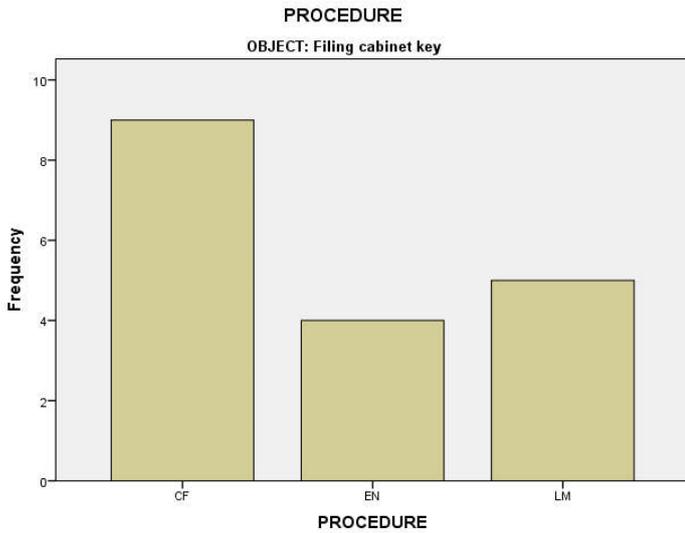


**Expected haptic EP:**

**Unsupported Holding (UH)**

Additional haptic EPs:

Contour Following (CF)  
Enclosure (En)  
Lateral Motion (LM)  
Pressure (Pr)

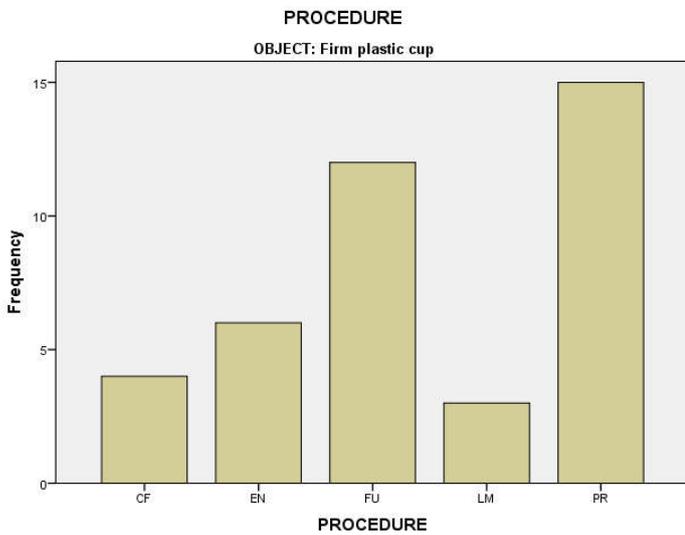


**Expected haptic EP:**

**Enclosure (En)**

Additional haptic EPs:

Contour Following (CF)  
Lateral Motion (LM)

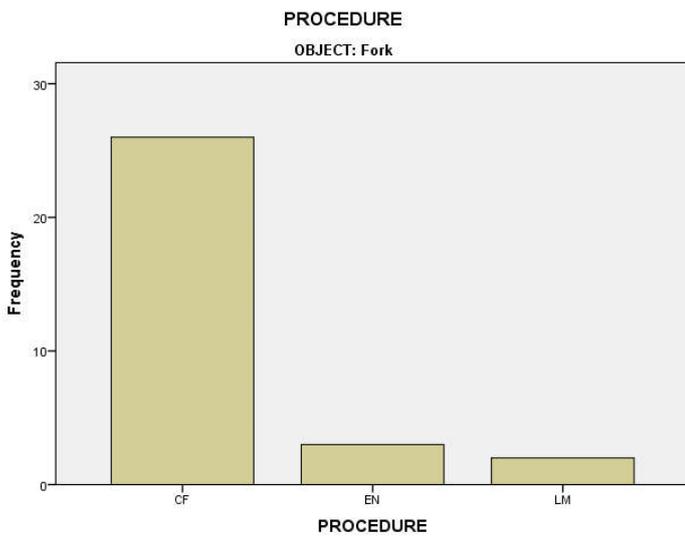


**Expected haptic EP:**

**Pressure (Pr)**

Additional haptic EPs:

Contour Following (CF)  
Enclosure (En)  
Function (Fu)  
Lateral Motion (LM)

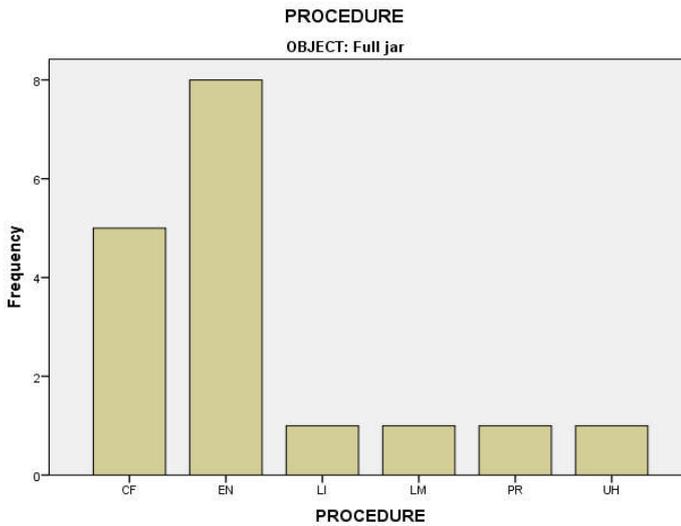


**Expected haptic EP:**

**Contour Following (CF)**

Additional haptic EPs:

Enclosure (En)  
Lateral Motion (LM)



**Expected haptic EP:**

**Unsupported Holding (UH)**

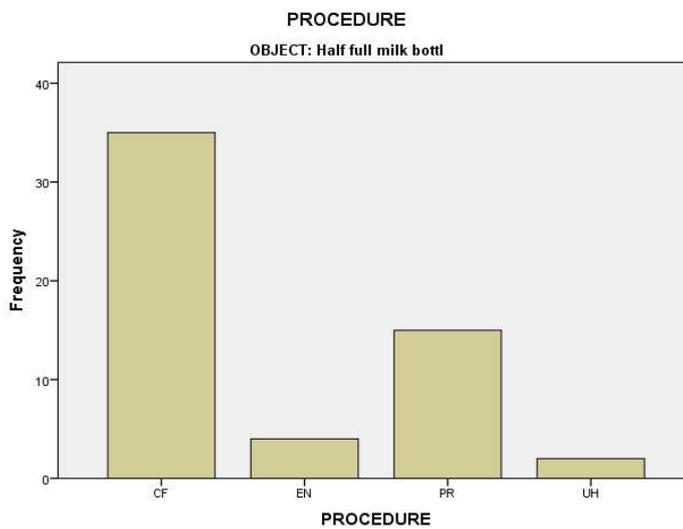
Additional haptic EPs:

Contour Following (CF)

Enclosure (En)

Lateral Motion (LM)

Pressure (Pr)



**Expected haptic EP:**

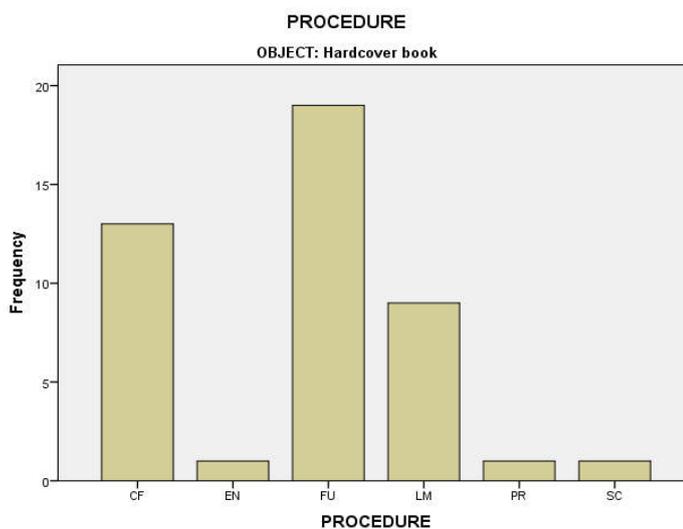
**Unsupported Holding (UH)**

Additional haptic EPs:

Contour Following (CF)

Enclosure (En)

Pressure (Pr)



**Expected haptic EP:**

**Pressure (Pr)**

Additional haptic EPs:

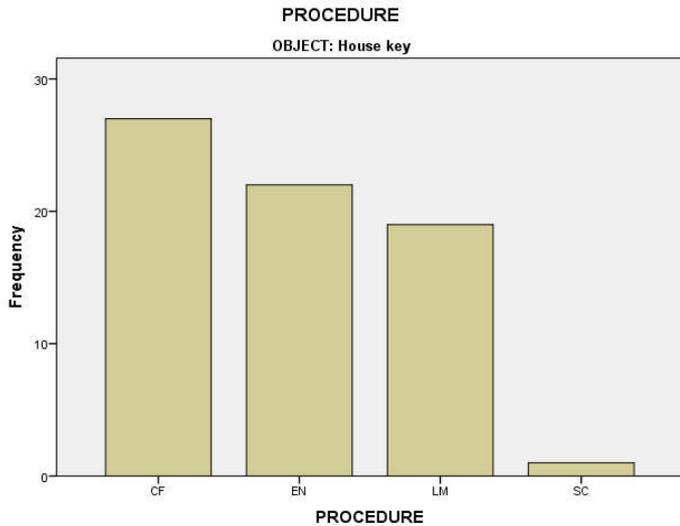
Contour Following (CF)

Enclosure (En)

Function (Fu)

Lateral Motion (LM)

Static Contact (SC)



**Expected haptic EP:**

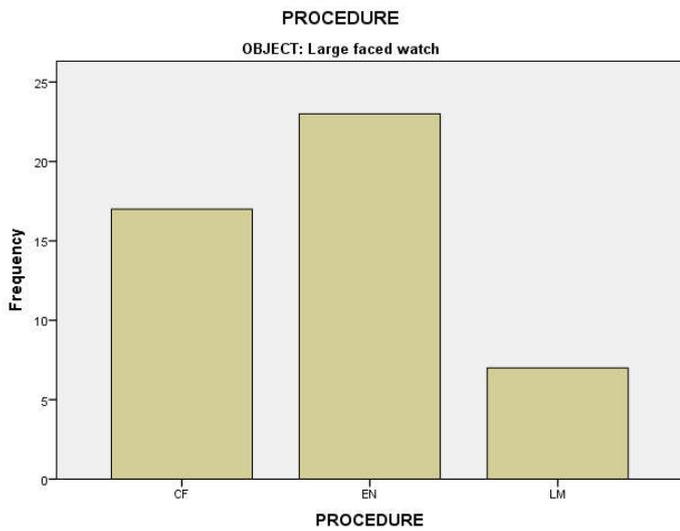
**Enclosure (En)**

Additional haptic EPs:

Contour Following (CF)

Lateral Motion (LM)

Static Contact (SC)



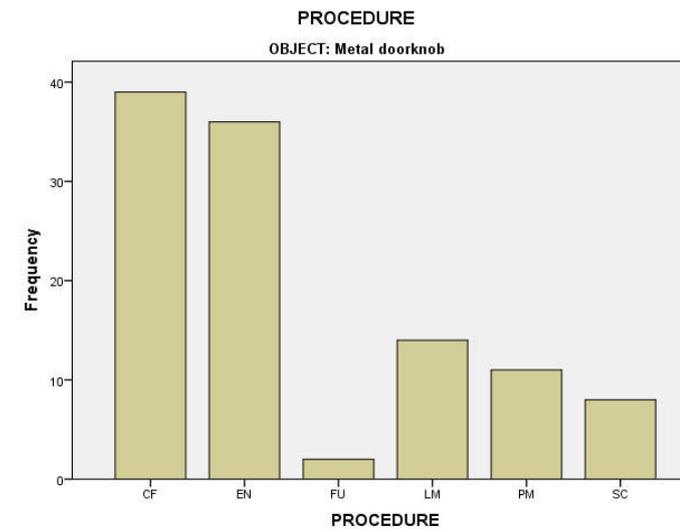
**Expected haptic EP:**

**Enclosure (En)**

Additional haptic EPs:

Contour Following (CF)

Lateral Motion (LM)



**Expected haptic EP:**

**Static Contact (SC)**

Additional haptic EPs:

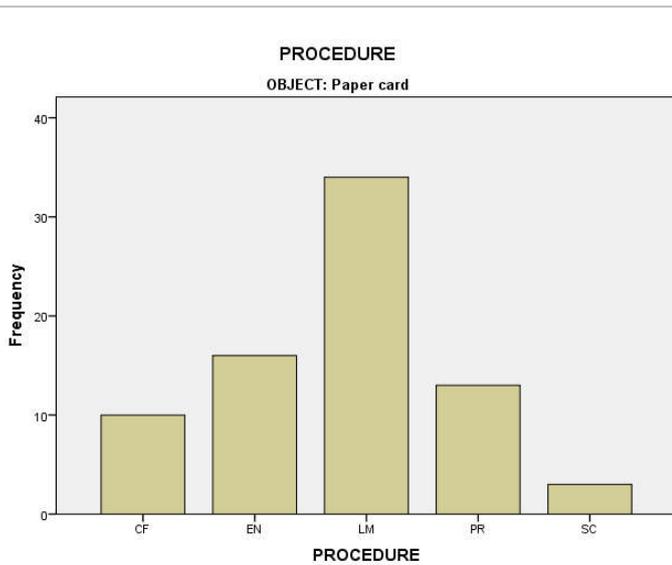
Contour Following (CF)

Enclosure (En)

Function (Fu)

Lateral Motion (LM)

Part Motion (PM)



**Expected haptic EP:**

**Lateral Motion (LM)**

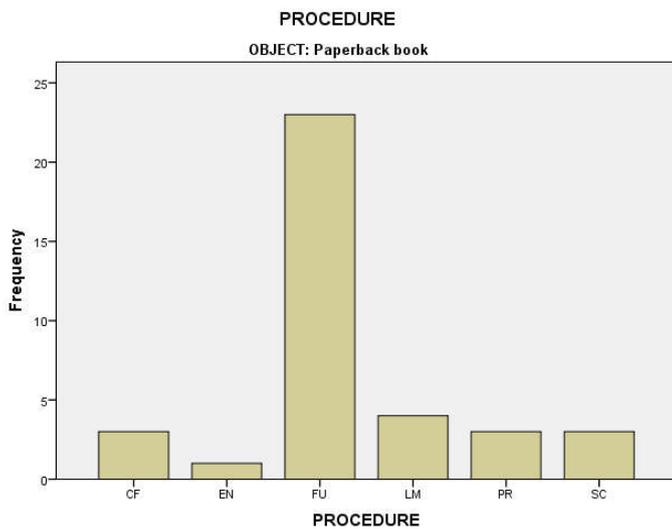
Additional haptic EPs:

Contour Following (CF)

Enclosure (En)

Pressure (Pr)

Static Contact (SC)



**Expected haptic EP:**

**Pressure (Pr)**

Additional haptic EPs:

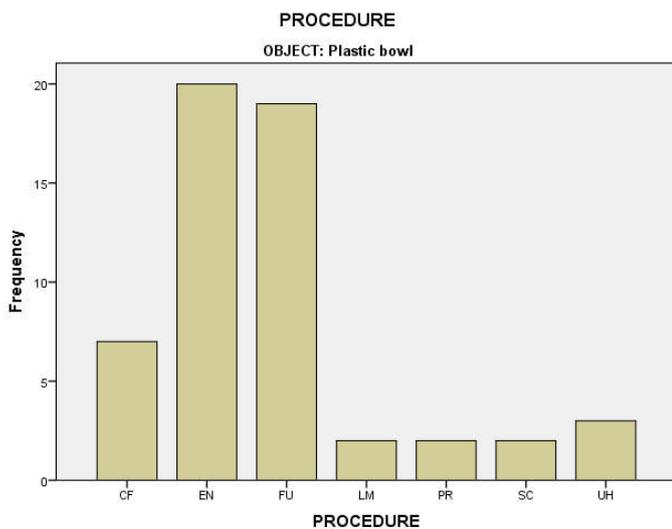
Contour Following (CF)

Enclosure (EN)

Function (Fu)

Lateral Motion (LM)

Static Contact (SC)



**Expected haptic EP:**

**Static Contact (SC)**

Additional haptic EPs:

Contour Following (CF)

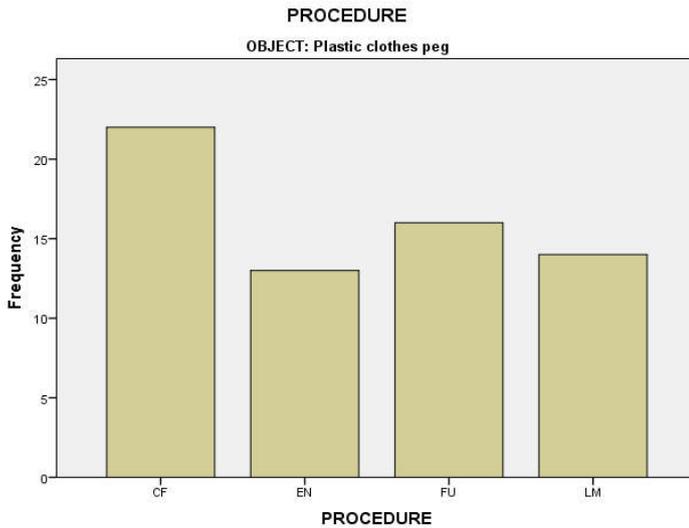
Enclosure (En)

Function (Fu)

Lateral Motion (LM)

Pressure (Pr)

Unsupported Holding (UH)

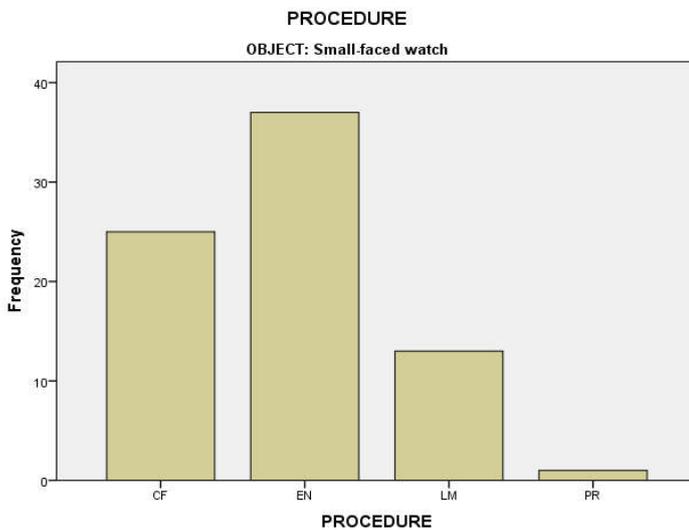


**Expected haptic EP:**

**Lateral Motion (LM)**

Additional haptic EPs:

Contour Following (CF)  
Enclosure (En)  
Function (Fu)

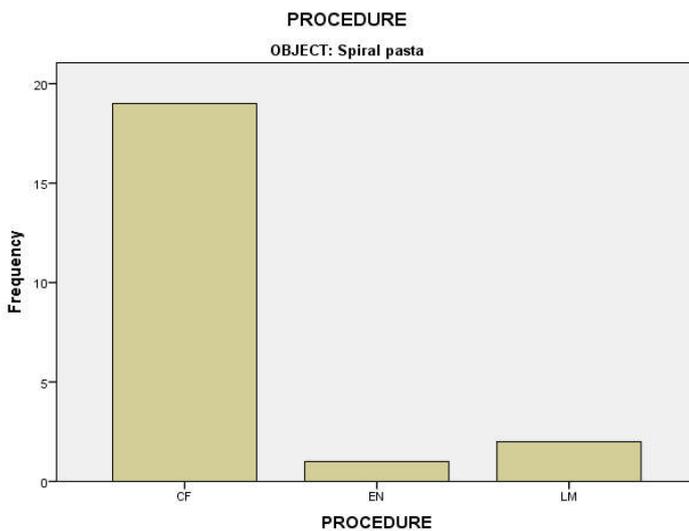


**Expected haptic EP:**

**Enclosure (En)**

Additional haptic EPs:

Contour Following (CF)  
Lateral Motion (LM)  
Pressure (Pr)

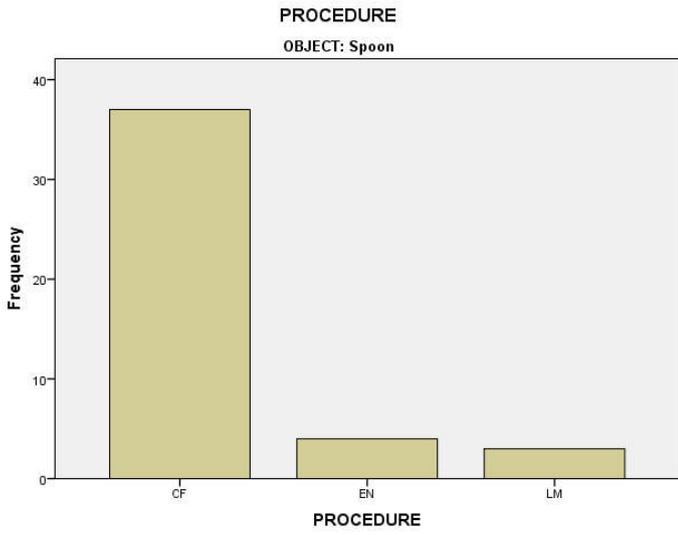


**Expected haptic EP:**

**Contour Following (CF)**

Additional haptic EPs:

Enclosure (En)  
Lateral Motion (LM)



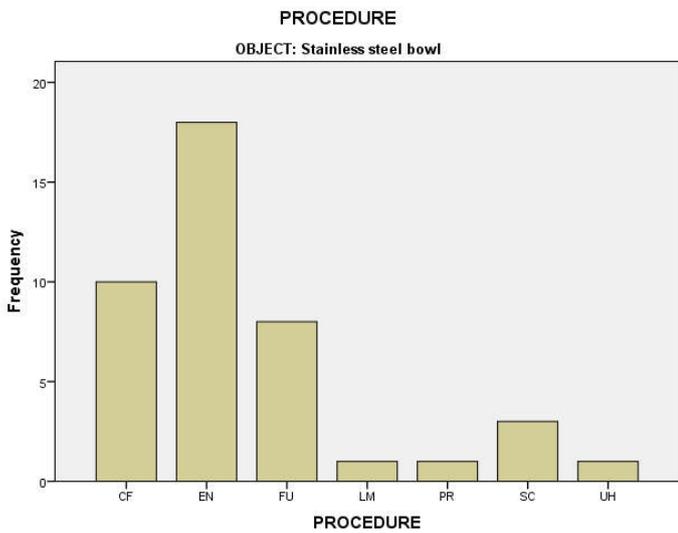
**Expected haptic EP:**

**Contour Following (CF)**

Additional haptic EPs:

Enclosure (En)

Lateral Motion (LM)



**Expected haptic EP:**

**Static Contact (SC)**

Additional haptic EPs:

Contour Following (CF)

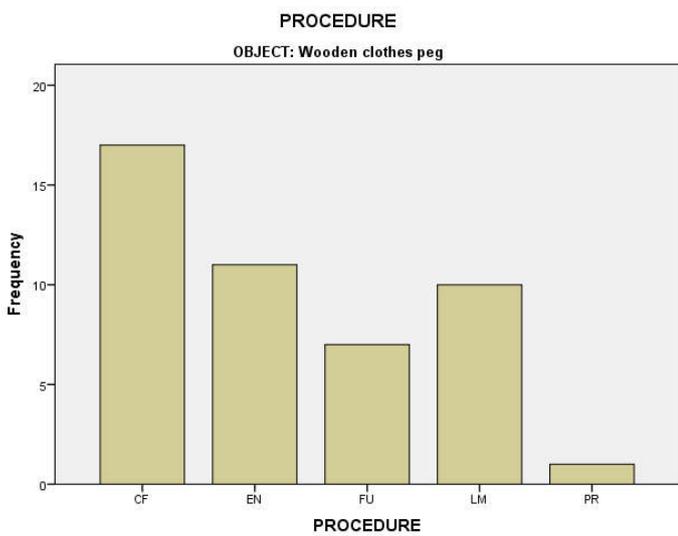
Enclosure (En)

Function (Fu)

Lateral Motion (LM)

Pressure (Pr)

Unsupported Holding (UH)



**Expected haptic EP:**

**Lateral Motion (LM)**

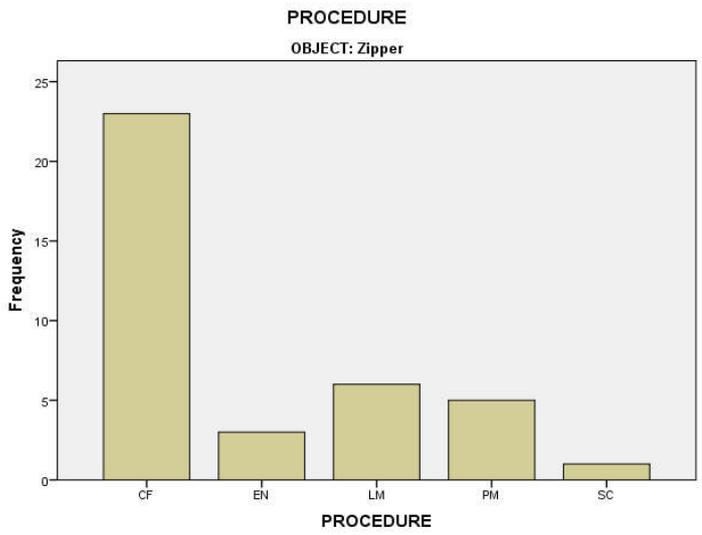
Additional haptic EPs:

Contour Following (CF)

Enclosure (En)

Function (Fu)

Pressure (Pr)



**Expected haptic EP:**

**Part Motion (PM)**

Additional haptic EPs:

Contour Following (CF)

Enclosure (En)

Lateral Motion (LM)

Static Contact (SC)

## Appendix C - Fabric Matching Test

### Fabric Matching Test (FMT) (Carey & Matyas, 2005)

#### Description

Texture discrimination is defined by a child's ability to match fabrics during the Fabric Matching Test (FMT). The test consists of two identical fabric wheels as shown in Figure 1. The fabrics are ordered from very smooth to rough and the child is required to indicate if two fabrics are the same or different using their index finger (Figure 2). The test has reliability, discriminative validity, and normative data for adults (Carey, Matyas & Oke, 2000) and high retest reliability and normative standards for children and adolescents aged 6 to 15 years (Overheu et al., 2017).

#### Equipment

Two fabric disks  
Test board with curtain and guide for the fabric disks  
Assessment form and pen  
Waist height table for participant  
(Booster cushion provided by examiner as required)  
Two chairs (one with armrests for the participant as required)



Figure 1. Identical fabric wheels. When placed in the presentation windows the comparison wheel is to the child's right.



Figure 2. Image of a participant actually exploring the test fabric.

#### Set Up

The child is seated in a chair opposite the examiner with the FMT frame and curtain set up on a waist-height table for the child. The elbow should be supported in a position of approximately 90 degrees flexion. The curtain is drawn to occlude vision, ensuring only the tactile system is being assessed. The child then places their hand in prone position through the curtain frame.

### **Test administration**

The therapist guides the child's index finger at a constant speed and pressure to feel the 'Test fabric' and the 'Comparison fabric' and asks if the fabrics feel the same or different. If the child states their comparison fabric is different to the test fabric, they instruct the therapist to turn their comparison wheel to either a smoother or rougher fabric. The child is required to match the two fabrics as closely as possible using discriminative touch. The child may revisit the fabrics as many times as they require until they are satisfied with their choice. Prior to commencing the child can perform 1-2 practices of the FMT to ensure instruction comprehension, which they indicate verbally. The FMT is performed on the left and right hand, with ten fabrics tested in pseudo-random order for each hand, requiring children to match a total of 20 fabrics. The FMT takes approximately 10-15 minutes to complete.

### **Verbal Instructions**

*(Explain the test using the standardised script as provided below)*

**In this activity I will ask you to match a set of everyday fabrics. When I say match, I'm asking you if the fabrics feel the same. I will pull this curtain across so you can't see.** *(Start with fabric wheels exposed)* **The fabrics look like this, they start smooth at number one** *(present one fabric wheel and point to surface 1)* **and become roughest at number 10** *(point to surface 10 then help child to explore each of the fabrics from 1-10 with both hands).* **These two wheels are the same** *(place wheels in display base and put lid on to show the two fabric windows).* **I am going to guide your hand like this** *(guide child's hand to demonstrate)* **to feel two fabrics: the first in my window (T) and the second in your window (C), then ask if they feel the same or different.**

**In your window, I will only ever start by putting the finest or roughest fabric** *(point to fabric 1 and then 10).* **So you either need to move up towards the rougher fabrics or down towards the smoother fabrics** *(demonstrate by moving wheels).*

**Let's have a go, I will put one fabric in my window** *(point to opening marked T on subject's left hand side and put fabric 9 in the opening)* **and one in your window** *(put fabric 1 in the comparison window marked C).* **Do the fabrics feel the same or different?** *(Practice trial is done with vision).* *(After child responds)* **Does your fabric feel rougher or smoother than mine? Ok so we need to move towards the smoother/rougher fabrics. I can move your wheel one fabric at a time or more than one if you think there is a big difference, you could say move two fabrics towards the rougher surfaces. If child selects to go smoother when you are on the smoothest, fabric 1, say; That one is the smoothest, I can't go any smoother, would you like to try the other way?**  
**If you feel like your finger needs a rest let me know. Do you understand?**  
**Do you have any questions?**

### **Test form**

Scores are recorded on a test form with the child given no indication of accuracy. On the test form write down the response from the child. This is the corresponding number from the fabric disk in the Comparison window. The Comparison window shows the disk that is moved to match with the Test window.

### **Scoring**

Raw scores are entered into a spreadsheet and the accuracy of the 10 presentations conducted on each hand is assessed using the Spearman's rank correlation coefficient (Rho). This is a nonparametric measure of association between two variables (Portney & Watkins, 2009).

### **Interpretation**

The higher the score the better the child's tactile discrimination ability i.e. a rho of 1 is all responses correct.

### **References:**

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## Appendix D - Jebsen Taylor Hand Function Test

### Jebsen Taylor Hand Function Test (JTHFT)

#### “Small objects” (Jebsen et al., 1969)

#### Description

The Jebsen Taylor Hand Function Test (JTHFT) has been widely used, has favorable psychometric properties, and has the advantage of normative scores for age, gender, and hand dominance. The JTHFT item selected for this study involves a pinch grip action.

#### Equipment

Tin can  
2 paper clips  
2 bottle caps  
2 5cent coins  
Test Board  
Test form  
Chair  
Waist height table  
(Booster cushion provided  
by examiner as required)  
Pen  
Stopwatch



#### Set Up

Align the test board with the edge of the table closest to the participant. The test board has an outline of objects used in the test to facilitate their placement for testing. Place the tin can in front of the participant, 12.5 cm (5") from edge of table. Line up two 5 cent coins, two bottle caps (with insides facing up) and two (1") paper clips (oriented vertically) as indicated on the test board towards the hand that is being tested. The objects are placed approx. 5 cm (2") apart. The paper clips are closest to the tested hand, and the 5 cent coins placed nearest the can.

#### Verbal Instructions

*(Explain the test using the exact phrasing as provided)*

**“Place your (right / left) hand flat on the table please. When I say “Go”, use your (right / left) hand to pick up these objects one at a time like this *(indicate picking up objects one at a time)* and place them in the can as fast as you can, beginning with this one *(indicate paper clip)*. Do you understand? Ready? Go.”**

#### Timing

Start the timer from “Go” until the sound of the last coin hitting the inside of the can.

### Test Form

Fill in the total time taken for the participant to place all the objects into the can.

### Scoring

The scores below indicate the time taken for typically developing children aged 5 to 10 years old to perform the test according to hand dominance and age.

### Interpretation

Scoring at or below the time indicated in Table 4 would imply normal healthy dexterity of the hand tested. Taking longer than the timings indicated in Table 4 above may indicate abnormally slow dexterity for the hand tested. Further testing may be conducted if the participant takes a longer than normal time to complete the test.

Table 4. Normative Data for Jebsen Taylor Hand Function Test 'Small Objects' (Beagley, Reedman, Sakzewski, & Boyd, 2016).

Age Group (n)		5Y 19	6-7Y 36	8-9Y 34	10Y 13
Subtest	Hand	Mean Times and Standard Deviations (sec)			
Small Objects	ND	8.27 (1.09)	7.27 (1.13)	6.94 (1.11)	6.86 (0.91)
	D	7.70 (1.12)	6.53 (0.91)	6.31 (0.82)	6.47 (1.26)

Note. Y=years; n=participant sample; ND=non-dominant; D=dominant. Adapted from (2016).

"Establishing Australian norms for the Jebsen Taylor Test of Hand Function in typically developing children aged five to 10 years: A pilot study," by S. B. Beagley, S. E. Reedman, L. Sakzewski and R. Boyd, *Physical & Occupational Therapy in Pediatrics*, 36, p. 95.

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- Jebsen, R., Taylor, N., Trieschmann, R., Trotter, M., & Howard, L. (1969). An objective and standardized test of hand function. *Archives of Physical Medicine and Rehabilitation*, 50, 311-319.

### Appendix E - Levels of difficulty

In the event of significant somatosensory deficit please attempt assessment of the child using Level 1. If they excel through this level move through the other levels as appropriate. In using the subtests in this way the therapist can establish if the full subtests of the *sense\_assess© kids* are an appropriate assessment for an individual.

Test	Level 1 (Easy)	Level 2 (Moderate)	Level 3 (Difficult)
TDT	Texture 300	Texture 260	Texture 170
FTORT	Click switch Crush plastic cup Metal doorknob Spoon Stainless steel bowl Fork	Firm plastic cup Half full milk bottle Hard cover book Plastic card Paper card Wooden clothes peg	Cylinder pasta Empty jar House key Small faced watch
WPST	Close to 90°	Extension	Flexion
Left hand	Between 70° - 110°	Between 30° - 69°	Between 151° - 180°
Right hand	Between 70° - 110°	Between 111° - 150°	Between 0° - 29°



## **APPENDIX F      Descriptive Study**

**For submission to: Physical & Occupational Therapy in Pediatrics; impact factor 1.255**

PhD candidate Belinda McLean accounted for 85 per cent of the intellectual property associated with the final manuscript, collectively Susan Taylor and the other authors accounted for 15 per cent.

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### **Somatosensory discrimination impairment in children with hemiplegic cerebral palsy**

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## **Abstract**

**Introduction:** Impairments in somatosensation are recognised as having a significant effect on upper limb function in children with cerebral palsy (CP). Our objective was to characterise somatosensory discrimination impairment of the upper limb across domains of touch detection and discrimination, proprioception and haptic object recognition in children with hemiplegic CP aged 6 to 15 years.

**Methods:** A cross-sectional observational study of somatosensory impairment in children with CP. The *sense\_assess© kids* was administered to 28 children, aged 6 to 15.5 years (mean age = 10.1yrs, SD = 2.4yrs), with hemiplegic CP (right hemiplegia n = 15) and Manual Ability Classification System Level I (n = 11) and II (n = 17).

**Results:** Twenty-three (82%) had impaired somatosensory discrimination in one or more somatosensory domains. Tactile discrimination was impaired in 18 (67%), body position sense in 20 (74%) and haptic object recognition was impaired in 21 (75%) participants.

**Conclusion:** Presence of somatosensory impairment in the upper limb of children with hemiplegic CP is high; with 82% having impairment and 71% having impairment across more than one domain. Evidence-based therapeutic interventions are needed to remediate somatosensory deficits and improve functional outcomes for children with neurological impairments.



## **Introduction**

Cerebral palsy (CP) is an umbrella term for an injury or malformation in the developing infant brain resulting in limitations in motor function and disturbances to sensation, perception, cognition, communication and behaviour (1). Limitations in motor function affecting primarily one side of the body is termed hemiplegia and is the most frequently occurring distribution of motor impairment in children with CP (2). Impairments in somatosensation have been reported for up to 97% of children with hemiplegic CP (HCP) (3). More recent studies have found that roughly two thirds of children with HCP assessed have reduced somatosensation and these impairments significantly impact motor function (4, 5). Recent neural imaging studies suggest that damage to somatosensory processing areas in the brain may be the primary cause of observable motor impairments (6, 7). Furthermore, there is some evidence that somatosensory function is important for the retention of functional gains following motor intervention .

Somatosensation has been defined as “*all aspects of touch and proprioception that contribute to a person’s awareness of his or her body parts and the direct interface of these with objects and the environment*” (9). Somatosensory discrimination is essential for interaction with the environment. It is needed to determine important properties of objects and tools for use and is part of the perception-action cycle that is a foundation for learning and cognitive development (10). The aspects of somatosensory discrimination that directly interface with objects and the environment include limb position sense- knowledge of where the limbs are positioned in space, tactile discrimination- the ability to discriminate between different surface properties such as texture, and haptic object recognition- the ability to discriminate and identify objects through touch (9). Somatosensory discrimination enables identification of different textures and objects without vision and sensing where our body is positioned in relation to its other parts, these are skills crucial for every day functioning (9).

The reported prevalence of somatosensory impairment in children with HCP varies from 31 to 97 percent (11). The varied rates may be a result of variability in methods of assessment (12). A recent clinimetric review of assessments

for tactile impairment sought to recommend a single battery of assessments to cover aspects of tactile registration and tactile perception (12). The recommended assessments included single point localisation, for tactile registration and the AsTex® (13) tactile discrimination test, the Klingels stereognosis method (14) and double simultaneous testing for tactile perception. In subsequent examination the tactile perception tests were found to have shortcomings such as a ceiling and learning effects. However, the authors suggested their use until more robust measures were available (15).

Measures of limb position sense have typically involved imposed movement around the proximal phalanx of the index finger in the flexion-extension plane, assessment response was the direction of movement (16), verbal confirmation of detection of movements of the index finger (17), or replication of an imposed limb position with vision occluded (18). While these assessments may be useful in determining deficits in limb position sense in children with HCP, the imposed movements are somewhat subjective and consequently there is a risk of administrator bias and responsiveness to change is unknown.

Because of the limitations in current measurement tools it remains unclear which assessments to use for evaluation of somatosensory impairment. However, it is clear that assessments which address the functional aspects of somatosensation are crucial to understanding individual somatosensory capacity. Measures addressing impairment in the domains of tactile discrimination and limb position sense exist in the current literature for adults (19). Quantitative standardised measures that have been developed for adult stroke survivors include the Tactile Discrimination Test (TDT) (20), the Wrist Position Sense Test (WPST) (21) and the functional Tactile Object Recognition Test (fTORT) (22). These subtests are part of *sense\_assess*©, an integrated method of assessment which has been designed to quantitatively measure somatosensory discrimination ability of the upper limb (23). These measures have been modified for use with children with HCP and combined for use in the assessment tool known as the *sense\_assess*© *kids* (24).

In summary, children with HCP commonly experience somatosensory deficits which impact on the functional use of their affected upper-limb. Currently there is no consensus in the literature about the frequency and functional impact of

these somatosensory impairments. A variety of assessments have been reported and existing recommended tools have weaknesses. This research aims to describe the functional somatosensory discrimination of children with HCP using the quantitative measures of tactile discrimination, wrist position sense and haptic object recognition developed by Carey and colleagues (20-22) and modified for use in children using the *sense\_assess© kids* (24). Gross motor dexterity was also measured as somatosensory impairment has been found to contribute to limitations in motor function (4).

## **Methods**

### ***Participants***

Participants attended a Cerebral Palsy Mobility Service at a tertiary children's hospital in Western Australia. Thirty-three children met the inclusion criteria of having HCP, being at least 12 weeks post their most recent botulinum toxin injection and at least 12 months post upper limb surgery at the time of assessment. Three children declined and 30 children enrolled in the study, two of whom were unable to complete the assessment. Recruitment took place from May 2013 to May 2015. This study received ethics approval from the Princess Margaret Hospital HREC (#2052). Children were assessed in their homes by an occupational therapist trained in the use of the *sense\_assess© kids*.

### **Outcome measures**

***Somatosensory discrimination.*** The *sense\_assess© kids* (Taylor et al., in preparation) measures the following aspects of somatosensory function: tactile registration; tactile discrimination; haptic object recognition; and wrist position sense. Criterion validity has been established in the component tests of the *sense\_assess©* in an adult population with and without stroke (20-22). The Protective Touch Test (PPT) is a measure of touch registration, the ability to detect the presence of a touch stimulus, at the level of protective touch using the Semmes Weinstein 4.56 monofilament (25). Texture discrimination is measured using the TDT (20). The TDT is a three-alternative forced choice response test where children are guided to feel several triplets of textures with their index finger with vision occluded, and are asked to identify the texture that is different in each set of three. The fTORT (22) is a

measure of haptic object recognition in which children are assisted to feel familiar and novel objects in a standardised manner with vision occluded and asked to identify the matching object on a response poster. The WPST (21) is a measure of wrist position sense and children are asked to indicate on a protractor where their wrist has been positioned out of vision. Normative data is available for the sense\_assess© *kids* and the tool has demonstrated clinical acceptability for children with hemiplegic CP aged 6 to 15 years (24).

**Gross manual dexterity.** The Box and Block test (BBT) (26) is a brief standardised measure of gross manual dexterity. Participants are required to move as many blocks as they can, one at a time over a partition, in one minute with one hand then the other. The BBT has been used previously with children with CP and has high test-retest reliability for typically developing children and adults ( $r = .84$ ;  $r = .96$ ), and high concurrent validity and high interrater reliability ( $r = .99$ ) in adults (27).

### ***Procedure***

Families were referred by their consultant paediatricians and invited to participate by the primary investigator (BM). Children's description of HCP was confirmed by the referring paediatrician. An occupational therapist (ST) visited the child's home to complete the assessment. The sense\_assess© *kids* takes approximately 40 minutes to administer and the (BBT) takes approximately five minutes. The Manual Ability Classification System (MACS) level (28) was identified by a 5 minute parent interview.

### ***Data analysis***

Analyses were conducted in age groups because somatosensory discrimination improves with age in typically developing children (29). Impairment was defined as being outside the 95% confidence intervals for 56 typically developing children aged 6 to 8yrs, 9 to 11yrs and 12 to 15yrs (30). The frequencies of combinations of impairments were recorded. Pearson product-moment correlation coefficients were assessed between age and each subtest of the sense\_assess© *kids* as well as between motor performance on the (BBT) and each subtest of the sense\_assess© *kids*. Data was missing for tactile discrimination for one child and for the (BBT) for two children. Analysis excluded missing data.



## Results

Twenty-eight children, 16 boys and 12 girls aged 6 to 15.5 years with a mean age of 10.1 years (SD = 2.4) and a MACS level of I (n = 11) or II (n = 17) participated in the study.

### *Somatosensory discrimination*

Twenty-three of the 28 children presented with impairment in somatosensory discrimination. Age was not associated with impairment in any of the somatosensory domains measured by the sense\_assess© kids: WPST ( $F(1,26) = 0.77, p = 0.39$ ); ftORT ( $F(1,26) 0.31, p = 0.58$ ); TDT ( $F(1,25) 0.10, p = 0.76$ ); PTT ( $F(1,26) = 1.07, p = 0.31$ ). Twenty of the 28 children (74%) demonstrated impaired wrist position sense (see Table 1).

Table 1 Performance of children with cerebral palsy on the Wrist Position Sense Test (average error score, in degrees) by age group, with reference values for performance in typically developing children

Age Range	Typically Developing (95% Confidence Interval of mean score)		Most impaired score	Cerebral Palsy sample (n= 28)		
	Lower bound Left hand	Upper bound Left hand		Best score	Median	Impaired N (%)
6 to 8 yrs. (n=9)	12.0	18.0	37.0	8.9	15.5	4 (44)
9 to 11 yrs (n=12)	9.2	13.2	41.0	10.2	21.0	11 (92)
12-15 yrs (n=7)	9.0	12.2	31.2	7.1	19.4	5 (71)
Total Impairment						20 (74)

*Note:* Lower scores reflect better performance on this test.

Twenty-one children demonstrated impaired haptic object recognition on the ftORT (see Table 2). Eighteen children demonstrated impaired tactile discrimination on the TDT (see Table 3). Six of the 28 children showed impairment in registering protective touch using the PTT. In all age groups for all domains of somatosensation there were children with HCP who demonstrated performance within the range of typically developing children.

Table 2 Performance of children with cerebral palsy on the functional Tactile Object Recognition Test by age group, with reference values for performance in typically developing children

Age Range	Typically Developing (95% Confidence Interval of mean score)		Most impaired score	Cerebral Palsy sample (n= 28)		
	Lower bound Left hand	Upper bound Left hand		Best score	Median	Impaired N (%)
6 to 8 yrs. (n=9)	39	40	3	41	34	7 (78)
9 to 11 yrs (n=12)	39	41	7	39	30.5	10 (83)
12-15 yrs (n=7)	40	41	10	41	38	4 (57)
Total Impairment						21 (75)

Note. Higher scores reflect better performance.

Table 3 Performance of children with cerebral palsy on the Tactile Discrimination Test by age group, and with reference values for performance in typically developing children

Age Range	Typically Developing (95% Confidence Interval of mean score)		Most impaired score	Best score	Median	Impaired N (%)
	Lower bound Left hand	Upper bound Left hand				
6 to 8 yrs. (n=9)	27.3	44.9	2.2	50.3	30.7	4 (44)
9 to 11 yrs (n=12)	52.1	67.9	-36.6	73.6	18.8	9 (75)
12-15 yrs (n=6)	64.9	84.1	-15.9	68.4	14.7	5 (83)
Total Impairment						18 (67)

Note. Higher scores reflect better performance.

### ***Somatosensory Impairment across domains***

Deficits also occurred in combination across somatosensory measures, with more than half of the children having impairment in three or more somatosensory domains (see Table 4). Impairment in protective touch only occurred in association with impairment across all of the somatosensory measures. When the number of somatosensory domains was compared with performance on the BBT, where lower scores indicate poorer performance, a clear decline in medians is observed with the poorest performance occurring for children with impairment in all four domains (see Table 4).

Table 4 Impairment of somatosensory discrimination across multiple domains, with the combinations of impaired domains presented

Combinations	Number	Sensory domains affected	Median performance on Box and Block*
No domains	5	N/A	35.0 (30-39)
One domain	3	Haptic object recognition (2) Limb position sense (1)	27.0 (10-42)
Two domains	4	Limb position & haptic object recognition (2) Limb position & texture discrim (1) Texture discrim & haptic object recognition (1)	18.5 (18-27)
Three domains	10	Limb position, haptic object recognition and texture discrimination (10)	18.0 (2-45)
Four domains	6	As above with the addition of protective touch.	5.5 (2-31)

Note. \*Median and range of scores for performance on the Box and Block Test is presented for each combination for the impaired hand. The score represents the number of blocks moved in one minute.

### ***Somatosensory Discrimination and Motor Performance***

Moderate to strong correlations were present between motor performance as measured by the BBT and the discriminative components of somatosensation in the sense\_assess© kids. Using a Pearson product-moment correlation the WPST and the TDT demonstrated a moderate correlation with motor performance ( $r(24) = -0.48, p = .01$ ) and ( $r(23) = 0.43, p = .03$ ) respectively, the fTORT showed a strong correlation with motor performance on the BBT ( $r(24) = 0.69, p = <.005$ ). The PTT has a ceiling and 22 children achieved the top score so no test was used to investigate this beyond that shown in Table 4.

### **Discussion**

We found that 82% of the 28 children in our sample had impaired somatosensation in at least one domain. This frequency of impairment is slightly higher but comparable with other recent findings (31). Not all children had impairment in all assessed somatosensory domains however we did find that more than two thirds of our sample had impairment in two or more domains.

Robust measures of somatosensory function with good discriminative properties are necessary to adequately identify somatosensory impairment in children (15). Our findings support the ongoing development of the sense\_assess© kids as one such tool (24). What is still required is a somatosensory capacity score in addition to discrete somatosensory functions because different combinations of impairment

exist. Whilst it is useful to have specific and detailed information on individual domains of somatosensory function, for the purposes of severity of overall impairment a summary capacity score would be beneficial.

We measured tactile registration at the level of protective touch only and found that 15% of our sample had impairment which is the same as reported for protective touch by Auld and colleagues (31). Tactile discrimination is not routinely assessed in children with HCP, despite previous reports indicating it is frequently impaired (29, 31) consistent with the 67% with reduced tactile discrimination ability in our study. The measures employed differed between studies and a learning effect is possible (15). Using a novel measure of haptic object recognition, we observed impairment in 75% of our population. This result is similar to the seven of nine children with HCP observed to have impairment by Cooper and colleagues (16). Our result is higher than that reported by two studies with larger sample sizes (31, 32) however in light of a recommendation for a more challenging measure of haptic object recognition their results may be underestimates (15).

Wrist position sense was measured using a quantitative approach and we observed reduced performance in 74% of our sample. Our finding is substantially higher than other studies that used a less precise method of measuring limb position such as reporting direction of movement or position matching (3, 16). The higher rate observed in our work most likely reflects the greater challenge of discriminating the position of the wrist and the greater sensitivity of an interval rather than an ordinal or categorical measurement scale (30). The high frequency and functional implications of impairment in limb position sense provide justification to include it as a domain of interest when looking at somatosensory function. The moderate to strong correlations observed between somatosensory discrimination and unimanual motor performance, as assessed by the BBT, provide evidence of the impact of somatosensory impairment on motor performance. This supports the findings of a study of 25 children with HCP that found a strong correlation between measures of tactile sensibility and an object pick-up test (33) and another of 15 children with HCP that found a relationship between tactile sensibility and fine control of fingertip force regulation (34). Our findings are also in agreement with Auld et al's (4) study that reported correlations between spatial tactile deficits and both unimanual and

bimanual performance in 52 children with HCP. Auld et al. (4) wished to determine the movement-free contribution of tactile deficits and excluded consideration of haptic object recognition whereas we are particularly interested in the aspects of somatosensory discrimination that inform movement and so included haptic object recognition. We observed that the combination of impairments was important for motor performance, the higher the number of impaired somatosensory domains the worse the performance on the BBT. To our knowledge this has not been described elsewhere.

There remains a need to determine what level of somatosensory impairment is functionally important. Typically developing children perform well on somatosensory tasks providing a narrow range of typical performance as a benchmark for impairment. However, of those who qualified as having impairment, some did so by only a small margin and it is not known if somatosensory intervention is clinically indicated for mild impairment. Taking the broader context of the child's performance in several somatosensory assessments may be worthwhile, as we found that motor function decreased with increasing numbers of impaired somatosensory domains.

Currently there are no evidence based interventions to improve somatosensory ability for children with HCP (35). There is a growing body of literature recommending interventions to address somatosensory impairment to be investigated in this group of children (5, 35-37). It is not yet known what effect interventions may have on a child's somatosensory function and subsequently hand function. However, with what we know about the role of somatosensory discrimination on motor performance it is essential that we investigate treatment options. The universal use of a standardised somatosensory assessment battery sufficiently sensitive to evaluate change would greatly facilitate comparison of treatment approaches, and for this reason we continue to strive for such an assessment battery.

### ***Limitations***

We did not assess the unimpaired limb of the children in our sample. Previous work has found that over half of children with HCP also have some form of impairment in tactile function of the unimpaired limb, (31) so that the 'unimpaired' limb should be

assessed with a view to intervention rather than providing a control comparison. Study participants were not a population based sample and may not be representative of the broader population of children with HCP with MACS levels I and II.

## **Conclusions**

Eighty-two percent of our sample had impaired somatosensory discrimination function. This result is consistent with the 50%-90% frequency of sensory impairment reported in the literature for children with HCP, although the domains of somatosensory impairment differed as did severity. The knowledge that children with HCP commonly have impairments in somatosensory discrimination function is well established. The expression of these deficits and their influence on function is less well understood. Knowing which aspects of somatosensory discrimination function have the greatest impact on a child's independent functioning and hand use is important in developing targeted interventions. Given the potential impact of somatosensation in the performance of functional tasks, it is essential that therapists routinely assess for somatosensory impairment across multiple domains of somatosensory discrimination function. Interventions to improve somatosensory discrimination function are underrepresented in the literature and future research is needed (4, 36).

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## **APPENDIX G      Pilot Intervention Study**

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PhD candidate Belinda McLean accounted for 85 per cent of the intellectual property associated with the final manuscript, collectively Susan Taylor and the other authors accounted for 15 per cent.



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# Somatosensory Discrimination Intervention Improves Body Position Sense and Motor Performance in Children With Hemiplegic Cerebral Palsy

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**OBJECTIVE.** This study examined the use of the adult neuroscience-based Sense<sup>®</sup> intervention with children with hemiplegic cerebral palsy (HCP) to improve upper-limb somatosensory discrimination, motor function, and goal performance.

**METHOD.** Seventeen children with HCP (9 boys, 8 girls; mean age = 10.2 yr) participated in this pilot matched-pairs trial with random allocation and 6-mo follow-up (intervention,  $n = 7$ ; control,  $n = 10$ ). The intervention group received Sense training 3x/wk for 6 wk (18 hr). Outcome measures included Goal Attainment Scaling, Sense\_assess<sup>®</sup> Kids, and the Assisting Hand Assessment.

**RESULTS.** The intervention group improved in goal performance, proprioception, and bimanual hand use and maintained improvement at 6-mo follow-up. The control group improved in occupational performance by 6-mo follow-up.

**CONCLUSION.** This study established the feasibility of using the Sense intervention in a pediatric setting and adds preliminary evidence to suggest that improving somatosensory function can improve motor function and goal performance among children with HCP.

McLean, B., Taylor, S., Blair, E., Valentine, J., Carey, L., & Elliott, C. (2017). Somatosensory discrimination intervention improves body position sense and motor performance in children with hemiplegic cerebral palsy. *American Journal of Occupational Therapy, 71*, 7103190060. <https://doi.org/10.5014/ajot.2016.024968>

**C**erebral palsy (CP) is an umbrella term for motor impairment arising from a nonprogressive insult to or malformation of the developing fetal or infant brain (Rosenbaum et al., 2007). Children with CP frequently have associated impairments that affect their upper-limb functioning, including somatosensory impairments (Rosenbaum et al., 2007). The prevalence of somatosensory impairments in children with hemiplegia has been reported to be more than 75% (Auld, Boyd, Moseley, Ware, & Johnston, 2012b; McLean, Taylor, Valentine, Carey, & Elliott, 2017). Impairment in tactile or somatosensory function has been shown to have a substantial impact on children's motor function and upper-limb use (Auld, Boyd, Moseley, Ware, & Johnston, 2012a).

*Somatosensory function* is defined as "all aspects of touch and proprioception that contribute to a person's awareness of his or her body parts and the direct interface of these with objects and the environment" (Dunn et al., 2013, p. S41). Somatosensation impairments in children with hemiplegic cerebral palsy (HCP) are commonly reported in the domains of haptic object recognition (or stereognosis), limb position sense (or proprioception) and two-point discrimination. These impairments affect functional sensibility (Majnemer, Bourbonnais, & Frak, 2008). Somatosensation is necessary for fine motor function and the development of dexterous movement for useful hand function (Blennerhassett, Carey, & Matyas,

2006; Majnemer et al., 2008). It has also been suggested that impairments in somatosensation may result in learned nonuse of an affected hand, leading to reduced exploration of the environment, altered sensorimotor development of the affected hand, and impaired bimanual function (Majnemer et al., 2008).

There are currently no evidence-based interventions to improve somatosensory function in children with neurological impairments. However, there is a growing body of evidence-based interventions to improve the somatosensory functioning of adults who have survived a stroke. A recent systematic review of interventions to improve tactile functioning recommended two approaches from the adult stroke literature for use with children (Auld, Russo, Moseley, & Johnston, 2014). One of the recommended approaches was transfer-enhanced training, known as *Sense*<sup>®</sup> *training*, by Carey, Macdonell, and Matyas (2011). *Sense* is based on principles of perceptual learning and learning-dependent neural plasticity and is designed to improve three important aspects of somatosensory discrimination function: tactile discrimination, haptic object recognition, and limb position sense.

In a randomized crossover control trial, *Sense* training was found to improve functional somatosensory discrimination in adult stroke survivors in the chronic phase post-stroke. Of the 50 participants, 59% halved their deficits or better, with improvements maintained at 6-mo follow-up (Carey et al., 2011). The learning principles used in *Sense* training are similar to motor learning principles, which have been shown to improve motor function and structural neuroplastic change in children with HCP (Sterling et al., 2013). Our study sought to pilot a child-friendly version of *Sense* with children with HCP. We hypothesized that the *Sense* intervention would be effective in improving somatosensory discrimination, upper-limb motor function, and occupational goal performance of children with HCP.

## Method

### *Participants*

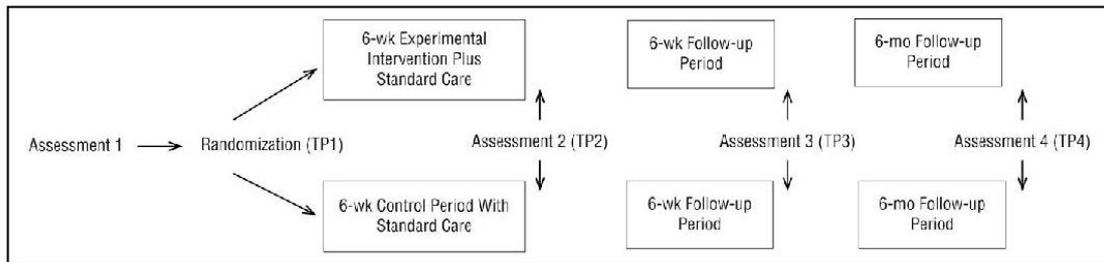
This pilot, single-blind, matched-pairs trial with random allocation recruited children with HCP ages 5–15 yr from a pediatric tertiary hospital in Perth, Western Australia, Australia. Children entered the study on a rolling recruitment basis from May 2013 to May 2015. Baseline assessments took place within 2 wk of each participant's commencement of the trial. Eligibility criteria were a confirmed diagnosis of HCP by a pediatrician and somatosensory discrimination impairment as measured by the *Sense\_assess*<sup>®</sup> *Kids* (Taylor, McLean, et al., 2017). Children were ineligible if they had upper-limb surgery in the previous 12 mo.

Children were assessed at least 12 wk after their most recent botulinum toxin therapy and were pair-matched for age and Manual Ability Classification System level (Eliasson et al., 2006). Children were assessed at baseline (Time Point 1, or TP1), after 6 wk of intervention or control (TP2), at 6-wk follow-up (TP3), and at 6-mo follow-up (TP4; see Figure 1). All assessments were undertaken at each time point, including goal setting and subsequent scoring, by an assessor blinded to group allocation. With the assessor, who was trained to identify tasks involving obvious somatosensory cues, children and their parents set goals important to them that involved the upper limbs. After completing the baseline assessment, children were randomized to a group by a third party not involved with the study.

The treatment group received the experimental *Sense* intervention 3×/wk for 6 wk (total dose = 18 hr) in the child's home or school setting. In both environments, distractions were removed with the assistance of the primary caregiver; in school settings, a quiet room was made available, and the same intervention equipment was used. Each session was 1 hr long and conducted by an occupational therapist trained in the *Sense* training approach. The treatment and control groups continued with their usual care through their local community provider. Families reported setting goals with a community occupational therapist or physiotherapist that involved problem-solving implementation but no formalized intervention.

The intervention protocol closely followed that described by Carey and colleagues (2011) in their Study of the Effectiveness of Neurorehabilitation on Sensation (*SENSe*) trial, with some minor changes to accommodate scheduling sessions in the community setting, predominantly in family homes. Children were trained in only two of the three components of somatosensory discrimination per 1-hr session to allow time for caregiver education. The *Sense* occupation-based training was incorporated into this trial to facilitate children's intrinsic motivation. The *Sense* occupation-based approach to somatosensory retraining involves the application of *Sense* principles in the context of valued activities identified by the participant (Mastos & Carey, 2010). Occupational task performance is an important element of *Sense* and involves emphasizing obvious somatosensory features in activities meaningful to the child, such as recognizing the change in texture that signals the presence of a buttonhole.

The ability to recognize obvious sensory attributes in occupational tasks is practiced in component training. Component and occupational training, as applied in *Sense*, use active exploration with vision occluded; feedback on performance (accuracy and method of exploration);



**Figure 1. Study design with assessment schedule.**

Note. TP = time point.

calibration with vision and with the less affected hand; anticipation trials in which the participant knows what to expect to feel (vision occluded); and repetition and progression from easy to more complex tasks to distinguish differences across a wide variety of stimuli in tactile discrimination, haptic object recognition, and limb position sense (Carey et al., 2011). Tactile discrimination is trained using a variety of graded textured surfaces such as rubber, glass, and leather that vary from smooth to rough with different surface properties. Haptic object recognition is trained with graded object sets that vary in the sensory attribute of interest such as size, weight, or crushability. Limb position sense is trained using graded positions of the wrist and then elbow, starting with the largest differences (e.g., near full flexion, neutral, and near full extension).

#### Primary Outcome

Goal performance was our primary outcome, measured using Goal Attainment Scaling (GAS; Kiresuk, Smith, & Cardillo, 1994) and the Canadian Occupational Performance Measure (COPM; Law et al., 1998). A recent evidence-to-practice commentary by Novak (2014) recommended that clinicians use the COPM and GAS together to monitor change in parent coaching interventions for which the evidence base is not well developed. Our research team has opted to use the GAS and COPM as primary outcome measures because tools for somatosensation are still under development and, from an occupational therapy perspective, our primary interest was whether the intervention facilitates improvements in occupational performance.

#### Secondary Outcomes

Somatosensation was measured using the *Sense\_assess Kids* (Taylor, McLean, et al., 2017). The assessment comprises three subtests that measure functional somatosensory discrimination ability. The tool was developed for use with adult stroke survivors (Carey et al., 2011) and adapted for use with children and adolescents. Two of the three components of the *Sense\_assess Kids*, the functional

Tactile Object Recognition Test and the Wrist Position Sense Test, were examined in this study; the third, tactile discrimination, was not. Tactile discrimination was instead measured using two alternative methods, which in this pilot trial resulted in insufficient data for analysis.

The functional Tactile Object Recognition Test is a standardized 14-item test of haptic object recognition in which children are presented with familiar and novel items out of sight and indicate what they are exploring using a response poster (Carey, Nankervis, LeBlanc, & Harvey, 2006). The Wrist Position Sense Test is a measure of wrist position sense in which a child's hand is moved through 20 standardized positions on a protractor scale out of the child's sight (Carey, Oke, & Matyas, 1996). The child indicates where the hand is pointing using a second protractor scale positioned in view immediately above the occluded hand. Psychometric testing continues, but reliability for the Wrist Position Sense Test has been determined for typically developing children using the Bland–Altman method (Bland & Altman, 2010). Using the Bland–Altman method, Taylor, Parsons, et al. (2017) determined that there were no consistent differences between time points, indicating the measure's reliability. The *Sense\_assess Kids* has normative standards for typically developing children ages 6–15 yr and has demonstrated construct validity and clinical acceptability for children with CP ages 6–15 yr (Taylor et al., 2015; Taylor, McLean, et al., 2017).

Motor performance was measured using the Assisting Hand Assessment (AHA; Krumlinde-Sundholm & Eliasson, 2003) and the Box and Block Test (B&B Test; Mathiowetz, Federman, & Wiemer, 1985). The AHA is a measure of how a child with HCP uses the more impaired hand in bimanual activities (Krumlinde-Sundholm & Eliasson, 2003). It has good construct validity and excellent test-retest reliability (.99), and it is responsive to change when used to assess children ages 18 mo–18 yr (Holmefur, Aarts, Hoare, & Krumlinde-Sundholm, 2009). The AHA is conducted as a play session and is video recorded for scoring at a later time.

The B&B Test (Mathiowetz et al., 1985) measures gross manual dexterity and unimanual capacity. In the test administration, a child is seated in front of a wooden box that has a central partition with blocks on the same side as the hand to be tested. Children are instructed to move as many blocks as they can, one at a time, in 1 min from one side of the partition to the other, ensuring that their fingertips pass over the partition. The B&B Test is a valid, reliable measure commonly used in intervention studies of children with CP (Platz et al., 2005). The AHA and B&B Test were selected to observe for changes in motor performance after the Sense intervention.

### Statistical Analysis

Because of the small number of participants and variability in baseline scores, raw scores were converted to individual change scores by subtracting the baseline score from each time point score. The Mann-Whitney *U* rank-sum test (Portney & Watkins, 2009) was used to determine the presence of between-groups differences in individual change scores for each measure. Friedman  $\chi^2$  repeated-measures analysis (Portney & Watkins, 2009) was used to identify the statistical significance of within-group differences, and the Wilcoxon signed-rank test (Portney & Watkins, 2009) was used to find the direction of any identified differences. Effect size is reported using *r* to describe a small ( $\geq .10$ ), medium ( $\geq .30$ ), or large ( $\geq .50$ ) effect (Cohen, 1992).

## Results

Seventeen children ages 6–15.5 yr were recruited for this study. Children were randomly allocated to the control ( $n = 10$ ) or intervention ( $n = 7$ ) group. The treatment group had fewer children as a result of the randomization process. We were not able to recruit suitable matches for each participant for the matched-pair design before cessation of this preliminary investigation. See Figure 2 for recruitment flowchart.

### Goal Performance

Goals in both groups were similar and included using a knife and fork, doing up buttons or a zipper, putting hair in a ponytail, and throwing and catching a ball. The somatosensory cues important to successful completion of each of these activities can be readily observed by a trained clinician. During the intervention, obvious somatosensory cues related to limb position, texture differences, and identifying when the hand was in contact with objects important for task performance were graded and emphasized for each task.

*Goal Attainment Scaling.* Significant between-groups differences existed that favored the intervention group on GAS at TP2 ( $r = .79, p < .01$ ) and TP3 ( $r = .58, p = .02$ ) but were not present at TP4. Within-group differences existed in both the intervention and the control groups from TP1 to all subsequent time points, with a greater magnitude of change in the intervention group (see Table 1). At TP2, the intervention group had achieved 12 of their 14 (86%) goals, and the control group had achieved 4 of their 20 (20%) goals, an expected or greater than expected outcome. The intervention group maintained their goals, and the control group achieved 10 of their goals (50%) at TP4.

*Canadian Occupational Performance Measure.* Similar between-groups and within-group differences were observed in COPM performance (see Table 1). Groups differed on COPM satisfaction, in that between-group differences existed only at TP2 ( $r = .5, p = .04$ ).

### Somatosensory Discrimination

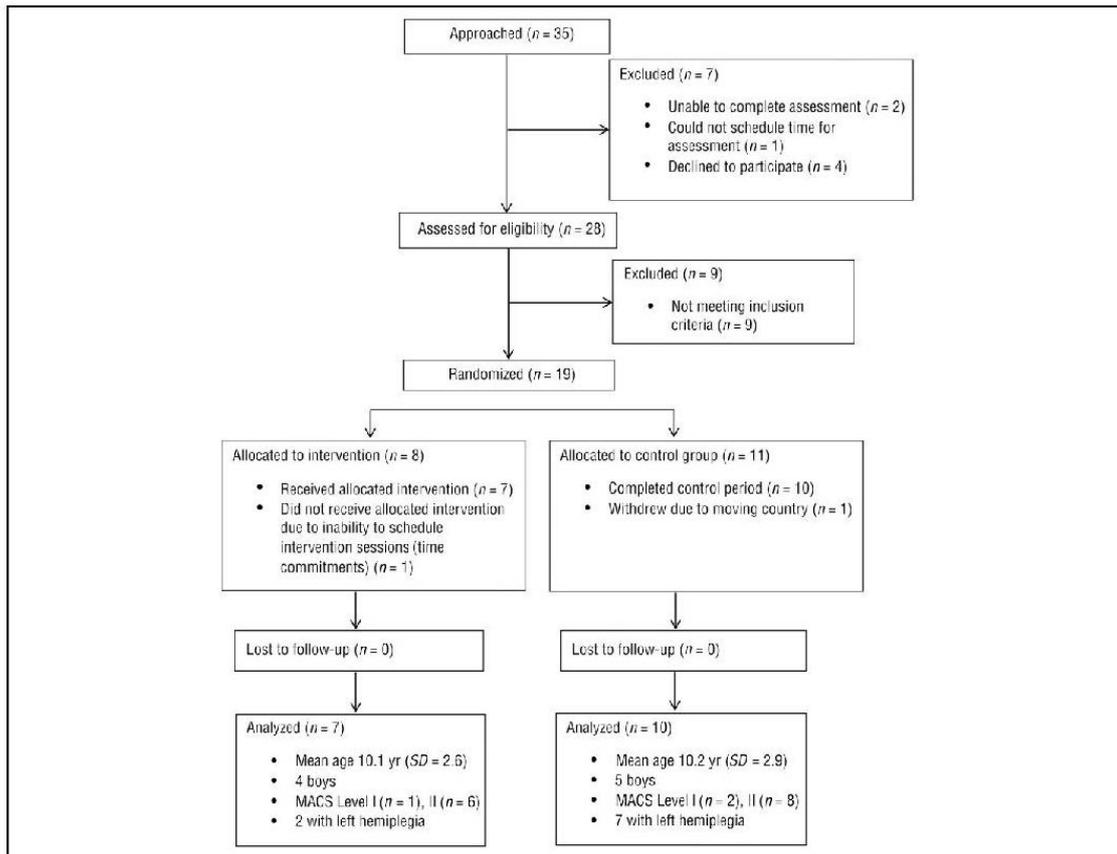
No significant between-groups differences were found across time points on the Wrist Position Sense Test. The intervention group had within-group differences indicating that improvements were maintained at TP4. No differences were found across time points for the control group (see Table 1). There were no significant between-groups differences across time points for the functional Tactile Object Recognition Test, but there was a significant within-group difference for the intervention group at TP2 (median [*Mdn*] = 2,  $r = .54, p = .04$ ) and the control group at TP3 (*Mdn* = 3,  $r = .47, p = .04$ ).

### Motor Performance

No significant between-groups differences were found across time points for the AHA, but the intervention group had within-group differences that indicated improvements were maintained at TP4 (see Table 1). No significant differences were found across time points for the control group. For the B&B Test, there were no significant between-groups differences across time points, but there was a significant within-group difference for the intervention group at TP3 (*Mdn* = 3,  $r = .64, p = .02$ ) and TP4 (*Mdn* = 6,  $r = .59, p = .03$ ) and for the control group at TP3 (*Mdn* = 1.5,  $r = .54, p = .02$ ) and TP4 (*Mdn* = 4,  $r = .60, p = .01$ ).

## Discussion

This study was an early investigation of a somatosensory discrimination intervention for children with HCP that used an established intervention protocol from another clinical group that was modified by this research team for use with children with HCP. Improvements were observed



**Figure 2. Recruitment flowchart.**

Note. MACS = Manual Ability Classification System; SD = standard deviation.

in the children's somatosensory discrimination, motor performance, and goal attainment.

Goal performance can be improved using a somatosensory discrimination approach. Children in our intervention group showed improvement on their goals immediately postintervention; they achieved an expected or greater than expected outcome on 86% of their GAS goals and maintained these improvements at the 6-mo follow-up. Children in the control group achieved an expected or greater than expected outcome on 20% of their goals immediately after the control period, increasing to 50% at the 6-mo follow-up. The act of goal setting can motivate self-action, particularly when goals are chosen by the person and described in detail (Locke & Latham, 2002). Goal setting alone has been found to direct attention to activities that will support the achievement of goals important to the person (Locke & Latham, 2006). This mechanism may account for some of the goal attainment observed in our control group.

In addition to self-selecting important goals, our intervention group had structured opportunity for practice with feedback and opportunities to enhance their sense of self-efficacy by demonstrating new skills to family members, which is also important for goal progress (Locke & Latham, 2002). The greater magnitude of and immediate response with maintenance of gains observed in the intervention group suggest that the intervention facilitated improvement. The improvement observed in the control group was smaller, but gains were still observed at long-term follow-up and may be explained by increased attention to important self-selected goals.

It is possible to improve and maintain improvement in limb position sense after Sense discrimination training. We found that limb position sense improved in the intervention group and was maintained at 6-mo follow-up. This improvement involved reduced variability and a steady improvement in the group median over time. A previous study showed that body position sense can be improved in the short

**Table 1. Within-Group Comparisons of Median Change Between Baseline and Each Time Point on Selected Outcome Measures**

Group and Result Type	Baseline (TP1)	TP2	TP3	TP4
GAS				
Control				
<i>Mdn</i>	0	4.56	9.3	24.81
<i>r</i>		.479	.53	.597
<i>p</i>		.042*	.018**	.011**
Intervention				
<i>Mdn</i>	0	37.2	37.2	37.2
<i>r</i>		.638	.634	.638
<i>p</i>		.017**	.018**	.017**
COPM Performance				
Control				
<i>Mdn</i>	0	0.7	1.3	3.0
<i>r</i>		.254	.547	.557
<i>p</i>		.256	.014**	.013**
Intervention				
<i>Mdn</i>	0	4.6	3.5	3.4
<i>r</i>		.632	.634	.635
<i>p</i>		.018**	.018**	.018**
Sense_assess Kids WPST				
Control				
<i>Mdn</i>	0	2.3	-0.2	1.5
<i>r</i>		.199	.125	.285
<i>p</i>		.374	.575	.203
Intervention				
<i>Mdn</i>	0	4.45	6.45	7.7
<i>r</i>		.632	.41	.632
<i>p</i>		.018**	.128	.018**
AHA				
Control				
<i>Mdn</i>	0	-1.0	2.0	2.5
<i>r</i>		.040	.205	.146
<i>p</i>		.859	.358	.515
Intervention				
<i>Mdn</i>	0	4.0	14.0	18.0
<i>r</i>		.588	.587	.587
<i>p</i>		.028*	.028*	.028*

Note.  $r \geq .10$  indicates a small effect size;  $\geq .30$ , a medium effect size; and  $\geq .50$ , a large effect size. AHA = Assisting Hand Assessment; GAS = Goal Attainment Scaling; COPM = Canadian Occupational Performance Measure; *Mdn* = median; TP = time point; WPST = Wrist Position Sense Test. \* $p < .05$ . \*\* $p < .02$ .

term by practicing a matching task, but the improvement was not maintained at 1-wk follow-up (Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2013). In this single practice session, integrating limb position sense with a meaningful activity was not considered, nor did the session meet the intensity requirements expected to effect lasting change (Sakzewski, Provan, Ziviani, & Boyd, 2015). Our approach to improving limb position sense was intensive and multimodal, and it involved calibration with and without vision and incorporated obvious limb position cues into children's

goal performance. The variety and intensity of practice paired with the structured focus on somatosensory discrimination may be the important difference in our approach.

The intervention group demonstrated a statistically significant improvement in median object recognition score and reduced within-subject variability immediately post-intervention, which was not maintained at 6-wk follow-up. The control group demonstrated a statistically significant improvement in haptic object recognition between baseline and the 6-wk follow-up measure, which did not replicate the reduced variability of the treatment group and was not maintained. A recent study by Kuo and colleagues (2016) found improvement in tactile outcomes after 82 hr of Hand Arm Intensive Bimanual Training with an additional 8 hr of somatosensory discrimination tasks with objects and shapes. They found that both their treatment (structured tactile tasks, vision occluded) and control (unstructured handling of tactile items, with vision) groups improved on tactile measures, and the authors suggested that the mechanism behind these changes could have been the intensive bimanual training alone or the incorporation of a diverse range of shapes and textures. No follow-up data were presented. The variability in our groups meant that we also had no between-groups differences, and whether the changes in the control group were the result of a learning effect is unclear. However, the absence of a reduction in variability suggests this is not the case, and the agent responsible for the change requires further investigation.

The Sense somatosensory discrimination intervention was associated with an improvement in motor function. Although there were no significant between-groups differences on the AHA, our intervention group demonstrated improvements immediately postintervention that were maintained at 6-mo follow-up, and our control group showed no change over time. Our data suggest that improvements in limb position sense may contribute to improved motor performance because the results on our measure of limb position sense mirrored the AHA results.

Kuo et al. (2016) demonstrated improved motor performance on the AHA as well, but improvement was observed in both treatment conditions, and the authors determined that whether the tactile training contributed to improvements in motor performance was not clear. We did not see the same improvement on the B&B Test as we did on the AHA, and both the intervention and the control groups made modest improvements over time. The differences observed between these assessments may be due to differences in the focus of measurement; the AHA measures how the more affected hand is used in bimanual tasks, and the B&B Test solely measures unimanual performance. An

important direction for future study will be to understand the elements of somatosensation that most contribute to gains in motor function. This study suggests that limb position sense is modifiable and plays an important role in upper-limb use of children with HCP.

Individual variation was observed in both the treatment and the control groups and may have contributed to the absence of statistically significant between-groups change. Variability such as we observed has been reported in studies examining intensive interventions for the upper limb (Eliasson, Krumlinde-Sundholm, Shaw, & Wang, 2005; Hoare et al., 2013; James, Ziviani, Ware, & Boyd, 2015; Sakzewski, Ziviani, & Boyd, 2011). In these examples, the effectiveness of the intervention was confirmed by within-group analysis while acknowledging the presence of variance. As recommended in a systematic review of upper-limb interventions for children with HCP, it is crucial that future studies examine child characteristics such as severity of impairment, side of hemiplegia, motivation, cognition, and cortical sensorimotor reorganization for their contribution to treatment outcome (Sakzewski, Gordon, & Eliasson, 2014).

## Limitations

This preliminary investigation of Sense training with children considered a very small sample. To accommodate this sample size, individual differences were investigated, and nonparametric statistics were used. Goal attainment and occupational performance were the primary outcomes considered because there is no gold-standard measure of somatosensation in children with CP. We consider the results of this study to be preliminary, and they should be used to orient the direction of future work. It is possible that this intervention did not meet adequate dosage requirements. Children in the intervention group received 18 hr of intervention in contrast to motor learning approaches, which demonstrate change with doses ranging from 30 to 60 hr. Whether higher doses are required for a somatosensory approach is not yet known. Larger samples and systematic manipulation of factors such as dosage are required.

## Implications for Occupational Therapy Practice

This study's findings have the following implications for occupational therapy practice:

- The sense somatosensory discrimination intervention improves children's body position sense, motor outcomes, and goal attainment, and improvements were maintained at long-term follow-up.

- Body position sense is modifiable and may be linked to improvements in motor outcomes.

These findings are preliminary, and further investigation is required to confirm them.

## Conclusion

This is the first pediatric trial of a novel somatosensory discrimination intervention with demonstrated efficacy in adult stroke survivors, and it provides evidence of feasibility and potential efficacy in children with HCP. Using a small pilot sample, we observed that it is possible to improve limb position sense, motor performance, and attainment of occupational goals and maintain improvements at 6-mo follow-up. Building on the knowledge that somatosensation is important for motor function, we provide preliminary evidence to suggest that improving somatosensory function can improve motor and goal performance outcomes for children with HCP. Occupational therapy practitioners are encouraged to consider using evidence-based approaches to improving somatosensory function to help children with HCP achieve their functional goals. Our findings suggest that applying the Sense approach is feasible, but its use would need to be closely monitored until further high-level evidence is available. ▲

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## **APPENDIX H      Intervention Protocol**

**For submission to: BMC paediatrics; impact factor 1.81**

PhD candidate Belinda McLean accounted for 85 per cent of the intellectual property associated with the final manuscript, collectively Susan Taylor and the other authors accounted for 15 per cent.

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**Discovering the sense of touch: Protocol for a randomised controlled trial examining the efficacy of a somatosensory discrimination intervention for children with hemiplegic cerebral palsy**

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## Abstract

**Introduction:** 75% of children with hemiplegic cerebral palsy (HCP) have impaired somatosensory function, which contributes to learned non-use of the affected upper limb. Currently, motor learning approaches are used to improve upper-limb motor skills in these children, with few studies having examined the effect of any intervention to ameliorate somatosensory impairments. Recently, *Sense*© training was identified as a goal directed intervention designed for adults that demonstrated statistically and clinically significant change in limb position sense, goal performance and bimanual hand-use in a pilot study of seven children with HCP. This paper describes a protocol for a Randomised Controlled Trial (RCT) of *Sense*© *for Kids* training, hypothesising that its receipt will improve somatosensory discrimination ability more than placebo (dose-matched Goal Directed Therapy via Home Program). Secondary hypotheses include that it will alter brain activation in somatosensory processing regions, white-matter characteristics of the thalamocortical tracts and improve bimanual function, activity and participation more than participants who received Goal Directed Therapy via Home Program.

**Methods:** A single blind, randomised matched-pair, placebo-controlled trial will randomly allocate participants aged 8 to 15 years with a confirmed description of hemiplegic CP (HCP) and somatosensory discrimination impairment, as measured by the *sense*©\_*assess kids*, to receive 3 hours a week for 6 weeks of either *Sense*© *for Kids* or Goal Directed Therapy via Home Program. Children will be matched on age and severity of somatosensory discrimination impairment. The primary outcome will be somatosensory discrimination ability, measured by *sense*©\_*assess kids* score, secondary outcomes will include degree of brain activation in response to a somatosensory task, measured by changes in the white matter of the thalamocortical tract, measured by diffusion magnetic resonance imaging (MRI), bimanual motor function, activity and participation.

**Discussion:** This study will assess the efficacy of an intervention to increase somatosensory discrimination ability in children with HCP and answer clinically important questions about the efficacy of intervening in somatosensation impairment to improve motor function compared with focusing on motor impairment directly and whether focusing on motor impairment alone can affect somatosensory ability.



## **Introduction**

Cerebral palsy (CP) is the most commonly occurring childhood physical disability, and is an umbrella term covering a variety of aetiologies with a combined prevalence of roughly 2.1 per 1000 live births (1). It is defined by motor impairment arising from an injury or malformation of the developing brain and is often accompanied by comorbidities such as impairment in sensation, perception, cognition, communication, and behaviour (2). Hemiplegic CP (HCP) is the most commonly occurring motor impairment subtype (3) and negatively impacts upper limb function. Recent reports indicate that more than 75% of children with HCP have impaired somatosensory function (4, 5).

Somatosensory function involves the detection, discrimination, and recognition of body sensations (6). According to the National Institutes of Health toolbox, somatosensation refers to *“all aspects of touch and proprioception that contribute to a person’s awareness of his or her body parts and the direct interface of these with objects and the environment”* p. S41 (6). This includes body position sense, haptic object recognition, and tactile discrimination (6). Somatosensation guides motor function in a feed forward manner: the more a child can perceive, the more they explore (move), and the more they can understand and interact with their environment (7, 8). Ascending somatosensory neural pathways provide tactile and proprioceptive information (9). By monitoring these forms of information, the central nervous system can adjust signals to descending motor pathways during grasp and associated manipulation of objects (10). In the upper limbs, both fine motor movements and tool use rely heavily on such feedback (7, 10, 11).

A clear link exists between somatosensory deficits and poor hand function in children with HCP (10, 12). This was recently demonstrated in a cross-sectional study by Auld et al. (12) where a moderate relationship between tactile function and hand performance was identified. Specifically, haptic object recognition and single point localisation had the greatest influence on unimanual capacity while haptic object recognition and recognition of double simultaneous stimulation had the greatest influence on bimanual function. In this study, impairment in somatosensory function accounted for one third of the variance in motor function (12). The significant contribution of somatosensation to motor function indicates that

therapeutic interventions that target somatosensation may have the potential to improve motor function in children with HCP.

It is recognised that damage to corticomotor tracts and thalamocortical sensory pathways both contribute to upper limb motor impairment in hemiplegia (13-15). Children with hemiplegia have different patterns of brain activation than typically developing peers during somatosensory tasks (16, 17). The reorganisation of motor pathways is well documented in children with hemiplegia, with a subset showing evidence of persistent and predominant ipsilateral motor pathway control of hand movements (18-27). Such reorganization is not always functionally advantageous: a noted decline in affected upper limb function is associated with the persistence of ipsilateral pathways in children who sustained injury in late gestation (27). However, studies investigating somatosensation using magnetoencephalography (MEG), functional magnetic resonance imaging (fMRI) and somatosensory evoked potentials (SEP) of the affected side have demonstrated that activation of the primary somatosensory cortices is still predominantly contralateral, and the contralateral pathway still functions, albeit with altered responses (16, 18, 28-32). This “interhemispheric dissociation” between somatosensory inputs and motor outputs may be a significant contributing factor to the impaired integration of sensorimotor function in a subset of children with hemiplegia (18).

Neuroplastic changes associated with improvement have been demonstrated in motor function following motor learning approaches such as constraint induced movement therapy (33). Several studies have provided a neurological basis for pursuing somatosensory intervention to improve upper limb function in children with HCP by demonstrating somatosensory pathways are active, albeit disorganised, and therefore possibly treatment responsive (17, 34). The core principles which inform motor learning approaches to upper-limb therapy are the same as principles of learning dependent neural plasticity such as repetition of a challenging but achievable task, practice repetition and feedback on performance (35, 36). It is reasonable to expect that when such principles are applied in a somatosensory intervention, neural plastic changes in the sensory regions of the brain will also be observed.

Upper limb function, is recognised by experts as a high priority area for treatment of children with hemiplegia (37). A large body of research has investigated therapeutic interventions and modes of delivery to maximise outcomes for this group of children (38). Recent research has predominantly focused on improving motor skills via motor learning approaches and has demonstrated that intensive goal-directed treatments have a positive effect on hand function (38). However, there is limited research into whether reducing developmental non-use and improving bimanual hand function might be more effectively achieved by treating any sensory impairments that are known to contribute to impaired motor function. A recent systematic review of interventions for tactile deficits that may be suitable for children suggested two approaches that were effective in adults post stroke (39). This study aims to investigate one of those recommended: *transfer enhanced somatosensory discrimination training*, known as *Sense©* training (36).

The principles of *Sense©* training stem from theories of perceptual learning and learning dependent neural plasticity (36). *Sense©* training involves repeated practice discriminating between graded stimuli in the somatosensory domains: body position sense, haptic object recognition and tactile discrimination using specially designed training tasks (36). In an RCT with cross over control, *Sense©* training was found to improve somatosensory discrimination function in adults (n = 50) who were a median of 48 weeks post stroke (36). In this trial, 59% of stroke survivors at least halved their somatosensory deficits six months' post treatment. Survivors also achieved transfer of training effects to untrained tasks. Seven training principles are operationalized in the training protocol: selection of training tasks; goal-directed attentive exploration of sensation without vision; feedback on the accuracy and method of exploration by therapist/vision; calibrate somatosensory perception via vision and/or touch of the unaffected hand; use of deliberate anticipation trials; variety of sensory tasks and practice conditions to facilitate transfer; and repeat and progress, as outlined in the training manual (40) and online video (41). *Sense©* is also applied to client-selected activities (occupations), with the aim for the client to learn strategies in how to use somatosensory skills to perform the activity most optimally and to transfer these strategies and skills learnt to untrained activities (40).

Hemiplegia can arise in infants with a variety of neurological pathologies such as white matter injuries, grey matter injuries, malformations of the brain as well as focal vascular insults (seen in ~9%) and no cerebral pathology can be identified on imaging in about the same proportion (42). It cannot be ignored that these aetiologies are highly varied in comparison to adult stroke survivors. Furthermore, most children with HCP have a somatosensory system that has never functioned normally in the extra-uterine world while an adult stroke survivor has received an insult to a previously well-functioning system. Despite these population differences, pilot work for this study demonstrated that *Sense*© training is feasible with children with HCP and warrants further investigation (43).

During our pilot, matched pairs, controlled trial, *Sense*© training was modified to increase suitability for a paediatric population of children with HCP (43). The principles of training remain the same and children progress through the same levels of graded somatosensory training as adults (36). To facilitate child engagement with the *Sense*© training, the principles of self-determination theory and family centred service were incorporated into the provision of *Sense*© *for Kids* training (44, 45). To improve the relevance of *Sense*© *for Kids* training to children with HCP and their families further modifications were implemented following consumer engagement (46). Focus groups and interviews were conducted and feedback from children and their families were integrated into changes to *Sense*© *for Kids* training. A consumer representative (EB) also vetted all aspects of this protocol paper and details of the intervention. These changes are aimed at reducing the scheduling demands on families and increasing the education provided to parents. Parent coaching will be used to facilitate maximal carryover of the benefits of therapy into everyday life following the completion of the formal intervention period (47).

Our pilot work suggests that children improve in trained somatosensory domains, motor performance, and in trained occupational tasks (43). A qualitative investigation of parent and child engagement suggests that improvements were also observed in untrained tasks requiring bimanual function. Improvements following *Sense*© *for Kids* training were maintained six months after training ceased and warrant further investigation with a larger sample (47).

In order to test the efficacy of the *Sense© for Kids* training, a “best practice” comparison intervention will be used to provide adequate control for ‘dosage’ and maintain the external validity of this trial (48). Further, it is considered unethical to withhold potentially effective interventions in controlled comparison conditions and Goal Directed Training delivered via Home Program is an evidence based intervention (38, 49) with a green light on the traffic light system of evidence for children with HCP (50). Because there are no evidence based somatosensory discrimination interventions for comparison, Goal Directed Training via Home Program will act as our control. Goal Directed Training is a motor learning approach which uses a child’s goals to allow problem solving and indirectly elicit movements needed to complete a task but does not include any direct somatosensory training: it is therefore a placebo intervention incorporating common features of *Sense© for Kids* training but no direct somatosensory training (51).

## **Method**

### ***Study design***

A single blind, matched pair, prospective randomised placebo-controlled trial with parallel groups is proposed comparing the effects of *Sense© for Kids* discrimination training with a dose matched, therapist supported Goal Directed Training via Home Program. The primary outcome measure is the *sense©\_assess kids* to assess changes in somatosensory discrimination. The *sense©\_assess kids* measures tactile registration, tactile discrimination, haptic object recognition, and body position sense of the upper-limb in children (52). The secondary outcome measures are brain imaging including functional magnetic resonance imaging (fMRI) and diffusion MRI to observe central nervous system (CNS) changes in response to intervention, the Assisting Hand Assessment (AHA) (53) to measure bimanual ability, Goal Attainment Scaling (GAS) (54) and the Canadian Occupational Performance Measure (COPM) (55) to monitor change in children’s self-selected goals.

## **Interventions**

### ***Sense© for Kids training description***

*Sense© for Kids* training is a structured and graded intervention program based on *Sense©* somatosensory discrimination training (36, 40). *Sense© for Kids* training will

be implemented in this study, as informed by the pilot work that explored the efficacy of *Sense*© somatosensory discrimination training with children with hemiplegia (43). *Sense*© for Kids training uses principles of perceptual learning and learning dependent neural plasticity to develop somatosensory discrimination capacity in aspects of sensation (56, 57). The aspects of somatosensation trained are body position sense, haptic object recognition and tactile discrimination. The principles of training are the same as in *Sense*© discrimination training (36) and include active exploration without vision, feedback on accuracy and method of exploration, anticipation trials, calibration with the less affected hand and with vision, repetition and progression from large to finer differences and transfer to occupational tasks. The equipment and training levels are based on the work of Carey et al. (36, 40), see Table 1 for details of the intervention.

### ***Goal Directed Home Program***

This study will follow current best practice descriptions of Goal Directed Training and be delivered using the model home program approach outlined by Novak and Cusick (58). See Table 1 for details of the intervention.

Table 1 TIDieR Guidelines comparing experimental and control interventions

Item	EXPERIMENTAL INTERVENTION	CONTROL INTERVENTION
Name	<i>Sense</i> © for Kids	Goal Directed Training via a Home Program
Why	<p><i>Rationale:</i> The ability to gain a sense of touch and use this information in goal-directed use of the arm and daily activities is supported by theories of perceptual learning and neural plasticity and may be enhanced by addressing somatosensory discrimination functions through intervention (36, 57). <i>Sense</i>© for Kids is a structured and graded intervention program based on <i>Sense</i>© somatosensory discrimination training (36).</p> <p><i>Theory: Underlying principles of Sense</i>© Principles of perceptual learning and learning-dependent neural plasticity inform <i>Sense</i>© training principles. <i>Sense</i>© is based on seven principles (41), with the theory underlying three core principles outlined. Goal directed attention and deliberate anticipation are important for learning and to facilitate links to somatosensory regions of the brain. Calibration across and within modality improve and create new somatosensory neural connections. Graded</p>	<p><i>Rationale:</i> Children with CP learn movements best when they are engaged in practicing real-life activities that are meaningful to them, based on self-identified goals and practice occurs in real-life environments where they are most comfortable. Parents are experts in knowing their own child.</p> <p><i>Theory: Underlying principles of Goal Directed Training</i> Dynamic systems theories of motor control, where movement emerges as a result of the interaction between the person’s abilities, the environment and their goal</p> <p><i>Underlying principles of Home Programs</i> The therapist coaches parents to build confidence and capabilities The child and parents are more motivated by self-set goals Programs set up in the home environment are more realistic Practice is embedded in family routine to</p>

Item	EXPERIMENTAL INTERVENTION	CONTROL INTERVENTION
	<p>progression within and across sensory attributes and tasks are used facilitate perceptual learning and transfer to novel stimuli (57).</p> <p><i>Sense© for Kids Essential Elements:</i></p> <p>Active exploration without vision of new and known stimuli where the child explores objects/ textures/ body positions with focus on discriminating differences.</p> <p>Anticipation is used for previously experienced stimuli; the child knows what to expect to feel and concentrates on attributes of difference without vision.</p> <p>Calibration occurs within and across modalities with comparison of what is felt by the impaired hand with the less affected hand and with vision. The child matches what they know from visual confirmation and calibration with the less affected hand with their impaired hand. They are prompted to imagine what the sensory stimulus is supposed to feel like based on this knowledge.</p> <p>Each level of stimulus difference is trained to an accuracy level of 75% correct responses before progressing to a more difficult level of difference.</p> <p>Transfer to untrained tasks is facilitated by training on a large variety of stimuli and integrating training principles into occupational tasks important the child.</p> <p>Occupational tasks are trained using grading of stimuli, feedback on distinctive features of difference and method of exploration. Additional information can be found in <i>SENSE: A Manual for Therapists</i> (40).</p>	<p>ease caregiver burden and so as to adapt the parenting style to the child</p> <p>Regular support and the evaluation of outcomes increases practice</p> <p><i>Goal Directed Training Essential Elements:</i></p> <p>Child-set goals about real-life activities the child wants or needs to perform and are deemed realistic for intervention</p> <p>Examination of the goal-limiting factors at the child, task and environment level</p> <p>Changing the task and environment to facilitate child-active independence task performance</p> <p>Establishment of a child-active motor practice schedule at the “just right challenge”, including intense repetition and variation.</p> <p><i>Home Program Essential Elements:</i></p> <p>Development of a collaborative partnership characterised by empowerment of parents</p> <p>Therapist is the coach, the parent carries out the intervention at home but adjusts it using their parental expertise to their unique child</p> <p>Goals are set by the child and parent</p> <p>A range of activities using Goals Directed Training principles are written down designed to help their child reach their goal</p> <p>Therapists actively support implementation to ensure the program continues to meet family needs and help identify successes (58).</p>
Materials	<p>Therapist: The <i>Sense©</i> training kit will be required to train the individual components of sensation. Materials for practice relating to occupational goals will vary depending on the child’s goal e.g. <i>If the goal is using a knife and fork, food items with varying textures will be required that provide the right level of difference of somatosensory feedback during cutlery use.</i></p> <p>A log book will be provided to all families as a reminder to complete home practice incorporating sense© principles into child’s goals, and as an opportunity to increase the challenge as the child improves.</p>	<p>A log book will be provided to all families as a reminder to practice, and as an opportunity to update the home program as the child improves.</p> <p>Materials for each child will vary depending on the child’s goal and which elements of the task and environment are being changed to enhance independent performance e.g. <i>If the goal is catching a tennis ball, materials required may initially include balloons and then light large balls as task modifications to facilitate catching practice at the “just right challenge”.</i></p>
Who	<p>CHILD: Sets goals</p> <p>THERAPIST: Identifies deficit in somatosensory function and works with child through component training in relevant domains (body position sense,</p>	<p>CHILD: Sets goals</p> <p>THERAPIST: Determines goal limiting factors and partners with the parent to develop a home-based practice schedule. Also offers coaching and support via home</p>

Item	EXPERIMENTAL INTERVENTION	CONTROL INTERVENTION
	haptic object recognition, tactile discrimination). Supports parent with incorporating sense© principles into child's goals. PARENT: Incorporates sense© principles into child's goal.	visits PARENT: Carries out the intervention with the child.
How	Home based	Home based
How Much	The total dose of <i>Sense© for Kids</i> will be three hours per week for six weeks with a home visit from a therapist for two hours a week and the family undertaking the remaining one hour of incorporating sense© principles into goal practice. (same dose)	The total dose of this intervention will be three hours per week for six weeks with a home visit from a therapist one hour a week and the family undertaking the remaining accumulative two hours per week of practice. (same dose)
Tailoring	Because children will set their own goals, the activities pertaining to the goal itself may differ but in all other aspects this intervention will remain the same for all participants.	Because children will set their own goals, the activities pertaining to the goal itself may differ but in all other aspects this intervention will remain the same for all participants.
How Well	This study will seek to define and measure fidelity of the <i>Sense© for Kids</i> intervention for: Clinician adherence to active ingredients Intervention receipt There is a home program component of <i>Sense© for Kids</i> training which focuses on incorporating somatosensory cues into occupational task performance and the facilitation of goal attainment by utilising these somatosensory cues within tasks.	This study will seek to define and measure fidelity of Goal Directed Therapy via Home Programs for: Clinician adherence to active ingredients Intervention receipt It is acknowledged that children receiving home programs will have incidental exposure to sensory stimuli through movement and interaction with objects during purposeful activity, however these stimuli will not be emphasised nor will the process of making sense of these somatosensory stimuli.

### **Treatment fidelity**

Two different types of intervention fidelity will be evaluated in this study. The first will assess clinician adherence to the active ingredients of each intervention protocol. Fidelity checklists containing the active ingredients of the respective intervention protocols have been developed to monitor treatment delivery against a priori criteria (see Appendix A) (59). Each criterion will be measured against a four-point Likert scale. Adherence to the intervention approach will be determined by the computation of a percentage score (60).

Each intervention session will be video recorded. Assessment of intervention fidelity will include the random selection of 10% of the recorded intervention sessions, and observed by independent third-party reviewers trained in

both intervention protocols. A fidelity rating of no less than 80% will be required to consider the intervention delivered to the intervention prototype (i.e. with fidelity).

The second fidelity measure is aimed at intervention receipt (59). This will be monitored through completion of home practice logs. Participants will be provided with a log book to record practice sessions and note challenges and successes. In addition, parents will be asked to video record their occupational sessions for review, feedback and problem solving with respect to the active ingredients of the respective intervention protocols. These sessions will be reviewed with the treating therapist during home visits. Parents will be asked to use readily available technology such as their mobile phone, if available, for the express purpose of feedback.

### ***Ethical Considerations***

The study will be undertaken at Perth Children's Hospital, the sole tertiary children's hospital in Western Australia. This study has been prepared in accordance with the principles and mandates set out in the Declaration of Helsinki 2008. Ethics approval has been obtained for this study through Tertiary Hospital Human Research Ethics Committees (ethics number 2014034). Parents and children will be provided with oral and written information about the study with opportunity to have their questions clarified before providing written assent/consent. Randomisation will occur after eligibility assessment. Children who receive botulinum toxin therapy will continue to receive this treatment, however their baseline assessments will be timed at least twelve weeks post their most recent Botulinum toxin-A injections and these treatments will be recorded.

### ***Primary and Secondary Objectives***

Our primary objective is to determine whether *Sense© for Kids* training, a somatosensory discrimination intervention, is more effective than placebo (Goal Directed Training via Home Programs) in improving somatosensory discrimination in children with HCP.

The specific hypotheses to be tested are:

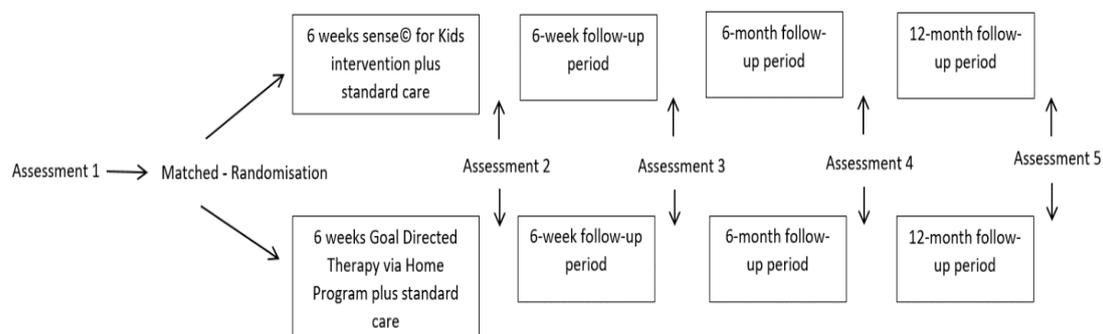
- Children receiving six weeks of *Sense© for Kids* training will have higher scores on *sense\_assess© kids* (52) compared to children who received dose matched goal directed therapy via home program.
- Children receiving six weeks of *Sense© for Kids* training will have altered fMRI activation of the cortical somatosensory processing centres in response to tactile stimulation of the affected limb. Such changes will be greater than any activation changes seen in children who received dose matched goal directed therapy via home program.
- Children receiving six weeks of *Sense© for Kids* training will have altered structural connectivity (as assessed with diffusion MRI) of somatosensory processing centres.
- Children receiving six weeks of *Sense© for Kids* training will have higher scores on the Assisting Hand Assessment (53) compared to children who received dose matched goal directed therapy via home program.
- Children receiving six weeks of *Sense© for Kids* training will have comparable scores on the Goal Attainment Scale (54) and Canadian Occupational Performance Measure (55) compared to children who received dose matched goal directed therapy via home program.

### ***Trial design***

The Consolidated Standards of Reporting Trials (CONSORT statement, 2010) for RCT's of non-pharmacological treatments will inform this single blind randomised placebo-controlled trial with a matched pair design (61). Matched pair designs are recommended to reduce covariate effects and strengthen comparisons between groups (62). Children will be matched on age and total sum score on the *sense\_assess© kids* (52). There will be two arms of this study, *Sense© for Kids* training and a dose matched Goal Directed Training via Home Program (Figure 1). Children will be randomised following baseline assessment to one of these treatment groups. The children in the *Sense© for Kids* training group will receive two therapist-directed one-hour treatment sessions per week for six weeks, plus a third hour per week of *Sense© for Kids* occupational training carried out by the primary caregiver (who will receive coaching and guidance from the therapist). Children in the Goal Directed Training via Home Program will receive one hour a week of therapist led Goal Directed Training and will undertake a further two hours per week

of home practice with primary caregiver support. Differences in therapist directed therapy time exists between these two interventions and reflect the nature of each intervention. The total dose of therapeutic activity is equal.

Figure 1 Study design with assessment schedule



### ***Recruitment***

Children will be recruited through the Cerebral Palsy Mobility Service at Princess Margaret Hospital.

### ***Participants***

#### *Inclusion criteria*

This study will include school aged children and youth:

- With a paediatrician confirmed description of HCP
- Aged eight to 15 years
- Who can follow assessment procedure (including fMRI)
- With a confirmed impairment in somatosensory discrimination function as assessed on the *sense\_assess©\_kids*.
- Who live within metropolitan Perth, Western Australia

#### *Exclusion criteria*

This study will not include children and youth who have:

- Upper limb surgery in the 12 months preceding baseline assessments
- MRI contraindications including: metal implants, implantable devices, significant anxiety issues, claustrophobia, or behavioural problems

- For children in receipt of Botulinum toxin-A for spasticity management, study commencement will begin 12 weeks after their most recent treatment to allow for Botulinum toxin-A “washout”.

### ***Withdrawal***

Children and their families are free to withdraw at any time. Any data collected prior to withdrawal will be retained and used for an intention-to-treat analysis.

### ***Allocation***

Minimisation will be employed to optimise the homogeneity of the two groups (63). Children will be matched for age ( $\pm$  6 months) and somatosensory discrimination capacity sum score (mild/moderate/severe). When a child is enrolled to the study without a match for age and somatosensory capacity, that child will be randomly allocated to a treatment group using an online randomisation form by a staff member not otherwise involved in the study. The next child enrolled who is a match for the unmatched participant will be automatically allocated to the alternate group. The process will be repeated for each matched pair; the first member always being allocated at random.

### ***Blinding***

The families and *treating* therapist(s) will not be blinded to group allocation, but families will be blinded to the study hypotheses. The therapist(s) responsible for assessment will be blinded to group allocation. If blinding is broken, this will be noted in the therapist’s treatment or assessment record and reported. To protect the blinding of assessors, participants will be coached not to discuss group allocation with assessors, and interventionists will not discuss study hypotheses with participants.

### ***Sample size***

To determine the sample size required for this study we used pilot data from seven children with HCP who have received the *Sense© for Kids* intervention. Data from the Wrist Position Sense Test (a component of the *sense\_assess©\_kids*, see below) were entered into G\* Power (64) and a two tail “Means: difference between two independent groups” power calculation was performed. With an intervention group mean of 15.94 and standard deviation 9.72; and control group mean 25.79 and

standard deviation 11.93 the calculated effect size was 0.9052. To detect this effect size, we need 42 subjects (21 in the intervention group and 21 in the control group) to have statistical power of 0.8 at the significance of 0.05. To account for attrition, this study will aim to enroll 50 children, with 25 in each of the control and intervention groups.

### ***Study protocol***

All outcomes will be measured within two weeks prior to commencement, again within two weeks following completion of intervention, then six months and 12 months' post intervention ( $\pm 2$  weeks; Figure 2). Assessment and intervention will take place in children's homes or at school, whichever is the most convenient for families, except for MRI assessments which will take place at Perth Children's Hospital. MRI data will be acquired at all time-points, except the 6 weeks follow up.

Figure 2 Outcome Measures

Outcome measure	Time-point					ICF Domain
	1	2	3	4	5	
sense_assess©_kids	•	•	•	•	•	Body structure/ function
Magnetic Resonance Imaging	•	•		•	•	Brain structure/ function
Assisting Hand Assessment	•	•	•	•	•	Activity
Goal Attainment Scaling	•	•	•	•	•	Activity and participation
Canadian Occupational Performance Measure	•	•	•	•	•	Activity and participation

Figure 2 outlines when each outcome measure will be obtained. Time-point one is the baseline assessment, time-point 2 is at completion of 6 weeks of intervention, time-point 3 is 6 weeks' post intervention completion follow-up, time-point 4 is 6 months post intervention completion follow-up and time-point 5 is the 12 month post intervention completion follow-up.

### **Outcome measures and procedure**

#### ***Body function and structure***

##### *sense\_assess© kids*

The sense\_assess© kids (65), is a battery of tests which measure functional somatosensory discrimination ability. The domains of somatosensation measured by the sense\_assess© kids include the Protective Touch Test (66, 67), the Tactile

Discrimination Test (68), the functional Tactile Object Recognition Test (69) and the Wrist Position Sense Test (70). The Protective Touch Test uses the 4.56 Semmes Weinstein monofilament to test tactile registration at the threshold of protective touch. The Tactile Discrimination Test is a forced choice test of tactile discrimination whereby children need to indicate in a series of presentations which surface out of three is different. The functional Tactile Object Recognition Test is a 14-item test of haptic object recognition with multiple versions in which children are presented with familiar and novel objects out of vision and indicate what they are exploring using a response poster with pictures of all possible items. The Wrist Position Sense Test is a measure of proprioception in which a child's hand is moved out of vision to 20 positions in random order in the flexion/extension plane of movement of the wrist using a lever and a protractor scale. Children indicate where their hand is positioned using a protractor scale immediately above their occluded hand. The *sense\_assess@kids* has high reliability and normative standards for typically developing children aged 6 to 15 years (71), and demonstrated construct validity and clinical acceptability for children with CP aged 6 to 15 years (52, 72).

### ***Magnetic Resonance Imaging***

Quantification of central neural change in response to intervention contributes to the understanding of the mechanisms that lead to sustained functional improvements. In this trial, we aim to quantify brain changes that accompany any clinical improvements. To this end, we intend on analysing three types of MRI: *structural MRI*, task-based *functional MRI* (fMRI), and *diffusion MRI* (dMRI).

MR imaging will be conducted on a 3 Tesla Siemens Magnetom Skyra scanner (Siemens, Erlangen, Germany) located at the Perth Children's Hospital (PCH), Nedlands, Western Australia. Scan types are listed in Table 2.

Table 2 List of scan types  
*MRI scans to be acquired at each time point. See text for details.*

	Type	Resolution	Additional Details
T1 MPR	Structural	1mm iso	3D
T2 FLAIR	Structural	1mm iso	3D
T2 Blade	Structural		
GRE	field map	3mm iso	for EPI distortion correction
EPI	Functional	3mm iso	80 frames

	Type	Resolution	Additional Details
EPI	Diffusion	2mm iso	8x b = 0 s/mm <sup>2</sup> 20x b = 1000 s/mm <sup>2</sup> 60x b = 2000 s/mm <sup>2</sup>

Prior to the initial scan each child will attend an MRI preparation session. This has been demonstrated to improve the success of sedation-free brain MRI scanning in children (73). The preparation session will include watching a presentation about the MRI experience, familiarisation with the fMRI task (see below) and practice in a mock MRI scanner. On each arrival at the PCH Radiology Department for MRI scans children will be familiarised with the scanning procedure, scanning devices, and receive 5-10 minutes of practice of the fMRI task. Following the MRI, participants will complete a simple questionnaire regarding the MRI experience including awareness of the stimuli, degree of concentration and comfort.

#### *Structural MRI*

Both high resolution T1 and T2 images will be acquired (Table 1). The participant will be able to watch a DVD of choice during anatomical sequences. Anatomical reporting will be conducted upon these images by a paediatric neuroradiologist. Baseline MRIs will be classified using the harmonized classification of magnetic resonance imaging, based on pathogenic patterns (MRI classification system or MRICS) proposed by the Surveillance of CP in Europe network (74). MRI Classification will be documented for each participant and utilised for subgroup data analysis. A paediatrician will meet with the participant and their caregiver to discuss anatomical findings and the primary treating physician will be informed of these results.

#### *Functional Magnetic Resonance Imaging*

Functional Magnetic Resonance Imaging (fMRI) will be utilised as an indirect measure of neuronal activation in the brain in response to a somatosensory stimulus. fMRI utilizes blood-oxygen-level-dependent (BOLD) contrast to indirectly measure neuronal activation in the brain. In neurorehabilitation, fMRI has been utilised to identify, quantify and map cortical activation associated with execution of particular tasks (15). fMRI has also been used in research as a physiological marker of brain plasticity in children with cerebral palsy, and small studies of motor function in

children with CP have demonstrated a significant change in task related cortical activation following constraint-induced therapy (75, 76). Correlation between somatosensory functional impairment post-stroke and central neural changes has been demonstrated using fMRI (36, 77).

Pre-intervention, fMRI activation patterns in response to somatosensory stimulation of both hands will be measured as a baseline, with focus on cortical somatosensory processing centres including S1 and S2. Post intervention fMRI somatosensory task-related activation will be measured and compared to pre-intervention results as an indicator of central neural change in response to therapy. This methodology is supported by literature that indicates that in order to measure neuroplasticity with fMRI, scans should be obtained during a task, both before and after intervention, for at least 20 people per experimental group (78).

In conjunction with the CSIRO and Florey Institute of Neurosciences and Mental Health, an fMRI protocol has been adapted for use in children with CP. This protocol consists of two acquisitions – one per hand. Each scan will consist of four 30-second ‘stimulation’ blocks, each preceded by a 30 second rest block. During stimulation blocks, a device presents a textured grid to the fingertips in a manner controlled for speed and pressure, alternated with no stimulus. A plastic texture grating is moved side to side across the fingertips of the second, third and fourth digits (77, 79). Within block, two different plastic texture grids will be delivered, with spacings of 1550 and 3000 micrometres between the gratings, alternating every five seconds. These patterns will be different every block to maintain attention of the participant. Participants will be instructed to pay attention to the differences between the two patterns presented in each block, but to remain still. A screen showing the words ‘FEEL’ or ‘REST’ will be shown to the participant during these respective blocks to cue attending the stimuli. The pressure of stimulus delivery is calibrated at the commencement of the scan via a weighted pulley system. To control for movement, the participant’s hand rests on a platform with custom openings for the fingertips and is immobilised in the device as the stimulus is moved from side to side under the fingertips. The control ‘REST’ condition of the paradigm is no presentation of the textured grid to the participant’s fingers, though it continues to be moved at a

constant speed to the side of the participant's hand (77, 79). The participant lies supine throughout.

### *Diffusion Magnetic Resonance Imaging*

Diffusion magnetic resonance imaging (dMRI) will be used to investigate brain microstructural changes within pathways delineated using fMRI driven diffusion tractography. dMRI data will be acquired using a multi-shell approach, which includes 8 non-diffusion weighted images, 20 diffusion weighted images at  $b=1000\text{s/mm}^2$ , and 60 diffusion weighted images at  $b=3000\text{s/mm}^2$ . Correction for susceptibility distortions will be performed using reverse phase-encoded non-diffusion weighted images. Fibre orientation distributions for tractography will be estimated using multi-shell multi-tissue constrained spherical deconvolution (80) implemented in MRtrix software. Fractional anisotropy (FA) will be estimated based on the  $b=1000\text{s/mm}^2$  shell.

### **Activity**

The *Assisting Hand Assessment* (AHA) (53) is a measure of how a child with HCP or brachial plexus palsy uses their involved hand for bimanual activity. The AHA has been found to have good construct validity, excellent test-retest reliability (0.99) and is responsive to change when used to assess children aged 18 months to 18 years (81). The assessment is conducted as a play session and is video recorded for scoring at a later time.

The *Canadian Occupational Performance Measure* (COPM) (55) is a measure of a client's self-perceived occupational performance over time. The COPM has been found to have good validity and reliability and is responsive to change (82) and has been found to have moderate reproducibility (83).

*Goal Attainment Scaling* (GAS) (54) is a technique used to quantify goals set in a rehabilitation setting. This goal setting technique enables the conversion of measurable goal attainment on a 5-point scale into t-scores which are normally distributed around a mean score of 50 and a standard deviation of 10. The GAS has been found to be a valid and reliable measure of goal attainment (84) with excellent inter-rater reliability ( $>.90$ ) and satisfactory concurrent validity (85).

## **Descriptive measures**

To describe our population the following scales and measures will be completed at baseline.

### ***GMFCS, MACS and CFCS***

The *Gross Motor Function Classification Scale- Expanded and Revised* (GMFCS-E&R) (86) is a five level scale describing gross motor function for children with CP aged 6 to 12 years and 12 to 18 years. The GMFCS describes a range of abilities from level I, where children are independently mobile, through to level V where children have limited ability to maintain head and trunk postures and are dependent on wheeled mobility with assistance from others (86).

The *Manual Ability Classification Scale* (MACS) (87) is a five level scale describing the ability of children with cerebral palsy aged 4 to 18 years to handle objects in everyday activities. The MACS describes a range of manual abilities from level I, where children handle objects easily and successfully, through to level V, where children do not handle objects and are severely limited in their ability to perform simple actions (87).

The *Communication Function Classification Scale* (CFCS) (88) is a five level scale describing the communication ability of individuals with CP. The CFCS describes a range of communication abilities from level I, where children are effective senders and receivers with familiar and unfamiliar communication partners, through to level V, where children are seldom effective senders or receivers with familiar communication partners (88).

### **Hypertonia Assessment Tool**

The *Hypertonia Assessment Tool* (HAT) is a six-item clinical assessment tool used to differentiate between spastic, dystonic and rigid paediatric hypertonia (89). The HAT allows for standardization of such clinical examination, noting that mixed tone, i.e. both spasticity and dystonia, are present in a large proportion of children with CP (89, 90). This information will be utilized in subgroup analysis to evaluate whether children with certain hypertonia subtypes demonstrate greater response to intervention than others. In doing so, children with CP can be directed to

interventions of greatest efficacy in the future. The HAT has good inter-rater test-retest reliability and validity for the identification of spasticity, and moderate agreement for dystonia (89, 90).

The *Selective Control of the Upper Extremity Scale* (SCUES) (91) is a measure of selective motor control for the upper limbs. SCUES is a short (< 15 minute) video based assessment that is administered by an occupational therapist or physiotherapist. SCUES examines selective motor control for the shoulders, elbow, wrist, and fingers/thumb. The examiner demonstrates a movement, passively moves the child to replicate the movement and determine passive range of motion, then the child replicates the movement. Performance is graded on the presence of mirror movements, movement of additional joints beyond target joint, presence of trunk movement, and the extent to which actual movement is equal to or less than passive range. SCUES has acceptable content validity, intrarater (>0.75) and inter-rater (0.72) reliability and construct validity (91).

### **Adverse Events**

Adverse events from intervention and activity based assessment are not anticipated. Adverse events due to imaging aspect of assessment may occur if there is a high degree of anxiety for children about being inside the MRI scanner. A data safety committee will be convened to monitor the risks associated with the large number of MRI scans proposed in this study. Children are not sedated during MRI scans and this research team has developed and piloted a familiarization package to allow children to experience what being inside an MRI scanner is like prior to their consenting to take part in this aspect of the study. The MRI assessment is introduced to children in a standard clinic room with their parents present by the staff members who will be with them on the day of their MRI assessments. All efforts will be made to help children feel comfortable with MRI assessment, however children can withdraw from the MRI assessment if they are experiencing distress and this will not limit their participation in the rest of the study. Any adverse events will be reported to the HREC and data safety committee.

## **Statistical methods**

As outlined in the CONSORT statement reporting for RCT's: between group comparisons will be conducted on intention to treat analysis (61). Missing data arising from incomplete observations and dropouts will be managed using multiple imputations. Multiple imputations is recommended for use in RCT's because it avoids bias found in last observation carried forward approaches while maintaining power (92).

Summary statistics will be reported for each time period for each group using means and standard deviations. For skewed data, medians and interquartile ranges will be reported.

A mixed-effects model with repeated measures will be used to assess within and between group differences. The corresponding baseline measure will be entered as a covariate in the model along with age and somatosensory capacity given these features were used in the randomisation process. The mixed model approach has the advantage of allowing for correlated data (repeated measures) and allows for missing observations within-subject. Model assumptions will be examined and if required transformations applied or non-parametric methods employed. Statistical significance will be set at 0.05.

### ***Structural MRI***

In the interests of measuring brain change, cortical thickness analyses will be undertaken using previously described methods (93, 94). In brief, for each participant, structural images from each time-point will be merged to generate a *single participant template* – a pseudo image whose orientation is unbiased with respect to time-point. Templates will be tissue-segmented using in-house software and manually corrected where required. Each template will then be used as a prior to further segment that participant's individual structural images. Cortical thickness will then be analysed in accordance with methods described in Pagnozzi et al. (94). An ANOVA will test for effects of time, treatment group, and an interaction between the two.

### ***Functional MRI***

First level statistical analyses will contrast blocks (FEEL > REST) on an individual subject basis. Owing to the heterogeneous size and location of brain lesions seen within most cohorts, inter-subject registration (required for voxelwise statistics) can be impossible to perform reliably (67). We plan to address this by performing region-of-interest analyses that measure the interhemispheric balance of activation between the sensorimotor cortices before and after therapy in the same child. S1, S2, and the dorsolateral prefrontal cortex (as delineated on single-participant templates) will be used. First-level results from all participants will be pooled into an ANOVA, to investigate (A) changes by time-point, to search for a general change in brain activation and (B) whether an interaction between time-point and treatment exists.

Task-related fMRI (t-fMRI) is considered an important neuroimaging modality in researching neuroplasticity. It is however recognised that t-fMRI presents a “unique set of challenges” (95) that impact data analysis and interpretation. These challenges include but are not limited to: t-fMRI result variability, inability to distinguish the biological process that underlie changes in activation including alternative explanations such as compensatory activation or strategic shifts, the challenge in presenting task equivalency, and specific challenges in data analysis (95). Many of these challenges of t-fMRI are even greater in children with CP in comparison to adult populations. As a group, the brain pathology and morphology of children with CP is highly heterogeneous and often markedly abnormal owing to the wide range of aetiological processes and the early stage of development at which these processes occur. Additionally, clear relationships between brain structure and a child’s function have been challenging to establish (96). These factors make standard functional neuroimaging analysis extremely challenging and at risk of limitation in the CP population (97). Reid et al. make the case that multimodal imaging enables these unique challenges to be addressed and increases the robustness of functional neuroimaging research (95).

This study attempts to reduce the influence of Reid’s cited confounds in a number of ways. First, we have selected long block lengths to reduce effects of abnormal haemodynamic responses. Second, we expect to scan a meaningful number of participants to reduce bias caused by a small number of unusual cases. Third,

participants will receive extensive task practice prior to entering the scanner, including in a mock-scanner scenario, reducing differential task anxiety and familiarity between scans. Fourth, we have selected a task with minimal active response required from the participant and should not be substantially more difficult for participants with poorer motor abilities (77, 79). Fifth, we avoid voxelwise analyses which may be invalidated by pathology. Finally, fMRI analyses will be interpreted in the context of independent clinical scores, measures of cortical thickness, and diffusion measurements of white matter.

### ***Traditional diffusion MRI***

Traditional diffusion MRI (dMRI) analysis methods make assumptions about brain structure-function relationships that may not hold in the context of significant brain pathology and cortical reorganisation, as occurs in children with CP (95). To overcome this challenge, surface-based-fMRI guided tractography will be utilised, as previously demonstrated in children with CP (95), to extract thalamocortical tracts.

Mean FA will be taken for thalamocortical tracts at each time-point, and entered into an ANOVA to test for (A) the effects of time-point, to test for brain changes, and (B) a time-point – treatment interaction, to test whether treatments evoke different degrees of brain change.

### **Discussion**

This study is a phase II comparative clinical trial (98), that builds on the findings of the recently completed phase I feasibility trial, reported by McLean et al. (43). This comparative clinical trial will make a substantial contribution to our current knowledge base by exploring the efficacy of a somatosensory discrimination approach for children with CP, as well as observing neural changes following a somatosensory intervention. This study will also be the first to compare a somatosensory intervention approach in children with HCP with a dose matched evidence based motor function focussed intervention, in this case goal directed home programs. This study will provide valuable insights into treatment effectiveness and the underlying mechanism for change in the use of somatosensory discrimination training and will add to existing literature concerning the use of home programs. If

children gain benefit from somatosensory discrimination training and increase their use of their affected hand and can transfer those skills to novel tasks, such as the children in our pilot work, this will improve functional independence and long-term outcomes for children with HCP. Understanding how a somatosensory approach may impact hand use, a child's functional independence and self-efficacy will be an important contribution. Further to this, knowing whether a home program alone, without emphasis on sensory stimuli involved in any purposeful activity could have an incidental effect on somatosensory function, will also be an important finding. Results of this study will be disseminated widely through publications, international academic conferences and elsewhere as guided by our consumer representative.

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## **APPENDIX I      Plain Language Summaries**

The following documents are plain language summaries and reports of somatosensory function emailed to the typically developing participants and children with cerebral palsy and their families after completion of the research studies reported in papers one, two, three and four, and appendices A, D, F and G.

## I.1 Invitation Letter (Prospective Typically Developing Participants)

**Curtin University**  
For all enquiries, please contact:  
Susan Taylor  
Bsc (OT) (Hons)  
PhD candidate  
School of Occupational Therapy and  
Social Work, Health Sciences

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Kent Street, Bentley, Perth  
Western Australia 6102



### Sense\_assess Kids Project

My name is Suzi Taylor. I am looking for young people to take part in a research project that aims to find out if an assessment designed for adults is suitable for use with young people. In order to find this out I would like to ask therapists to do this assessment with someone like you.

#### **What is the assessment?**

The sense\_assess kids is an assessment tool used to measure feeling in the hands and wrists. Young people that may be assessed with the tool may be people with cerebral palsy, acquired brain injury or autism. Body sensations measured by the assessment involve touch, how well you can guess objects using the hand, how well you can guess the position of your hand and wrist. It takes approximately thirty five minutes to do.

#### **What are the benefits of this research?**

If I find that the assessment is suitable for young people it will be used by therapists with people like you to help plan your treatment sessions. There are not many assessment tools out there to measure feeling in the hands, so it will be great if the sense\_assess kids is suitable. If it isn't, the information collected in this study will still help in the modification of this tool to make it better and more suitable for young people.

#### **What is required of you?**

The sense\_assess kids involves four test activities that are listed below. It is a seated assessment involving testing of the hand only. You will be assessed at the centre that you usually visit for therapy. Your parent or carer is welcome to watch the assessment and feedback will be provided on how you have performed on the score sheet. Score sheets from the sense\_assess kids will be coded, which means that your name does not appear on the score sheet therefore the information is secret.

#### **The sense\_assess Kids**



**Protective Touch Test** uses a light fishing line string pressed gently against the skin



**Tactile Discrimination Test (TDT)** measures if someone can choose between different textures using their finger



**Functional Tactile Object Recognition Test (fTORT)** measures how well a child can recognise objects through touching, without using vision



**Wrist Position Sense Test (WPST)** measures how well a child can guess what position their wrist is in without looking at it

### Your rights

Participation in this project is your choice and you are free to leave the study at any time and for any reason. Your privacy will be respected at all times. All information collected will be stored in a locked cabinet at Princess Margaret Hospital forever, even after the project is finished. The results of this research may be published in therapy journals; however your name will never be revealed.

### Further use of your assessment scores

The coded sense\_assess kids test results collected from participants will only be used for the purposes of this study. However, there may be a case where other researchers like me wish to use the results for future research projects or to make it available to other researchers. You have the option to say if you do not want this to occur.

### Risks/Restrictions

The sense\_assess kids is described as a safe test and poses no known risks to you.

### Participant instructions

If you would like to participate in the sense\_assess kids project please sign the consent form at the end and return it to your therapist.

If you have any questions you can ask your therapist or me, my number is 0418 317 257 and my email is [susan.taylor4@health.wa.gov.au](mailto:susan.taylor4@health.wa.gov.au)

Thank you for reading

Yours sincerely,

**Susan Taylor**

Chief investigator  
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*This research project has been reviewed and given approval by the Curtin University Human Research Ethics Committee (approval number). Should you wish to make a complaint and would like to talk to an independent person, you may contact the Human Ethics Committee (Secretary), phone: 9266 2784, email: [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au), mail: c/- Office of Research and Development, Curtin University of Technology, GPO Box U1987, Perth WA 6845.*

## I.2 Introductory Letter (Typically Developing Participants and their Families)

### **Curtin University**

For all queries, please contact:  
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Bachelor of Science (Hons)  
Occupational Therapy  
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Curtin University  
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Government of **Western Australia**  
Department of **Health**  
Child and Adolescent Health Service

Dear participant and family of participant,

Thank-you for being the typically developing participants of a research study related to the assessment of sensation in children with neurodevelopmental disorders (Curtin University Human Research Ethics Committee protocol approval number HR 87/2014). We would like to extend our gratitude and appreciation to those who participated and gave so generously of their time after school and on weekends. In Perth metropolitan and rural areas we received overwhelming participation with 32 children aged 6-15 years completing the Sense Assess Kids at two different time points (18 females and 14 males with an even spread across each year of age and 90% with a right hand dominance).

We are still in the process of analysing data however preliminary findings show that the assessment designed for adult stroke survivors is suitable for use with children and youth, which was our main aim to find out. This research has also helped identify the hand movements a young child uses in order to identify what they are feeling with their hands when they cannot see.

Analysing such a large amount of data does take time however the associated research article will be prepared for submission to a scientific journal and we look forward to the comprehensive results being available once published. Without your support, research projects such as ours would not be possible. Thank-you for contributing to this research and supporting the development of the Sense Assess Kids.

**Kind regards,**

**Susan Taylor**  
Bsc (OT) (Hons)

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Delivering a **Healthy WA**

### I.3 Summary of Test Scores for Typically Developing Participants (Parents)

#### An investigation of hand movements in children and youth Sense Assess Kids test scores

<b>Participant name:</b>	<b>Assessed by:</b>
<b>Date of initial assessment:</b>	<b>Date of reassessment:</b>

The Sense Assess Kids has been developed to measure sensation of the upper limb in children. Its purpose is to provide therapists with an assessment tool to aid in early and accurate detection of sensory impairments. The current study you and your child have just been involved in has provided us with typically developing information for children aged 6 to 15 years.

The following results are for the right and left hand and indicate a maintenance or difference in scores over two time-points (2-3 weeks). The Sense Assess Kids is still in the tool development phase and we are using your child's scores to map typical development. Many factors may affect test scores for children and adolescents including time of day, motivation to complete test and length of test. Having a low score on any of the test items does not indicate your child has poor sensation as this tool is still under development. If you are concerned please contact us directly for further discussion.

**The Fabric Matching Test (FMT)** is a measure of one's ability to match a set of common fabrics through 'tactile discrimination'. The participant feels two fabrics with their index finger and decides if they are the same or different. Tactile discrimination is important to identify different surface textures such as feeling a button and button-hole on fabric and knowing which one is which. For adults, a score close to 1.00 indicates intact tactile discrimination. In this study we have been collecting data for typically developing children. Results from this study indicate that our 31 typically developing children had an average score of 0.93 with scores ranging from 0.55 to 0.98 depending on age.

#### Scores for tactile discrimination:

**Right hand**  
**Time-point 1**  
**Time-point 2**

**Left hand**  
**Time-point 1**  
**Time-point 2**

**The functional Tactile Object Recognition Test (fTORT)** is designed to test recognition of objects through touch. The test contains 14 object sets – the objects are common, everyday objects. Each object set consists of two objects, that differ in one sensory feature e.g. weight, and a distracter object that differs in more than one sensory feature eg weight and shape. Pictures of object sets are displayed on a poster and test objects are provided to be felt. Object recognition is useful to identify objects in the hand without looking; such as choosing a particular pen you want from inside a pencil case.

For the fTORT a score of 36 to 39 out of 42 indicates intact object recognition for typically developing 6 to 15 year olds. Results from this study indicate that our 31 typically developing children had an average score of 39 with scores ranging from 28 to 42 depending on age. Please note the fTORT was only completed at 1 time-point.

#### Scores for object recognition:

**Right hand**  
**Time-point 1** /42 correct

**Left hand**  
**Time-point 1** /42 correct

**The Tactile Discrimination Test (TDT)** measures one's ability to pick differences in finely graded plastic texture grids. The child feels the texture grids with their index finger. For the TDT Version 1 they are asked to choose the different one out of three textures. At this stage we do not know what indicates intact tactile discrimination for typically developing children however results from this study show that our 31 typically developing children had an average score of 14 out of 20 with scores ranging from 8 to 20 depending on age.

For the TDT Version 2 children are asked to choose the rougher one out of two textures. Results from this study indicate that our 31 typically developing children had an average score of 29 out of 32 with scores ranging from 18 to 32 depending on age.

**Scores for tactile discrimination**

**TDT version 1**

**Right hand**

**Time-point 1** /20

**Time-point 2** /20

**Left hand**

**Time-point 1** /20

**Time-point 2** /20

**TDT version 2**

**Right hand**

**Time-point 1** /32

**Time-point 2** /32

**Left hand**

**Time-point 1** /32

**Time-point 2** /32

**All correspondence to:**

**Susan Taylor** Bsc (OT), PhD candidate

Chief investigator (Curtin University)

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[Catherine.elliott@health.wa.gov.au](mailto:Catherine.elliott@health.wa.gov.au)

*This research project has been reviewed and given approval by the Curtin University Human Research Ethics Committee (HR87/2014). Should you wish to make a complaint on ethical grounds and would like to talk to an independent person, you may contact the Human Ethics Committee (Secretary), phone: 9266 2784, email: [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au), mail: c/- Office of Research and Development, Curtin University of Technology, GPO Box U1987, Perth WA 6845.*

I.4 Certificate of Appreciation (Typically Developing Participants)



## I.5 Information Letter (Participants with Cerebral Palsy and their Families)



**Curtin University**



Government of **Western Australia**  
Department of **Health**  
Child and Adolescent Health Service

Dear participant and family,

Thank-you for participating in our research: Discovering the sense of touch implications for the development and recovery for children with cerebral palsy (Curtin University Human Research Ethics Committee protocol approval number HR 87/2014). We would like to extend our gratitude and appreciation to those who participated and gave so generously of their time after school and on weekends. In Perth metropolitan and rural areas we received overwhelming participation with 28 children aged 6-15 years completing the Sense Assess Kids at four different time points (10 females and 18 males with an even spread of ages).

In your envelope we have included a Research summary for your child to read, a Summary of Sense Assess Kids test scores for your information and a Certificate of appreciation. If you have any questions about any of these documents please don't hesitate to contact myself or Belinda. We are still in the process of analysing data however preliminary findings show that the assessment designed for adult stroke survivors is suitable for use with children and youth with cerebral palsy, which was our main aim. This research has also helped identify the type of hand movements a young child with cerebral palsy uses in order to identify what they are feeling in their hands without looking.

Analysing such a large amount of data does take time however the associated research article will be prepared for submission to a scientific journal and we look forward to the comprehensive results being available once published. Without your support, research projects such as ours would not be possible. Thank-you for contributing to this research project and supporting the development of the Sense Assess Kids and SENSE Training.

Kind regards,

**Susan Taylor**

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## I.6 Summary of Test Scores for Participants with Cerebral Palsy (Parents)

### Discovering the sense of touch Summary of Sense Assess Kids test scores

<b>Participant name:</b>	<b>Assessed by: Susan Taylor</b>			
<b>Date of assessment:</b>	<b>Time point 1:</b>	<b>Time point 2:</b>	<b>Time point 3:</b>	<b>Time point 4:</b>
<b>Hand assessed:</b>				

The Sense Assess Kids has been developed to measure somatosensation (*body sensations*) of the upper limb in children. Its purpose is to provide therapists with an assessment tool to aid in early and accurate detection of somatosensory impairments. Note: The Sense Assess Kids is currently under development for use children with cerebral palsy aged 6 to 15 years.

The following results are for the right/left (assisting) hand only and indicate a maintenance or difference in somatosensory scores over four assessment time-points (6 months). Through previous research we have an average test score to describe how a child without cerebral palsy (CP) aged 6 to 15 years performs on the Functional Tactile Object Recognition Test, Wrist Position Sense Test, Fabric Matching Test and Tactile Discrimination Test. From the results below you can see how your child performs compared to peers without CP. If you have questions about your child's scores please contact us directly for further discussion.

**The Fabric Matching Test (FMT)** is a measure of one's ability to match a set of common fabrics through 'tactile discrimination'. The participant explores two fabrics with their index finger and decides if they are the same or different. Tactile discrimination is important to identify different surface textures such as feeling a button and button-hole on fabric and knowing which one is which. The closer a score is to 1, the more choices a child got correct. The average test score for children without CP is 0.93 with scores ranging from 0.76 to 0.98. Scores below this range may indicate reduced tactile discrimination. Your child's score for each of the 4 time points is in the table below.

#### Scores for tactile discrimination:

<b>Right hand</b>
<b>Time point 1</b>
<b>Time point 2</b>
<b>Time point 3</b>
<b>Time point 4</b>

These scores indicate an improvement between time point 1 and 4.

**The Functional Tactile Object Recognition Test (FTORT)** is designed to test recognition of objects through the sense of touch. The test contains 14 object sets – the objects are common, everyday objects. Each object set consists of two objects that differ in one sensory feature e.g. weight, and a distracter object that differs in more than one sensory feature e.g. weight and shape. Object sets are displayed on a poster and test objects are provided to be felt. Object recognition is useful to identify objects in the hand without looking, such as choosing a particular pen you want from inside a pencil case. Children are given a score out of a possible 42, therefore the higher the test score the better the object recognition. The average test score for children aged 6 to 8 years without CP is between 36 and 38 out of 42; 9 to 11 years without CP is between 38 and 39 out of 42; 12 to 15 years without CP is 39 out of 42. Scores below this range may indicate reduced haptic object recognition. Your child's score for each of the 4 time points is in the table below.

#### Scores for object recognition:

<b>Right hand</b>
<b>Time point 1</b> 6/42
<b>Time point 2</b> 6/42
<b>Time point 3</b> 9/42
<b>Time point 4</b> 9/42

These scores indicate no significant change over the 4 time points.

**The Wrist Position Sense Test (WPST)** measures a child's ability to sense the position of their hand and wrist when they can't see it. The test involves placing the child's hand in 20 different positions and the child matching this position as closely as they can. Wrist position sense helps a child know where their hand is in space and is important for ball

sports, handwriting and many other activities involving coordination. Scores are given in degrees away from actual correct position, therefore the lower the test score the better the wrist position sense. The average test score for children aged 6 to 8 year olds without CP is between 8° and 21°; 9 to 11 year olds without CP is between 7° and 15°; 12 to 15 year olds without CP is between 6° and 14°. Scores above this range may indicate reduced wrist position sense.

**Scores for wrist position sense:**

<b>Right hand</b>
<b>Time point 1</b> °
<b>Time point 2</b> °
<b>Time point 3</b> °
<b>Time point 4</b> °

These scores indicate no significant change over the 4 time points.

**The Tactile Discrimination Test (TDT)** measures the ability to pick differences in finely graded plastic textures like lines. The child feels the texture grids with their index finger and they are asked to choose the different one out of three textures. The average test score for children aged 6 to 8 years without CP is 12 out of 20 with scores ranging from 8 to 15; 9 to 11 years without CP is 14 out of 20 with scores ranging from 9 to 18; 12 to 15 years without CP is 15 out of 20 with scores ranging from 12 to 16. Scores below this range may indicate reduced tactile discrimination. Please note the TDT was not completed at every time-point.

**Scores for tactile discrimination TDT**

<b>Right hand</b>
<b>Time point 1</b> /20
<b>Time point 2</b> /20
<b>Time point 3</b> /20
<b>Time point 4</b> /20

These scores indicate a slight improvement over the 4 time points.

**If you would like to discuss these scores further all correspondence is to:**

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*This research project has been reviewed and given approval by the Princess Margaret Hospital Human Research Ethics Committee (2052EP). If you have any concerns or complaints regarding this study, you can contact the Director of Medical Services at PMH (Telephone No: (08) 9340 8222). Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study.*

**I.7 Research Summary for 6 to 7 year olds (Participants with Cerebral Palsy)**

  <b>Curtin University</b> Government of <b>Western Australia</b> Department of <b>Health</b> Child and Adolescent Health Service	
  Curtin University	<p>Suzi is doing a project with children who have cerebral palsy; that is why we asked you to help us.</p>
	<p>Suzi asked your mum or dad if you could complete some activities...</p> <p>And</p> <p>...You said yes!</p>
	<p>Suzi Taylor came to visit you and you did these activities with her.</p>
	<p>We gave you a code and wrote the results of the activities down on paper.</p> <p>You can ask your mum or dad to read your results to you.</p>
	<p>Suzi will write about these activities in a short story. The story will be about the Sense Assess Kids and how easy or hard young people with cerebral palsy found the activities.</p>
<p><del>Jane Smith</del></p>	<p>Suzi would like to share this story with many others as they will find it very interesting.</p> <p>Your name will not go in this story, only information about the Sense Assess Kids activities will be in it.</p>

	<p>The activities did take a lot of your time...</p>
	<p>...so we wanted to say thank-you because you did such a great job helping us!</p>
	<h2>QUESTIONS</h2> <p>If you have any questions about the activities, it is fine to let your mum or dad know and they can ask me. My phone number and email address are just below.</p>
	<h2>SUZI'S CONTACT DETAILS</h2> <p>You can phone Suzi on <b>0418 317 257</b></p> <p>Or email <a href="mailto:susan.taylor4@health.wa.gov.au">susan.taylor4@health.wa.gov.au</a></p> <p><b>Suzi's supervisors:</b>  A/ Prof Sonya Girdler <a href="mailto:sonya.girdler@curtin.edu.au">sonya.girdler@curtin.edu.au</a>  A/Prof Catherine Elliott  <a href="mailto:catherine.elliott@health.wa.gov.au">catherine.elliott@health.wa.gov.au</a></p>

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