Short title: Somatosensory impairment in children with cerebral palsy

**Full title:** Somatosensory discrimination impairment in children with hemiplegic cerebral palsy as measured by the sense\_assess<sup>©</sup> *kids*.

Authors: Belinda McLean PhD<sup>1,2</sup>, Susan Taylor PhD<sup>1,3</sup>, Jane Valentine PhD FRACP FAFRM<sup>2</sup>, Leeanne Carey PhD FAOTA FOTARA<sup>4,5</sup>, Ashleigh Thornton PhD<sup>2,6</sup>, Catherine Elliott PhD<sup>1,2,7</sup>

<sup>1</sup>School of Occupational Therapy, Social work and Speech Pathology, Curtin University, Perth, Australia; <sup>2</sup>Kids Rehab Western Australia, Child and Adolescent Health Service, Perth, Australia; <sup>3</sup>Occupational Therapy Department, Child and Adolescent Health Service, Perth, Australia; <sup>4</sup>Occupational Therapy, School of Allied Health, Human Services and Sport, La Trobe University, Melbourne, Australia; <sup>5</sup>Neurorehabilitation and Recovery, The Florey Institute of Neuroscience and Mental Health, Melbourne, Australia; <sup>6</sup>Division of Paediatrics, University of Western Australia, Perth, Australia<sup>7</sup>Telethon Kids Institute, Perth, Australia

 Susan Taylor ORCID ID
 0000-0001-5057-8140

 Jane Valentine ORCID ID
 0000-0003-1854-7381

 Leeanne Carey ORCID ID
 0000-0001-6376-8613

 Ashleigh Thornton ORCID ID
 0000-0003-3712-3330

 Catherine Elliott ORCID ID
 0000-0002-5324-8216

Correspondence to Belinda McLean ORCiD ID: 0000-0001-5456-8695 Postal address: C/O School of Occupational Therapy and Social Work GPO Box U1987 Perth WA 6845 E-mail: Belinda.Mclean2@health.wa.gov.au

# Acknowledgements

The authors would like to thank the participants for their precious time, the Cerebral Palsy Mobility Service team at Perth Children's Hospital for their support and assistance with this project, as well as Professor Eve Blair and Dr Richard Parsons for their guidance in preparing this manuscript.

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the <u>Version of Record</u>. Please cite this article as <u>doi: 10.1111/1440-1630.12729</u>

The authors would like to acknowledge support from the Perth Children's Hospital Foundation and Australian Postgraduate Awards (to BM and ST), the Western Australian Health Translation Network Early Career Research Fellowship (BM), the National Health and Medical Research Council (NHMRC) project grant #1022694; NHMRC partnership grant #1134495) and James S. McDonnell Foundation Collaborative Award (# 220020413) (to LMC).

# Conflict of interest declaration

The authors have no conflict of interest to declare.

Abstract: 203 Nain document word count: 4,589 Number of references: 47 Tables: 3 Keywords: Cerebral palsy, upper extremity, proprioception, touch, touch perception, stereognosis

#### DR. BELINDA MCLEAN (Orcid ID: 0000-0001-5456-8695)

DR. SUSAN TAYLOR (Orcid ID: 0000-0001-5057-8140)



**Introduction:** To characterise somatosensory discrimination impairment of the upper-limb across domains of tactile discrimination, limb position sense and haptic object recognition using the sense\_assess© kids and examine associations with upper-limb motor performance in children with hemiplegic cerebral palsy (CP).

**Methods:** The sense\_assess<sup>©</sup> kids was administered at one timepoint to 28 children, aged 6 to 15.5 years (M= 10.1, SD= 2.4), with hemiplegic CP (right hemiplegia n=15) and Manual Ability Classification System Levels I (n=11) and II (n=17). Unimanual motor performance was quantified using the Box and Block Test.

**Results:** Tactile discrimination was impaired in 18, limb position sense in 20, and haptic object recognition was impaired in 21 of 28 children. Over eighty percent (23/28) of children had impaired somatosensory discrimination in one or more domains. Low to moderate correlations were observed between each measure of somatosensory discrimination and motor performance. Manual ability classification was associated with limb position sense and haptic object recognition. A moderate inverse correlation (r = -.57, p < .01) exists between the number of somatosensory domains impaired and motor performance.

**Conclusion:** The frequency of somatosensory impairment in the upper limb of children in our sample was high and associated with manual ability, suggesting a need for routine assessment of somatosensation in this population.

Keywords: Cerebral palsy, upper extremity, proprioception, touch, touch perception, stereognosis

Word counts: abstract, 203/300; main document, 4589.

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 (Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). 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 (Stanley, Blair, & Alberman, 2000). Impairments in somatosensation have been reported for up to 97% of children with hemiplegic CP (HCP) (Van Heest, House, & Putnam, 1993). Previous studies have found that roughly two thirds of children with HCP have reduced tactile sensation and these impairments significantly impact motor function (Auld, Boyd, Moseley, Ware, & Johnston, 2012a, 2012b; Klingels et al., 2012). A recent editorial described CP as a sensorimotor disorder and highlighted the importance of considering somatic sensation to enhance the relevance of therapies for this group of children (Dan, 2020).

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" (Dunn et al., 2013). Tactile registration is the initial awareness of an external stimulus and does not involve interpretation of stimulus meaning (Williamson & Anzalone, 2001). Somatosensory discrimination, a more complex function than registration, is essential for interaction with the environment. It is needed to determine important properties of objects and tools and is part of the perception-action cycle that is a foundation for learning and cognitive development (Gibson & Pick, 2000). The aspects of somatosensory discrimination that directly interface with objects and the environment include limb position sense - the knowledge of where one's 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, often referred to as haptic ability or stereognosis (Dunn et al., 2013). Somatosensory discrimination enables identification of different textures and objects as well as knowledge of where our body is positioned in relation to its other parts without vision. These skills are crucial for everyday functioning (Dunn et al., 2013).

The reported frequency of somatosensory impairment in children with HCP varies from 50 to 97 percent in stereognosis alone, with limb position and two-point discrimination also

frequently impaired (Majnemer, Bourbonnais, & Frak, 2008). Somatosensation is important for use of the upper limb in daily actions, motor function and gains following motor and upper-limb interventions for children with CP. Tactile sensation and limb position sense have been found to account for one-third of the variance in motor performance among a cohort of 52 children with CP (Auld, Boyd, et al., 2012a). In addition, in a single-case study of 16 children, better tactile sensation and limb position sense were associated with greater motor learning gains following intervention (Robert, Guberek, Sveistrup, & Levin, 2013). Impaired tactile sensation and limb position sense are also predictive of functional gain after upperlimb surgery, with those children most severely affected unlikely to benefit functionally from procedures to correct deformity (Zancolli, 2003). Emerging evidence has demonstrated that somatosensory discrimination is modifiable and can lead to gains in bimanual hand-use (McLean et al., 2017).

With the assertion that CP is a sensorimotor disorder it is important to consider somatosensory function that interfaces with motor function (Dan, 2020). This paradigm cements the importance of haptic ability and limb position sense as areas for assessment and intervention. Limb position sense is an important aspect of somatosensory discrimination which is typically measured by imposed movement around the proximal phalanx of the index finger in the flexion-extension plane with assessment based on the subject's response as to the detection, direction, or replication of movement or position (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Klingels et al., 2010; McLaughlin et al., 2005). While these assessments may be useful in determining deficits in limb position sense in children with HCP, they are somewhat subjective and responsiveness to change is unknown. The sense\_asses© kids is a norm referenced, functional measure of somatosensory discrimination proposed by Taylor et al. (2017) that examines protective touch, tactile discrimination, haptic object recognition and limb position sense. The sense\_assess© kids was selected for this current work because it is a measure that covers the aspects of somatosensory function of interest.

Despite children with HCP frequently having impairment, somatosensory functions are not routinely assessed in clinical practice and therapists lack confidence in the measures they are using (Walmsley et al., 2018). The interaction between somatosensory functions and motor performance in children with HCP should be examined in depth, including any associations that may make identification of children likely to have impairment easier for clinicians (McLean, 2018). This research aims to characterise the frequency and nature of impairments

in somatosensory discrimination in a sample of children with HCP using the sense\_assess© kids (Taylor et al., 2017) and to examine associations between somatosensory impairment and motor performance quantified by a measure of gross manual dexterity and manual ability as classified by the Manual Ability Classification System (Eliasson et al., 2006).

# Method:

**Participants.** Participants in this cross-sectional study attended a Cerebral Palsy Mobility Service at a tertiary children's hospital [removed for peer review]. Children were included in this study if they had HCP, were aged between six and 15 years and, were at least 12 weeks post their most recent botulinum toxin injection (if relevant) and at least 12 months post any upper-limb surgery at the time of assessment. Recruitment took place from May 2013 to May 2015. Data were collected as part of a larger registered trial, ACTRN12614000314628. This study received ethics approval from the [removed for peer review] HREC (2052). Informed consent was received from parents and children assented to the study and all eligible children were invited to participate. Children were assessed in their homes by an occupational therapist trained in the use of the sense\_assess© kids and only the most impaired hand was assessed because this was to be the target of intervention in the larger study.

**Outcome measures. Somatosensory discrimination.** The sense\_assess© kids is a standardised, norm referenced assessment of functional somatosensory capacity of the upper limb of children with CP (Taylor, McLean, et al., 2018). The sense\_assess© kids contains a measure of tactile registration, The Protective Touch Test (Weinstein, 1993); a measure of tactile discrimination, the Tactile Discrimination Test (Carey, Oke, & Matyas, 1997); a measure imb position sense, the Wrist Position Sense Test (Carey, Oke, & Matyas, 1996); and a measure of haptic object recognition, the functional Tactile Object Recognition test (Carey, Mak-Yuen, & Matyas, 2020). Construct validity has been established in the component tests of the sense\_assess© in adult and paediatric populations with and without neurological conditions (Carey, Nankervis, LeBlanc, & Harvey, 2006; Carey et al., 1996; Carey et al., 2020; Carey et al., 1997; Taylor, 2017). Protective Touch Test 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 (Weinstein, 1993) across dorsal and palmer aspects of the hands (Taylor, McLean, et al., 2018). The Protective Touch

Test has high interobserver agreement of 99 to100% in typically developing children (Taylor, McLean, et al., 2018). The Tactile Discrimination Test (Carey et al., 1997; Taylor, McLean, et al., 2018) 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 Tactile Discrimination Test is assessed on accuracy of identifying the different texture with the score converted to an Area Under the Curve threshold score - a higher score indicates better performance. The functional Tactile Object Recognition Test (Carey et al., 2020; Taylor, Girdler, et al., 2018) measures haptic object recognition by assisting children to feel familiar and novel objects in a standardised manner with vision occluded and asking them to identify the matching object on a response poster. Haptic object recognition is assessed on accuracy and a higher score indicates better performance. The functional Tactile Object Recognition Test is a unidimensional scale with excellent internal consistency (Carey et al., 2020) and has demonstrated construct validity for children with CP (Taylor, Girdler, et al., 2018). The Wrist Position Sense Test (Carey et al., 1996; Taylor et al., 2020) 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. The Wrist Position Sense test uses an error score based on how far away from the test position a response pointer is positioned on a protractor scale, and lower scores indicate better performance. The Wrist Position Sense Test has high retest reliability (Taylor, McLean, et al., 2019), good discriminative validity, has demonstrated construct validity and responsiveness for children with CP (Taylor et al., 2020).

**Gross manual dexterity.** The Box and Block Test (Araneda et al., 2019; Mathiowetz, Federman, & Wiemer, 1985) is a brief standardised measure of gross manual dexterity. Participants are required to move blocks one at a time over a partition as quickly as they can. The number of blocks moved in one minute is recorded for each hand. Lower scores indicate less manual dexterity. The Box and Block Test 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 (Jongbloed-Pereboom, Nijhuis-van der Sanden, & Steenbergen, 2013).

**Procedure.** Families with a child meeting the inclusion criteria were referred by their consultant paediatricians and invited to participate by the primary investigator [removed for

peer review]. An occupational therapist [removed for peer review] visited the families' home to complete the assessment. The sense\_assess© kids takes approximately 40 minutes to administer and the Box and Block Test takes approximately five minutes. The Manual Ability Classification System (MACS) level (Eliasson et al., 2006) was identified by a 5-minute parent interview.

Statistical Analysis. Somatosensory discrimination improves with age in typically developing children (Taylor et al., 2016), therefore we conducted an analysis of variance to examine differences between age groups in our sample. Impairment was defined as being outside the 95% confidence intervals of normative data which was obtained from 56 typically developing children aged 6 to 8yrs, 9 to11yrs and 12 to 15yrs (Taylor et al., 2017; Taylor et al., 2015; Taylor et al., 2020). Frequencies of combinations of impairments were recorded. Pearson correlation coefficients were used to examine associations between the Tactile Discrimination Test and the Wrist Position Sense Test with motor performance on the Box and Block Test because these data were normally distributed as determined by the Shapiro-Wilke test (W=.97, p=.64; W=.94, p=.09; W=.95, p=.29 respectively). Spearman Rank Correlation was used to examine associations between the functional Tactile Object Recognition Test and motor performance on the Box and Block Test because the data was not normally distributed for the former (W= .87, p < .01). Spearman's Rank Correlation was used to examine associations between the number of impaired somatosensory discrimination domains and performance on the Box and Block Test. Mann Whitney U was used to analyse the effect of manual ability classification level and current botulinum toxin therapy receipt or non-receipt on somatosensory discrimination.

# Results

Thirty-three children met the inclusion criteria. Three children declined, and 30 children enrolled in the study, two of whom were physically unable to complete the assessment. 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. Fifteen children had received botulinum toxin therapy in the past and one child had upper-limb surgery more than 12 months prior to participation in this study. All 28 children completed the Protective Touch Test, functional Tactile Object Recognition Test and the Wrist Position Sense Test, 27 completed the Tactile Discrimination Test- one child was unable to complete

the measure; and 26 completed the Box and Blocks Test, two were not asked to complete the measure in error.

**Somatosensory discrimination.** Twenty-three (82%) 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: Wrist Position Sense Test (F(1,26) = 0.77, p = 0.39); functional Tactile Object Recognition Test (F(1,26) = 0.77, p = 0.39); functional Tactile Object Recognition Test (F(1,26) = 0.77, p = 0.39); functional Tactile Object Recognition Test (F(1,26) = 0.76); Protective Touch Test (F(1,26) = 1.07, p = 0.31). Twenty of the 28 children (74%) demonstrated impaired wrist position sense (see Table I). Twenty-one (75%) children demonstrated impaired haptic object recognition on the functional Tactile Object Recognition Test (see Table II). Eighteen (67%) children demonstrated impaired tactile discrimination on the Tactile Discrimination Test (see Table III). Six of the 28 children (21%) showed impairment in registering protective touch using the Protective Touch Test. In all age groups there were children with HCP who demonstrated performance within the range of typically developing children (please see tables I, II and III).

# [Insert tables I, II and III here please]

**Combinations of Somatosensory Discrimination Impairment.** More than half of the children (16/28) had impairment in three or more somatosensory domains (see figure I). Impairment in protective touch only occurred in association with impairment across all the discriminative somatosensory measures.

### [Insert figure I here please]

**Somatosensory Discrimination and Motor Performance.** A moderate negative correlation (rs= -.57, p<.01) (Mukaka, 2012) exists between the number of impaired somatosensory domains and performance on the Box and Block Test with a discernible decline in median scores as the number of impaired somatosensory domains increases (see Figure I). Low to moderate correlations were present between motor performance as measured by the Box and Block Test and the discriminative components of somatosensation in the sense\_assess© kids. The Wrist Position Sense Test and the Tactile Discrimination Test demonstrated a low negative and low positive correlation with motor performance (rs (24) = -0.48, p=.01) and (rs (23)=0.43, p=.03) respectively. The functional Tactile Object Recognition Test showed a moderate correlation with motor performance on the Box and Block Test (rs = 0.66, p= <.01).

Somatosensory Discrimination and Manual Ability. There was also a significant association between Manual Ability Classification Level (MACS) and motor performance with children in MACS level I obtaining higher scores (Mdn= 36) than those in MACS level II (Mdn= 15) (U= 5.5, p<.01). Manual ability classification was also associated with limb position sense with children in MACS level I, achieving lower average error scores (Mdn=13.2) than MACS level II (Mdn= 20.7), (Z= 2.6, p=.01). Haptic object recognition was similarly associated with children in MACS level I, demonstrating greater accuracy (Mdn= 39) than MACS level II (Mdn= 29), Z= -2.4, p=.02). MACS level did not show a significant association with tactile discrimination or protective touch scores. There was no significant association with having received botulinum toxin for muscle spasticity in the past on either somatosensory discrimination (Protective Touch Test U= 87, p= .65; Wrist Position Sense Test U= 71, p= .23; functional Tactile Object Recognition Test U= 96, p= .96; Tactile Discrimination Test U= 88, p= .90) or motor performance (U= 54, p= .15).

#### Discussion

The frequency of impairment in somatosensation we observed in our sample of children with HCP using the sense assess<sup>©</sup> kids is slightly higher but comparable with previous findings (Auld, Boyd, et al., 2012b). 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 in a study of 52 children with HCP (Auld, Boyd, et al., 2012b). In both studies children with impaired protective touch were found to have impairments across all measured somatosensory domains and we would suggest these children be prioritised for intervention. Our finding that 67% of children in our study had reduced tactile discrimination ability is consistent with previous reports indicating it is frequently impaired (Auld, Boyd, et al., 2012b; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2008). Using a novel measure of haptic object recognition, we observed impairment in 75% of our sample. This result is similar to a study in which seven of nine children with HCP were observed to have impairment (Cooper et al., 1995). Our result is higher than that reported by two studies with larger sample sizes 63% (N=52) (Auld, Boyd, et al., 2012b) and 51% (N=55) (Yekutiel, Jariwala, & Stretch, 1994) however in light of a recommendation for a more challenging measure of haptic object recognition (Auld, Ware, Boyd, Moseley, & Johnston, 2012) their results may be underestimates.

Wrist position sense was measured using a quantitative approach and we observed reduced performance in 74% of our sample. We had substantially more children with reduced performance than other studies. Assessments of limb position sense in those other studies used a less precise method of measuring limb position such as subjective reporting or/subjectively determined direction of movement or position matching (Cooper et al., 1995; Van Heest et al., 1993). The higher rate observed in our work most likely reflects the greater sensitivity of an interval rather than an ordinal or categorical measurement scale (Taylor et al., 2017; Taylor et al., 2020) and is consistent with differences between clinically oriented tests and the Wrist Position Sense Test in adults with stroke (Carey, Matyas, & Oke, 2002).

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 children with HCP 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, or if conversely those children would make the greatest functional gain in such an intervention. 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.

Twenty-three (82%) of the 28 children in our sample had an impairment in one or more domains of somatosensory function. Not all children had impairment in all assessed somatosensory domains, however we did find that more than two thirds (71%) of our sample had impairment in two or more domains. While this frequency of impaired somatosensory discrimination function is important, we also observed that a higher number of impaired somatosensory domains was associated with poorer unimanual motor performance.

The low to moderate correlations observed between somatosensory discrimination and gross manual dexterity in our sample of 28 children with hemiplegic CP, further support the role of somatosensory impairment in poor motor performance and begs further study with children from a broader range of manual abilities. This is further supported by the findings of a study of 25 children with HCP that found moderate to strong correlations between measures of tactile sensibility and an object pick-up test (Krumlinde-Sundholm & Eliasson, 2002), a study of 15 children with HCP that found a relationship between tactile sensibility and fine control of fingertip force regulation (Gordon & Duff, 1999), and a study of 52 children with HCP

that reported correlations between spatial tactile deficits and both unimanual and bimanual performance (Auld, Boyd, et al., 2012a). A preliminary investigation by this research team found that improvements in limb position sense co-occurred with improvements in bimanual hand-use, with gains in both maintained at 6-months post intervention (McLean et al., 2017). Collectively these studies highlight important associations between somatosensory functions and upper-limb motor performance with emerging evidence suggesting somatosensation is modifiable and may improve bimanual hand function. These findings emphasise the need to address somatosensory discrimination impairments when planning therapy/strategies to improve upper-limb motor function.

The number of somatosensory domains that were impaired influenced the quality of motor performance, such that the higher the number of impaired somatosensory domains the worse the performance on the Box and Block Test. Clinically this may be useful for prioritising treatment for children with HCP, particularly with literature suggesting outcomes and maintenance of gain for motor learning interventions and upper-limb surgery are associated with somatosensory function (Robert et al., 2013; Zancolli, 2003). Investigations of the efficacy of interventions for somatosensory impairment and subsequent motor learning are required to confirm this.

We did not find a significant association with past botulinum receipt and somatosensory discrimination impairment. Clinics providing specialist upper-limb assessment for spasticity management may not have an advantage in identifying children at risk of somatosensory impairment. However, while this current study found that MACS level was associated with somatosensory discrimination impairment, previous work examining haptic exploratory procedures in children with HCP found no association between manual ability and accuracy in somatosensory tasks (Taylor, Girdler, et al., 2019). Taken in combination these findings support the need for routine assessment of somatosensory function for children with HCP in primary and tertiary service settings.

Currently there is only emerging evidence for interventions targeting somatosensory ability for children with HCP (Kuo et al., 2016; McLean et al., 2017; Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2013). There is a growing body of literature recommending interventions to address somatosensory impairment be investigated in this group of children (Auld, Russo, Moseley, & Johnston, 2014; Dan, 2020; Klingels et al., 2012; Majnemer et al., 2008; Van de Winckel et al., 2013). Early investigations suggest that somatosensory discrimination functions are modifiable and result in improvements in bimanual hand-use (McLean et al., 2017). However, this has only been observed in a small sample and more research is needed to investigate treatment options and their impact on somatosensation, motor function, hand use, and occupation. To this end investigations into the impact of somatosensory discrimination impairment on occupational performance are also warranted. 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 provide evidence for the sense\_assess© kids.

Limitations. We only assessed somatosensory function of the more impaired 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 (Auld, Boyd, et al., 2012b). This suggests that the 'unimpaired' limb should be assessed with a view to intervention rather than providing a control comparison. This study presents baseline assessment data of a study that investigated an intervention that utilises the less impaired hand as a 'reference' hand to calibrate somatosensory inputs and the impaired hand was prioritised for assessment since it was to be the focus of intervention. The sense\_assess© kids has been modified for use with children aged 6-15 years however children with CP are increasingly diagnosed before 2 years of age making clinical awareness of the high incidence of somatosensory impairment, the development of assessments for young children and integrated intervention approaches a priority for future work in this area.

# Conclusions

Impaired somatosensory discrimination function is common in children with HCP, with 82% of our sample having impairment. A moderate association was found between the number of impaired somatosensory domains and unimanual motor performance as well as low to moderate correlations between each somatosensory discrimination domain and motor performance. The knowledge that children with HCP commonly have impairments in somatosensory discrimination function is well established. However, the expression of these deficits and their influence on function is less 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 impact of somatosensation on manual ability, 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.

# **Key Points for Occupational Therapy**

- Assessment of somatosensory function is essential as part of routine clinical care for children with HCP.
- Impairment across a greater number of somatosensory domains results in poorer unimanual motor performance.
- Characterising somatosensory impairments has implications for targeted assessment and treatment for upper-limb function.

### Authors declaration of authorship contribution

All authors made substantial contributions to the conception and design of the work (BM, JV, LC, CE); OR the acquisition, analysis, or interpretation of data (BM, ST, JV, AT, LC, CE); All authors were involved with drafting the work (BM) or revising it critically for important intellectual content (ST, JV, AT, JC, CE). All authors gave final approval of the version to be published. All agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved as outlined in the ICMJE recommendation.

#### Data availability statement

The data that support the findings of this study are available from the corresponding author, BM, upon reasonable request.

### Funding statement

The authors would like to acknowledge support from the [removed for peer review] and Australian Postgraduate Awards [removed for peer review], the [removed for peer review] Early Career Research Fellowship [removed for peer review], the National Health and Medical Research Council (NHMRC) project grant [removed for peer review]; NHMRC partnership grant #1134495) and James S. McDonnell Foundation Collaborative Award [removed for peer review].

# **Declaration of interest**

The authors report no conflicts of interest.



# Acknowledgements

The authors would like to thank the participants for their precious time, the [removed for peer review] team at [removed for peer review] for their support and assistance with this project, as well as [removed for peer review] for their guidance in preparing this manuscript.



**Table I:** Performance of children with cerebral palsy on the Wrist Position Sense Test by age group, and with reference to performance in typically developing children.

	Typically	Cerebral Palsy $(N = 28)$ affected hand				
	Developing					
	(95% Confidence					
	Interval of mean					
	score)					
Wrist	Lower- upper bound	Poorest	Best	Median	Impaired (%)	
Position	Left hand	score	score			
Sense Test						
(average						
error score,						
in degrees)	D					
6 to 8 yrs.	12 – 18	37.0	8.9	15.5	4 of 9 (44%)	
n= 9						
9 to 11 yrs.	9.2 – 13.2	41.0	10.2	21.0	11 of 12 (92%)	
n= 12						
12 to 15 yrs.	9 – 12.2	31.2	7.1	19.4	5 of 7 (71%)	
n= 7						

Total Impairment	20 of 28 (74%)

Note: Lower scores reflect better performance on this test.

**Table II:** Performance of children with cerebral palsy on the functional Tactile Object

 Recognition Test by age group, and with reference to performance in typically developing children.

	Typically Developing	Cerebral	Palsy $(N = 2)$	28) affected	l hand
	(95% Confidence				
	Interval of mean				
C	score)				
functional	Lower - upper bound	Poorest	Best	Median	N impaired
Tactile Object	Left hand	score	score		(%)
Recognition					
Test					
(summed					
accuracy score)					
6 to 8 yrs.	39-40	3	41	34	7 of 9 (78%)
n= 9					
9 to 11 yrs.	39 – 41	7	39	30.5	10 of 12
n= 12	-				(83%)
12 to 15 yrs.	40 - 41	10	41	38	4 of 7 (57%)
n= 7					
Total Impairment					21 of 28
				(75%)	

Note: Higher scores reflect better performance. Maximum accuracy score is 42.

**Table III:** Performance of children with cerebral palsy on the Tactile Discrimination Test by age group, and with reference to performance in typically developing children.

	Typically Developing	Cerebral	Palsy (N=	27) affected	hand
	(95% Confidence				
	Interval of mean				
	score)				
Tactile	Lower - upper bound	Poorest	Best	Median	Impaired (%)

Discrimination	Left hand	score	score		
Test					
6 to 8 yrs.	27.3 - 44.9	2.2	50.3	30.7	4 of 9 (44%)
n= 9					
9 to 11 yrs.	52.1 - 67.9	-36.6	73.6	18.8	9 of 12 (75%)
n= 12					
12 to 15 yrs.	64.9 - 84.1	-15.9	68.4	14.7	5 of 6 (83%)
n= 6					
Total Impairment	18 of 27				
					(67%)

Note: Higher scores reflect better performance.





Figure legend: Scores are labelled to indicate the children's MACS level. The combinations of impairments are as follows: no domains- not applicable; one domain- Haptic object recognition (n=2), Limb position sense (n=1); two domains- Limb position & haptic object

recognition (n=2), Limb position & texture discrimination (n=1), Texture discrimination & haptic object recognition (n=1); three domains- Limb position, haptic object recognition and texture discrimination (n=10); and four domains- Limb position, haptic object recognition, texture discrimination, protective touch (n=6).

# References

- Araneda, R., Ebner-Karestinos, D., Paradis, J., Saussez, G., Friel, K. M., Gordon, A. M., & Bleyenheuft,
   Y. (2019). Reliability and responsiveness of the Jebsen-Taylor Test of Hand Function and the
   Box and Block Test for children with cerebral palsy. Developmental Medicine & Child
   Neurology, 61(10), 1182-1188. doi:https://doi.org/10.1111/dmcn.14184
- Auld, M., Boyd, R., Moseley, G. L., Ware, R., & Johnston, L. M. (2012a). Impact of tactile dysfunction on upper-limb motor performance in children with unilateral cerebral palsy. *Archives of Physical Medicine & Rehabilitation, 93*(4), 696-702. doi:10.1016/j.apmr.2011.10.025
- Auld, M., Boyd, R., Moseley, G. L., Ware, R., & Johnston, L. M. (2012b). Tactile function in children with unilateral cerebral palsy compared to typically developing children. *Disability & Rehabilitation, 34*(17), 1488-1494. Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=s3h&AN=77399344&site=ehost-live
- Auld, M., Ware, R., Boyd, R., Moseley, G. L., & Johnston, L. M. (2012). Reproducibility of tactile assessments for children with unilateral cerebral palsy. *Physical & Occupational Therapy In Pediatrics*, 32(2), 151-166. doi:10.3109/01942638.2011.652804
- Auld, M. L., Russo, R., Moseley, G. L., & Johnston, L. M. (2014). Determination of interventions for upper extremity tactile impairment in children with cerebral palsy: a systematic review. *Dev Med Child Neurol, 56*(9), 815-832. doi:10.1111/dmcn.12439
- Carey, L., Nankervis, J., LeBlanc, S., & Harvey, L. (2006). *A new functional Tactual Object Recognition Test (fTORT) for stroke clients: Normative standards and discriminative validity.* Paper presented at the 14th International Congress of the World Federation of Occupational Therapists, Sydney, Australia.
- Carey, L., Oke, L., & Matyas, T. A. (1996). Impaired limb position sense after stroke: A quantitative test for clinical use. *Archives of Physical Medicine & Rehabilitation, 77*(12), 1271-1278. doi:10.1016/s0003-9993(96)90192-6
- Carey, L. M., Mak-Yuen, Y. Y. K., & Matyas, T. A. (2020). The Functional Tactile Object Recognition Test: A Unidimensional Measure With Excellent Internal Consistency for Haptic Sensing of Real Objects After Stroke. *Frontiers in Neuroscience, 14*(948). doi:10.3389/fnins.2020.542590

- Carey, L. M., Matyas, T. A., & Oke, L. E. (2002). Evaluation of impaired fingertip texture discrimination and wrist position sense in patients affected by stroke: Comparison of clinical and new quantitative measures. *Journal of Hand Therapy*, 15(1), 71-82. doi:https://doi.org/10.1053/hanthe.2002.v15.01571
- Carey, L. M., Oke, L. E., & Matyas, T. A. (1997). Impaired Touch Discrimination After Stroke: A Quantiative Test. *Neurorehabilitation and Neural Repair, 11*(4), 219-232.
- Cooper, J., Majnemer, A., Rosenblatt, B., & Birnbaum, R. (1995). The determination of sensory deficits in children with hemiplegic cerebral palsy. *Journal of child neurology, 10*(4), 300.
- Dan, B. (2020). Cerebral palsy is a sensorimotor disorder. *Developmental Medicine & Child Neurology, 62*(7), 768-768. doi:10.1111/dmcn.14542
- Dunn, W., Griffith, J. W., Morrison, M. T., Tanquary, J., Sabata, D., Victorson, D., . . . Gershon, R. C.
  (2013). Somatosensation assessment using the NIH Toolbox. *Neurology, 80*(11 Suppl 3), S41S44. doi:10.1212/WNL.0b013e3182872c54
- Eliasson, A. c., Krumlinde-sundholm, L., Rösblad, B., Beckung, E., Arner, M., Öhrvall, A. m., & Rosenbaum, P. (2006). The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine & Child Neurology*, 48(7), 549-554. doi:10.1111/j.1469-8749.2006.tb01313.x
- Gibson, E., J., & Pick, A., D. (2000). An Ecological Approach to Perceptual Learning and Development. New York, New York: Oxford University Press.
- Gordon, A. M., & Duff, S. V. (1999). Relation between clinical measures and fine manipulative control in children with hemiplegic cerebral palsy. *Developmental Medicine & Child Neurology*, *41*(9), 586-591. doi:10.1111/j.1469-8749.1999.tb00661.x
- Jongbloed-Pereboom, M., Nijhuis-van der Sanden, M. W. G., & Steenbergen, B. (2013). Norm scores of the Box and Block Test for children ages 3-10 years. *The American Journal of Occupational Therapy*, *67*(3), 312-318. doi:http://dx.doi.org/10.5014/ajot.2013.006643
- Klingels, K., De Cock, P., Molenaers, G., Desloovere, K., Huenaerts, C., Jaspers, E., & Feys, H. (2010). Upper limb motor and sensory impairments in children with hemiplegic cerebral palsy. Can they be measured reliably? *Disability & Rehabilitation, 32*(5), 409-416. doi:10.3109/09638280903171469
- Klingels, K., Demeyere, I., Jaspers, E., De Cock, P., Molenaers, G., Boyd, R., & Feys, H. (2012). Upper limb impairments and their impact on activity measures in children with unilateral cerebral palsy. *European Journal of Paediatric Neurology*, *16*(5), 475-484. doi:http://dx.doi.org/10.1016/j.ejpn.2011.12.008

Krumlinde-Sundholm, L., & Eliasson, A.-C. (2002). Comparing tests of tactile sensibility: Aspects relevant to testing children with spastic hemiplegia. *Developmental Medicine and Child Neurology, 44*(9), 604-612. Retrieved from http://search.proquest.com/docview/195587295?accountid=14681

Kuo, H.-C., Gordon, A. M., Henrionnet, A., Hautfenne, S., Friel, K. M., & Bleyenheuft, Y. (2016). The effects of intensive bimanual training with and without tactile training on tactile function in children with unilateral spastic cerebral palsy: A pilot study. *Research in Developmental Disabilities, 49-50*, 129-139. doi:https://doi.org/10.1016/j.ridd.2015.11.024

- Majnemer, A., Bourbonnais, D., & Frak, V. (2008). The role of sensation for hand function in children with cerebral palsy. In A. C. Eliasson & P. A. Burtner (Eds.), *Improving Hand Function in Children with Cerebral Palsy: theory, evidence and intervention* (pp. 134-146). London: Mac Keith Press.
- Mathiowetz, V., Federman, S., & Wiemer, D. (1985). Box and block test of manual dexterity: Norms for 6–19 year olds. *Canadian Journal of Occupational Therapy, 52*(5), 241-245. doi:10.1177/000841748505200505
- McLaughlin, J. F., Felix, S. D., Nowbar, S., Ferrel, A., Bjornson, K., & Hays, R. M. (2005). Lower extremity sensory function in children with cerebral palsy. *Pediatric Rehabilitation, 8*(1), 45-52. doi:10.1080/13638490400011181
- McLean, B. (2018). Discovering the sense of touch: somatosensory discrimination in children with cerebral palsy. (Doctor of Philosophy). University of Western Australia, Nedlands.
- 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. *The American Journal of Occupational Therapy*, 71(3).
- Mukaka, M. M. (2012). Statistics corner: A guide to appropriate use of correlation coefficient in medical research. *Malawi medical journal : the journal of Medical Association of Malawi, 24*(3), 69-71. Retrieved from https://pubmed.ncbi.nlm.nih.gov/23638278
- Robert, M. T., Guberek, R., Sveistrup, H., & Levin, M. F. (2013). Motor learning in children with hemiplegic cerebral palsy and the role of sensation in short-term motor training of goaldirected reaching. *Developmental Medicine & Child Neurology*, 55(12), 1121-1128. doi:10.1111/dmcn.12219

- Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., & Bax, M. (2007). The Definition and Classification of Cerebral Palsy. *Developmental Medicine & Child Neurology, 49*, 1-44. doi:10.1111/j.1469-8749.2007.00001.x
- Smorenburg, A. R. P., Ledebt, A., Deconinck, F. J. A., & Savelsbergh, G. J. P. (2013). Practicing a matching movement with a mirror in individuals with spastic hemiplegia. *Research in Developmental Disabilities, 34*(9), 2507-2513. doi:https://doi.org/10.1016/j.ridd.2013.05.001
- Stanley, F., Blair, E., & Alberman, E. D. (2000). *Cerebral palsies : epidemiology and causal pathways* London: London : Mac Keith.
- Taylor, S. (2017). Evidence Based Adaptation and Evaluation of a Somatosensory Assessment for use with Children with Cerebral Palsy. (Doctor of Philosphy). Curtin University, Perth, Western Australia.
- Taylor, S., Carey, L., Mak, Y., Tan, A.-M., McLean, B., Girdler, S., & Elliott, C. (2017). Sense\_assess© kids Administration manual.
- Taylor, S., Elliott, C., Girdler, S., Valentine, J., Carey, L., McLean, B., . . . Parsons, R. (2015). Construct validity of the wrist position sense test and functional tactile object recognition test . Paper presented at the American Academy of Cerebral Palsy and Developmental Medicine, Austin, Texas, United States of America. http://dx.doi.org/10.1111/dmcn.9\_12887
- Taylor, S., Elliott, C., McLean, B., Parsons, R., Falkmer, T., Carey, L., . . . Girdler, S. (2020). Construct validity, reliability and responsiveness of the Wrist Position Sense Test for use with Children with cerebral palsy. Manuscript submitted for publication.
- Taylor, S., Girdler, S., McCutcheon, S., McLean, B., Parsons, R., Falkmer, T., . . . Elliott, C. (2019).
   Haptic Exploratory Procedures of Children and Youth with and without Cerebral Palsy.
   *Physical & Occupational Therapy In Pediatrics, 39*(3), 337-351.
   doi:10.1080/01942638.2018.1477228
- Taylor, S., Girdler, S., Parsons, R., McLean, B., Falkmer, T., Carey, L., . . . Elliott, C. (2018). Construct validity and responsiveness of the functional Tactile Object Recognition Test for children with cerebral palsy. *Australian Occupational Therapy Journal, 65*(5), 420-430. doi:10.1111/1440-1630.12508
- Taylor, S., McLean, B., Blair, E., Carey, L. M., Valentine, J., Girdler, S., & Elliott, C. (2018). Clinical acceptability of the sense\_assess© kids: Children and youth perspectives. Australian Occupational Therapy Journal, 65(2), 79-88. doi:10.1111/1440-1630.12429
- 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

- Taylor, S., McLean, B., Falkmer, T., Carey, L. M., Girdler, S., Elliott, C., & Blair, E. (2019). Assessing body sensations in children: Intra-rater reliability of assessment and effects of age. *British Journal of Occupational Therapy*, 82(3), 179-185. doi:10.1177/0308022618786933
- Van de Winckel, A., Klingels, K., Bruyninckx, F., Wenderoth, N., Peeters, R., Sunaert, S., ... Feys, H.
  (2013). How does brain activation differ in children with unilateral cerebral palsy compared to typically developing children, during active and passive movements, and tactile stimulation? An fMRI study. *Research in Developmental Disabilities*, *34*(1), 183-197.
  doi:http://dx.doi.org/10.1016/j.ridd.2012.07.030
- Van Heest, A. E., House, J., & Putnam, M. (1993). Sensibility deficiencies in the hands of children with spastic hemiplegia. *The Journal of Hand Surgery, 18*(2), 278-281.
- Walmsley, C., Taylor, S., Parkins, T., Carey, L., Girdler, S., & Elliott, C. (2018). What is the current practice of therapists in the measurement of somatosensation in children with cerebral palsy and other neurological disorders? *Australian Occupational Therapy Journal*, 65(2), 89-97. doi:10.1111/1440-1630.12431
- Weinstein, S. (1993). Fifty years of somatosensory research: from the Semmes-Weinstein monofilaments to the Weinstein Enhanced Sensory Test. *Journal of Hand Therapy 6*(1), 11-22.
- Williamson, G. G., & Anzalone, M. E. (2001). Sensory integration and self-regulation in infants and toddlers : helping very young children interact with their environment. (1st ed.. ed.).
   Washington, DC: Washington, DC: Zero To Three.
- Wingert, J. R. P. M. P. T., Burton, H. P., Sinclair, R. J. P., Brunstrom, J. E. M. D., & Damiano, D. L. P. P.
  T. (2008). Tactile sensory abilities in cerebral palsy: deficits in roughness and object
  discrimination. *Developmental Medicine and Child Neurology*, *50*(11), 832-838. Retrieved
  from

https://link.library.curtin.edu.au/gw?url=https://www.proquest.com/docview/195608484?a ccountid=10382

- Yekutiel, M., Jariwala, M., & Stretch, P. (1994). Sensory deficit in the hands of children with cerebral palsy: A new look at assessment and prevalence. *Developmental Medicine & Child Neurology, 36*(7), 619-624. doi:10.1111/j.1469-8749.1994.tb11899.x
- Zancolli, E. A. (2003). Surgical management of the hand in infantile spastic hemiplegia. *Hand Clinics, 19*(4), 609-629. doi:https://doi.org/10.1016/S0749-0712(03)00034-9