Can a Novel 'Serious Gaming' Technology Improve Upper Limb Sensation and Function in Children with Cerebral Palsy? A Population-based Cohort Study and Pilot Randomised Controlled Trial

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Abbreviations

3D	Three Dimension or Three Dimensional
ACPR	Australian Cerebral Palsy Register
AHA	Assisting Hand Assessment
ANZCTR	Australia and New Zealand Clinical Trials Register
AR	Augmented Reality
AT	Assistive Technology
AUD	Australian dollar
AVG	Active Video Game
Ax	Assessment
Bi	Bilateral
CAD	Computer Aided Design
CNS	Central Nervous System
CONSORT	Consolidated Standards of Reporting Trials
COPM	Canadian Occupational Performance Measure
CP	Cerebral Palsy
CP QOL	Cerebral Palsy Quality of Life
CT	Computed Tomography
Dom	Dominant
GMFCS	Gross Motor Function Classification System

GST	Good and Services Tax
HREC	Human Research Ethics Committee
ICC	Intraclass Correlation Coefficient
ICF	International Classification of Functioning, Disability, and Health
IR	Infrared
JTHFT	Jebsen-Taylor Hand Function Test
L	Left
LED	Light emitting diode
MACS	Manual Ability Classification System
MCID	Minimal Clinically Important Difference
MRI	Magnetic Resonance Imaging
ND	Non-dominant
OGS	OrbIT Gaming System
OT	Occupational Therapist
PCB	Printed circuit board
PNS	Peripheral Nervous System
PT	Physiotherapist
QOL	Quality of Life
RCT	Randomised Controlled Trial
R	Right
RA	Research Assistant

SA	South Australia
SACPR	South Australian Cerebral Palsy Register
SD	Standard deviation
SE	Standard error
SG / SGs	Serious Game / Serious Games
SWM	Semmes Weinstein Monofilaments
TDC	Typically Developing Children
TGA CTN	Therapeutics Goods Administration Clinical Trial Notification
TPD	Two-point discrimination
Uni	Unilateral
UniSA	University of South Australia
USA	United States of America
USB	Universal Serial Bus
UTN	Universal Trials Number
VR	Virtual Reality
WCH	Women's & Children's Hospital
WCHF	Women's & Children's Hospital Foundation
WCHN	Women's & Children's Health Network
WHO	World Health Organisation

Abstract

Cerebral palsy (CP) is a permanent condition and the most common cause of physical disability in childhood (Reddihough, 2011; Herbert *et al.*, 2016). Affecting approximately two per 1,000 live births in Australia (ACPR, 2016), the consensus definition for the condition recognises that CP is also accompanied by disturbances of sensation (Rosenbaum *et al.*, 2007). The primary aim of this PhD thesis was to investigate an upper limb somatosensory intervention for children with CP with a known sensory impairment, and was divided into two stages.

Stage 1 involved recruiting children living with CP from the *South Australian Cerebral Palsy Register* and assessing them for tactile sensory acuity. This was the first time a population-based sensory assessment of children living with CP was conducted in South Australia. Informed by the literature, each child was assessed using validated and reliable sensory tests (tactile registration, proprioception and tactile perception) and a test of functional hand motor skills (the Jebsen-Taylor Hand Function Test, JTHFT). Thirty six children (22 males, age = 10 ± 3.3 years) with either unilateral (n=23) or bilateral (n=13) CP completed the tests satisfactorily. Twenty eight (78%) children recorded a tactile deficit in one or more modality, which is comparable to the literature. Confirming previous reports in the literature, tactile sensory impairments were also recorded in the less-affected (dominant) hand for 52% of children with unilateral CP, and tactile registration deficits were associated with an increased likelihood of tactile perception deficits.

Stage 2 of the research involved a six-week home-based 'serious games' randomised controlled trial (RCT) using the OrbIT Gaming System (OGS). The OGS is a haptic accessible computer gaming system that requires coordinated and integrated upper limb use, whereby deliberate and targeted contextualised vibration stimulation is delivered to the child's hands during gameplay. It was hypothesised that children randomised to treatment with active vibration feedback (Group A) would have significantly better sensory and functional outcomes post-trial compared to children using the OGS but receiving no active vibration stimulation (Group B). This research is the first application of a technology-based intervention directed at improving somatosensory dysfunction in children with CP.

Eighteen children (12 males, age = 10.7 ± 3.4 years) participated in the trial, with 10 children randomised to Group A and eight to Group B. Statistical modelling revealed a significant between group difference for the more affected non-dominant (ND) hand for the test of stereognosis, between baseline (A_1) and the immediate post-trial assessment (A_2), which did not persist at follow-up assessment (A_3) one month later. Since all 18 children participated in a forced bimanual integrated upper limb task, a secondary exploratory analysis and re-modelling was conducted. This analysis revealed a strong statistically significant difference between baseline (A_1) and follow-up (A_3) assessments for the ND hand for the total time taken to complete the JTHFT (p = 0.001), however, a Type II error cannot be ruled out.

This series of studies has added to our knowledge of upper limb sensory loss in children with CP – its prevalence, nature and potential amelioration. The primary recommendation from this PhD is that sensory impairments should continue to be more intensively addressed, and that systems such as the OGS warrant further research to explore effectiveness and utility through an appropriately powered trial.

Declaration

I certify that this thesis does not incorporate without acknowledgment any material previously submitted for a degree or diploma in any university; and to the best of my knowledge and belief, does not contain any material previously published or written by another person except where due reference is made in the text.

David Anthony Hobbs

8th November 2018

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In conclusion, I confirm that I have written this thesis in its entirety, with guidance from my PhD supervisors, *without* the use of a professional editor.

“*Don't tell me the sky's the limit when there are footprints on the moon*”

\- Paul Brandt

Dedication

I dedicate this thesis to my wonderful wife Jodie and our three beautiful children – Nicole (11), Thomas (9) and Eleanor (6). This has been an eight year journey and there were many times towards the end when my thesis took me away from you, or meant that I couldn't be with you, and for this I apologise. It was very difficult not be with you and I will try to not let that happen again.

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Statement of Ethical Conduct

All research that contributed to the studies contained within this PhD were reviewed and approved by relevant Australian Human Research Ethics Committees (HREC), and are acknowledged within this thesis where appropriate.

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1. Introduction

Cerebral palsy (CP) is a permanent condition and the most common cause of physical disability in childhood (Reddihough, 2011; Herbert *et al.*, 2016). The term comes from two words used to define the condition – *cerebral* relating to or of the brain, and *palsy* referring to involuntary muscle tremors. Affecting more than 34,000 people in Australia and 17 million people worldwide, the *Australian Cerebral Palsy Register* (ACPR) reports the prevalence of individuals with CP born between 1993 and 2009 to be 2.1 per 1000 live births (95%CI 2.0 – 2.2)(ACPR, 2016). In South Australia, where this project is based, the reported prevalence rate is 2.0 per 1000 live births – marginally higher than that for Victoria (1.9 per 1,000 live births), but notably less than the rate reported for Western Australia (2.7 per 1000 live births) (ACPR, 2016), the three longest-standing Australian CP Registers. According to the ACPR, males are over-represented in the cohort of all children diagnosed with CP (57%), compared to males representing 51% of all Australian births (ACPR, 2016, pg. 7). There is currently no cure for CP and the highest risk factors for the condition include low birth weight, intrauterine infections, multiple gestation (Odding, Roebroeck, & Stam, 2006) and pre-term delivery, with birth asphyxia playing only a minor role (Longo & Hankins, 2009). In 2007, the annual economic impact of CP in Australia was estimated to be AUD$1.47 billion, or 0.14% of the Gross Domestic Product (Arnfield, Guzzetta, & Boyd, 2013), not including the cost of lost well-being.

In 2006 an international committee of pre-clinical and clinical experts convened to review the definition, classification, and the state of the science for CP. After a series of workshops and commentary, this committee produced what is now considered the consensus definition for CP, stating that "*cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems*" (Rosenbaum *et al.*, 2007, pg. 9). Historically viewed as a physical disability, Rosenbaum (2003) and others had previously highlighted that CP comorbidities were at least as important as the prevailing motor disabilities, hence their specific mention and inclusion in the revised definition. Most health conditions are commonly

described in terms of aetiology and their impact upon an individual's body structures and functions (impairments). However, an individual with CP is also an individual with activity and participation goals, subject to environmental barriers and facilitators, and influenced by a range of personal factors and experiences.

The Constitution for the World Health Organization (WHO) states that health "*is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity*" (WHO, 1946, pg. 1). This definition has not changed since first drafted, however, in 2001 the WHO introduced a new framework entitled the 'International Classification of Functioning, Disability, and Health', more commonly referred to as the ICF (WHO, 2001). The ICF provides a standard language and framework for the description of health and health-related states. The significant step forward encapsulated within the ICF is the shift in thinking and focus – the ICF considers health and functioning rather than disability and aetiology.

The ICF framework broadly consists of two parts: *Functioning and Disability* and *Contextual Factors*. Functioning and Disability includes *Body Functions and Structures* (the anatomy and physiology/psychology of the body, including psychological functions), *Activity* (the execution of a task or action by an individual) and *Participation* (involvement in a life situation) (WHO, 2001), as shown in Figure 1-1. The Contextual Factors include *Environmental Factors* (the physical, social and attitudinal environment in which people live and conduct their lives) and *Personal Factors* (which include gender, age, coping styles, social background, education, profession, past and current experience, overall behaviour pattern, character and other factors that influence how disability is experienced by the individual) (WHO, 2001).

The framework identifies the three levels of human functioning classified by the ICF: functioning at the level of body or body part, the whole person, and the whole person in a social context (WHO, 2001). Disability, therefore, involves a dysfunction at one or more of these same levels: impairments, activity limitations and participation restrictions. The ICF framework recasts the notions of 'health' and 'disability', recognising that while functional status may be related to a health condition, if we consider the reverse case, knowing the health condition does not predict functional status. More significantly, the reframing acknowledges that from time to time anyone

can experience a decrease in their health status and therefore experience some form of disability, even if temporarily. In doing so, the ICF 'mainstreams' the experience of disability, recognising it as a universal human experience, shifting the focus from cause to the impact it places on all health conditions (Kostanjsek, 2011).

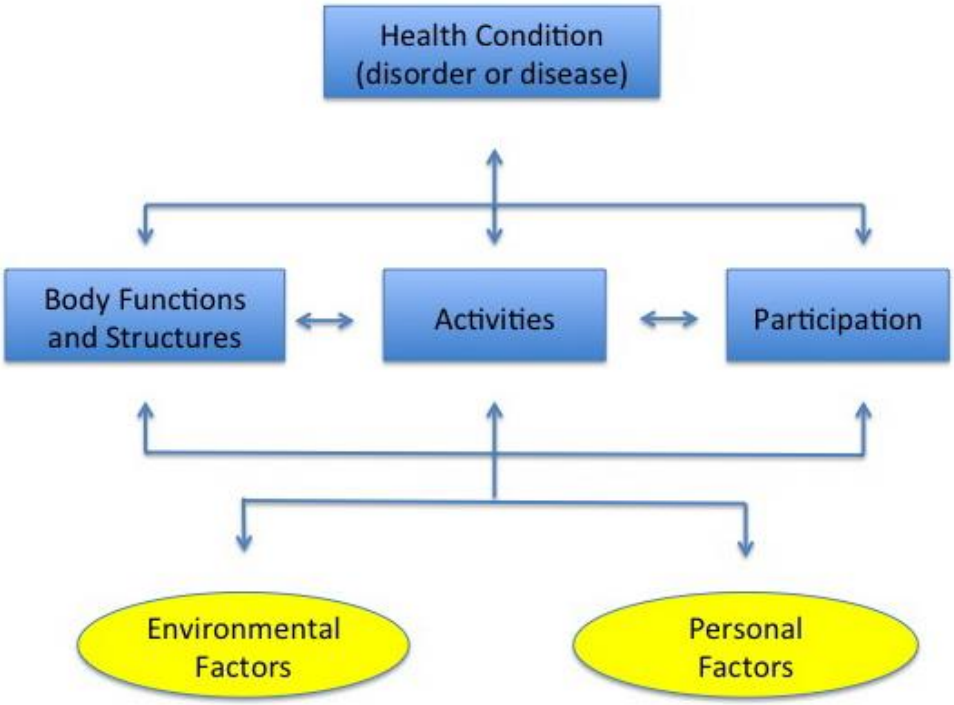

Figure 1-1 – The International Classification of Functioning, Disability, and Health Framework (ICF) (Source: https://commons.wikimedia.org/wiki/File:Version_of_the_ICF.jpg)

1.1 Sensation – A Possibly Neglected Impairment?

Our sense of touch has been described as "*our first language*", the first system that functions in-utero, the system that connects and bonds us to others (Fisher, Murray, & Bundy, 1991, pg. 108), and the most mature sensory system at birth (Clayton, Fleming, & Copley, 2003, pg. 44). The term 'somatosensation' describes the ability of the body to detect and perceive body surface sensations such as touch, pressure, pain, vibration, temperature, joint position or proprioception, and object recognition (Walmsley *et al.*, 2017, pg. 89). The consensus definition of CP makes specific mention of "*disturbances of sensation*", which was a neglected area of clinical investigation and understanding for this population until the pioneering work of Dr's J.P.M Tizard and Bronson Crothers last century. The authors referred to the "*glaring*

gap" (pg. 228) that represented no orderly attempt to collect evidence of sensory deficits when they carefully re-examined 44 cases of children with hemiplegia from their own hospital (Tizard & Crothers, 1952). They acknowledged that without knowing the sensory status of the hands of the children they were working with they were unknowingly forcing them "*to develop skills in hands with gross sensory defects, which made the whole effort absurd*" (pg. 228). They proposed that most children with inadequate growth on the affected side would show sensory disorders, but without a careful sensory study, there was no way to forecast the prevalence or sensory status of children with CP.

Tizard and Crothers' case audit led them to conduct what is generally acknowledged as the first published study that specifically aimed to quantify sensory deficits for this population (Tizard, Paine, & Crothers, 1954). The authors assessed 106 children with hemiplegia, reporting that more than 53% recorded a sensory disturbance, with the tests of stereognosis and two-point discrimination (TPD) being the most challenging tests for the cohort. The authors observed that in some instances the sensory deficit was the main reason a child with CP did not use their affected arm or hand. Additionally, all 16 children with a skeletal undergrowth had impaired sensation and visual fields, as they had proposed two years earlier. Following their sensory study and an additional analysis of four surgical cases involving the hand – where the only successful case involved a child with intact sensation – Tizard *et al.* (1954) concluded that no child with hemiplegia should have an operation on their involved limb without a sensory assessment to first identify and quantify their sensory loss.

To appreciate not only the significance of Tizard *et al.*'s work, but also how it was received and accepted by the professional community at the time, it is worth noting that the next study to be published in this field began with an open apology to Tizard, Paine and Crothers. The first sentence of the paper by Leslie Hohman, Lenox Baker and Ruth Reed (Hohman, Baker, & Reed, 1958) declares that "*the senior author would like to open this paper with an apology to his peers and especially to Dr Bronson Crothers*" (pg. 1), because professionals within the audience doubted the prevalence of sensory disturbances reported when Tizard *et al.* presented their study at the American Academy of Cerebral Palsy Conference in 1954. Following their own

independent study, Hohman *et al.* reported sensory deficits in 72% of their cohort of 47 children (Hohman *et al.*, 1958), a higher prevalence than that of Tizard *et al.*

Since 1952 more than 25 studies have assessed and analysed upper limb sensation in children with CP (discussed and summarised in Chapter 2). The literature has identified and reported somatosensory impairments in the upper limbs of children with CP, but varies in methodological quality and reporting standards across the years, making it difficult to group the studies for a number of reasons. These reasons include the lack of a definition as to what constitutes an impaired sense, poor cohort profiling and reporting, the lack of a standardised sensory testing suite, and a lack of detail relating to the testing protocols that were used. The identification and diagnosis of a sensory deficit in a child with CP and the subsequent estimated prevalence of that deficit depends to a large extent on which sensory modalities were tested, and the manner in which they were tested (Yekutiel, Jariwala, & Stretch, 1994).

Within the WHO ICF context, a sensory deficit would be considered an impairment that potentially leads to limitations in the activity domain and restrictions in participation. The processing and interpretation of sensory information is a fundamental property of understanding and interacting with the world around us. Intact upper limb sensation is critical for arm and hand proprioception, fine manual dexterity (Gordon & Duff, 1999), for engaging with the world through touch (grasping/holding and touching objects, etc.), and for avoiding potentially hazardous situations (such as pain receptors to warn of extreme hot/cold surfaces, etc.). Kenney (1966) theorised in the 1960s that a sensory impairment would have to result in a certain degree of motor deficit (pg. 46), with Cooper *et al.* (1995) recognising sensory input as an essential component of motor function and motor control (pg. 300), proposing that sensory deficits may constitute limits on the functional outcome of children with CP (pg. 301). Sensory information such as visual, cutaneous and proprioceptive inputs are known to be essential for the initiation and execution of refined hand movements (Majnemer, Bourbonnais, & Frak, 2008, pg. 138), facilitating the execution of precise hand, grasp, grip and finger movements. The authors went on to say that the impact of sensory deficits on motor performance cannot be overlooked (Majnemer *et al.*, 2008).

Moreover, it has been reported that for cases of severe sensory dysfunction, limb neglect may lead to a non-use phenomenon resulting in limb function deterioration, with decreased or absent afferent brain stimuli appearing to compromise motor learning as well as body image (Majnemer *et al.*, 2008, pg. 142). Consequently, the effect of decreased tactile exploration coupled with decreased afferent stimuli results in changes to early mapping of somatosensory and associated brain structures (Clayton *et al.*, 2003; Majnemer *et al.*, 2008). Effective use of the upper limb is important as it is known to impact on educational outcomes, participation in activities of daily living and vocational options for many children with CP (Boyd, Morris, & Graham, 2001). Wingert *et al.* (2008) acknowledged that cutaneous inputs are used to both localise and characterise qualities of touch (pg. 832).

While sensory impairments in the upper limbs of children with CP are now generally acknowledged, the area of sensory training in this population is not. This is in contrast to the field of post-stroke rehabilitation, where a number of studies have focussed solely on sensory retraining (Dannenbaum & Dykes, 1988; Yekutiel & Guttman, 1993; Chen, Liang, & Shaw, 2005), recognising the capacity of the nervous system to modify its organisation and to re-learn and adapt to new experiences – a process known as neuroplasticity. Indeed, following an initial trial to retrain sensory function in the hands of post-stroke survivors and a follow-up sensory assessment of children with CP, Yekutiel *et al.* observed that the "*efficacy of sensory training has been demonstrated in chronic stroke patients (Yekutiel & Guttman, 1993), but the question of its effect in children with CP has remained unanswered since it was raised 35 years ago by Crothers and Paine*" (Yekutiel *et al.*, 1994, pg. 623).

The evidence for sensory impairments in the hands of children with CP has been established, despite the lack of a consensus suite of tests, varied prevalence rates reported, definitions and classification as to what constitutes an impaired sense, the range of tests applied, and the testing protocols used. What is not well established is the area of somatosensory training for children with CP with a known sensory loss to improve upper limb hand function.

1.2 Human Anatomy Relevant to Tactile Sensation

To provide a background to the in-depth appraisal of sensory impairments in children with CP (Chapter 2), it is necessary to summarise the relevant anatomy and physiology of the human sensory system, and the role of sensation in motor control. The sensation of touch is detected on hairless or glabrous skin (epidermis), such as the palms of the hands and finger tips, via a number of highly specialised receptors. A major component of the peripheral nervous system (PNS), these receptors detect and relay afferent information from the body to the central nervous system (CNS), via the spinal cord to the brain. The receptors of relevance to this work are termed 'mechanoreceptors' due to their ability to detect physical or mechanical stimuli, as opposed to cutaneous receptors that detect pain (nociceptors) and temperature (thermoreceptors). Located beneath and within both the epidermis and dermis, are four main types of mechanoreceptors, as shown in Figure 1-2.

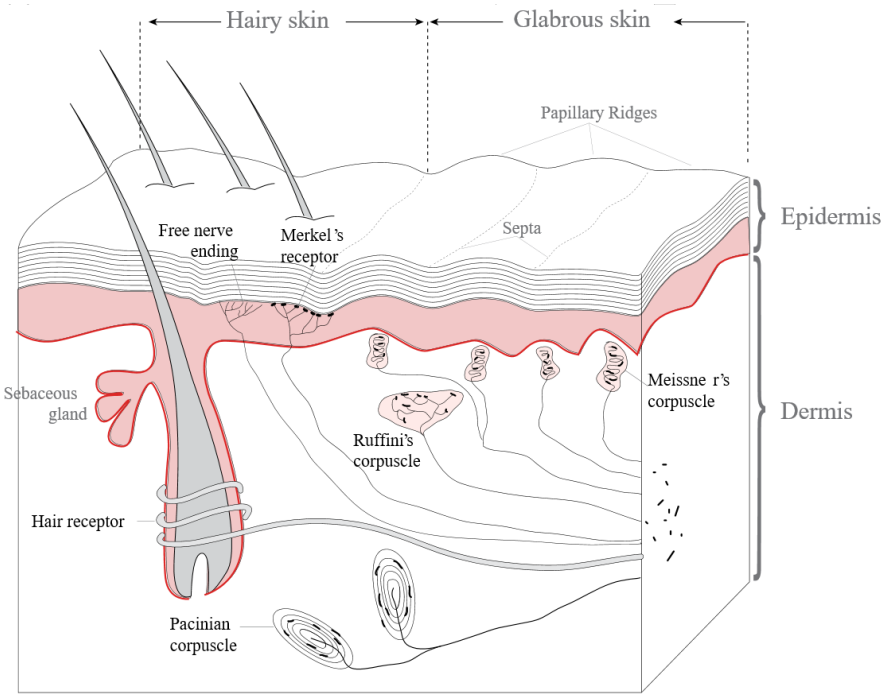

Figure 1-2 – The anatomy of human glabrous skin, showing the location and nature of mechanoreceptors to detect mechanical stimuli (Source: https://commons.wikimedia.org/wiki/File:Skin_proprioception.svg)

The four types of mechanoreceptors that detect external stimulation relating to touch, pressure, vibration, and cutaneous tension are called Meissner's corpuscles,

Pacinian corpuscles, Merkel's disks, and Ruffini's corpuscles (Purves *et al.*, 2001). Each mechanoreceptor has distinctly different response properties (that is, they either rapidly or slowly adapt to a given stimulus) and serve distinctly different perceptual functions (Johnson, Hsiao, & Yoshioka, 2002), and each receptor has a specific anatomical location and function as shown in Table 1.

Table 1 – Location and function of the four types of mechanoreceptors for sensing and relaying cutaneous information in the human hand (adapted from Purves et al. (2001), Chapter 9, The Somatic Sensory System, Table 9.1, The Major Classes of Somatic Sensory Receptors)

Receptor Type	***Location***	***Sensory Function***
Meissner's corpuscles	Principally glabrous skin	Light touch, pressure (dynamic)
Pacinian corpuscles	Subcutaneous tissue	Deep pressure, vibration (dynamic)
Merkel's disks	All skin, hair follicles	Light touch, pressure (static), form and texture perception
Ruffini's corpuscles	All skin, dermis	Stretching of skin

Within the skin of the fingertip, Meissner’s corpuscles lie beneath the epidermis of the fingers and palms, and as shown in Figure 1-2, are elongated receptors formed by a connective tissue capsule that comprises several lamellae of Schwann cells (Purves *et al.*, 2001), the principal glia of the PNS. These corpuscles are connected sideways with the epidermal cells on each side, optimally placed to register sideways shearing of the skin and are most commonly found in the fingertips (Carpenter & Reddi, 2012) and account for approximately 40% of the sensory innervation of the human hand (Purves *et al.*, 2001). Meissner’s corpuscles are particularly efficient in detecting and relaying information about low-frequency vibrations (30–50Hz) that occur when textured objects are moved across the skin (Purves *et al.*, 2001), so are principally involved in motion detection and control of hand grip.

Pacinian corpuscles are located deeper in the subcutaneous tissue and make up 10–15% of the cutaneous receptors in the hand. They are larger than Meissner's corpuscles and differ in their morphology, distribution, and response threshold (Purves *et al.*, 2001). However, both the aforementioned corpuscles provide information primarily about the dynamic qualities of a given mechanical stimulus. The

concentric layers of the Pacinian corpuscle imply a non-directional sensitivity to mechanical deformation (Carpenter & Reddi, 2012), and they are also more sensitive to disturbances at higher frequencies than Meissner's corpuscles, in the range of 250–350Hz (Purves *et al.*, 2001). Because of their lower response threshold (compared to Meissner's corpuscles), Pacinian corpuscles are involved in the discrimination of fine surface textures or other moving stimuli that produce high-frequency vibration of the skin (Purves *et al.*, 2001).

Merkel's disks (or 'Merkel's discs' in some texts) are located at the interface between the epidermis and the dermis and are aligned with the papillae that lie beneath the dermal ridges (Purves *et al.*, 2001). They are extremely sensitive to skin deformation (Carpenter & Reddi, 2012), account for about 25% of the mechanoreceptors of the hand, and are particularly dense in the fingertips. In contrast to Meissner's and Pacinian corpuscles, these receptors detect sustained rather than dynamic touch and pressure, and it is understood that Merkel's disks play a key role in the static discrimination of shapes, edges, and rough textures (Purves *et al.*, 2001).

Ruffini's corpuscles (or 'Ruffini endings') are elongated, spindle-shaped capsules located deep in the skin of the hand, as well as in ligaments and tendons. The long axis of the corpuscle is usually oriented parallel to the stretch lines in skin meaning Ruffini's corpuscles are sensitive to cutaneous stretching produced by digit or limb movements (Purves *et al.*, 2001), hence contributing to the kinesthetic or proprioceptive sense of finger position and movement (Boundless, 2018). Ruffini's corpuscles account for about 20% of the receptors in the human hand (Purves *et al.*, 2001).

To complement the four different cutaneous mechanoreceptors, human skeletal or striated muscle contains another class of receptor that provides sensory information arising from within the body itself. Specialised muscle proprioceptors, such as muscle spindles and Golgi tendon organs, detect changes in muscle length and tension, respectively, providing kinaesthetic information about limb position and other body parts in space (Purves *et al.*, 2001; Carpenter & Reddi, 2012) to the CNS. Wingert *et al.* (2009) describes proprioception as a complex somatosensory modality that incorporates inputs from muscle, joint and cutaneous afferent fibres, and that it

consists of two components, namely, the sense of limb movement (referred to as kinesthesia) and static limb position (referred to as joint-position sense)(pg. 447).

Once a tactile stimulus is detected by the relevant receptor(s), peripheral nerves transfer the sensation to the part of the brain responsible for receiving the relevant information, the thalamus. Strategically located on top of the brainstem in the centre of the cerebral hemispheres (shown in Figure 1-3), the neurons of the thalamus project entirely to the cerebral cortex, hence acting as a relay for afferent signals to the cortex (Carpenter & Reddi, 2012). Often referred to as the brain's 'sensory switchboard', the thalamus does not interpret but conveys information to the relevant sensory areas of the brain, namely the somatosensory cortical area or S1, also called Brodmann areas 3a, 3b, 2, and 1 (Carpenter & Reddi, 2012), located in the dorsal section of the frontal lobe.

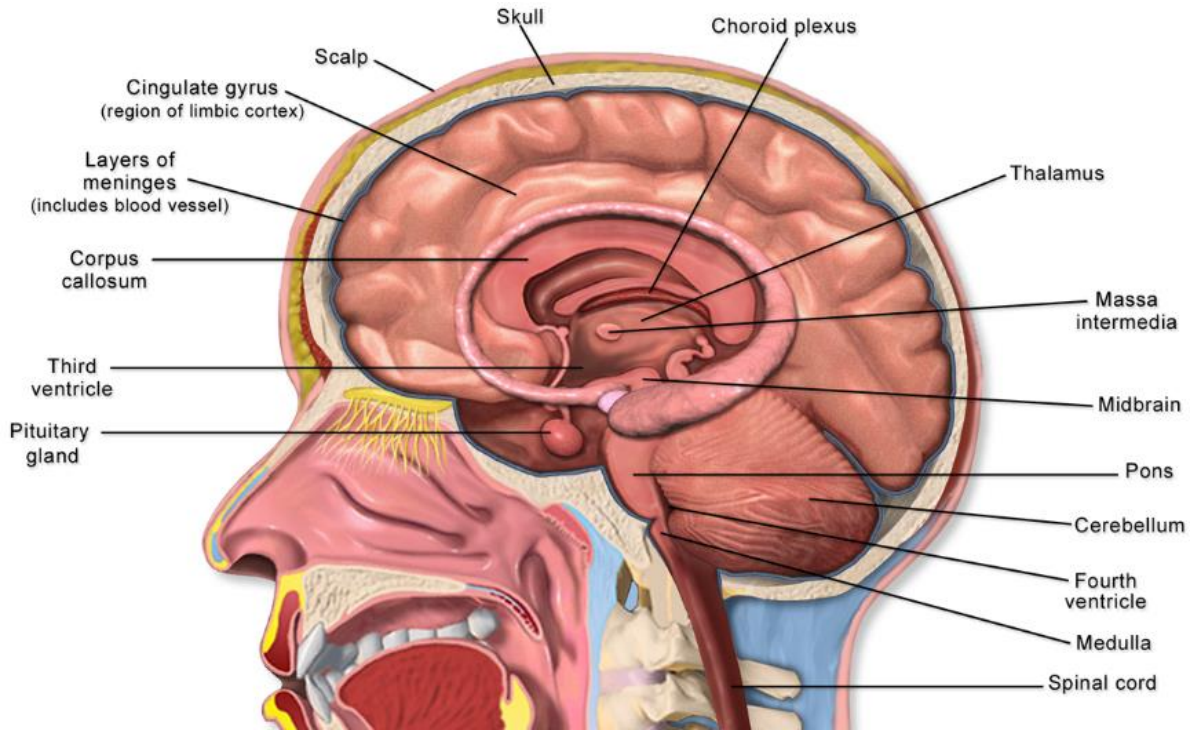

Figure 1-3 – The anatomy of the human brain, showing the location of the thalamus relative to the rest of the brain (Source: https://commons.wikimedia.org/wiki/File:Brain_Anatomy.png)

While area 3b is generally referred to as the 'primary somatosensory cortex' and referred to as 'SI' since it receives the bulk of the thalamocortical projections, all four areas are involved in processing tactile information (Purves *et al.*, 2001). The parietal

lobe, a part of the cerebrum that is positioned on top of the brain, posterior to the frontal lobe, is responsible for integrating and interpreting information relating to touch, and is where sense or meaning is made of a given stimulus. The postcentral gyrus, located within the lateral parietal lobe, is the main sensory receptive area for the sense of touch (Boundless, 2018).

Experimental brain research has determined that a relationship exists between the different areas of the skin on the human body and the primary somatosensory cortex – in essence a 'map' of the human body exists within the contralateral somatosensory cortex of the brain (Carpenter & Reddi, 2012). The representation of the human body within the primary sensory cortex is known as the sensory homunculus, which translates literally to "little man" (Purves *et al.*, 2001). Distinct and separate maps exist for both sensory and motor function, as shown in Figure 1-4.

Figure 1-4 has been removed due to copyright restrictions

Figure 1-4 – The motor (red, left) and sensory (blue, right) homunculus for the human body (Source: https://garyborjesson.wordpress.com/2014/03/06/meet-your-homunculus/)

When these brain maps were first determined it was observed that neighbouring locations on the surface of the body were represented by neighbouring regions of the cortex, but the proportionality was starkly different (Carpenter & Reddi, 2012). The

homunculus defined by the mapping procedures resulted in a grossly enlarged face and hands compared to the torso and proximal limbs (Purves *et al.*, 2001), highlighting that the area of the cortex that is dedicated to the sensation of the human body is proportional to the sensitivity of that body part. The grossly enlarged representation of the hands and human face can be explained by the need for fine, controlled and coordinated manipulation of the hands, and facial expression and speech, which require more central (and peripheral) circuitry to govern them (Purves *et al.*, 2001).

An impairment in sensation can result when there is a disruption or damage to any of the anatomy discussed earlier, such as at the receptor level where a stimulus is first detected, through to integration and processing within the brain and cortex itself. For children with CP, the disruption and resulting impairment results from the location, size and timing (pre-term compared to post-term) of the brain interference, lesion or abnormality of the immature brain. Quoting the work of Clayton *et al.* (2003), Majnemer *et al.* (2008) states that impaired sensation in children with CP is likely the result of damage or malformation of cortical and sub-cortical brain structures, such as the parietal lobe and thalamus (pg. 140).

1.3 Gap Statement

There is a paucity of research investigating training to positively influence impaired somatosensory function in the hands of children with CP. Therefore, this PhD research focusses on: (1) quantifying the nature and prevalence of upper limb somatosensory impairments in a population-based cohort of children living with CP in South Australia, as this has not been done before, and (2) developing and investigating an intervention that targets upper limb sensory impairments, through the rigour of a randomised controlled trial, using a home-based custom-designed serious gaming system. Consequently, the original contribution to research that this thesis presents is an understanding of somatosensory function for children living with CP in South Australia, and the novel application of a serious gaming system that targets somatosensory function.

The aims of this PhD thesis were:

1. To recruit and assess children living with CP in South Australia for upper limb somatosensory function;
2. Determine if the type of CP and the side of the lesion (including the resultant hand dominance) have an influence on the nature and extent of sensory impairment that is identified;
3. Determine the level of correlation between sensory impairment and level of activity (function) in the upper limb;
4. To compare the sensory and motor performance of the dominant and non-dominant (ND) limbs of children with CP;
5. To determine if children with CP with a known upper limb sensory impairment can improve their somatosensory function through using a novel and haptic home-based computer gaming system called the OrbIT Gaming System (OGS).

The hypotheses for this PhD thesis were:

1. Sensation is impaired in the upper limb(s) of children with CP, compared to age-matched typically developing peers;
2. The type of CP and the site of the lesion influences the nature and extent of sensory impairment;
3. The level of sensory impairment correlates with the level of upper limb (dys)function;
4. Sensory impairments will be more prominent on the ND side compared to the dominant side; and
5. Upper limb somatosensory function can be significantly improved through engaging with an accessible home-based computer gaming system. It is hypothesised that children randomised to treatment with vibration stimulation will have significantly better sensory and functional outcomes than children receiving no associated vibration stimulation during gameplay.

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2. Literature Review

This chapter provides an appraisal and synthesis of the literature relevant to upper limb somatosensory assessment studies involving children with CP, as a current analysis of this area does not appear in the literature. In 2008 a summary of studies that assessed sensation for this population was provided in the book chapter by Majnemer *et al.* (2008), but a number of studies were not captured in this analysis, and six studies have since been published.

For this literature review, the studies and their main outcomes are discussed, analysed and presented in chronological order, and tabulated and summarised in Table 2. To enable cross-referencing between the summary that follows and Table 2, the bold number in square brackets (e.g.: **[8]**) after each study is introduced refers to the allocated study number in the table. Relevant information pertinent to each study appears in the table, including a description of each cohort, specific details of each test that was conducted, and the key outcomes from each study. This review encompasses all literature prior and leading up to the conclusion of the overall project, with the discussion in subsequent chapters highlighting new research in light of the findings.

2.1 A Review of Published Somatosensory Studies for the Upper Limb of Children with Cerebral Palsy

The early work of J.P.M. Tizard and Bronson Crothers was pivotal in that it highlighted the need to understand, appreciate and record tactile sensory function for children with CP (Tizard & Crothers, 1952) **[1]**. Their first publication, a historical case note review of 44 children, provides no specific details on the cohort that was assessed in terms of age, sex, or the type of CP, nor does it provide any details of the assessments that were reviewed, which included measurements of the affected limb, vision, and “other sensory” tests (pg. 228). However, this initial work was important as it laid the foundations for both the evidence of sensory agnosia for this population, and instilled a conviction within the researchers to carefully and thoroughly examine sensory agnosia in a more formal and studious manner.

Following their early work, both clinicians committed to determining an effective method for critically examining the sensory status of children with CP, including the testing of young children and children with a cognitive impairment. Two years later, at the American Academy of Cerebral Palsy Conference in 1954, Tizard, Richmond Paine and Crothers published and presented on what is widely recognised as the first article to document and report the prevalence of sensory disturbances in this population (Tizard *et al.*, 1954) **[2]**. This time a range of tests were conducted, including touch, pain, position sense, temperature, passive motion, vibration, location sense, stereognosis, sharp-dull discrimination, two-point discrimination (TPD), and texture recognition. Details on the cohort profile (age and sex) and the specific criteria for how a sensory impairment was classified or justified (in terms of comparing results with a threshold level or 'normal' result) were not reported.

Their study identified a range of sensory impairments in more than half (54%) of their cohort of 106 children with hemiplegia, with the modalities most affected being, in order, stereognosis, TPD, and position or passive motion sense (the specific details are presented in Table 2, for all studies). Tizard and Crothers' earlier suspicion (Tizard & Crothers, 1952) that most children with inadequate limb growth on the affected side will show sensory disorders was confirmed in their later work when all children with skeletal undergrowth (n=16) were identified as having impaired sensation and visual fields. Tizard *et al.* (1954) reported no correlation between sensory dysfunction and motor disability, degree of muscular underdevelopment on the involved side, mental status, the frequency or severity of convulsive seizures, or the age of acquisition (for the group with acquired CP), and observed that stereognosis and TPD could be satisfactorily tested in an intelligent child of five years of age or older. The authors also presented four surgical case studies that involved transplantation of the flexor carpi ulnaris tendon, reporting that only one child benefited from the surgery, and that this particular child did not have a sensory deficit (Case 4). They concluded that no child with hemiplegia should have surgery on their involved upper limb without first conducting a sensory assessment to identify and quantify the sensory status of the limb.

When Tizard *et al.* (1954) presented their work it was met with scepticism amongst clinicians, as evidenced by the open apology that introduces the work of Hohman *et*

al. (1958), the next study to follow on from Tizard *et al.*'s work. Hohman *et al.* (1958) reported on sensory disturbances in 47 children with CP between the ages of six and 16. The examinations were conducted on the upper extremity only and most frequently on the hand, with tests including form discrimination, roughness, sharpness/dullness, light touch, wetness/dryness, hot/cold, TPD (from 0.5 to 38.1mm), measurement of length, position sense, weighing perception, visual field defects, localisation, and speed of response. While a methodology for each test was reported, criteria for how a sense was deemed to be impaired was not.

Hohman *et al.* (1958) **[3]** reported sensory defects in 34 children (72%), with loss of form sense, impairment in TPD, and loss of position sense being the most common sensory defects, in that order. All subjects with a sensory defect had a deficiency in one of the three major modalities just mentioned, and all of the subjects that showed a loss of position sense had both form and TPD involved as well, apart from one subject who did not lose TPD. The authors concluded that their study confirmed the results of Tizard *et al.* (1954), and further identified that the type of sensory defect was mainly of the cortical parietal lobe variety (form sense, TPD and position sense).

Later that same year, Tachdjian and Minear conducted a similar study to Tizard *et al*. and Hohman *et al.*, reporting on the sensory examination of 96 children with CP (Tachdjian & Minear, 1958) **[4]**. Their cohort was mostly children aged between six and 19 years, of average or normal intelligence. The assessment was extensive and comprehensive, including the testing of 13 sensory modalities, muscle tests of the upper extremities (the details of which are not explicit, and this was not done for all subjects), length and girth measurements of the upper extremities, skin temperature measurements, and an assessment of the functional use of the involved hand (graded as none, poor, fair, good and normal). As for the earlier studies, specific details relating to how a sense was deemed to be impaired were not reported.

Sensory deficits were identified in 40 children (42%), with stereognosis, TPD and position sense being the most common modalities affected, the same three tests and precedence ordering reported by both Tizard *et al.* (1954) and Hohman *et al.* (1958). Normal sensation was reported in the non-involved extremity of the hemiplegic cohort. The authors also reported an inverse relationship between the extent of sensory loss and functional use of the involved limb, with almost 88% of children

who had no use of the affected hand identified as having sensory loss, and children with ‘normal’ hand use demonstrating no sensory loss.

Similar to Tizard *et al.*, Tachdjian and Minear also investigated underdevelopment of the affected upper limb (compared to the unaffected limb), reporting the average total shortening being 34.9mm (range: 12.7 to 76.2mm), with the average atrophy being 44.5mm for the arms and 25.4mm for the forearms (pgs. 87-88). The authors identified that the extremities that had the greatest shortening or atrophy were also functionally evaluated as ‘poor’. Skin temperature measurements revealed slight differences between limbs (a constant temperature difference of 0.56-1.11ºC was reported, with the affected limb being cooler than the unaffected limb), but this was only observed in five of the 96 children. The authors noted that this result probably indicated normal upper limb circulation and hence did not support the theory that decreased blood flow was a cause for atrophy and shortening.

From an orthopaedic perspective, the authors reported that 15 of the 40 children with a sensory deficit had had surgery, and that none of the 15 hands showed any noticeable or appreciable functional improvement post-surgery. Echoing comments by Tizard *et al.* (1954), the authors believed that the initial “*extremely impaired*” sensory status of the hand adversely affected the outcome of the surgery, and that “*the motor and sensory status of the involved extremity and of the patient as a whole should be carefully evaluated*” (Tachdjian & Minear, 1958, pg. 90) prior to any surgery being undertaken.

Jones (1960)’s **[5]** study was the first to use a mixed age cohort, with half the 54 participants referred to as adults (aged over 16 years, but no further age information provided) and the other half aged between 19 months and 12 years. Aware of the sensory publications in the literature to date, Jones chose a battery that tested cortical-sensory modalities, namely tests relating to exteroceptive, proprioceptive, and cortico-sensory function, as listed in Table 2. Jones’ study identified sensory deficits in 39 (74%) participants, citing that the most common modalities affected on the involved side were stereognosis, TPD and position or passive movement. The author also reported that the frequency of a given impaired modality varied with age, with light touch impaired in 75% of children less than six years old, compared to only 25% of adults, with similar findings reported for tests for pain or sharp/dull

discrimination. With reference to earlier studies, Jones recommended that a re-evaluation of the sensory function of a child as they mature is essential, particularly prior to any hand surgery. The author stated that a sensory deficit is often associated with poor function of the involved hand, and indicated that a current study (and future publication described later in this chapter, Barrett and Jones (1967)), involving multisensory stimulation training on the development of motor function in young children with hemiplegia was encouraging.

Monfraix, Tardieu, and Tardieu (1961) **[6]** assessed a cohort of 92 children for disturbances of manual perception using five common objects and 12 geometric shapes that were placed into the child's hand but not manipulated. Their work was both innovative and important as it was the first sensory study to use a control group (218 children aged two and a half to eight and a half years old), meaning the authors could determine which shapes typically developing children of a given age could accurately identify and how many. The authors used the term 'gnosic disorder' to describe a disorder of perception, and the control data enabled them to calculate a given child's 'gnosic quotient', which was calculated as: (child's 'gnosic' age ÷ child's mental age) x 100 (pg. 550), enabling the ability to classify a child as being either 'mild' or 'severe', with severe defined as a 'gnosic quotient' < 50.

For the group with hemiplegia (n=22), 14 were classified as either having moderate (n=7) or severe (n=7) agnosia, with five recording bilateral disorders of perception. The side of the hemiplegia did not affect the frequency of the gnosic disorder, and mild gnosic disorders were noted in the unimpaired side. Hence, the work by Monfraix *et al.* (1961) is the first in the literature to report sensory disturbances on the unaffected side, potentially due to the brain lesion being bilateral for approximately 30-40% of children diagnosed with hemiplegia. A result that surprised the authors was the identification of upper limb gnosic disorders when "... *the motor disorders were clearly confined to the lower limbs*" (pg. 552), highlighting that sensory deficits may be present in the absence of motor issues. The study concluded that sensory deficits were present in all types of CP, but were more frequent and severe in children with spasticity or rigidity compared to children with athetoid CP, perhaps more frequent when the right side is affected compared to the left, and are more frequent in cases of hemiplegia compared to diplegia.

A sensory assessment of 19 children with CP that were all older than five years old by Kenney (1963) **[7]** reported that 14 (73%) children had some deficit in sensation. Tests of sensation included stereognosis (using nine objects), TPD, position sense (flexing or extending the fingers or toes), sharp and dull (using the point and blunt end of a safety pin), hot and cold (using a test tube with hot and cold water), palm writing/graphesthesia (both numbers and letters were written with a pencil on the subjects palm) and the size of coins (where the child was asked to determine the differences in coin size). Details relating to how a sensory deficit was identified were not reported. Twenty seven children ranging in age from five to 14 years, from two starkly different socio-economic institutions, were used as a control group for comparison, with no differences reported between the two normal sub-groups.

While 73% of the CP cohort showed a sensory deficit in one or more tests, very few sensory disturbances were identified in children with athetoid CP. Athetosis arises from an injury to the basal ganglia, which is known to specialise in processing information related to movement, and could potentially explain this result. Five of the six children with athetosis had difficulty recognising the difference in coin size, but tested as 'normal' in all other tests. The test that most participants failed was size of coins, followed by TPD, palm writing, sharp and dull, and stereognosis, consequently making it the first study to not list astereognosis as the most common sensory deficit. Astereognosis is associated with an injury to the parietal lobe (Irving, 1968), meaning it's possible that only a few of Kenney's cohort had lesions in this area of the brain. No specific details of the profile of the cohort was given (in terms of average age, age range, sex, type of CP and side affected by the lesion), and no explanation was offered to explain why certain subjects were excluded in the reporting of the results for the sensory tests (19 subjects were tested but none of the total responses sum to 19).

Kenney highlighted that surgery on a hand experiencing sensory loss may not yield useful function because it lacks normal sensation and hence the corresponding stimulus that sensation provides to motor activity. The author further noted that the "*question arises as to whether the patient with a sensory loss can be trained to overcome the loss or to compensate for it. It has not yet been demonstrated that it is possible to do so. The attempt should be made to teach the patients object*

recognition and differences in shape, texture, size and functions of various things" (Kenney, 1963, pg. 194).

A few years later the same author published a paper that summarised the field to date, including brief references to work that pre-dates that of Tizard and Crothers (1952). Citing '*Recent Advances in Cerebral Palsy*', edited by R.S. Illingworth and published in 1958, Kenny briefly noted the work of 'Phelps' from 1942 (no further details are provided), who described sensory deficits in a group of children with hemiplegia (Kenney, 1966, pg. 47). Kenney also highlighted the work of G.E. Woods from 1958, who identified astereognosis in children with visual field defects but no defects for children with athetoid CP. Kenney reported that Woods "*ascribed sensory defects to damage of the parietal lobe*" (pg. 47). Additionally, Kenney's paper highlighted the work of Critchley (1949) and the importance of "*the phenomenon of tactile inattention*" (pg. 45) for people with cerebral lesions, particularly of the parietal lobe, which became a key design consideration for the intervention for the current study (Chapter 5, section 5.2.7).

The study conducted by Wigfield (1966) **[8]** assessed the oldest cohort to date (age range: 16 to 31 years, average age = 20.5 years), due to the fact that this study was conducted within a Vocational Training Centre for young adults and the assessments related to employability into areas such as light engineering, woodworking, commercial practice, and domestic science. Not strictly a paediatric study, the cohort of 64 mostly male participants included 14 children (22%) aged 18 years of age or younger (16(1), 17(3), and 18(10)).

Wigfield reported that 19 of the 23 hemiplegic participants (83%) had impaired stereognosis, which was an overrepresentation for astereognosis considering this modality was impaired in only 25 (39%) of participants overall. Sense of touch (30%) and weight discrimination (22%) were the next most frequent modalities affected. Similar to Monfraix *et al.* (1961), Wigfield's results indicated that sensory disturbances may appear more frequently in cases of hemiplegia compared to diplegia, and also confirmed the presence of deficits on the unaffected side within the hemiplegic group. Given the older age of the cohort and the presence of sensory impairments recorded, the author concluded that childhood impairments most likely persisted throughout life.

One of the largest studies to date to assess children with CP for sensory disturbances across a number of modalities was conducted by Wilson *et al.*, published over three papers and seven years (Wilson & Wilson, 1967a, 1967b; Breakey, Wilson, & Wilson, 1974) **[9-11]**. The study involved 120 children with congenital CP (comprised of two equal groups, as annotated in Table 2) and a control group of 60 non-neurologically impaired children. The first study (Wilson & Wilson, 1967a) reported the results of light pressure threshold tests (conducted on the tip of the index finger and on the palm of the preferred, not necessarily dominant, hand) and TPD. The study revealed that 59 children (49%) had one or more sensory defects – of these 39 had one defect, 17 had two defects, and three had sensory losses in each of the three functions tested. The CP group differed from the control group on two of the three threshold measures, but not on pressure applied to the tip of the index finger. Furthermore, no significant differences were found between the two CP groups' means on any of the three threshold measures, across all ages. In contrast, the control group showed significant differences in thresholds as a function of age – the younger group was significantly more sensitive than the older group on palm-pressure measures and less sensitive on TPD. Finger-tip-pressure results showed the same trend, with younger children being more sensitive than older ones, but the results were not significant.

Forty one children (34%) were identified as having a TPD deficit, and there were 41 incidences of pressure defects in either the finger (n=16) and/or the palm (n=25). Concomitance calculations revealed that a finger-tip-pressure defect was more likely to occur in the presence of a palm defect than in its absence, with a significant concurrence also found between TPD and a palm-pressure defect. No correlation was found between intelligence and the severity of involvement on somatosensory functioning, however correlations between severity scores and sensory scores indicated a significant relationship between TPD and severity in the athetoid group alone. The authors concluded that while a sizable number of children with CP in their study did show a sensory defect, a larger number had sensory thresholds fall within normal limits as defined by their control group. Following the analysis of their TPD and light touch testing, the authors concluded that correlation calculations indicated an apparent dissociation between pressure and TPD thresholds for the CP group,

suggesting that these two tests are tapping substantially independent functions (Wilson & Wilson, 1967a).

Their second study assessed stereognosis using 15 objects, and was assessed for both size and form (Wilson & Wilson, 1967b). A significant effect due to diagnosis was found for both size and form error, but there was no significant difference in performance between the two CP groups, despite the difference in brain lesion location between the sub-groups (spasticity vs. athetosis). Age was not a significant variable for the CP groups, however, there was a significant difference in performance between the younger and older control groups with respect to size error, suggesting an underlying developmental trend in relation to haptic size discrimination as evidenced by the demonstration of an age affect in the control data. Similar to Monfraix *et al.* (1961), the authors investigated "mental age" correlations as opposed to chronological age correlations for the CP groups. The performance of the athetoid group did not vary with mental age, whereas the spastic group showed improved stereognosis performance with increasing mental age. That is, the spastic groups showed a decrease in size error as a function of increasing mental age, with their ultimate performance being comparable to that of the athetoid group across their entire mental age range. The authors noted that the "developmental lag" for the athetoid group was of a different order to that of the spastic group, potentially supporting the position that children with athetosis are less impaired in sensory and perceptual functions than children with spasticity (pg. 67), which was also noted by Kenney (1963).

Comparing the two CP sub-groups, a higher proportion of children with spasticity (18%) showed size discrimination defects than those with athetosis (11%), but both sub-groups performed similarly with respect to haptic form discrimination (31% and 30%, respectively) and an equal amount (8%) showed a deficit in both form and size discrimination. A size defect was significantly related to the non-occurrence of a palm defect and form discrimination was significantly correlated with TPD for both CP groups.

The third and final study in the series was an assessment and evaluation of visual perception, including a test of limb localisation, intra- and cross-modality pattern discrimination, concept formation and a picture-word association (referred to as an

'*Ammons Full Range Picture Vocabulary Test*') (Breakey *et al.*, 1974). The limb localisation task was performed using a movable shallow tray and calibrated in degrees of arc. With vision excluded, each child was asked to return their arm to a position from which it had been moved by the experimenter, with the error of return measured in degrees of arc. The authors reported that 23 of the 120 CP participants found the task physically impossible, with 19 children (83%) belonging to the athetoid group. Additionally, the group with spasticity recorded more visual defects (71%) than the group with athetosis (48%) when refractive errors were taken into account.

Lesný highlighted the importance of functional tactile and kinaesthetic afferent signals with respect to complex movement when laying the foundations for his study of TPD of children with CP (Lesný, 1971) **[12]**, the only researcher to assess a single modality. The study of 143 children, mostly with hemiplegia and diplegia (representing 70% of the cohort) and a control group of 30 healthy children, investigated the discriminative sensibility of TPD using Weber's scissors. Lesny stated that TPD was a function of the parietal cortex and that the test had an established and recognised norm, referencing the early work of Vierordt from 1887, who conducted TPD testing at eight sites on the upper limb in girls aged six and eight years, and in adult women. Lesný reported that the older control group (aged 11-15 years, n=16) recorded significantly better TPD at the tip of the right third finger but significantly worse TPD at the middle of the left palm and the acromion of the right hand, compared to the younger control group (aged 6-10 years, n=14). Statistical analysis revealed that the greatest disturbances of TPD for the CP cohort were found on the most distal parts of the upper limbs, in particular, the tip and centre of the third finger. Lesný also identified that in CP hemisyndromes, disturbances of TPD were also present on the unaffected side, similar to Monfraix *et al.* (1961) and Wigfield (1966).

More than two decades later, Lesný and four other colleagues published the results of a study on TPD disturbance in 220 children with CP (Lesný *et al.*, 1993) **[15]**, aged between seven and 14 years. The same eight sites for TPD testing as described in Lesný's 1971 paper were used, and it appears that the group were compared to the same control group as the 1971 cohort. The authors reported a decrease in TPD sensitivty in the CP cohort compared to the control group for most

forms of CP (although TPD was not measured in children with hypotonia), and that the decrease was greater among children with spasticity compared to athetosis, as Monfraix *et al.* (1961) and Kenney (1963) had reported previously. In agreement with the earlier 1971 study, Lesný *et al.* (1993) reported decreased TPD on the unaffected side for children with hemiplegia, and that the decreases were greater at the distal points (tip and middle of the third finger) compared to proximal points just as the carpal joint and centre of forearm. The decrease in TPD was less amongst children with quadriplegia than children with diplegia, leading the authors to conclude that a sensory disorder is independent of a motor disorder. The most marked changes of TPD were found in children with diplegia and children with hemiplegia on their involved side.

Uvebrant (1988) **[13]** conducted a wide ranging retrospective, population-based study of a large cohort of children with hemiplegic CP, where sensory appreciation was one aspect. For this reason the cohort size varies between tests, depending on who was able to be tested during follow-up. The sensory battery included stereognosis, grapheasthesia, TPD, pain, position sense/proprioception, temperature, light touch, and vibration. The criteria for, and classification of, stereognosis and grapheasthesia was considered ‘good’ if the child scored 4-5, ‘moderately impaired’ if they scored 2-3, and ‘poor’ if they scored 0-1, when either five objects were presented or five figures were traced on the hand. TPD of the index finger was classified as being ‘good’ when a distance of 0-4mm was recognised as two points, ‘moderately impaired’ for distances of 5-7mm, and ‘poor’ for distances greater than 8mm.

Uvebrant (1988) reported that sensation related to pain, temperature, vibration and position sense were all largely preserved in the majority of cases. No child recorded a complete loss of any of these sensations, with the minimum percentage of children who recorded ‘normal’ sensation being 84% (for pain) and the maximum being 93% (for vibration). The most impaired sensory modailities were grapheasthesia, followed by TPD, followed by stereognosis, with the pre-term congenital CP group (mainly sub-cortical brain injury) being the least affected and the post-natal CP group (mainly cortical brain injury) being the most affected, particularly for TPD and grapheasthesia. Children with CP who were born at term always recorded sensory

deficits that fell between the aforementioned groups, for every test and for every classification within each test.

Complete astereognosis (identifying zero objects) was present in 19% of congential CP cases and 25% of post-natal CP cases, for an overall frequency of 20% for the whole cohort. Undergrowth of the affected upper limb was common, with 79% of children having a limb that was greater than three millimetres shorter than their unaffected upper limb, and 88% of children having a lower limb that was more than five millimetres shorter than their unaffected leg. Uvebrant (1988) reported that a normal CT scan was a common finding for children with a mild disability, whereas those with moderate disabilities showed unilateral ventricular enlargement. However, a CT finding of cortical/subcortical cavities was significantly correlated with severe impairments relating to hand motor function, stereognosis, gait, mental retardation and epilepsy (pgs. 75-76).

Bolanos *et al.* (1989) **[14]** conducted a comparison study of TPD and stereognosis involving 51 children with CP and 170 controls between the ages of six and 20 years. The Weber TPD test was chosen as it was identified as the most reliable test of altered sensibility of the hand when related to function (Moberg, 1962), and a TPD width of 5mm was used following personal communication with Erik Moberg. Additionally, a minimum age was set for the study to address the issue of "disordered sensation" in normal children under the age of seven years (pg. 374). The authors acknowledged that while stereognosis was recognised as the most common sensory deficit in the hands of children with CP, particularly in hemiplegia, it was also a test of the child's cognition and verbal abilities in recognising the various shapes used during the test. Cut off levels were determined for both tests, being two or more incorrect answers for TPD and one or more incorrect answer for stereognosis.

Thirty two (63%) children recorded a TPD deficit compared to 20 (39%) children for the stereognosis test, which is counter to almost all previous studies with the exception of Uvebrant (1988), where stereognosis is typically the most impaired modality. TPD had a higher sensitivity than stereognosis for detecting tactile sensation in CP, but it had a slightly poorer specificity. In terms of the location of sensory deficits for TPD, the middle phalanges was where most errors occurred, but

the CP group had a significantly higher incidence of defects over the distal phalanges, as reported earlier (Lesný, 1971; Lesný *et al.*, 1993). The authors reported no evidence of a significant sex difference in sensitivity for either test, and no evidence of an age difference in sensitivity for the TPD test. However, the authors noted there was some suggestion of less sensitivity among children younger than 13 years of age compared to children older than 13 for the stereognosis test.

A hospital-based study by Van Heest, House, and Putnam (1993) **[16]** investigated sensibility in the hands of 40 children with only congenital spastic hemiplegia (mostly right side involved), including an assessment of the relative size of both upper limbs at four key areas (forearm length, forearm-hand length, arm circumference and forearm circumference). The sensory assessment included stereognosis (using 12 objects), TPD (using 6mm spacing, applied at the finger tips) and proprioception (movement of the fingers up and down) on both sides of the body. Thirty nine children (97%) recorded a stereognosis deficit, justified by the authors as being 'intact' if all 12 objects were correctly identified, 'mildly deficient' if eight to 11 objects were correctly identified, 'moderately deficient' if five to seven objects were correctly identified, or 'severely deficient' if less than four objects were correctly identified. Similarly, TPD was reported as being deficient in 90% of children, with TPD being assessed as being 'intact' if all five trials were correctly identified, 'impaired' if one to four trials were correctly identified, or 'absent' if none of the five trials were correctly identified. Proprioception was impaired in 46% of children, with the same criteria for TPD being applied for five movements of the finger either up or down (also graded as being 'intact', 'impaired', or 'absent'). Statistical analysis identified that the severity of the stereognosis deficit was directly correlated with an impairment of TPD, but not with an impairment of proprioception.

In terms of upper limb size differences, all 40 children recorded a size discrepancy in their affected upper limb in at least two parameters, compared with their unaffected upper limb, with the spastic limb significantly smaller in all parameters. The authors stated that a severe size discrepancy between limbs for children with spastic hemiplegia is a clinical clue that severe sensory impairments exist. Children with severe stereognosis deficits had a significantly smaller limb in all four limb measures compared to children with mild or moderate stereognosis deficits, and there was a

trend toward increased TPD and proprioception deficits in children with increasing size discrepancy.

Van Heest *et al.* (1993)'s study reported the highest incidence of somatosensory impairment for all studies to date (97%), with the high rate potentially due to a more stringent test of stereognosis and the fact the cohort was only children with spastic hemiplegia, and not a mixed cohort. Being a hospital-based study, it's possible that the cohort included children with more involved upper limbs, such as Manual Ability Classification System (MACS) Levels III to V compared to MACS Levels I to II, who hence performed worse. On the test of stereognosis, the authors noted that the average score for the dominant limb was 11.8 out of 12 (pg. 280), meaning the children understood the testing procedure but couldn't identify the item with their ND hand. The authors also noted that their four year study only involved assessing children at one time interval (like most studies), and that the effects of growth on size discrepancy and sensory status were not known. The study by Van Heest *et al.* (1993) also confirmed the reporting of the earliest studies (Tizard *et al.*, 1954; Hohman *et al.*, 1958; Tachdjian & Minear, 1958) in terms of the frequency of different modalities affected, with stereognosis most affected (39 of 40 children), followed by TPD (36/40), and then proprioception (18/40). The authors highlighted the likelihood that surgical outcomes may be limited to "*improvements in gross motor function, appearance or hygiene*" (pg. 281) and not better overall function when the sensory status of the hand is poor.

A study of 55 children with mixed CP types was conducted by Yekutiel *et al.* (1994) **[17]**, specifically assessing stereognosis, TPD and location of touch. This study used a control group of 15 children, however it wasn't a typically developing group. The 15 children had had poliomyelitis during infancy, with severe lower limb involvement, and their hands were assessed to provide 'normal data'. The authors highlighted the importance of identifying sensory deficits amongst children with CP, not just in terms of establishing realistic functional goals with respect to surgical decisions, as advocated by many earlier studies, but due to the possibility that sensory impairments "*may be partly remediable by sensory training*" (pg. 620), which Kenney (1963) had posited more than 30 years earlier.

The authors reported that the cohort could be reliably tested for stereognosis, particularly when familiar objects were used, but this was not the case for TPD (using the Disk-criminator and distances of 5 and 10mm) with only 36 children able to be reliably tested. The test for location of touch was abandoned because it wasn't understood by the children. The authors used a more lenient criteria for 'normal' stereognosis performance (that is, two incorrect responses from 20 tests was acceptable for stereognosis, and four incorrect responses from 40 tests was acceptable for TPD, based on the results of the post-polio group), and reported sensory impairments in 28 children (51%). The relative TPD impairment (24 of 36 children, 67%), mostly at 5mm, was more than double the level of stereognosis impairment (15/55, 27%), which is counter to the literature, but similar to Bolanos *et al.* (1989)'s study. This result is presumably due to the more lenient stereognosis scoring system used by the authors.

The sensory results were not correlated with age or cognitive function (termed 'mental retardation' by the authors), and the hemiplegic cohort showed the most sensory loss, confined exclusively to the affected hand. The authors stated that their research confirmed all previous work in this area, citing Tizard and Crothers (1952)'s original work, and highlighted that impaired sensation is common for this cohort, particularly amongst children with hemiplegia and less so for children with athetosis, which is a common theme within the literature. Yekutiel *et al.* (1994) acknowledged that prior sensory studies and their reported prevalence rates depend on the modalities being tested and the method of administration and application. Similar to previous research, the authors advocated an appreciation and understanding of a child's sensory status prior to training, rehabilitation and surgery, and supported exploring 'systemic sensory training' (pg. 623) to improve hand sensation following their successful earlier work with chronic post-stroke patients (Yekutiel & Guttman, 1993).

Cooper *et al.* (1995) **[18]** noted that current treatment for children with CP focused on the identified motor deficits, without regard for any underlying sensory deficits. Recognising the earlier work of Bolanos *et al.* (1989), the authors emphasised the importance of sensory testing prior to a rehabilitation program aimed at improving hand function. The objective of their study was to determine the presence and extent

of sensory deficits in school-aged children with only hemiplegia using a formal clinical sensory battery, including somatosensory evoked potentials. Only nine mostly male children were recruited to the study, making Cooper *et al.* (1995)'s study the smallest published study within the literature, but highly cited due to the clinical battery that was used. The sensory assessment included pressure sensitivity, TPD, moving two-point discrimination (MTPD), stereognosis, proprioception and directionality, and included a control group of 41 children.

Cooper *et al.* (1995) reported sensory impairments in eight (89%) children, with stereognosis again the most impaired modality, followed by proprioception. The study identified that the affected hand was significantly impaired across all five modalities, with sensory disturbances also reported for the unaffected hand for all modalities except MTPD for six of the nine children, emphasising the importance of testing sensation bilaterally. Statistical analysis revealed no significant differences between the dominant and non-dominant sides when investigating sensory function. The extent of sensory loss did not correlate with motor deficit severity (assessed via neurologic examination and via grasp patterns), whereas somatosensory evoked potential abnormalities did correlate with motor function assessment. Further, abnormal somatosensory evoked potentials were found to be a predictor of severe sensory deficits in four of five cases, but normal somatosensory evoked potentials was not a predictor of normal sensation. The authors endorsed and emphasised the importance of assessing sensory function using a standard, comprehensive clinical battery of tests, concluding that the likelihood of sensory impairment in one or more modalities on either side for children with hemiplegia is underappreciated and needs to be identified by rehabilitation specialists to maximise each child's functional potential.

Recognising the importance of sensory information when performing manual dexterity tasks, and knowing the literature related to tactile sensory deficits of the upper limbs amongst children with CP, Gordon and Duff (1999) **[19]** conducted a study to examine which clinical measures best relate to how this population applies fingertip forces during a precision grip-lift task. Tactile sensibility, pinch strength, manual dexterity and spasticity were studied, as was how each child adjusted their fingertip force to the object's texture, the degree of anticipatory control of the grip

force output, and the transition from grasping to lifting during the grip-lift task. The grip-lift task represents a complex combination of motor, sensory, and motor-control tasks.

Their study involved 15 children with hemiplegia only, compared to 15 age-matched control children who grasped and lifted an object whose surface texture varied from fine sandpaper to rayon material while their fingertip forces were recorded. The force coordination was then compared with tactile sensibility, grip strength, manual dexterity, and spasticity using correlational and regression analyses. Sensory assessments included TPD, pressure sensitivity, and stereognosis (based on the '*Manual Form Perception Test*' using eight shapes). Pinch strength (assessed using a dynamometer), spasticity (assessed using the Modified Ashworth Scale 0-4 scale) and manual dexterity (assessed using six subtests from the Jebsen-Taylor Hand Function Test or JTHFT) were also assessed.

The tactile assessment identified that TPD was four times larger in the CP group compared to the control group, with one subject unable to discern two points 15mm apart, which is the limit of operation for the *DiskCriminator*. Pressure sensitivity was also significantly impaired in CP group, though to a lesser extent than for TPD. The control group generally had intact stereognosis with nearly all subjects correctly identified all eight objects (range: 7-8 objects), while the CP group displayed significant impairment in comparison (range: 0-5 objects). Palmar pinch strength was significantly lower for the CP group, and the CP group were significantly slower (approximately 10 times) in the timed manual dexterity tasks of the JTHFT. A correlation analysis showed that TPD, stereognosis and manual dexterity each significantly correlated with pinch strength, and a multiple regression analysis indicated that spasticity and TPD were the strongest individual predictors of static grip force adaptation when considered separately.

Gordon and Duff (1999) stated that their results confirm the importance of intact tactile sensation and the role it plays with respect to sensory adaptation of fingertip forces and anticipatory force scaling. Furthermore, they highlighted that the strong correlation between TPD and grip-force adaptation "*indicates that fine discriminatory ability is related to the ability to differentiate the force output based on the object's texture*" (pg. 590). The authors concluded that impairments related to grasping in

children with hemiplegic CP are largely, but not exclusively, due to impaired sensory mechanisms.

A study of 25 children with mild or moderate hemiplegic CP by Krumlinde-Sundholm and Eliasson (2002) **[20]** sought to evaluate and identify the most relevant tests of tactile sensibility that can be used with children with hemiplegic CP. The aim was to know which tests were most sensitive and whether children with hemiplegic CP could adequately participate in the testing. Their work drew on the literature to date, with the aim of developing and validating a series of sensory tests that could be adopted for future studies to facilitate a means for consistent assessment. In essence, this study was aiming to achieve consensus for the field of sensibility testing for children with a neurological impairment and to apply rigour to the tests and the associated methods. Using TPD as an example, the authors cited the many different instruments, methods, distances tested, and lack of procedural details reported in the literature, as reasons why comparisons cannot be reliably drawn. An analysis of the literature reveals that the same can be said for tests of stereognosis, which, along with the test of TPD, the authors recognised as being the most common tests administered.

The authors used a control group of 19 age-matched typically developing peers and all children were examined on both sides for touch sensibility (Semmes–Weinstein Monofilaments, SWM), TPD (at 3mm and 7mm), stereognosis (six familiar objects and 10 flat plastic geometric forms), and a test of motor function (the ‘Pick up’ test, with and without vision). The authors stated that the two different tests of motor function that assessed the performance of the same hand with and without vision was indicative of the amount of somatosensory loss, due to the fact that tactile cues are required to guide the necessary motor actions, such as finding an object, grasping it, and taking it from the box. Should a child struggle with this test without vision, the authors stated this indicated the level of visual guidance the child’s motor performance relied on. Dexterity, spasticity, and bimanual task performance (for eight everyday tasks that involved using two hands) was also assessed.

Following their analysis, the authors reported that three particular tests were most useful when testing tactile sensibility: TPD, stereognosis of familiar objects only, and functional sensibility assessed through the ‘Pick up’ test. Of these tests, the authors

stated that TPD at 3mm appeared to be the most sensitive test, being able to identify minor deficits, in agreement with the work of Bolanos *et al.* (1989). Stereognosis of forms (using geometric shapes) was not recommended as a test as it not only requires the ability to manipulate the shape, but also to identify the form, and geometric forms are known to be more difficult to recognise than common objects, as reported by Stilwell and Cermak (1995). Supporting the fact that geometric forms are more difficult to recognise than common objects, more than half (58%) of the control group, who were considered to have normal sensibility, made one or two mistakes with stereognosis of forms, regardless of age. The authors concluded that geometric form recognition cannot be viewed as a test of tactile sensibility alone. The children who could not perform the 'Pick-up' test at all with vision occluded also showed low results for the stereognosis of objects and TPD. Contrary to previous research, and ignoring the stereognosis of forms test, all children with hemiplegic CP demonstrated intact sensation on their dominant side for all tests, with seven children demonstrating bilateral intact sensation. For four children with severely reduced touch sensitivity, this was a predictor of their performance overall, as those same four children also registered deficits for all others tests of sensibility. For the ND hand, tactile sensibility was significantly correlated with hand function in terms of the dexterity of hand.

In recommending the most suitable tests, Krumlinde-Sundholm and Eliasson (2002) recognised a hierarchical relationship that reflected a varying degree of involvement and input from other components that can affect the test outcome, such as motor function or vision. Their three test approach was independent of age and chosen for the following reasons: TPD, to measure subtle qualities of sensibility that do not require motor function; stereognosis of familiar objects, because it involves a practical use of sensibility combined with motor function and form perception; and functional sensibility assessed through the 'Pick-up' test (with and without vision), because it assesses the influence of both the motor and sensory components of hand function (pg. 611).

A study by Arnould, Penta, and Thonnard (2007) **[21]** assessed 101 children with mixed but mainly hemiplegic CP for both motor and sensory impairments. Motor impairments were assessed by measuring grip strength, gross manual dexterity (Box

and Blocks Test), and fine finger dexterity (Purdue Pegboard Test), and the tests of sensation included pressure detection using SWM, stereognosis as per Cooper *et al.* (1995), and proprioception as per Cooper *et al.* (1995). Additionally, manual ability was measured using the ABILHAND-Kids questionnaire. Results were compared to age and sex matched healthy children, although this particular cohort (the number of children and their sex) is not described in their paper.

The control group enabled normative results for all motor and sensory tests to be established, with the authors considering a 'significant' sensory impairment being when a raw score was lower than the fifth percentile of the distribution observed for the control group (pg. 710). Consequently, the authors considered a child to have a significant sensory impairment if they detected 166mg for tactile pressure detection (the first blue filament in a 20-piece SWM filament kit, which is referred to as 'diminished light touch'), correctly identified nine objects from 10 for stereognosis, and correctly identified seven movement directions from ten for proprioception. These cut off scores are in agreement with other published studies to date, such as Cooper *et al.* (1995) for pressure detection, Van Heest *et al.* (1993) and Cooper *et al.* (1995) for proprioception, and Bolanos *et al.* (1989) and Van Heest *et al.* (1993) for stereognosis, except that in this case the authors considered these thresholds or cut off scores as not just representing abnormal sensation, but sensation that was significantly impaired.

The authors reported bilateral sensory impairments across all CP types (tetraplegia, diplegia and hemiplegia), with the tetraplegic cohort recording more frequent impairments on the dominant side (followed by the hemiplegic group and then the diplegic group), but the hemiplegic group recorded more frequent impairments on the ND side (followed by the tetraplegic group and then the diplegic group). The most commonly affected modality on the ND side was stereognosis (38%), followed by tactile pressure (33%) and proprioception (15%). For the dominant hand, the most commonly affected modality was tactile pressure (21%), followed by stereognosis (20%) and proprioception (4%). Compared to the control group, stereognosis and tactile pressure were both significantly impaired, but proprioception was not. One potential explanation for this is that the cohort with diplegia, who performed best on all sensory tests, recorded intact sensation with respect to proprioception on both

upper limbs. The authors observed that manual ability was significantly but only moderately correlated with motor impairments and stereognosis for both hands, while no significant relationship was found with respect to pressure detection and proprioception. Motor impairments were markedly more prevalent than sensory impairments for all CP types, with fine finger dexterity being the most prevalent motor impairment on both hands.

Wingert *et al.* conducted a study published over two papers that investigated tactile sensory abilities such as roughness, object discrimination (Wingert *et al.*, 2008) **[22]**, joint-position sense and kinesthesia (Wingert *et al.*, 2009) **[23]**, with a cohort of 38 children with CP (diplegia (21) and hemiplegia (17)). The mostly female cohort was assessed for object recognition (five common objects, four embossed geometric shapes, and eight embossed capital letters) and roughness (via paired horizontal gratings of various groove widths), and joint position sense and kinesthesia in the transverse plane, with and without vision, for the arm and foot. A control group of appropriately age-matched healthy children was used as a comparison.

The authors treated their CP cohort as two different groups, recognising the unique brain lesions that are associated with different CP sub-types. They reported that both cohorts had significantly higher thresholds for groove width difference for both hands compared to the control group. For children with diplegia, roughness discrimination was the only modality that differed between hands, whereas for children with hemiplegia, significant between hand differences were recorded for all assessed tasks. In agreement with previous research, sensory deficiencies were recorded bilaterally for both CP groups, with the ND hand recording greater sensory impairments, as would be expected. Owing to the degree of bilateral upper limb tactile sensory impairments that were recorded, Wingert *et al.* (2008) suggested that a child's tactile guidance was likely affected, providing diminished sensory cues when touching objects and contributing to their "awkward dexterity" (pg. 837).

The authors extended their work by looking at joint-position sense and kinesthesia, assessing both the upper and lower limbs using a custom device that rotated in the transverse plane (Wingert *et al.*, 2009). Similar to the work conducted previously by Breakey *et al.* (1974) and Krumlinde-Sundholm and Eliasson (2002), the authors conducted their tests with and without vision to assess how each child used their

somatosensory inputs to complete the necessary tasks. The movements required for the upper limb were forearm pronation and supination, and for the lower limb it was hip internal and external rotation.

Wingert *et al.* (2009) reported that when vision was allowed, no group differences were detected for either test. However, when vision was occluded, significant joint-position sense deficits were detected for all children with CP for both lower limbs and the ND upper limb, but not the dominant upper limb. This was despite the fact that all children with CP had relatively mild motor involvements, being classified as either Gross Motor Function Classification System (GMFCS) Level I or II and MACS Level I or II. When analysed, the joint-position sense error was biased towards the direction of internal rotation. Deficits related to kinesthesia were also detected on the ND upper limbs for both CP groups, including bilaterally for the lower limbs for children with hemiplegia. Age was not found to have an effect when the cohort was stratified based on being younger or older than 13 years old. The authors noted that the difference in performance accuracy between tasks highlighted the role that visual compensation played for children with pervasive deficits in proprioception, and emphasised the importance of optimising vision for people with CP. This extended to how vision is used when learning and practicing movements.

A similar, earlier study that does not appear in Table 2 because it only assessed kinesthesia and no other tactile assessments, used a standard kinesthesiometer with 24 children with CP (either spastic quadriplegia or athetoid quadriplegia) aged eight to fifteen years, and 12 age matched controls (Opila-Lehman, Short, & Trombly, 1985). The study, which was the first to ‘gamify’ the assessment (that is, the researchers turned the exercise into a game to increase the child’s interest in the task), identified that children with CP performed significantly worse than their typically developing peers when their upper limb was passively moved to align with set targets in terms of absolute error. Within the CP cohort, children with spastic CP performed significantly worse than children with athetoid CP, and all three groups tended to underestimate the target they were attempting to align with.

Holmström *et al.* (2010) **[24]** investigated the relationship between hand function, brain lesions, and corticoprojections in children with unilateral CP, where TPD, mirror movements, the Box and Blocks (B&B) test and the Assisting Hand Assessment

(AHA) were used to assess hand function. All children were classified as MACS Level I or II and GMFCS Level I. The study used magnetic resonance imaging (MRI) to assess the type, location and extent of the brain lesion, and single-pulse transcranial magnetic stimulation (TMS) to provide information on the organisation of corticomotor projections.

The study identified that the combination of type, location, and extent of the lesion was a stronger predictor of hand function than just the lesion type alone. It concluded that the most favourable hand function was seen in children who had white-matter damage of immaturity with white-matter loss and contralateral projections. Children with ipsilateral projections to the hemiplegic hand had the most impaired hand function, however some children in this motor projection group had good bimanual ability, indicating that an ipsilateral pattern can be associated with a good "*assisting hand*" (pg. 150).

All children with poor TPD performance were in the group with poorer hand function, whereas children with normal tactile discrimination were in the group with more favourable hand function. In general, hand function was better in children with mixed projection than in those with ipsilateral projection. The authors noted that impaired sensory functions could be a contributing factor to poorer hand function, due to their effect on sensorimotor integration. Additionally, the authors proposed that impaired sensory function may be caused by a dissociation of sensory input and motor output to different hemispheres (pg. 150), as suggested by others, including Guzzetta *et al.* (2007). A surprising finding from the study was the performance of the cohort's dominant hand when assessed by the Box and Blocks test. When compared to typically developing children of a similar age, most of the children (15 or 88%) recorded a lower test score than would be expected for their age, which was not related to bilateral brain lesions when the MRI scans were visually examined.

A study of 81 children aged five to 15 years old with unilateral CP by Klingels, Demeyere*, et al.* (2012) **[25]** reported that only 69 children (85%) could satisfactorily complete the tests of sensation, with a "*lack of understanding*" being the reason provided (pg. 478). This is contrary to most other studies but in agreement with Yekutiel *et al.* (1994)'s reporting of the TPD test. Klingels *et al.*'s cohort comprised children with either a congenital (n=69) or acquired (n=12) brain lesion, and were

classified as either MACS Level I (n=29), II (n=36) or III (16). It's unclear from the article if both hands were assessed, but descriptors and criteria for 'intact', 'impaired' or 'absent' sensation were provided. Passive range of motion (PROM), muscle tone, muscle strength, grip strength, the Melbourne Assessment, the AHA, and the ABILHAND-Kids questionnaire were also tested or administered.

Tests of sensation included exteroception or light touch (on the thumb, index finger and palm), proprioception, TPD using an Aesthesiometer®, and stereognosis using 6 objects. Stereognosis (65%) and TPD (58%) were the modalities most commonly impaired, and the only two modalities that recorded an 'absent' result, with the other two modalities only recording 'impaired' or 'intact' results. The authors recognised this reflected a hierarchical functional anatomical organisation within the CNS, with touch and movement sense being basic sensory functions, while TPD and stereognosis represent more cortical (perceptual) functions. Children with a higher MACS level had significantly less impairments compared to children with a lower MACS level, but there were no significant differences between the two CP groups (congenital & acquired) for the sensory tests. However, children with congenital lesions had significantly less motor impairments compared to children with acquired lesions.

Together with wrist strength, stereognosis and proprioception were two of the three important variables that predicted the Melbourne Assessment when analysed using multiple regression modelling. Likewise, wrist strength and stereognosis were the two significant predictors for the ABILHAND-Kids questionnaire. With respect to the questionnaire, children aged older than 10 years performed significantly better than children aged less than 10 years, but no significant differences were found between the two CP groups. The authors reported that internal rotation was more frequently limited than external rotation, which supports Wingert *et al.* (2009)'s findings with respect to joint-position sense error bias mentioned earlier.

The same authors also reported on a one-year follow-up study with the same cohort, however, did not assess for tactile deficits 12 months later (Klingels, Feys*, et al.*, 2012). The authors reported an age related improvement in grip strength for both hands, and that motor impairments, movement quality and ND hand involvement in bimanual tasks did not spontaneously improve over one year. However, the authors

noted that children with a high manual ability level (a high MACS Level) and children with congenital lesions may learn adaptive movement strategies over time resulting in increased fine motor skills, evidenced by the significant improvement in performance for the JTHFT.

The first study to assess an Australian cohort of children with only unilateral CP was conducted by Auld *et al.* (2012b) **[26]**. Fifty two child, classified as being MACS Levels I (36) and II (16) and GMFCS Level I (34) and II (18), were assessed using one test of tactile registration (SWM, using the full 20-filament kit), five tests of spatial-tactile perception (single-point localization, double simultaneous, both static & moving TPD, and stereognosis with 9 common objects), and one test of texture perception (using a device called the AsTex®). The authors reported tactile sensory impairments in 40 (77%) children, with stereognosis being the most common modality impaired (63%), followed by double simultaneous (58%). Confirming the results of previous studies, the authors reported that more than half the cohort (54%) recorded tactile sensory deficits on their dominant side. Forty per cent of the cohort recorded deficits in both tactile registration and perception, with the dominant hand of the CP group performing statistically significantly poorer than either hand of the control group. The authors noted that while there were no significant performance differences between children with left or right hemiplegia, there was a greater proportion of children with left hemiplegia who recorded a combined tactile registration and perception deficit compared to children with right hemiplegia (52% vs. 31%). Additionally, more children with right hemiplegia registered a deficit in the less-severe test of tactile perception compared to children with left hemiplegia (45% vs. 26%).

Given the different modalities tested (tactile registration, spatial tactile perception, and texture perception) and the correlation values reported between the variables (the highest was 0.76), Auld *et al.* (2012b) advocated for a battery of tests across different modalities as previously reported by Cooper *et al.* (1995) and Krumlinde-Sundholm and Eliasson (2002). A ‘mini’ battery including one test of registration (SWM), one test of unilateral spatial perception (single point localisation) and one test of bilateral spatial perception (double simultaneous) was proffered.

Kurtaran *et al.* (2015) **[27]** investigated both hand sensation and function in 36 children with mixed CP and 18 healthy controls, assessing stereognosis, light touch, pain, TPD, graphesthesia, as well as grasp force and a test involving the lifting of two different masses. The authors reported significant differences between all measures of hand function related to lifting objects and grasp force for the ND hand compared to the control group, with significant differences for the dominant hand reported for the heavier mass (400g) and hand grasp force. The ND hand performed the test of stereognosis significantly worse compared to the control group, and there was a significant negative correlation between hand functional status and stereognosis for the ND hand. No significant differences involving the dominant hand were reported for stereognosis. Light touch was impaired in only 6% of children, substantially less than impairments recorded for graphesthesia (53%). Kurtaran *et al.* (2015) also reported that TPD was non-testable in a surprising 67% of children (n=24), but similar to both Yekutiel *et al.* (1994) and Klingels, Demeyere*, et al.* (2012). The authors also reported that children with higher functional activity levels and who were ambulatory also had higher levels of grasp force.

The work by Lesný (1971) and Lesný *et al.* (1993) were the only two sensory studies that assessed a single modality, that of TPD. Two other studies have assessed a single sensory modality, stereognosis, for two different purposes. Kinnucan, Van Heest, and Tomhave (2010) investigated motor function (using the JTHFT) and stereognosis in children with hemiplegia and triplegia, identifying that stereognosis impairment was significantly correlated with an impairment in motor function. This study, like others, reported mild impairments for stereognosis on the unaffected side. The study by Petersen *et al.* (2016) investigating the effect of treatment on stereognosis, discussed in more depth later, also reported mild and moderate impairments in stereognosis in the unaffected limb of children with hemiplegia.

2.1.1 Literature Review Summary

Summarising the literature to date, the aforementioned studies highlight that somatosensory deficits are common, with overall cohort prevalence rates ranging from 42% (Tachdjian & Minear, 1958) to 97% (Cooper *et al.*, 1995). Stereognosis is the modality most frequently impaired, with TPD and proprioception the next most

frequently affected modalities. With respect to testing, stereognosis of common objects has greater validity compared to stereognosis of forms, which is not recommended. A number of studies reported sensory deficits on the dominant or unaffected limb for children with hemiplegic CP (Monfraix *et al.*, 1961; Wigfield, 1966; Lesný, 1971; Lesný *et al.*, 1993; Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002; Arnould *et al.*, 2007; Wingert *et al.*, 2008; Auld *et al.*, 2012b), highlighting the importance of evaluating hand function bilaterally for children with hemiplegia, as advocated by Cooper *et al.* (1995). Multiple authors (Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002; Auld *et al.*, 2012b) have recommended and endorsed a clinical 'battery' that tests multiple sensory modalities.

Themes through the literature include children with acquired CP having more impairments that may also be more severe than children with congenital CP; children with hemiplegia tending to have more impairments on their ND side compared to children with diplegia; children with athetosis having fewer impairments compared to children with other forms of CP, particularly spastic CP (which was also noted by Majnemer *et al.* (2008), pg. 140); that neither age or sex influences impaired sensation (Majnemer *et al.*, 2008), and that children with left hemiplegia (right side brain injury) are more involved than children with right hemiplegia. This last point with respect to the side of involvement was also reported by Brown *et al.* (1987). Multiple authors have also reported being unable to administer a given sensory test to their whole cohort, (Breakey *et al.*, 1974; Yekutiel *et al.*, 1994; Klingels, Demeyere*, et al.*, 2012; Kurtaran *et al.*, 2015) because the child simply could not complete the task or due to a lack of understanding of the test. Additionally, a strong theme from multiple studies was to adopt a cautious approach to upper limb surgery without knowing or fully appreciating the extent of sensory loss in advance, which is explored in more detail in section 2.1.3.

A thorough exploration and analysis of the somatosensory literature for children with CP, from initial scepticism to acceptance of the role and prevalence of sensory deficits, afforded an appreciation of the progression of ideas and theories that emerged over the years. As the value and significant impact of sensation on function gained momentum, calls to re-train sensation to alleviate existing deficits as part of a formal rehabilitation program appear to remain unheeded.

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Table 2 – Summary and analysis of all somatosensory assessment studies for the upper limb of children with CP, 1952 – 2015

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
1. Tizard and Crothers (1952), England/USA	44; NR; NR	NR; NR	Vision and 'other sensory' (no further information provided)	N	Vision: 19/44 Other sensory: 22/44 Vision + sensory: 10/44	Assessed growth of affected limb, reporting inadequate growth in 29/44. Advocated conducting a sensory study to determine and quantify sensory disorders.
2. Tizard *et al.* (1954), England/USA	106; NR; NR	Hemiplegia, congenital and acquired; NR	Touch, pain, temperature, position sense, passive motion, vibration, location sense, sharp/dull discrimination, TPD, stereognosis, and texture recognition	N	Overall: 57/106 (54%) *Major modalities impaired* – Stereognosis: 44/106 TPD: 32/106 Position or passive motion sense: 22/106	*Acquired* hemiplegia and *congenital* hemiplegia groups differed in types of sensation affected, with impairment slightly more frequent in the acquired group. Impaired sensation sometimes major reason for disuse of the arm; severity of sensory involvement not correlated with the severity of motor disability (except where there was no motor residue). All children with skeletal under-growth (n=16) had impaired sensation and visual fields. More disturbances found in children with spasticity compared to athetosis.
3. Hohman *et al.* (1958), USA	47; NR; 6 – 16 yrs	Infantile hemiplegia (23), quadriplegia (15), unilateral athetosis (5), triplegia (2), paraplegia (2); NR	Form discrimination (thick/thin wooden blocks and thin leather geometric designs), roughness, sharp/dull, light touch, wet/dry, hot/cold, TPD (at 0.5, 0.75 & 1.5 inches, or 12.7, 19.1 & 38.1mm), measurement of length, position sense, weighing perception, visual field defects, localisation, and speed of response	N	Overall: 34/47 (72%) *Major modalities impaired* – Form discrimination: 28/47 TPD: 26/47 Position sense: 14/47	All subjects with impaired position sense also had impaired TPD and form, except one. Impairment frequency per sub-group – 18/23 (hemi), 12/15 (quad), 2/5 unilateral athetosis, 1/2 tri- and para-plegia.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
4. Tachdjian and Minear (1958), USA	96; 40 females; 6 – 19 yrs	Spastic hemiplegia (64), spastic quadriplegia (24), athetoid quadriplegia (7), athetoid hemiplegia (1); NR	Stereognosis (button, key, safety pin, marbles, pen, wooden shapes: blocks, triangles, circles, ovals), position sense, TPD, graphesthesia, weighing perception, vibration sense, texture (rough & smooth), wet/dry, localisation of tactile stimuli, hot/cold, sharp/dull, light touch, length measurement + muscle tests of the upper extremities (no details provided), length and girth measurements of the upper extremities, skin temp measurements, and an assessment of the functional use of the involved hand	N	Overall: 40/96 (42%) *Major modalities impaired* – Stereognosis: 40/96 TPD: 31/96 Position sense: 16/96	All subjects with impaired stereognosis, TPD, and position sense *also* had impaired graphesthesia, weighing perception, localisation of stimuli, sharp/dull discrimination, temp., and length measurement. High degree of correlation b/n hands with a sensory impairment and hands rated functionally as being impaired (39/40). Inverse ratio b/n extent of sensory loss and functional use of limb. Normal sensation reported on ND side; temp differential of 0.56-1.11^{o}C b/n limbs, with involved limb cooler for only 5 subjects.
5. Jones (1960), USA	54; NR; Under 6 yrs (14), 6 – 12 yrs (13), over 16 yrs (27)	Most had spastic hemiplegia, few with quadriplegia (athetoid or mixed) – details not provided; NR	*Exteroceptive:* Light touch, sharp/dull, pain, hot/cold, wet/dry; *Proprioceptive:* passive movement (position), pressure, vibration; *cortico-sensory:* rough/smooth, texture, weight/length, size (thickness), shape (2 dims), form (3 dims, object discrimination), & tickle/scrape	N	Overall: 39/54 (74%)	Most common modalities affected were stereognosis (form discrimination), TPD, and position/passive movement in children aged over 6 yrs. Some sensory modalities were age related, such as light touch being affected in 75% of children < 6 yrs, 60% in children aged 6-12 yrs., and only 26% for adults.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
6. Monfraix *et al.* (1961), France	92; 60 male; 3 – 15 yrs	Hemiplegia (22), 'bilateral syndromes' (52), ataxia (9); impairment confined to lower limbs (9)/ NR	5 common objects (cube, marble, pencil, cotton reel, box), 12 geometrical shapes (round, square, triangle, lozenge, hexagon, octagon, oval, semi-circle, star, cross, trapezium, rectangle)	Y, 218 (2.5 to 8.5 yrs)	Overall: 47/92 (51%) Hemiplegia: 14/22 (64%) (7 moderate, 7 severe) Bilateral: 27/52 (52%) (18 moderate, 9 severe) Ataxia: 4/9 (44%) (all moderate) Lower limbs: 2/9 (22%) (all moderate)	Gnosic disturbances appear bilaterally when motor damage is unilateral. Severe agnosia appeared more common when motor damage was right-sided. Disturbances more frequent and more severe for cases involving spasticity or rigidity compared to athetosis. Disturbances also present when motor disorders appeared clearly confined to lower limbs.
7. Kenney (1963), USA	19; NR; Over 5 yrs	NR; NR	Stereognosis (rubber ball, wooden cube, metal key, metal knife, fork, spoon, crayon, cotton, pen), TPD (compass), position sense, sharp/dull, hot/cold, size of coins, & graphesthesia	Y, 27 (5 – 14 yrs)	Overall: 14/19 (73%) *Major modalities impaired* – Size of coins: 14/18 TPD: 6/14 Graphesthesia: 6/15	Very few disturbances were identified in children with athetosis. Noted that hand surgery may not improve function due to lack of sensory stimulus to motor activity. Questioned if sensory deficiencies could be overcome by training.
8. Wigfield (1966), England	64; 44 males; 16-31 yrs (ave. = 20.5 yrs)	Right hemi (13), left hemi (8), double hemi (2), paraplegia (17), monoplegia (3), quadriplegia (4), athetosis (12), cerebellar ataxia (2), flaccid CP (1), educationally subnormal (2); NR	Test of visual field, touch (sharp/blunt points), TPD (divider points to dorsum, not palm), stereognosis (empty match box, cotton reel, chalk, 3-penny piece, pack of sweets), graphesthesia (on the dorsum, not palm, using letters: A, B, F, C, Q, R, W, M and X), weight discrimination (wooden blocks), drawing a house, spatial relationships	N	For hemiplegic subjects: Astereognosis: 19/23 (83%) Sense of touch: 7/23 (30%) Weight discrimination: 5/23 (22%)	Assessments were made with respect to employability (light engineering, woodworking, commercial practice, domestic science). Impairments recorded on dom side for 4 of the 23 hemi subjects (stereognosis, touch, weight discrimination). Only 1 subject from whole cohort (n=64) recorded 9/9 for graphesthesia. Few deficits identified in athetoid group. Given age of cohort and the presence of sensory impairments, author concluded that childhood impairments likely persisted throughout life.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
9. Wilson and Wilson (1967a), USA; and **10.** Wilson and Wilson (1967b), USA; and **11.** Breakey *et al.* (1974), USA	120; 60 female; 7 – 21 yrs	Spastic hemi- and quadriplegia (60) and quadriplegic athetosis (60); NR	Light touch or pressure (using a modified Von Frey aesthesiometer on the index finger + palm) and TPD (using modified Vernier calipers)(1967a); stereognosis (15 objects, tested for form and size: 4 basic geometric shapes in two different sizes, and 7 unique shapes) (1967b); and visual perception, including a test of limb localisation, intra- and cross-modality pattern discrimination, concept formation and a picture-word association (1974)	Y, 60	Stereognosis: 55/120 (form: 37/120, size: 18/120) TPD: 41/120 Light touch: 41/120 (finger (n=16), palm (n=25)) Non-refractive visual defects: 71% of spastic group and 48% of athetoid group	Light touch & TPD: no sig. diff. b/n 2 CP groups, including for age – but sig. diff. for control group as a function of age for pressure-palm & TPD. Pressure-finger defect more likely in presence of palm defect; sig. concurrence b/n TPD & palm defect. Sig. relationship b/n TPD & impairment severity for athetoid group alone. Stereognosis results sig. diff. b/n CP groups and control, but not b/n CP groups. Size defect sig. related to non-occurrence of palm defect; form defect sig. related with TPD for both CP groups. Analysis indicated a dissociation b/n pressure & TPD thresholds for the CP groups, suggesting the tests are tapping substantially independent functions. Limb localisation task impossible for 4 children in the spastic group and 19 children in the athetoid group.
12. Lesný (1971), Czechoslovakia	143; NR; 6 – 15 yrs	Hemiplegia (65), diplegia (35), generalised athetosis (18), tetraplegia (14) and hemiathetosis (11); NR	TPD (using Weber's scissors at 8 different sites: tip of third finger, centre of third finger, centre of palm, carpal joint, middle of forearm, elbow, middle of arm, & acromion)	Y, 30 (6-10 yrs (14) and 11-15 yrs (16))	Cohort or sub-cohort percentages not reported.	Age related TPD differences noted for control groups – older group more sensitive at tip of right third finger, less sensitive at middle of left palm and acromion of right hand. Greatest disturbances were found on most distal parts of the ULs, particularly tip & centre of the third finger. Disturbances present on both sides for hemi group.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
13. Uvebrant (1988), Sweden	From 148 to 114 (cohort size varies); ~ 59% male; 6 – 15 yrs	Hemiplegia, grouped as congenital (preterm or term) and postnatal; NR	Stereognosis (5 objects), graphesthesia (5 figures), TPD (index finger), pain, position sense/proprioception, temperature, light touch, vibration *[*] TPD evaluated as: good = 0-4mm, moderately impaired = 5-7mm, poor > 8mm*	N	Stereognosis: impaired in 44%, complete astereognosis in 20% Graphesthesia: impaired in 51% (ave.) TPD: impaired in 46% (average) *[*]* Pain: reduced = 16% Position sense: reduced = 12% Temp.: reduced = 12% Light touch: reduced = 11% Vibration: reduced = 7%	For stereognosis, grapheasthesia & TPD, preterm congenital CP group *least affected,* postnatal CP group *most affected*, particularly for TPD & grapheasthesia. Undergrowth of affected upper limbs was common (96%); degree of undergrowth correlated largely to severity of sensory and motor impairment. Motor dysfunction was moderate (31%) or severe (19%) in half the cohort. Pain, temp., vibration & position sense mostly preserved in all cases (84-93% recorded 'normal'). Normal CT was a common finding for children with mild disability, unilateral ventricular enlargement in those with moderate disability, & cortical/subcortical cavities in those with severe disabilities.
14. Bolanos *et al.* (1989), USA	51; NR; 6 – 20 yrs (mean = 12.2 yrs)	Main types include spastic (47), diplegia (20), quadriplegia (15), hemiplegia (10), and triplegia (3); NR	TPD (5mm) and stereognosis (using 3 plastic shapes, a triangle, square and circle, not objects)	Y, 170 (6 – 20 yrs)	TPD: 32/51 Stereognosis: 20/51	TPD had higher sensitivity than stereognosis for detecting tactile defects, but slightly poorer specificity. Most TPD errors occurred over the middle phalanges; CP group had a sig. higher incidence of defects over the distal phalanges. No sig. sex diff. for either test; no significant age diff. for TPD.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
15. Lesný *et al.* (1993), Czechoslovakia	220; NR; 7 – 14 yrs	Classic diplegia (46), mildly spastic diplegia (23), hemiplegia (86), quadriplegia (25), dyskinesia (26), right (10) & left sided (4) hemiathetosis; NR	TPD (at 8 different sites, as per Lesný (1971))	Y, 30 (same control group as Lesný, 1971)	Prevalence rates not reported. Sig. diff. reported b/n CP type (bilateral & hemi) and controls per CP sub-type, extremity, and assessment site	Decreased TPD sensitivity was greatest for children with spastic CP compared to athetoid CP. Decreased TPD on dom side for hemiplegic children, decreases were greatest at distal (tip and middle of the third finger) compared to proximal points Most marked changes in TPD: diplegic children; on ND side of hemi children
16. Van Heest *et al.* (1993), USA	40; 24 males; 11 yrs (ave.)	Only congenital spastic hemiplegia; 18 left-sided, 22 right-sided; NR	Stereognosis (12 objects: cube, key, pencil, penny, marble, string, button, safety pin, pill, rubber band, spoon, and paper clip), TPD (6mm spacing at finger tips), and proprioception (movement of the fingers up and down); also measured relative limb size at 4 different points in both ULs	N	Overall: 39/40 (97%) *Major modalities impaired* – Stereognosis: 39/40 TPD: 36/40 Proprioception: 18/40	Stereognosis deficit severity directly correlated with impairment of TPD but not with impairment of proprioception. All children had a size discrepancy in affected vs. unaffected UL in at least two parameters – spastic limb sig. smaller for all parameters. Children with severe stereognosis deficits had sig. smaller UL in all four measures compared to children with mild or moderate stereognosis deficits. Trend toward increased TPD and proprioception deficits in children with increased size discrepancy.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
17. Yekutiel *et al.* (1994), India	55; NR; 6 – 17 yrs	Mixed or ill-defined (15), spastic quadriplegia (14), spastic diplegia (12), spastic hemiplegia (8), athetosis (6); NR	Stereognosis (10 familiar objects, presented in the following order: pencil, Dinky car, handkerchief, tea-spoon, book, ping-pong ball, comb, wooden cube, stone, key), TPD (5mm, 10mm and one-point stimuli at four locations), location of touch (blunt pencil) *[*] Control group was not 'normal', but post-polio children*	Y, 15 *[*]*	Overall: 28/55 (51%) *Major modalities impaired* – TPD: 24/36 Stereognosis: 15/55	Stereognosis could be tested on whole cohort, but not TPD (only 36/55), and location of touch test was abandoned as children did not understand test. Used a 10% error (2 incorrect / 20 for stereognosis, 4 / 40 for TPD) to set a 'normal' performance benchmark. TPD at 5mm was most difficult to discriminate. No correlation with age or cognitive levels; diagnostic category showed sig. diff. for children with athetoid compared to hemiplegic CP, with later showing most sensory loss, but only on ND side.
18. Cooper *et al.* (1995), Canada	9; 7 males; 4 – 18 yrs (mean = 11.25 yrs)	Only hemiplegia (5 right-sided, 4 left-sided); NR	Pressure sensitivity (SWM), TPD (static, 1-12mm, & moving, using the Disk-criminator), stereognosis (5 shapes: circle, triangle, square, diamond, octagon + 5 everyday objects: toothbrush, tennis ball, 4-inch comb, large cup, candy in wrapper), proprioception (thumb & 2 fingers), and directionality (using SWM)	Y, 41 (21 female, 4 – 16 yrs, mean = 9.88 yrs)	Overall: 8/9 (89%) *Major modalities impaired* – Stereognosis: 7/9 (ND hand), 3/9 (dom hand) Proprioception: 6/9 (ND hand), 3/9 (dom hand)	Left-hemi children more severely involved (neurologically) compared to right-sided children. Stereognosis and proprioception chief modalities affected. Reported sensory loss on both sides. Extent of sensory loss did not correlate with motor deficit severity, whereas somatosensory evoked potentials closely related to motor function. Authors endorsed clinical 'battery' testing multiple sensory modalities & testing both hands of hemi children.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
19. Gordon and Duff (1999), USA	15; 10 males; 8 – 14 yrs	Only hemiplegia; NR	TPD (Disk-criminator, 2-15mm), pressure sensitivity (using SWM on index finger + thumb), and stereognosis (based on the 'Manual Form Perception Test' using 8 shapes), also tested pinch strength, spasticity (Modified Ashworth Scale) and manual dexterity (6 subtests from JTHFT)	Y, 15 (8 – 14 yrs)	TPD: 4 x larger than controls Pressure sensitivity: impaired, but less so than TPD Stereognosis: range 0-5 out of 8 (controls: range 7-8)	TPD, stereognosis, and manual dexterity (via JTHFT) all sig. correlated with pinch strength. TPD, along with spasticity, were the strongest individual predictors of static grip force. Pressure sensitivity wasn't useful singular predictor of any measured parameter, but TPD was. Cohort approx. 10 times slower than controls with JTHFT.
20. Krumlinde-Sundholm and Eliasson (2002), Sweden	25; NR; 5 – 18 yrs	Only hemiplegia – 21 (mild) and 4 (moderate), 14 right side hemi; NR	SWM (5-filament kit), TPD (3 and 7mm), stereognosis (6 paired familiar objects: Lego brick/eraser, wooden bead/ paper pellet, coin/shirt-button, and 10 flat forms, from the 'Manual Form Perception Test'), functional sensibility (Pick-up test with and without vision), dexterity, spasticity (Ashworth), bimanual task performance	Y, 19	Overall: 18/25 (72%) [*excluding stereognosis of forms*] For ND side only: Stereognosis: 11/25 (objects) & 24/25 (forms) TPD: 18/25 (3mm) & 13/25 (7mm) Pick-up test: 10/25 SWM: 5/25	Poor SWM performance predicted poor performance in all other sensory tests. Most useful tests: stereognosis (familiar objects), TPD (3mm), functional sensibility (Pick-up test, with & without vision). Only stereognosis of forms identified impairments in dom hand (15/25) – other tests showed dom hand performed as 'normal'. More than half of control group made mistakes on this test, test not recommended as being beneficial for testing tactile sensibility. Correlation b/n age and (i) Pick-up test results for controls (older children performing better) & (ii) stereognosis of forms for dom hand.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
21. Arnould *et al.* (2007), Belgium	101; 59 males; 6 – 15 yrs (mean = 10 yrs)	Tetraplegia (31), diplegia (20), hemiplegia (50; 25 left and 25 right); NR	Pressure (SWM), stereognosis (as per (Cooper *et al.*, 1995)), proprioception (as per (Cooper *et al.*, 1995)), grip strength, gross manual dexterity (Box and Block Test), fine finger dexterity (Purdue Pegboard Test), manual ability (ABILHAND-Kids)	Y, but no details provided	All CP types combined: SWM: 21% (dom) and 33% (ND) Stereognosis: 20% (dom) and 38% (ND) Proprioception: 4% (dom) and 15% (ND)	Sensory impairments identified on both sides for children with hemi. Tetraplegic group performed worst on all measures (sensory & motor) for dom hand, but better on ave. than hemi group for ND hand. Diplegic group performed best for all sensory measures on both hands. Hemi group performed best for motor measures on dom hand. Manual ability sig. but moderately correlated with motor impairments and stereognosis, but not with pressure detection and proprioception. Motor impairments markedly more prevalent than sensory impairments for all CP types.
22. Wingert *et al.* (2008), USA and **23.** Wingert *et al.* (2009), USA	38; 22 females; diplegia: 7.3 – 34.3 yrs (mean = 14.8 yrs) hemi: 8.6 – 26.5 yrs (mean = 13.75 yrs)	Hemiplegia (17) diplegia (21); all subjects were GMFCS Level I or II and MACS Level I or II	Object recognition (using 5 common objects (key, penny, pencil, spoon, and button), 4 embossed geometric shapes (triangle, square, circle, star), and 8 embossed capital letters (A, O, W, J, U, L, T, I)), roughness via paired horizontal gratings of various groove widths (grating: 22mm wide x 38mm long, groove width = 0.25mm width, groove height = 0.5mm high) (2008), and joint position sense and kinesthesia, with and without vision, for arm and foot (2009)	Y, 21 (11 males, mean = 14.83 yrs)	Cohort or sub-cohort percentages not reported.	Both cohorts had sig. increased thresholds for groove width difference with both hands compared to control. For diplegic group, only roughness discrimination differed b/n hands. For hemi group, sig. b/n hand diff. recorded for all tasks. Sensory deficiencies recorded on both sides for both groups. Tactile sensory impairments in both ULs probably impact tactile guidance of hands, possibly contribute to awkward dexterity. Sig. joint position sense deficits for all limbs except dom UL for both groups with vision occluded. Kinesthesia deficits present in ND UL for both groups. Joint position sense and kinesthesia deficits noted on dom side for hemi group.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
24. Holmström *et al.* (2010), Sweden	17; 9 male; 7 – 16 yrs (mean = 11.4 yrs)	Unilateral CP; all subjects GMFCS Level I, MACS Level I (6) and II (11)	TPD (fingertips of digits II–IV), mirror movements, and hand function via the Box and Blocks Test and Assisting Hand Assessment (AHA) *[*] TPD evaluated as: normal = at least 3mm, decreased = 5mm, poor = not able to discriminate at 5 - 7mm*	N	TPD: 8/17 (47%) *[*] 6 = poor, 2 = decreased*	All children with 'poor' TPD were classified as MACS Level II and in group with poorer hand function. Strong negative correlations b/n TPD & performance on Box and Blocks Test & AHA. Combination of type, location, and extent of lesion is a stronger predictor of hand function. Most favourable hand function identified in children with white-matter damage of immaturity with mild white-matter loss and contralateral motor projections; children with ipsilateral projections had the most impaired function. Dom hand performed worse on Box and Blocks Test than expected for age of child for 88%.
25. Klingels, Demeyere*, et al.* (2012), Belgium	81; 43 male; 5-15 yrs (mean = 9 yrs 11 mths)	Unilateral CP (congenital = 69, acquired = 12), left-sided (36), right-sided (45); MACS Level I (29), Level II (36), and Level III (16)	Exteroception (thumb, index and hand palm), proprioception (index finger), TPD (Aesthesiometer®) and stereognosis (6 objects from 12). Also assessed passive range of motion (PROM), muscle tone, muscle strength, grip strength, and the Melbourne Assessment, Assisting Hand Assessment (AHA) and ABILHAND-Kids	N	Overall: 45/69 (65%) *Major modalities impaired –* Stereognosis: 65% TPD: 58% Proprioception: 21% Exteroception: 9%	Only 69 of 81 children could complete sensory assessments. TPD & stereognosis were only modalities that recorded an 'absent' result, other modalities were 'impaired' or 'intact' only. For ABILHAND-Kids, children > 10 yrs performed sig. better than children < 10, and no sig. diff. found b/n congenital & acquired CP groups. Also, no sig. diff. b/n congenital & acquired groups for sensory tests. Children with a lower MACS level or acquired lesion had significantly more impairments and activity limitations.

Author(s) (year), Country	*Cohort number (n); Sex (n); Cohort age (years)*	*CP type (n); GMFCS and MACS*	*Sensory and other assessment tests conducted*	*Control group (Y/N), (n)*	*Somatosensory deficit prevalence (n/n or %)*	*Comments / Notes*
26. Auld *et al.* (2012b), Australia	52; 29 male; 8 – 17 yrs (median = 12 yrs)	Unilateral CP; GMFCS Level I (34) and II (18), MACS Level I (36) and II (16), 23 left-sided CP	Tactile registration: SWM (20-filament kit) Spatial-tactile perception: single-point localization (SWM), double simultaneous (SWM), static (STPD) & moving TPD (MTPD)(using the Disk-Criminator), and stereognosis (9 common objects) Texture perception: AsTex	Y, 34 (20 males, 5 – 17 yrs, median = 9 yrs, 9 left handed)	Overall: 40/52 (77%) *Major modalities impaired* – Stereognosis: 63% Double simultaneous: 58% Single-point localisation: 40% STPD: 31% MTPD: 27% Texture: 17%	ND hand of CP group performed worse than ND hand for control group for all tests except MTPD; dom hand for CP group performed sig. poorer than either hand for control group for all tests except MTPD. Tactile registration deficits clearly associated with increased likelihood of tactile perception deficits. No sig. diff. b/n child with L vs. R CP for any test, but trend noted that LCP (R side brain lesion) have more severe sensory deficits. 28 (54%) children recorded sensory impairments on dom side. Authors endorsed clinical 'battery' testing multiple modalities due to low correlation b/n tactile variables, meaning no redundancy within test battery.
27. Kurtaran *et al.* (2015), Turkey	36; 24 male; (ave. = 6.28 ± 1.95 yrs)	Hemiplegia (6), diplegia (13), total involvement (17); NR	Stereognosis (10 objects; spoon, pencil, eraser, napkin, key, wood, ball, money, fork, pencil sharpener), light touch, pain, TPD, graphesthesia, lifting two objects (200g & 400g) & grasp force (Jamar dynamometer)	Y, 18 (12 male, ave. = 6.61 ± 1.61 yrs)	Graphesthesia: 53% TPD: impaired (5%), non-testable (67%) Superficial sensation: 6%	Sig. negative correlation b/n hand functional status & grasp force of both hands. Sig. negative correlations b/n hand functional status & lifting (both masses) for both hands, & stereognosis performance for ND hand.

Notes: USA = United States of America; yrs. = years; NR = not reported/information absent from article; CP = cerebral palsy; TPD = two-point discrimination; L = left; R = right; b/n = between; hemi = hemiplegia; UL = upper limb; dom = dominant; ND = non-dominant; ave. = average; med. = median; SWM = Semmes-Weinstein Monofilaments; JTHFT = Jebsen-Taylor Hand Function Test; GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System; dims = dimensions; sig. diff. = significant difference(s); g = grams; approx. = approximately; temp. = temperature.

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2.1.2 Sensory Re-Training for Children with Cerebral Palsy

More than fifty years ago, Kenney (1963) noted that sensory deficiencies may play a large role in the practical motor function of the hands, and queried if it was possible to overcome sensory deficits through training, noting that it "*has not yet been demonstrated that it is possible to do so*" (pg. 194). Kenney stated that children with sensory deficits should be taught to recognise objects, including differences in shape, texture, size and function.

Being based in the United States, it is possible that Kenney wasn't aware of the work of French researchers Monfraix and Tardieu (1961), who investigated a training and re-educating program involving four children with sensory deficits due to CP, with a focus on manual perception (shape recognition). The process involved testing the child as per their earlier study (Monfraix *et al.*, 1961), but this time highlighting where and when mistakes were made, meaning incorrect shape identification was being immediately corrected. One technique involved passing the same object between hands (ND hand first, followed by the dominant hand) to recognise that the same object was being held each time. Monfraix and Tardieu (1961) underlined the value and importance of re-education, even in severe cases of sensory agnosia (pg. 555), and that frequent practice involving putting objects into the child's hands as often as possible, as early as possible, was critical (pg. 556). They noted that the re-education process is sometimes prolonged (one of the case studies presented went for over two and a half years) but should be sustained as "*fallings off*" are frequent when the re-education program is interrupted (pg. 557). Following their study the authors concluded that if the agnosia was caused by a lesion, re-education produces only long-term success, whereas if the reason for the agnosia is lack of use, results are obtained much quicker (pg. 557).

Around the same time, Jones and Ogg advocated for and highlighted that an integrated sensory approach (one that involves visual, auditory, tactile and kinaesthetic stimuli) was essential for motor function development (Jones & Ogg, 1966). They also recommended the stimuli should be both cutaneous and proprioceptive, and similar to Monfraix and Tardieu (1961), that it should begin as early as possible to avoid the deprivation of sensory experiences (Jones & Ogg, 1966). Following this work, Barrett and Jones (1967) published what appears to be

the second study within the CP literature that investigated sensory re-training, this time in the United States. The novel intervention involved a “sensory story” that used an interactive, repetitive, hands-on story that was read to six toddlers with hemiplegic CP (age range: 19-60 months, four female, four right-side hemiplegia) in a pre-nursery/school setting. The story was personalised (used the child’s name), used contrasting sensory words (e.g. soft/stiff, smooth/sharp, hot/cold), and involved the touching and handling of story objects using both hands as the story was read. The number of sessions ranged from eight to 32 (mean = 18) over a period of three to five months, with five of the six children having diminished light touch in their affected hand.

Barrett and Jones (1967)’s study philosophy was akin to Forster and Shields’ sensory rehabilitation conditioning experiment that involved repeatedly exposing an adult with a sensory deficit post cerebral vascular accident, to tactile sensory experiences while providing verbal cues and positive reinforcement when a task was successfully completed (Forster & Shields, 1959). While time consuming and involved, the essence of the experiment was to enable new engrams to be laid down, where vision was an important mechanism for assisting this process, a critical aspect noted by Wingert *et al.* (2009) and more recently referred to as “*visually enhanced touch*” (pg. 1852) by Auld and Johnston (2018). Barrett and Jones conducted their study with young children as they recognised that the timing of sensory training may be crucial, taking into account ‘critical periods’ in learning, and that encouraging the child to use their affected hand early was important.

Following Friedman two-way analysis, the authors reported that the application of their sensory story resulted in a significant increase in the spontaneous reach and grasp of the affected hand in young children with hemiplegia during training sessions (pg. 453), which appeared to carry over into play activities at other times. In addition to increased use of the affected hand, the authors observed that their repeated ‘sensory story’ appeared to also increase verbal responses and attention span, though neither was formally measured.

2.1.3 Upper Limb Somatosensory Deficits and Surgery

Tizard *et al.* (1954)'s landmark paper included a cautionary note regarding the understanding and appreciation of hand sensory function prior to hand surgery, highlighted by their case study analysis. This important aspect of hand function was echoed by Tachdjian and Minear (1958), who assessed 15 children with sensory deficits post hand surgery with none showing appreciable functional improvement, and became a theme of other studies (Kenney, 1963; Bolanos *et al.*, 1989; Van Heest *et al.*, 1993; Yekutiel *et al.*, 1994). Not long after Tizard *et al.* and Tachdjian and Minear's work, orthopaedic surgeons Goldner and Ferlic advocated an understanding and appreciation of the sensation of the hand in their preliminary assessment prior to surgical treatment for children with an upper limb involvement (Goldner & Ferlic, 1966). Jones (1960) recommended that understanding a child's sensory function as the child matures was essential, particularly prior to surgery. Decades later, Bolanos *et al.* (1989) reiterated that pre-operative sensory testing would assist in setting realistic functional goals for the child, family, therapists and physicians, adding that if sensory deficiencies were present then the child may be required to use (or be instructed to use) visual feedback to compliment hand use, even after physical deformities have been surgically corrected (pg. 371). Bell-Krotoski, Weinstein, and Weinstein (1993) noted the value and role of vision as an educational technique to compensate for tactile deficiencies of the hand. Similarly, Wingert *et al.* (2009) identified vision as a probable compensatory strategy for limb use by children with CP, and advocated that vision should be engaged and relied on while movements are learnt and practised (pg. 452).

With respect to hand surgery, Eliasson *et al.* reported on a study of 32 children and young adults (aged six to 20; median = 12 years), who underwent a range of hand surgeries including muscle releases and tendon transfers (Eliasson, Ekholm, & Carlstedt, 1998). The authors reported improved hand function for all children when examined and assessed nine months post-surgery, with the main advantage post-surgery being a more functional position of the hand, with increased extension and forearm supination (pg. 612). The authors reported increased functionality of handgrips, grip strength and dexterity, but no change in tactile sensibility, assessed via TPD (using a paperclip on the tip of the thumb, index and middle finger and a threshold of 3-4mm as 'normal') and stereognosis (using six paired but different

objects, with two points awarded for a correct guess, and scoring at least 10 points considered 'normal'). Eliasson *et al.* (1998) reported that children with impaired hand sensibility benefited from the surgery to the same extent as children with 'normal' sensibility (pg. 618), which appears to contradict the cautionary approach from the literature to corrective surgery for hands with poor sensibility.

Contrastingly, in the same year that Eliasson *et al.* (1998) reported no improvement in stereognosis ability, Dahlin *et al.* reported on a six year study involving 36 children and young adults (aged five to 25; median = 15 years) with hemiplegic CP. All participants had surgery on their more involved hand, with significant stereognosis improvements observed at six and 18 months post-surgery compared to pre-surgery performance (Dahlin, Komoto-Tufvesson, & Sälgeback, 1998). Stereognosis using four different objects was assessed at the relevant time periods, but the same four objects were not used during the follow-up assessments. The authors couldn't identify the reason for the stereognosis improvements but observed that in general the hand was more efficient post-operatively, meaning the test objects were more easily exposed to the palm and fingertips (pg. 338). This led them to postulate that the in-hand manipulation was causing increased tactile stimulation of specific areas of the hand and their corresponding cortical projectional areas in the brain, causing increased formation of synapses due to the afferent stimuli. They suggested that "…*stereognosis can be improved following surgical reconstruction of the upper extremity in cerebral palsy due to functional cerebral reorganization induced by the modified afferent inflow*" (Dahlin *et al.*, 1998, pg. 339).

In support of Dahlin *et al.*'s work related to improved hand efficiency, Carlson and Brooks (2009) demonstrated that upper limb position and the ability to manipulate an object is an important factor for stereognosis function. Their study involving 21 typically developing adults (aged 18 to 55; mean = 30 years) placed each participant's ND upper limb in a hemiplegic hand position simulator, and assessed stereognosis performance. The authors used 12 different objects and Van Heest *et al.* (1993)'s protocol over three trials – using the simulator, not using the simulator (normal hand position), followed by using the simulator again. Carlson and Brooks reported significantly different stereognosis performance between all three trials, with the mean scores for each trial being 7.6 objects (trial one), 11.7 (trial two), and 9.3

(trial three), also suggesting a learning effect between trials one and three. Consequently, the stereognosis performance of 'healthy' adults when using the hemiplegic hand position simulator, which caused decreased mobility and altered hand/wrist position, caused stereognosis performance to significantly decrease. They concluded that Dahlin *et al.*'s significant results could be due to improved hand function following surgery, as their results identified. Similar work by Bensmail *et al.* (2009) investigating wrist position and grip force in typically developing adults (aged 28 to 39; mean age = 28 years) demonstrated that a change in horizontal wrist position adversely affects grip force scaling, particularly when the wrist is hyperextended.

In contrast to Dahlin *et al.*'s study, Petersen *et al.*'s recent study assessing the effect of treatment on stereognosis for 63 children (aged four to 16 years) with hemiplegic CP reported no statistically significant improvement in stereognosis function following either operative or non-operative treatment (Petersen *et al.*, 2016). Treatment included surgery with rehabilitation, Botulinum toxin injection with rehabilitation, and rehabilitation alone, with the authors stating that all treatments are intended to improve hand function, appearance and hygiene. Their baseline stereognosis assessment using 12 objects identified impairments in 92% of children on their affected side and in 38% of children on their unaffected or dominant side, once again highlighting the nature of bilateral impairments amongst a hemiplegic cohort. Intact stereognosis function was defined as identifying all 12 objects correctly, a mild impairment was identifying between nine to 11 objects, a moderate impairment was identifying five to eight objects, and a severe impairment was identifying four objects or less. The cohort demonstrated stereognosis impairments across all classifications, with the majority of the cohort (43%) having severe impairments on their affected side. The authors also reported that stereognosis function did not appear to be influenced by age.

With specific reference to Dahlin *et al.* (1998)'s work, Petersen *et al.* proffered their different and more rigorous stereognosis test to explain different outcomes between the studies, with Dahlin *et al.* using less objects (only four) and a protocol that involved using four different objects at different time points, whereas Petersen *et al.* used the same 12 objects each time. Petersen *et al.* also noted the greater time

period between surgery and testing for Dahlin *et al.*'s study, potentially enabling more time for sensory re-education than for their own study. Additionally, Petersen *et al.*'s cohort was younger, whereas Dahlin *et al.*'s study had participants aged up to 25 years. In his commentary on the conflicting observations discussed in this section, Seruya suggests that a dose-dependent phenomenon may be occurring, and that improvement may need to surpass a threshold before being quantified via a test of stereognosis (Seruya, 2016).

2.1.4 Somatosensation and the Lower Limbs

Apart from a brief reference by Monfraix *et al.* (1961), noting that sensory deficits were identified in the upper limbs of their cohort when the motor disorders of CP appeared clearly confined to lower limbs, few studies have investigated sensory function in the lower limbs of children with CP. One of the first such studies was conducted by McLaughlin *et al.* (2005), who assessed the lower limbs of 62 children with mixed CP (mostly spastic diplegia) and 65 typically developing children. Their work acknowledged that of Dannenbaum and Dykes (1988), which recognised the role that sensory stimuli plays with respect to motor actions, and Umansky (1973), who hypothesised that decreased or absent afferent cerebral inputs affect both motor learning and functional body image, which was proposed six years earlier by Barrett and Jones (1967), who also wondered if timing aligned with critically important developmental periods was crucial. The consequence of decreased or absent afferent inputs for individuals with sensory deficits are that they will fail to incorporate the involved body part into a complete and functional body image, resulting in limited use of the involved body part. McLaughlin *et al.* (2005) noted that Umansky's hypothesis was supported in the literature following studies of both animals and humans with congenital or acquired sensory deficits, where failure to use the de-afferented limb resulted in a learned disuse phenomenon, resulting in a greater deficit of motor capability in the involved limb (McLaughlin *et al.*, 2005, pg. 46). Given the importance of afferent sensory inputs, McLaughlin *et al.* proffered that the variability reported in the literature in terms of performance among children with apparently similar motor deficits may be due to underlying and unrecognised sensory deficits.

The testing battery that McLaughlin *et al.* (2005) used included testing for light touch (using a cotton ball), pain sensation (sharp and dull ends of a safety pin), position sense of the big toe (up or down), position sense of the knee, vibration sense (using a 128Hz fork) and direction of scratch (using the wooden end of a cotton applicator). The authors identified that sensory testing was feasible in children with CP from as young as five years of age. The tests for direction of scratch, toe position and vibration sense were the most impaired modalities within the CP group compared to the control group, which the authors acknowledge are traditionally anatomically assigned to the dorsal columns. Children with spastic diplegia recorded fewer deficits compared to the total cohort, with the authors also reporting that the disturbance of vibration sense did not appear to be associated with spastic diplegia.

While McLaughlin *et al.*'s study focussed solely on sensation of the lower limbs, as mentioned earlier, Wingert *et al.* (2009)'s study (Table 2, **[23]**) assessed joint-position sense and kinesthesia in all limbs for their cohort of children with hemiplegia and diplegia. The authors reported significant deficits for joint-position sense in the lower limbs bilaterally for both CP groups, and bilateral kinesthesia deficits in the lower limbs for the hemiplegic group only, when vision was occluded.

2.2 Conclusion

This chapter has reviewed and appraised the current literature relating to upper limb somatosensory assessment studies involving children with CP. A number of themes emerged from the literature, such as: sensory deficits being bilateral for children with hemiplegic CP; children with acquired CP having more impairments that may also be more severe than children with congenital CP; children with hemiplegia tending to have more impairments on their ND side compared to children with diplegia; children with athetosis having fewer impairments compared to children with other forms of CP, particularly spastic CP; and children with left hemiplegia being more involved than children with right hemiplegia.

With respect to the sensory assessments, stereognosis was the modality most frequently impaired, with stereognosis of common objects reported as having greater validity compared to stereognosis of forms. Multiple authors report being unable to

administer a given sensory test to their whole cohort, and multiple studies advocated a cautious approach to upper limb surgery without knowing the extent of sensory loss in advance. However, as discussed in section 2.1.3, the literature contains conflicting reports on this aspect that remain unresolved.

Additionally, a number of studies have highlighted the role of vision (Monfraix & Tardieu, 1961; Jones & Ogg, 1966; Barrett & Jones, 1967; Bell-Krotoski *et al.*, 1993; Wingert *et al.*, 2009), particularly the role it plays as an educational technique to compensate for tactile deficiencies of the hand. Atkinson (2002) acknowledged that vision is the main sensory system that guides our actions, while Brown *et al.* (1987) recognised the importance of both sensory and vision inputs when stating that "*sensory input is as important in motor learning as hearing is in the learning of speech, although vision may help overcome sensory deficits of the limb*" (pg. 299). Moreover, the 'Apartment Block Theory' framework recently proposed by Auld and Johnston (2018) noted the "*need to continuously and consciously capitalise on vision in tactile training endeavours*" (pg. 1852).

3. Overall Study Design and Study Methods

This chapter introduces the study design and discusses the assessment measures and tools that were used throughout the overall study. Each assessment measure is introduced and described, with rationale and justification for why a particular measure was used, and identification of where other studies in the literature have used the same tool with children with CP.

3.1 Overall Study Design

The overall study was divided into two main sections, referred to as Stage 1 and Stage 2. Stage 1, presented in detail in Chapter 4, was the initial recruitment and assessment phase of the project. The aim of this stage was to assess children living with CP in South Australia to determine the nature and prevalence of upper limb somatosensory function for this cohort, as a study of this nature had not been done before. The assessments that occurred during Stage 1 were referred to as A_0 assessments as they were screening assessments for the stage to follow.

Stage 2, presented in detail in Chapter 6, was the study intervention. During this stage children with a confirmed somatosensory impairment identified in Stage 1 were invited to participate in a home-based randomised controlled trial (RCT) using the customised serious gaming system, which is described in detail in Chapter 5. The Stage 2 RCT had two different arms and three assessment points, which are described in detail in Chapter 6. In brief, the assessment points occurred over a 14 week period: a baseline assessment (A_1); an 'immediate' post-intervention assessment (A_2) 10 weeks later; and a follow-up assessment (A_3) a further four weeks later. The overall study is shown schematically in Figure 3-1.

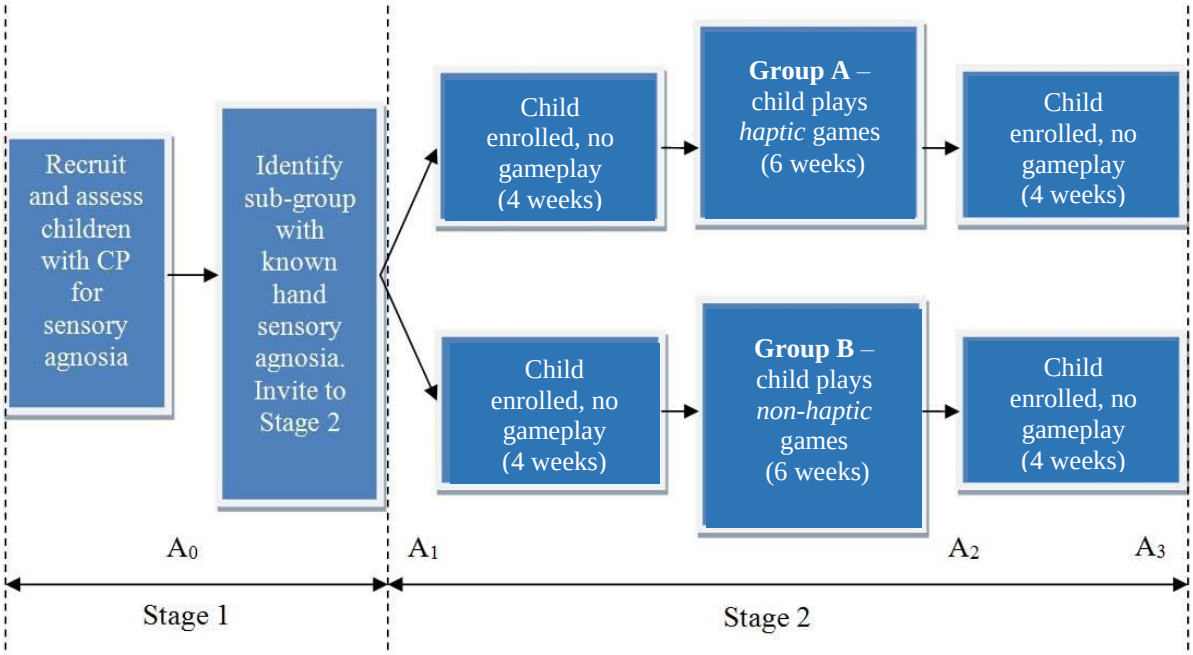

Figure 3-1 – An overview of the overall study, highlighting the two project stages, the relative assessment points (A_x), and the two arms of the randomised controlled trial

3.2 Somatosensory Assessment Measures

3.2.1 Informed, Relevant and Valid Assessment Measures

The literature review presented in Chapter 2 (and summarised in Table 2) reported the outcomes of all upper limb somatosensory assessment studies involving children with CP from 1952 – 2015, identifying the breadth of tests that were administered over the years and the modalities that each test was assessing. This initial research assisted in identifying the most appropriate tests for this population and was informed by two key publications that investigated appropriate, clinically relevant and reliable tactile sensory assessments for children with CP. These publications were Klingels *et al.* (2010)'s study of upper limb motor and sensory measurement reliability, and the clinimetric review of tactile sensory assessments by Auld *et al.* (2011). The aim of Auld *et al.*'s review was to identify and examine the clinimetric properties of a range of assessments that had been administered to test tactile registration and perception in children with CP and to provide recommendations for clinical practice. This work was important because of the lack of a standardised testing protocol and procedures in this area.

Auld *et al.*'s review identified two key areas of sensory assessment to be evaluated: tactile registration or sensation, defined as "*the initial awareness of sensory information*" (pg. 414), citing the work of Kandel, Schwartz, and Jessell (2000) and Williamson and Anzalone (2001), and tactile perception, defined as the ability "*to understand, interpret, or give meaning to sensory stimuli*" (pg. 416), citing the work of Koppitz (1970). The authors used the *CanChild Outcome Measures Rating Form Guidelines* (Law, 2004), which enabled an assessment of the clinical utility, reliability, validity, and test responsiveness for each sensory test.

From all sensory assessments published in the literature, Auld *et al.*'s review identified two appropriate tests of sensory registration: Semmes-Weinstein Monofilaments (SWM) and exteroception as per Klingels *et al.* (2010), and six tests of sensory perception: Single Point Localization, Double Simultaneous, Two Point Discrimination (TPD), graphesthesia, stereognosis, and Manual Form Perception (Auld *et al.*, 2011). The authors concluded that there wasn't a single tool sufficiently broad in scope to address all the required constructs of a comprehensive tactile assessment framework, and consequently recommended a combined battery of assessments as part of a comprehensive examination of tactile function. As reported in Chapter 2, this was also the conclusion of previous researchers (Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002).

Refining the battery of assessments, Auld *et al.* recommended using SWM for testing sensory registration, despite the increased expense and time to administer, compared to exteroception as per Klingels *et al.* (2010). The recommended tests for tactile perception were Single Point Localization, TPD (using the Disk-Criminator® in preference to a paper clip, and adopting the static application method), Double Simultaneous (incorporating proximal vs. distal and left vs. right areas of the body), and stereognosis (as per Klingels *et al.* (2010) compared to that of Cooper *et al.* (1995)). Auld *et al.*'s recommendations formed the basis for their tactile sensory assessment study published soon after (Auld *et al.*, 2012b). The process of preparing for the study, the setting in which the assessments occurred, and the chosen tactile assessments administered are described in the following sections.

3.3 Somatosensory Assessment Preparation, Setting and Tests

All Stage 1 assessments were conducted at the Paediatric Rehabilitation Department of the Women's and Children's Hospital (WCH) in North Adelaide by a qualified and registered Occupational Therapist (OT) or Physiotherapist (PT). An OT assessment room at the hospital was booked for each assessment to minimise noise and other distractions, and a height adjustable table was used where possible. All the assessments were conducted with the child in a sitting position with their parent(s) present.

Prior to the study beginning, an assessment familiarisation and training session was held with a number of OT staff from the Rehabilitation Department, which was led by the author and one of the project supervisors (SH). The session was designed to address questions that staff had with respect to the assessments to be used, to review and pilot the assessment recording form for the study (see Appendix A), and to agree on a consistent approach to conducting the assessment sessions. The session was conducted following recommendations in the literature with respect to improving interrater reliability by standardising the test procedure and refining the scoring criteria by providing assessment training (Klingels *et al.*, 2010). All staff involved in direct contact with the children had the necessary Australian national police clearance/background checks required to work within a paediatric rehabilitation facility.

While one staff member was recruited to the project and appointed as the designated OT for the study, the group training session group was conducted with multiple therapists in case the appointed OT was unavailable for a given assessment, and to leverage the skills and experience of a number of Allied Health Professionals when suggesting changes to the study assessment form. Following the training session a number of formatting/layout changes were suggested to improve assessment fluency, and a small description of each test was included under the heading for each test.

All Stage 2 assessments were similarly conducted at the WCH with the exception of one non-metropolitan child who lived three hours from Adelaide. To assist this family and to reduce time away from school and travel involved, the OT conducted this

child's A_2 and A_3 assessments at their regional school. In terms of assessors, three OTs conducted the Stage 1 assessments as the designated OT took maternity leave mid-way through the project and cover couldn't be provided by one extra OT. For similar reasons two OTs and one of the project supervisors (SH) completed the Stage 2 assessments.

For all sensory tests except stereognosis, the dominant hand was tested first, followed by the non-dominant (ND) hand, as per other studies (Klingels *et al.*, 2010, pg. 412). The sensory tests, which are described in detail in the following sections, represented a hierarchy of perceptual difficulty from a brain processing perspective, from simple to complex. Consequently, the testing sequence was:

1. A simple test of tactile registration, on the child's first (index) finger and thumb, using the SWM (section 3.3.1);
2. A test of discrimination/perception using the *AsTex* device (described in section 3.3.2), assessing not only tactile registration, but if the child being assessed can judge if the sensation is rough or smooth;
3. A test of proprioception sensing thumb position in space (section 3.3.3);
4. A test of stereognosis or haptic perception using 12 objects, six per hand (section 3.3.4); and
5. A test of hand motor function, assessed using the Jebsen Taylor Test of Hand Function (JTTHF)(section 3.3.5), where the ND hand was always tested first, followed by the dominant hand, as per the test kit and original instructions (Jebsen *et al.*, 1969).

Each assessment and how it was administered is described in the following sections.

3.3.1 Semmes-Weinstein Monofilaments (SWM)

The first test conducted for Stage 1 and 2 was a test of light touch or tactile registration using SWM (Stoelting Company, Wood Dale, IL, USA, 60191). SWM are a standardised, non-invasive, motor-free way to assess and measure cutaneous light touch through an objective and repeatable process and are recognised as the best hand-held instrument to monitor sensory change, particularly if the same tester uses

the same instrument to repeatedly test an individual over time (Tubiana, Thomine, & Mackin, 1996). Consequently, SWM kits have been used to test sensory registration in many studies involving children with CP (Cooper *et al.*, 1995; Gordon & Duff, 1999; Krumlinde-Sundholm & Eliasson, 2002; Arnould *et al.*, 2007; Auld *et al.*, 2012b), and as noted earlier, the clinimetric review of tactile assessments for children with CP by Auld *et al.* (2011) recommended the use of SWM for tactile registration.

Each filament (typically made from nylon) is numbered and is sequentially colour coded (from green to red/red-lined) corresponding to a particular filament thickness or diameter, with 'green' being the smallest diameter (least stiff) filament and 'red' or 'red-lined' being the largest diameter (stiffest) filament. Consequently, when the filament is applied normal to the skin surface in the prescribed manner, it will bow or bend when it reaches its maximum stiffness or peak force (Bell-Krotoski *et al.*, 1993), as shown in Figure 3-2. The number assigned to each filament (Table 3, 'Filament Size') represents the logarithm of 10 multiplied by the force in milligrams that is required to bow the filament (Tubiana *et al.*, 1996), meaning the stiffness of the filament is directly proportional to the diameter of the filament, which correlates to an applied force. The subject being assessed has their vision occluded and is asked to respond with 'yes' (or through a gesture if they are non-verbal) to indicate they felt an applied stimulus.

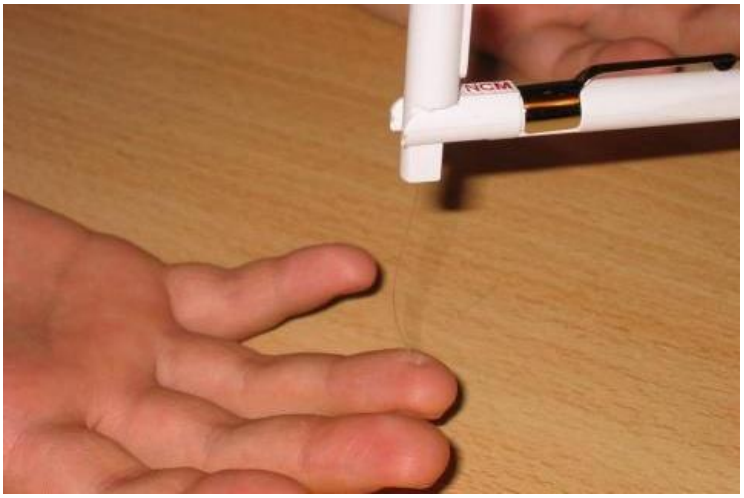

Figure 3-2 – A SWM being applied to the pad of the distal phalanx of the fourth digit on the right hand (Source: http://www.gms-books.de/book/living-textbook-hand-surgery/chapter/nerve-injury-classification-clinical-assessment)

SWM typically come in two forms – a 'mini kit' form that contains five monofilaments and a 'full kit' that contains 20 monofilaments. For Stage 1 of this study a five-filament test kit was used whereas a full 20-filament test kit was used for Stage 2. The decision to use a five-filament test kit for Stage 1 was due to the fact that Stage 1 was a screening test for entry into the intervention that was Stage 2. The literature reports that the overall number of monofilaments can be reduced (from twenty to five) with no significant loss in sensitivity for the test overall as the variation in the range of force for one filament may overlap with its nearest neighbour within the 20-filament kit (Tubiana *et al.*, 1996). A 20-filament kit was used for Stage 2 in an attempt to record a deeper sense of tactile registration for this particular cohort. For this test children were required to wear a blindfold, which was well tolerated. Table 3 provides an overview of the five-filament test kit, identifying filament colour, size, target force, diameter and the associated clinical threshold sensation meanings.

Table 3 – An overview of the five-filament SWM mini-kit (adapted from the Operation Manual, pgs. 4 and 5)

Filament Colour	***Filament Size***	***Target Force (g)***	***Filament Diameter (mm)***	***Hand Threshold***
Green	2.83	0.07	0.127	Normal
Blue	3.61	0.41	0.178	Diminished Light Touch
Purple	4.31	2	0.305	Diminished Protective Sensation
Red	4.56	4	0.356	Loss of Protective Sensation
Red-lined	6.65	300	1.143	Deep Pressure Sensation Only

Notes: g = grams; mm = millimetres.

Auld *et al.*'s clinimetric review reported that SWM demonstrated high content validity, adequate criterion validity, and consistently high intra-rater reliability for different observers, but that inter-rater reliability and test-retest reliability varied across studies and populations (pg. 422). Extensive research by Bell-Krotoski *et al.* (1995) identified that the 2.83 filament was a good predictor of 'normal' light touch recognition for the hands.

3.3.1.1 SWM Procedure and Scoring

The standard instructions as per the SWM Operation Manual were followed for this study. Tactile registration was assessed as being intact for a particular filament size when the child correctly identified three stimuli from three consecutive applications. That is, if a child identified two or less stimuli from three applications then the next largest filament in the sequence would be applied three times, until three correct responses were received for three applications of a given filament. The filament size that was correctly identified three times following three applications was recorded.

3.3.2 Two Point Discrimination (TPD)

TPD, also a motor-free assessment, is the ability to determine that two nearby objects that are touching the skin are in fact two distinct points of contact rather than one. The test can be static or dynamic (also called 'moving two-point discrimination' or MTPD) and is conducted with vision occluded. Within a 'normal' population, Louis *et al.* (1984) reported that moving TPD is a more sensitive test than stationary TPD, meaning that two points can be distinguished at a smaller distance when moving compared to when they are stationary.

The CP literature reports a number of ways to assess TPD, such as using the points of a divider (Hohman *et al.*, 1958; Wigfield, 1966); lead-point calipers (Tachdjian & Minear, 1958); lead points of a compass (Kenney, 1963); modified Vernier Calipers (Wilson & Wilson, 1967a); Weber's scissors (Lesný, 1971); a calibrated (Finnell *et al.*, 2004) or bent (Bolanos *et al.*, 1989) paperclip, or a device such as an Aesthesiometer® (Klingels, Demeyere*, et al.*, 2012) or Disk-Criminator® (Yekutiel *et al.*, 1994; Cooper *et al.*, 1995; Gordon & Duff, 1999; Klingels *et al.*, 2010; Auld *et al.*, 2012b).

A TPD study of 112 typically developing children aged two to 13 years by Cope and Antony (1992) reported 'normal' values that ranged from 2 – 5mm for the fingertip, with an average value of 2.7 ± 0.7mm for the left hand and 2.6 ± 0.7mm for the right (pg. 252). These values are comparable with those reported by Louis *et al.* (1984), another study of 'normal' TPD in subjects aged four to 92 years, as well as 'normal' values reported by Vierordt and cited by Lesný (1971)(pg. 331). Tubiana *et al.* (1996)

cites ‘normal’ TPD being less than 6mm, referencing the ‘American Society for Surgery of the Hand Clinical Assessment Recommendations’ (pg. 350), adding that the threshold criteria is correctly guessing seven from ten stimuli. The 70% success rate is also noted by Auld *et al.* (2011), but for distances 3 – 5mm (pg. 422).

Within the CP literature, differing values have been used to classify ‘normal’ TPD. Uvebrant (1988) evaluated TPD as ‘good’ for distances between 0 – 4mm, ‘moderately impaired’ for distances between 5 – 7mm, and ‘poor’ for distances greater than 8mm. Bolanos *et al.* (1989) tested TPD using a distance of 5mm, Van Heest *et al.* (1993) used a distance of 6mm, while Krumlinde-Sundholm and Eliasson (2002) tested TPD at 3 and 7mm. In another study, Eliasson *et al.* (2009) considered TPD to be ‘good’ at 3mm, ‘reduced’ if 7mm of spacing was discriminated, and ‘poor’ if 7mm of spacing was not able to be discriminated. Similarly, Holmström *et al.* (2010) used classified ‘normal’ TPD for distances up to 3mm, ‘decreased’ at 5mm, and ‘poor’ or ‘not able to discriminate’ for distances of 5 – 7mm. Sanger and Kukke used Johnson–Van Boven–Phillips or ‘JVP’ domes instead of traditional TPD methods for their study, citing that the domes method was more sensitive to spatial cues (Sanger & Kukke, 2007). Their testing protocol involved using a series of the plastic domes with ridges spaced at 3.0, 2.0, 1.5, 1.2, 1.0, and 0.75mm apart.

Wingert *et al.* (2008) highlighted some of the limitations of TPD, including the variation associated with the applied application force (between testers and across trials) and the issue with stimulation pressure being non-synchronous when applied, citing the work of Lundborg and Rosen (2004). Lundborg and Rosen emphasised that lack of standardisation with respect to TPD testing, and stated that all assessments should be accompanied by a detailed description of how the test was administered. From an equipment perspective, Finnel and colleagues found no statistically significant difference between instruments used to measure static TPD with healthy individuals aged 18-59 years, when using properly calibrated paper clips compared to the Disk-Criminator® (Finnell *et al.*, 2004).

Auld *et al.* (2011)’s clinimetric review reported that the Disk-Criminator® had criterion validity (high correlation with object recognition), construct validity (high correlation with accuracy and time to recognise an object) and high reliability, and that a static paperclip had a high correlation with position sense (pgs. 422, 426). The authors

also reported adequate intra-rater, inter-rater, and test-re-test reliability, but that the results varied from excellent to poor across methodologies (pg. 426).

The device used to assess TPD for this study was the AsTex® device (Australian Patent No. 2008229741). The AsTex® was invented at The University of Melbourne as a tool for measuring tactile sensitivity quickly, accurately and repeatedly (Miller *et al.*, 2009). Miller and colleagues developed the AsTex® to measure and evaluate hand sensation in adult's post-stroke, reporting excellent test-retest (ICC = 0.98) and inter-rater reliability (ICC = 0.81) in neurologically 'normal' subjects (Miller *et al.*, 2009). It was used for the first time with a paediatric cohort (typically developing children and children with CP) by Auld *et al.* (2012b), with 'normal' values reported in section 3.3.7. In terms of the device itself, unlike the Disk-Criminator® or a modified paper clip, the AsTex® is a Perspex rectangular slab measuring 390 x 100mm that has a central strip of laser cut grooves with parallel vertical ridges that logarithmically decline in width from 2.50 to 0.21mm along its centre length (Miller *et al.*, 2009), as shown in Figure 3-3.

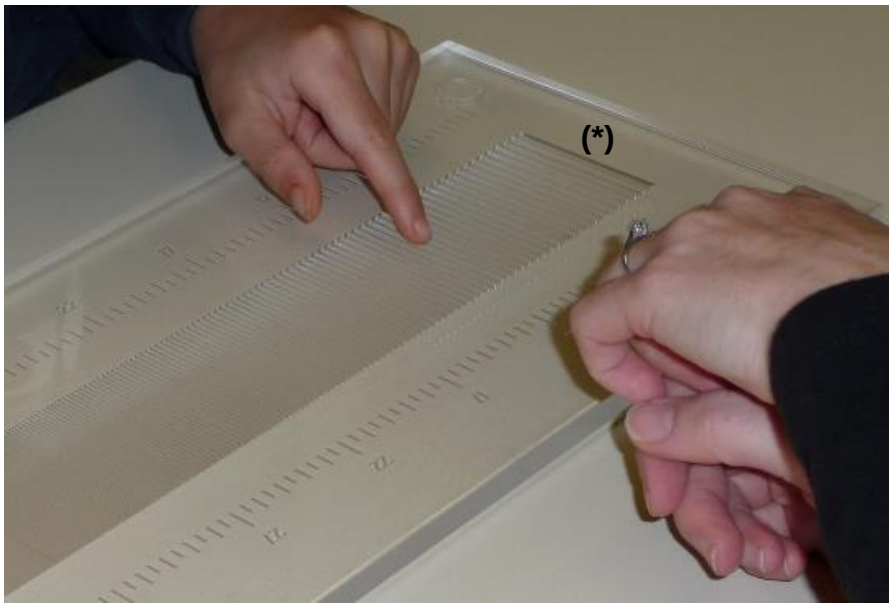

Figure 3-3 – One of the trial participants familiarising themselves with the AsTex® device before performing the test of TPD. The asterisk (*) indicates the 'rough' or coarse end of the device

3.3.2.1 TPD Procedure and Scoring

With vision occluded, children were seated in front of the AsTex® with the distal pad of their index finger on the end of the AsTex® where the vertical grooves are widest and furthest apart (the 'rough' or coarse end). They were then asked to slowly slide their finger across the grooves, medially to laterally, stopping their finger when the surface texture feels "smooth". This procedure was repeated three times and the mean of the three values obtained was converted to a measure of the texture discrimination index (TDI) in millimetres for that finger using the table provided in the '*Procedure for Sensory Assessment of the Hands using the AsTex*®', from Dr Kimberly Miller and Prof Mary Galea. A familiarisation trial is allowed prior to the assessment being conducted. The start (0cm) of the grooved central strip represented a TDI of 2.50mm, the halfway point (18.5cm) represented a TDI of 1.43mm, and the end (37.0cm) represented a TDI of 0.21mm. Consequently, the resultant score for this test was a decimal number between zero and 37 per hand, which was then converted to the nearest TDI value using the table provided.

Due to prior experiences that one of the supervisors (SH) had with the AsTex® device from another Adelaide-based study with typically developing university students (Causby, 2016), there was concern with respect to how the formal instructions for the test, as per the manual, would be interpreted and understood by children. Consequently, two tests of TPD using the AsTex® device were conducted, with Test 1 using the official instruction: "*Stop your finger at the point where the strip "feels smooth" and hold your finger in that position until I can record the value*", compared to Test 2, which used the following instruction: "*Stop your finger at the point where you can't feel individual lines anymore and hold your finger in that position until I can record the value*".

For Stage 1 the two AsTex® tests were conducted one after the other, in the order described above (section 3.3). For Stage 2, the two tests were separated in time to discern if the instruction given to each child influenced the way they reported TPD, without the possibility of a learning effect with the device contaminating the results. That is, the first AsTex® test was Test 2 in the protocol and the second AsTex® test was Test 6.

3.3.3 Proprioception

Proprioception is the ability to sense the position and motion of one's limbs and the relative position of other neighbouring body parts in space (Purves *et al.*, 2001). With respect to hand assessment, Tubiana *et al.* cite Omer (1981)'s description of proprioception as the ability to identify both the positional and directional change in finger movements when passive interphalangeal joint movements are made. As noted in a recent systematic review of the assessment, proprioception has had an inconsistent application and use within the literature and represents a complex range of inputs involving both the peripheral and central nervous systems related to joint position, joint range of motion, and force specification (Hillier, Immink, & Thewlis, 2015). The review identified the *Rivermead Assessment of Somatosensory Perception*, which contains within it a test of distal proprioception of the thumb, as being the most valid and reliable tool, but with low precision due to the binary nature of the scoring and possible tester manipulation influence. By clinical convention, the thumb is usually tested because it is the most important digit with respect to opposable actions of the hand (grip, grasp, pinch, etc), so directly relatable to hand use and fine motor control.

3.3.3.1 Proprioception Procedure and Scoring

To conduct this test, the assessing therapist first stabilised the child's thumb at the proximal joint and gently held the distal phalanx on the lateral edges, as shown in Figure 3-4. The therapist demonstrated that they were passively moving the thumb (distal interphalangeal joint) either up (extension) or down (flexion). Care was taken not to apply pressure on the flexion or extension surfaces to avoid triggering pressure detection. With vision occluded, the child was asked to detect when the thumb was moved and to discriminate whether it was being paused in an 'up' or 'down' position.

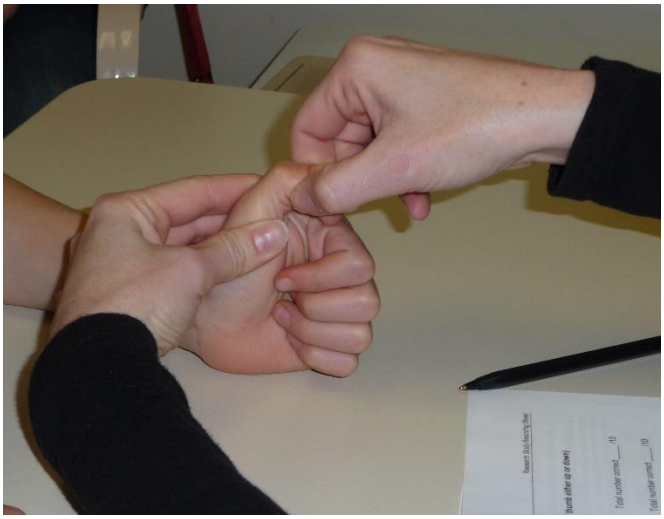

Figure 3-4 – A therapist performing the test of proprioception with a child's left thumb during a Stage 1 assessment

For this assessment ten movements were performed in a random sequence and with random timing. Accuracy of discrimination was recorded as one for identifying the thumb orientation correctly, and zero for an incorrect response. The range of possible scores for this test were integer values between zero and 10 per hand.

3.3.4 Stereognosis

Irving (1968) defined stereognosis as the ability to recognise objects using only tactile sensation (pg. 23), without the use of other cues such as vision or hearing. In the literature it has been referred to as "motor enhanced tactile perception" (Auld *et al.*, 2012a) or 'haptic perception' because it involves a combination of motor (manipulation) and tactile skills and abilities to correctly identify the form and hence the object within the hand. In simple terms, stereognosis is the ability to put your hand into your pocket or handbag without looking to identify your car keys when other objects such as loose change or a pen are also present, for example. Irving (1968) stated that astereognosis is associated with an injury to the parietal lobe, and that stereognosis ability was "*clearly dependent on an intact post-central gyrus*" (pg. 24).

As noted earlier, in terms of clinimetric validity, Auld *et al.* (2011)'s clinimetric review compared Klingels' protocol (Klingels *et al.*, 2010) with that of Cooper's (Cooper *et*

al., 1995), recommending Klingels' protocol as the preferred test for stereognosis because it had better clinical utility and in-depth clinimetric support (Auld *et al.*, 2011). Auld *et al.* reported adequate inter-rater reliability (ICC = 0.78) and excellent test-retest reliability (ICC = 0.86) for Klingels *et al.* (2010) method. The literature review presented earlier identified that stereognosis of common objects was more valid than stereognosis of forms.

Klingels' protocol involved using 12 different objects that represented two different groups of six objects, which is almost identical to the protocol used by Van Heest *et al.* (1993). The first group of six objects was designed to be dissimilar or clearly different from each other, and Klingels' object list was replicated for this study. The dissimilar stereognosis objects were a key, a clothespin, a marble, a comb, a spoon and a ball, as shown in Figure 3-5(a). The second group of six objects was represented by three pairs of objects similar to each other in size and in shape, and again Klingels' object list was replicated for this study. The group of similar objects were a pencil and a pen, a coin and a button, and a paperclip and a safety pin, as shown in Figure 3-5(b). The second group of similar objects presented a more challenging test as it required the ability to detect subtle form differences.

(a) **(b)**

Figure 3-5 – The two groups of six objects that were used for the test of stereognosis as per Klingels *et al.* (2010)'s protocol, showing (a) the dissimilar group of six objects and (b) the similar group

3.3.4.1 Stereognosis Procedure and Scoring

For each test of stereognosis, six objects were selected for each hand, three from the first group and three from the second. The objects were randomly selected by the assessing therapist, either prior to or during the test session, and offered to the child with their vision occluded. Like other researchers, Bolanos and colleagues recognised that stereognosis was also a test of the child's cognition and their verbal abilities – the ability to recognise the object being handled and then to be able to name it (Bolanos *et al.*, 1989). Consequently, prior to the test beginning the 12 objects were shown to the child and during the test a laminated sheet of A4 paper showing all 12 objects was placed in front of the child so they could point to an object to name it if they were non-verbal, similar to Jones (1960).

Rather than use a blindfold for this test a custom stereognosis testing box was created by the author using a cardboard box. A cut out and material drape was made on one side (enabling the child to insert their hand into the box while not being able to see into the box due to the material drape) and a fold down flap on the opposite side (for the therapist to place the object in the child's hand), as shown in Figure 3-6. As per the Klingels *et al.*'s guidelines, the child's ND limb was always tested first and both hands were tested using a random presentation of the 12 objects. For each presentation a score of one was given if the object was correctly identified and zero if the object could not be identified. The range of possible scores for this test were integer values between zero and six per hand. Petersen *et al.* (2016)'s study used a clinically significant change in stereognosis score being an improvement in correctly identifying two or more objects.

Figure 3-6 – The custom made cardboard box used for the test of stereognosis. The child would put their hand in the box through a cut out that was covered with a drape on one side, and the therapist would lower the flap on their side and put the object in the child's hand

3.3.5 The Jebsen-Taylor Hand Function Test

The Jebsen-Taylor Hand Function Test (JTHFT), sometimes shortened to the 'Jebsen-Taylor' or Jebsen Hand Function Test, is a series of seven unilateral, timed, multi-task standardised evaluation measures of functional hand motor skills, as described in Table 4. The test is designed to provide a short, objective test of hand function commonly used in activities of daily living. The test was developed by Jebsen and colleagues (Jebsen *et al.*, 1969) to assess and evaluate adults with neurologic or musculoskeletal conditions involving the hands (Mercuri, Fedrizzi, & Cioni, 2011).

Table 4 – The seven tests of the Jebsen Taylor Test of Hand Function (JTHFT)

Test Number	*Test details*
1	Writing a short sentence (24 letters, 3rd grade reading difficulty)
2	Turning over five 3 x 5 inch index cards
3	Picking up small common objects (two paper clips, two regular sized bottle caps, and two 1c coins) and placing them inside a can
4	Simulated feeding – scooping up five kidney beans using a teaspoon and placing them inside a can
5	Stacking four standard sized checkers on top of each other
6	Picking up and placing five large, light cans
7	Picking up and placing five large, heavy cans

Taylor, Sand, and Jebsen (1973) conducted the original test of the JTHFT with typically developing children aged six to 19 years, using an un-modified version of the adult JTHFT. Taylor *et al.* observed that the writing task was difficult and hence excluded this particular test when considering the youngest cohort (pg. 130). The authors also noted that overall test completion times decreased with increasing age (pg. 131), that females were statistically faster than males (pg. 132), and that a practice effect was not observed when statistically tested with a cohort of children with a stable hand impairment (n=20), including children with CP (n=11)(pg. 133).

Reedman *et al.* (2015) also noted a statistically significant difference between males and females, but only for nine year olds (again, with females being faster than males), for their cohort (pg. 296). Reedman *et al.* (2015) tested their data across discrete age groups to discern the development of manual dexterity with increasing age, identifying that five year olds were significantly different to all other age groups so excluded them from further analysis (pg. 296), but found no statistically significant differences between the older age groups for the total test score for either hand (pg. 296), allowing for the pooling of data. The final cohort ended up being 71 children aged between six years, zero months and 10 years, 11 months, with the authors concluding that the total score for either hand (dominant or ND) in typically developing children for the JTHFT had good test-retest reliability (pg. 298), and that

the Minimal Clinically Important Difference (MCID) was 5.09 seconds for the dominant hand and 5.87 seconds for the ND hand (pg. 300). Thus, the authors state that changes in the total test score for the JTHFT of at least those values could be attributed to an intervention (pg. 301). However, it is unclear how this outcome relates to children with a neurological impairment such as CP given the heterogeneous nature of the condition, and the fact that the smallest error measurement and the smallest real difference, which are used to calculate the MCID, will both be greater for an impaired population.

From an ICF perspective (WHO, 2001), the JTHFT measures capacity in the domain of 'activity' (Mercuri *et al.*, 2011) and has been used with children with CP in previous studies (Gordon & Duff, 1999; Eliasson *et al.*, 2009; Kinnucan *et al.*, 2010; Gordon *et al.*, 2011; Auld *et al.*, 2012a; Fong *et al.*, 2013), with Eliasson *et al.* (2009) observing that the test-retest data for the JTHFT was fairly strong (pg. 316), citing the early work of Taylor *et al.* (1973). To reduce the possibility of bias an official JTHFT kit was purchased to ensure the correct items (in terms of object size, mass and material) were being used, which was highlighted in the literature as a crucial consideration by Reedman *et al.* (2015)(pg. 301).

3.3.5.1 Jebsen-Taylor Hand Function Test Procedure and Scoring

The seven tasks (Table 4) are conducted with each hand separately while being timed by the administering therapist. Training is not required and the ND hand is always tested first, followed by the dominant hand, meaning a score in seconds is obtained for each hand. As identified by Mercuri *et al.*, for studies involving children with CP the maximum time allowed for each test is two minutes (120 seconds) to reduce frustration for the child, as per a recent study by Rich *et al.* (2017), and the hand writing/copying of text (Test 1) is typically omitted (Mercuri *et al.*, 2011). Reedman *et al.* (2015) noted that the hand writing task was typically omitted for children younger than eight years and zero months, and that only the dominant hand was tested when this test was conducted (pg. 295).

These two modifications to the traditional JTHFT procedures (limiting each test to 120 seconds and omitting Test 1) were implemented for this study for all children for

consistency and the ability to compare results to other CP studies. Figure 3-7 shows one of the trial participants conducting the JTHFT. Being a timed activity involving six different tasks, the range of scores per hand and per test were integer values between zero and 120 seconds, meaning the total score per hand for all six tests were integer values between zero and 720 seconds per hand. All assessors familiarised themselves with the test by reading the Jebsen documentation, exploring the Jebsen kit, and watching online videos of the test being conducted to prepare for the testing process.

(a) **(b)**

Figure 3-7 – A child from the study conducting the JTHFT: (a) Test 3, picking up small common objects, such as bottle caps and coins; (b) Test 5, stacking checkers

3.3.6 Assessment Tests Overview and Summary

The assessments that were used for this study are summarised in Table 5 to provide an overview of how each child was assessed.

Table 5 – A summary of the assessment outcome measures that were used during the study

Test	*Range of Results per Hand (min – max) (unit)*	*How Assessed / Scored*
SWM	2.83 – 6.65 (0.07 – 300g force)	Lowest filament value that was correctly identified three consecutive times
TPD (AsTex®), Test #1	0 – 37.0 (cm) = 2.5 – 0.21 TDI (mm)	AsTex® number (0 – 37) for which the lines "*feel smooth*"
TPD (AsTex®), Test #2	0 – 37.0 (cm) = 2.5 – 0.21 TDI (mm)	AsTex® number (0 – 37) for which the child "*can't feel individual lines anymore*"
Proprioception	0 – 10 responses	Number of times the child's thumb was correctly identified in a given orientation (up or down)
Stereognosis	0 – 6 items	Number of times an object presented was correctly identified
JTHFT	0 – 120 secs per test, 0 – 720 secs overall	Individual test time and total overall test time to complete the required tasks (6 tasks in total)

Notes: SWM = Semmes-Weinstein Monofilaments; TPD = two-point discrimination; TDI = texture discrimination index; JTHFT = Jebsen-Taylor Hand Function Test; min = minimum; max = maximum; g = grams; cm = centimetres; mm = millimetres; secs = seconds.

3.3.7 Normative Results and Cut-off Scores for each Sensory Test

Informed by the literature review, normative or cut-off results for each test were identified to determine what constituted an atypical or abnormal result. The term 'cut-off' in this context follows the definition provided by Bolanos *et al.*, as the upper limit of the number of incorrect answers expected from control or TDC patients, such that a response greater than this limit or value are not acceptable as normal (Bolanos *et al.*, 1989, pg. 373). Table 6 presents the normative or cut-off data from relevant CP studies, derived from an analysis of the data obtained from the control group for each study.

Table 6 – A summary of published normative and/or cut-off values for the four sensory tests used for this study

Test: ***Author:***	***SWM***	***TPD***	***Proprioception***	***Stereognosis***
Bolanos *et al.* (1989)	N/A	Cut-off > 2 incorrect responses @ 5mm	N/A	Cut-off > 1 incorrect response
Van Heest *et al.* (1993)	N/A	Intact only if all 5 correct responses @ 6mm	Intact only if all 5 trials correctly identified	Only intact if all objects correctly identified
Bell-Krotoski *et al.* (1995)	Normal if detect 2.83	N/A	N/A	N/A
Cooper *et al.* (1995)	Abnormal if threshold > 2.83	Cut-off value = 3mm for all fingers. Abnormal if ≥ 3mm	Cut-off value = 5 (out of 5). Scoring ≤ 4 was *abnormal*	Cut-off value = 9 (for 10 objects). Scoring ≤ 8 was *abnormal*
Arnould *et al.* (2007)	Controls: median scores < 2.83 for both hands	N/A	Controls: median and IQR = 10 (for 10 objects) for both hands	Controls: median and IQR = 10 (for 10 objects) for both hands
Kinnucan *et al.* (2010)	N/A	N/A	N/A	Only intact if all objects correctly identified
Klingels, Demeyere*, et al.* (2012)	N/A	Intact if TPD was ≤ 5mm	N/A	Only intact if all objects correctly identified
Auld *et al.* (2012b)	TDC: all median and IQR values were < 2.83 for both hands	TDC: AsTex® TDI index finger; median = 0.27mm (IQR = 0.21 – 0.64 for ND and 0.21 – 0.47 for dom hand)	N/A	TDC: median and IQR = 9 (for 9 objects) for both hands
Petersen *et al.* (2016)	N/A	N/A	N/A	Only intact if all objects correctly identified

Notes: N/A = information not provided or absent from article; TDC = typically developing children; IQR = interquartile range; SWM = Semmes-Weinstein Monofilaments; TPD = two-point discrimination; TDI = texture discrimination index; dom = dominant; ND = non-dominant.

Therefore, the following sensory thresholds were used to discern *abnormal* sensory function, per hand, based on the literature:

- For **SWM**, a pressure sensitivity threshold greater than 2.83 (green filament), as per Cooper *et al.* (1995), Bell-Krotoski *et al.* (1995), Arnould *et al.* (2007) and Auld *et al.* (2012b);
- For the **AsTex device (TPD)**, a value lower than 30.5cm on the device (or a TDI > 0.64mm) for the ND hand, and a value lower than 33.0cm (or a TDI > 0.47mm) for the dominant hand, as per Auld *et al.* (2012b);
- For **proprioception**, 1 incorrect response or more, so a score of 9 or less as per Van Heest *et al.* (1993), Cooper *et al.* (1995) and Arnould *et al.* (2007);
- For **stereognosis**, 1 incorrect response or more, so a score of 5 or less as per Van Heest *et al.* (1993), Arnould *et al.* (2007), Kinnucan *et al.* (2010), Klingels, Demeyere*, et al.* (2012), Auld *et al.* (2012b) and Petersen *et al.* (2016).

3.4 Quality of Life (QOL) and Upper Limb Manual Ability Questionnaires

In addition to the tests reported in section 3.3, two questionnaires were also administered during Stage 2, one to measure quality of life (QOL) and the other to measure upper limb manual ability. These are described in the following sections.

3.4.1 The Cerebral Palsy Quality of Life (CP QOL) Questionnaire

Apart from the sensory and motor outcome measures described earlier, it was important to measure any impact or effect that the intervention had on the general well-being of the child during and following the trial. The reason for assessing a study participant's well-being or quality of life pre- and post-trial was to account for and measure potential changes in self-perception following the intervention. Additionally, each CP QOL questionnaire includes specific questions relating to hand and arm use, and certain daily activities that relate to the upper limbs, which are reported in section 6.3.10.4.

The Cerebral Palsy Quality of Life (CP QOL) questionnaire was developed by a team from the University of Melbourne and the Royal Children's Hospital in Melbourne, and has evidence of internal and test-retest reliability, and strong psychometric properties and clinical utility (Carlon *et al.*, 2010). The CP QOL questionnaire is free to download after registration. The systematic review by Carlon *et al.* (2010) identified the CP QOL-Child questionnaire to have the highest quality assessment rating when compared to four other CP specific measures, as well as being the only measure that wholly fulfilled the criteria of QOL being measured across broad domains and being a measure of well-being (pg.6). However, like all measures of quality of life for children with CP, it lacks data on responsiveness or sensitivity to change (Carlon *et al.*, 2010). The tool was designed to assess QOL for children aged four to 18 years (hence, appropriate for this study) and uses four different questionnaires, categorised by the age of the child, as follows:

- A CP QOL-**Child** questionnaire for parents or the primary care giver to complete if their child is aged between 4 and 12 years;
- A CP QOL-**Child** questionnaire for the child to complete (self-report) if they are aged between 9 and 12 years;
- A CP QOL-**Teen** questionnaire for adolescents to complete (self-report) if they are aged between 13 and 18 years old;
- A corresponding CP QOL-**Teen** questionnaire for parents or the primary care giver to complete if their adolescent is aged between 13 and 18 years old.

All CP QOL instruments ask questions and assess the wellbeing of the child/teenager across various domains of life:

- The seven domains of the **Child** questionnaire are: 1 ‘Social wellbeing & acceptance’, 2 ‘Feelings about functioning’, 3 ‘Participation & physical health’, 4 ‘Emotional wellbeing & self-esteem’, 5 ‘Access to services’, 6 ‘Pain & impact of disability’ and 7 ‘Family health’.
- The **Teen** questionnaire has very similar domains of assessment, but is tailored for an older cohort. Most of the domains have similar headings except for the following, with the changes noted in italics: first (‘*General* wellbeing &

participation'), third ('*Communication* & physical health'), fourth ('*School wellbeing*'), and sixth ('*Social Wellbeing*').

With respect to the overall study design, the CP QOL questionnaires were administered during the A_1 (Stage 2, baseline measure) and A_2 (Stage 2, immediate post-trial measure) assessments. Most times the questionnaire was completed during the assessment, however a few parents requested to complete the questionnaire at home due to the time it took to complete the form. Completed questionnaires were returned via post.

3.4.1.1 Processing and Scoring the Cerebral Palsy Quality of Life (CP QOL) Questionnaire

From a data cleaning and processing perspective, all responses from the hard copy forms were translated into a custom Excel spreadsheet for data recording and manipulation, and every QOL score was double-checked by the author to ensure it was entered correctly. If a response was marked midway between two numbers, the higher of the two numbers was chosen (that is, a circle or mark between the numbers '3' and '4' was scored as '4'). Care was taken during scoring to ensure that the right questions from the form were grouped into the right domains as the questionnaires do not number each question, and that the correct re-coding process was followed (the coding process varies depending on the question being asked), based on the respective manual instructions (Davis E *et al.*, 2013; Waters E *et al.*, 2013). Issues from the trial related to the administration of the questionnaire are discussed in Chapter 6, section 6.3.10.1.

3.4.2 The ABILHAND-Kids Questionnaire

When identifying and choosing a tool from the literature to assess upper limb function the following criteria guided the selection of the most appropriate tool: (1) valid for children with CP; (2) accurately assess a child's ability to manage activities of daily living, (3) relatively quick to administer to reduce time burden on families; and (4) appropriate for children with a unilateral or bilateral involvement, meaning a single tool could be used for the mixed cohort from this study. The last criteria meant

that the AHA, which is used in many CP studies, could not be considered as it was developed for children with only a unilateral involvement.

The ABILHAND-Kids questionnaire[1] is a tool to measure the manual ability of children with an upper limb impairment. Developed by researchers from the Laboratory of Rehabilitation and Physical Medicine at the Université catholique de Louvain in Belgium, the questionnaire measures a child's ability to manage activities of daily living that require the use of both upper limbs, irrespective of the strategies involved. Klingels, Demeyere*, et al.* (2012) reported that the questionnaire had a high level of reliability and validity following the work of Arnould *et al.* (2004) with children with CP aged 6-15 years, with Bleyenheuft *et al.* (2017) also recognising that it had been 'calibrated' in this age range.

The questionnaire is completed via parent report and not by the child. Parents are asked to assess their perceived child's difficulty when assessing 21 items of daily living (such as opening a jar of jam or sharpening a pencil), as either 'Easy', 'Difficult' or 'Impossible'. A fourth option is available (a 'Question Mark') if the parent cannot estimate their child's ability to complete the activity because they have never done it. According to the instructions, activities that the child hasn't attempted in the last three months are not scored and entered as missing responses. The authors of the tool observed that parents were able to report a finer perception of their child's manual ability to complete a task than the child, with a higher reliability (R=0.94) and good reproducibility over time (R=0.91) (Arnould *et al.*, 2004). Furthermore, during the development of the functional scale (from 74 original items to the final 21) involving more than 100 children with CP and their families and experts, the item difficulty hierarchy was consistent between the parents and the experts. Additionally, the ABILHAND-Kids measures were found to be significantly related to school education (mainstream vs. special education program), type of CP (diplegia and hemiplegia vs. tetraplegia), and gross motor function (Arnould *et al.*, 2004). The questionnaire has been used in the literature with respect to CP sensory and motor assessment studies previously (Arnould *et al.*, 2007; Klingels, Demeyere*, et al.*, 2012; Zoccolillo *et al.*, 2015; Preston *et al.*, 2016; Bleyenheuft *et al.*, 2017).

[1] See: http://www.rehab-scales.org/abilhand-kids.html

As for the CP QOL questionnaire, the ABILHAND-Kids questionnaire was administered during the A_1 and A_2 assessments. For all children, parents completed this questionnaire during the assessment session. To avoid potential systematic effects when parents complete the form, ten different ABILHAND-Kids questionnaires with a random ordering of the activities to be rated have been developed. This meant parents completed different forms at the two time points of the study, with the exception of one family who was inadvertently given the same form on both occasions. However, given there was a period of six weeks between A_1 and A_2, a systematic effect for this particular child is unlikely.

3.4.2.1 Processing and Scoring the ABILHAND-Kids Questionnaire

As per the ABILHAND-Kids questionnaire test package instructions, each child's pre- and post-trial responses were entered into the online analysis section of the website[2], and a Rasch analysis was conducted. This enabled a pre- and post-trial comparison of the calculated 'Patient Measure' (in logits), where the more positive the measure the higher the child's manual ability.

3.5 Evaluation of the Gaming System by Families

A one-page custom form, called the '*Participant Experience Questionnaire*' was developed to evaluate the serious gaming intervention post-trial (Appendix B). It was designed to be completed simply and easily by the child and/or their parents at the completion of the trial when the author collected the system after six weeks. To reduce perceived bias due to the presence of the author when the system was being collected, the form was typically left with families along with a pre-paid envelope to return at their leisure. However, some families were happy to complete the form when the gaming system was being collected.

The form asked each child/family to rate the gaming system (out of ten) using a linear scale, asked for examples of positive or negative experiences during the trial,

[2] See: http://www.rehab-scales.org/abilhand-kids-rasch-analysis-cerebral-palsy.html

and asked if other therapy programs or interventions occurred during the trial that may affect the trial results (such as regular or irregular physiotherapy, occupational therapy or Botulinum toxin treatment).

3.6 Conclusion

This chapter has presented and discussed the assessments that were administered for this research, stating the rationale for choosing each test, providing evidence of the validity of each assessment item, and highlighting where they have been used with children with CP within the literature.

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4. Stage 1 – Determining the Prevalence of Upper Limb Sensory Impairments in Children with Cerebral Palsy

4.1 The Stage 1 Study

Since the aim of this PhD research was the development and trialling of an intervention for children with CP with a known upper limb somatosensory impairment, the first stage was to conduct a study to assess and quantify the sensory status of a cohort of children living with CP in the state of South Australia. As mentioned earlier, this was referred to as Stage 1 of the PhD research.

While South Australia keeps and maintains a CP Register (formally known as the *South Australian Cerebral Palsy Register* (SACPR), based at the WCH, the somatosensory status of children on the Register is not recorded. At the time this study began there was no published research from Australia on the prevalence or severity of somatosensory impairments for children with CP, until Auld and colleagues published their Queensland-based research in 2012 (Auld *et al.*, 2012b). Consequently, this research was an important starting point for the overall project, and represented the first time such a study occurred in South Australia.

4.1.1 Study Aims

The specific aims of the Stage 1 study were to:

1. Recruit and assess children living with CP in South Australia for upper limb somatosensory function;
2. Determine if the type of CP and the side of the lesion (including the resultant hand dominance) have an influence on the nature and extent of sensory impairment that is identified;
3. Determine the level of correlation between sensory impairment and level of activity (function) in the upper limb; and
4. To compare the sensory and motor performance of the dominant and non-dominant (ND) limbs of children with CP.

4.1.2 Study Hypotheses

The hypotheses for the study were that:

1. Sensation is impaired in the upper limb(s) of children with CP, compared to age-matched typically developing peers;
2. The type of CP and the site of the lesion influences the nature and extent of sensory impairment;
3. The level of sensory impairment correlates with the level of upper limb (dys)function; and
4. Sensory impairments will be more prominent on the ND side compared to the dominant side.

At the conclusion of Stage 1 a sub-group from the overall cohort would be identified as having a known somatosensory deficit compared to age-matched typically developing peers. Children belonging to this sub-group would then be invited to participate in Stage 2 of the PhD research (Chapter 6), which was a six week home-based intervention using the OrbIT Gaming System. At the time this research began there was a paucity of research activity that focussed on tactile sensory interventions for children with a known sensory loss.

4.1.3 Inclusion and Exclusion Criteria

Children living in South Australia with all types of CP who satisfied the following inclusion and exclusion criteria were invited to participate in the study via a direct mail out from the SACPR:

- A confirmed diagnosis of CP, hence registered on the SACPR;
- Aged five to 15 years and attending a mainstream school;
- No previous hand surgery;
- No Botulinum toxin A (BoNT-A) in the three months prior to assessment, which was also criteria used by Auld *et al.* (2012b), Wingert *et al.* (2008) and Klingels, Demeyere*, et al.* (2012), with the latter two studies requiring a six month BoNT-A free window; and
- The ability to follow and respond to verbal instructions.

4.1.4 Funding

A grant application for funding Stage 1 of the research was made to the Women's & Children's Hospital Foundation (WCHF) in June 2011, via a research project grant, for work to commence in 2012. The application underwent assessment and scientific review and was ranked 10th out of 39 applications. It was subsequently funded in full (AUD$45,342 + GST).

4.1.5 Ethics Approval

Prior to beginning the study, ethics approval was obtained from two Human Research Ethics Committees (HRECs) – the HREC from SA Health and the HREC from the Women's & Children's Health Network (WCHN). This was necessary because the sensory assessments were taking place on site at the WCH, and because the SA Health HREC has continuing oversight of the ethics of the activities of the Register.

Final ethics approval for the study was obtained from the WCHN HREC on the 21st December 2011 (protocol number REC2441/12/14) and SA Health on the 8th February 2012 (protocol number 480/11/2014).

4.1.6 Participant Recruitment

In early 2012, SACPR staff identified 262 children from the Register that had a confirmed diagnosis of CP and who were aged between five and 15 years of age (birth years 1997 – 2007). Working with SACPR staff, the author prepared and sent information packs to all eligible families containing: (i) a letter of introduction from the SACPR on behalf of the research team, (ii) a study information sheet and (iii) a consent form (see Appendix C). Information packs were mailed out in April 2012. The recruitment response and the study flow for Stage 1 is detailed in section 4.2.1. As described earlier (Chapter 3, section 3.3), the testing assessment represented a hierarchy of perceptual difficulty from a brain processing perspective, beginning with a test of tactile registration, followed by tests for discrimination/perception, proprioception, stereognosis, and a test of hand motor function.

4.1.7 Data Entry and Integrity Check

The assessing therapist recorded test results directly on to a hard copy Stage 1 assessment recording sheet during each session (Appendix A). At the conclusion of a session, data were transferred into a Microsoft Excel spreadsheet that was customised for the purposes of this study. The therapist who conducted the session entered the data or this was done by the author – for the latter case, the therapist would scan the assessment sheet and email it to the author.

Once all data were entered into a single Excel spreadsheet, every data point for all Stage 1 participants was crosschecked against the entry on the hard copy form. Nine transcription errors were identified from the 864 data points for Stage 1 (36 participants, each with 24 data points), representing a 1% error. Once all Excel data points matched the hard copy assessment sheets, the data were formatted and a meeting was arranged with the consulting statistician for the project to discuss the necessary statistical analyses.

4.1.8 Statistical Methods and Analysis

The statistical analysis used mixed effects modelling and focussed on the test performance between each hand (dominant and ND), within each group (unilateral and bilateral), as well as between each group. Standard measures of association (correlation) using simple linear regression and standard errors for repeat observations were also conducted to determine correlations between each sensory test and the total time taken for the JTHFT. Welch's Test for Unequal Variances (or Welch's t-test) was used to determine the mean and standard deviation for the individual tests of the JTHFT, which is a test that is recommended when two different samples have unequal variances as well as unequal sample sizes. For the test of tactile discrimination (using the AsTex® device), median and interquartile ranges were calculated for each hand and each group. Statistical significance was set to p=0.05 for all test. Data analyses were performed and modelled using SPSS (Version 23) or Microsoft Excel.

4.1.9 Manual Ability Classification System (MACS) Classification

During each assessment session the assessing therapist would assess and classify each child's MACS level. The MACS classification scale is used to classify, into one of five different classification levels, the way a child with CP uses both of their hands when handling objects in everyday activities (Eliasson *et al.*, 2006). Level I represents the most able hand function and is equivalent to hand function as per typically developing children, Level III represents a child that handles objects with difficult, slow performance, with limited success with respect to quality and movement, whereas Level V represents the poorest or most involved hand function, where a child is not able to handle objects or complete simple hand actions.

MACS is not an indication of a child's maximal hand ability but instead represents their typical hand performance, and classifies the collaborative use of both hands. When the system was developed it was tested on 168 children with CP aged four to 18 years old, and is appropriate for children with a unilateral or bilateral involvement. The authors report an intraclass correlation coefficient (ICC) for all ages as being 0.97 (0.96-0.98) (Eliasson *et al.*, 2006, pg. 552). MACS was shown to have a high and statistically significant Spearman correlation coefficient with the self-care domain of the *Pediatric Evaluation of Disability Inventory* (PEDI) in a study of 61 children aged five to 14 in a school setting (Kuijper *et al.*, 2010, pg. 617).

4.1.10 Participant Brain Scan Classification

Post each sensory assessment session, and in accordance with the ethics approval granted for the project, any previously recorded brain scan information (either magnetic resonance imaging (MRI) or computed tomography (CT) scan) for all children was accessed through the WCH patient records system, to determine and classify the type and nature of the brain lesion. Each scan was assessed and classified by one of the project supervisors (RR), who had undertaken specialised training with respect to brain lesion classification for children with CP. The classification was based on research by Krageloh-Mann and Horber (2007), which is summarised in Table 7.

Table 7 – The brain scan classification system used for the study

Number/Code	*Brain Imaging Classification (from Krageloh-Mann and Horber (2007))*
1	Brain mal-developments
2	Periventricular white matter injury
3	Cortical/subcortical lesions
4	Miscellaneous
5	Normal
999	Missing / not done / no imaging available

4.2 Results

4.2.1 Stage 1 Response and Study Flow

Following the mail out to 262 families, 49 (19% response rate) signed and returned the consent form for the study. Three additional families phoned the author to register their interest in the study, but noted that they did not believe that their child would be suitable for the study due to attention or behavioural reasons. After consent, four families were lost to follow up and three withdrew their child from the study, prior to attending an assessment session. As a result, 42 children attended a sensory assessment session at the WCH, as shown in Figure 4-1.

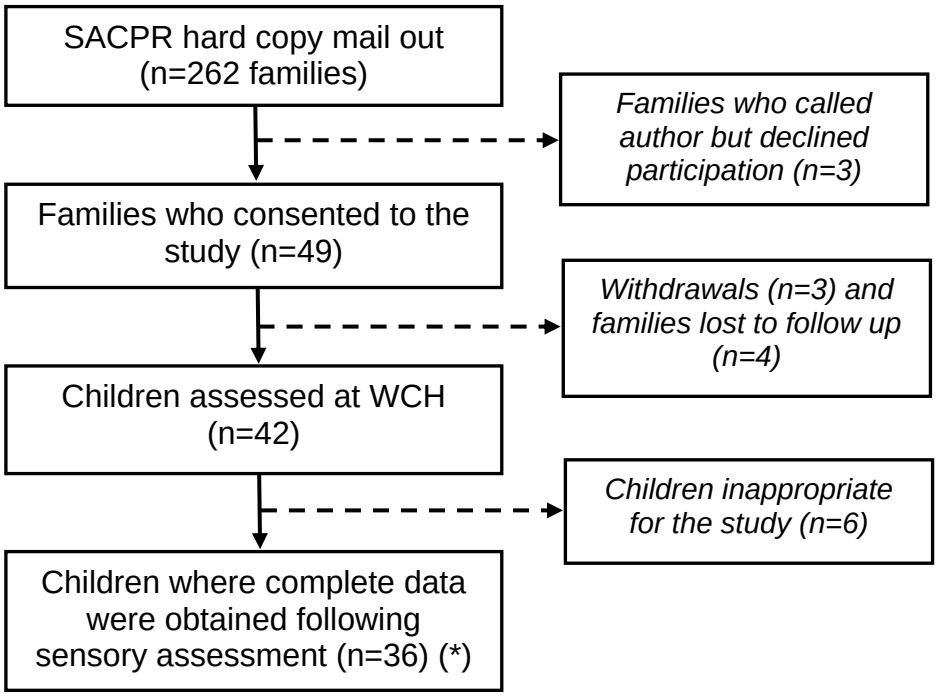


Figure 4-1 – A flow diagram indicating recruitment and assessment of children for the Stage 1 study[(*) participant #31 completed 22 of the 24 tests]

Three families withdrew their child from the study after consent due to feeling overcommitted with existing extracurricular family activities, and four failed to respond to communication and could not be contacted post-consent. Following study consent and prior to the child attending the WCH for their sensory assessment, the assessing therapist checked each child's WCH patient record to verify the child's date of birth, CP classification (to identify if the child had a unilateral or bilateral

involvement), and to record each child's ND side. The first assessment occurred in late May 2012 and the last assessment occurred in early April 2014. The window of assessment remained open for this length of time to increase recruitment to the study.

From the 42 children who were assessed, six children were excluded from further analysis for reasons outlined in Table 8. Consequently, complete data were obtained for 36 children, with the exception of one child (#31) who refused to conduct the second AsTex® test but completed all other tests (as shown in Table 11). This meant that 22 of the possible 24 test results were assessed for this particular child.

Table 8 – Reasons for excluding participants from Stage 1 and the characteristics of each child (age, CP type, MACS Level and brain scan classification)

Number Excluded	***Reason for Exclusion***	***Child Profile (# = participant number)***
3	Behavioural; uncooperative and/or refused to perform the required tasks or respond to instructions	• 5.0 year old male, unilateral CP, MACS III, scan = cortical / subcortical lesions (#36) • 4.8 year old male, unilateral CP, MACS II, scan = normal (#26) • 7.7 year old female, unilateral CP, MACS III, cortical / subcortical lesions (#23)
2	Inconsistent responses and an inability to perform the required tasks	• 5.3 year old male, diplegic CP, MACS V, scan = miscellaneous (#3) • 13.0 year old male, diplegic CP and high tone, MACS V, scan = cortical / subcortical lesions (#22)
1	Physical/anatomical – the child did not have a left hand	• 9.2 year old male, diplegic CP, MACS II, no scan information available (#14)

Notes: MACS = Manual Ability Classification System; CP = cerebral palsy; (#X) = participant number X.

4.2.2 Stage 1 Cohort Overview

Table 9 provides an overview of the Stage 1 cohort of 36 children. Most participants were male (n=22, 61%); approximately two-thirds of the cohort were classified as having a unilateral involvement (n=23, 64%); most had a left side involvement/right side brain injury (n=23, 64%); and most were classified as MACS Level II (n=19, 53%). Most brain scans were classified as having cortical/subcortical lesions (n=13, 36%) or periventricular white matter injury (n=9, 25%), with missing or no scan available for 10 children (28%). Twenty four children (67%) were aged eight years of age or older, and 12 children (33%) were aged less than eight years old. While children across all five MACS Levels were assessed, both children classified as MACS Level V were excluded from the study due to inconsistent responses during the assessment sessions and an inability to perform the required tasks (Table 8).

Table 9 – An overview of all Stage 1 participants (n=36)

Age (years ± SD)	10 ± 3.3
Sex	**22 males (61%)**
ND side (Left : Right)	**23** : 13
MACS Level (n)	I(9); **II(19)**; III(5); IV(3); V(0)
CP type (unilateral : bilateral)	**23** : 13
Brain imaging classification (n) (Table 7)	Cortical/subcortical lesions (13); missing or no information (10); periventricular white matter injury (9); brain maldevelopments (2); miscellaneous (1); normal (1)

Notes: **bold font** indicates predominant classification; ND = non-dominant; SD = standard deviation; MACS = Manual Ability Classification System.

Table 10 provides an overview of the Stage 1 cohort based on CP classification, providing a breakdown of the unilateral sub-cohort vs. the bilateral sub-cohort.

Table 10 – An overview of all Stage 1 participants, by CP classification

	Unilateral (n=23)	***Bilateral (n=13)***
Age (years ± SD)	9.7 ± 3.1	10.5 ± 3.9
Sex	13 males	**9 males**
ND side (Left : Right)	13 : 10	**10** : 3
MACS Level (n)	I(3); **II(14)**; III(4); IV(2); V(0)	I(6); II(5); III(1); IV(1); V(0)
Brain imaging classification (n) (Table 7)	**Cortical/subcortical lesions (12)**; periventricular white matter injury (4); missing or no information (4); brain maldevelopments (2); miscellaneous (1)	Missing or no information (6); periventricular white matter injury (5); cortical/subcortical lesions (1); normal (1)

Notes: **bold font** indicates predominant classification; ND = non-dominant; SD = standard deviation; MACS = Manual Ability Classification System.

4.2.3 Stage 1 Sensory and Motor Assessment Results

The raw data for all Stage 1 assessments appear in Table 11, grouped by CP classification, and then by participant number. Reading the table from left to right, the columns indicate the test order, representing a hierarchy of perceptual difficulty from a brain processing perspective as described earlier (Chapter 3, section 3.3).

The following notes explain the acronyms and abbreviations used in Table 11.

Notes: No. = number; F = female; M = male; R = right; L = left; yrs = years; mths = months; Uni = unilateral; Bi = bilateral; CP = cerebral palsy; Ax = assessment; MACS = Manual Ability Classification System; ND = non-dominant; Dom = dominant; ***X*** = refused test; Nil = no result / threshold not detected; TX = test number X; SWM = Semmes Weinstein Monofilament; (f) = index finger; (th) = thumb; Brain Scan as per Table 7.

Table 11 – Sensory and motor assessment results for the Stage 1 cohort (n=36), grouped by CP classification

Subject No.	Gender	Age at Ax	CP Type	ND side	MACS Level	Brain Scan	SWM				AsTex®				Proprioception		Stereognosis		Jebsen Taylor Hand Function Test (Tests 2-7)(seconds)												Total (ND)	Total (Dom)
							ND(f)	Dom(f)	ND(th)	Dom(th)	T1, ND	T1, Dom	T2, ND	T2, Dom	ND	Dom	ND	Dom	T2, ND	T2, Dom	T3, ND	T3, Dom	T4, ND	T4, Dom	T5, ND	T5, Dom	T6, ND	T6, Dom	T7, ND	T7, Dom		
1	F	10 yrs 11 mths	Uni	R	II	3	Nil	2.83	Nil	2.83	18.5	36.6	21.8	32.0	3	10	2	5	120	7	120	12	120	24	120	15	120	4	120	6	720	68
2	F	8 yrs 5 mths	Uni	L	II	3	2.83	2.83	2.83	2.83	24.5	30.8	25.2	28.3	10	10	6	5	120	4	120	6	120	9	120	5	120	3	120	4	720	31
4	M	8 yrs 2 mths	Uni	L	III	3	Nil	2.83	Nil	2.83	19.7	25.3	25.3	35.7	4	9	0	6	120	6	120	9	120	12	120	5	120	5	120	6	720	43
5	M	7 yrs 7 mths	Uni	R	II	999	2.83	2.83	2.83	2.83	33.3	30.7	31.0	32.5	8	10	4	6	18	8	18	9	32	13	13	5	11	5	9	5	101	45
6	M	14 yrs 6 mths	Uni	R	IV	3	2.83	2.83	2.83	2.83	25.7	28.0	35.7	35.0	9	10	4	6	19	4	120	7	120	10	23	5	12	4	18	5	312	35
11	F	5 yrs 9 mths	Uni	R	II	1	2.83	2.83	2.83	2.83	33.0	36.7	37.0	37.0	10	10	2	6	18	5	71	9	120	12	72	5	9	4	11	5	301	40
12	M	12 yrs 8 mths	Uni	R	II	3	2.83	2.83	4.56	2.83	33.7	33.3	36.7	37.0	7	10	3	6	15	5	15	6	30	9	13	4	8	4	8	4	89	32
16	M	6 yrs 1 mth	Uni	R	I	2	2.83	2.83	2.83	2.83	34.7	32.7	37.0	37.0	10	10	6	6	6	9	7	8	44	19	8	6	7	6	11	9	83	57
25	M	12 yrs 1 mth	Uni	L	II	999	2.83	2.83	2.83	2.83	33.5	30.3	35.3	37.0	10	10	5	5	24	5	57	10	84	12	18	5	23	4	20	4	226	40
27	F	9 yrs 6 mths	Uni	L	II	2	2.83	2.83	2.83	2.83	28.7	34.7	31.0	29.0	8	10	5	6	13	8	10	17	25	18	11	7	9	6	7	6	75	62
29	M	8 yrs 7 mths	Uni	L	IV	1	2.83	2.83	2.83	2.83	27.3	25.3	32.3	34.0	3	4	4	2	11	5	22	21	80	120	21	6	25	12	120	7	279	171
30	F	14 yrs 9 mths	Uni	R	III	3	2.83	2.83	2.83	2.83	22.3	30.7	32.7	36.0	10	10	4	5	48	4	44	6	120	13	106	4	10	2	120	3	448	32
31	M	6 yrs 2 mths	Uni	L	III	999	2.83	2.83	2.83	2.83	29.0	36.0	***X***	***X***	6	9	0	6	27	9	86	13	120	17	120	15	120	5	120	8	593	67
32	M	7 yrs 6 mths	Uni	L	II	3	Nil	3.61	Nil	2.83	18.0	26.0	7.0	30.0	8	8	1	5	120	8	120	11	120	19	120	9	120	6	120	6	720	59
33	F	13 yrs 4 mths	Uni	R	II	3	2.83	2.83	2.83	2.83	36.3	33.0	26.7	25.7	6	10	6	6	18	3	42	7	41	12	40	3	52	3	12	3	205	31
37	M	12 yrs 11 mths	Uni	L	II	4	2.83	2.83	2.83	2.83	35.7	37	36	35.3	3	10	1	6	24	3	59	5	120	10	9	2	11	3	7	3	230	26
38	M	6 yrs 5 mths	Uni	L	II	2	2.83	2.83	2.83	2.83	23.3	26.7	31.0	34.7	10	9	4	6	120	4	120	6	120	36	38	2	23	3	120	5	541	56
41	F	6 yrs	Uni	L	I	3	2.83	2.83	2.83	2.83	24.7	15.3	34.7	36.3	10	10	5	5	8	4	18	6	120	33	8	3	6	2	6	3	166	51
42	F	8 yrs	Uni	L	II	3	3.61	2.83	4.31	2.83	27.0	32.2	29.0	34.7	6	10	1	6	120	6	120	7	120	26	120	3	120	4	120	4	720	50
43	F	15 yrs 11 mths	Uni	R	I	3	2.83	2.83	2.83	2.83	28.2	30.0	27.3	26.8	10	10	6	6	4	3	8	5	26	8	4	1	3	2	4	2	49	21
44	M	11 yrs 2 mths	Uni	L	II	2	2.83	2.83	2.83	2.83	24.8	24.8	24.8	28.5	10	10	6	6	6	5	11	10	120	120	3	2	7	5	7	5	154	147
46	M	9 yrs 1 mth	Uni	L	II	3	2.83	2.83	2.83	2.83	26.7	15.7	33.3	10.7	9	10	4	5	11	4	23	7	120	16	6	2	10	3	9	3	179	35
47	F	8 yrs 5 mths	Uni	R	III	999	Nil	2.83	Nil	2.83	24.7	18.3	16.0	34.0	0	10	2	5	120	3	120	8	120	14	120	3	120	3	120	4	720	35
7	F	14 yrs 7 mths	Bi	R	IV	5	2.83	2.83	2.83	2.83	33.0	32.3	26.3	26.2	2	3	6	6	8	7	14	12	120	12	16	7	11	5	7	5	176	48
8	F	5 yrs 6 mths	Bi	R	II	3	2.83	2.83	2.83	2.83	29.3	27.7	28.3	29.7	3	6	4	5	9	9	11	10	29	17	8	6	6	6	7	6	70	54
9	M	14 yrs 6 mths	Bi	L	II	2	3.61	3.61	3.61	3.61	22.8	22.2	34.7	36.3	5	4	2	3	21	13	57	20	120	120	120	32	20	25	12	10	350	220
10	F	7 yrs 5 mths	Bi	L	I	999	2.83	2.83	2.83	2.83	36.8	37.0	37.0	36.3	10	10	6	6	8	7	9	11	12	14	4	17	5	6	6	6	44	61
13	M	11 yrs	Bi	L	II	999	2.83	2.83	2.83	2.83	6.7	11.3	36.0	37.0	8	10	6	6	8	10	10	11	63	120	11	6	8	6	5	5	105	158
15	M	15 yrs 6 mths	Bi	L	II	999	3.61	2.83	2.83	3.61	25.3	27.5	28.0	33.2	10	10	4	6	120	7	120	10	120	12	120	6	114	5	120	5	714	45
19	M	7 yrs 3 mths	Bi	L	I	2	2.83	2.83	2.83	2.83	37.0	37.0	37.0	37.0	10	10	6	6	9	5	6	7	32	9	6	4	4	4	4	4	61	33
21	M	6 yrs 6 mths	Bi	R	II	999	2.83	2.83	2.83	2.83	26.0	30.7	32.3	35.0	10	7	5	5	12	10	11	9	90	76	11	7	8	6	8	6	140	114
28	F	4 yrs 11 mths	Bi	L	III	999	2.83	2.83	2.83	2.83	27.7	28.3	33.3	26.7	5	5	4	4	37	23	30	13	120	120	38	25	20	11	21	13	266	205
34	M	14 yrs 6 mths	Bi	L	I	2	2.83	2.83	2.83	2.83	34.0	30.3	34.0	33.8	10	10	6	6	7	4	13	8	69	22	3	1	5	4	5	4	102	43
39	M	12 ys 10 mths	Bi	L	I	999	2.83	2.83	2.83	2.83	37.0	37.0	32.0	31.7	9	10	6	5	10	7	15	11	120	29	3	2	5	6	6	6	159	61
45	M	13 yrs 3 mths	Bi	L	I	2	2.83	2.83	2.83	2.83	33.5	32.5	35.2	33.5	10	10	6	6	4	5	10	10	12	17	3	1	5	4	5	3	39	40
49	M	8 yrs 5 mths	Bi	L	I	2	2.83	2.83	2.83	2.83	24.7	19.3	30.3	26.2	10	10	6	6	6	7	9	12	23	27	10	6	5	5	5	5	58	62

4.2.4 Children Who Recorded Intact Upper Limb Somatosensation

Using the sensory threshold criteria outlined earlier for 'normal' somatosensory function (Chapter 3, section 3.3.7), eight (22%) children (participants #10, #16, #19, #34, #43, #44, #45 and #49) recorded intact somatosensory function. That is, all eight children detected the lightest SWM (green, 2.83) for both the finger and thumb, scored 10 out of 10 for both hands for proprioception, and six out of six for both hands for stereognosis. The age range was six years and one month through to 15 years and 11 months. Results for the AsTex® will be discussed in section 4.2.5.2. The eight children who recorded normal somatosensory function appear in Table 12, along with each child's ND hand JTHFT ranking (right hand column), which is discussed in section 4.2.5.6 (Table 17).

Table 12 – An overview of the eight Stage 1 participants identified as having 'normal' somatosensory function, grouped first by CP classification and secondly by JTHFT ND Hand Ranking

Subject #	*Sex*	*Age at Ax*	*CP Type*	*ND side*	*MACS Level*	*Brain Scan*	*JTHFT ND Hand Ranking*
43	F	15 yrs 11 mths	Uni	R	I	3	3
16	M	6 yrs 1 mth	Uni	R	I	2	8
44	M	11 yrs 2 mths	Uni	L	II	2	14
45	M	13 yrs 3 mths	*Bi*	L	I	2	1
10	F	7 yrs 5 mths	*Bi*	L	I	999	2
49	M	8 yrs 5 mths	*Bi*	L	I	2	4
19	M	7 yrs 3 mths	*Bi*	L	I	2	5
34	M	14 yrs 6 mths	*Bi*	L	I	2	11

Notes: F = female; M = male; R = right; L = left; yrs = years; mths = months; Uni = unilateral; *Bi* = bilateral; CP = cerebral palsy; Ax = assessment; MACS = Manual Ability Classification System; ND = non-dominant; JTHFT ND Hand Ranking = Jebsen Taylor Hand Function Test non-dominant hand ranking (where 1 represents the fastest time for the cohort and 36 represents the slowest time), see section 4.2.5.6 (Table 17).

From a CP classification perspective, three of the 23 (13%) children in the unilateral group and five of the 13 (38%) children in the bilateral group recorded normal somatosensation. Within the sub-group of eight children who recorded normal somatosensation the predominant classifications were: seven (88%) children were MACS Level I (the eighth was MACS Level II), six (75%) had a left side involvement

(right side brain injury), six (75%) were classified as having periventricular white matter injury, six (75%) were male, and five (63%) had a bilateral involvement. From an imaging and neuro-anatomical perspective, white matter injury is known to be associated with favourable hand function, as noted earlier (Holmström *et al.*, 2010).

While the number of children within the cohort with intact sensation is small (n=8), this sub-group differs from the overall cohort (n=36) in that it is over-represented by children with the highest level of hand function, MACS Level I (sub-group proportion = 88% compared to cohort proportion = 23%), periventricular white matter injury (75% compared to 36%), and bilateral CP (63% compared to 36%). Additionally, unlike the children who recorded intact sensation from the unilateral group, all five of the children from the bilateral group had the same ND side (left), the same MACS classification (Level I) and four of the five had a periventricular white matter injury (the fifth did not have a brain scan record).

4.2.5 Children Who Recorded Abnormal Upper Limb Somatosensation

Twenty-eight children (78%) recorded abnormal sensory assessment results – 20 of the 23 (87%) children with unilateral CP and eight of the 13 (62%) children with bilateral CP. These children represented all MACS Levels for the study (I(2); II(18); III(5); IV(3)), both sexes (16 males, 12 females), all ages (four years, 11 months through to 15 years, six months), and both ND sides of the body (17 left, 11 right).

4.2.5.1 Results for the SWM Test of Light Touch (Tactile Registration)

Eight children (22%) from the cohort recorded abnormal tactile registration (SWM filament > 2.83 or 0.07g force) for either the finger and/or thumb for either hand. Six children had unilateral CP and two had bilateral CP. The impairment always involved the ND hand and was primarily for both testing locations (six of eight cases). The dominant hand was involved on three occasions, and never in isolation but always in association with an abnormal ND SWM result. Four children recorded ‘Nil’ SWM results for both testing locations, meaning they did not register the stiffest filament (6.65 or 300g force), and all four children had unilateral CP.

All children who recorded abnormal tactile registration also recorded abnormal test results for tactile perception (proprioception and stereognosis) for that particular hand. For the unilateral CP group (n=6), all children also scored less than half on either test (five or less for proprioception; three or less for stereognosis), and five of the six children scored the maximum possible (worst) score of 720 seconds for their combined JTHFT ND hand score. For the bilateral CP group (n=2), an abnormal result for the SWM test resulted in scores of half or less for one child for both proprioception and stereognosis (#9) but only a score of four out of six for the ND hand for stereognosis for the second child (#15).

4.2.5.2 Results for the AsTex® Test (Tactile Discrimination)

Administering the test of tactile discrimination using the AsTex® device was very problematic for this particular cohort. The assessing therapist reported that children were confused by the test, despite the pre-test instructions and familiarity provided. Due to being blind-folded, children would either run their finger at an angle and hence off the grooved section of the AsTex® and onto the smooth side surface thinking they'd finished the test (when this occurred the therapist would stop the test and repeat it), or the child would move their finger along the grooved section too rapidly, 'racing' to the end of the grooves. When the latter occurred the therapist would repeat the test and ask the child to slow down, or guide their finger along the grooved central strip to ensure it stayed on the grooved section for its entirety. The assessing therapist reported that in most instances for the latter case the child did not change their behaviour and continued to rapidly move their finger over the grooves, and in their opinion, they felt this test and both interpretations of it (Test 1 and Test 2) were of little value. Despite the feedback, data for this test were collected and analysed for completeness, so also appear in Table 18 and Table 19.

As reported in Table 11, individual variability was generally high for this test. Table 13 reports the median and interquartile ranges (IQR) for each test, per hand and per CP group. The results of this test are discussed in more detail in the Discussion (section 4.3), and compared to results published by Auld *et al.* (2012b).

Table 13 – Median and interquartile AsTex® texture discrimination index (TDI) values for both CP groups and both hands, per test administered

	AsTex® TDI – Test 1		***AsTex® TDI – Test 2***	
	ND	***Dom***	***ND***	***Dom***
Unilateral CP Group				
Median	0.86	0.64	0.60	0.37
IQR	0.47 – 1.03	0.47 – 0.96	0.34 – 0.96	0.27 – 0.70
Bilateral CP Group				
Median	0.70	0.64	0.44	0.44
IQR	0.4 – 0.96	0.5 – 0.83	0.34 – 0.64	0.24 – 0.70

Notes: CP = cerebral palsy; TDI = texture discrimination index; IQR = interquartile range; ND = non-dominant; Dom = dominant.

4.2.5.3 Results for the Test of Proprioception

Twenty-two children (61%) from the cohort recorded an abnormal result (a score < 10) for the test of proprioception for either hand; 15 children (65%) with unilateral CP and seven children (54%) with bilateral CP. Within the unilateral group, the ND hand performed worse on 13 of 15 occasions and within the bilateral group the ND hand performed worst on four of seven occasions. For eight children, abnormal results were recorded for both hands (four from each CP group).

Within the unilateral group, if an abnormal result was recorded for the ND hand for proprioception, an abnormal result was also recorded for the same hand for the test of stereognosis (a score < 6) 93% of the time (13 of 14 occasions). For the bilateral group, this situation occurred 50% of the time (three of six occasions).

4.2.5.4 Results for the Stereognosis Test

Twenty-five children (69%) from the cohort recorded an abnormal result (a score < 6) for the test of stereognosis for either hand; 19 children (83%) with unilateral CP and six children (46%) with bilateral CP. Abnormal results were restricted only to the ND hand for 10 of 19 children (53%) from the unilateral group and one child (17%) from the bilateral group, and in both hands for nine children from the unilateral group (47%) and four children from the bilateral group (67%).

For the unilateral group, stereognosis scores ranged from two to six for the dominant hand and from zero to six for the ND hand. In comparison, for the bilateral group, stereognosis scores were less spread, ranging from three to six for the dominant hand and from two to six for the ND hand. This is shown graphically via two frequency distribution plots of stereognosis score per hand, for both the unilateral (Figure 4-2) and bilateral groups (Figure 4-3). For comparative purposes, both figures appear together on the following page.

For the unilateral group, dominant hand scores were typically high (five or six), indicating close to 'normal' hand performance, whereas ND hand scores were poorer, with results recorded across the full range of score possibilities. For the bilateral group, both hands performed similarly, with the ND hand performing slightly poorer (this hand recorded a greater range of results, with less scores of five or six) compared to the dominant hand. Three children (8%) recorded a higher test score using their ND hand compared to their dominant hand – two children from the unilateral group (#2, #29) and one from the bilateral group (#39). The largest discrepancy was for child #29, who scored four for their ND but only two for their dominant hand.

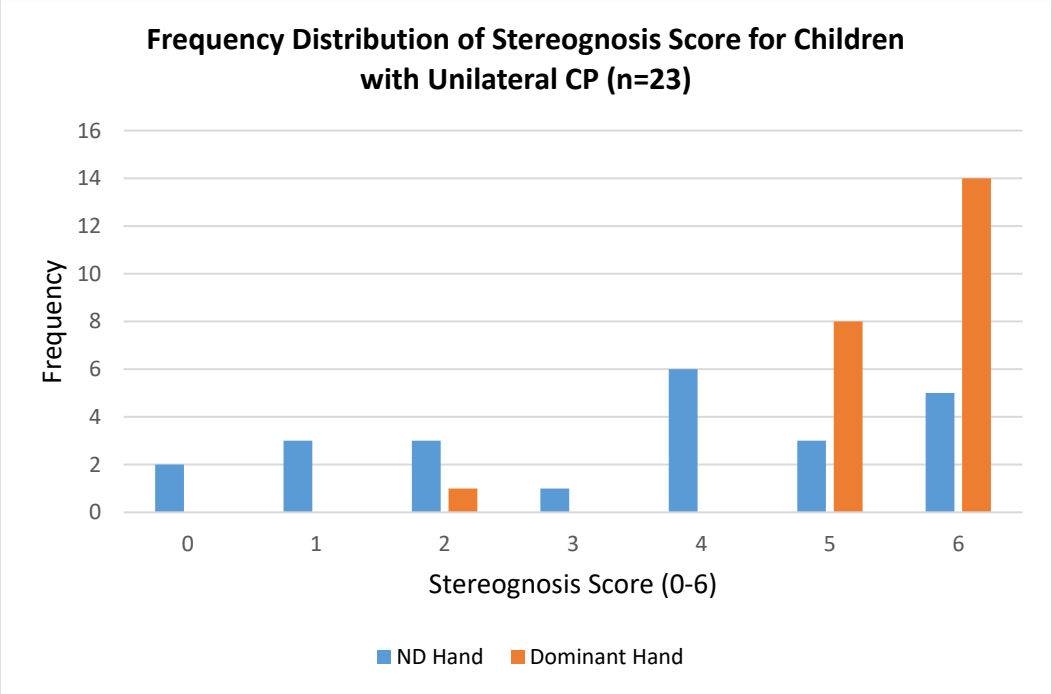


Figure 4-2 – The frequency distribution of stereognosis score per hand for the unilateral group (n=23) (blue = ND hand, orange = Dominant hand)

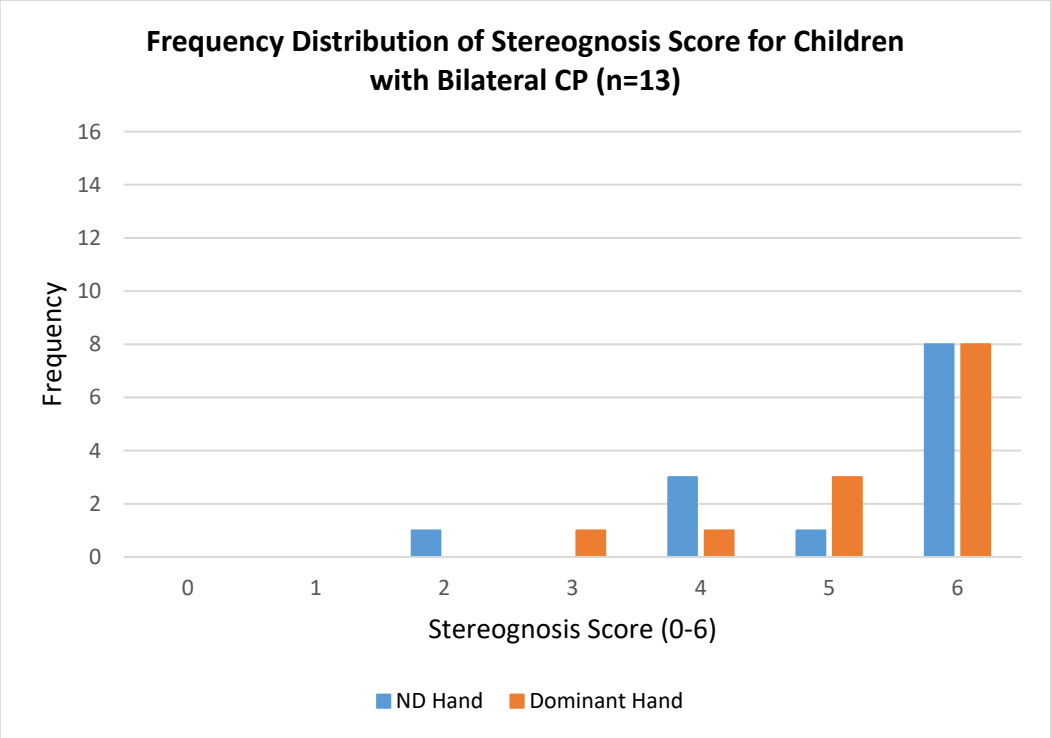


Figure 4-3 – The frequency distribution of stereognosis score per hand for the bilateral group (n=13) (blue = ND hand, orange = Dominant hand)

4.2.5.5 Results of the Jebsen Taylor Hand Function Test (JTHFT)

As mentioned in Chapter 3 (section 3.3.5.1), two changes were made to the guidelines for the standard JTHFT, namely that Test 1 (the hand writing test) was omitted and a time limit of 120 seconds was set for each test to limit frustration. This meant six of the seven Jebsen-Taylor tests were conducted as part of this assessment.

Table 14 shows the mean times, standard deviations, and range of test scores for each test per hand, for both cohorts, including the total test time for all six tests. The last two right hand columns of Table 14 show data published by Reedman *et al.* (2015) for a typically developing cohort (TDC) aged six years and zero months through to ten years and eleven months (n=71). While this represents a narrower age range than the cohort for the present study, it also provides an indication of paediatric TDC hand performance when using the JTHFT. Welch's Test for Unequal Variances (or Welch's t-test), which is a modification of the Student's t-test, was used to determine statistical significance between the test results (that is, comparing the results from this study to Reedman *et al.*'s TDC results). Welch's t-test is recommended when two different samples have unequal variances as well as unequal sample sizes, as is the case in this scenario.

As shown in Table 14, the mean total time taken for the JTHFT for both hands for both groups was significantly longer when compared to the mean total time for each respective hand of TDC (p values indicated in Table 14, all less than 0.05). In particular, the dominant hand for children with unilateral CP was significantly slower compared to the dominant hand for a child without CP (p=0.004). The most significant result was recorded for the ND hand for children with unilateral CP (p<0.0001) compared to TDC.

Table 14 – The mean ± standard deviation and range of test scores for the JTHFT for all children for their ND and Dominant hands per test, compared to normal values as per Reedman *et al.* (2015). All test values in seconds.

JTHFT Test No.	*Test Times (secs) (Range)*					
	Unilateral Group (n=23)		***Bilateral Group (n=13)***		***Normal (n=71) (*)***	
	ND	***Dom***	***ND***	***Dom***	***ND***	***Dom***
2	48.3 ± 48.2 (4 – 120)	5.3 ± 1.9 (3 – 9)	19.9 ± 30.1 (4 – 120)	8.8 ± 4.7 (4 – 23)	4.82 ± 1.01	4.42 ± 0.95
3	63.1 ± 46.0 (7 – 120)	8.9 ± 3.8 (5 – 21)	24.2 ± 30.6 (6 – 120)	11.1 ± 3.0 (7 – 20)	7.06 ± 1.10	6.36 ± 0.88
4	94.0 ± 37.9 (25 – 120)	25.3 ± 30.1 (8 – 120)	71.5 ± 43.8 (12 – 120)	45.8 ± 43.8 (9 – 120)	9.93 ± 2.06	8.56 ± 1.44
5	53.6 ± 49.4 (3 – 120)	5.1 ± 3.6 (1 – 15)	27.2 ± 40.6 (3 – 120)	9.2 ± 9.2 (1 – 32)	4.36 ± 0.93	3.81 ± 0.71
6	46.3 ± 49.7 (3 – 120)	4.3 ± 2.0 (2 – 12)	16.6 ± 28.6 (4 – 114)	7.2 ± 5.4 (4 – 25)	3.52 ± 0.54	3.28 ± 0.51
7	57.8 ± 54.7 (4 – 120)	4.8 ± 1.7 (2 – 9)	16.2 ± 30.3 (4 – 120)	6.0 ± 2.6 (3 – 13)	3.57 ± 0.49	3.40 ± 0.50
Total time	363 ± 257 ***p*<0.0001**	54 ± 36 ***p*=0.004**	176 ± 186 ***p*=0.0171**	88 ± 65 ***p*=0.0073**	33.25 ± 4.25	29.85 ± 3.54

Notes: JTHFT = Jebsen Taylor Hand Function Test; No. = number; ND = non-dominant; Dom = dominant; secs = seconds; (*) Normal values from Reedman *et al.* (2015), TABLE 2, pg. 298, 'Test' scores; *p* values calculated using Welch's t-test comparing means, standard deviations, and sample sizes with data from Reedman *et al.* (2015) for the ND and Dom hands, respectively.

One of the study aims was to determine the level of correlation between sensory impairment and the level of activity of the upper limb. Within the literature, this is usually demonstrated by comparing stereognosis with the total time for the JTHFT. Due to the nature of the scoring for these tests – where a high score for stereognosis and a low score (time) for the JTHFT indicates good/normal hand function – an inverse relationship exists between these two measures. Figure 4-4 shows a scatter plot of the stereognosis result versus the total time taken to complete the JTHFT, for the ND hand for both groups (the unilateral group is shown in blue, and the bilateral group is shown in orange).

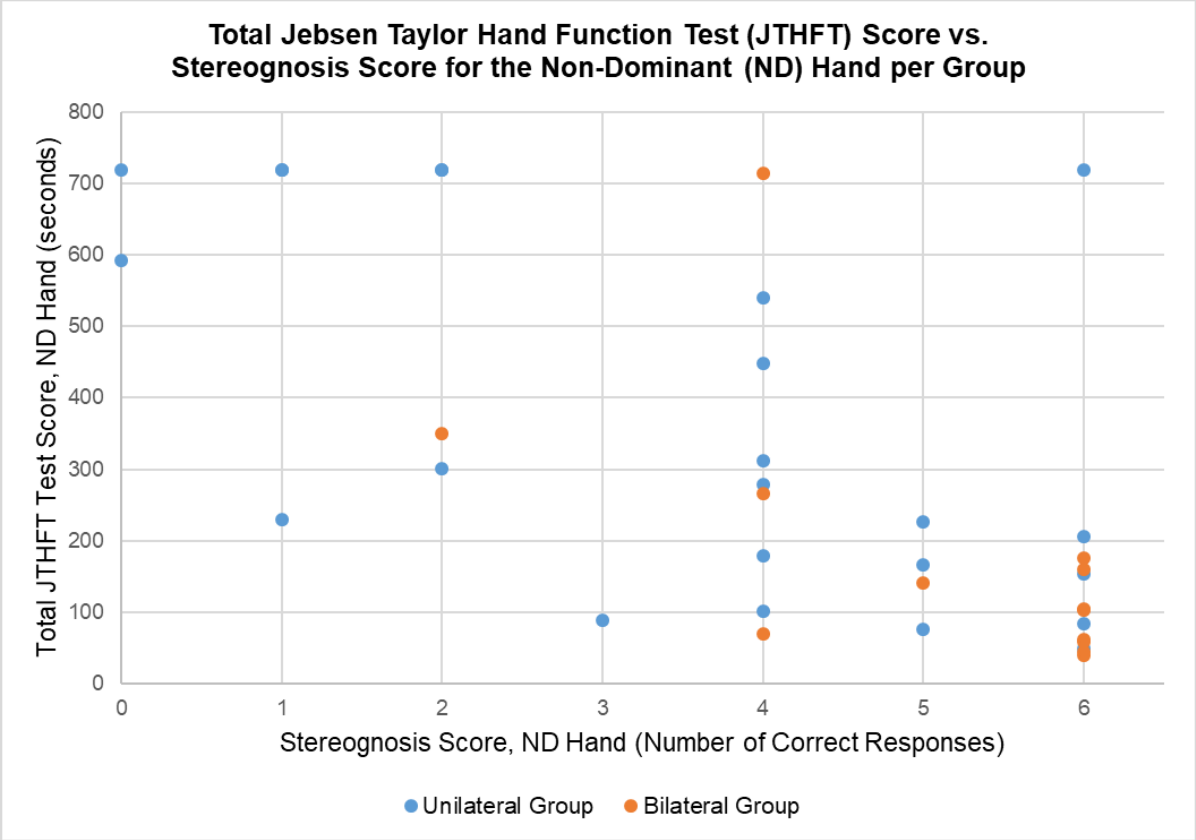


Figure 4-4 – A scatterplot showing correlation between stereognosis score and the total time for the Jebsen Taylor Hand Function Test (JTHFT) for the ND hand, per group (blue = Unilateral group, orange = Bilateral group)

As indicated in Figure 4-4, a grouping of data occurs in the bottom right portion of the scatterplot, representing a good stereognosis score associated with a comparatively good JTHFT score, with only one outlier (child #2 from the unilateral group). Generally, a poor stereognosis score tended to be associated with a high JTHFT score, but not always as indicated by child #12 from the unilateral group, who scored three for stereognosis but only 89 seconds for the total JTHFT time, and child #2 who scored six for stereognosis but the maximum score possible (720) for the JTHFT test. Measures of association are analysed in more detail in section 4.2.5.9.

For the unilateral group, the total time taken for the JTHFT was always greater for the ND hand compared to the dominant hand, as would be expected for such a task and the unilateral nature of this particular group. However, for the bilateral group, there were four instances where the total time taken for the JTHFT was less for the ND hand than the dominant hand – for children #10, #13, #45 and #49. The

differences in total time ranged from 53 seconds (child #13) to 1 second (child #45), which is essentially equivalent hand performance for child #45 given that the difference is spread over six different tests. Three of the four children (#10, #45 and #49) recorded intact sensation according to the criteria outlined in section 4.2.4 and were classified as MACS Level I (child #13 was MACS Level II). All four children had a ND left side (right-side brain injury).

4.2.5.6 The Number of Sensory Modalities Affected per Hand, per Group

Using the results for tactile registration (SWM) and perception (proprioception and stereognosis), an analysis of the number of sensory modalities that were affected per child was conducted for this study. For children with unilateral CP, 92% (n=21) of the cohort recorded an impairment for their dominant hand in one modality or less, compared to 43% (n=10) for the ND hand, as shown in Table 15.

Table 15 – The number of sensory modalities (light touch, proprioception and stereognosis) affected per hand for children with unilateral CP (n=23)

No. of sensory modalities affected (n)	*ND hand (n)*	*%*	*Dom hand (n)*	*%*
0	4	17	11	48
1	6	26	10	44
2	7	31	1	4
3	6	26	1	4

Notes: No. = number; ND = non-dominant; Dom = dominant

For children with bilateral CP, 69% (n=9) of the cohort recorded an impairment involving one modality or less for either hand, as shown in Table 16. Only 31% (n=4) of the bilateral group recorded impairments in two or more modalities for their ND hand, compared to 57% (n=13) for the unilateral group. For the dominant hand, again, 31% (4) of children with bilateral CP recorded impairments in two or more modalities compared to only 8% (n=2) for children with unilateral CP.

Table 16 – The number of sensory modalities (light touch, proprioception and stereognosis) affected per hand for children with bilateral CP (n=13)

No. of sensory modalities affected (n)	*ND hand (n)*	*%*	*Dom hand (n)*	*%*
0	5	38	6	46
1	4	31	3	23
2	3	23	3	23
3	1	8	1	8

Notes: No. = number; ND = non-dominant; Dom = dominant

Table 17 shows the Stage 1 cohort ranked by the total time taken for the ND hand to complete the JTHFT, divided into quartiles (nine children per quartile). The top (fastest) grouping completed all six JTHFT tasks in less than 90 seconds (mean = 63.1 ± 17.4 seconds), contains more children with bilateral than unilateral CP, MACS Level I classification, and more children with a brain scan classification of periventricular white matter injury. Additionally, six of the nine children in this group recorded intact upper limb sensation (highlighted in green).

The second grouping completed all six JTHFT tasks in a time between 101 and 179 seconds (mean = 142.4 ± 32.0 seconds) and contains mostly children with MACS Level II classification and children with zero or one impaired sensory modality.

The third grouping completed all six JTHFT tasks in a time between 205 and 448 seconds (mean = 290.8 ± 74.8 seconds), contains mostly children with unilateral CP, predominantly MACS Level II (range: Levels II to IV), and children with between one and three impaired sensory modalities.

The fourth and slowest grouping is almost exclusively children with unilateral CP, who completed all six JTHFT tasks in a time between 541 and 720 seconds (median time = 720 seconds, IQR = 66.5[3]), who predominantly have a ND left side (right side brain injury), brain scan classification of cortical/subcortical lesions, and mostly two to three impaired sensory modalities. An exception to the last statement is child #2 (ranked 32nd out of 36), who recorded zero impaired sensory modalities for her ND hand, however this child recorded one impaired sensory modality for her dominant hand, for the test of stereognosis.

[3] Data for the fourth grouping had a non-normal distribution, so median and IQR values are reported.

Table 17 – The Stage 1 cohort ranked by total time taken (in seconds) to complete the JTHFT for the ND hand, divided into quartiles

Rank	*Subject No.*	*Sex*	*Age*	*Group*	*ND Side*	*MACS Level*	*No. of Mod. Affected (ND side)*	*Brain Scan Class.*	*Total Time (secs), ND Hand*
1	45	M	13 y 3 m	Bi	L	I	0	2	39
2	10	F	7 y 5 m	Bi	L	I	0	999	44
3	43	F	15 y 11 m	Uni	R	I	0	3	49
4	49	M	8 y 5 m	Bi	L	I	0	2	58
5	19	M	7 y 3 m	Bi	L	I	0	2	61
6	8	F	5 y 6 m	Bi	R	II	2	3	70
7	27	F	9 y 6 m	Uni	L	II	2	2	75
8	16	M	6 y 1 m	Uni	R	I	0	2	83
9	12	M	12 y 8 m	Uni	R	II	3	3	89
10	5	M	7 y 7 m	Uni	R	II	2	999	101
11	34	M	14 y 6 m	Bi	L	I	0	2	102
12	13	M	11 y	Bi	L	II	1	999	105
13	21	M	6 y 6 m	Bi	R	II	1	999	140
14	44	M	11 y 2 m	Uni	L	II	0	2	154
15	39	M	12 y 10 m	Bi	L	I	1	999	159
16	41	F	6 y	Uni	L	I	1	3	166
17	7	F	14 y 7 m	Bi	R	IV	1	5	176
18	46	M	9 y 1 m	Uni	L	II	2	3	179
19	33	F	13 y 4 m	Uni	R	II	1	3	205
20	25	M	12 y 1 m	Uni	L	II	1	999	226
21	37	M	12 y 11 m	Uni	L	II	2	4	230
22	28	F	4 y 11 m	Bi	L	III	2	999	266
23	29	M	8 y 7 m	Uni	L	IV	2	1	279
24	11	F	5 y 9 m	Uni	R	II	1	1	301
25	6	M	14 y 6 m	Uni	R	IV	2	3	312
26	9	M	14 y 6 m	Bi	L	II	3	2	350
27	30	F	14 y 9 m	Uni	R	III	1	3	448
28	38	M	6 y 5 m	Uni	L	II	1	2	541
29	31	M	6 y 2 m	Uni	L	III	2	999	593
30	15	M	15 y 6 m	Bi	L	II	2	999	714
31	1	F	10 y 11 m	Uni	R	II	3	3	720
32	2	F	8 y 5 m	Uni	L	II	0	3	720
33	4	M	8 y 2 m	Uni	L	III	3	3	720
34	32	M	7 y 6 m	Uni	L	II	3	3	720
35	42	F	8 y	Uni	L	II	3	3	720
36	47	F	8 y 5 m	Uni	R	III	3	999	720

Notes for this table appear over the page.

Notes for Table 17 (previous page): Green highlight = intact upper limb tactile sensation; No. = number; F = female; M = male; R = right; L = left; y = years; m = months; Uni = unilateral; Bi = bilateral; CP = cerebral palsy; ND = non-dominant; MACS = Manual Ability Classification System; Mod. = modalities; Class. = classification; secs = seconds. Brain Scan Class. as per Table 7.

4.2.5.7 Sensory Impairments Recorded on the Dominant Side for Children with Unilateral Cerebral Palsy

More than half the cohort with unilateral CP (12 of 23 children, 52%) recorded a sensory deficit for their dominant or less-affected hand (children #1, #2, #4, #25, #29, #30, #31, #32, #38, #41, #46, #47). With respect to the test of light touch (SWM), only one child (#32) registered a score that was above the normal threshold for the thumb, detecting the next stiffest filament (blue, 0.407g of force), which is classified as 'diminished light touch' according to the operation manual.

For the test of proprioception, five children scored less than 10 for their dominant hand (children #4, #29, #31, #32, and #38). The range of scores was four to nine correct responses, with three children recording a score of nine. For the test of stereognosis, nine children scored less than six for their dominant hand (children #1, #2, #25, #29, #30, #32, #41, #46, #47), with scores ranging from two to five. Child #29 recorded the lowest proprioception score (4) as well as the lowest stereognosis score (2) and the highest JTHFT score (171 seconds) for their dominant hand, yet recorded normal tactile registration for both hands, highlighting why multi-modality testing is important.

4.2.5.8 Within and Between Group Statistical Analysis

Table 18 shows the aggregated data for each hand (dominant and ND), for each test, and the within and between group analysis. Considering the unilateral CP group first (n=23), a between hand comparison identified a result approaching statistical significance (p=0.05) for the SWM test of the thumb but not the finger (p=0.56). Six of the 23 children in this group recorded an abnormal SWM result, mostly for the thumb, and abnormal results were mostly confined to the ND side. The dominant hand performed statistically significantly better than the ND hand for the tests of proprioception (p<0.001), stereognosis (p<0.001), and for the total time for JTHFT

(p<0.001), which is not unexpected given the hemispherical involvement and unilateral limb use associated with this group.

For the bilateral group (n=13), the between hand comparison identified no statistically significant differences for any test. This is not an unexpected result given children with a bilateral involvement tend to use both their hands to engage in activities. The test that yielded the smallest non-significant p value was the total time taken to complete the JTHFT (p = 0.17). While the mean scores for the two hands differ by a factor of two (ND hand mean = 176 seconds, dominant hand mean = 88 seconds), this non-significant result is partly due to the large variance, and hence standard deviation, associated with the cohort (176 ± 186 compared to 88 ± 65). Using the two means and standard deviations, a post-hoc power analysis was conducted at 80% power, generating an estimated sample size of n=43. Consequently, the study was underpowered in terms of generating a statistically meaningful result for this test.

A between group comparison identified no statistically significant differences between the two CP groups for either test of tactile registration (p=0.32 for the thumb and p=0.41 for the finger) or proprioception (p=0.59). However, a significant result between the two CP groups was identified for the test of stereognosis (p=0.001) and for the total score for the JTHFT (p=0.001).

Table 18 – Statistical analysis for the Stage 1 study, per test result, within and between CP groups

Test	*Unilateral (n=23)*		*Bilateral (n=13)*		*β (95% CI): Group[1] (Uni vs Bi)*	*β (95% CI): Dom vs ND[1] (within Uni)*	*β (95% CI): Dom vs ND[1] (within Bi)*
	ND	*Dom*	*ND*	*Dom*			
SWM (Thumb)	3.00 ± 0.51	2.83 ± 0.00	2.89 ± 0.22	2.95 ± 0.29	0.11 (-0.10, 0.32) (*p*=0.32)	-0.17 (-0.34, 0.00) (*p*=0.05)	0.06 (-0.15, 0.27) (*p*=0.58)
SWM (Finger)	2.87 ± 0.18	2.86 ± 0.16	2.95 ± 0.29	2.89 ± 0.22	-0.06 (-0.20, 0.08) (*p*=0.41)	-0.03 (-0.11, 0.06) (*p*=0.56)	-0.06 (-0.17, 0.04) (*p*=0.27)
AsTex® (Test 1)	27.53 ± 5.44	29.13 ± 6.29	28.75 ± 8.31	28.71 ± 7.50	-1.22 (-5.63, 3.19) (*p*=0.59)	1.60 (-0.55, 3.76) (*p*=0.14)	-0.05 (-2.91, 2.82) (*p*=0.98)
AsTex® (Test 2)	29.40 ± 7.44	32.14 ± 5.98	32.65 ± 3.52	32.49 ± 4.10	-3.25 (-7.15. 0.65) (*p*=0.10)	2.74 (-0.10, 5.58) (*p*=0.06)	-0.16 (-3.85, 3.53) (*p*=0.93)
Proprioception	7.39 ± 3.00	9.52 ± 1.31	7.85 ± 3.00	8.08 ± 2.69	-0.45 (-2.12, 1.21) (*p*=0.59)	**2.13 (1.16, 3.11) (*p*<0.001)**	0.23 (-1.07, 1.53) (*p*=0.73)
Stereognosis	3.52 ± 2.02	5.48 ± 0.90	5.15 ± 1.28	5.38 ± 0.96	**-1.63 (-2.57, -0.69) (*p*=0.001)**	**1.96 (1.21, 2.70) (*p*<0.001)**	0.23 (-0.76, 1.22) (*p*=0.65)
Total JTHFT Score	363 ± 257	54 ± 36	176 ± 186	88 ± 65	**187 (76, 299) (*p*=0.001)**	**-309 (-403, -215) (*p*<0.001)**	-88 (-213, 37) (*p*=0.17)

Notes: [1]Assessed using mixed effects model with group, dominant side and a group X side interaction term included as fixed effects and subject ID included as a random intercept; summary statistics are mean ± standard deviation; ND = non-dominant; Dom = dominant, Uni = unilateral; Bi = bilateral; JTHFT = Jebsen Taylor Hand Function Test.

4.2.5.9 Measures of Association between the Sensory Tests and the Jebsen Taylor Hand Function Test

Measures of association between the sensory test results and the total score for the JTHFT were analysed for the cohort and appear in Table 19. Correlations were analysed for the overall cohort, the ND and dominant sides, and for the type of CP (unilateral and bilateral).

There was no association between the total score for the JTHFT and the test of tactile registration for the thumb for any sub-group that was analysed. However, there was a statistically significant and positive association between the total score for the JTHFT and tactile registration for the finger for the overall group (p=0.03), the ND side (p=0.04), and the bilateral CP group (p<0.001). As mentioned earlier, abnormal results for the SWM test of registration were mostly confined to the ND side, and if they were present on the dominant side, it was always associated with an abnormal ND result. The two children with bilateral CP who recorded abnormal SWM results for their finger on their ND side also recorded the two highest scores for the JTHFT (350 and 714 seconds, respectively).

Statistically significant inverse associations were recorded between the test of proprioception and the total score for the JTHFT for the overall cohort (p=0.004), the dominant side (p<0.001) and the unilateral CP group (p=0.002). The association was trending towards significance but not statistically significant (p=0.08) for the results on the ND side.

Strong, inverse associations were recorded for the test of stereognosis and the total score for the JTHFT across all sub-groups considered: the cohort overall (p<0.001), the side of involvement (ND, p<0.001 and dominant, p=0.005) and the type of CP (unilateral, p<0.001 and bilateral, p=0.03), with the association being strongest for the unilateral group (β=-0.72). With the exception of one category, the magnitude of the β coefficient for the stereognosis association was the largest β coefficient for every test (only the β coefficient for the association between the SWM (finger) and the bilateral group had a larger correlation, that is, β=0.76 compared β=-0.61).

Table 19 – Standardised measures of association (correlations) for sensory tests versus the Jebsen Taylor Hand Function Test[1]

	Overall (n=36)	ND	Dom	Unilateral (n=23)	Bilateral (n=13)
Monofil-Thumb	β=0.24, *p*=0.26	β=0.20, *p*=0.45	β=0.32, *p*=0.30	β=0.27, *p*=0.33	β=0.19, *p*=0.48
Monofil-Finger	**β=0.44, *p*=0.03**	**β=0.55, *p*=0.04**	β=0.36, *p*=0.24	β=0.28, *p*=0.35	**β=0.76, *p*<0.001**
AsTex® (Test 1)	**β=-0.35, *p*<0.001**	**β=-0.47, *p*=0.001**	**β=-0.33, *p*=0.03**	**β=-0.43, *p*<0.001**	β=-0.25, *p*=0.05
AsTex® (Test 2)	**β=-0.46, *p*=0.002**	**β=-0.62, *p*=0.001**	β=0.05, *p*=0.76	**β=-0.50, *p*=0.005**	β=-0.20, *p*=0.13
Proprioception	**β=-0.41, *p*=0.004**	β=-0.31, *p*=0.08	**β=-0.61, *p*<0.001**	**β=-0.56, *p*=0.002**	β=-0.16, *p*=0.54
Stereognosis	**β=-0.71, *p*<0.001**	**β=-0.66, *p*<0.001**	**β=-0.63, *p*=0.005**	**β=-0.72, *p*<0.001**	**β=-0.61, *p*=0.03**

Notes: [1]Assessed using simple linear regression with robust standard errors to account for repeat observations within individuals. The β coefficients are standardised estimates and equivalent to Pearson r correlations; ND = non-dominant; Dom = dominant.

4.2.5.10 The Severity of Somatosensory Impairments Recorded

Within the literature, a number of authors have not only compared their cohort of children with CP to control or normative data, but have also assessed and graded the severity of sensory loss recorded. Severity relates to the degree of impairment of a given sense. Criteria for 'severe' sensory deficits within the literature typically relate to the test of TPD and a distance in millimetres (as noted in Chapter 3, section 3.3.2), however, the following criteria can serve as a guide for tactile registration and perception:

- Tactile registration (SWM for either finger or thumb): a result greater than the blue filament, meaning the child felt the purple filament (4.31, 2.041g force, 0.305mm filament diameter) or higher. Purple correlates with 'diminished protective sensation';
- Tactile perception (stereognosis): Petersen *et al.* (2016) classified 'severe' stereognosis impairment as correctly identifying four objects or less from 10; Uvebrant (1988) classified 'poor' performance as correctly identifying one object or less from five; and Klingels, Demeyere*, et al.* (2012), which was the basis for this study, described stereognosis as being 'absent' for correctly identifying three objects or less from six.

With respect to tactile registration, six children (#1, #4, #12, #32, #42, and #47) or 17% recorded a severe deficit for their ND hand only, and all six children belonged to the unilateral group (MACS Level II (4), MACS Level III (2)). For all six children, the ND thumb was involved, and for four of the six both the ND thumb and finger were involved. All six children also recorded severe tactile perception deficits for their ND hand, three recorded scores of less than half (5) for the test of proprioception, and five of the six children recorded the maximum possible score (720 seconds) for the JTHFT for their ND hand. The ND side was left for three children and right for three children. Brain scan information was available for five of the six children, and all five had cortical/subcortical lesions, which was a similar finding to Uvebrant (1988). Additionally, five of the six children ranked in the bottom quartile of Table 17.

With respect to tactile perception, and using the criteria provided by Klingels, Demeyere*, et al.* (2012) for the test of stereognosis, 11 children (31%) recorded poor to very poor performance for this test (#1, #4, #9, #11, #12, #29, #31, #32, #37, #42,

and #47); 10 children from the unilateral group and only one child from the bilateral group. Seven of the 11 children have a ND left side, which is the same proportionality as the cohort overall. For nine of the 10 children from the unilateral group, this result was for the ND hand, as expected. For the child from the bilateral group, the result was for both hands. The worst scores (scoring zero or one) were confined to the ND hand of the unilateral group (n=5). Six of the ten children (60%) recorded a severe tactile registration deficit in the presence of a severe tactile perception deficit.

4.3 Discussion

This study, referred to as Stage 1 of the overall PhD project, assessed a cohort of 42 children with CP for somatosensory acuity, specifically addressing Study Aim 1. Despite the Stage 1 recruitment window being kept open for almost 24 months (May 2012 – April 2014), only a modest cohort was recruited. Three families originally consented to this study but then withdrew their child due to feeling overcommitted with existing extracurricular family activities, indicating how 'full' and busy the lives of these children can be when juggling regular activity and therapy. During the study each child was assessed bilaterally using standard somatosensory tests, namely, tactile registration for the index finger and thumb using SWM, TPD using the AsTex® device, proprioception using the distal phalanx of the child's thumb, tactile perception using stereognosis with six objects, and a test of functional hand motor skills using the JTHFT.

Overall, 36 children with either unilateral (n=23) or bilateral (n=13) CP completed the tests satisfactorily, with six children excluded from the results and data analysis mainly due to behavioural issues (Table 8). Other authors have also recorded being unable to administer certain sensory tests with their cohort (Breakey *et al.*, 1974; Yekutiel *et al.*, 1994; Klingels, Demeyere*, et al.*, 2012; Kurtaran *et al.*, 2015), as reported in Chapter 2. Eight children (22%) recorded results that indicated 'normal' sensation (Table 12). Seven of these children were MACS Level I, six had a ND left side (right side brain injury), six were male, six were classified as showing periventricular white matter injury, and five were children with bilateral CP. The literature reports no significant somatosensory differences due to sex, such as

Bolanos *et al.* (1989), meaning the 75% result for males with 'normal' sensation is likely due to the small cohort and not a significant result. Five of the children with 'normal' sensation also recorded the five fastest ND hand times for the JTHFT when the overall cohort is ranked from fastest to slowest (Table 17), and four of the five children had bilateral CP. The finding that most children with 'normal' sensation had a brain scan classification showing periventricular white matter injury is consistent with the fact that sensory processing is mainly cortical/subcortical in nature and that white matter injury is associated with favourable hand function (Holmström *et al.*, 2010; Arnfield *et al.*, 2013).

Addressing one aspect of Study Aim 2, considering the severity of somatosensory loss, children with unilateral CP recorded more severe sensory loss compared to children with bilateral CP. Only children with unilateral CP recorded severe tactile registration loss, and 91% of children with a severe tactile perception loss belonged to the unilateral CP group. Five of the six children with severe tactile registration loss had a brain scan showing cortical/subcortical lesions, an area of the brain known to be involved in sensory processing and associated with severe impairment (Uvebrant, 1988). The second aspect of Study Aim 2 is addressed in section 4.3.4.

Addressing Study Aim 3, better hand function and performance were associated with better stereognosis results for the overall cohort, both sub-groups, and for both hands, with the strongest association being for the unilateral group. This result supports the work of Kinnucan *et al.* (2010), who also demonstrated a statistically significant inverse correlation between the total time for the JTHFT and stereognosis for the ND hand (pg. 1320). The tests of tactile registration (SWM) for the finger and proprioception were also associated with JTHFT results for the overall cohort.

Addressing Study Aim 4, children with unilateral CP performed statistically significantly worse using their ND hand compared to their dominant hand for the tests of proprioception, stereognosis and total JTHFT test scores, however, children with bilateral CP performed equally well with either hand across all tests, but still poorer than TDC. Comparing results across CP groups, statistically significant differences were identified for the test of stereognosis and total JTHFT time, with the bilateral group performing better in terms of overall test performance. With respect to

ND hand function and the JTHFT test, when the cohort is ranked from fastest to slowest (Table 17), eight of the nine slowest performers have unilateral CP.

4.3.1 Appraisal of the Cohort

Following personal communication with the SACPR (Gibson & Scott, 2017), aggregate analysis of the Register at the time of recruitment (for birth years 1997 – 2007) was conducted. There were 380 children on the Register in April 2012, with 232 (61%) being male. Overall, 145 were classified as having a unilateral involvement (38% – 69 right-sided hemiplegia and 76 left-sided hemiplegia) and 202 were classified as having a bilateral involvement (53% – 114 with diplegia, 83 with quadriplegia, and 5 with triplegia). The remaining classifications, in decreasing order, were: ataxia (9), unknown (8), dyskinetic dystonic (7), monoplegia (5), dyskinetic athetoid (3), and hypotonia (1), which combine to represent the remaining 9% of the children on the Register at the time.

Comparing the Stage 1 cohort to all children who were on the Register at the time shows that our study recruited the same proportion of males (61% in both cases) but a greater proportion of children with a unilateral involvement (64% compared to 38% on the Register). Within the unilateral cohort, a similar proportion of children with left unilateral CP were recruited to the study from all children with left unilateral CP on the Register. In terms of the greater proportion of unilateral children compared to bilateral children being recruited to the study, it's possible that families of children with unilateral CP were more likely to agree to the study because they are more aware of their child's functional deficits, limitations, and unilateral hand use compared to children with a bilateral involvement.

Detailed information from the Register relating to mean age, MACS Level or side of involvement (apart from the unilateral group) was not available to compare. During the years 1993-2007, the minimum and maximum prevalence of CP in South Australia was 1.41 and 1.97, respectively, per 1,000 live births (Gibson *et al.*, 2012, pg.12). As of the 31st December 2016, there were 774 children on the Register, with the proportion of CP classifications remaining essentially the same as April 2012 (40% with a unilateral involvement, 52% with a bilateral involvement, and 8% were

classified as having ataxia, unknown, dyskinetic dystonic, monoplegia, dyskinetic athetoid or hypotonia) (Gibson & Scott, 2017). However, males represented 56% of the children on the register at the end of 2016, compared to 61% in April 2012 (Gibson & Scott, 2017).

4.3.2 Results Compared to the Published Literature

The cohort of children with unilateral CP recruited to the study was representative of children on the SACPR, with most having a ND left side. However, this is counter to both the Swedish population-based study and other cases reported by Uvebrant (1988), which all report right-sided hemiplegia being more common (pg. 50). The reason for this difference is not known. The sex ratio of 22 males to 14 females (1.6:1) for the current study is slightly higher than that reported by Uvebrant (1988), who assessed a much larger cohort (n=149, 99 males, male to female ratio = 1.4:1, pg. 50). The predominant ND side for Uvebrant's cohort was the right side (53%, n=151), whereas the predominant ND side for the current study is the left side.

The somatosensory deficit prevalence for this study was 78% (28 of 36 children), which is comparable to the results of previous published studies, such as Hohman *et al.* (1958) (cohort size n=47; deficit prevalence rate=72%), Jones (1960) (n=54; 74%), Kenney (1963) (n=19; 73%), Krumlinde-Sundholm and Eliasson (2002) (n=25; 72%) and Auld *et al.* (2012b) (n=52; 77%). Such results affirm the importance and relevance of somatosensory assessment of this population (Walmsley *et al.*, 2017).

This study identified and confirmed the presence of somatosensory impairments in the dominant hand for children with unilateral CP, as previously reported in the literature (Monfraix *et al.*, 1961; Wigfield, 1966; Lesný, 1971; Lesný *et al.*, 1993; Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002; Arnould *et al.*, 2007; Wingert *et al.*, 2008; Auld *et al.*, 2012b). In this study, 52% of children with unilateral CP recorded an impairment on their dominant side, which is in agreement with the 54% reported by Auld *et al.* (2012b).

Also in agreement with both Krumlinde-Sundholm and Eliasson (2002) and Auld *et al.* (2012b), tactile registration deficits were associated with an increased likelihood of tactile perception deficits, with all eight children (22%) who recorded an abnormal

SWM result also recording an abnormal tactile perception result and a poor time for the JTHFT. The result of approximately one in five children recording an abnormal tactile registration result for their ND hand is higher than that reported by Kurtaran *et al.* (2015) (6%), Klingels, Demeyere*, et al.* (2012) (9%), and Uvebrant (1988) (11%), comparable with Krumlinde-Sundholm and Eliasson (2002) (25%), and less than results reported by Arnould *et al.* (2007) (33%).

In agreement with many previous studies (Tizard *et al.*, 1954; Hohman *et al.*, 1958; Tachdjian & Minear, 1958; Wigfield, 1966; Wilson & Wilson, 1967b; Van Heest *et al.*, 1993; Cooper *et al.*, 1995; Arnould *et al.*, 2007; Auld *et al.*, 2012b; Klingels, Demeyere*, et al.*, 2012), stereognosis was the modality most often impaired (69%), followed by the test of proprioception (61%), similar to the results reported by Cooper *et al.* (1995). Additionally, Cooper *et al.* (1995) are one of the few groups to investigate the number of sensory modalities that were impaired per hand for their unilateral cohort, reporting a greater number of impaired modalities for the ND hand compared to the dominant hand (pg. 305). The present study confirms this result, with two or more modalities affected in the ND hand for 57% of the cohort, compared to 8% for the dominant hand (Table 15). However, this was not the case for children with bilateral CP, with 31% of the cohort recording two or more impaired modalities on both sides of the body (Table 16).

4.3.3 Hand Function and the Jebsen Taylor Test

The JTHFT is primarily a test of functional hand motor skills, however, it is known and recognised that sensory inputs are an important component for successful motor function and control (Cooper *et al.*, 1995, pg. 300). Additionally, previous research has shown statistically significant inverse correlations between stereognosis performance and some of the individual and total JTHFT scores (Kinnucan *et al.*, 2010). If the total time taken to complete the JTHFT for the ND hand is considered, all eight children with intact hand sensation appear in the top half of results when the cohort is ranked, with five of the children filling the first five positions (Table 17).

Twenty-two children (61%) required at least one test to be halted at 120 seconds because they could not complete it in the time allowed – 16 children (73%) with unilateral CP and six children (27%) with bilateral CP. A study comparing motor

function (assessed using the JTHFT) and stereognosis in the upper limb of children with CP by Kinnucan *et al.* (2010) found that 19 of 41 children (46%) with either unilateral or triplegic CP could not complete one or more JTHFT tasks in the time allowed. The proportional differences between the present study and the study by Kinnucan *et al.* (2010) may be explained by the fact that Kinnucan *et al.* allowed each child 180 seconds to complete each task, which is an extra 50% of time.

Kinnucan *et al.* (2010) reported the mean time to complete the JTHFT for their mostly unilateral CP cohort was approximately 330 seconds for the ND hand and 46 seconds for the dominant hand (Figure 2, pg. 1319), while Rich *et al.* (2017) reported mean times of 234.2 ± 167.5 seconds (ND) and 53.2 ± 27.3 seconds (dominant), respectively (pg. 967) for their unilateral cohort. Results for the unilateral group from the present study compare favourably with that of Kinnucan *et al.* (2010), despite the differences in time allowed to complete each task. It may be the case therefore that if a child with CP cannot complete a given Jebsen Taylor task in 120 seconds, it is unlikely they will complete it in 180 seconds, indicating that time is not a critical element for this test, rather functional ability. The results from the present study for the dominant hand (54 $\pm$ 36 seconds) compare favourably with that reported by Rich *et al.* (2017), but are higher for the ND hand (363 ± 257 seconds), indicating that the children from the present study had more severe hand limitations.

Of the 22 children who could not complete at least one JTHFT, nine (41%) recorded 120 seconds for at least half the tests (three or more), with eight of these children belonging to the unilateral group. Six children (27%), all from the unilateral group, had to have all six tests halted and consequently scored the maximum possible score of 720 seconds for their ND hand. Five of these six children also performed poorly on the SWM test, with the exception of participant #2. Only one child who tested as having normal somatosensation (#44) also scored 120 seconds (for both hands) for one of the tests (Test 4). Twenty-one of the 22 children recorded a score of 120 seconds for their ND hand, with participant #13 (with bilateral CP) the only child who recorded a score of 120 seconds for their dominant hand (Test 4) and not their ND hand. A post-study review of this child's assessment notes identified that the assessing therapist had written that while both hands were involved, the child wrote with one hand but used their other hand as the lead hand for some tasks.

From a test difficulty perspective, an examination of the data highlights that Test 4 (simulated feeding) was the most challenging test for children from both cohorts using either hand. Fifteen of the 23 unilaterally involved children (65%) and five of the 13 bilaterally involved children (38%) recorded the maximum score for this test using their ND hand. Additionally, Test 4 was the only test within the suite that children recorded a score of 120 seconds for when using their dominant hand, irrespective of group (unilateral or bilateral). Statistically, Test 4 recorded the highest mean time and largest standard deviation for both hands for both groups. This was also the case for the cohort from Kinnucan *et al.* (2010)'s study, where the mean time for the ND hand for this particular test was approximately 105 seconds, compared to 94.0 ± 37.9 for the ND hand for the present study. Test 4 is also the most difficult test (and hence has the highest mean and largest standard deviation for all tests, excluding the hand writing test) for typically developing children, as indicated by Reedman *et al.* (2015)'s data in Table 14 and as published by Taylor *et al.* (1973)(see Table 1, pg. 131 and Table 2, pg. 132, (Taylor *et al.*, 1973)). Reedman *et al.* noted that Test 4 demonstrated low reliability, poor reproducibility and that it was the most difficult test for children, often requiring multiple practices (pg. 300).

4.3.4 Side of Involvement / Non-Dominant Side

Further exploring Study Aim 2, Auld *et al.* (2012b) reported no statistically significant differences between children with a left or right side involvement on any specific test for their unilateral cohort of 52 children who were predominantly MACS Level I. However, the authors observed a trend towards children with a ND left side (right side brain injury) having more severe sensory deficits (Auld *et al.*, 2012b, pg. 1493), which was also reported by Cooper *et al.* (1995), but for a much smaller cohort (n=9, pg. 306). Earlier work by Monfraix *et al.* (1961) reported that severe agnosia seemed more common when the motor damage was right-sided, but that severe agnosia was significantly associated with spastic compared to athetoid CP (pg. 551), While the cohort for this study is smaller than Auld *et al.'s*, the results of the present study do not suggest that children with a ND left side are over represented in the sub-group that can be defined as having more severe sensory deficits (section 4.2.5.10).

Additionally, as noted earlier (Table 12), this study identified that children with a ND left side were proportionally over represented in the sub-group of children who recorded normal somatosensory function (six of the eight (75%) children with intact sensation have a ND left side, whereas 23 (64%) children from the overall cohort have a ND left side). However, when the cohort is ranked according to the overall time taken for the ND hand to complete the JTHFT (Table 17), seven of the nine (78%) children ranked in the bottom quarter of the cohort have a ND left side.

Research published by Okuda *et al.* (1995) highlighted the asymmetrical role of the human somatosensory cortex with respect to "*conveying highly organised sensory information to the motor cortex*" (pg. 496) when investigating complex finger movements. An injury to the left somatosensory cortex resulted in bilateral hand clumsiness, whereas an injury to the right somatosensory cortex resulted in only the left hand being affected (pg. 497). However, the study was small (four subjects) and the subjects were post-stroke adults (aged 57 – 66 years), between six and 12 months post stroke onset. More recently, Riquelme *et al.* (2014) demonstrated that somatosensory processing is different among individuals with CP aged five to 29 years. The authors examined a small bilateral CP cohort with lateralised motor impairments, identifying that participants with 'right-dominant motor impairments' showed brain activity that was more similar to healthy controls compared to participants with 'left-dominant motor impairments' (pg. 7). However, more research needs to be conducted to understand the relationship between lesion site and impairment within a CP population.

4.3.5 Tactile Discrimination and the AsTex® Results

As was reported earlier (section 4.2.5.2), the test for tactile discrimination using the AsTex® device was not successful during this study. Causby (2016) also reported difficulty using this device for his PhD research, with university student participants finding the test counter-intuitive and subject to variation.

The spread or dispersion in the data is most evident when the AsTex® results for the unilateral group only are compared to published data from Auld *et al.* (2012b) (Table 20), who reported median and IQR AsTex® values for the index finger for 52 children with unilateral CP and 34 TDC (pg. 1490). For Test 1, the median values and IQR

ranges for both hands (index fingers) are greater for the present study compared to that of Auld *et al.*, indicating a higher tactile discrimination threshold and greater data spread. For Test 2, the median value is comparable (ND hand) or lower (Dom hand) to that of Auld *et al.*'s with a greater data spread for the ND hand, but comparable spread for the Dom hand, which is inconsistent. One conclusion that may be drawn from the data obtained from the present study and the comparison with Auld *et al.* (2012b) is that instructions provided for Test 2 appear to produce a more accurate result compared to the instructions provided for Test 1.

Table 20 – Comparison of results for children with unilateral CP from the current study with that of Auld *et al.* (2012b) for the test of tactile discrimination using the AsTex® device

	ND finger (TDI) (mm)		***Dom finger (TDI) (mm)***	
Study	***Median***	***IQR***	***Median***	***IQR***
Auld *et al.* (2012b), TDC	0.27	0.21 – 0.64	0.27	0.21 – 0.47
Auld *et al.* (2012b), CP	0.60	0.40 – 0.83	0.44	0.24 – 0.68
Unilateral CP, Test 1	0.86	0.47 – 1.03	0.64	0.47 – 0.96
Unilateral CP, Test 2	0.60	0.34 – 0.96	0.37	0.27 – 0.70

Notes: CP = cerebral palsy; TDC = typically developing children; TDI = texture discrimination index; mm = millimetre; IQR = interquartile range; ND = non-dominant; Dom = dominant.

4.4 Conclusion

4.4.1 Study Aim 1

"*Recruit and assess children living with CP in South Australia for upper limb somatosensory function*" (section 4.1.1).

This study recruited and assessed 42 children with CP for somatosensory acuity from a population-based CP Register in South Australia (Figure 4-1). Each child was assessed using standard sensory tests, namely, tactile registration (index finger and thumb), TPD, proprioception (thumb), tactile perception (stereognosis), and a test of functional hand motor skills (JTHFT). Six children were excluded from the study, mainly due to behavioural issues (Table 8), with the remaining 36 children (Table 9) completing the tests satisfactorily (mean age: 10 ± 3.3 years; 22 males; unilateral CP (n=23), bilateral CP (n=13); and MACS Levels: I(9), II(19), III(5), IV(3)).

Only eight children (22%) recorded intact upper limb sensation (section 4.2.4), meaning the somatosensory deficit prevalence for this study was 78%. This is comparable to the results of previous published studies, such as Hohman *et al.* (1958) (cohort size n=47; deficit prevalence rate=72%), Jones (1960) (n=54; 74%), Kenney (1963) (n=19; 73%), Krumlinde-Sundholm and Eliasson (2002) (n=25; 72%) and Auld *et al.* (2012b) (n=52; 77%). Similarly, this study recorded somatosensory impairments in the dominant hand for 52% of children with unilateral CP (section 4.2.5.7), as previously reported (Monfraix *et al.*, 1961; Wigfield, 1966; Lesný, 1971; Lesný *et al.*, 1993; Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002; Arnould *et al.*, 2007; Wingert *et al.*, 2008; Auld *et al.*, 2012b).

4.4.2 Study Aim 2

"*Determine if the type of CP and the side of the lesion have an influence on the nature and extent of sensory impairment that is identified*" (section 4.1.1).

Children with unilateral CP recorded more severe sensory loss compared to children with bilateral CP. Only children with unilateral CP recorded severe tactile registration loss, and of the cohort that recorded severe tactile perception loss, 91% belonged to the unilateral CP group. However, the results of the present study do not suggest that children with a ND left side are over represented in the sub-group that can be

defined as having more severe sensory deficits (section 4.2.5.10), with 75% of children with intact sensation having a ND left side. Contrastingly, when the cohort is ranked according to the overall time taken for the ND hand to complete the JTHFT (section 4.2.5.6, Table 17), 78% of children ranked in the bottom quarter of the cohort have a ND left side.

4.4.3 Study Aim 3

"*Determine the level of correlation between sensory impairment and level of activity (function) in the upper limb*" (section 4.1.1).

Better hand function and performance was strongly associated with better stereognosis results for the overall cohort, both sub-groups, and for both hands, with the strongest association being for the unilateral group (Table 19). Additionally, the tests of tactile registration (SWM) for the finger and proprioception were also associated with JTHFT results for the overall cohort.

4.4.4 Study Aim 4

"*To compare the sensory and motor performance of the dominant and ND limbs of children with CP*" (section 4.1.1).

Children with unilateral CP performed statistically significantly worse using their ND hand compared to their dominant hand for the tests of proprioception, stereognosis and total JTHFT test scores, however, children with bilateral CP performed equally well with either hand across all tests (Table 18), but still significantly poorer than TDC. Comparing results across CP groups, statistically significant differences were identified for the test of stereognosis and total JTHFT time, with the bilateral group performing better in terms of overall test performance. With respect to ND hand function and the JTHFT test, when the cohort is ranked from fastest to slowest (Table 17), the fastest performing group represents an even mix of children with unilateral and bilateral CP, but eight of the nine slowest performers have unilateral CP.

5. The Design, Development, Testing and Piloting of a Serious Games Intervention for Children with Cerebral Palsy

This chapter introduces the field of serious games and describes the co-design, development, and evaluation processes for the intervention that was the focus of this PhD research. An overview of the field of using computer games to engage children with CP is provided in section 5.1, followed by the design requirements and specifications that were developed for this particular research (section 5.2). A fully standalone custom-made gaming solution was developed, wherein the software (section 5.3) and hardware design, development, and evaluation processes (section 5.4) are described, including the overall system integration (section 5.5), specific technical and clinical features (section 5.6), and the overall system overview and summary (section 5.7).

5.1 Using Computer Gaming to Engage End Users in Rehabilitation Activities

Engaging children with CP in meaningful therapy or exercise can be difficult, despite the merits of the intervention, the potential therapeutic benefits that accompany compliance, and the best intentions of family and rehabilitation specialists to motivate and encourage the child. A key aspect of the intervention for this project was the development of a haptic and accessible computer gaming system that included a range of challenging and engaging games that children with CP would want to play. As noted by Golomb and colleagues *"…rehabilitation that incorporates play also aids in motivation*" (Golomb *et al.*, 2011, pg. 392). The aim was to turn therapy into 'play', making it a fun, engaging and motivating activity and not something that is seen as 'work'.

Computer gaming, or just 'gaming', is an incredibly popular pastime. However, despite the profile and advocacy of groups such as *Game-Accessibility.com*[4] in Europe and the *AbleGamers Foundation*[5] in the US, commercial or off-the-shelf

[4] See: http://gameaccessibility.com
[5] See: http://www.ablegamers.org/

gaming systems generally remain inaccessible for people living with an impairment. From a business and marketing perspective this represents an untapped market, with *AbleGamers* alone representing more than 33 million gamers with either an acquired or congenital disability (AbleGamers, 2018). However, from a social perspective, this means many people with an activity limitation are excluded from participating in gaming, and are often excluded from peer conversations involving games as they cannot contribute their first-hand experience. For children, this has a greater impact given the appeal of games to a younger audience.

Using commercial gaming systems to facilitate and augment therapy is not a new concept. 'Serious games' (SGs) or 'serious gaming' is the recognised industry term for an electronic or computer gaming application where the primary objective of the game is not one of pure entertainment. Consequently, gaming for health, leadership, education or training are all examples of SGs. Different terms have been used to describe SGs used within a health or rehabilitation context, such as health gaming, interactive computer play (Sandlund, McDonough, & Hager-Ross, 2009), 'Exer-gaming' ('Exer' from the word 'exercise'), rehab gaming, active video gaming (AVG), '*Wiihabilitation'* (specifically when the *Nintendo Wii* system is used) and aspects of virtual or augmented reality (VR or AR), depending on the application.

By their very nature, the participatory aspect of computer gaming allows the player to be both engaged and distracted by the game, as they become immersed in the challenge of the game activity. Moreover, they create "*fun and engaging environments that motivate the child to exercise*" (Sandlund *et al.*, 2009, pg. 173). Rehabilitation practitioners began taking an interest in SG technology when it was recognised that gaming actions could be used to substitute the boredom often associated with rehabilitation program exercises including stretching, strengthening or mobilisation (Sharan *et al.*, 2012), skill acquisition (Annema *et al.*, 2010), and that it could also act as a distraction from pain (Pearson & Bailey, 2007; Annema *et al.*, 2010). Sandlund *et al.* (2012) described the current range of interactive technologies (described in section 5.1.1) as potentially being "*excellent tools to increase motivation for practice in rehabilitation*" (pg. 926). Staiano and Flynn (2014) concluded that this combination of entertainment and distraction could "…*be just as useful to completing therapy and restoring positive mood as the actual physical*

improvements attained" (pg. 361), while Sandlund *et al.* (2009) note the role SGs can play in delivering home-based rehabilitation, reducing travel time and hospital or clinic costs. A review of SGs used with children with CP by Bonnechère *et al.* (2014) reported that SGs have been used as a treatment option for children with CP since 1998 (pg. 1905).

Deutsch *et al.* critically analysed the literature within a rehabilitation setting for evidence of games being able to increase energy expenditure and exercise intensity, identifying that there was preliminary evidence of moderate energy expenditure for post-stroke survivors with moderate motor deficits and children with CP with mild deficits (Deutsch *et al.*, 2015, pg. 35). Dunne *et al.* highlighted the need for engaging children with CP in an immersive and engaging environment, noting that "…*a large challenge in administering therapy, however, is to maintain the child's interest and enthusiasm during these exercises*" (Dunne *et al.*, 2010, pg. 1751). Consequently, the aim of most SG interventions within a health setting is to utilise technology to engage and motivate end users, while requiring them to perform movements or actions akin to a therapy intervention. Most therapists acknowledge 'buy-in' (engagement and motivation) is a significant challenge, especially when it comes to a paediatric rehabilitation program. Sandlund *et al.* (2012) reported parents' perceptions of SG interventions, with parents reporting positively on how gaming can promote motivation, stimulation, social interaction, and reduce the burden and effort required by them to supervise training at home. The authors also reported that parents wanted games that addressed specific rehabilitation movements, that were individualised (for specific motor functions and skills), and unobtrusive. The literature also notes that SG interventions should complement and not replace the role of the therapist, allowing them to be more effective and helpful for their patient (Burdea, 2003; Annema *et al.*, 2010).

In a non-paediatric population, a 2012 Cochrane Review that investigated VR for stroke rehabilitation concluded that the technology was significantly more effective than conventional therapy in improving upper limb function and activities of daily living, but not grip strength or gait speed (Laver *et al.*, 2012, pg. 523). The authors hypothesised that VR was an enjoyable and motivating therapy when explaining the reason for the effectiveness (pg. 529) and noted very few side effects such as pain,

dizziness and headaches (pg. 528), and no serious adverse events. However, questions remain as to the most appropriate population (in terms of age, their interest in technology, and the stage of their recovery) that will benefit from such an intervention and what the intervention purpose should be. A recent systematic review of studies from January 2000 – August 2016 that investigated AVGs as a tool for physical, psychological and cognitive rehabilitation for older patients (mean age ≥ 60 years) identified mixed results from 19 studies that met their inclusion criteria (Zeng *et al.*, 2017). While positive rehabilitation effects were reported, along with a focus on physical functioning across the studies, the authors raised concerns about the quality of study design being undertaken and the limited number of studies to assess.

5.1.1 Using Commercial Gaming Systems for Serious Gaming Applications

From a game platform perspective, most commercial gaming systems, such as the *Sony Playstation* or *Microsoft Xbox*, are not readily accessible nor applicable for children with a disability, with game accessibility issues highlighted in the literature (Bierre *et al.*, 2005; Henschke, Hobbs, & Wilkinson, 2012). Typically, some form of modification or adaptation is required before a commercial game can be incorporated into a rehabilitation program. Most barriers to commercial gaming systems for children with hand impairments are two-fold.

Firstly, the hardware (that is, the joystick, controller or ‘nunchuk’ depending on the system being used) is a barrier because of the assumption and expectation that a user’s hands and fingers can hold and manipulate the physical interface. Children with CP typically lack the fine motor skills associated with finger coordination, control and dexterity to consistently and reliably access the full range of buttons and the joystick on a commercial controller required for competitive gameplay. As an example, the *Microsoft Xbox 360* controller, shown in Figure 5-1, has more than 11 different buttons of varying size, shape and location, all of which require activation when interacting with the system. The primary ‘in-game’ buttons are the coloured circular buttons labelled A, B, X and Y located on the right hand side of the controller. For a child with a right hand or thumb involvement, this presents an immediate challenge to gameplay in terms of accessing and pressing these buttons. Similarly, the left thumb joystick (labelled ‘3’ in Figure 5-1) is the primary controller

for almost every *Xbox* game, meaning a child with a left hand or thumb involvement would have difficulty controlling most if not all *Xbox* games. While some buttons may be accessible (such as the four primary buttons mentioned earlier, or the 'bumper' buttons labelled '5' and '10' in Figure 5-1) most commercial games utilise all available inputs and functionality of the controller. This means access to the full range of input options and not a sub-set is required to play a game successfully.

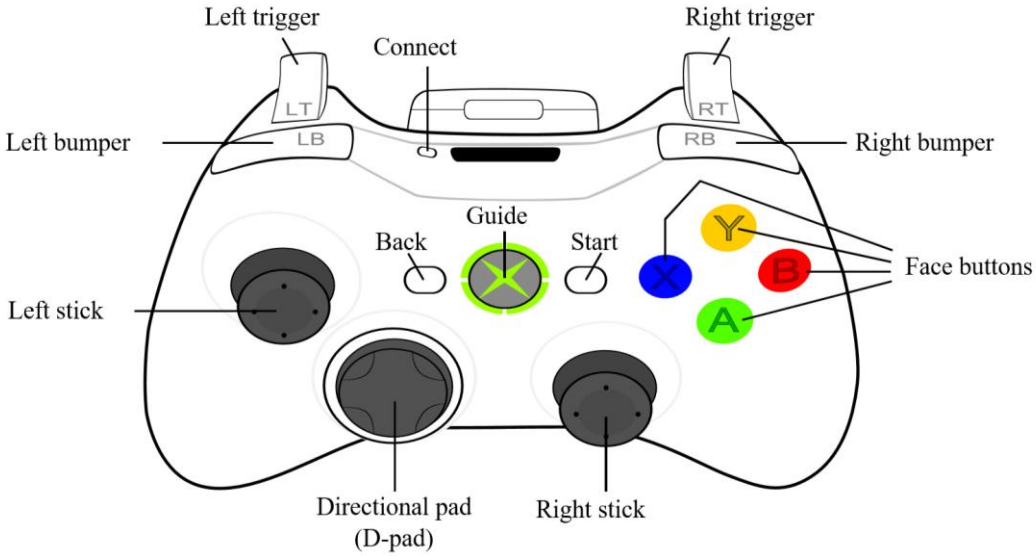


Figure 5-1 – A *Microsoft Xbox 360* controller showing the number, location and range of different buttons and thumb joysticks (sticks) and pads. The 'face buttons' (shown in colour and labelled A, B, X and Y) are the major buttons on the controller (Source: https://commons.wikimedia.org/wiki/File:360_controller.svg)

Secondly, the games themselves are designed and programmed based on the premise that the full range of buttons and joystick/thumb pad features are available, and hence the games themselves, through software, reinforce the hardware obstacles. As an example, to progress past a certain obstacle and to reach the next level within a game may require rapid, multiple and coordinated bilateral button presses combined with joystick manipulations, which can be extremely difficult or impossible for a child with a hand impairment. It is the combination of timing (pressing a particular button at a precise moment), digit specificity (requiring finger or thumb coordination and control) and repeated activations (digit dexterity) that cause issues for players with a hand and finger impairment. Staiano and Flynn (2014) noted that certain game features of commercially available systems may place limits on the ability of a player with a disability to play the game (pg. 361).

Anecdotally, even if the hardware obstacles can be overcome there is considerable frustration and disappointment for the child if they cannot independently succeed within the game for themselves. Compensation strategies include asking a typically developing sibling or friend to assist by completing the difficult part of the game for them, which is disempowering.

The use of commercial gaming systems for physical rehabilitation of people with a neurological impairment only became a viable option when new controllers – and hence the control input mechanism for the game – became available. In October 2003, Sony introduced the first gesture based control device – the *EyeToy,* shown in Figure 5-2. This unique camera plugged into the *Sony PlayStation2* and used computer vision to detect the presence of a player, and in turn used the player's movements to control the game. This was a quantum leap forward in terms of game interaction and control, with all commercial gaming systems up until that point using hand held controllers with multiple buttons played in a seated position. The *EyeToy* eliminated fine motor skills such as hand and finger movements for button pressing, using the player's stature and gross movements, typically in a standing position, to drive game character control.

Figure 5-2 – The *Sony EyeToy* (Source: https://commons.wikimedia.org/wiki/File:PS2-Eyetoy.jpg)

According to Bonnechère *et al.*'s most recent systematic review (Bonnechère *et al.*, 2016), a publication from Turkey using the *EyeToy* with a post-stroke population was the first commercial gaming system used within a physical rehabilitation context, highlighting the work of Yavuzer *et al.* (2008). While this technology was the first gesture-based controller on the market and the first published study in the literature implemented with an impaired population, uptake has been poor with only 8% of

studies using the *EyeToy* for a rehabilitation intervention (Bonnechère *et al.*, 2016, pg. 278). An earlier systematic review of the therapeutic uses of AVGs for all populations and ages identified 6% of studies used the *EyeToy* for an intervention (Staiano & Flynn, 2014, pg. 353).

Since the first SG publication in 2008, use of commercial gaming systems as a physical rehabilitation intervention for people with a neurological impairment has rapidly increased, with Bonnechère *et al.* (2016) investigating their use with patient groups including CP, stroke, Parkinson's disease, balance training, weight loss and ageing. This review identified that SGs are at least as efficient in terms of outcomes as conventional therapy and advantageous in areas including increasing patient motivation, decreasing or preventing monotony and boredom, providing feedback to the patient, and allowing double-task (training of more than a single activity) training (pg. 287). The review results are even more surprising given that none of the games used in any of the trials had clinical underpinnings or rationale – they were mainstream entertainment games. The authors argue that better therapeutic outcomes could be obtained if clinical requirements are considered and used as a basis for the game (pg. 287), and note that gaming interventions could have a role in maintaining rehabilitation benefits at home (pg. 277).

Introduced in late 2006, the *Nintendo Wii* has become the most popular commercial gaming system used within a physical rehabilitation setting, with 79% of studies involving children with CP (Bonnechère *et al.*, 2016, pg. 278) and 69% of AVG studies for all populations (Staiano & Flynn, 2014, pg. 353) using a *Wii* or *Wii Fit* (an exercise program that incorporates a peripheral known as the *Wii Balance Board*). What made this system successful was the unique functionality of the controller, called the *Wii Remote* or *Wiimote* (Figure 5-3). It was the first controller to incorporate three-axis microelectromechanical-system (MEMS) accelerometers into it, meaning it could measure movement in three dimensions[6], and this movement in space controlled the game character rather than a joystick controlled by a thumb or fingers, or the player's body (e.g.: the *Sony EyeToy*).

When coupled with Nintendo's *Wii Sports* games package, the *Wiimote* controls a baseball bat, a tennis racquet or a golf club, without the need for many (if any) button

[6] See: https://www.technologyreview.com/s/408183/hack-the-nintendo-wii/

presses. Similar to the *EyeToy*, when playing the *Nintendo Wii* the player is typically upright compared to sitting passively in front of a computer screen. Post-release, a wrist strap was added to the *Wiimote* to tether it to the player's limb should the player drop or accidentally let go of the controller when performing gross arm movements. However, this unique interface – being upright in an immersive 3D environment with the game being controlled through upper limb gestures – has also been known to result in serious injuries amongst the general (typically developed) population. Documented injuries including head trauma (Wells, 2008), higher than average hand lacerations and bruising (Sparks, Chase, & Coughlin, 2009), and fractures (Fysh & Thompson, 2009).

Figure 5-3 – The *Nintendo Wii Remote* (*Wiimote*) controller, which is held in the palm of the player's hand, and the accompanying wrist strap (Source: https://commons.wikimedia.org/wiki/File:Wiimote.png)

Within a rehabilitation context, the upright player holds the *Wiimote* in the palm of their hand making it an extension of their arm, promoting gross upper limb movement with minimal button presses and eliminating fine hand and finger control and coordination. Where grasp is not possible due to the impairment, the *Wiimote* is typically strapped to the involved hand as reported in the literature (Yong Joo *et al.*, 2010). To interact with any of the games, the player points the *Wiimote* at the screen, requiring balance combined with shoulder, elbow and wrist movements coupled with focussed attention and concentration. A feasibility study using the *Nintendo Wii* for upper limb motor rehabilitation in a post-stroke population reported that all subjects found the system enjoyable and comparable to, if not better than, conventional therapy, with most subjects agreeing that they would recommend it to

another patient (Yong Joo *et al.*, 2010). Yong Joo *et al.* (2010) reported two subjects withdrawing due to lethargy and fatigue after the first session, and a further three cases of mild pain and soreness that lasted less than 24 hours, which did not prevent the subjects from further participation (pg. 439).

Microsoft's response to Sony's *EyeToy* and Nintendo's *Wiimote* was the *Kinect* (Figure 5-4), another camera based technology that uses gesturisation as an input to the game, similar to the *EyeToy*. Used in 13% of all CP studies where a commercial gaming system has been used for physical rehabilitation purposes (Bonnechère *et al.*, 2016) and less than 2% of all AVG studies (Staiano & Flynn, 2014), the first *Kinect* camera was introduced in late 2010 and has been used as a SG input for both children and adults. Aside from gaming, it has also been piloted with children with severe impairments as a way of encouraging movement and expression by facilitating the creation of digital artwork (Diment & Hobbs, 2014). All three gesture or 'natural movement' systems (*EyeToy*, *Wiimote* and *Kinect*) promote upright gaming combined with gross movement of the upper limbs, torso and the whole body itself, with little emphasis or focus on fine motor control and coordination.

Figure 5-4 – The *Microsoft Xbox 360* Kinect camera (Source: https://commons.wikimedia.org/wiki/File:Xbox-360-Kinect-Standalone.png)

Staiano and Flynn's systematic review of all therapeutic uses of AVGs for all ages and across all populations up until July 2013 identified 11 of 64 studies involving children with CP (Staiano & Flynn, 2014, pgs. 356-7). The number of participants in each study ranged from one to 18 (109 participants in total) and the mean age, depending on how age was reported, ranged from 9.4 to 36 years (four studies) or from five to 15 years (three studies), and was not reported in four studies. The number of sessions per study (range: 1 – 146 sessions) and the length of the intervention (range: 32 minutes – 14 months) varied greatly between studies, with

few incorporating a follow up assessment after the intervention ended and few involving a RCT. The studies reported improvements in both fine and gross motor function, visual discrimination, hand function, finger range of motion and grip, balance, and an environment for providing moderate-intensity levels of activity. The authors reported that games that focused on a specific skill or deficit proved useful in terms of achieving a rehabilitation goal, including for CP studies (pg. 361), and that AVG interventions were accepted and enjoyed by all, with no serious adverse events reported. The low-cost, in-home nature of AVGs were identified as being appropriate for home-based rehabilitation interventions, as well as being very successful in terms of compliance and attendance, particularly for children (pg. 362).

Bonnechère *et al.*'s overlapping systematic review up until the end of 2015 identified 16 studies where a commercial gaming system was used with children with CP. The number of participants ranged from eight to 62 (329 participants in total), with interventions ranging from single sessions to 12 weeks (84 sessions) (Bonnechère *et al.*, 2016, pg. 278). Excluding single sessions, the mean intervention was 6.4 weeks (n=14, range: 3 – 12 weeks) and the mean number of sessions was 4.3 per week (n=9, range: 2 – 7 sessions). The most common system used was the *Nintendo Wii* (10 studies), and studies reported significant improvements in gross motor function, subject motivation and participation, balance, motor skills and movement control. Only one study reported on a quality of life measure (PedsQL or *Pediatric Quality of Life Inventory*) when used with a mixed cohort (aged six to 29 years) with acquired brain injury, reporting no statistically significant difference between scores at baseline and after 12 weeks (de Kloet *et al.*, 2012).

Given the significant content and thematic overlap between the reviews by Staiano and Flynn (2014) and Bonnechère *et al.* (2016), it is surprising that only six of the 11 studies identified by the former review also appear in the latter, especially considering all five studies that were overlooked used commercial gaming systems. Bonnechère *et al.* (2016) identified one study that was overlooked by Staiano and Flynn (Sharan *et al.*, 2012), and both studies overlooked the work of Li *et al.* (2009), who trialled a modified *Sony EyeToy* system within a hospital and home setting. This study investigated eliciting hand and arm movements of the hemiplegic upper limb, particularly reaching activities. Child and caregiver satisfaction levels, measured

through a questionnaire, were positive and indicated the system was fun and enjoyable to interact with.

Two studies that used the *Microsoft Kinect* with children with CP reported on fine dexterity outcomes, with one study reporting significant improvements for all 11 subjects following an eight week intervention (Luna-Oliva *et al.*, 2013), and the other reporting no significant improvements for 22 subjects following a four week intervention (Zoccolillo *et al.*, 2015). Zoccolillo and colleagues reported that SGs were more effective in terms of significantly and clinically improving upper limb skill quality (evaluated using the QUEST – the *Qualities of Upper Extremities Skills Test*), but that conventional therapy was more effective in terms of improving manual ability and activities of daily living (evaluated using the ABILHAND-Kids, pg. 673). Data revealed that the ND side was moved significantly less (compared to the healthy side) during the SG intervention, but that the difference was not significant during conventional therapy. However, the number of overall upper limb movements performed during the trial was three times higher during the gaming intervention (pg. 673), indicating increased engagement of the ND side. Zoccolillo *et al.* described SGs as "*… a feasible and well-accepted exercise to be performed by children with CP as a complementary strategy to CT [conventional therapy] in order to increase the amount of paretic arm movements*" (pg. 675).

More recently, Page *et al.* (2017) published contradictory findings following their systematic review of the benefit of AVGs on motor skill development for non-TDC and adolescents. Using the PRISMA (*Preferred Reporting Items for Systematic Reviews and Meta-analysis*) protocol, the authors identified eight studies involving children with CP, seven of which also appeared in the review by Bonnechère *et al.* (2016) (the eighth study, by Bilde *et al.* (2011), was omitted because it used a non-commercial gaming system) and three that also appeared in the review by Staiano and Flynn (2014) (with two studies omitted and three not included because they were published outside the search window for Staiano and Flynn). Page *et al.* reported significant improvements in balance (three studies) and the 10 minute walk test (one), but non-significant improvements for balance (two), upper limb coordination (one), the six minute walk test (one) and walking, running, and jumping (one). Page *et al.* also reported that the *Nintendo Wii* was the most common platform

used for both CP and all AVG studies (used in 75% and 74% of studies, respectively).

While not a serious gaming intervention, Choi and Lo (2011) combined VR technology with a commercial haptic interface, the *Phantom Omni*® (*SensAble Technologies Inc.*, Wilmington, MA, USA), to train and assess the hand writing of two seven year old children with CP. The aim was to investigate whether the provision of computer-assisted training and variable force feedback (through either a 'guiding' or 'dragging' influence, pgs. 1706-7) was feasible for improving the handwriting ability of children with CP when they drew a series of ten Chinese characters. Following the two week trial the authors noted that both subjects generally tended to increase their writing speed through repeated practice, and decrease their overall writing time after the intervention, suggesting an improvement in fine motor control and handwriting accuracy (pg. 1704). However, skill retention was not well achieved in the short period of the study and handwriting legibility appeared to improve slightly for only one subject.

The authors described a number of short-comings with their study, including the need for a quiet, isolated and "distraction-free" room (given the nature of the task, being handwriting), the need to capture and maintain the subject's patience and interest, and a counter-intuitive system interface issue that meant that the students didn't write where they were watching, which caused some confusion. While some metrics did improve, it's likely the study duration wasn't long enough to adequately train the children in a new skill, and the specific effect that the haptic device had on the subjects' improvement is difficult to ascertain.

5.1.2 Using Customised Gaming Systems for Serious Gaming Applications

The alternative to using a commercial gaming system for a SG intervention is to use a part or wholly custom-made or custom designed system. A wholly custom designed system implies that both the controller (hardware) and the gaming system (software) are custom designed, compared to a system where a custom controller interfaces with a commercial gaming system, or where a commercial hardware option interfaces with a custom gaming system.

A systematic review of 'interactive computer play' (ICP) for the motor rehabilitation of children with sensorimotor disorders by Sandlund *et al.* (2009) appears to be the first to examine non-commercial gaming systems as an intervention, while it wasn't a specific aim of their review (one included study used the *Sony EyeToy*). Using research design and methodological quality criteria established by the *American Academy for Cerebral Palsy and Developmental Medicine* (AACPDM), Sandlund *et al.* (2009) conducted their review between January 1995 and May 2008. The authors identified 16 studies that involved 257 participants (162 children with CP), and does not overlap articles reviewed by previously reported systematic reviews (Staiano & Flynn, 2014; Bonnechère *et al.*, 2016; Page *et al.*, 2017). The most common intervention length was four weeks (range: 3 – 12 weeks) and the mean session length was 60 minutes (range: 15 – 90 minutes) (pg. 175), with the authors noting that 13 studies (81%) reported positive effects, meaning the children found the computer intervention fun and motivating. However, Sandlund *et al.* (2009) concluded that the evidence for ICP as a rehabilitation intervention for paediatric sensorimotor disorders is scarce and inconclusive, noting a discipline dominated by case studies, uncontrolled trials, small sample groups, poor reporting (lack of inclusion and exclusion criteria), non-blinded assessments, and insufficient statistical evaluation and power calculations (pgs. 176-7). The authors noted the promise of the area and encouraged further, more rigorous research.

Prior to their 2016 review of commercial gaming systems for rehabilitation purposes, Bonnechère *et al.* (2014) conducted a review of all SG interventions for children with CP up until early December 2013. Using a structured *PICOS* (*Population, Intervention, Control, Outcome and Study* design) approach (pg. 1902), the review identified 31 studies that met the criteria, of which 18 used a custom SG intervention. The 18 studies included four RCTs, seven cohort studies, and seven single case studies, totalling 206 children, with more than half participating in a RCT (125 children). The authors rated the quality of the studies out of 32 using a published ratings scale (Downs & Black, 1998), reporting generally high mean quality for the RCTs (26.7 ± 2.5), but poor quality for the single case studies (11.6 ± 1.3). Two of the RCTs reported a difference between the SG and control groups – Akhutina *et al.* (2003), who reported improved spatial functioning using a virtual game environment coupled with supportive non-computer tasks, and Chen *et al.* (2012) who reported

increased knee muscle strength after using a VR cycling trainer coupled to a computer.

Across all studies reviewed, Bonnechère *et al.* (2014) reported a wide variation in SG interventions in terms of frequency of the intervention, the number of sessions and the duration of each session, and the types of games deployed. Bonnechère *et al.* (2014) noted the need to develop SGs that focused on one particular aspect of a rehabilitation program, and, recognising the heterogeneity of CP, recommended targeting a particular sub-group of children with CP. Drawing parallels to the evidence that underpins and supports current CP interventions, the authors concluded that the use of SG "*shows enough evidence to be included within conventional treatment of CP children since it proved to be efficient for increasing patients' motivation*" (pg. 1910).

Few studies report on the use of haptic gaming devices, with the exception of the *New Jersey Institute of Technology Robot-Assisted Virtual Rehabilitation* or NJIT-RAVR, which was based on the Haptic Master (Moog, The Netherlands), a six degree of freedom admittance-controlled robot (Qiu *et al.*, 2009, pg. 2). The NJIT-RAVR was trialled with two subjects with spastic hemiplegia, with one showing upper limb improvements following a three week trial (using the *Melbourne Assessment of Unilateral Upper Limb Function* and upper extremity range of motion test). However, the quality of this study was poor when assessed by Bonnechère *et al.* (2014), scoring only 12 out of 32.

Omitted from the above review is the work of Dunne *et al.* (2010), who developed a gaming system based on a large *Microsoft* interactive multi-touch display device, coupled with a wearable accelerometer to sense movement and rotation. The large display provided a visually attractive interface for the children to play with, creating an immersive and engaging environment to capture the child's attention while providing a platform to perform therapeutic exercises. The overall system was designed in conjunction with therapists and clinicians, with the authors reporting on a few of the custom games developed for the project, noting that they were coded to target both unilateral and bilateral tasks. The system tracked trunk flexion via the accelerometer, and when a pre-defined threshold for truck flexion was reached during gameplay – meaning the child was leaning forward to engage in the game

rather than extending their upper limb – an on-screen warning was triggered. The intention was that the warning would highlight that a compensatory movement or strategy was being performed (as the child may not be aware that they were doing it), and prompt self-correction. The authors do not report on the success or otherwise of the system when it was trialled to know how effective the system was in highlighting upper body compensatory strategies to the child, and if this caused behavioural change.

Similarly overlooked in the review by Bonnechère *et al.* (2014) was the work of Wade and Porter (2012), who investigated the ability of a custom gaming system with a unique controller to influence sitting ability in non-ambulant children with GMFCS classification Level IV or V. The study utilised a customised controller that detected centre of pressure movements using four pressure sensors sandwiched between two boards, such that when the centre of pressure shifted, the controller moved the game character in that particular direction. Using a randomised, cross-over trial, the small (n=13), unblinded study reported statistically significant improvements with respect to box sitting (namely, shoulder girdle position and spinal profile, which indicates a more upright posture) and five elements of the *Sitting Assessment for Children with Neuromotor Dysfunction* (or SACND), for both reach and rest phases. The authors concluded that their results suggest that engaging children in a meaningful therapeutic activity – one that requires coupling of upper body leaning to control a computer game – can help to improve sitting ability in children with CP.

More recently, Preston *et al.* (2016) reported on a six-week, home-based RCT that used a computer-assisted arm rehabilitation gaming technology with children with CP aged five to 12 years as part of a post-Botulinum toxin treatment program for spasticity management. The trial followed promising earlier pilot work of the custom technology within the home (Weightman *et al.*, 2011) and a school (Preston *et al.*, 2014). A pre-trial power calculation indicated the need to recruit 58 children in total to detect a large effect at 5% level of significance with 80% power (pg. 1007). The study used a control group that received usual post-Botulinum toxin treatment, with the intervention group receiving the gaming technology. Three assessment points (at baseline, six and then 12 weeks post-trial) formed the basis of the trial, with primary outcome measures being the ABILHAND-Kids questionnaire and *Canadian*

Occupational Performance Measure (COPM). Fifteen children were recruited to the trial, with eight children randomised to the intervention (gaming) group. The authors reported no group differences in mean ABILHAND-Kids scores across time points, but did report a statistically significant improvement in the COPM across time points (with children from both groups recording improved results), which was not greater than the MCID. This project deployed four games as part of the trial, with the mean level of system engagement being seven minutes per day. The most active user engaged for just less than 11 minutes per day on average, despite the researchers suggesting children should aim for 30 minutes of use per day. Post-trial feedback from children and families noted that the games were not as engaging as they could have been to promote high levels of sustained use. The researchers reported no adverse events and made a number of recommendations following their trial, such as providing competitive and collaborative play opportunities (through multiplayer or online gaming, so children can play against their peers in real time), introducing games in turn after a set period (to improve game longevity and interest), and reducing the burden for families who participate in similar trials (to improve questionnaire return rates and usage diaries). The authors concluded that their gaming technology did not appear to benefit arm function, but due to the low recruitment rate (15 children instead of 58), cautioned that a Type II error could not be ruled out.

5.1.3 Summary of Serious Gaming for Rehabilitation Applications

Summarising the SG literature to date, a strong theme is that SGs are successful in terms of being a fun, engaging and motivating intervention for a clinical population when used as an intervention akin to a rehabilitation program. Parents' perceptions of SGs in this context are positive (Sandlund *et al.*, 2012), as is end user feedback, with few adverse events reported. However, study designs (in terms of the length, duration, the number of sessions for an intervention, and the primary outcome measure(s) used) and results are variable and lack consistency. Reviews conclude that SGs are a "*highly promising area*" (Sandlund *et al.*, 2009, pg. 178), "*improve patients' motivation*" (Bonnechère *et al.*, 2014, pg. 1910), and "*at least as efficient as conventional therapy*" (Bonnechère *et al.*, 2016, pg. 287), yet caution that the evidence for positive effects is poor (Sandlund *et al.*, 2009), that the risk of bias is

great, meaning more robust research is needed (Page *et al.*, 2017), and that incorporation of SGs into a traditional rehabilitation program requires determining "*the ideal prescription for the duration and frequency of gameplay for each patient*" (Staiano & Flynn, 2014, pg. 362).

To the best of the author's knowledge, there are no publications available that report on the use of SGs (commercial or custom-made) to primarily influence sensory function for a neurologically impaired population, such as children with CP. All SG publications and applications to date have focussed on improving an aspect of motor function (for either the upper and/or lower limbs, including posture and balance), hence the novelty and contribution to new knowledge that is the current intervention to improve somatosensory function of the upper limbs for children living with CP.

5.1.4 Comparing Commercial and Custom-made Gaming Systems for Serious Games Interventions

For this thesis, the decision as to which SG system to use for the intervention was based on a few key factors, including the budget for the project, the local skill set available to work on the project, the timeframe for the intervention, and the specific aims of the intervention. Table 21 compares the two approaches that can be taken with respect to using either a commercial or custom-made SG system for a rehabilitation intervention, outlining the advantages, disadvantages, and key considerations for each.

Given the unique focus of this particular project – that is, piloting a SG system that provides specific contextually relevant afferent haptic stimulation to the hands of children with CP with a known somatosensory impairment, coupled with motivational gameplay, cognitive engagement, visual stimuli, and forced integrated bimanual upper limb use, a custom-made SG approach was chosen. The coupling of forced integrated bimanual use means two hands are always in contact with the controller, ensuring the ND hand is active and engaged during gameplay, and present to receive haptic input.

Table 21 – Comparing the advantages and disadvantages of using either a commercial or custom-made SG system for a rehabilitation trial

Serious Gaming Intervention Option	*Advantages*	*Disadvantages*
Commercial	**Cost** – commercial gaming systems and the associated games are relatively cheap compared to a custom designed solutions. In Australia, new gaming systems retail for between AUD$300-$500 (depending on the system, add-ons and hard drive size) and new games retail for between AUD$80-$100. Second hand systems can be significantly cheaper and still appropriate for trial use;	**Game appropriateness** – commercial games are made for mainstream entertainment use, and not with therapeutic or clinical goals in mind (Preston *et al.*, 2016). As Bonnechère *et al.* (2016) noted, the fact that commercial systems are having an impact as a rehabilitation intervention despite not being designed for an end user with an impairment implies that tailored solutions "*may lead to even better therapeutic outcomes*" (pg. 287). Customised solutions can target a particular movement, range of movements or activity, and reward the movement (Sandlund *et al.*, 2012);
	Acceptance – commercial systems have mass-market appeal, recognition, and profile. Consequently, playing with commercial systems represents the end user '*mainstreaming*' their behaviour and '*doing what everyone else is doing*', which is important for children with impairments. As Bierre *et al.* (2005) notes, gamers with a disability "*are consumers, and access to gaming is a quality of life issue*" (pg. 2);	**End user progress / trial statistics** – an important part of any device related clinical trial is the ability to track end user progress, interaction and use. Commercial systems generally can't be interrogated for end user performance to determine clinical improvement or deterioration, apart from game high scores, which only provides a single, coarse measure and is acknowledged as being insufficient in the literature (Staiano & Flynn, 2014, pg. 362);
	Quality – end user 'buy in' to the system and especially the games is a critical element in terms of motivation, enjoyment and long-term system use. Commercial games incorporate high quality 'movie-like' graphics, engaging story lines, and game detail that is difficult to reproduce at a research level for trial purposes;	**Software accessibility** – commercial games can be difficult to play at an optimal level if all the features cannot be accessed (through the controller), if the game isn't intuitive to play or follows a complex story line, requires fast or repetitive actions, or doesn't provide adequate auditory cues, as noted by Bierre *et al.* (2005);

	Availability – commercial systems are available online and in-store, with little to no waiting period once the decision is made to implement one in a study;	**Hardware accessibility** – commercial gaming controllers are designed for a mainstream market and are typically inaccessible for an end user with hand impairments;
	Reliability – companies that specialise in gaming systems (*Sony*, *Microsoft*, and *Nintendo*) spend millions on development and testing, meaning the systems rarely crash, cause conflicts between hardware and software, or don't work straight out of the box.	**Adaptability** – commercial games lack the ability to be adapted as a participant improves, which is an important consideration for any therapist who wishes to extend or provide new challenges to a participant as they improve, as noted by Staiano and Flynn (2014)(pg. 362).
Custom-made	**Software** – complete control over the games and gameplay means clinical or therapeutic outcomes and goals can be written into the game at the 'story-boarding' phase of the project and then implemented within the game to target a particular aspect of the rehabilitation (Bonnechère *et al.*, 2014). Known accessibility issues can be addressed from the outset, relating to graphics contrast and the colours used, sounds and how they are implemented, speed of the game, intuitiveness of the game, game scoring, game reward structure, game complexity, and game or level progression. Additionally, a custom solution enables control over the integration between hardware and software;	**Resources / skill set availability** – as is the case with many professions, staff with specific skill sets require training, experience and availability. Designing and developing an all-in-one custom gaming system requires skills that include game development, digital media and animation, computer science, information technology, software engineering, product/industrial design, and electronic engineering. When applied in a SG context with children with an impairment, a multi- and trans-disciplinary team should also incorporate biomedical/ rehabilitation engineering, physiotherapy, occupational therapy, paediatric rehabilitation medicine, and the user themselves to ensure the system is designed and developed appropriately;
	Hardware – a Universal Design (Story, 1998) philosophy and accessibility intent can be part of the design fabric from the outset, meaning accessibility can be maximised from the beginning, and unique access issues for a particular population can be addressed;	**Cost** – there is significant cost associated with designing a custom gaming system from scratch compared to purchasing a commercial gaming system (Preston *et al.*, 2016). In most cases the majority of costs relate to personnel with specific skills, as well as necessary costs for prototyping and product development;

End user progress / trial statistics – as the games are coded 'in-house' it is possible to capture, measure, store and then evaluate and report on any and all aspects of the overall system and individual games, as noted by Sandlund *et al.* (2009). This includes not only what is tracked, but how the information is collected, how often it is collected, and how it is presented to researchers and clinicians;

Flexibility and adaptability – a custom designed system affords the ability to tailor a given game to a particular user (either manually or automatically) depending on their progress or achievement within the game, their capability, or their rehabilitation program;

Co-design – to improve the appeal and acceptance of the system, a co-design approach can be adopted with the intended end user group for all aspects of the system to improve buy-in, motivation, acceptance, and to ensure the system is designed and tailored appropriately. Additionally, therapists and clinicians can be consulted and incorporated into the co-design process, to ensure the right movements or actions are being conducted.

Time – designing, developing, testing and trialling a complete custom gaming system is time consuming (Preston *et al.*, 2016) and can take years, not weeks or months, meaning the lead-time is long and significant resources are required to be directed towards project management and coordination;

Quality and appeal – one of the significant software trade-offs with this approach is the difference between a commercial quality game and one that is custom-made, including compromises with graphics, animations, sounds, game mechanics, game story lines, general game appeal and overall game polish and quality. This aspect is heavily dependent on the skills, talents and experiences of the staff who work on the project. Similarly, from a hardware perspective, the form, functionality and appeal of the controller needs to be high to ensure appropriate access and engagement from the end user;

Reliability – once complete, the overall gaming system needs to function as intended and meet or exceed the expectation of the end user to minimise frustration and annoyance. This means the system needs to be sufficiently robust, stable and integrate all components (the games, the controller, and the system that runs and monitors all activity) seamlessly.

5.2 Conceptualising, Designing and Developing an Integrated Accessible Custom-made Serious Gaming System

An integrated and accessible custom-made gaming system is one that promotes independent access through appropriately designed hardware, integrated with appropriately designed games that are engaging yet challenging, that does not require dexterous fine finger and thumb movement, control and coordination. In essence, the software (games) and hardware (controller) are designed in tandem and integrated to promote accessibility, realising that these two aspects are intimately coupled.

5.2.1 Computer Gaming System Requirements

Hand function is known to depend on more than just physical functioning, with Majnemer *et al.* (2008) noting it also depends on behavioural (concentration, attention), social-emotional (motivation), cognitive, and perceptual (somatosensory integration) components (pg. 142). Citing the work of Eliasson (2005), the authors stated that hand function in the presence of a sensory-motor deficit can be optimised through training strategies that capitalise on strengths in other component areas (pg. 142). The specific aim of this project was to design an accessible home-based SG system for a child with a hand impairment due to CP that encouraged and motivated active engagement, cognitive buy-in, required bimanual hand use, and delivered a range of afferent haptic stimuli to the child's ND hand that complemented and reinforced gameplay actions.

The specific requirements, specifications and features of the SG system are described in sections 5.2.2 to 5.2.11, namely, that the system needed to be home-based, use the Microsoft Xbox platform, adopt a unique 'no button' gaming philosophy, incorporate high quality games with appropriate appeal, deliver a range of haptic feedback to the child's palms and fingers during gameplay, provide haptic isolation between the left and sides of the controller, promote bimanual or two-handed use, incorporate a single button for menu selection, have high aesthetic appeal, and incorporate the Principles of Universal Design.

5.2.2 Home-based

Compared to a hospital or clinic trial, a home-based SG system isn't supported while on trial, requiring it to be both robust and tamper-proof. However, as Bilde *et al.* (2011) reported, more intensive and longer SG training sessions can occur while the child is at home due to increased convenience, since the child can use it in short bursts or for extended periods, without burdening families with additional travel. As reported earlier (section 5.1.1), both Staiano and Flynn (2014) and Bonnechère *et al.* (2016) reported on the value and merit of a home-based study in their reviews. The system was specified to be standalone and un-networked (offline), meaning families did not require an Internet connection for the trial. This requirement minimised the technology and connectivity expectations for families considering participating in the trial, and reduced the possibility of network errors, modem drop-outs, or program/operating system updates occurring during the trial, diminishing system performance and the child's experience.

5.2.3 Using the *Microsoft Xbox* Platform

Flinders University teaches Computer Game Development as part of its Computer Science and Information Technology Awards, and through an agreement with *Microsoft*, uses the *Xbox* system and XNA[7] programming language for teaching purposes. As all Flinders students and graduates are familiar with XNA as a platform for game development, this platform was chosen for the software side of the project to facilitate rapid game development. Games were initially coded using XNA 3.0, before upgrading to XNA 4.0 when the language was updated. Based on this decision, the hardware (controller) for the project was required to interface with a *Microsoft* technical package to avoid system conflicts and to facilitate effective hardware and software communication. This meant that all controller designs would need to be based on the internal circuit board of a *Microsoft Xbox 360* wired controller (Figure 5-5) to ensure functional compliance.

[7] See: https://msdn.microsoft.com/en-us/library/bb203894.aspx

Figure 5-5 – The *Microsoft Xbox 360* wired controller, on which the controller hardware needed to be based. The left thumb-stick is highlighted (dashed purple circle), as is the green 'A' button (purple arrow). (Source: https://commons.wikimedia.org/wiki/File:Xbox-360-Wired-Controller.jpg)

5.2.4 Game Development and Game Philosophy

The central principle that guided all game development for this project related to accessibility and game playability. As noted earlier, commercial gaming systems are inaccessible for many children with a hand impairment, with button activation a known issue due to the size, shape, and location of the buttons on commercial controllers. However, button presses are only required when the software requires or expects a button press to occur, with typical button actions being for shooting or jumping. From a design perspective, removing the requirement for a button press would remove the need for game actions to be mapped to one or more button activations.

Consequently, the central guiding principle for all game development was that all games should be coded such that button actions were *not* required. This meant that all game control, and hence all game actions, were based on joystick or controller movements *only*. When coupled with the eventual system controller, this paradigm encouraged sustained and integrated bimanual use that didn't require fine digit movement and/or control. Eliminating button control from all games provided a basis for game development that simultaneously required significant effort with respect to the design, conceptualisation, and development of the games to ensure features such as game appeal, re-playability, player interest and intuitive gameplay were part

of the gaming experience despite the modified control system. Every game developed for the project was required to conform to the following requirements:

- Game control via a joystick/controller only. Typically, but not always, this translated to only four control options for movement – forward, backward, left and right;
- Offer a variety of games, as per Li *et al.* (2009), that appeal to individual preferences such as game genre, styles, interests and age, to reduce the possibility of game fatigue;
- Provide an engaging experience where relevant game actions (such as collecting a reward or bumping into an object) produced a corresponding haptic event felt via the controller, thereby coupling a visual stimulus (the gameplay) with a reinforcing afferent stimulation;
- Ensure haptic events were: (a) often (specified as enabling the child to experience a haptic event at least once every 10 seconds of gameplay) and (b) contextualised to the particular game event, providing an opportunity for the child to experience a range of vibration intensities and durations while playing, and not a single, repetitive burst of vibration each time;
- Increase game difficulty and complexity to provide challenge and engagement, but at a slower rate compared to commercial games, while still providing a degree of challenge and sense of progression and achievement within the game;
- Integrate with a 'Central Games Catalogue' so that common system features (such as data logging, hand position monitoring and the ability to pause the system) could be coordinated centrally;
- Ensure all games are classified as 'G' for a 'general audience', as defined by the Australian Government Federal Legislation "*Guidelines for Classification of Computer Games 2012, Classification (Publications, Films and Computer Games) Act 1995*", dated 11 September 2012. This is the lowest, least impactful rating that a game can receive, and appropriate for the target audience for this study.

5.2.5 Game Quality and Appeal

It was acknowledged that the appeal (and hence attractiveness) of the overall system when on trial would be based on the quality of the games on offer and their ability to maintain the child's interest and enthusiasm while providing an immersive and engaging environment, as noted by Dunne *et al.* (2010). If the games were interesting, challenging and enticing, then the necessary buy-in would be achieved, with Harris and Reid (2005) observing that game variability, challenge and competition resulted in higher volitional scores (assessed using the *Pediatric Volitional Questionnaire*). The hardware (controller) would facilitate and promote physical access to the system but the games would be the reason the child ultimately used and engaged with it. An iterative approach to game development was adopted that included focus group evaluations with typically developing children and children with CP (section 5.3).

5.2.6 Maximise Afferent Stimulation to the Child's Palms and Fingers

The games were required to provide afferent cutaneous stimulation via haptic or vibration feedback via the controller, meaning the player could experience a tactile sense of the game during gameplay. Vibration sense is the modality that is most preserved in children with CP (Uvebrant, 1988), and vibration is known to activate the primary and secondary somatosensory cortices (Coghill *et al.*, 1994). Vibration feedback increases the realism of games by applying forces that are similar to or representative of those that would be felt if actually performing the task (Geerdink *et al.*, 2004), hence why commercial gaming systems incorporate haptic feedback into their systems to enhance 'game immersion'. Moreover, Orozco *et al.* (2012) noted that the overall gaming experience comprises physical, mental, social and emotional aspects, with haptic technology "*creating a deeper physical feeling of playing a game, improving the physical skills of the players, and imitating the use of physical artefacts*" (pg. 220). The form of the controller was required to maximise palm and finger contact during use to ensure that all stimulation was being delivered to the correct part of the open hand. Given the constrained game control mechanic described earlier (section 5.2.4), this meant that during gameplay the player's hands would always be resting on the controller (not pressing a button), so in a more receptive position to receive the vibration.

To facilitate rapid prototyping and development, and given that the controllers were based on an *Xbox 360* controller board, the afferent stimulation was to be delivered via standard vibration motors found in these controllers. Each *Xbox 360* contains two motors, one in each handle hold on each side of the controller, shown in Figure 5-6.

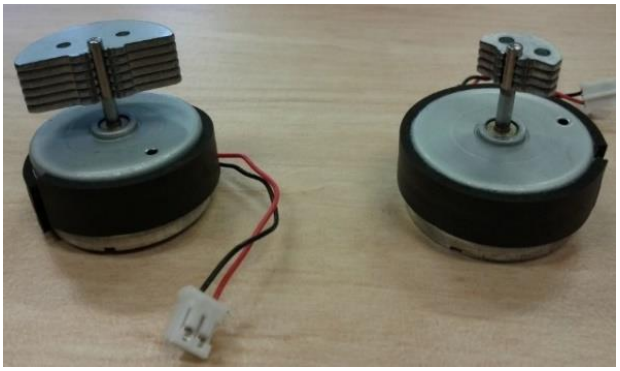


Figure 5-6 – The large (left) and small (right) counterweighted vibration motors taken from a commercial *Microsoft Xbox 360* controller

The motor on the right hand side of Figure 5-6 comes from the right hand side of the controller and has smaller counterweights compared to the motor from the left side of the controller. The two motors are used in isolation and in combination to deliver fine/smooth or rough/strong vibrations, depending on the desired game effect. The motor with the larger counterweights was used for this project to ensure that a large stimulus could be delivered to the child during use, considering it was being designed for children with a sensory deficit.

5.2.7 Haptic Isolation between the Left and Right Hand

An important clinical requirement for the controller design was to avoid the phenomenon of "*tactile inattention*" (Critchley, 1949) or "*sensory extinction*" (Brozzoli *et al.*, 2006), which is a failure to report a stimulus on the impaired side when a stimulus is simultaneously delivered to both sides of the body. Critchley noted that this phenomenon was particularly relevant when there is an injury to the parietal lobe (pg. 550). Consequently, the system, through the controller, was required to deliver afferent stimulation *only* to the ND hand of the child during use.

5.2.8 Promoting Bimanual or Two-Handed Use

One of the most important aspects of the project was to ensure the child always used both hands when using the controller, meaning the child would need to physically and cognitively engage and attend to their ND hand in order to use the system. Clinically, this introduced an element of upper limb bimanual integration – a way to engage the neglected ND hand in dynamic, purposeful activity, which is known to be an effective intervention for children with CP (Novak *et al.*, 2013; Shierk, Lake, & Haas, 2016). Additionally, for children with poor ND hand control, there needed to be provision for a strap or other mechanism that would position the ND hand appropriately on the surface of the controller to receive afferent vibration stimulation (section 5.2.6). Ideally, the child would self-manage this aspect to engender a sense of independence.

5.2.9 Incorporating a Single 'Out-of-Game' Button

Rather than employ a 'dwell' feature for system selections, a single 'out-of-game' button was specified for the controller, enabling the child to make a selection when they needed to (such as choosing a game from the main menu). The action of pressing a button to select a game shouldn't be game or time dependent, meaning the child is not penalised within the game if they take too long to complete this task.

5.2.10 High Aesthetic Appeal

With the system being deployed into family homes, it was hoped that children would not only use the system, but also take ownership of it and be proud of it. This aspect of the project was to be tested through focus group evaluation (section 5.3) and by involving children with and without a disability during the development process to trial, evaluate and critique the system. By adopting a co-design process, it embraced the '*nothing about us, without us*' philosophy of participation (Charlton, 1998), meaning the intended end users were co-designers in the process.

5.2.11 Ergonomic and Universal Design Considerations

The overall system needed to be independently operable by a child with a known hand impairment, meaning accessibility was a core requirement. Cook and Hussey (2002) describe the human/technology interface as the "*boundary between the human and the assistive technology*" (pgs. 44-45) – where and how the intended user interacts with the technology. From the outset, human factors insights and the seven Principles of Universal Design (*Equitable Use*, *Flexibility in Use*, *Simple and Intuitive Use*, *Perceptible Information*, *Tolerance for Error*, *Low Physical Effort*, and *Size and Space for Approach and Use*) developed by Story (1998) were used by the author to guide and direct the controller conceptualisation and development. Through professional links and networks, the author approached Industrial Design colleagues at the University of South Australia's School of Art, Architecture and Design[8] when it came to developing the controller (section 5.4).

While a focus of the form of the controller was to maximise accessibility and incorporate the Principles of Universal Design where possible, the author ensured these principles were applied to the project as a whole and not just the controller, as detailed in section 5.6.8 and published in the literature (Hobbs, Walker*, et al.*, 2015). Many children with a hand impairment can struggle with inter-limb coordination, control and movement when using their hands. Consequently, the controller was required to provide a stable base so that it didn't tip or rock during use, requiring it to be stable wherever it was used (dining room/kitchen table, wheelchair tray top, etc).

5.3 Software Development and Evaluation

The software for the system and the necessary computer games for the project were developed entirely by graduates and students of Flinders University who specialised in Game Development, and supervised by the author with co-supervision from Dr Brett Wilkinson (a Flinders University game development colleague). Starting in early 2011, all games were conceptualised, designed and developed by either the Research Assistant employed on the project or by undergraduate students for their final year Honours project in conjunction with and under direct supervision of the

[8] See: http://aad.unisa.edu.au/

author. Games developed by final year project students were supervised and tracked weekly or fortnightly by the author to monitor progress and academic milestones.

From a game development perspective, the team used Scrum-based agile software[9] engineering practices to rapidly prototype each game. The game evaluations provided feedback that assisted with improvements to the current games as well as the design of future games, particularly where the feedback was general and could be applied across other games. Game developers wrote and shared common sub-routines, such as those that enabled individual games to communicate with the Central Games Catalogue (section 5.6.1) and report information such as high scores, game time and the amount of vibration delivered, which provided coding and software consistency. This approach developed not only a strong sense of comradery within the software team but also a way of ensuring any minor or major code changes could be quickly and efficiently shared. From the outset, an important aspect was to seek external feedback on the quality and relevance of the games developed for the project. Consequently, a combination of meetings with the author coupled with the game evaluation sessions identified positive aspects and areas for improvement, which were then fed back into the development process to improve the overall outcome. As such, evaluation occurred in three stages: (i) formal and informal feedback from the author and within the team, (ii) formal feedback from typically developing children (section 5.3.1), and (iii) formal feedback from two teenagers with CP (section 5.3.2).

To facilitate rapid game development, given the games were developed at Flinders University and the controller was being developed and prototyped at the University of South Australia, all games were coded to respond to the left thumb stick and the out-of-game button was coded to respond to the green ‘A’ button (both highlighted in Figure 5-5). This meant game development could progress while the custom controller was being conceptualised, designed and developed in parallel, as the controller was required to replicate the joystick action of the left thumb-stick and to respond to presses of the green ‘A’ button.

[9] For an explanation of Agile and Scrum methodologies, see: https://www.qasymphony.com/blog/agile-methodology-guide-agile-testing/

To improve the attractiveness, appeal and longevity of the System, a suite of fifteen different games across different game genres and targeted at different age groups were developed. A brief overview of each game is provided in Table 23 (section 5.3.3), with all game credits/attributions provided in Appendix D. All fifteen games adopted the control mechanic discussed earlier (section 5.2.4), meaning the child kept their hands on the controller at all times while playing their game of choice.

To engage users actively in the game experience, appropriate interactive elements and a rewarding story progression was required. Sweetser and Wyeth proposed the idea of game enjoyment while outlining their '*GameFlow*' model and its eight core elements; concentration, challenge, skills, control, clear goals, feedback, immersion and social interaction (Sweetser & Wyeth, 2005). All eight elements were incorporated into the development of the games for the system, with 'social interaction' achieved by making the final system one that was shared within the family (using different player log ins) and through the system-wide high score table (which displayed who the leading scorers were for each game, section 5.6.1). Feedback was provided through visual, auditory and haptic methods, ensuring a coupling between the playing of computer games that are cognitively engaging with meaningful, contextually relevant and appropriate tactile sensory cues to the palms and fingers to assist with immersion and afferent stimulation.

Most games adopted the control mechanic that was specified, where forward, backward, left and right were the primary control inputs, like the major points on a compass. However, some of the games were coded to accept the full rotational range of 360 degrees as a control input, namely *Sunday Driver*, *Marine Life*, *Snake*, *Space Stuntz* and *Swimma* (for short descriptions, see Table 23). This meant that a movement of the controller that was midway between a movement that was 'forward' and 'right' – namely 'northeast' to use the compass analogy – would produce a movement in that particular direction. Where a game didn't use the full range of rotational inputs for control, the movement was interpreted as either a 'forward' or 'right' movement only. These two different methods of control input were dependent on the game that was being played, as for some games it didn't make sense to offer control besides the four basic control options. It also provided a degree of variety with respect to how a particular game is played and controlled, meaning it

challenged the child when they used the system to think about a logical way to control their game character.

5.3.1 Games Evaluation by Typically Developing Children

Due to the inherent inaccessibility of commercial gaming systems, children with CP can lack firsthand experience to draw on. Consequently, a typically developing cohort of the same age as the intended target audience was sought for the first step of game evaluation, recognising that some limitations are associated with this approach, such as the optimal timing of movements required to perform a game action, acceptable game speed, and assumed knowledge. This group of children without a disability would be in a position to evaluate and critique the games from two important perspectives:

- They would have relevant commercial games experience and knowledge, so would be able to make an informed comparison between the games for this project and commercial games – hence, they would be 'content' specialists in this area; and
- They would be familiar with using a commercial gaming controller having all inputs available to them for gameplay (that is, all buttons, thumb sticks and thumb pads). This means they could make an informed comparison between a commercial game and how it is controlled and the games for this project given the restrictive control mechanic.

The decision to trial the games initially with typically developing children was also driven by the fact that the accessible controller development lagged game development, and consequently was not available to be trialled at this stage.

Through personal networks and existing collaborations, a convenience sample of participants were recruited from two local schools for the game evaluations. The first round of testing involved 31 primary school students aged four to 13 years (15 females), and the second round of testing, conducted a year later, involved 17 high school students aged 14 to 16 years (8 females). Information packs about the study were provided to all families, and consent to participate was provided by the child's main parent/caregiver. Children could assent to the study if they were old enough to

read and understand the ‘plain English child’s version’ of the information sheet for the study. Permission to conduct the user evaluations was provided by Flinders University’s Social and Behavioural Research Ethics Committee (‘*User evaluation of custom-made and interactive computer games*’, project number 5234) for both schools, and the Government of South Australia’s Department for Education and Child Development Research Unit for the high school (‘*User evaluation of custom-made and interactive computer games*’, DECD CS/12/20.7) since the high school was a public (government) school. Both evaluations were conducted and coordinated by the author and Dr Wilkinson (Wilkinson & Hobbs, 2015; Hobbs *et al.*, 2018).

An unused classroom was utilised for both evaluations. This provided enough room for up to ten laptops to be set up while still being able to access mains power and suitable desk space. Each participant was assigned a unique two-digit ID as a log in. This two-digit number formed the first part of the filename for the log files that the system generated for each participant, allowing their gameplay to be tracked. Participants were assigned a laptop each and asked to play as many games as they liked in the time allowed, which was typically one hour. It was a conscious decision not to segregate participants during the evaluation as when the system is eventually deployed, it is expected the child enrolled in the trial will share and discuss the games with their family, peers and siblings to engender a sense of ownership, competition and buy-in. Allowing the typically developing game evaluation participants to play the games within one room meant that a sense of competition and comparison could occur.

Each round of game testing and evaluation provided an opportunity to also test a number of aspects of the overall system, apart from the games themselves. These included the performance and stability of the laptops chosen for the trial; the stability of the games catalogue, at that point in development; the performance and accuracy of the data logging system; and the stability of the overall system when running for up to 5 hours continuously.

The laptop chosen for the trial was an off the shelf *HP ProBook 4730s* (Intel Core i5 (2nd Gen) 2450M / 2.5 GHz, AMD Radeon HD 6490M, 1GB GDDR5 SDRAM graphics card, 17.3” display). This device was chosen because it was Windows

based, had enough processor speed to run the games, a dedicated graphics card, and a larger than standard screen (17.3 inches) to facilitate player immersion. Each laptop was loaded with identical software and a USB-corded *Microsoft Xbox 360* controller for Windows was used to control the games. All joystick control was routed through the left thumb-stick, and the green 'A' button represented the out-of-game button.

Beside each laptop was a set of simple one-page instructions for each game, which briefly identified the theme of the game and the controls required to play it. The instructions were prepared for the average aged student playing the game and used few words and mostly images to indicate how to play a particular game. Some of the younger children required additional support to understand some of the game requirements (that is, the instructions were read or more fully explained to the younger children). The primary school cohort tested and evaluated the first six games developed for the project (in August 2011), whereas the high school cohort tested and evaluated eight games one year later (in August 2012) – six new games and two from the earlier primary school evaluation. Figure 5-7 shows a photo from the high school evaluation.

Figure 5-7 – A group of high school students during the games evaluation

Participants were allowed up to one hour to play the games. For the primary school cohort, this was a continuous hour of play, but for the high school cohort the hour was divided into two half hour sessions with an alternate paper-based activity being conducted in a separate room. The paper-based activity asked students to brainstorm, develop and story-board their own game, from any genre, that used the same controls and game development philosophy as this project.

At the beginning of each evaluation session the author provided an introduction, which included a background to the project and the expectations of the session. A verbal summary of the games was provided for the primary school cohort to provide the participants with a context for the games they were to play. Participants were allowed to select the order of play that suited their game interests, however, they were encouraged to play all six games and were able to go back to previous games they enjoyed toward the end of the session. The game designers and developers were not present during either evaluation session. Prior to each session starting, participants were asked a series of background questions asking their age, sex, how often they played games, the devices they used, the genre of games they enjoyed, and what their favourite games were via a custom '*Participant Evaluation Form*' (see Appendix E, side 'A').

Side 'B' of the '*Participant Evaluation Form*' (Appendix E) listed questions that addressed a range of game evaluation aspects, such as whether they enjoyed the games and if they needed to read the instructions to be able to play them. Responses to questions that produced a 'yes' or 'no' answer were collated and presented as a percentage that agree with the particular question being asked. Participants were also asked to rate their interest in the game they were playing (out of ten), meaning average ratings per game and an overall rating could be calculated. All comments and feedback were given to the individual game designers for consideration and possible incorporation into future game development.

For the primary school cohort, the author and Dr Wilkinson circulated the room asking participants their impressions of the games by asking each child questions from the '*Participant Evaluation Form*' as they played them. Responses were written directly onto the evaluation form as the child responded, and then transferred into an *Excel* spreadsheet post-evaluation for analysis. Due to time restrictions, each

primary school participant's response was recorded for at least two of the six possible games they played. For the high school cohort, each participant recorded their own responses to the questions on the sheets provided, and they were encouraged to do this after they had played a given game enough times to be able to critique it.

During gameplay, player activity was logged and stored locally on each laptop. The information logged included the game length, *X* and *Y* locations of the thumb stick, and the duration and intensity of vibration events. The system created an individual log file for each game per player, where the student's unique two-digit code formed the first part of the log file filename.

5.3.1.1 Games Evaluation Results – Typically Developing Cohort

An analysis of the log files indicated that the primary school cohort (n=31) played a total of 362 individual games (average number of games per student: 11.7; range: 4 – 21 games), while the high school cohort (n=17) played 246 games (average: 14.5; range: 6 – 28). The participants provided a wealth of qualitative feedback and information on all the games, including comments on the graphics and artwork quality, sounds, game storyline, opportunities for improvements such as including 'power-ups' or extra lives, and what aspects of a particular game they liked and didn't like.

Across both evaluations, most participants described the integration and use of haptic vibration feedback to complement gameplay as being 'good', where 'good' was the highest possible response. This particular question helped to validate the process that was used to ensure the haptic feedback was both appropriate and meaningful for all games. Responses to game evaluation questions that could be quantified are shown in Table 22. As indicated in the table, both cohorts enjoyed the game evaluation, with most games rated as being enjoyable (88%) and showing high replay-value (85% for the primary school cohort compared to 77% for the high school cohort). The largest discrepancy between the two cohorts was in response to the question: "*would you buy this game if it were available in a store?*". Nearly three-quarters of the younger cohort said they would buy the game, compared to 38% of the older cohort. This discrepancy can be explained by the fact that most of the older

cohort were game-savvy enough to know that the games for this project were based on similar games available on the market at no cost. As one participant said: "*I like the game a lot, but why would I buy it when I can download it for free?*". Additionally, younger children wouldn't actually buy the games, their parents would, so this question may not have had as much relevance to the younger cohort.

Table 22 – A summary of responses to sample questions asked during the two game evaluations with typically developing children (n=48)

Evaluation Question ('yes' response only)	Primary School cohort (n=31)	High School cohort (n=17)
Did you enjoy playing the game?	88%	88%
Would you play the same game again?	85%	77%
Would you buy the game if in a store?	74%	38%
Average interest in game (/10)	7.3	7.0
(Range)	(5.9 – 8.7)	(4.5 – 8.2)

Note: The table above appears in Hobbs *et al.* (2018), which is currently *in press*.

5.3.2 Games Evaluation by Children with Cerebral Palsy

At two different time points during the game development process (December 2011 and March-April 2013), two teenage children with CP volunteered to test and evaluate the games in an extended, home-based trial. Both children came to know about the overall project through participating in Stage 1 of the study, and both expressed an interest in being 'testers' for the games as they were being developed.

The first trial was undertaken by a 13 year old girl with spastic hemiplegia (left side dominant, MACS Level 2), and the second by a 14 year old boy with diplegia (right side dominant, MACS Level 1). Permission to conduct the user evaluations was provided by Flinders University's Social and Behavioural Research Ethics Committee ('*User evaluation of custom-made and interactive computer games*', project number 5234 for the teenage girl, and '*Testing and evaluating an interactive, haptic, computer gaming system for children with cerebral palsy*', project number 5930 for the teenage boy).

As the controller was still in development, both children were provided with a standard Windows *Microsoft Xbox 360* controller with all joystick control routed through the left thumb stick and the green 'A' button, identical to the earlier school

evaluations. The teenage girl was able to use the controller confidently due to her left side being her dominant side, and the teenage boy had a mild left side impairment. Hence, both teenagers were able to use a standard *Xbox* controller without duress or discomfort, and neither child reported a problem during their trial. The teenage boy had previous gaming experience and enjoyed car racing games (using a steering wheel instead of a standard controller and foot pedals, both of which are commercial accessories for gaming systems). The teenage girl had limited games experience.

5.3.2.1 Games Evaluation Results and Feedback – Children with CP

The first participant played 69 games on 12 days over two weeks and her favourite game was *Space Stuntz*, which she played 24 times. *Driving Maniac* was her second favourite game (21 games), and *Marine Life*, admittedly targeted at much younger children, was her least favourite game (played three times). An attempt to further analyse the log files post-trial identified that the logging system malfunctioned during the trial – game instances were recorded, but game detail (game duration, vibration received, joystick position, etc) was not. Post-trial, the logging system was reviewed and changed to avoid the error happening again.

The second participant played with the gaming system sporadically, playing 78 games on only three days over a three-week period. The total amount of time spent using the system was 76 minutes and 23 seconds, which comprised 67 minutes and 28 seconds of gameplay and 8 minutes, 55 seconds minutes spent in the system menu. The longest time spent playing one game was 7 minutes, 53 seconds (*BiPlane 1922*) and *Snake* was the game that was played the most (six instances). The total amount of vibration that was delivered was 15 minutes, 45 seconds, of which 49 seconds was delivered when navigating the menu system and 14 minutes, 56 seconds was delivered during actual gameplay. A written assessment and detailed feedback was received from the first participant only, who rated the system highly. The second participant didn't complete the formal assessment due to school commitments, which helped to explain why they only played with the system for three out of 21 days.

The first participant reported the games as being "*exciting*", "*mostly creative*", "*quite fun*", and described the game vibration events as "*very creative and easily felt*". She

identified spelling errors within the games that needed to be corrected and that some games needed more work as they "*were not very good*". However, for games she enjoyed, she commented, "*that you wanted to try to beat the previous score that you have achieved*", indicating a degree of replay-value for some of the games. Interestingly, this participant also reported that the controller she was given for the trial was "*the easiest controller for games I [have] dealt with while playing games online*". Given this participant was using a standard *Xbox* controller, this comment is presumably reflective of the modified control inputs required to engage with the system and the fact that button presses weren't required, coupled with this participant's limited gaming experience. The participant concluded by saying she "*had the best 2 weeks of my life playing the games, from early morning to afternoon*". The participant's mother reported anecdotally that her daughter preferentially chose to play games on the system instead of her normal favourite past time, which was competitive swimming.

5.3.3 Final Software System Design

All feedback and comments from the games evaluation sessions were collated into *Excel* and given to each individual game developer and the project Research Assistant to improve individual games and the overall system. The author would routinely review, critique and suggest changes to the games after a student game developer finished working on the project (for their University credit), working with the project RA to improve the overall quality and to ensure that feedback from each evaluation was incorporated. Not every game that was initiated was incorporated into the final Games Catalogue, with the author not choosing a particular game if it was not mature enough, still had bugs, or lacked appeal (in the author's opinion). At the end of the game development phase, 15 games met the approval of the author and were included in the final Games Catalogue. Each game is briefly described in Table 23, with full game credits and attributions (for both game development and game artwork) in Appendix D.

Table 23 – An overview of the games that were developed for the project (for full game credits and attributions, see Appendix D)

Game name	Brief game overview / summary
A Bridge Too Far	Similar to 'Temple Run', the main character has to navigate an endless pathway, jumping gaps and collecting gems and coins.
Alex Adventure	This side-scroller game has the main character, Alex, explore themed landscapes while collecting carrots and avoiding obstacles.
Alien Attack	Similar to 'Space Invaders', the player must cleanse each planet in the solar system of alien spaceships.
BiPlane 1922	This 3D flight simulator has the player fly over an English countryside, avoiding obstacles while navigating through farm barns. Levels are presented from different perspectives, such as the cockpit and chase-cam.
DragonFly Dodge	This side-scroller game has the main character, a dragonfly, fly over a stream collecting coins while avoiding frogs, reeds, birds and rocks.
Driving Maniac	This vertical-scroller game has players avoiding obstacles and challenges on the road, such as cars, road works and lane changes.
Sunday Driver	This 3D exploratory driving game has players searching for hidden objects and avoiding enemies, before progressing to the next world.
Marine Life	This swimming game has players attempt to move up the food chain by eating other underwater creatures while avoiding predators.
Move Gravity	This puzzle game requires players to combine multiple asteroid masses in space to form a single mass, taking into account gravitational forces and black holes.
Planet Fall	This action game requires players to control a laser and rocket-shooting moon lander, to stop meteorites from reaching the ground.
Snake	Similar to the 'Snake' game on Nokia mobile phones, players must move a snake around the screen, trying to eat as many objects as possible while avoiding running into themselves or the screen edges.
Space Stuntz	This 3D space ship simulator has players zoom through an endless tunnel of rings while avoiding asteroids and other objects.
Squirrel	This running game has players control a squirrel as it climbs a never-ending tree, collecting coins and avoiding tree knots and branches.
Swimma	This side-scroller game requires players to control a snorkeler, collecting as gems and air bubbles, while avoiding predators.
The Fancy World	This dress-up game challenges players to suitably dress their character for a given event, such as going to the movies or the beach.

Note: The table above appears in Hobbs *et al.* (2018), which is currently *in press*.

With respect to the main menu screen for the system, two different interfaces were explored. The first used a rotating carousel, with games being selected by moving either left or right around the carousel until the game of choice was highlighted. Each game was represented by a cube and the 'game cube' was wrapped in artwork from the game to convey a sense of what the game was about. The second, and final, main menu screen adopted a more intuitive and contemporary interface, shown in Figure 5-8. This interface used a four by four grid or matrix to present all 15 games to the child at once, using the same 'game cube' technique as the earlier interface. A smartphone on the right hand side of the main menu serves two purposes: showing a short video preview of a selected game (top half) and the high score table for a particular game (bottom half). When the 'Incentivised Games Catalogue' was developed (section 5.6.6), the smartphone also kept track of the time left until a game would be available (Figure 5-21).

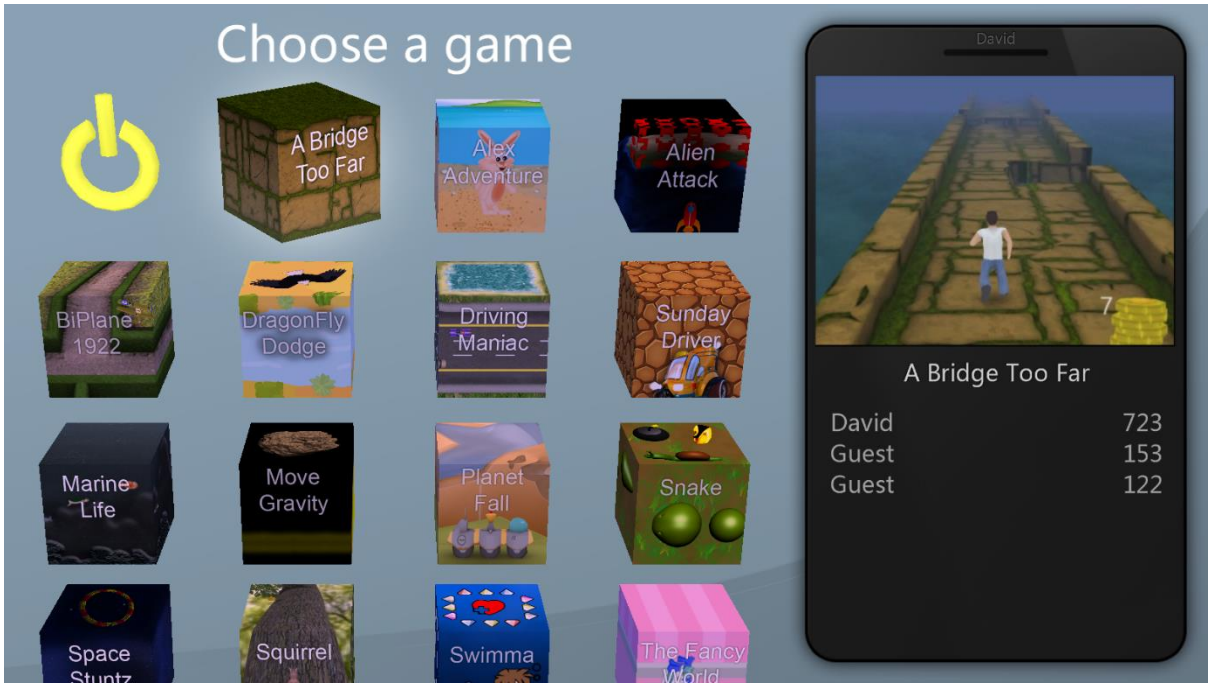


Figure 5-8 – The main menu screen and final version of the Games Catalogue, showing all 15 games developed for the eventual trial. Each game is represented by a cube that is wrapped in artwork from the game, providing a visual indicator of the game. Additionally, a smartphone on the right hand side of the main menu shows a short video preview of the game that is highlighted (in this instance, '*A Bridge Too Far*'), in combination with a high score table for that particular game (Source: Brad Wesson)

5.4 Designing, Developing and Evaluating an Accessible Gaming Controller

The design of the gaming system controller was conducted at the University of South Australia (UniSA), within the Industrial Design section of the School of Art, Architecture and Design. The engagement began with the author presenting the design brief for the project to interested students at a closed-session to provide an overview of the project and the necessary detail relating to accessibility requirements and hardware specifications listed in section 5.2.1, and the need to interface with the current game development work. The author co-supervised and co-directed the project as an external supervisor alongside A/Prof Sandy Walker at the UniSA.

The design, development and testing of the controller followed three distinct stages:

- Stage 1: an initial four month (short term) pilot project involving four students (August – November 2011),
- Stage 2: a more focussed and in-depth (long term) project at the Masters level, involving two students over a full academic year (2012), and
- Stage 3: the refinement of the final design for the RCT involving a new graduate over a 10 month period (January – October 2013).

5.4.1 Stage 1 – Initial Pilot Project

After the initial presentation by the author, four students chose to work on the project, each conceptualising a distinct design idea for the controller. Each student was given feedback and direction in terms of their particular design, and then presented their results and a working prototype to the author at the conclusion of the four month pilot project. The design that had the closest agreement to the design brief is discussed in the following sections, with the three alternative designs (referred to as Designs 1, 2 and 3) appearing in Appendix F. All four designs were based on a *Microsoft Xbox 360* controller to allow communication with the games being developed.

The inspiration for the preferred controller design (by Max Hughes) was a well-known item of assistive technology, a Trackball Mouse[10]. Shown in Figure 5-9, this particular design was simple, clean and intuitive. Using the controller required the child to place their open hands onto either side of the spherical ball (either side of the central grey strip), and rotate the ball in the direction they wished their game character to move. Vibration motors were mounted to the inside of each hemisphere, underneath where the hands would be placed.

(a) **(b)**

Figure 5-9 – The preferred pilot project controller design: (a) Computer Aided Design (CAD) model (Source: Max Hughes), and (b) working prototype being used

This controller design was appealing and had many positive aspects, particularly:

- A very simple and intuitive design form that was readily understandable;
- Excellent functionality – with the hands and arms in a neutral position when seated, this controller design only required very simple and easy wrist and forearm movements to control the game character;
- The form maximised an open hand in contact with the curved controller surface, meaning the player's palms and fingers were in contact with the controller as required to maximise afferent haptic stimulation (section 5.2.6);
- The sphere self-centred when released;
- The design promoted upper limb coordination and coupling as both hands were required to work together to produce either identical or opposite

[10] See: http://www.trackballmouse.org/

movements for the controller to function correctly. That is, when moving forward/backward the movements were identical for both hands, but when moving sideways one hand would move up while the other moved down, and vice versa;

- Good to very good haptic vibration isolation delivered via the motors mounted onto the inside surface of each hemisphere;
- The white hemispheres of the controller were attached to the central grey joystick mounting with magnets, meaning access to the internal electronics and motors was quick and easy, should tool-less access be required. When hands were placed on the two hemispheres during gameplay, this also meant the controller was positively locked; and
- Excellent stability in all directions – this particular design had the most stable base (a requirement, as detailed in section 5.2.11) because all movement and control occurred over the base of the controller at all times due to the ball being mounted on a central pivoting joystick.

The drawbacks of this particular design were:

- The out-of-game button, while contrasting in colour (red on grey in the Computer Aided Design (CAD) model), needed to be larger, prouder, and more obvious. For the prototype, the button was connected but not mounted on the circular base (as can be seen in Figure 5-9(b)) due to time constraints;
- The controller could be used with one hand placed on top of the controller, straddling the two hemispheres, yet bimanual use (section 5.2.8) was a requirement; and
- Not intuitively knowing where to place your hands when using the controller. The utility of the controller could be improved if hand position ‘locators’ could be added to the spherical design to intuit where hands should be placed.

The uniqueness of this particular design – namely, the way that it encouraged open handed use, bimanual coordination and coupling (when used with both hands), a very stable base, and intuitive hand and wrist movements – was encouraging. This design was short-listed to progress to the next stage of the project.

5.4.2 Stage 2 – Further Controller Design Iteration

The author invited the student designers for both the preferred controller design and Controller Design 1 (Appendix F) to continue and to extend their pilot designs into the following academic year as the major project for their Masters of Design (Industrial Design) project. Figure 5-10 shows how both initial prototype controllers would look if they were deployed for a trial.

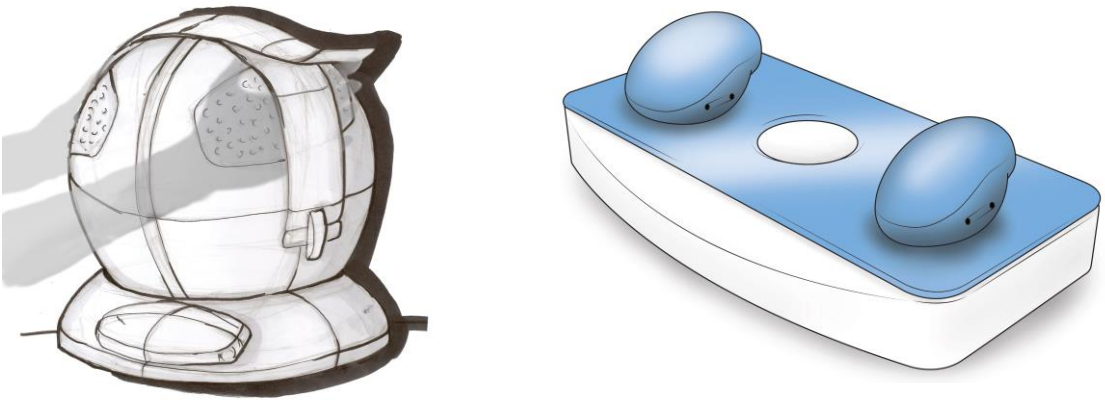
(a) **(b)**

Figure 5-10 – Two of the Stage 1 initial pilot controller designs, (a) The preferred design and (b) Controller Design 1, as they would look if deployed for a trial (Sources: Tom Whitby)

The first step of Stage 2 was to conduct a review of the controllers, individually and with the author. This involved revisiting the problem statement and analysing how each design met the projects requirements (section 5.2.1). A 'paired comparison analysis' was undertaken to identify where design time, energy and resources are best directed during an iterative design and development process. The analysis highlighted the importance of each of the functional requirements of the controller, indicating that, in rank order, two-handed use, isolated haptic feedback, USB connection to a laptop, robustness, an accessible out-of-game button, support for the ND hand, and a joystick that self-centres were key areas to focus on. The UniSA uses a structured five-stage Industrial Design Project 'Stage-Gate' New Product Development Process (Walker & Hobbs, 2014) to steer and supervise projects to completion, and A/Prof Sandy Walker and the author closely supervised each stage.

During Stage 2 both controller designs were routinely reviewed, assessed and supervised by A/Prof Walker and the author. At key milestones (such as 'Stage-Gate' presentations) broader feedback was solicited from other professionals within the team, namely the PhD supervisors of this project (Prof Reynolds, A/Prof Hillier and A/Prof Russo), the assessing therapist from the WCH, Dr Wilkinson, and the RA who was grant funded for the game development part of the project. This was to ensure that a broad multi- and trans-disciplinary audience was able to provide input and direction given their respective backgrounds and expertise.

Modifications to the preferred controller design included a streamlined base, providing an indication of where the player's hands should be placed (hand pads), a more accessible out-of-game button, and a way to support the ND hand (Figure 5-11(a)). The underside of the hand pads became the mounting points for the motors to deliver haptic feedback, ensuring proximal and targeted vibration to the hands. The initial inspiration for Controller Design 1 was computer peripherals, but through re-imagining the interface, the student drew inspiration from indoor rock climbing handholds to modify the look and shape of the design. The controller base was also streamlined from a large circle to a rectangular shape, and the contrasting out-of-game button was placed centrally between the two handholds (Figure 5-11(b)).



(a) **(b)**

Figure 5-11 – The Stage 2 designs of both controllers: (a) Preferred design (Source: Max Hughes) and (b) Design 1 (Source: Tom Whitby)

5.4.2.1 Controller Evaluation by Children with Cerebral Palsy

An important part of any design and development within the disability and AT sector is evaluation by the target audience, and it was also a specific stage in the 'Stage-Gate' process (Stage 4). Approval for the evaluation with children with CP was obtained from the UniSA's Human Research Ethics Committee (ethics protocol number: 0000029359, title: 'Cerebral Palsy Game Controller') in August 2012.

Due to time restrictions relating to the academic year and milestones that both students needed to satisfy, children with CP that were known to the supervisors of this research were approached and invited to participate. Three teenagers with CP agreed to participate and were booked into sessions on two separate days. On the first evaluation day (Day 1), only one of the two participants attended, meaning the user evaluation was conducted with two teenage children with CP, who both had commercial games experience, and who both participated in Stage 1 of the study (Chapter 4). One participant was female (MACS Level III, hemiplegic CP, dominant side = left, age = 14 years 11 months) and the other participant was male (MACS Level 1, diplegic CP, dominant side = right, age = 13 years 10 months). The second evaluation, Day 2, was ten days after Day 1 due to participant availability.

The sessions began with an introduction to the project by the author, a brief presentation by the two students on their controller designs using CAD models, and an opportunity to trial each controller. Participants were seated at a table in an unused classroom, with the controller and laptop placed in front of them, as shown in Figure 5-12. During the controller trial phase, pre-prepared questions about the controller design that solicited the participant's feedback, thoughts and feelings were asked (see Appendix G). Controller use, ease of use and hand position were studied. The sessions were conducted separately to eliminate comments or observations from one participant biasing or leading the other, and each session lasted between 30 and 45 minutes, depending on the participant. The author led the evaluation sessions while the students observed proceedings, took photos, and asked questions relating to their particular controller design.

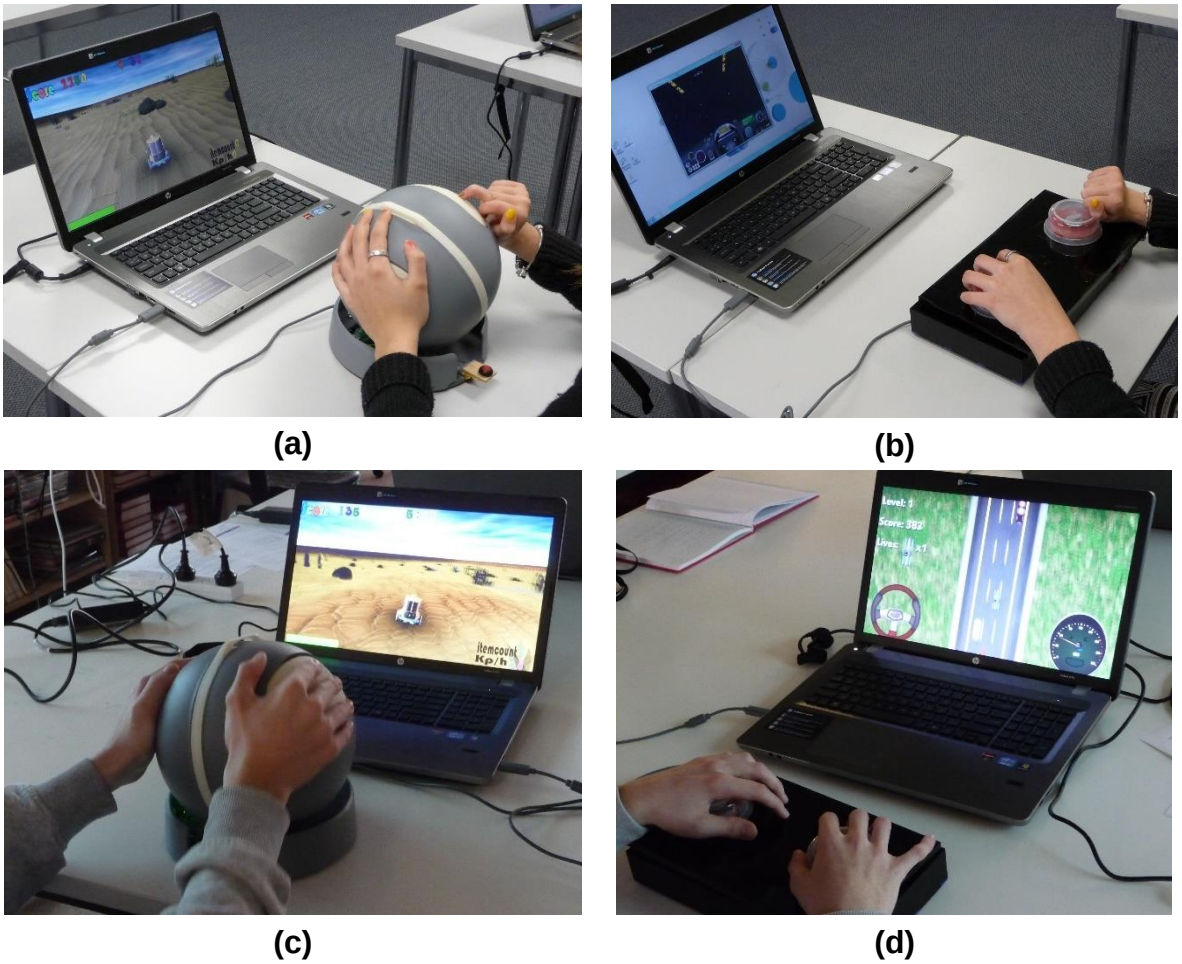


Figure 5-12 – Photos from the controller evaluation days. Day 1, a participant with CP using (a) The preferred controller, and (b) Design 1; and Day 2, a participant with CP using (c) The preferred controller, (d) Design 1 (Sources: Tom Whitby)

The evaluation sessions were scheduled at a time when both designs could be presented beyond the conceptualisation phase and at the early prototype phase, to maximise the ability to incorporate feedback from the target population. This meant that both controllers used 'mock prototypes' for the evaluation as neither design was mature. Both mock prototypes functioned as required but differed in look and feel compared to what the final prototype design would eventually be. This difference was communicated to the participants by drawing their attention to CAD model imagery of the intended designs shown on a large screen during each trial (sample images shown in Figure 5-13). The images showed the controller in different colours, how possible ND hand straps could be attached, and the possible form and location of the out-of-game button. Both participants were asked to comment on the use of

the controllers while playing with each mock prototype, and on the aesthetic and visual appeal by studying the CAD models.

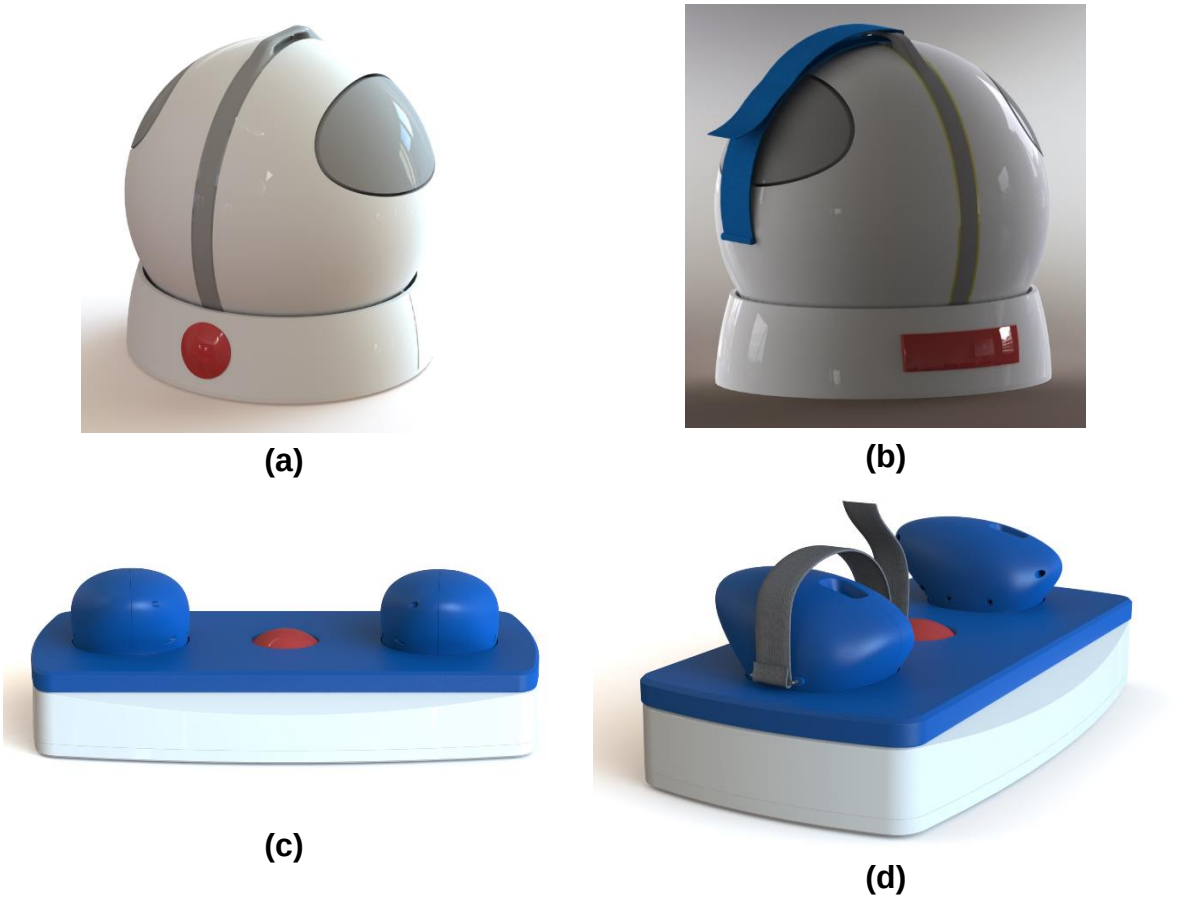


Figure 5-13 – Examples of the CAD models shown to participants during the controller evaluation; (a) The preferred design without hand strap, (b) The preferred design with hand strap, (c) Design 1 without hand strap, and (d) Design 1 with hand strap (Sources: Max Hughes and Tom Whitby)

Both participants thought the controllers looked simple and easy to use, using descriptive words such as "*novel*", "*creative*", "*new*", "*different*" and "*great*" for both controllers. Both participants indicated that they wanted to buy the controllers, with their parents commenting that if they were robust and performed as desired, they would spend "*around AUD$100 or more*" if they thought it would help their child play computer games. A pragmatic condition of the parents' comments related to the controller actually working for their child – that is, if it was *accessible* for their child and *worked* for their child, then they would buy it. Colour, colour combinations, durability and a compact design were highlighted as important features of both designs by both participants and parents.

The preferred design's spherical form was identified as being more "*instantly natural*" and "*intuitive*". During the evaluation the research team noticed that both participants were able to play and perform well with both controllers when playing 2D games. However, there was a stark difference in performance and gameplay when 3D games where played (such as *Space Stuntz*). The preferred design's spherical controller was much more intuitive, natural and easier to use, with the perception of depth created by the game more readily mapped to the controller movements. When using Design 1, which utilised sliding planar movements for control, both participants tried to twist, rotate or lift the top plate from the base when attempting to control their 3D game character. This resulted in the mock controller breaking, with the top plate detaching from the base.

5.4.2.2 Controller Evaluation by the Supervisory Team

Following the evaluation sessions with children with CP, the PhD supervisory team, A/Prof Walker, the assessing therapist from the WCH, Dr Wilkinson and the game developer for the project assessed and trialled both prototype designs. The preferred design's operation was logical, smooth, and intuitive. The child simply rested their hands on the oval pads on the outside of the sphere, which placed their wrists, hands and fingers in a neutral position when sitting in front of the controller. The pads used a 'floating pad' design for haptic isolation that also delivered vibration stimulation via motors mounted on the underside of the pads, directly under the hands. The controller self-centred, making it intuitive and easy to use. The contrasting out-of-game button was easily located on the front of the controller and the form of the controller removed the need to form a hand grip. The strap design for this controller held the ND hand against the oval pad through a central slot at the top of the controller, which was relatively easy to self-administer and tighten if already located on the controller.

Design 1's planar sliding control was non-intuitive to use, especially when playing 3D games, despite a novel sliding mechanism design that meant the motion was smooth to operate as well as being self-centring. The handholds were comfortable but required a hand that could grip around an object to use the controller effectively. The contrasting out-of-game button located midway between the handholds was easy to

access, and the strap design for this controller was easy to independently use and understand.

Comparing the two designs, the operation and spherical form of the preferred design was chosen for the eventual trial due to its smooth and intuitive use, and the fact that both end users found it more natural to use and rated it higher. The final CAD models and working physical prototypes for both designs are shown in Figure 5-14. Owing to its spherical or ‘orb’ shape, the preferred controller design was nicknamed ‘Orby’. Both controllers were showcased to the public as part of an end of year UniSA Exposition, where attendees could play a few select games using the controllers. Both controller designs won prizes on the night for design excellence.

Two of the most challenging design requirements for the controller related directly to two of the most important clinical criteria for the project: haptic isolation to avoid sensory extinction (section 5.2.7) and ensuring bimanual upper limb use at all times during gameplay (section 5.2.8). Both these issues were studied during the end user evaluation sessions and tested by the author with each prototype. The requirement for haptic isolation was challenging because it required delivering yet quarantining haptic vibrations to one particular side of the controller, while dampening them from reaching the opposite side. With respect to bimanual use, the form of the controller was used to intuit and direct how the child used the controller, but Design 1 explored using short-range proximity sensors mounted into the handholds to detect the presence of hands on the handholds, and incorporated this feature into the final design. The recesses that house the proximity sensors can be seen in Figure 5-14(c) and (d), indicated by the purple arrows.

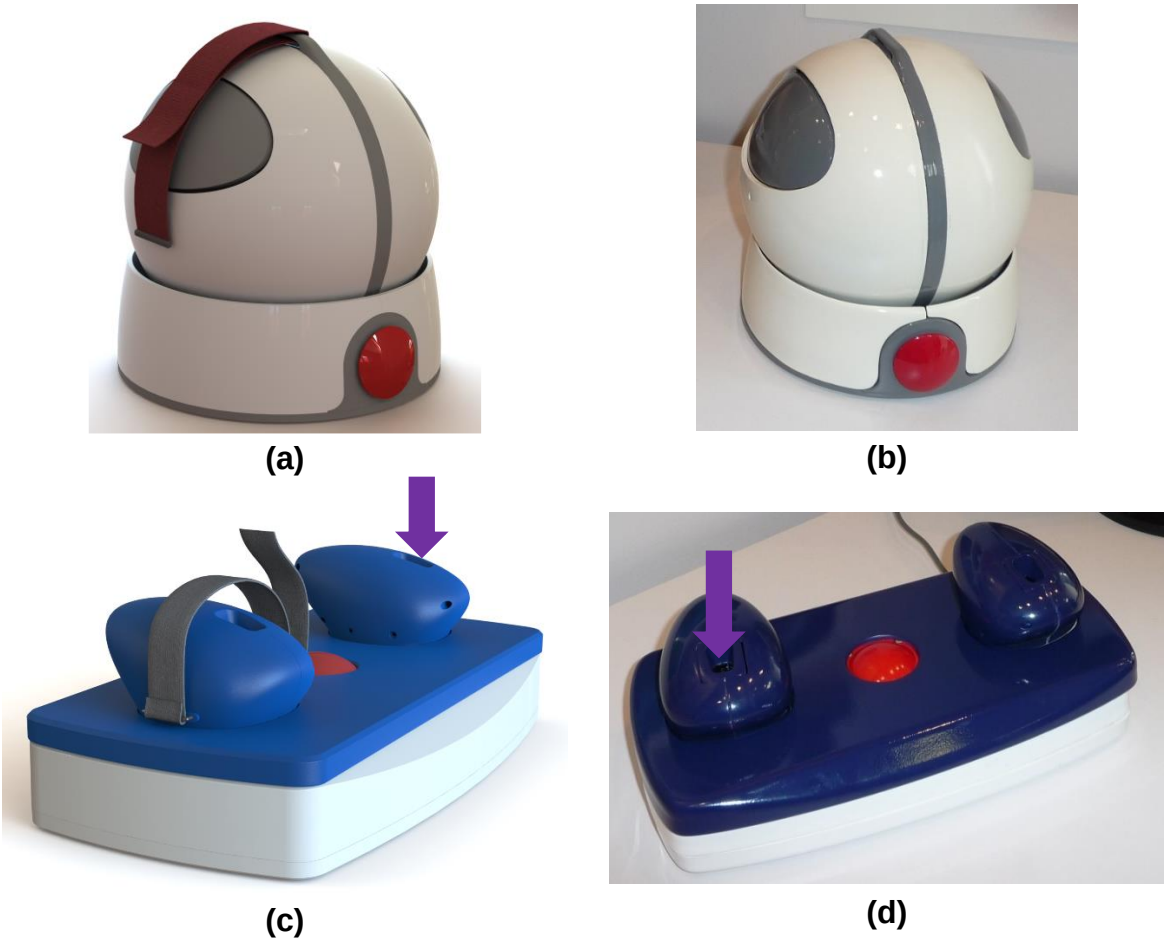


Figure 5-14 – (a) Final CAD model for 'Orby' (Source: Max Hughes); (b) The 'Orby' physical prototype; (c) Final CAD model for Design 1 (Source: Tom Whitby); and (d) Design 1 physical prototype. The purple arrows shown in (c) and (d) indicate the locations of the short-range proximity sensors

5.4.3 Stage 3 – Final Controller Design for the Randomised Controlled Trial

With 'Orby' being the preferred controller design, the final stage of the development process for the controller was to reassess the strengths and weaknesses of the design, with a view to manufacturing a number of units for simultaneous trial deployment. This part of the project was funded through a competitive grant from the Channel 7 Children's Research Foundation (Chapter 6), with the author being the Chief Investigator. The re-design process and readiness for manufacture is briefly summarised below, but described in more detail in Appendix H.

5.4.3.1 Controller Re-evaluation

A re-evaluation of the 'Orby' controller by the author and designer (Max Hughes) identified a number of areas for improvement, namely:

- *Oval pad design*: during the UniSA Exposition of the controllers (mentioned in section 5.4.2.2) attendees thought the grey oval pads – used to indicate hand position and provide haptic stimulation via motors mounted on the underside of the pads – were buttons. Pushing on the pads caused them to break indicating a weakness in the design, even though they were used inappropriately. Hence, the oval pad design needed reconsideration;
- *Haptic feedback*: the haptic isolation between the two sides was better from first to second prototype, but could be improved. Reconsidering where the vibration motors could be positioned was necessary considering the oval pad design was being revisited;
- *Proximity sensors*: Design 1's use of recessed proximity sensors (Figure 5-14) to detect hand position proved insightful and this concept was to be incorporated into the 'Orby' re-design for the trial;
- *Surface texture*: the surface of both Stage 2 prototype controllers (Figure 5-14) was smooth to touch, yet the end users would be children with a somatosensory impairment. A textured surface, particularly the oval pads where the hands are placed, would provide constant passive afferent feedback to the child during use and was to be investigated;
- *LED lighting*: an element of one of the alternative controller designs (Appendix F, Design 3, James French) was LED lighting. Lighting was to be incorporated into the final design, to provide visual confirmation that the controller is plugged in and ready to go and to augment the design aesthetic;
- *ND hand support*: the support/strapping design for the ND hand could be improved, mainly from an aesthetic perspective; and
- *Component access*: the original 'Orby' controller was fastened and positively locked during use with strong rare earth magnets. This meant that access to any internal circuitry or components was tool-less, quick, easy, and non-obvious (users didn't realise the controller 'internals' could be accessed this way). This feature was to be retained for the trial in case 'on the run' maintenance needed to be performed.

The grant funding for the project enabled the designer (Max Hughes) to be employed part-time as a Research Assistant and provided a specific budget to manufacture the controller through a short production run. A brief summary of the design and functional changes that were incorporated into the final controller design follows.

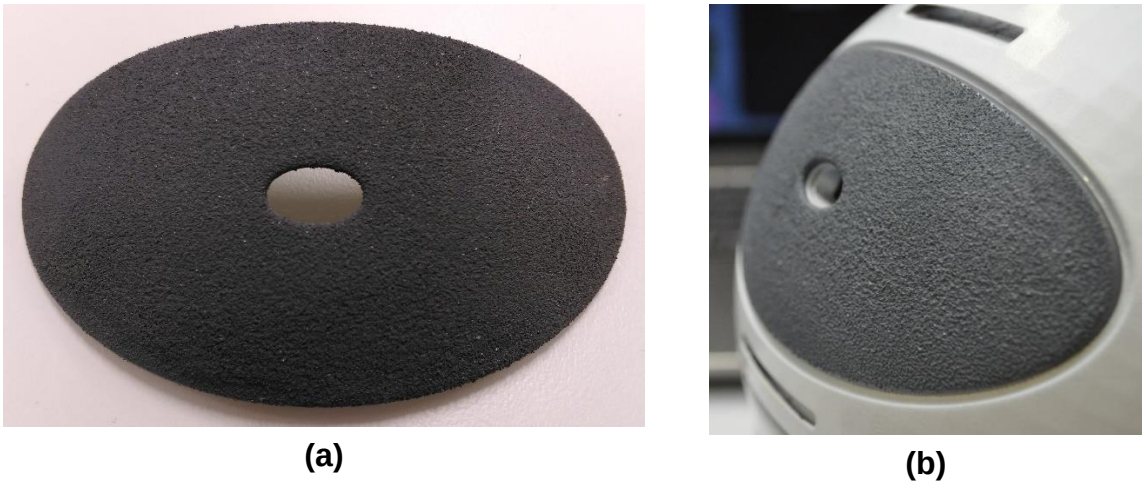
5.4.3.1.1 Improved Haptic Isolation

The Stage 2 'Orby' controller used a 'floating pad' design in combination with high density foam to minimise vibration across the controller to ensure the left and right sides were isolated from each other. The major shortcoming of this particular design was the delicacy of the suspension mechanism for the oval pad. Working with A/Prof Walker and an industry-based Industrial Design mentor, commercial vibration dampeners or vibration mounts were suggested as a way to dampen vibration from one side of the controller to the other, as well as increasing the path of travel for the vibration from one side to the other. Three cylindrical mounts in a triangular configuration were connected to each side of the central joystick mount (six mounts in total per controller), as shown in Appendix H.

5.4.3.1.2 Oval Pad Surface Texture

After being 3D printed, the oval pads were individually treated to provide a non-smooth surface texture. In partnership with the chosen controller manufacturer (see 5.4.3.2), a combination of grit blasting, hand sanding, and hand painting were used to achieve the desired finish, which was required to be non-smooth without being sharp or unpleasant to touch. Recognising that some children with CP exhibit hyper tactile responsivity or "*tactile defensiveness*" (Clayton *et al.*, 2003, pg. 46), it was important to get the surface texture right to avoid the controller being rejected by the child during the trial.

The final design and surface texture of the oval pads is shown in Figure 5-15(a), with Figure 5-15(b) showing the pad in place when fixed to the outside of the controller. During controller manufacturer (section 5.4.3.2), this particular item was the one that was most closely inspected and most often rejected due to a poor or abrasive finish, due to the fact it was manually hand treated and not machine produced.



(a) **(b)**

Figure 5-15 – (a) The textured oval pads, and (b) A close-up of the pad when mounted on the outside of the 'Orby' controller

5.4.3.1.3 Hand Detection via Proximity Sensors

A low profile distance measuring sensor (*Sharp*, GP2Y0D805Z0F), which uses both a photo diode and infrared emitting diode and has proximity sensor applications, was used for hand detection. The sensor was recessed 10mm into the textured oval pad (Figure 5-15(b)) inside the controller, due to the operating range being 5-50mm and the need to ensure that a hand on the controller surface would not be below the detection threshold of the sensor. Details of the sensor and its mounting location within the controller are shown in Appendix H.

The sensor was powered by connecting it to the 5V supply on the *Xbox* controller board, and from a system perspective, each sensor was connected to the location normally reserved for the left and right 'bumper' buttons (labelled '5' and '10' in Figure 5-1). When the sensors were covered, indicating the child's hands were in the correct position, this was equivalent to a player holding down both bumper buttons when using a standard *Xbox* controller. From a design perspective, integrating the sensors in this way made use of existing inputs on the *Xbox* board that weren't being used. Consequently, the Central Games Catalogue (section 5.6.1) responsible for monitoring hand position was interrogating bumper button switch presses when monitoring for correct hand position. Sandlund *et al.* (2012) reported suggestions from parents such as 'rewards' for specific movements that aligned with rehabilitation goals, such as Dunne *et al.* (2010) using an accelerometer to detect correct posture. In this instance, correct hand position was rewarded with gameplay.

5.4.3.1.4 Use of LED Lighting

One of the desirable but non-essential requirements of the controller was for it to incorporate lighting to provide visual feedback. The final 'Orby' design has a gap between the moveable spherical 'orb' and the circular shroud around the base. The gap is small (typically 1-5mm) and allowed the 'orb' to freely rotate without interfering with the base. After exploring design options to illuminate the controller (such as fibre-optic lighting vs. LED lighting) and provide visual feedback to the player during gameplay, due to time and budget constraints, simple LED lighting was chosen. During ideation, one suggestion was to provide different lighting effects via the controller to reinforce gameplay. However, one of the supervisors (SH) advised the animated lighting would be distracting for the child rather than reinforcing, so the idea was rejected.

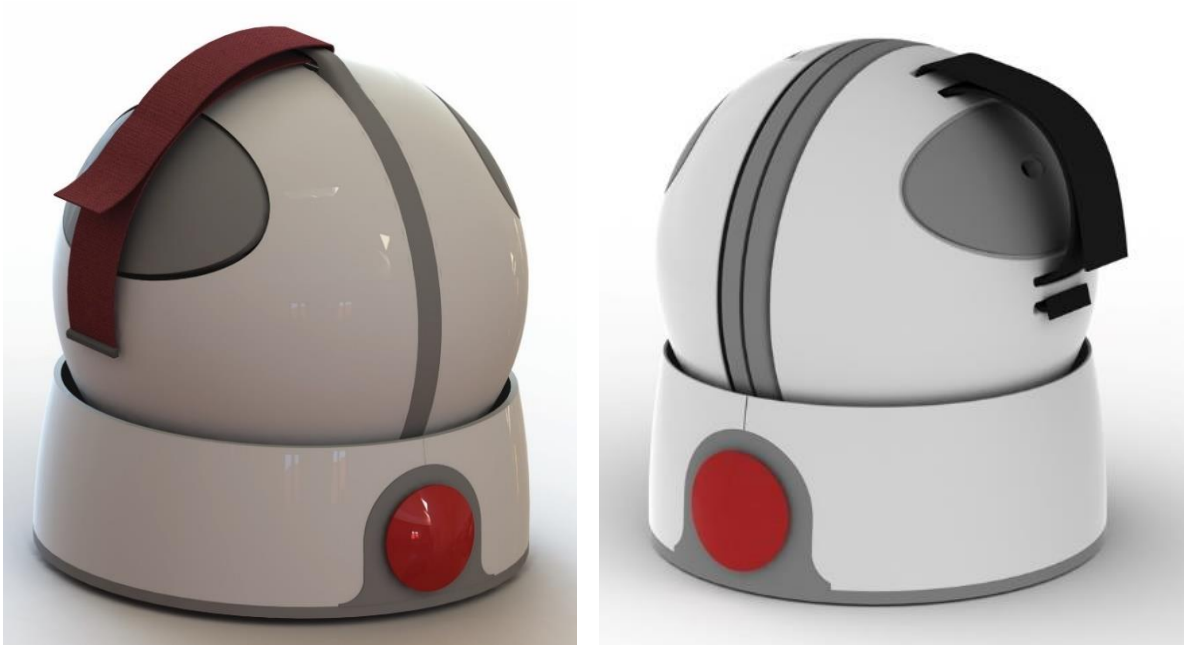
To avoid a clash of colours, green round wide angle (130º) LEDs were chosen for lighting, to match the existing small green LED that indicates power on the standard *Xbox* board (Figure H3, Appendix H), and powered via direct connection to the *Xbox* board. The LEDs were mounted to the inside of the circular shroud, four per side, positioned towards the front of the controller, with the intention of projecting light onto the bottom of the 'orb' that sits within the base during use. In this way, the lighting served to provide visual confirmation that the controller was plugged in and ready for use as well as an element of visual aesthetic.

5.4.3.1.5 ND Hand Support / Strap

The intention of the ND hand support or strap was to provide *assistance* to the ND hand to position it onto the spherical surface. The strap was not intended to tightly secure or pin the hand to the controller surface, as this would encourage passivity rather than active attention or use of the ND hand. That is, the child would be more likely to ignore their ND hand if they felt if it was tightly secured, which would be counterproductive to requiring the child to attend to their ND hand during gameplay.

The original Stage 2 'Orby' controller used a hand strapping design that was pinned at the base, beneath the oval pad, looped through a central slot on top of the controller, and then folded back on itself and secured with Velcro, as shown in Figure

5-16(a). The Stage 3 final ‘Orby’ controller used a similar strap design except the tethering and restraining mechanism now used grooved slots that the strap slid through, keeping the strap and its attachment points to a given hemisphere, as shown in Figure 5-16(b). The straps were custom-made by a local orthotics and prosthetics workshop (Orthotics and Prosthetics South Australia, Daw Park). The intention of both designs was that once the base of the strap was fitted to the controller, the other end of the strap could be independently looped and folded back on itself by the child using their dominant hand.



(a) **(b)**

Figure 5-16 – (a) The CAD model of the original ‘Orby’ controller strap design, and (b) A CAD model of the final ‘Orby’ controller strap design (Sources: Max Hughes)

Throughout Stage 3, each design change was prototyped and tested (in isolation and combination) using the rapid prototyping and 3D printing facilities at the UniSA. This was done to ensure each design element that was changed, and the final overall design, was appropriate, had the correct fit and tolerance, and was ready for small scale manufacture. Appendix H details relevant design and development elements including the vibration mount testing, haptic motor and proximity sensor location, the new ND hand strap design, and testing of a complete assembled test unit.

5.4.3.2 Controller Manufacture

With the Stage 3 controller design finalised, five Australian companies were approached for quotes to manufacture the 21 parts required for each controller, with a view to producing approximately 16 controllers in total, depending on budget constraints. Having access to multiple controllers was planned to allow parallel trials to occur and to cater for faults and potential breakages if they occurred. Four companies replied to the request for quote, and prices ranged from AUD$15,886 to AUD$27,231 (exclusive of GST). One Adelaide-based company (*Ellex Precise*, Gillman, South Australia) provided greater insight to the project after initial contact and demonstrated a deeper understanding of what was required, as well as a willingness to collaborate on this project compared to the other companies, and was chosen as the preferred supplier. *Ellex Precise's* quote was AUD$19,840 (ex. GST), which included painting external components and custom texturing each oval pad.

Ellex Precise advised that 3D printing the components was going to be the best and most efficient way to produce a short production run and used a *PROJET HD 3500 Plus 3D* printer to print all components. Owing to the number of components that needed to be printed per controller, the 21 components were laid out on three separate print beds, meaning it took almost 48 hours to print all the components for a single controller, including cooling time, prior to painting the necessary components. The components were delivered, inspected, and rejected (if faulty, warped or had a poor finish) or accepted and assembled as they arrived over a period of eight weeks, typically with parts for two complete controller units being delivered each week.

All the electronics components for the project were sourced through the Engineering Services Group at Flinders University, owing to their ability to leverage discounts with preferred suppliers (if grouped in a large order), quickly compare prices and volumes with different national suppliers, and obtain quick delivery of components. The bill of materials for the controller electronics totalled $2,083 (incl. GST) (Appendix H). The largest contribution to this cost was the proprietary components required, namely the *Microsoft Xbox* controllers (to provide 16 printed-circuit boards and 32 large vibration motors, two for each 'Orby' controller), and 16 *Logitech* 'Attack 3' joysticks (a ruggedised joystick module to provide movement). The cost per 'Orby' controller was $1,555, inclusive of GST (Appendix H).

5.4.3.3 Controller Assembly

The controllers were assembled as the 3D printed componentry was delivered, inspected, and accepted. This was done primarily by the designer with assistance from the author. Each controller required a number of fasteners and other componentry for assembly, as detailed in Appendix H. A small piece of black rubber cord (approx. 25mm in length) was used to prevent the ends of the grey hemisphere connectors knocking or 'chattering' during vibration events. The noise was heard during final testing and the cord served to hold the ends apart to prevent them touching each other and to prevent an acoustic cue during vibration events, as shown in Appendix H. Six clear rubber feet were glued to the controller base to provide support, friction, and stability during controller use (Appendix H).

5.5 Overall System Integration

Throughout the design and development process, software and hardware integration was a key component that was managed and facilitated by the author, who was the common researcher, supervisor and link across all aspects of the overall project. Despite the software and hardware teams working at different sites, beginning their projects at different times, and progressing at different rates, a smooth integration of the final system was achieved through using a commercial *Microsoft Xbox 360* controller and the associated XNA programming language. This meant each team could conduct tests of integration during development, knowing the specific inputs or outputs that were expected.

During system integration, the overall system was nicknamed 'OrbIT', a conjunction of the 'Orb' part of the 'Orby' controller nickname and the fact that the games run on a laptop or an item of 'IT'. The OrbIT Gaming System (OGS) took two and half years to go from initial concept within the author's imagination to the first trial with a child with CP as part of the RCT. The final system is shown in Figure 5-17.

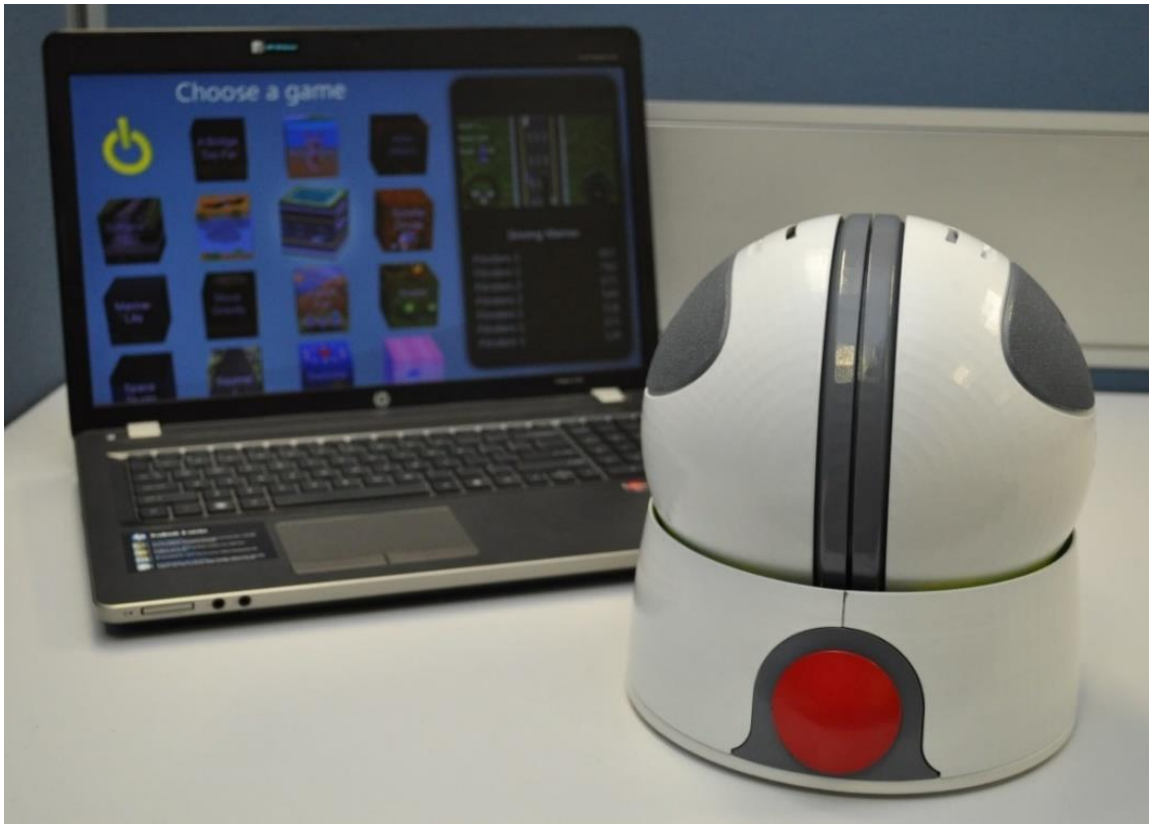


Figure 5-17 – The final OrbIT Gaming System (OGS), showing the Stage 3 'Orby' controller in the foreground and the laptop that runs the software and games in the background

5.6 OrbIT Gaming System (OGS) Features

In addition to the accessibility and clinical/rehabilitation features mentioned earlier, the OGS has the following features:

5.6.1 A Central Games Catalogue to Coordinate and Monitor the System

A 'Central Games Catalogue' was developed to coordinate and monitor the overall gaming system, which provided the framework for the games and supported many of the integrated features. The Central Games Catalogue was responsible for the following activities:

- Track and log all game activity (section 5.6.4), a requirement of the trial;

- Track and log all high scores and display them on the smartphone in the main menu screen to provide feedback on which games had been played and who had achieved the highest score(s) to date;
- Deliver unilateral haptic feedback to the child during system use (section 5.2.7), based on their individualised profile. The profile would identify the group to which the child was randomised and the child's ND side. The 'Guest' profile mirrored the settings of the child that the OGS was given to, meaning if a child was randomised to Group A and their ND side was 'left', the 'Guest' profile would also vibrate on the left side;
- During gameplay, monitor all game activity and if the out-of-game button is pressed, the system displays a generic 'pause' pop-up (Figure 5-18) that enables the child to take a break, adjust the volume, or exit the game;

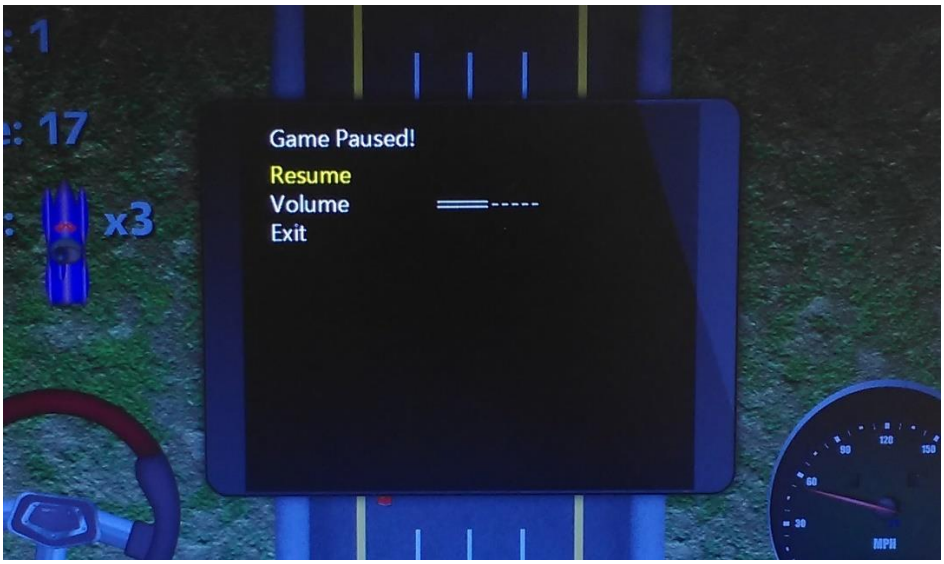


Figure 5-18 – The system-wide 'pause' pop-up that is displayed when the out-of-game button is pressed during gameplay

- During gameplay, continually poll the status of the proximity sensors that monitor hand position and display a pop-up alert if the child's hands aren't detected (Figure 5-19). The OGS can detect if one or both hands are removed or misplaced on the controller, and display the necessary prompt based on which hand(s) is not in the correct position; and

Figure 5-19 – The pop-up that is shown when the system detects that the child's hands are not in the correct position. In this instance, both hands have been removed from the controller, as both arrows are shown (Source: Brad Wesson)

- If the OGS crashes or 'hangs' for whatever reason, the system detects this event and automatically re-starts and re-boots. Additionally, if a vibration event persists and doesn't terminate as it should (all vibration events have a specific duration, as described in section 5.6.3), the system terminates the vibration event so the controller doesn't vibrate uncontrollably and without reason.

5.6.2 Game Randomisation through Procedural Generation

A feature of the OGS was that 14 of the 15 games used procedural generation to algorithmically generate level design and events in real-time to provide game variety. Thus, a child couldn't use their previous game experience to know exactly what to do if they played a particular game again. This feature addressed potential game boredom or 'game fatigue' if a game was played often. For example, a track or course would change from game to game, meaning the child couldn't memorise the hand movements or strategies required to succeed within the game – they had to dynamically respond to the gameplay. Additionally, game collectables and enemies

would appear in different positions, meaning the child had to fully explore each level or ‘world’ as items moved around, requiring concentration and attention.

The only exception to the above method of coding games was *Move Gravity*, the only puzzle-based game within the games catalogue. If a child is unsuccessful on their first attempt to solve the puzzle presented to them, the level repeats, providing an opportunity to learn from their previous experience. This occurs three times before the game ends. Hard coding and not procedurally generating content meant the child could learn from their previous attempts and scaffold their learning.

5.6.3 Haptic Feedback

All 15 games and the main menu screen delivered haptic feedback to the child during use to create a sense of immersion and to provide afferent stimulation to the ND hand (section 5.2.7). The vibration was characterised by two key features – vibration *intensity* and vibration *duration*, thus providing *graded* haptic feedback to the child. This provided a range of sensory cues to the child rather than a coarse ‘one size fits all’ vibration for all events. For some children, low intensity vibrations may be subthreshold.

During game development an important step was determining how and when haptic feedback would be delivered during gameplay. The author was involved with all levels of game development and worked with all game developers to define ‘look-up’ tables for each game. An example ‘look-up’ table is shown in Table 24 for *Dragonfly Dodge*, which was coded by Mr Chad Lundstrom. Within this game, the child would lose a life if they were eaten by either a bird or frog. Consequently, these game events were rated as the ‘worst’ scenarios within the game, and hence corresponded to the highest haptic intensity of 1.0 being delivered for a period of half a second. Colliding with an object (either rocks or reeds) was identified as the next ‘worst’ event that could happen within the game and was assigned a haptic level of 0.5, which lasted for the duration of the player being ‘stunned’ after the collision. Collecting a bonus within the game (a coin or an extra life) was assigned a haptic level of 0.25 for half a second.

Table 24 – An example 'look-up' table detailing haptic feedback for the game *Dragonfly Dodge* (Credit: Chad Lundstrom)

Vibration Event	***Vibration Intensity (0 – 1.0)***	***Vibration Duration (seconds)***
Eaten by a bird or frog	1.0	0.5
Collision with a rock or reed	0.5	Duration of 'stun'
Collection of a coin or extra life	0.25	0.5

Throughout game development each haptic feedback 'look-up' table was reviewed and tested using a standard *Xbox 360* controller to ensure that the vibration events were appropriate and matched what was expected for each game event. Where there was a perceived mismatch between what was felt and what was seen on the screen, the haptic feedback was adjusted accordingly. The coupling of haptic feedback to particular game events was an area that was specifically examined during the games evaluation sessions (sections 5.3.1 and 5.3.2) to ensure that such mismatches were minimised. The child's individual profile during the RCT identified whether they received haptic feedback or not (depending on their group allocation) and their ND side.

5.6.4 Data Logging

A requirement of the OGS was the ability to log all game activity when on trial, to determine how often it was used, by whom, when, the duration of gameplay, and the amount of vibration delivered (if randomised to Group A). During the development of the system, different logging methods were explored to accurately capture system and game metrics while at the same time optimising file size to avoid filling the hard drive of the laptops while on trial, and potentially losing important data. Additionally, automatically logging all system activity reduced the burden on families to have to keep a diary of system use, as highlighted in the literature (Preston *et al.*, 2016).

Frequency-based logging and event-based logging were two methods explored. The first method provides a common way to capture data if the right frequency is chosen – that is, a sampling frequency that is fast enough to capture all relevant game events but slow enough not to log unnecessary detail and create unnecessarily large data files. The second method provides a way of minimising file size when no activity

occurs because it only logs events or activities as they occur. During early system development, both methods were trialled and tested in-house by Mr Martin Henschke for the earliest game developed, *Space Stuntz*. Event-based logging proved to be superior in terms of minimising file size while capturing all essential data and was consequently chosen as the logging method and implemented during the trial. Data logging was tested a number of times during the games evaluation sessions (section 5.3), with an error identified in the way data were being recorded during one of the home-based trials with a teenager with CP (section 5.3.2.1). The way the data were logged, stored and analysed was improved when Mr Brad Wesson was employed on the project, and a custom program was written to interrogate the log files post-trial. An example log file from the trial appears in Appendix D.

5.6.5 Clinical Trial 'Demonstrator' Mode

To improve the integrity of the trial and to remove the possibility of author / demonstrator bias based on group randomised, a special 'Demonstrator' ('Demo') profile was developed for all orientation and training sessions when the OGS was deployed and set up for trial (Chapter 6, section 6.2.6). Prior to deployment, the off-site randomiser would notify the project Research Assistant of the randomisation outcome. If the child was randomised to the haptic group (Group A), the Research Assistant would also be told which side was the child's ND side. The Research Assistant would then prepare a laptop for the author to deploy with the child's individual profile pre-loaded into the OGS, where the child's profile would be programmed to function as per their randomisation allocation, and with 'Demo' mode enabled. In this configuration, when the OGS was first turned on, a choice of three log-in names would be presented instead of two – the child's name, 'Guest', and 'Demo'.

The 'Demo' log-in was a special profile that enabled the author to deliver the pre-trial training and study overview protocol (see Appendix I) without knowing which group the child was allocated to. By default, the 'Demo' profile was non-haptic (Group B). Consequently, when introducing and explaining how the OGS worked and while providing an overview of the OGS, the author was blinded as to the child's allocation.

The ‘Demo’ profile also enabled access to every game within the catalogue, enabling the author to deliver a consistent protocol for all children, even after the ‘Incentivised Games Catalogue’ was introduced (described in section 5.6.6). Once the OGS orientation and training overview was finished, and after any remaining questions were answered, the ‘Demo’ mode was disabled, removing it from the log-in screen. This meant the main log-in screen only showed the child’s name and ‘Guest’, as per the default set-up.

5.6.6 The ‘Incentivised Games Catalogue’

The ‘Incentivised Games Catalogue’ was developed mid-trial to increase engagement with the OGS, in response to what was perceived as a low level of initial engagement with the system during the first part of the RCT. As shown earlier (Figure 5-8), when the OGS was first deployed, the child could play any of the games they wanted, in an effort to provide game variety and choice. While this approach initially seemed sound, it didn’t reward effort or encourage long term engagement.

To address this problem, an ‘Incentivised Games Catalogue’ was developed. The suggestion for this approach was made by a colleague to the author who advised that engagement may increase if children had to ‘earn’ the games rather than be given them all upfront, with no effort on their behalf. Most commercial games adopt such an approach, using level accomplishment and/or high scores as a metric for releasing new levels or opening up bonuses within a game, which requires time and effort to achieve. However, when time and effort are put in, a reward is earnt.

To implement such an approach required an appropriate reward structure. Rather than use high scores or accomplishments within a game as a reward structure like commercial games, the time spent engaging with the OGS was seen as a better key metric. This also meant that if a child wasn’t able to get past a certain level or couldn’t accomplish a particular task in a game, they weren’t disadvantaged from accessing and unlocking future games. The addition of new games was advocated by Li *et al.* (2009) to reduce a gradual loss of novelty for a home-based system and

to address declining engagement. Implementing the 'Incentivised Games Catalogue' for the OGS required a few key decisions to be made:

- *Time*: the length of time a child had to engage with the OGS for prior, to the next game being 'unlocked' or released, was a balance between reward for effort (the incentive) and persistence with the task at hand (always using two hands to play games). After reviewing the amount of time children spent playing games during the evaluation sessions, the game release 'time variable' was set to 30 minutes. That is, a new game was 'unlocked' or released every time 30 minutes of *cumulative gameplay* was accrued.
- *How time was measured*: a novel application of the 'time variable' was that time was only counted when (a) the child that the OGS was given to was playing a game, as identified through the profile log in; and (b) the child was actually playing a game, not sitting idle in a game or system menu. This meant that the 'time variable' was silent whenever anyone logged into the 'Guest' profile, meaning players other than the child themselves couldn't unlock games.
- *Game release order*: the game release order was determined based on the ratings that the games received when they were trialled in-home and during the games evaluations sessions (section 5.3). Again, this was a balance between generating and maintaining interest in the OGS, keeping the child interested for long enough that they wanted to keep engaging and playing games until they released every game. Consequently, the most popular game from the trials and focus groups were released last. To promote interest in unlocking the games, two games were released after the first 30 minutes of gameplay, with one game being released every subsequent 30 minutes. Therefore, to unlock every game the child had to engage and play games for at least 270 minutes, or four and a half hours.
- *The initial number of games*: another important consideration was the initial number of unlocked games that were given to the child, and which of the 15 games were part of the initial release. The initial set of unlocked games represented different game genres and game diversity, as well as games that would help to generate game time when played. The initial set of unlocked

games was set at five – *Alien Attack*, *DragonFly Dodge*, *Snake*, *Space Stuntz* and *The Fancy World*, as shown in Figure 5-20.

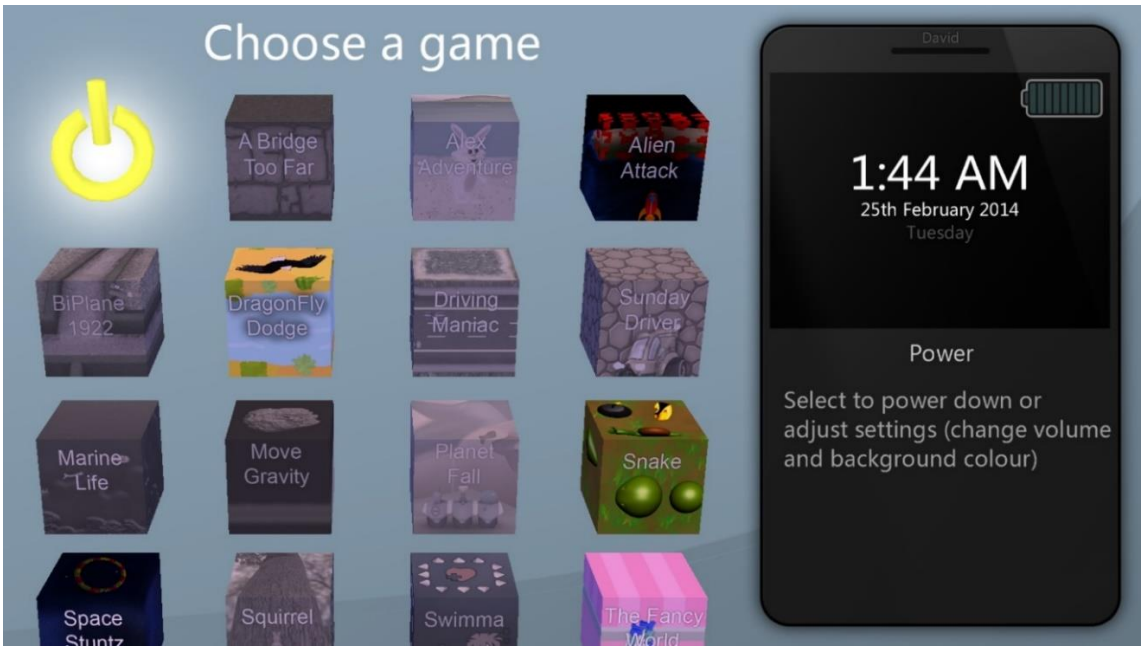


Figure 5-20 – The main menu screen when the 'Incentivised Games Catalogue' feature is activated. Unlocked games are shown in colour while locked games are greyed out (Source: Brad Wesson)

- *Visual communication and game release integrity*: it was important all players understood which games could and could not be played. As shown in Figure 5-20, colour was used to communicate which games were unlocked (the cube representing these games was brightly coloured) and which games weren't (the cube representing these games was greyed out). The smartphone in the main menu screen also communicated if a game was unlocked or not, and if the game was locked, how soon it could be released. However, if a child didn't understand this concept, couldn't read, didn't notice that the smartphone was telling them the status of a particular game, or if they wanted to try playing a game that was locked, they would receive the prompt shown in Figure 5-21. As can be seen in the background of Figure 5-21, the smartphone contains the same information as the prompt in terms of how much longer a game has to be played before it becomes unlocked.

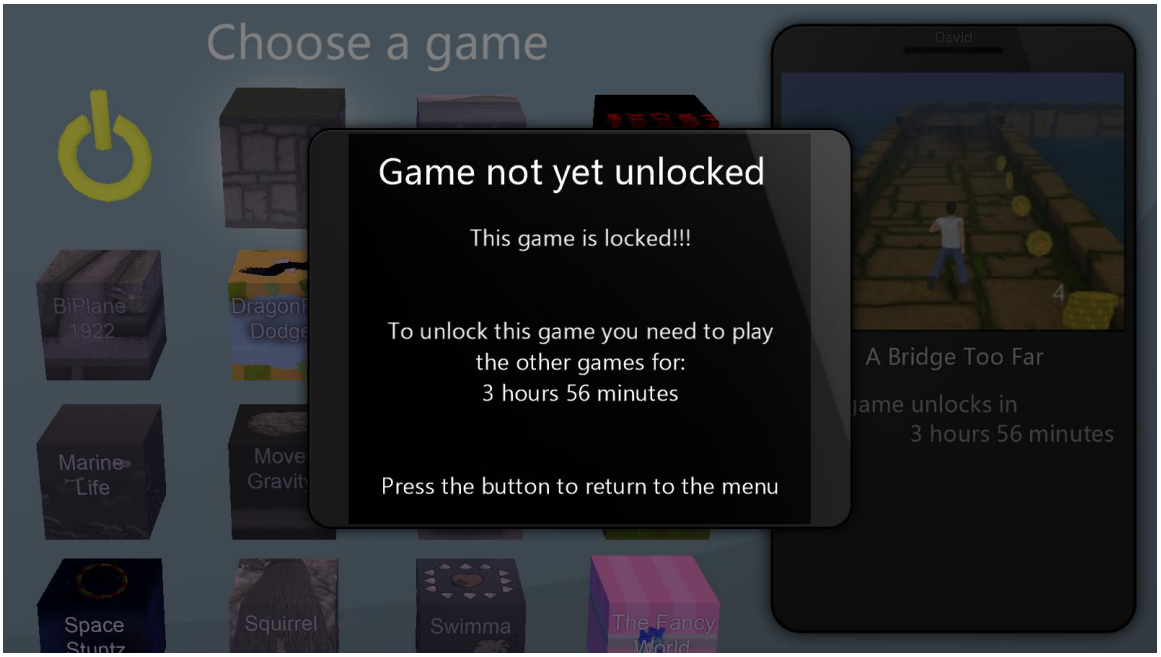


Figure 5-21 – The prompt that is received if a player tries to play a game that is still locked (Source: Brad Wesson)

- *Encouraging the child to release games*: as mentioned earlier (5.3.3), the smartphone in the main menu screen was integrated into and coupled with the Central Games Catalogue. It served two functions – showing a short video preview of the highlighted game in the top section (typically around 30 seconds of game highlights) and keeping track of all high scores in the form of a 'High Score Table' in the bottom section. When the 'Incentivised Games Catalogue' was activated, the smartphone also kept track of when a particular game could be unlocked, as shown in Figure 5-21. To maintain the child's interest, the video preview feature still functioned on the smartphone even if a particular game was locked, to generate interest and enthusiasm in the game and to encourage the child to unlock it.

5.6.7 Other OGS Features

To improve the longevity of the OGS and to increase both game challenge and 'buy in' from the child, in addition to the use of procedural generation (section 5.6.2) three games were coded to 'evolve' in real-time or change control paradigms based on

player progress and achievement within the game. Examples of real-time game evolution and ‘adaptability’ are described in Appendix D.

The OGS was designed to be robust and maintenance free while on trial. From a power perspective, ‘Orby’ was based on a USB corded *Xbox 360* controller, meaning it was powered directly via the laptop, rather than with batteries. This eliminated situations where a child would forget to dock and charge the controller after use.

5.6.8 Incorporating and Embodying the Principles of Universal Design

The intention of the OGS was to provide an accessible computer gaming system for a child with a hand impairment due to CP that achieved active engagement and cognitive buy-in, rewarded bimanual upper limb use and correct hand position by allowing gameplay, and delivered a range of afferent haptic vibration stimuli to the child’s ND hand. Consequently, the overall design was a combination of accessibility and clinical rationale. With the seven Principles of Universal Design (Story, 1998) as a framework, Table 25 provides a summary of how the principles were incorporated into the final design of OGS.

Table 25 – Summary of how the Principles of Universal Design were incorporated into the OrbIT Gaming System (OGS)

Universal Design Principle	*Incorporation and Embodiment of the Principles within the OGS*
Principle 1: *Equitable Use*	The 'Orby' controller is appropriate and applicable for children and adults with a hand impairment / limitation due to other conditions besides CP, such as stroke, as well as people without an impairment. This is achieved through the size and spherical form of the controller, meaning fine finger control and grip are not prerequisites for controller use. Additionally, there was a focus was on making the controller look mainstream rather than like a 'clinical device' that might stigmatise the user.
Principle 2: *Flexible in Use*	The size of the controller sphere meant children of all ages could place a hand on the pad on each half of the controller's surface, without crossing over to the other half of the controller. This was important as it meant that proper use would avoid the phenomenon of sensory extinction (Critchley, 1949; Brozzoli *et al.*, 2006), as mentioned in section 5.2.7. Moreover, the delivery of haptic stimulation is controlled through software, with the OGS capable of delivering stimulation to just the left side, just the right side, both sides, or neither. The controller facilitates a wide range of abilities through the ND hand strap, which encourages volitional ND hand control during use. The software was designed to ensure that each game provided a gentle lead in, with a game complexity 'ramp' that provided challenge and complexity that accommodated different children's abilities and experiences. This enabled each child to experience a sense of succeeding within each game, particularly the early stages, before greater challenges was provided later in the game. Most of the games were coded so that fine, precise movements weren't a requisite for successful gameplay, meaning that success and game progression wasn't coupled to hand capability. Additional flexibility was provided by offering two methods to pause games activity: the child could press either the out-of-game button to pause their game or simply remove their hands to activate the proximity sensors and achieve the same outcome. When the child resumed playing games, the OGS provided a three second countdown to lead them back into gameplay.
Principle 3: *Simple and Intuitive Use*	All OGS interaction, gameplay and menu navigation is achieved through moving the controller forward, backward, left or right, through intuitive 'rolling of the dome' motion. This means the child can rest their hands on the controller surface and move it as necessary, without worrying about actuating buttons with fine finger movements. All games were designed so they required little background knowledge to play them, meaning they could be played without needing to read any instructions beforehand. This aspect and other gameplay features were tested during group evaluation sessions with young and teenage children (sections 5.3.1). Consequently, when the OGS was on trial, no instructions were provided, meaning the child could discover and explore each game.

The final main menu design adopted a contemporary tablet-style look, using the universal computer symbol for 'on/off' in the top left hand corner position, and each of the fifteen games were represented by a cube wrapped in artwork from the game. Consequently, if the child couldn't read the game name, they could identify the game through the game graphics/artwork. Additionally, to the right of the games matrix layout another contemporary device, a smartphone, showed a short video of each game, helping the child to identify a particular game by seeing a video of it, and displayed the high score table for each game.

Principle 4: *Perceptible Information*

The OGS was designed to target and appeal to multiple senses, including visual, auditory and tactile. The games use bright colours, graphics and relevant sounds and music to engage children during gameplay, with game sounds and haptic feedback coupled to game actions, such as hitting an object or losing a life. Consequently, for every game action, the visual stimulus is coupled with both acoustic and haptic feedback.

Two grey oval textured pads contrast against the white surface of the controller to indicate hand position, and the large contrasting easily accessible bright red button on the front of the controller provides a distinguishable visual cue for the out-of-game button. A 'click' sound is heard when the button is pressed, providing an auditory cue to accompany the switch press. By placing the proximity sensor in the centre of each pad, when the controller is used correctly the child receives passive tactile feedback. An additional textural element relating to the controller resulted during manufacturing – the 3D printing process resulted in a non-smooth finish, which provides a different texture to the oval pad.

Principle 5: *Tolerance for Error*

The OGS needed to be robust and to perform as expected on trial to minimise adverse events. However, the OGS was based on a commercial laptop along with standard *Microsoft Windows* install programs and the ability to access the Internet, which was not part of the trial. Consequently, prior to deployment each laptop was pre-configured when switched on to auto-start the Games Catalogue, disable the Wi-Fi, and ignore all keyboard inputs (meaning the 'Esc' key or 'Ctrl-Alt-Del' couldn't be used to close the Games Catalogue and access the laptop itself either by accident or through deliberate tampering). All Windows icons were removed from the Desktop, the background was set to black, and the Windows Toolbar was hidden from view, meaning only a blank screen was seen while the Games Catalogue was loading at the start. Administrator access to each laptop was hard-coded into the software, with '`quit459`' the passcode that was required to access the laptop Desktop.

Within the Games Catalogue, the only mistake that could be made was an unintended game selection. If this happened, the child could easily cancel or exit the game they accidentally chose, log in again by choosing their named profile, and try and select the correct game. So while an error could occur (choosing the wrong game), the outcome (exiting the incorrectly chosen game and re-choosing the intended game) resulted in only a time delay. The Games Catalogue could only be shut down by choosing the yellow 'On/Off' icon from the main menu screen, and then selecting the 'Shutdown' icon.

Principle 6: *Low Physical Effort*	The 'Orby' controller provided a simple, intuitive and effortless way to interface with the OGS. When the OGS was set up at the correct height the child's shoulders, arms, forearms and wrists were in neutral positions, and the spherical surface of the controller combined with the height of the joystick mount within the controller promoted low physical effort to move or rotate the controller. Additionally, the out-of-game button on the front of the controller required minimal effort to activate, and could be pressed with a thumb, finger, palm, heel or back of the hand. If a child could not position and hold their hands on the grey oval pads to operate the controller, a strap could be fitted to assist with holding the ND hand in place. The strap also addressed fatigue issues if the child tired. Additionally, repetitive in-game actions were avoided with the software randomising game events and game activity, meaning dynamic hand actions were required to respond to each new game scenario.
Principle 7: *Size and Space for Approach and Use*	The OGS was designed to be used within the home in a seated position with the controller positioned on a table, desk, or wheelchair tray top. Rubber feet on the bottom of the controller provided a degree of friction with the surface the controller was placed on to minimise movement during use. The spherical surface of the controller allowed for hands of varying size, and once seated and set up, the child only needed to interact with the controller and not the laptop, meaning all operable items were within comfortable reach. The USB cord that connected the controller to the laptop was long to provide flexibility in terms of setup. This meant that the author and parents could vary the distance between the controller and the laptop screen for optimum viewing for each child. This also provided flexibility in terms of how the games were viewed, such as if a family wished to connect the laptop to their large screen TV via the HDMI output on the laptop, to view the games on a larger, more immersive screen.

5.7 OrbIT Gaming System Overview and Conclusion

The OGS was conceptualised, designed, developed, tested, evaluated, refined, manufactured and deployed for the RCT in two and a half years. The following provides an overview of the OGS. OrbIT:

- Is a stand-alone, integrated, custom-made, home-based, accessible computer gaming system that was designed to be independently operable by a child with a hand impairment due to CP, that was developed using a co-design process (sections 5.3.2 and 5.4.2.1) and a trans- and multi-disciplinary team of professionals;
- Features a novel, accessible controller nicknamed ‘Orby’, that was designed using Universal Design principles to promote integrated, forced bimanual hand use (section 5.2.8) while providing graded afferent haptic feedback (section 5.6.3) to the ND hand only (section 5.2.7), and provision to support a child’s ND if required (section 5.4.3.1.5);
- Contains 15 different games (Table 23) that only require joystick movement to play, removing the need for in-game button activity that is difficult and limiting for children with a hand impairment. This meant the child’s hands were always engaged on the controller during gameplay, ready to receive haptic feedback. Additionally, the games were intuitive and were provided *without* instructions, meaning each child could discover and explore the games at their own pace, learning and adapting as they did so;
- Promotes focussed attention, system longevity, and dynamic hand movements that respond to visual stimuli, with games that are coded using procedural generation (section 5.6.2). Additionally, some of the games are coded to adapt dynamically based on the child’s score, achievements or choices within the game (Appendix D);
- Through the Central Games Catalogue, coordinates and monitors the overall system (section 5.6.1), tracks and logs all game activity using event-based logging (section 5.6.4), monitors hand position (section 5.4.3.1.3), and also enables the author to train and orientate the child to the OGS without knowing which group the child was randomised to (section 5.6.5);

- Provides flexibility in terms of the games that can be deployed through the development of an ‘Incentivised Games Catalogue’ (section 5.6.6), which restricts the child to a few games initially, requiring them to play games (and hence spend time using the OGS) to release games that are locked. Games are released based on how much time the child who was allocated the OGS actually spends playing games, not their ‘Guest’ or their time spent in a menu. The release of games can be varied (it was set to 30 minutes for the RCT), as can the release order of the games; and
- Uses a smartphone interface to show a short video preview of each game within the main menu (Figure 5-8) to indicate which games have been played and to track high scores for all games, including indicating who has achieved the high score for a particular game.

This chapter has introduced the concept of using SG for health and rehabilitation purposes, provided an overview of the literature to date, and detailed the design process that was used to define core system requirements and specifications for an accessible, custom-designed, co-designed SG system called OrbIT. A custom-designed system was pursued in preference to a commercial gaming system due to the unique requirements for this particular intervention, namely, an accessible system that delivers graded and contextualised vibration stimulation only to the ND hand of the player. Post-trial, the United States-based group *AbleGamers Charity* (part of the *AbleGamers Foundation*), reviewed and assessed an OGS unit, providing an independent review (see Appendix J).

5.8 Author’s Contribution to the Work Presented

Given the team of people that contributed to the development of the OGS, the following lists the specific contributions of the author:

Overall Project:

- Developed the overall project concept and direction, initiated projects for both the hardware and software parts of the project, including applying for and

receiving funding for the project from within the University (Faculty Establishment Grant) and externally (the Channel 7 Children's Research Foundation) to support and conduct the work; and

- Developed technical specifications and requirements to integrate the hardware and software parts of the project, facilitated integration between the two parts of the project, and supervised and directed all aspects of the project.

Software:

- Co-drafted all ethics applications for the games evaluations and responded to ethics queries/requests for changes, including final ethics reports;
- Alongside Dr Brett Wilkinson, coordinated, planned, set up and facilitated the games evaluation sessions within schools with typically developing children, including collecting and analysing primary data, and co-designing the '*Participant Evaluation Form*';
- Planned, coordinated, set up and deployed both home-based gaming trials involving children with CP, including collecting and analysing primary data;
- Provided day-to-day supervision, direction and guidance for the Research Assistant working on the project, from introducing disability and understanding what CP and rehabilitation is, to suggesting game modifications, new games and game concepts, game features that required haptic elements, ensuring the games were appropriate for children aged five to 15 years, ensuring the necessary Principles of Universal Design were implemented in each game, assessing and recommending data logging methods, main menu formats, and the mechanics and features of the 'Incentivised Games Catalogue';
- Worked alongside each game developer to ensure they understood what disability, rehabilitation and CP were, while guiding and providing input to the story-boarding phase to ensure each game had a logical flow, clear purpose, and logical reward structure that also corresponded to haptic game events;
- Worked alongside digital artists to choose appropriate imagery and graphics for each game;
- For all student projects, where games were developed for the project for credit, provided feedback, critique and supervision as necessary, including for

all presentations, reports and theses submission, as the primary supervisor or as a co-supervisor for the student along with Dr Brett Wilkinson.

Hardware:

- Developed and documented the necessary hardware specifications and project requirements in terms of interfacing the hardware and software;
- Initiated the pilot controller project at the University of South Australia and worked directly with all four students, providing direction and supervision as necessary, alongside A/Prof Sandy Walker;
- For the second stage of controller development, alongside A/Prof Sandy Walker, provided supervision, direction and guidance as necessary, specifically at key 'Stage-Gate' design phases of the project, and particularly around understanding disability, hand impairments, and CP, and incorporating rehabilitation and clinical requirements into the controller, such as bimanual use, haptic isolation, and hand positioning to avoid grip actions. Participated in prototype concept refinement, providing design direction and ranking in terms of design priorities and design features;
- Co-wrote the ethics application for the evaluation by children with CP with A/Prof Sandy Walker, approached families to be involved, and led/facilitated the evaluation session;
- For the third stage of controller development, alongside Max Hughes, supervised and directed the next stage of controller evaluation and development, including evaluation of different vibration mount orientations, assessment of different quotes for manufacture, choosing the most appropriate manufacturer and meeting with them to agree on a production schedule, component scrutineering and acceptance, assisting with 'Orby' controller assembly (mostly performed by Max Hughes), and post assembly inspection and testing, including trouble-shooting of the 'Orby' controllers when they didn't operate as expected.

5.8.1 Recognising the Contributions of Others

For the software part of the project, the attribution and credit for all games appears in Appendix D, for both the games themselves and the game artwork and assets (if a digital graphic artist worked alongside a game developer). All game developers were responsible for the coding of individual games, working under direction of the author, and ensuring a common format was followed to enable the game to seamlessly integrate into the overall Games Catalogue. Only the Research Assistants that were employed to work on the project through grant funding (Mr Martin Henschke and Mr Brad Wesson) worked with the author on the main menu system and integration of individual games into the Games Catalogue. The author did not code any of the games or the software.

For the hardware part of the project, the author co-supervised, along with A/Prof Sandy Walker, individual Industrial Design students who conceptualised, designed, sketched and 3D CAD modelled their respective designs. The author provided supervision and direction and did not design or CAD model any of the prototype controllers.

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6. Stage 2 – The Pilot Randomised Controlled Trial of the OrbIT Gaming System (OGS)

6.1 The Stage 2 Study

The aim of Stage 1 was to identify the somatosensory status of the hands of a cohort of children living with CP in South Australia using established and valid assessments. The results of this phase flowed directly into the Stage 2 study, which was a home-based RCT of the OrbIT Gaming System (OGS), aimed specifically at children with CP with known somatosensory impairments.

6.1.1 Study Aim

The aim of the study was to determine if children with CP who have a known upper limb somatosensory impairment can have that sense of touch improved through using a novel and haptic home-based computer gaming system called the OGS. The gaming system is designed to captivate and engage the interest and motivation of children while providing contextually relevant and graded afferent stimulation to their hands through a customised and accessible controller.

As discussed in section 5.2.6, the ‘active’ element for the RCT intervention was the use of haptic or vibration stimulation. Vibration sense is the modality that is most preserved in children with CP (Uvebrant, 1988), and vibration is known to activate the primary and secondary somatosensory cortices (Coghill *et al.*, 1994). Additionally, with respect to computer gaming, vibration feedback is known to increase the realism of games by applying forces that are similar to or representative of those that would be felt if actually performing the task (Geerdink *et al.*, 2004), and hence enhance ‘game immersion’.

The primary outcomes for this study were the sensory assessments, as the ‘impairment level’ outcome measure, and the JTHFT, a valid measure of hand motor function, as the ‘activity level’ outcome measure. Secondary measures included a validated quality of life questionnaire (the CP-QOL) and a validated parent- and child-self report hand function questionnaire (the ABILHAND-Kids). When the study

was first conceived in 2010, to the best of the research team's knowledge, this was the first RCT investigating the use of technology (serious gaming) that was specifically directed at improving sensory dysfunction in the hands of children with CP, and represented a gap in the intervention literature for this population. This gap was confirmed through a systematic review of the intervention literature four years later by Auld *et al.* (2014), with the authors recommending that interventions based on (1) stimulus specific training, (2) transfer enhanced training and (3) mirror therapy showed the most promise for a paediatric population (pg. 831). Recently, interventions piloting two of these approaches, namely, mirror-based training (Auld *et al.*, 2017) and transfer enhanced training (McLean *et al.*, 2017), have been published, with both approaches showing early promising results for tactile perception (Auld *et al.*, 2017) and proprioception (McLean *et al.*, 2017). These studies are discussed in more detail in the Discussion (section 6.4).

6.1.2 Study Hypothesis

The hypothesis for Stage 2 was that somatosensory function in children with CP with a known sensory loss can be significantly improved through a home-based computer gaming system that couples a bilateral upper limb activity with an opportunity to experience a range of appropriate afferent (sensory) inputs. Through the use of vibration stimulation delivered in this context, it is hypothesised that children randomised to treatment with vibration stimulation would have significantly better sensory and functional outcomes than children having no associated vibration stimulation.

Specific areas of investigation for this study included:

1. The feasibility of deploying the OGS into a family home in an unsupervised and 'child-led' format, as a way of engaging children with CP to use both their hands;
2. Child and parent acceptance of the OGS;
3. OGS usability and engagement; and
4. The effectiveness of the OGS to improve hand function.

6.1.3 Study Inclusion and Exclusion Criteria

Given that this study followed on directly from Stage 1, the criteria for inclusion or exclusion into Stage 2 were the same as for Stage 1 (Chapter 4, section 4.1.3). The only criteria that were different to Stage 1, or specific to Stage 2, were:

- **Age:** the age requirement for recruitment into Stage 1 was children five to 15 years of age and sensory assessments began in May 2012. Stage 2 began recruiting in May the following year, with the first OGS deployed in mid-September 2013. Hence, the cohort was typically 18 months older than they were for Stage 1, shifting them up to the six and a half to 16.5 years old age bracket. The child's age was calculated as the time from their date of birth to their set-up date for the OGS, expressed in whole years and whole months. Half months were rounded up to the nearest whole month, meaning that a child aged eight years, two months and two weeks on their set-up day would be recorded as being eight years and three months old, or 8.25 years.
- **Hand function and the ability to use the 'Orby' controller:** during each Stage 1 assessment, the assessing OT was asked to provide a professional opinion on the child's ability to use and manipulate the 'Orby' controller for the eventual trial at the conclusion of the Stage 1 assessment. As mentioned earlier (section 5.4.2.2), the assessing OT was part of the overall team that reviewed and provided feedback on the controller design at key milestone stages, so was able to provide insightful feedback. This meant that one child was excluded from the trial due to 'fisting' of their ND hand, which would cause problems when using the controller, as described in section 6.2.5.
- **Children who were involved in the evaluation of the OGS during development:** children with CP who volunteered and participated in either the games evaluation (section 5.3.2) or prototype controller evaluation (section 5.4.2.1) sessions were excluded from being invited through to Stage 2 due to their involvement in the development of the OGS, and the possible bias this may have created with respect to being blinded as to the exact nature of the study (and the role of afferent stimulation via vibration feedback).

6.1.4 Funding

An application for funding for Stage 2 of the research was made to the *Channel 7 Children's Research Foundation* (Ch7CRF) in July 2012 via a research project grant, for work to commence in 2013. The application (Project ID 13700) underwent assessment and scientific review and was funded to the value of AUD$65,000. The author was the Chief Investigator for the grant application and project work.

6.1.5 Ethics Approval

Prior to beginning the RCT, ethics approval was obtained from the WCHN HREC. The ethics application and Site Specific Assessment (SSA) were submitted in November 2012 and final ethics approval was received on the 26th February 2013 (protocol number REC2530/12/15, HREC/12/WCHN/100, and SSA/12/WCHN/101). The final SSA review and approval was received on the 4th March 2013.

6.1.6 Trial Registration

With respect to trial registration, an application for a *Universal Trial Number* (UTN) was made and the RCT was allocated a UTN of U1111-1127-0623. The RCT was then registered with the *Australian New Zealand Clinical Trials Registry* (ANZCTR), and allocated the registration number 12612000186853, and through the *Therapeutics Goods Administration Clinical Trial Notification* (TGA CTN) Scheme, and allocated a CTN number of 092/2013 (Appendix L).

6.1.7 Data Entry and Integrity Check

The assessing therapist recorded test results directly on to a hard copy Stage 2 assessment recording sheet during each session (Appendix M). At the conclusion of each session, data were transferred into a Microsoft Excel spreadsheet that was customised for the purposes of this study. The therapist who conducted the session entered the data or this was done by the author – for the latter case, the therapist would scan the assessment sheet and email it to the author.

Post-trial, a 100% data audit was conducted for all sensory and motor assessment results. Sixteen children completed all three assessments (72 data points per assessment), one child completed just the A_1 assessment (24 data points), and one child completed A_1 and A_2 only (48 data points). Of the 1,224 total data points, 11 errors (0.90%) were identified – four data interpretation errors (the therapist interpreted the SWM instructions incorrectly for two children) and seven data transcription errors. Once all Excel data points matched the corrected hard copy assessment sheets, the data were formatted and a meeting was arranged with the consulting statistician for the project to discuss the necessary statistical analyses. The same statistician who was consulted for Stage 1 was also consulted for Stage 2, to ensure project consistency.

6.1.8 Statistical Methods and Analysis

The statistical analysis used random effects modelling, investigating 'Group' (A or B) and 'visit' (A_1, A_2 or A_3) interactions for each child and for each hand (dominant and ND). OGS usage was investigating using Independent Sample t-Tests or a paired samples t-test, depending on the variables being analysed. Statistical significance was set to p=0.05 for all tests. Data analyses were performed and modelled using SPSS (Version 23) or Microsoft Excel. Analysis was on an 'intention-to-treat' basis.

6.2 The Stage 2 Randomised Controlled Trial (RCT)

6.2.1 Trial Design

The RCT was designed with two arms, Group A and Group B, and lasted 14 weeks. The difference between the two arms was the status of the haptic motors built into the gaming controller. **Group A** was the *intervention* group where the OGS was deployed with the haptic motors *enabled*, providing active afferent haptic vibration stimulation to the child's ND hand during game play. **Group B** was the *control* group, where the haptic motors were *disabled.* It is recognised that Group B still received passive cutaneous stimulation through touching the 'Orby' controller. Consequently, the delivery of *active* afferent haptic vibrational stimulation to the ND hand of the child during game play was the differentiator between the two groups.

For both arms of the trial, the same physical OGS was deployed for six weeks (42 days) and a single researcher (the author/lead researcher) conducted an introduction, orientation and training session on Day 1 with participants and their families using the 'Demo' profile (Chapter 5, section 5.6.5), while reading from a standardised protocol (Appendix I).

Across the trial there was a difference between the Games Catalogues that the children were provided, which extended across both trial arms. Due to reasons described in the results section, and as described earlier (Chapter 5, section 5.6.6), the original Games Catalogue that was deployed was re-programmed half way through the trial into a new 'Incentivised Games Catalogue', where children were required to spend time playing games in order to release 'locked' games, to improve engagement and system longevity.

Upon being identified as an appropriate participant for Stage 2 based on the results of the Stage 1 study (see section 6.2.5), children were invited to an assessment session at the WCH with the assessing OT. The testing framework for the Stage 2 assessments were the same as for Stage 1, with the exception of the SWM test, where a 20-filament test kit was used instead of the 5-filament test kit, as explained earlier (Chapter 3, section 3.3.1). The only other modification to the Stage 2 protocol was a change to the testing order with respect to the *AsTex*® device. During Stage 1 the two tests using the *AsTex*® device were conducted back-to-back, after the SWM test. For Stage 2, the two *AsTex*® tests were separated – the *AsTex*® Test 1 was conducted directly after the SWM test, but the *AsTex*® Test 2 was conducted at the end of the session, after the JTHFT, to separate them temporally. The two tests were de-coupled to discern if there was a difference between the results for each test. The recording sheet for the Stage 2 assessments highlighting these differences appears in Appendix M.

The first assessment for Stage 2 was referred to as the A_1 assessment – the baseline assessment prior to beginning the RCT, as shown in Figure 6-1. With the OGS being home-based, the location for the trial intervention was each child's home. All assessments took place at the WCH, with the exception of one child who lived in regional South Australia (his A_2 and A_3 assessments were conducted in his local school by the assessing OT to minimise travel time and inconvenience to the family).

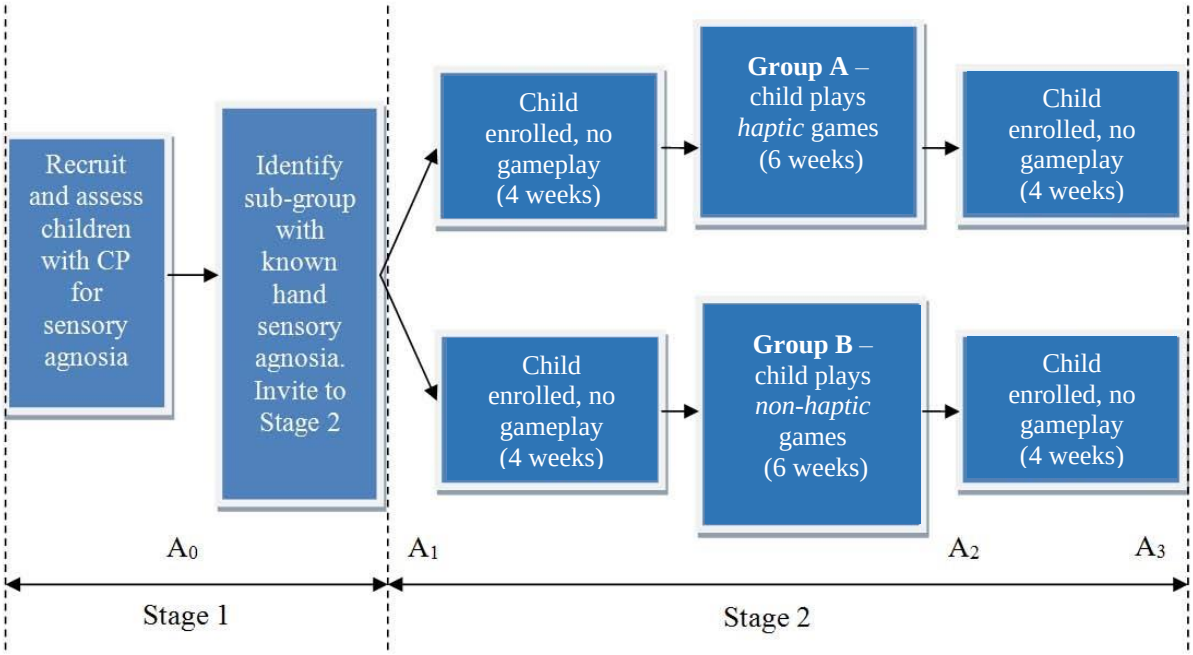


Figure 6-1 – An overview of the complete study, showing Stage 1 and Stage 2, including the different arms and assessment points of the Stage 2 RCT

From a trial timing perspective, participants were considered enrolled in the trial once they had their A_1 assessment. A four week rest period followed the A_1 assessment, where the child was enrolled in the trial but did not have access to the OGS for logistics reasons, allowing sufficient time for the units to be deployed. After four weeks, the author visited each child in their home and provided them with a standard introduction, orientation and training session, as describe earlier. The session was directed towards the child, but all family members who were present at the time were encouraged to observe and ask questions. In all instances, the child sat alongside the author throughout the session and viewed the screen at the same angle as the author. The time this took is reported in section 6.3.5.4.

At the completion of the standard introduction, orientation and training session, the author positioned the child in front of the OGS, adjusted the screen angle (if necessary), positioned the 'Orby' controller so it was easily accessed by the child, and watched the child interact with the OGS for the first few minutes. This was done to confirm that the child understood the demonstration, knew how to navigate the menu screen, start a game of their choosing, and successfully play it. The author also stayed for a few minutes once the child was set up in case a family member had

a question or in case the child questioned the vibration of the controller when they used it, if they were randomised to Group A.

At the end of the six week (42 day) trial, the author collected the OGS and left families with a single page '*Participant Experience Questionnaire*' (Appendix B) to complete and return in a reply paid envelope. This form asked the child if they enjoyed playing the OGS computer games (using a 10-point scale), to list any positive or negative outcomes from the trial, and to record if they received any physiotherapy, occupational therapy or Botulinum toxin during the trial, to account for any possible confounders to the study. On the same day the OGS was collected, most children attended the WCH for their A_2 assessment. This was the 'immediate' post-trial assessment. Four weeks later the child returned to the WCH for their A_3 'follow-up' assessment. There was 14 weeks between A_1 and A_3.

6.2.2 Additional Stage 2 Measures

Apart from the same Stage 1 sensory and hand function assessments, two other measures were introduced for Stage 2: the CP QOL and ABILHAND-Kids questionnaires, as indicated earlier (Chapter 3, section 3.4). The questionnaires were administered during the A_1 and A_2 assessment sessions to detect any changes following being a participant of the trial. On most occasions the questionnaire was completed by the parent during the assessment session while their child completed the tests and handed directly to the assessing therapist.

6.2.3 Trial Blinding

By using the 'Demo' mode (Chapter 5, section 5.6.5) to introduce and demonstrate the OGS to each child, the author was blinded as to group allocation when the orientation and training session occurred, ensuring that the author treated both groups exactly the same during the session. Additionally, the author read from a standardised protocol, meaning all participants heard the same information, received the same instructions, and saw the same games demonstrated. This is reported in section 6.3.5.4.

The children themselves were blinded as to the exact nature of the intervention under investigation to control for the placebo effect. That is, participants were not aware that one version of the OGS provided afferent haptic feedback during use and that another version didn't. Additionally, the assessing OTs were blinded as to group allocation and the consultant statistician was blinded as to which group was the intervention and which was the control.

6.2.4 Trial Follow-Along

After the initial set-up and demonstration of the OGS with the child and their family on Day 1, all parents were telephoned midway through the trial (end of week 3, start of week 4) to ensure that the system was operating correctly, to ask how often the OGS was being used, and to ascertain if any difficulties were being encountered. Families were able to contact the lead researcher via email and phone to report problems if any occurred during the trial.

6.2.5 Trial Recruitment

As reported in Chapter 4, 49 children formally consented to the Stage 1 study, with 42 children attending a sensory assessment session at the WCH. Six children were excluded for behavioural reasons, meaning complete data were recorded for 36 children (the only exception was subject #31 who completed 22 of the required 24 tests – this child refused to complete the second *AsTex*® test).

From this cohort of 36 children, eight children recorded intact or 'normal' somatosensation and were consequently excluded from Stage 2, meaning 28 children recorded abnormal or impaired somatosensory function. From this group of 28 children, two declined further participation in the study (subjects #30 and #33) and one child was excluded following professional advice from the assessing therapist (subject #2), who advised that their ND hand 'fisting' would cause problems when using the controller. This advice was valuable and was only possible because the assessing therapists were also consulted during the design and development of the intervention, particularly the controller (Chapter 5, section 5.4.2.2). Consequently, 25 children received written invitations in the mail (Appendix K) to

participate in the RCT of the OGS, as shown in Figure 6-2 (section 6.3.4), the *Consolidated Standards of Reporting Trials* (CONSORT) flow diagram for the study.

6.2.6 Trial Randomisation and OGS Set-Up

As consent forms for the RCT were received by the author, the child's relevant information was entered into an Excel spreadsheet, namely, their participant number (column A), first name (column B) and ND side (column C). The order of receiving consent forms in the mail became the trial randomisation order. Randomisation was conducted offsite by one of the PhD supervisory team (SH) who was not involved in the study recruitment or intervention (concealed allocation) to ensure the author was blinded as to a child's group allocation and hence unbiased during each child's introduction, orientation and training session with the OGS.

Randomisation occurred in blocks of ten using computer software (sequence generation), such that within a given block of ten children, five children would be randomly assigned to either Group A or Group B. SH would randomise each child as per the trial randomisation order and the outcome from the computer sequence, and put 'A' or 'B' in column D of the Excel spreadsheet to indicate the group they were assigned to. Without the author knowing, this spreadsheet was then emailed only to the project Research Assistant, to ensure that blinding and group allocation were not compromised. The Research Assistant would set up each laptop so the child's name and profile were loaded onto the laptop they would receive. The profile would add the child's first name to the log-in screen, along with 'Guest', and if they were allocated to the haptic group (Group A), it would deliver haptic vibration to their ND hand (as per the side indicated in column C of the Excel spreadsheet). The Research Assistant would enable the 'Demo' feature of the System and notify the author that a given laptop was set-up as per the randomisation requirements for the trial participant and ready for deployment.

During each introduction, orientation and training session the author would assess each child's ability to independently use the 'Orby' controller. The strap, and provision for a strap, wasn't mentioned or highlighted during the session as the aim was for the child to use their ND hand as much as possible if they could, without

knowing an option to support the ND hand was available. If the child struggled, the author would discuss hand strap options with the parents and fit a strap to the controller or leave a strap with the family to fit at a later date. Most parents were keen to see how long their child would persist without a strap and delayed using the strap if they could. Children who required a strap for their ND hand for the trial are discussed in section 6.3.11.

6.3 Results

6.3.1 The Stage 2 Cohort

Of the 25 children who were invited into Stage 2, 19 families completed and returned the consent form (76% response rate). One family withdrew their consent and declined participation in the RCT after their consent was received but prior to randomisation, due to existing family commitments, meaning 18 children participated in the trial. Apart from technical equipment issues (reported in section 6.3.12) that occurred during the trial, no adverse issues were reported.

The RCT cohort (n=18) ranged in age from six years and three months to 16 years and four months, with 16 children living within the Adelaide metropolitan area. Table 26 provides an overview of the cohort along with a comparison to the overall Stage 1 cohort (n=36) from which they had been recruited. As shown in Table 26, the RCT cohort was comparable to the Stage 1 cohort in that it recruited predominantly MACS Level II children, males, children classified as having a unilateral involvement, children with a left ND side, and children with a brain scan classification of '3' (cortical/subcortical lesions according to Krageloh-Mann and Horber (2007)). From a hand function classification (MACS Level) perspective, of the seven children who did *not* reply to the invitation to participate in the trial, six were Level II and one was Level III.

Table 26 – An overview of the cohort for the Stage 2 RCT (n=18), compared to the Stage 1 cohort they were recruited from

Category	***Stage 2 Cohort (n=18)***	***Stage 1 Cohort (n=36)***
Average age (years ± SD)	10.7 ± 3.4	10 ± 3.4
Median age (years)	9.6	8.8
MACS Level (n)	I(2), **II(10)**, III(3), IV(3)	I(9), **II(19)**, III(5), IV(3)
Sex	**12 males** (67%)	**22 males** (61%)
Unilateral : Bilateral	**13** : 5	**23** : 13
ND side (Left : Right)	**11** : 7	**23** : 13
Brain scan classification (n) [1]	1(2), 2(3), **3(7)**, 4(1), 5(1), 999(4)	1(2), 2(9), **3(13)**, 4(1), 5(1), 999(10)

Notes: **bold font** indicates predominant category or classification; SD = standard deviation; MACS = Manual Ability Classification System; [1] brain classification as per Krageloh-Mann and Horber (2007), see Table 7.

6.3.2 Randomisation Outcome

Randomisation occurred in two tranches – the first group of 10 children who started their trial between September 2013 and January 2014, and a second group of eight children who started their trial between February and October 2014, with the 18th child completing their trial in late November 2014. During the second group randomisation, the 19th and 20th allocation spaces were left open and unallocated, but remained available should extra children be recruited to the trial at a later date, which did not occur for this study.

For the first group of 10 children, five were allocated to each group as blocked. For the second group of eight children, five were allocated to Group A and three to Group B, meaning that 10 children were allocated to Group A and eight to Group B overall. Table 27 provides an overview of the cohort per randomisation group for Stage 2. Group A had a slightly older cohort with more males, a similar MACS Level distribution and unilateral : bilateral split as Group B, and more children with a ND left side. An Independent Sample t-Test was used to compare the ages of the children randomised to each Group, with the result not being statistically significant (p = 0.559). There was one withdrawal from each group.

Table 27 – An overview of all Stage 2 RCT participants, by group allocation

Category	***Group A (haptic) (n=10)***	***Group B (non-haptic) (n=8)***
Average age (years ± SD)	11.1 ± 3.1	10.1 ± 4.0
Median age (years)	10.5	8.9
MACS Level (n)	I(1), **II(5)**, III(2), IV(2)	I(1), **II(5)**, III(1), IV(1)
Sex	**8 males**	4 males
Unilateral : Bilateral	**7** : 3	**6** : 2
ND side (Left : Right)	**7** : 3	4 : 4
Withdrawals from trial	1	1
Games catalogue (old : new)	4 : **6**	**5** : 3
Brain scan classification (n) [1]	1(1), 2(2), 3(2), 4(1), 5(1), 999(3)	1(1), 2(1), **3(5)**, 4(0), 5(0), 999(1)

Notes: **bold font** indicates predominant category or classification; SD = standard deviation; MACS = Manual Ability Classification System; [1] brain classification as per Krageloh-Mann and Horber (2007), see Table 7.

From an OGS perspective, nine children received the original (or old) Games Catalogue and nine children received the 'Incentivised' (or new) Games Catalogue. This translated to six Group A and three Group B children receiving the new Games Catalogue, as shown in Table 27.

6.3.3 Additional Therapy Children Received During the RCT

The '*Participant Experience Questionnaire*' (Appendix B), reported in Chapter 3 (section 3.5), was also used to record any instances of therapy or treatment during the trial (Question 4). Children involved in the study were allowed to continue to undertake activities typical for them without change. Five families reported the following:

- Four children received weekly or twice weekly physiotherapy (reported as 'hydrotherapy' for two children), and
- One child received occupational therapy for 45 minutes every fortnight.

6.3.4 CONSORT 2010 Flow Diagram and Reporting

The Consolidated Standards of Reporting Trials (CONSORT) flow diagram for the trial appears in Figure 6-2, identifying the flow of participants from the initial pool of children (n=42) for Stage 1, through to the 18 children that were randomised for the Stage 2 RCT.

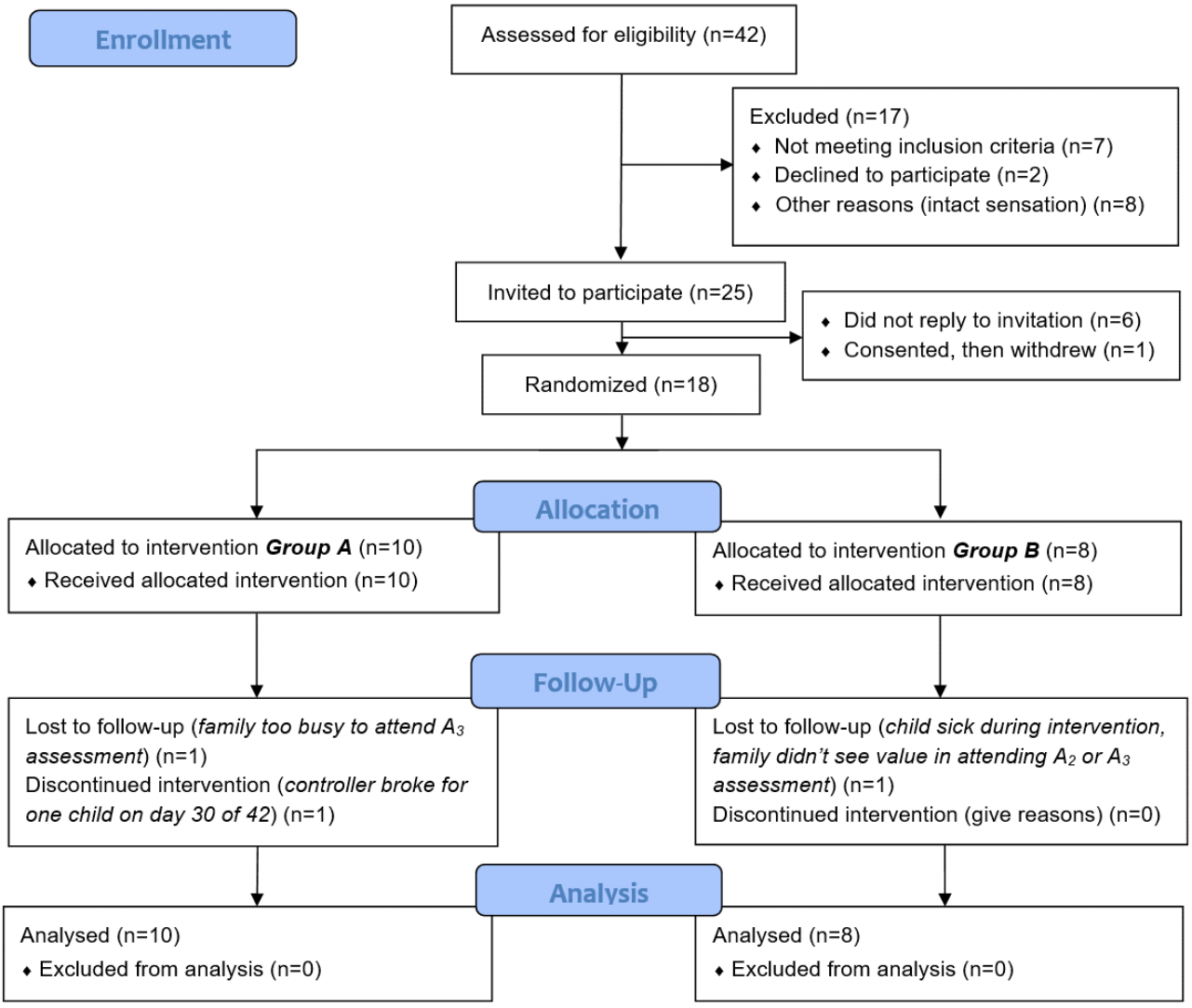


Figure 6-2 – The CONSORT 2010 Flow Diagram for the 18 children randomised to either Group A (vibration) or Group B (no vibration) for the Stage 2 RCT

6.3.5 Trial Fidelity

6.3.5.1 Compliance with the six week OGS Intervention

All 18 children who were allocated and set up with the OGS completed the six week trial. As reported in the CONSORT flow diagram, an 'Orby' controller broke for one participant (#37) on Day 30 of his trial, with the family not reporting the breakage until the OGS was collected on Day 42, meaning this child discontinued his intervention early. Problems that arose during the trial, including instances where a controller broke and was replaced, are reported in section 6.3.12.

6.3.5.2 Stage 2 Withdrawals

As reported in the CONSORT flow diagram, two children withdrew from Stage 2 post-OGS intervention. One Group A child (#4) attended their A_2 assessment but not their A_3 assessment. The family reported being too busy with other siblings and extra curricula activities to fit in the last assessment, despite the assessing OT offering to conduct the A_3 assessment within the family home to eliminate the need for travel to the WCH.

A post-trial analysis identified that this particular child used the OGS the least of all children, with only 117 mins of total game time accrued over seven of the 42 days, which could indicate a reason for their withdrawal (a lack of engagement and use by the child). This child's engagement and usage is substantially less than both the average total game time and daily usage for the whole cohort (377 minutes and 13 days), and all Group A children (403 mins and 14 days). This child was one of only two children who required a strap for their ND hand to use the controller, and was classified as having MACS Level III hand function. The parents reported that their son struggled and persisted with using the controller, and that the strap assisted but did not solve the problem associated with his particular hand impairment. Using the '*Participant Experience Questionnaire*' (Appendix B and section 6.3.13), the parents reported that their son "*would have liked to play more but had difficulty keeping the left hand in position*". This family rated the OGS 5/10, which was the second lowest rating received.

The second child (#9) to withdraw from Stage 2 was from Group B. One of the parents withdrew this child from the trial at the conclusion of the six week OGS intervention, meaning they did not attend their A_2 or A_3 assessments. The mother reported that their son was sick and off school for more than a week during the trial, so didn't think he engaged with it as much as he could have. Unfortunately, this child also experienced two 'Orby' controller failures during his trial, and although a new controller was swapped in to replace the broken controller each time (see section 6.3.12), and the trial extended to cover the days where the OGS was not available, their overall trial experience wasn't as positive as other children. This family gave the OGS the lowest rating using the '*Participant Experience Questionnaire*' (see section 6.3.13), which was 4/10.

A post-trial analysis identified that this particular child accrued 337 mins of total game time over 10 days. These values are less than the average for both the total game time and total number of days used for the whole cohort (377 minutes, 13 days), but close to the average for all Group B children (340 mins, 12 days). However, despite the parent's impression of how much her son used the OGS, this child's overall usage places them seventh overall when all children are ranked based on total time using and engaging with the OGS, as shown in Table 28. Both children who withdrew from the trial were early enrolees, male, and were allocated an OGS that used the original, non-incentivised Games Catalogue.

6.3.5.3 Compliance with Trial Assessment Timeframes

Excluding withdrawals, 14 (82%) of the 17 children who attended their immediate-post intervention A_2 assessment at the WCH did so on the same day or within one day of the OGS being collected by the author. For the remaining three children, the time between the A_2 assessment and the OGS collection was two days, five days, and 34 days, with the significant delay (34 days) for one child (#32) being due to a severe stomach virus that affected the child and both parents, combined with the Christmas-New Year closure period. As noted earlier, one child (#9) did not attend their A_2 assessment.

For participant #32, their A_2 assessment was closer to being an A_3 assessment (given it was approximately a month after their trial finished), but rather than omitting this particular child's A_2 assessment, it was decided to conduct it as soon as practical (which was 34 days later), and still have their 'follow up' A_3 assessment a further month later.

The average time between the A_2 and A_3 assessments was 34 days or almost five weeks, with the longest time being 53 days and the shortest being 17 days. Excluding withdrawals, four (25%) of the 16 children who remained in the trial returned to the WCH for their follow-up A_3 assessment four weeks after their A_2 assessment (±1 day). Only two children returned for their A_3 assessment in less than 28 days (17 days and 25 days). Reasons for the increased time between assessments were the juggling of busy family schedules, children returning to

school, and interruptions due to the Christmas and New Year break, where either staff were on annual leave or families were away on holiday.

Table 28 – OGS usage, ranked according to Total System Time (mins) over the six week trial, showing each child's age, sex, MACS Level, CP type, group allocation, number of days used, and the Games Catalogue type

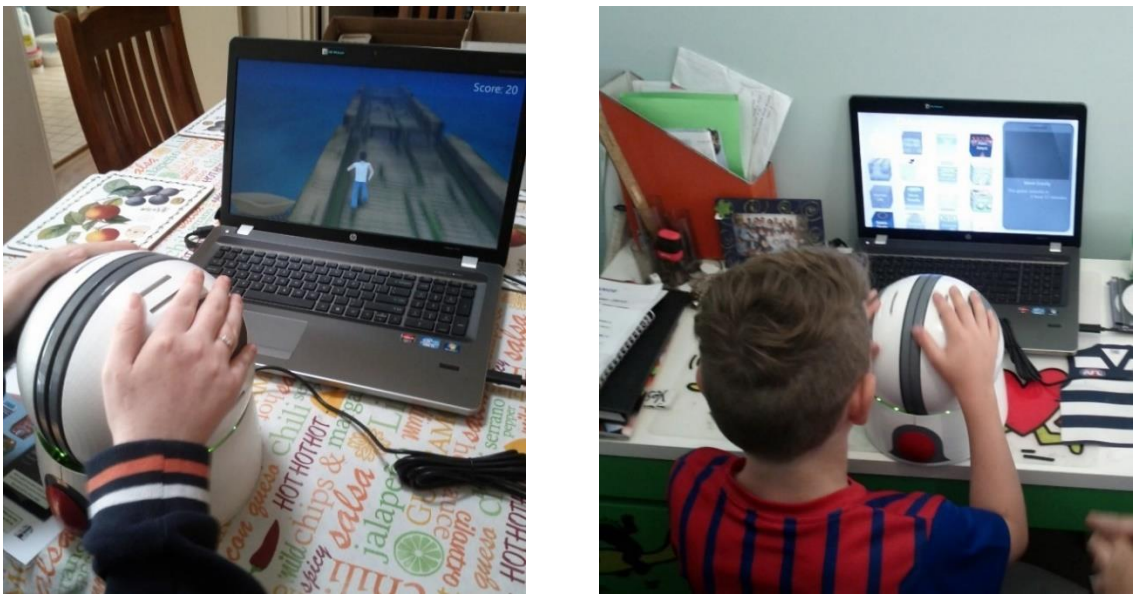
Subject #	*Age (yrs)*	*Sex (M / F)*	*MACS Level*	*CP Type (Uni / Bi)*	*Grp (A / B)*	*Total No. of Days Used*	*Games Cata-logue (Old / New)*	*Total System Time (mins)*
38	7.2	M	II	Uni	A	41	New	1140
21	8.2	M	II	Bi	A	14	New	877
46	10.5	M	II	Uni	B	9	New	566 (#)
8	6.8	F	II	Bi	B	13	Old	496
12	14.3	M	II	Uni	A	9	New	482
11	7.1	F	II	Uni	B	22	Old	431
9	15.8	M	II	Bi	B	10	Old	337
6	16.6	M	IV	Uni	B	12	New	333
47	9.0	F	III	Uni	B	13	New	303
29	9.8	M	IV	Uni	A	11	Old	302
27	11.2	F	II	Uni	A	17	New	292
31	7.7	M	III	Uni	A	17	New	238
37	13.5	M	II	Uni	A	13 (*)	Old	221
7	15.8	F	IV	Bi	A	5	Old	202
39	13.8	M	I	Bi	A	7	New	159
41	6.5	F	I	Uni	B	7	Old	158
32	8.7	M	II	Uni	B	6	Old	136
4	9.3	M	III	Uni	A	7	Old	117

Notes: ***Red*** font indicates a child who withdrew from Stage 2, post OGS intervention; M = male, F = female; (*) the 'Orby' controller broke on Day 30 for this child, meaning they engaged with the OGS for 13 of the 30 days available; (#) the profile for this child was tampered with, so this value represents all OGS usage, regardless of group allocation, as described in section 6.3.12 and Appendix N; Old = original Games Catalogue; New = Incentivised Games Catalogue; CP = cerebral palsy; Uni = unilateral, Bi = bilateral; MACS = Manual Ability Classification Scheme; Grp = Group; A = haptic system, B = non-haptic system.

6.3.5.4 OGS Set-Up and Participant Orientation and Training using the Study Protocol

Setting up each child with the OGS was unique and depended on the child and their home environment. The main requirements for the OGS were access to mains power (to ensure the laptop ran smoothly and didn't drain the laptop battery) and a flat, stable surface for the laptop and controller. On most occasions, the OGS was set up on the table in the family dining room (Figure 6-3(a)), in the lounge room, or the study (Figure 6-3(b)) on a desk or table, meaning it was in an open space and easily visible within the home. On one occasion it was set up in a child's bedroom as that was the only location that had space available.

Once a location was chosen (in conjunction with the family) the OGS was set up and running within minutes. A post-trial analysis identified that the average time to read through the study protocol and orientate the child to the OGS using the 'Demo' profile was 16.1 ± 1.9 minutes (range: 13.1 – 19.6 mins). The time was dependent on how many questions the child asked, how impatient the child was to have their turn on the OGS, if other family members had questions, and the child's familiarity with playing computer games.



(a) **(b)**

Figure 6-3 – The OGS set up in the family home. (a) On the dining room table and (b) On a desk in the study

6.3.6 Sensory and Motor Assessment Results

The raw data for the Stage 2 sensory and motor function assessments for the RCT appear in Table 29. Similar to the Stage 1 results, reading Table 29 from left to right indicates the test order, with the exception of the results for the second AsTex® test, which was conducted last (as discussed in section 6.2.1), but shown next to the results for the first AsTex® test. In this way, the presentation of the test results represent a hierarchy of perceptual difficulty from a brain processing perspective, as described in Chapter 3.

The following colour coding has been used in Table 29 to improve readability:

Colour coding:

12	Child (#) received the new incentivised games Catalogue
Bi	Child has bilateral (Bi) CP Type
A	Child was randomised to Group A
	No data – child withdrew from trial

The following notes accompany Table 29:

Notes: F = female; M = male; R = right; L = left; yrs = years; mths = months; Uni = unilateral; Bi = bilateral; CP = cerebral palsy; Grp = Group; Dosage = amount of time (in minutes), spent engaging with the OGS; Ax = assessment; **W/D** = withdrawn; f = finger; th = thumb; MACS = Manual Ability Classification System; Scan = brain classification as per Krageloh-Mann and Horber (2007), see Table 7; ND = non-dominant; Dom = dominant; Propriop. = Proprioception; Stereo. = Stereognosis; ***X*** = refused test; Nil = no result/threshold not detected; TX = test number X.

The random effects modelling for the RCT follows, with Table 30 reporting the summary statistics and estimated effects of treatment for the sensory and motor tests, for the *ND hand*, and Table 31 reporting the summary statistics and estimated effects of treatment on the sensory and motor tests for the *dominant hand*.

Table 29 – The sensory and motor function assessment results for the Stage 2 cohort (n=18)

Subject #	Gender	Ax No.	Ax Date	DOB	CP Type	Grp (A / B)	Dosage (mins)	ND side	MACS	Scan	Monofil				AsTex				Propriop. (10)		Stereo. (6)		Jebsen Taylor Hand Function Test (Tests 2-7)												Total Hand Scores	
											ND(f)	Dom(f)	ND(th)	Dom(th)	T1, ND	T1, Dom	T2, ND	T2, Dom	ND	Dom	ND	Dom	T2, ND	T2, Dom	T3, ND	T3, Dom	T4, ND	T4, Dom	T5, ND	T5, Dom	T6, ND	T6, Dom	T7, ND	T7, Dom	Total (ND)	Total (Dom)
4	M	A1	11/06/2013	12/05/2004	Uni	A	117	L	III	3	6.65	2.83	6.65	2.83	34.0	34.7	35.7	37.0	2	9	0	5	120	5	120	9	120	24	120	2	120	4	120	3	720	47
		A2	1/11/2013								4.56	3.61	4.56	3.61	28.0	36.3	35.3	37.0	1	10	1	5	120	4	120	8	120	25	120	2	120	4	120	5	720	48
		A3	W/D																																	
6	M	A1	2/05/2014	5/01/1998	Uni	B	333	R	IV	3	2.83	2.83	2.83	2.83	21.0	35.3	17.7	27.8	10	10	5	6	24	5	77	11	120	8	39	5	13	5	12	5	285	39
		A2	1/10/2014								2.44	2.44	2.44	2.36	21.3	21.7	22.3	29.0	6	10	2	6	27	4	92	8	110	14	13	5	10	5	10	4	262	40
		A3	30/10/2014								3.61	3.61	3.61	3.61	26.7	29.0	26.3	33.3	9	10	5	6	17	3	38	6	120	10	27	4	12	6	10	5	224	34
7	F	A1	11/06/2013	12/12/1997	Bi	A	202	R	IV	5	2.83	3.61	2.83	3.22	28.0	26.7	32.7	32.0	10	10	6	6	6	5	13	11	120	15	120	3	6	5	5	4	270	43
		A2	8/11/2013								2.83	2.83	2.83	2.83	37	35.3	37	37	8	10	6	6	7	4	13	9	120	9	4	3	8	5	7	6	159	36
		A3	6/12/2013								2.83	2.83	2.83	2.83	34.3	36.2	36.3	33.0	10	10	6	6	6	5	13	9	120	12	4	4	6	5	7	6	156	41
8	F	A1	17/06/2013	24/12/2006	Bi	B	496	R	II	3	2.83	2.83	2.83	2.83	27.5	23.2	27.0	22.7	10	10	5	6	7	7	10	7	22	40	3	2	6	5	7	6	55	67
		A2	9/12/2013								2.83	2.83	2.83	2.83	25.7	24.3	20.7	25.7	10	10	4	5	5	6	10	9	26	20	4	6	4	4	5	5	54	50
		A3	14/01/2014								2.83	2.83	2.83	2.83	19.5	25.5	21.7	21.5	9	10	6	6	5	7	9	9	11	15	3	4	4	6	6	5	38	46
9	M	A1	11/06/2013	25/12/1997	Bi	B	337	L	II	2	3.22	2.83	2.83	2.83	13.3	19.3	16.7	17.3	6	9	4	5	14	10	50	30	120	120	120	15	12	11	11	8	327	194
		A2	W/D																																	
		A3	W/D																																	
11	F	A1	24/06/2013	5/10/2006	Uni	B	431	R	II	1	2.83	2.83	2.83	2.83	27.0	33.2	31.0	29.0	8	10	4	6	28	4	120	7	60	15	8	3	11	3	7	3	234	35
		A2	16/12/2013								2.83	2.83	2.83	2.83	33.3	36.8	35.3	37	10	10	5	6	14	3	120	7	65	20	18	2	6	3	7	3	230	38
		A3	7/02/2014								2.83	2.83	2.83	2.83	33.2	35.2	30.8	34.8	10	10	4	6	15	4	48	6	40	9	27	2	6	3	15	3	151	27
12	M	A1	19/07/2013	15/10/1999	Uni	A	482	R	II	3	2.83	2.83	2.83	2.83	28.8	28.3	32.2	31.0	9	9	4	6	7	4	15	7	30	13	5	2	5	3	4	3	66	32
		A2	25/03/2014								2.83	2.83	2.83	2.83	29.3	36.7	33.3	36.3	10	10	4	6	6	4	15	8	27	8	6	4	7	3	5	4	66	31
		A3	6/05/2014								2.83	3.22	3.61	3.22	36.3	36.2	35.3	34.0	9	9	4	6	11	4	20	8	54	8	7	3	7	3	5	3	104	29
21	M	A1	16/09/2013	3/01/2006	Bi	A	877	R	II	999	2.83	2.83	2.83	2.83	34.3	32.7	31.7	31.7	9	10	5	6	5	6	11	8	120	30	9	3	6	4	6	4	157	55
		A2	15/04/2014								2.83	2.83	2.83	2.83	23.0	28.8	24.0	24.7	9	10	6	5	6	8	12	7	120	19	6	5	6	4	5	5	155	48
		A3	16/05/2014								2.83	2.83	2.83	3.22	33.8	31.7	30.7	29.3	10	9	5	6	7	8	11	8	12	26	8	5	6	5	5	3	49	55
27	F	A1	30/09/2013	22/12/2002	Uni	A	292	L	II	2	2.83	2.83	2.83	2.83	32.3	36.7	32.3	34.0	10	10	6	6	8	10	8	11	33	25	3	1	4	4	4	5	60	56
		A2	15/04/2014								2.83	2.83	2.83	2.83	36.0	33.0	22.7	23.8	10	10	6	6	7	10	10	8	58	13	12	9	7	4	5	5	99	49
		A3	20/05/2014								2.83	2.83	2.83	2.83	29.5	31.7	29.8	27.0	10	10	6	6	15	11	9	9	23	26	10	6	6	4	4	4	67	60
29	M	A1	7/06/2013	16/12/2003	Uni	A	302	L	IV	1	2.83	2.83	2.83	2.83	24.3	27.0	21.3	22.3	3	8	2	2	8	4	18	8	120	55	6	15	9	4	16	5	177	91
		A2	15/11/2013								2.83	2.83	2.83	2.83	25	20.7	30	28.3	7	8	3	4	7	4	23	11	120	120	23	4	7	3	6	4	186	146
		A3	19/12/2013								2.83	2.83	2.83	2.83	20.5	19.7	32.0	30.3	1	7	3	5	10	4	16	11	120	120	20	4	7	4	10	5	183	148
31	M	A1	11/03/2014	7/08/2006	Uni	A	238	L	III	999	4.31	2.83	4.31	2.83	37	37	37	37	4	9	2	5	40	9	73	11	120	18	21	14	120	6	120	6	494	64
		A2	30/05/2014								4.31	4.31	2.83	2.83	32.0	37.0	29.0	36.7	5	9	4	6	39	7	70	11	120	90	120	9	120	6	120	4	589	127
		A3	27/06/2014								4.56	2.83	4.56	2.83	29	37	X	X	X	X	1	5	29	9	120	11	120	43	120	7	41	6	120	6	550	82
32	M	A1	21/06/2013	9/02/2005	Uni	B	136	L	II	3	4.56	2.83	4.56	2.83	28.3	27.3	31.0	31.0	8	10	1	6	120	5	120	12	120	15	120	3	120	4	120	4	720	43
		A2	14/01/2014								Nil	2.83	Nil	2.83	9.5	26.5	6.5	21.7	6	9	1	6	120	12	120	14	120	12	31	7	46	6	120	5	557	56
		A3	7/03/2014								4.56	2.83	4.56	2.83	9.3	28.5	7.3	24.3	10	10	0	6	120	8	120	7	120	14	120	5	30	4	30	4	540	42
37	M	A1	7/06/2013	12/04/2000	Uni	A	221	L	II	4	2.83	2.83	2.83	2.83	35.0	35.3	35.7	37.0	3	10	1	6	18	3	107	7	35	9	16	2	12	2	10	3	198	26
		A2	22/11/2013								2.83	3.61	2.83	3.84	36	35.8	36	37.0	8	10	1	6	12	2	57	7	120	11	12	2	7	3	8	2	216	27
		A3	20/12/2013								2.83	2.83	2.83	3.84	37.0	37.0	37.0	37.0	10	10	2	6	11	3	50	8	31	9	20	2	6	3	6	2	124	27
38	M	A1	21/06/2013	11/11/2006	Uni	A	1140	L	II	2	2.83	2.83	2.83	2.83	22.0	24.0	18.7	25.0	10	10	6	6	31	7	51	5	120	21	120	1	120	3	120	3	562	40
		A2	4/03/2014								2.83	2.83	2.83	2.83	13.5	33.2	35.7	37.0	9	10	5	5	51	5	79	6	120	20	44	5	27	5	51	4	372	45
		A3	8/04/2014								2.83	2.83	2.83	2.83	29.7	34.7	36.0	37.0	10	10	5	6	25	4	70	6	120	24	24	6	22	6	16	5	277	51
39	M	A1	30/09/2013	21/06/2000	Bi	A	159	L	I	999	2.83	2.83	2.83	2.83	36.3	31.0	35.0	36.7	10	10	4	5	11	9	11	11	120	114	3	2	7	6	6	6	158	148
		A2	6/05/2014								2.83	3.61	2.83	3.61	37.0	35.3	30.0	27.0	10	10	5	5	9	8	14	11	28	11	4	6	8	6	7	6	70	48
		A3	10/06/2014								2.83	2.83	2.83	2.83	37.0	37.0	30.8	29.3	10	10	6	5	9	7	17	14	42	45	7	5	6	6	6	5	87	82
41	F	A1	7/06/2013	26/03/2007	Uni	B	158	L	I	3	2.83	2.83	2.83	2.83	30.3	35.7	37.0	37.0	10	8	6	6	3	6	5	14	120	20	8	2	6	3	5	3	147	48
		A2	1/11/2013								2.83	2.83	2.83	2.83	27.8	30	29.7	29.8	10	10	6	6	8	4	14	6	120	16	11	2	6	2	5	4	164	34
		A3	6/12/2013								2.83	2.83	2.83	2.83	34.5	31.0	37.0	37.0	10	9	6	6	5	4	12	7	120	29	6	3	6	3	5	3	154	49
46	M	A1	10/09/2014	14/04/2004	Uni	B	566	L	II	3	2.36	2.36	2.36	2.36	30.3	29.3	29.3	33.7	10	10	6	6	8	4	42	8	82	25	10	2	9	6	9	4	160	49
		A2	24/11/2014								2.36	2.36	2.36	2.36	32.3	37.0	34.7	37.0	10	10	5	5	10	4	19	8	30	26	7	5	10	7	7	5	83	55
		A3	19/12/2014								2.44	2.36	2.44	2.36	32.3	28.3	36.0	36.3	10	10	5	5	9	3	15	10	45	33	9	5	9	4	8	4	95	59
47	F	A1	28/01/2014	20/04/2005	Uni	B	303	R	III	999	Nil	2.83	Nil	2.83	24.7	22.8	20.3	28.2	4	10	1	5	120	4	120	7	120	11	107	7	120	3	120	4	707	36
		A2	10/06/2014								Nil	2.83	Nil	2.83	17.0	26.3	10.8	25.0	6	10	0	6	120	4	120	6	120	17	120	8	120	6	120	6	720	47
		A3	27/06/2014								Nil	2.83	Nil	2.83	16.0	16.3	16.7	21.3	5	10	1	6	120	4	120	7	120	19	120	9	120	8	120	5	720	52

Table 30 – *Non-dominant* hand summary statistics and estimated effects of treatment (Group B versus Group A) at visits 2 (A_2) and 3 (A_3) on sensory and motor tests for 18 children with CP randomised to vibration treatment (Group A) or not (Group B)

Test	*Visit 1 (A_1)*		*Visit 2 (A_2)*		*Visit 3 (A_3)*		*β (95% CI) Group X Visit 2*[1]	*β (95% CI) Group X Visit 3*[1]
	A	*B*	*A*	*B*	*A*	*B*		
Monofil (Thumb)	3.36±1.25	3.01±0.71	3.00±0.55	2.66±0.24	3.11±0.60	3.18±0.78	0.24 (-0.31, 0.79) (*p*=0.40)	0.16 (-0.37, 0.70) (*p*=0.55)
Monofil (Finger)	3.36±1.25	3.07±0.70	3.15±0.68	2.66±0.24	3.02±0.58	3.18±0.78	0.09 (-0.34, 0.53) (*p*=0.67)	0.23 (-0.19, 0.66) (*p*=0.28)
AsTex (Test 1)	31.21±5.17	25.31±5.73	29.68±7.63	23.85±8.56	31.91±5.37	24.50±9.73	-0.81 (-7.13, 5.51) (*p*=0.80)	-2.43 (-8.85, 3.99) (*p*=0.46)
AsTex (Test 2)	31.21±6.21	26.25±7.28	31.30±5.06	22.86±11.25	33.50±2.95	25.12±10.75	-4.10 (-11.09, 2.89) (*p*=0.25)	-4.51 (-11.74, 2.72) (*p*=0.22)
Proprioception	7.00±3.50	8.25±2.25	7.70±2.83	8.29±2.14	8.75±3.15	9.00±1.83	-0.89 (-2.87, 1.09) (*p*=0.38)	-0.33 (-2.38, 1.73) (*p*=0.76)
Stereognosis	3.60±2.22	4.00±2.00	4.10±1.91	3.20±2.20	4.22±1.86	3.86±2.41	***-1.21 (-2.20, -0.23) (p=0.02)***	-0.42 (-1.42, 0.58) (*p*=0.41)
Total JTHFT score	286.2±227.1	329.4±251.7	263.2±226.2	295.7±249.9	177.4±155.8	274.6±254.8	-11 (-77, 55) (*p*=0.74)	5 (-61, 72) (*p*=0.87)

Notes: [1]Group X visit interaction term from a random effects model with group (A or B), visit (1, 2 or 3) and group X visit interaction terms included as fixed effects and subject ID included as a random intercept. Treatment effects at each visit from this model are adjusted for any differences between groups at baseline. Two separate measures were performed for each subject (one on the dominant side, and one on the non-dominant side). Summary statistics are mean ± standard deviation; JTHFT = Jebsen Taylor Hand Function Test.

Table 31 – *Dominant hand* summary statistics and estimated effects of treatment (Group B versus Group A) at visits 2 (A_2) and 3 (A_3) on sensory and motor tests for 18 children randomised to vibration treatment (Group A) or not (Group B)

Test	*Visit 1 (A_1)*		*Visit 2 (A_2)*		*Visit 3 (A_3)*		β (95% CI)	β (95% CI)
	A	B	A	B	A	B	Group X Visit 2[1]	Group X Visit 3[1]
Monofil (Thumb)	2.87±0.12	2.77±0.17	3.09±0.41	2.70±0.17	3.03±0.35	2.87±0.37	-0.29 (-0.63, 0.05) (*p*=0.09)	-0.06 (-0.40, 0.28) (*p*=0.71)
Monofil (Finger)	2.91±0.25	2.77±0.17	3.21±0.53	2.71±0.21	2.87±0.13	2.87±0.37	-0.37 (-0.77, 0.03) (*p*=0.07)	0.14 (-0.27, 0.54) (*p*=0.50)
AsTex (Test 1)	31.34±4.63	28.27±6.17	33.21±5.03	28.95±5.99	33.44±5.59	27.69±5.82	-1.87 (-6.73, 2.99) (*p*=0.45)	-3.61 (-8.54, 1.33) (*p*=0.15)
AsTex (Test 2)	32.37±5.20	28.33±6.15	32.48±5.75	29.30±5.90	32.13±3.72	29.81±7.10	0.08 (-5.13, 5.29) (*p*=0.98)	0.20 (-5.19, 5.58) (*p*=0.94)
Proprioception	9.50±0.71	9.63±0.74	9.70±0.67	9.86±0.38	9.38±1.06	9.86±0.38	-0.03 (-0.53, 0.47) (*p*=0.91)	0.37 (-0.15, 0.89) (*p*=0.17)
Stereognosis	5.30±1.25	5.75±0.46	5.40±0.70	5.71±00.49	5.67±0.50	5.86±0.38	-0.19 (-0.86, 0.48) (*p*=0.58)	-0.39 (-0.97, 0.39) (*p*=0.41)
Total JTHFT score	60.2±35.8	63.9±53.6	60.5±41.0	45.7±8.6	63.9±37.2	44.1±10.9	-3.73 (-28.3,20.9) (*p*=0.77)	-7.4 (-32.5,17.6) (*p*=0.56)

Notes: [1]Group X visit interaction term from a random effects model with group (A or B), visit (1, 2 or 3) and group X visit interaction terms included as fixed effects and subject ID included as a random intercept. Treatment effects at each visit from this model are adjusted for any differences between groups at baseline. Two separate measures were performed for each subject (one on the dominant side, and one on the non-dominant side). Summary statistics are mean ± standard deviation; JTHFT = Jebsen Taylor Hand Function Test.

6.3.7 Statistical Analysis of Sensory and Motor Test Results

During this trial, Group A (n=10) children received the haptic version of the OGS, while Group B (n=8) children received the non-haptic version. Additionally, nine children received the Original Games Catalogue and nine children received the Incentivised one, as shown in Table 27.

As reported in Table 30, the statistical modelling comparing sensory and motor assessment results by Group (A and B) and visit (A_1, A_2 and A_3) for the *ND hand* identified only one statistically significant result, which was for the test of stereognosis between the Groups at the immediate follow-up A_2 assessment (visit 2). This result revealed children in Group A (haptic group) performing better in the A_2 stereognosis assessment than children in Group B (non-haptic group), combined with the fact that Group B children performed worse in stereognosis during their A_2 assessment. This significant between Group difference did not persist at the follow-up A_3 assessment (visit 3). There was a trend for both groups to record improved results for the test of proprioception and for the JTHFT from A_1 through to A_3, but only Group A children also recorded a trend towards improved stereognosis results. As reported in Table 31, the statistical modelling comparing sensory and motor assessment results by Group (A and B) and visit (A_1, A_2 and A_3) for the *dominant hand* identified no statistically significant results for any measure between the groups at the different assessment points.

Working with the consulting statistician, a secondary exploratory analysis and remodelling was conducted, this time investigating differences between the assessment measures at the different time points, per hand, when all children are considered as a single cohort (n=18). Analysis revealed a statistically significant improved result for the SWM test for the ND thumb between A_1 and A_2 (β coefficient = -0.27, CI (-0.54, -0.01), $p = 0.043$), which did not persist at A_3 (β coefficient = 0.37, CI (-0.21, 0.31), $p = 0.712$), and a statistically significant improved result for the ND hand for the total time taken to complete the JTHFT between A_1 and A_3 (β coefficient = -58.03, CI (-91.22, -24.84), $p = 0.001$). The total time taken for the JTHFT for the ND hand for the cohort appears in Table 32, with the cohort ordered by change in total test times between A_1 and A_3, which were 14 weeks apart. A negative time indicates a decreased (or improved) total time at A_3 compared to A_1.

Table 32 – Total JTHFT times for the ND hand for the Stage 2 cohort (n=16), comparing total time taken for the follow-up assessment (A_3) to the baseline assessment (A_1), ordered by difference in total JTHFT time

Subject No.	*Age (yrs.)*	*CP Type*	*Group*	*Scan*	*ND side*	*MACS*	*Dosage (mins)*	*JTHFT $A_3 - A_1$ (secs)*
38	7.2	Uni	A	2	L	II	1140	-285
32	8.7	Uni	B	3	L	II	136	-180
7	15.8	Bi	A	5	R	IV	202	-114
21	8.2	Bi	A	999	R	II	877	-108
11	7.1	Uni	B	1	R	II	431	-83
37	13.5	Uni	A	4	L	II	221	-74
39	13.8	Bi	A	999	L	I	159	-71
46	10.5	Uni	B	3	L	II	566	-65
6	16.6	Uni	B	3	R	IV	333	-61
8	6.8	Bi	B	3	R	II	496	-17
29	9.8	Uni	A	1	L	IV	302	6
27	11.2	Uni	A	2	L	II	292	7
41	6.5	Uni	B	3	L	I	158	7
47	9	Uni	B	999	R	III	303	13
12	14.3	Uni	A	3	R	II	482	38
31	7.7	Uni	A	999	L	III	238	56

Notes: No. = number; yrs. = years; CP = cerebral palsy; Uni = unilateral; Bi = bilateral; Group = RCT Group (A or B); ND = non-dominant; L = left; R = right; Scan = brain scan classification as per Krageloh-Mann and Horber (2007), see Table 7; MACS = Manual Ability Classification System; Dosage = OGS usage; mins = minutes; JTHFT = Jebsen Taylor Hand Function Test; secs = seconds.

Sixteen children appear in Table 32, since child #4 from Group A and child #9 from Group B withdrew and did not attend their A_3 assessment. As reported earlier (Chapter 3, section 3.3.5), the MCID for the JTHFT is 5.87 seconds for the ND hand (Reedman *et al.*, 2015), meaning 10 children recorded a clinically significant improvement for this test (the top portion of Table 32). All four children with bilateral CP and seven of the nine children classified as being MACS Level II belong to the group that recorded clinically significant results for the JTHFT.

Welch's t-Test was used to compare the children who recorded clinically significant results (n=10) to those that did not (n=6) from Table 32, to determine if the amount of time each child engaged with the OGS ('Usage' in Table 32) was a significant factor. The group that improved clinically where shown to engage with the OGS for 456.1 ± 330.8 minutes (7.6 ± 5.5 hours), while the group that did not improve clinically, engaged with the OGS for 295.8 ± 106.9 minutes (4.9 ± 1.8 hours), however, the difference in usage is not statistically significant (p=0.183).

Similarly, there was no statistically significant difference between each group based on age (group that improved: n=10, average age = 10.8 ± 3.8 years, group that did not improve: n=6, average age = 9.8 ± 2.8 years, p = 0.558). Figure 6-4 shows a graph of the total time taken to complete the JTFHT for the ND hand, for all Stage 2 children with complete data (n=16), with each child plotted in a different colour (legend shown below the x-axis). The black dashed line represents the median JTHFT results for the Stage 2 cohort overall, which were 187.5 seconds (A_1), 175 seconds (A_2), and 152.5 seconds (A_3), for a median improvement of -35 seconds overall between A_3 and A_1. The median improvement between A_3 and A_1 was 27.5 seconds for the unilateral group and 89.5 seconds for the bilateral group.

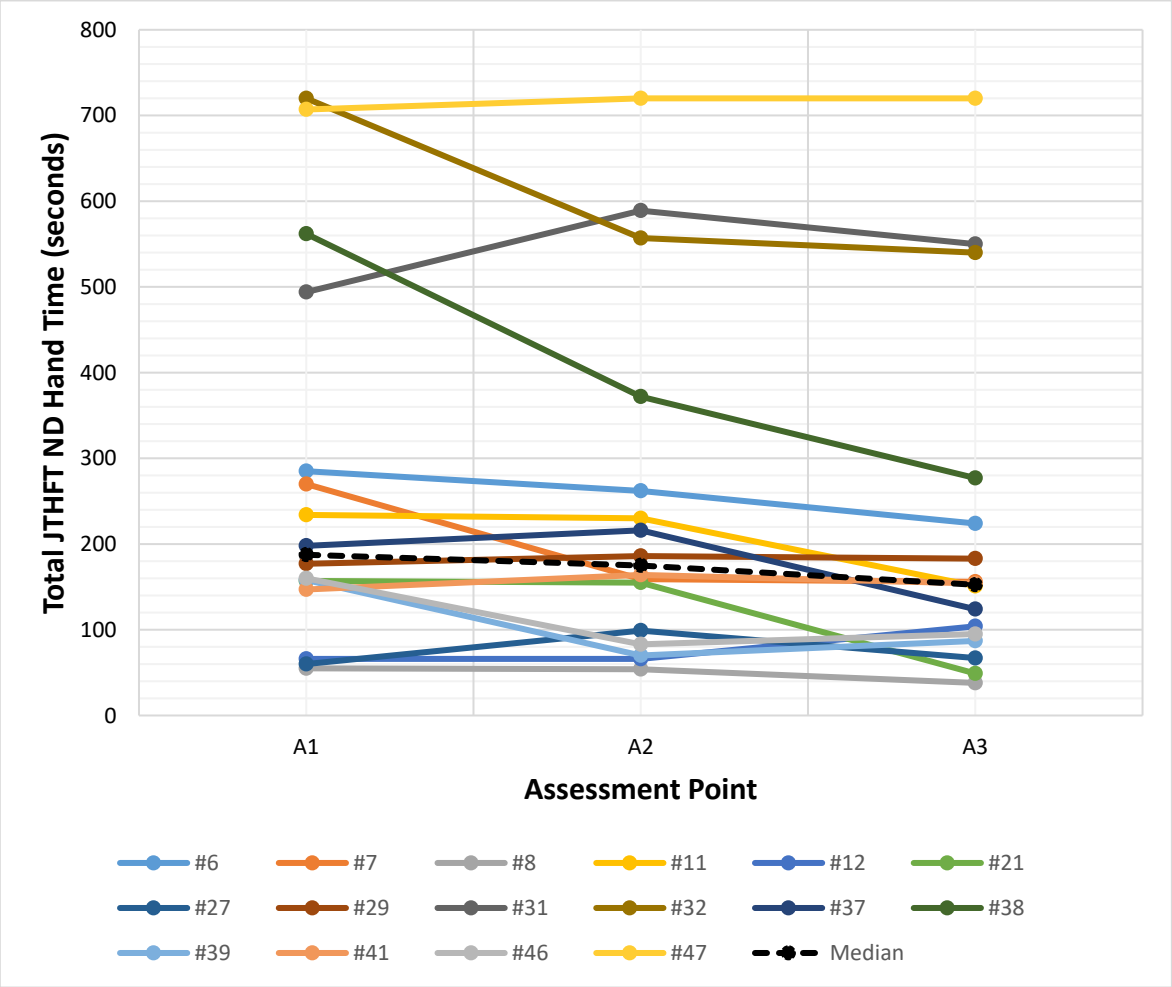


Figure 6-4 – Graph of Total Time Taken for the JTHFT for the ND Hand per Stage 2 child with complete data, for the assessment points A_1, A_2 and A_3. The black dashed line represents the median results for all children

6.3.8 ABILHAND-Kids Questionnaire Results

Table 33 shows the results of the Rasch analysis processing for the ABILHAND-Kids questionnaire, ordered by their calculated logit change and trial Group (A or B). Two children (#9 from Group B and #31 from Group A) do not appear in Table 33 as post-trial ABILHAND-Kids forms were not received for these children. The eight children who received the 'Incentivised' Games Catalogue are highlighted in blue.

Table 33 – The results of the ABILHAND-Kids questionnaire Rasch analysis (n=16) from the RCT, ordered by change in logit and group allocation (A or B)

Subject No.	*CP Type*	*Scan*	*ND Side*	*MACS Level*	*Pre-Patient Measure (logits)*	*SE (logits)*	*Post-Patient Measure (logits)*	*SE (logits)*	*Change (logits)*	*Usage (mins)*
Group A (haptic group, n=9)										
21	Bi	999	R	II	1.911	0.535	4.342	0.765	2.43	877
4	Uni	3	L	III	0.217	0.489	1.24	0.52	1.02	117
37	Uni	4	L	II	1.52	0.445	2.395	0.484	0.88	221
7	Bi	5	R	IV	0.68	0.416	1.203	0.426	0.52	202
39	Bi	999	L	I	2.009	0.57	2.365	0.489	0.36	159
27	Uni	2	L	II	1.967	0.471	1.963	0.456	0	292
29	Uni	1	L	IV	0.102	0.448	-0.164	0.411	-0.27	302
12	Uni	3	R	II	1.571	0.438	1.203	0.426	-0.37	482
38	Uni	2	L	II	1.318	0.471	-0.52	0.437	-1.84	1140
Group B (non-haptic group, n=7)										
6	Uni	3	R	IV	-0.332	0.412	0.34	0.412	0.67	333
8	Bi	3	R	II	1.571	0.428	2.172	0.468	0.60	496
46	Uni	3	L	II	1.384	0.431	1.763	0.446	0.38	566
32	Uni	3	L	II	1.384	0.431	1.571	0.438	0.19	136
11	Uni	1	R	II	2.499	0.566	2.685	0.61	0.19	431
41	Uni	3	L	I	0.68	0.416	0.68	0.416	0	158
47	Uni	999	R	III	3.9	0.663	1.963	0.456	-1.94	303

Notes: No. = number; CP = cerebral palsy; Scan = brain classification as per Krageloh-Mann and Horber (2007), see Table 7; ND = non-dominant; MACS = Manual Ability Classification Scheme; Uni = unilateral, Bi = bilateral; L = left; R = right; SE = standard error; Change = (Post – Pre) Patient Measure; Usage = OGS usage; mins = minutes; blue 'Subject No' = child received new 'Incentivised' Games Catalogue; red font = decrease in logit change; green font = increase in logit change.

As can be seen in Table 33, 10 children recorded 'Patient Measure' logit changes that increased, four recorded changes that decreased, and two recorded no change between their pre- and post-trial measure. Of the 10 children to record logit increases, five were randomised to Group A and five to Group B. The average increase for Group A (n=9) was +1.04 logits (range: 0.36 – 2.43), which is more than

double the average increase for Group B (n=7), which was +0.41 logits (range: 0.19 – 0.67). For the four children that recorded decreased logit scores, three were randomised to Group A (average decrease = -0.82, range: -1.84 to -0.37) and one to Group B (-1.94). As a cohort (n=16), the overall average change was a +0.18 logit increase.

Using an Independent Samples t-Test, there was no difference in ABILHAND-Kids scores between groups at either the A_1 assessment (Group A = 1.255 ± 0.743, Group B = 1.5837 ± 1.340, p = 0.358) or the A_2 assessment (Group A = 1.559 ± 1.454, Group B = 1.596 ± 0.826, p = 0.258). There was also no significant difference between the change in logit scores between the groups (Group A = 0.303 ± 1.167, range: -1.84 to 2.43; Group B = 0.013 ± 0.893, range: -1.94 to 0.67; p = 0.491).

In terms of CP type, all children with bilateral CP (n=4) only recorded *increases* in logit scores (average increase = +0.98 logits, range: 0.36 to 2.43), whereas children with unilateral CP (n=12) recorded an average logit decrease of -0.09 (range: -1.94 to 1.02). Following personal e-mail communication with the developers and authors of the ABILHAND-Kids questionnaire in September 2015 (Dr Carlyne Arnould and Dr Yannick Bleyenheuft), the reported MCID for this measure is +0.71 logits, with this work now appearing in the literature (Bleyenheuft *et al.*, 2017), while Preston *et al.* (2016) used a change that was greater than the standard error or SE (0.44 logits) to be clinically significant.

Using the Bleyenheuft *et al.* (2017) MCID criteria, three children (#4, #21 and #37) achieved a clinically significant increase according to the ABILHAND-Kids measure, with all three children belonging to Group A. Using the same criterion, two children recorded a clinically significant decrease (#38 and #47), one from each group. However, using the SE criterion as per Preston *et al.* (2016), six children (#4, #6, #7, #8, #21 and #37) achieved a clinically significant increase, four from Group A and two from Group B, and the same two children noted earlier achieved a clinically significant decrease (#38 and #47). With the exception of child #4 (who withdrew from the trial), the group of six children who achieved a clinically significant increase using the lower SE criteria also all appear in the top section of Table 32, which ranks the cohort in terms of clinically significant improvements in ND hand times for the JTHFT.

Further, with the exception of child #38 (who engaged with the OGS the most and recorded the largest improvement in JTHFT times), every child who appears in the top section of Table 32 also recorded a positive logit change, and every child who recorded a negative or no logit change appears in the bottom section of Table 32, where clinically significant JTHFT changes were not recorded, as shown in Table 34.

Table 34 – A comparison of pre- and post-RCT results for the ABILHAND-Kids and JTHFT test times for the ND hand, ordered by change in JTHFT score (n=15)

Subject #	*Age (yrs)*	*ABILHAND-Kids logit score change*	*JTHFT A_3-A_1 (ND hand)(secs)*
38	7.2	-1.84	-285
32	8.7	0.19	-180
7	15.8	0.52	-114
21	8.2	2.43	-108
11	7.1	0.19	-83
37	13.5	0.88	-74
39	13.8	0.36	-71
46	10.5	0.38	-65
6	16.6	0.67	-61
8	6.8	0.6	-17
29	9.8	-0.27	6
27	11.2	0	7
41	6.5	0	7
47	9	-1.94	13
12	14.3	-0.37	38

Notes: yrs. = years; JTHFT = Jebsen Taylor Hand Function Test; A_1 = baseline assessment; A_3 = follow-up assessment; secs = seconds.

A study by Klingels, Demeyere*, et al.* (2012) that investigated upper limb impairments in 81 children with only unilateral CP reported that children older than 10 years of age (n=43) performed significantly better than children younger than 10 years old (n=38) when assessed using the ABILHAND-Kids questionnaire (pg. 478). However, when the current cohort was analysed using Welch's t-Test, the difference between the two sub-groups based on age (older group: n=7, mean result = 0.35 ± 0.42, range = -0.37 to +0.88; younger group: n=8, mean result = 0.04 ± 1.35, range = -1.94 to +2.43) was not statistically significant (p=0.535). The older sub-group only recorded one instance (child #12) of a negative ABILHAND-Kids result compared to three in the younger sub-group, with a greater variation in age (younger cohort aged 6.5 to 9.8 years, older cohort aged: 10.5 to 16.6 years), but the numbers for the

current study are too small compared to those of Klingels *et al*. to draw any further comparisons.

6.3.9 OGS Usage

An analysis of the log files identified that the overall average OGS usage for the trial for all 18 participants was 377 ± 267 minutes or 6.3 ± 4.5 hours (median usage = 302.5 minutes; range: 117 – 1140 minutes), representing an average usage of 63 minutes per week, or 9 minutes per day. This value is greater than the seven minutes per day reported by Preston *et al.* (2016) for their home-based SG intervention cohort (n=8), with the authors reporting no significant differences between the gaming group and their control group. The average 'Guest' OGS usage was 145 ± 142 mins (2.4 ± 2.4 hours). The overall average OGS usage was not statistically significantly different from the usage by those children who also recorded clinically significant JTHFT results (section 6.3.7, Table 32, p=0.528, using an Independent Samples t-Test), or the group that did not record clinically significant JTHFT results (p=0.300). A graph of OGS usage per trial participant, for both the child ('Participant'), and any friend or family members who played the OGS using the 'Guest' profile, is shown in Figure 6-5. The graph shows total OGS usage as a function of trial order, from first OGS deployment to the last. Consequently, Figure 6-5 also shows when the new 'Incentivised' Games Catalogue was introduced and the affect this had on OGS usage.

As shown in Figure 6-5, assuming that the child who received the OGS *always* used their individual profile when they played games and that everyone else used the 'Guest' profile, in almost all instances the child played with the OGS more than their family members and/or friends. When analysed this difference was statistically significant (p<0.001, using a Paired Samples t-Test). There were only two instances where the 'Guest' profile logged more time with the OGS than the child enrolled in the trial, and this occurred once for each type of Games Catalogue deployed, and once for each group (Group A or B). An analysis of the log files to calculate total OGS usage, per Group allocation and Games Catalogue that was deployed, for both the child and the Guest profile, is shown in Table 35.

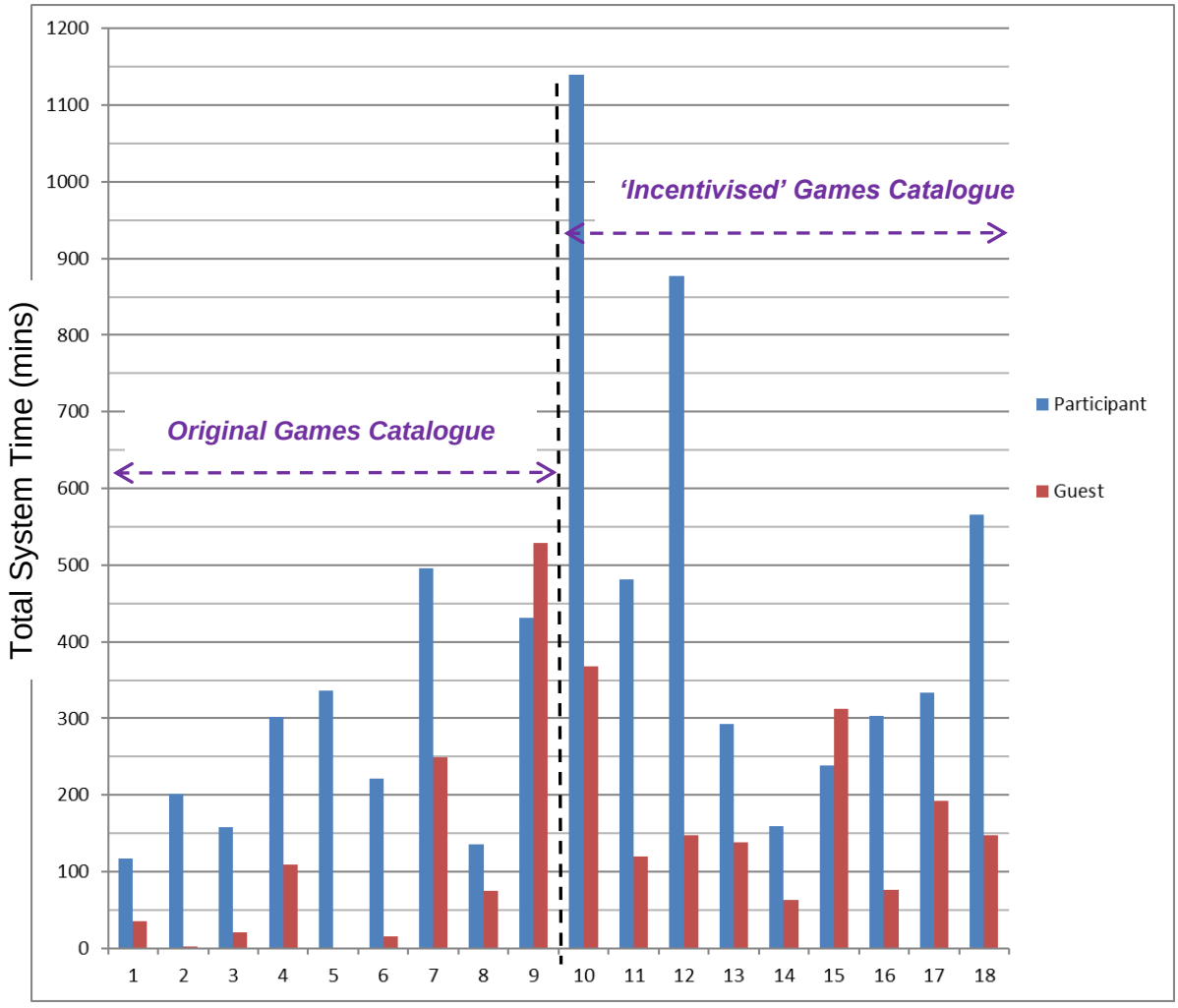


Figure 6-5 – OGS usage (in minutes) for both the child ('Participant', in blue) and their family and friends ('Guest', in red), for both the Original Games Catalogue (first 9 children) and the incentivised Games Catalogue (second 9 children)

Comparing OGS usage between Groups (A and B) identified that child usage and engagement as a function of their Group was not statistically significant (p = 0.070, using an Independent Samples t-Test). Investigating OGS usage as a function of the Games Catalogue each child received revealed that switching to an Incentivised Games Catalogue meant that children used and engaged with the OGS on average for an extra 221 minutes (median increase = 112 minutes) and for five extra days (median increase = 3 extra days) over the six week trial period. Additionally, when all children are ranked according to OGS usage (Table 28), four of the top five children received the Incentivised Catalogue. When OGS usage is analysed based on the Games Catalogue each child received, the result is not statistically significant (p=0.057). However, from a clinical perspective, a median increase of 112 minutes of

participation in a bilateral upper limb intervention would be viewed as a positive improvement in terms of therapy delivered for children with limited hand function.

Table 35 – An analysis of OGS usage, comparing overall OGS usage per 'Child' and 'Guest' profile, as a function of group allocation and Games Catalogue

System Usage (mins)	***Child***	***Guest***
Overall		
Average usage (n=18)	377 ± 267	145 ± 142
Group (A or B)		
A: Average OGS usage (n=10)	403 ± 340	131 ± 122
B: Average OGS usage (n=8)	340 ± 143	161 ± 171
Catalogue (Original or Incentivised)		
Original: Average OGS usage (n=9)	267 ± 133	115 ± 174
Incentivised: Average OGS usage (n=9)	488 ± 326	174 ± 103

Notes: mins = minutes.

Comparing the mean and standard deviation of the usage for both Catalogues, the comparatively much smaller standard deviation associated with the Original Catalogue indicates that this group all engaged with the Catalogue to a similar level. The Incentivised Catalogue was able to engage children for longer (indicated by the higher mean time), but not consistently across individuals (as indicated by the much larger standard deviation). A box and whisker plot of Games Catalogue usage is shown in Figure 6-6, with the outlier (o^1) for 'NewCat' (the Incentivised Catalogue) being subject #38, who used the OGS the most – 1140 minutes, or more than 27 minutes per day.

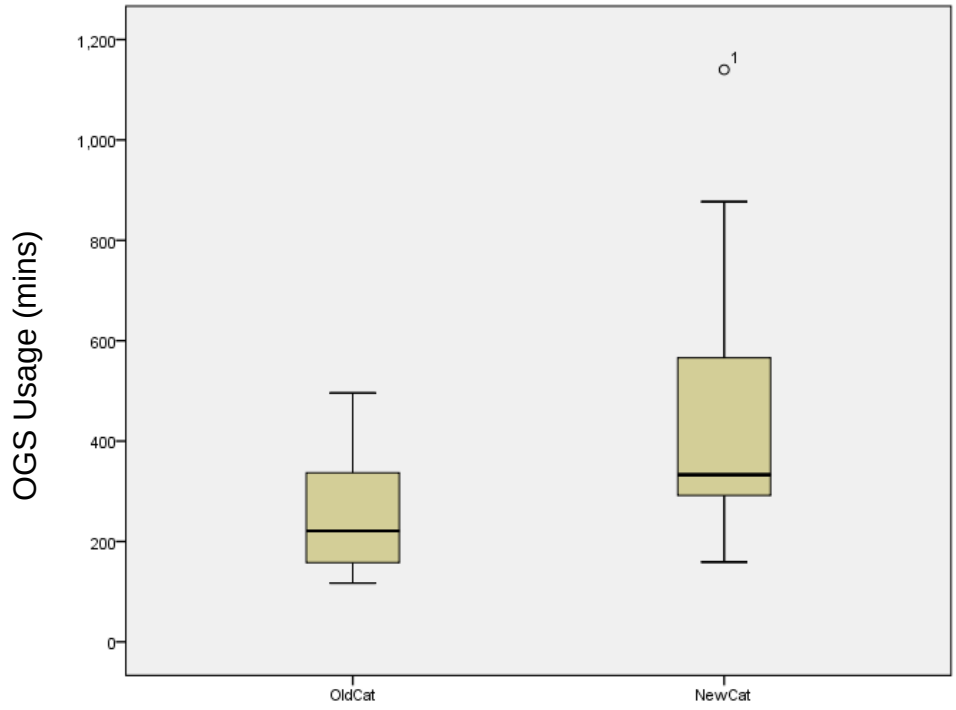


Figure 6-6 – Box and whisker plot of OGS usage, comparing the Original Games Catalogue ('OldCat') to the Incentivised Games Catalogue ('NewCat')

In terms of Guest OGS usage, there were no statistically significant differences when comparing Guest usage between groups (p= 0.467) or Games Catalogues (p=0.331).

6.3.9.1 Investigating Possible OGS Usage Differences due to Sex

The Stage 2 cohort was one third female (six females within a cohort of 18 children), and all six females completed the trial as both withdrawals were male. Most females were randomised to Group B and most received the original Games Catalogue, as shown in Table 36. Males typically used the OGS more than females (409 ± 315 mins compared to 314 ± 130 mins), but their variability in usage was much greater. Despite males filling four of the top five places when all children are ranked in terms of overall OGS usage (see Table 28), usage with respect to sex is not statistically significantly different (p=0.113). Similarly, females consistently rated the OGS higher (8.3 ± 1.9 compared to 6.8 ± 1.8), with all three 'ten out of ten' scores provided by females, but this result is also not statistically significant (p=0.960). A box-plot of

OGS is shown in Figure 6-7. Overall ratings for the OGS are presented in section 6.3.13.1.

Table 36 – Comparison of male and female OGS usage, rating, group allocation and Games Catalogue

	Female (n=6)	***Male (n=12)***
OGS Usage		
Average usage (mins) (n=18)	314 ± 130	409 ± 315
System Rating		
Average rating (/10)	8.3 ± 1.9 (n=6)	6.8 ± 1.8 (n=11)
Range (min – max)	6 – 10	4 – 9
Group Allocation		
A (n=10)	2	8
B (n=8)	4	4
Games Catalogue		
Original (n=9)	4	5
Incentivised (n=9)	2	7

Notes: min = minimum; max = maximum; mins = minutes.

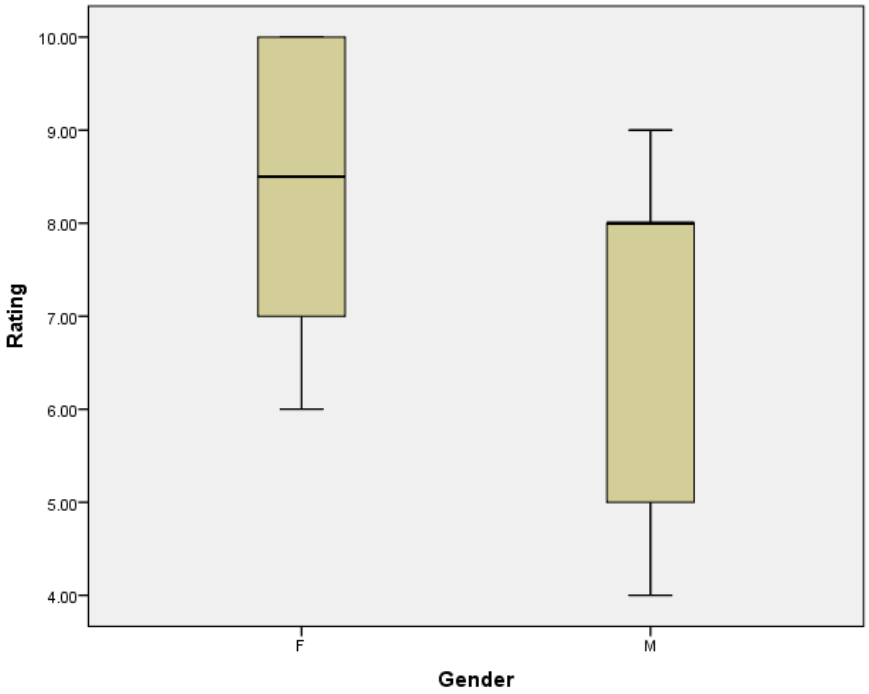


Figure 6-7 – Comparison of OGS ratings per child sex (F = female (n=6), M = male (n=11))

6.3.10 CP QOL Questionnaire Responses

With respect to the CP QOL questionnaire, 16 children completed both a pre- and post-trial form allowing a comparison to be made (participants #9 and #31 did not complete and return a post-trial form). As mentioned earlier (Chapter 3, section 3.4.1), self-reporting was based on the age of the child, and regardless of the child's age a parent always completed one of the questionnaires. Consequently, eight parents completed the '*CP QOL-Child Primary Caregiver Questionnaire (4-12 years)*' for their children (participants #8, #11, #21, #29, #32, #38, #41 and #47), three parents and children both completed the respective '*CP QOL-Child Primary Caregiver and Child Report Questionnaire (9-12 years)*' (participants #4, #27 and #46), and five parents and teenagers both completed the respective '*CP QOL-Teen Primary Caregiver and Adolescent Self Report Questionnaire*' (participants #6, #7, #12, #37 and #39). Child #29 was 9.5 years old when they attended their A_1 assessment, so old enough to self-report, but was not given a questionnaire to complete, which was an oversight. Data entry, cleaning and scoring was as per each questionnaire manual, respectively (Davis E *et al.*, 2013; Waters E *et al.*, 2013).

6.3.10.1 Administering the CP QOL Questionnaire

From an analysis perspective, on a few occasions an individual question was left blank or skipped, but on one occasion (for subject #39) two large groups of questions (11 in the first instance and 16 in the second – 27 questions in total) were skipped and not answered. This was presumably because the pages of the questionnaire were stuck together, and the parent/caregiver accidentally turned over two pages instead of one. This situation was rare and only occurred to this degree once. Where a particular question was skipped or overlooked on either the pre- or post-trial QOL form, the corresponding question, if it was answered, was not included in the analysis to ensure only 'like' questions were being compared at the two time points.

From a utility perspective, a few issues arose during the course of this study with respect to the questionnaire. Firstly, while the questions appear in sections (such as '*Family and Friends*' and '*Health*') none of the questions were numbered, making document use and cross-referencing difficult and laborious. Secondly, in the '*Primary*

Caregiver Questionnaire (4-12 years)' document, the 'Communication' section on page 7 has three questions, but the second question (question 23) has both a typo (it begins with the word '*they*' instead of '*the*', as does question 24) and doesn't have a coloured number scale with which to rate the question, as shown in Figure 6-8.

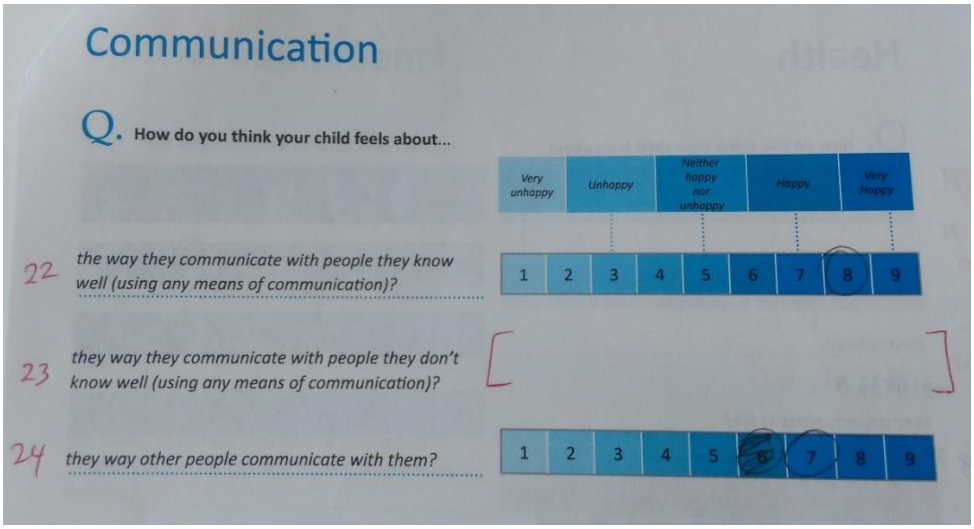
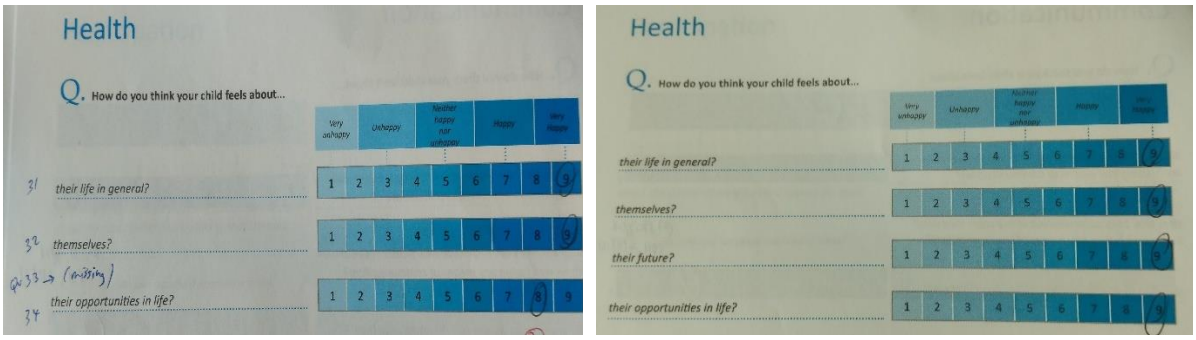


Figure 6-8 – The 'Communication' section of the CP QOL '*Primary Caregiver Questionnaire (4-12 years)*' document for question 23, with the coloured number scale missing (author annotations in red pen)

Consequently, only one parent out of 12 scored this question as intended on both the pre- and post-trial questionnaire for their child. For the analysis, this question was omitted if it wasn't answered correctly on both the pre- and post-trial questionnaires, meaning it was omitted in most circumstances.

During the course of this study, all the CP QOL questionnaires were updated to version 2 (in July 2013), meaning that some families completed version 1 at visit A_1 and then version 2 at A_2. Interestingly, the omission noted in Figure 6-8 was not detected and remains in version 2 of the CP QOL, but an additional question was added to the 'Health' section (page 8, question 33) of the '*Primary Caregiver Questionnaire (4-12 years)*' document. This question asks parents to rate how they think their child feels about '*their future*'. To enable a comparison between the pre- and post-trial scores, and to ensure questions accurately mapped across forms, the pre-trial form was adjusted and offset to cater for the new question, as shown in Figure 6-9 (a) and (b).



(a) **(b)**

Figure 6-9 – The 'Health' section of the '*Primary Caregiver Questionnaire (4-12 years)*' document, showing: (a) Version 1 (original), and (b) Version 2 (latest version), of the document, with the new question about '*their future*' added to version 2

The update to version 2 of the documents also introduced a 17th page and five additional questions (questions 86-90) to the '*Quality of Life Questionnaire for Adolescent (CP QOL-Teen), Primary Caregiver Questionnaire*' document, which focused on the happiness, health and financial situation of the parent/caregiver. Again, due to form differences, these particular questions couldn't be compared across time points if the earlier version of the questionnaire was given to parents/caregivers at the A_1 visit, and the updated version was given for the A_2 visit.

The results for the respective questionnaire responses appear in the following sections and tables.

6.3.10.2 CP QOL-Child Primary Caregiver and Child Report Questionnaire Results

Table 37 shows the '*CP QOL-Child Primary Caregiver Questionnaire (4-12 years)*' parent only responses (n=8) and Table 38 shows the '*CP QOL-Child Primary Caregiver and Child Report Questionnaire (9-12 years)*' parent and child responses for five of the questionnaire domains (n=3). The maximum score for each domain is 100 points, meaning the higher the score, the higher the QOL. This holds true for four of the five domain areas – '*Social wellbeing & acceptance*', '*Feelings about functioning*', '*Participation & physical health*', and '*Emotional wellbeing & self-esteem*', but not the fifth domain '*Pain and impact of disability*', where a lower score equates to a higher QOL. To improve interpretation and understanding of the

following three CP QOL tables, green numbers represent an increase or positive change, and red numbers represent a decrease or negative change, with respect to QOL post- and pre-trial ratings. As per the reporting guidelines for this particular tool, an overall or single QOL score is not calculated and individual domain scores are reported separately as each domain reports on a substantially different area of life (Swift, 2017).

Table 37 – Parent only responses for the five domain areas of the '*CP QOL-Child Primary Caregiver Questionnaire (4-12 years)*' (n=8)

Subject No.	*Report*	*Time Point*	*Social wellbeing & acceptance*	*Feelings about function*	*Participation & physical health*	*Emotional wellbeing & self-esteem*	*Pain & impact of disability*
8	Parent	Pre	80.2	67.0	69.3	80.0	31.3
		Post	80.2	72.7	67.0	77.5	45.3
		Change	0.0	5.7	-2.3	-2.5	14.1
11	Parent	Pre	95.8	93.2	90.9	100.0	4.7
		Post	81.3	75.0	72.7	85.0	4.7
		Change	-14.6	-18.2	-18.2	-15.0	0.0
21	Parent	Pre	77.1	83.0	65.9	77.1	18.8
		Post	75.0	88.9	75.0	89.6	18.8
		Change	-2.1	5.9	9.1	12.5	0.0
29	Parent	Pre	93.8	85.2	87.5	97.5	0.0
		Post	94.8	73.9	81.8	92.5	0.0
		Change	1.0	-11.3	-5.7	-5.0	0.0
32	Parent	Pre	100	88.6	89.8	97.5	12.5
		Post	97.9	90.9	93.2	100	40.6
		Change	-2.1	2.3	3.4	2.5	28.1
38	Parent	Pre	99	94.3	96.6	97.5	34.4
		Post	99	94.3	87.5	100	23.4
		Change	0.0	0.0	-9.1	2.5	-11.0
41	Parent	Pre	81.8	81.3	84.1	90	7.8
		Post	80.7	78.1	86.4	90	9.4
		Change	-1.1	-3.2	2.3	0	1.6
47	Parent	Pre	87.5	83	83	90	29.7
		Post	93.8	79.5	94.3	97.5	20.3
		Change	6.3	-3.5	11.3	7.5	-9.4

Notes: No. = number; Report = person completing the questionnaire; Pre = pre-trial; Post = post-trial; Change = (Post score – Pre score); **green** numbers = increase/positive change in QOL score (Post – Pre); **red** numbers = decrease/negative change in QOL score (Post – Pre).

As shown in Table 37, across the five domains parents rated four children as having decreased QOL scores and three children as having increased QOL scores in at least three domains, post-trial compared to pre-trial. The eighth child (#38) recorded increases in two domains, no change in two domains, and a decrease in one domain. The domains '*Social wellbeing & acceptance*', '*Feelings about function*', and '*Participation & physical health*' recorded the most instances of a decreased rating, indicating a lower quality of life rating was reported post-trial compared to pre-trial, for four children each (range: -1.1 to -14.6, -3.2 to -18.2, and -2.3 to -18.2,

respectively). Conversely, the domain '*Participation & physical health*' as well as '*Emotional wellbeing & self-esteem*' recorded the most instances of an increased rating, indicating a higher QOL rating was reported post-trial compared to pre-trial, for four children each (range: 2.3 to 11.3 and 2.5 to 12.5, respectively). For the fifth domain, '*Pain & impact of disability*', parents reported QOL score increases for three children (range: 1.6 to 28.1), no change for three children, and decreases for two children (range: -9.4 to -11), post-trial. However, an increase or decrease in QOL score for the first four domains did not correlate with a decrease or increase in the '*Pain and impact of disability*' domain. As an example, child #32 showed slight improvements in three domain areas, but recorded the most negative change in the '*Pain and impact of disability*' domain. Similarly, child #38 showed the largest positive change in their '*Pain and impact of disability*' domain, but also recorded a negative change for their '*Participation & physical health*' domain.

Table 38 shows the parent and child QOL scores for the three children who were younger than 12 years of age and able to self-report. A common theme was for all three children to rate their own QOL per domain higher than their parents for the first four domain areas, indicating a difference in perception between their opinion of their own life with CP and that of their parent. This is most evident in the post-trial ratings, but holds true for the pre-trial ratings as well. The results for the '*Pain & impact of disability*' domain were mixed, with parents and children disagreeing on all levels of this domain, including pre-trial, post-trial, and any change between ratings. If the individual domains are studied, all three children and one of the three parents reported higher levels of QOL post-trial, with only increases or increases in three or more domains recorded. One parent reported deceased values across all five domains, whereas their child (#27) reported increases for four of the five domains.

Table 38 – Parent and child responses for the five domains of the '*CP QOL-Child Primary Caregiver Questionnaire (4-12 years)*' (n=3)

Subject No.	*Report*	*Time Point*	*Social wellbeing & acceptance*	*Feelings about function*	*Participation & physical health*	*Emotional wellbeing & self-esteem*	*Pain & impact of disability*
4	Parent	Pre	83.3	85.2	77.3	97.5	29.7
		Post	92.7	87.5	90.9	100	12.5
		Change	9.4	2.3	13.6	2.5	-17.2
	Child	Pre	100	93.8	100	100	6.3
		Post	100	100	100	100	0
		Change	0	6.2	0	0	-6.3
27	Parent	Pre	71.9	70.5	64.8	72.5	20.3
		Post	68.8	64.8	51.1	67.5	21.9
		Change	-3.1	-5.7	-13.7	-5	1.6
	Child	Pre	83.3	68.8	75	70.8	70.3
		Post	93.8	90.6	100	95.8	75
		Change	10.5	21.8	25	25	4.7
46	Parent	Pre	80.2	77.3	64.8	77.1	29.7
		Post	79.2	77.3	77.3	79.2	46.4
		Change	-1	0	12.5	2.1	16.7
	Child	Pre	70.8	64.6	72.7	100	33.3
		Post	68.8	79.2	81.8	100	10.4
		Change	-2	14.6	9.1	0	-22.9

Notes: No. = number; Report = person who completed the questionnaire; Pre = pre-trial; Post = post-trial; Change = (Post score – Pre score); **green** numbers = increase/positive change in QOL score (Post – Pre); **red** numbers = decrease/negative change in QOL score (Post – Pre).

6.3.10.3 CP QOL-Teen Primary Caregiver and Adolescent Self Report Questionnaire Response Results

Table 39 shows the parent and teen QOL life scores for five children who were adolescents and able to self-report. The adolescent version of the CP QOL questionnaire differs from the child CP QOL version, with all five domains scored positively (due to a reverse coding process), meaning that across all five domains an increase in score represents an increase in QOL.

Similar to the younger group who self-reported, all five teenagers consistently rated their own QOL higher than their parent's ratings across all domains. Three parents and four teenagers reported higher levels of QOL post-trial in three or more domain areas, and all five domain areas recorded increases in QOL from five or more reports. The domain that received the most number of positive change reports was

‘*Communication & physical health*’ (nine increased scores post-trial from 10 reports, range: 0.8 to 16.4), and the domain that received the least number of positive change reports was ‘*Social wellbeing*’ (five increases, three no change, and two decreases).

Table 39 – Parent and teen responses for the five domains of the '*CP QOL-Teen Primary Caregiver and Adolescent Self Report Questionnaire*' (n=5)

Subject No.	*Report*	*Time Point*	*General wellbeing & participation*	*Communication & physical health*	*School wellbeing*	*Social wellbeing*	*Feelings about functioning*
6	Parent	Pre	45.2	50	51.6	76.8	45
		Post	47.6	46.9	53.1	89.3	37.5
		Change	2.4	-3.1	1.5	12.5	-7.5
	Teen	Pre	64.9	68	58.9	71.4	75
		Post	60.7	71.1	62.5	76.8	50
		Change	-4.2	3.1	3.6	5.4	-25
7	Parent	Pre	33.9	37.5	48.4	64.3	32.5
		Post	34.5	40.6	45.3	58.9	42.5
		Change	0.6	3.1	-3.1	-5.4	10
	Teen	Pre	53.6	69.6	85.9	87.5	60
		Post	58.9	76.8	82.8	87.5	70
		Change	5.3	7.2	-3.1	0	10
12	Parent	Pre	60.6	67.2	65.6	80.4	52.5
		Post	59.4	68	60.9	73.2	75
		Change	-1.2	0.8	-4.7	-7.2	22.5
	Teen	Pre	66.1	63.3	50	73.2	75
		Post	66.7	67.2	67.2	85.7	62.5
		Change	0.6	3.9	17.2	12.5	-12.5
37	Parent	Pre	61.3	60.9	57.8	80.4	65
		Post	77.4	77.3	78.1	89.3	77.5
		Change	16.1	16.4	20.3	8.9	12.5
	Teen	Pre	92.3	89.8	82.1	92.9	92.5
		Post	93.5	93	91.1	92.9	97.5
		Change	1.2	3.2	9	0	5
39	Parent	Pre	75	79.5	70.8	82.1	77.5
		Post	79.4	84.1	95.8	89.3	77.5
		Change	4.4	4.6	25	7.2	0
	Teen	Pre	98.2	98.4	100	100	82.5
		Post	97.6	100	98.4	100	92.5
		Change	-0.6	1.6	-1.6	0	10

Notes: Report = person who completed the questionnaire; Pre = pre-trial; Post = post-trial; Change = (Post score – Pre score); **green** numbers = increase/positive change in QOL score (Post – Pre); **red** numbers = decrease/negative change in QOL score (Post – Pre).

6.3.10.4 Specific CP QOL Questions Related to the Upper Limbs

Each CP QOL questionnaire contained specific questions relating to the child's use of their hands and arms, and certain daily activities that relate to the upper limbs. Each questionnaire also asked how happy the child was (Child form), or if they were concerned about their CP (Teen form). Table 40 highlights the specific questions and the section within each questionnaire from which each question comes.

Table 40 – Specific CP QOL questions that relate to hand or arm use and happiness

Questionnaire Form	***Question No.***	***Questionnaire Section***	***Question relates to (*)***
CP QOL-Child	35	Health	The way they use their arms
	37	Health	The way they use their hands
	38	Health	Their ability to dress themselves
	39	Health	Their ability to drink independently
	52	Final Questions	How happy is your child?
CP QOL-Teen	56	Health	The way they use their arms and hands
	58	Health	Their ability to dress themselves
	59	Health	Their ability to eat or drink independently
	55	Health	Is your teenager concerned about having cerebral palsy?

Notes: (*) questions written above from the parent's perspective, but for the child report form the questions are written in the first person for the child, e.g. "the way you use your arms", etc; No. = number.

With respect to the questions listed in Table 40, Table 41 shows the comparative change in QOL ratings that were recorded post-trial per question, as rated by the parent and trial participant (child or teenager). The CP QOL-Child form was completed by 11 parents and three children, and the CP QOL-Teen form was completed by five parents and five teenagers. Table 41 reports changes as either a rating increase, rating decrease, or no change, with the 'no change' report including situations where the parent and/or participant had already rated a particular question the maximum possible value on both occasions.

Table 41 – How parents and children/teenagers responded to specific CP QOL questions about hand or arm use and happiness

CP QOL Form	*Question No.*	*Reporter*	*n*	*Rating Increase*	*Rating Decrease*	*No Change*
Child	35	Parent	11	6	2	3
		Child	3	-	-	3
	37	Parent		5	3	3
		Child		1	-	2
	38	Parent		2	5	4
		Child		-	-	3
	39	Parent		1	1	8 (*)
		Child		1	-	2
	52	Parent		1	1	9
		Child		-	-	3
Teen	56	Parent	5	3	1	1
		Teen	5	1	1	3
	58	Parent		2	-	3
		Teen		1	1	2 (*)
	59	Parent		2	-	3
		Teen		1	1	2 (*)
	55	Parent		3	-	2
		Teen		3	1	1

Notes: (*) indicates one reporter skipped this question in each case.

For the younger cohort, the largest increase in QOL ratings were for questions 35 and 37 relating to arm and hand use. However, this positive increase did not translate through to increased ratings for daily activities that involve the hands and arms, such as question 38 (which received the most number of decreased ratings) and question 39 (where ‘no change’ was recorded the most).

For the older cohort, parents tended to rate their teenager’s use of their arms and hands higher post-trial compared to pre-trial (question 56), with some translation through to their rating of activities of daily living (questions 58 and 59), evidenced by the frequency of increased or ‘no change’ ratings and no decreased scores. Teenagers recorded responses across all three categories, with a slightly higher preference for ‘no change’ (questions 56, 58 and 59). Parents and teenagers both indicated they were less concerned about CP (question 55) post-trial compared to pre-trial, with this particular question receiving six instances of an increased rating, three ‘no change’ ratings, and one decreased rating. From an overall cohort

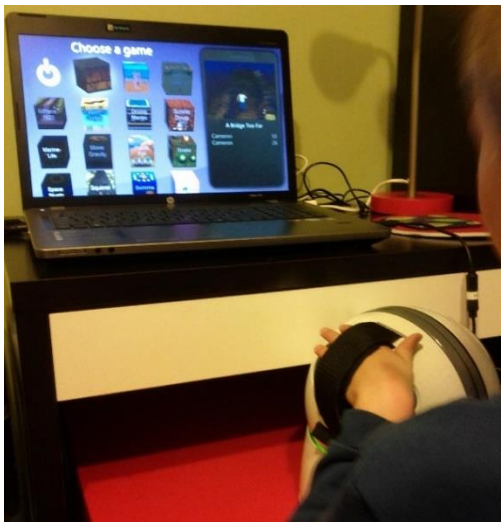
perspective (n=16), only one parent and one teenager reported being less happy or having more concerns about CP post-trial compared to pre-trial.

6.3.11 Children Who Required a ND Hand Strap During the Trial

Only two children required a strap to be fitted to their 'Orby' controller to support their ND hand during the trial. The two children were:

- Child #4 – MACS Level III, aged 9 years 1 month, left ND side, as shown in Figure 6-10(a); and
- Child #47 – MACS Level III, aged 8 years 9 months, right ND side, as shown in Figure 6-10(b).

It was obvious from the orientation and set up session for child #4 that this particular child may struggle without a strap, but the author and parents agreed that it was worth allowing the child to persist without one to begin with. However, the next day the family called and asked for a strap for their child, so the author returned to the family home and fitted a strap (see Figure 6-10(a)) on Day 2. For child #47, the mother requested a strap after a week and a half of her daughter using the OGS. A strap was posted to the mother and fitted accordingly on Day 14, and the child continued the study with a strap as shown in Figure 6-10(b).



(a) **(b)**

Figure 6-10 – Photos of the two children who required a strap to use the OGS during the trial (Source: (b) child's mother)

6.3.12 Trial Issues and Equipment Problems

A number of technical issues did arose from the trial, as detailed in Table 42. Most issues were hardware related and relatively straightforward to address, with the most serious issue being the snapping of the pin within the 'Orby' controller that connected the spherical 'orb' to the controller base, via the joystick mount (issue #2 in Table 42). This is examined in more detail in Appendix N. From a trial integrity perspective, the software issue (issue #4 in Table 42) that resulted when someone deliberately tampered with the OGS laptop meant that one child experienced both haptic and non-haptic gaming, which is explained in more detail in Appendix N.

Each entry in Table 42 identifies the trial issue or problem, the effect this had on the trial, how often the issue arose, the reason for the issue, how it was resolved, and the result of the actions that were implemented.

Table 42 – List of trial issues or equipment breakages, the effect these had on the trial, the reasons they occurred, and how each issue was resolved

Equipment Problem / Software Issue	***Effect on Trial / Child and Frequency of Occurrence***	***Reason for Fault / Issue, Resolution, and Result of Resolution***
1. 'Orby' controller faulty and not working as expected – either left/right or up/down movements not working	*Effect:* Controller was unsuitable and unusable for trial, leading to the orientation and set-up session being abandoned and rescheduled. Luckily, the issue was discovered during early OGS orientation/set-up and not during the trial proper. Hence, the trial start date was delayed by three days and six days, respectively, for the children affected. *Frequency*: Twice, the fourth and fifth OGS deployments.	*Reason:* Loose connection between joystick controls and *Xbox* PCB. *Resolution:* Author implemented a more rigorous controller assembly and functionality check prior to future OGS orientation and set-up sessions. Author also took a back-up 'Orby' controller to subsequent set-ups in case a fault was found at the child's home during set-up. *Result:* No further occurrences once a more rigorous controller assembly and functionality check was established pre-deployment.

2. ‘Orby’ controller breakages (1)	*Effect:* Spherical ‘orb’ part of the controller snapped off and detached from mounting, falling into the shrouded base section, rendering the OGS unusable. Child’s trial paused on each occasion. *Frequency:* Four times, for three children. Further details appear in Appendix N.	*Reason:* Examination of the broken controllers identified that they all broke in an identical fashion, with identical failure mechanisms observed (Figure 6-11(b), (d)). The component that broke was the 3D printed pin (Figure 6-11(a)), which connects the base of the controller, via the ruggedised joystick mount, to the top of the controller, via the vibration mount section. The pin was breaking at the abrupt change in pin shape geometry (purple arrow in Figure 6-11(a)), due to a high stress concentration at the transition region. Appendix N contains further details. *Resolution:* Author replaced 3D printed pins with machined aluminium pins (see Figure 6-12). *Result:* No further breakages once pin material changed.
3. ‘Orby’ controller breakages (2)	*Effect:* One side of an ‘Orby’ controller came away from the grey centrepiece, introducing a degree of ‘play’ into the top of the controller, but not affecting the function of the controller overall. Trial unaffected and the OGS still functioned as expected. *Frequency:* Once.	*Reason:* Upon inspection, it was noted that the glue holding three of the four magnets had become unstuck from the centrepiece. *Resolution:* Magnets were re-glued. *Result:* No further faults or ‘play’ in the controller.
4. Software issue	*Effect:* Child’s name no longer appeared on the main log-in screen. Mother called the author to notify that the OGS looked ‘different’. *Frequency:* One child	*Reason:* OGS laptop and child’s profile was tampered with. More details are provided in Appendix N. *Resolution:* No intervention, trial continued as per family wishes. *Result:* Child was randomised to Group B (non-haptic), but the tampering caused a profile flip, meaning the child received haptic feedback for part of the trial.

Notes: OGS = OrbIT Gaming System; PCB = printed circuit board; 3D = three dimensional.

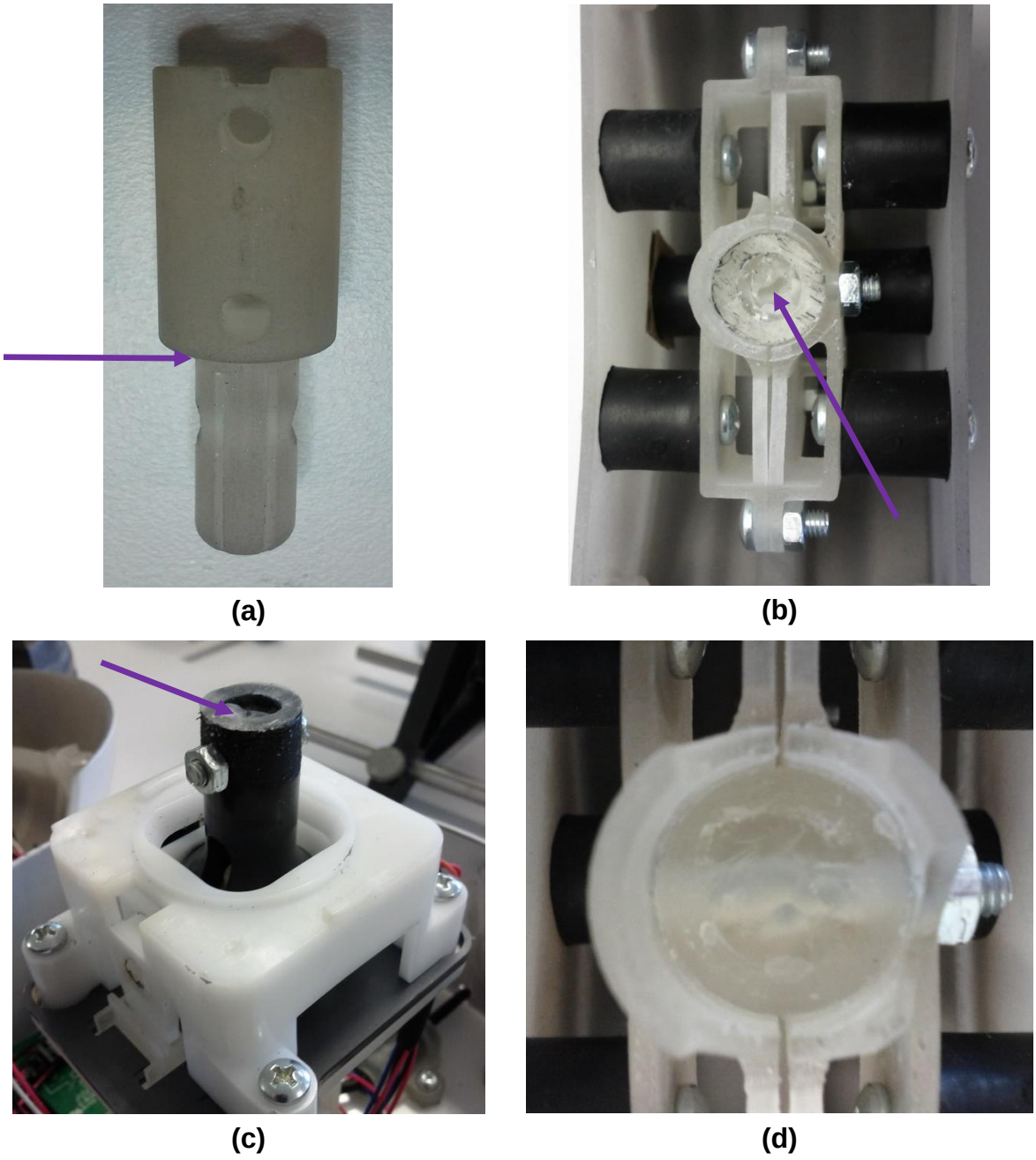


Figure 6-11 – Examination of the broken ‘Orby’ controllers: (a) The 3D printed pin, shown in its native orientation when mounted inside the controller. The purple arrow highlights the abrupt change in pin shape, leading to high stress concentrations in this area and a weakness in the design; (b) Looking into the top of the ‘Orby’ controller with the top of the broken pin still in place (purple arrow); (c) The ruggedised joystick mount within the controller, with the base of the broken pin still in the black joystick mount (purple arrow); and (d) A close up of the top of the pin from Figure 6-11(b), snapped off and stuck in the top of the ‘Orby’ controller

Figure 6-12 – The new machined aluminium pins (left and right), either side of a 3D printed pin (middle)

6.3.13 Results from the 'Participant Experience Questionnaire'

Seventeen of the 18 (94%) children completed and returned the '*Participant Experience Questionnaire*' form at the end of the RCT. The family that did not return their form was from regional South Australia. This child was randomised to Group A and received the Incentivised Games Catalogue. The author contacted the family about the form post-trial, and sent a reminder, but the evaluation was not returned.

6.3.13.1 OGS Ratings

Question 1 on the '*Participant Experience Questionnaire*' form asked families and children to rate how much they enjoyed playing the computer games on a linear scale out of 10. The average OGS rating was 7.4 ± 1.9 out of 10 (median rating = 8.0, n=17). OGS rating as a function of Group allocation and Games Catalogue ('Old' = Original Catalogue and 'New' = Incentivised Catalogue) is shown in Figure 6-13, with the average ratings and n values for each group appearing within each coloured bar.

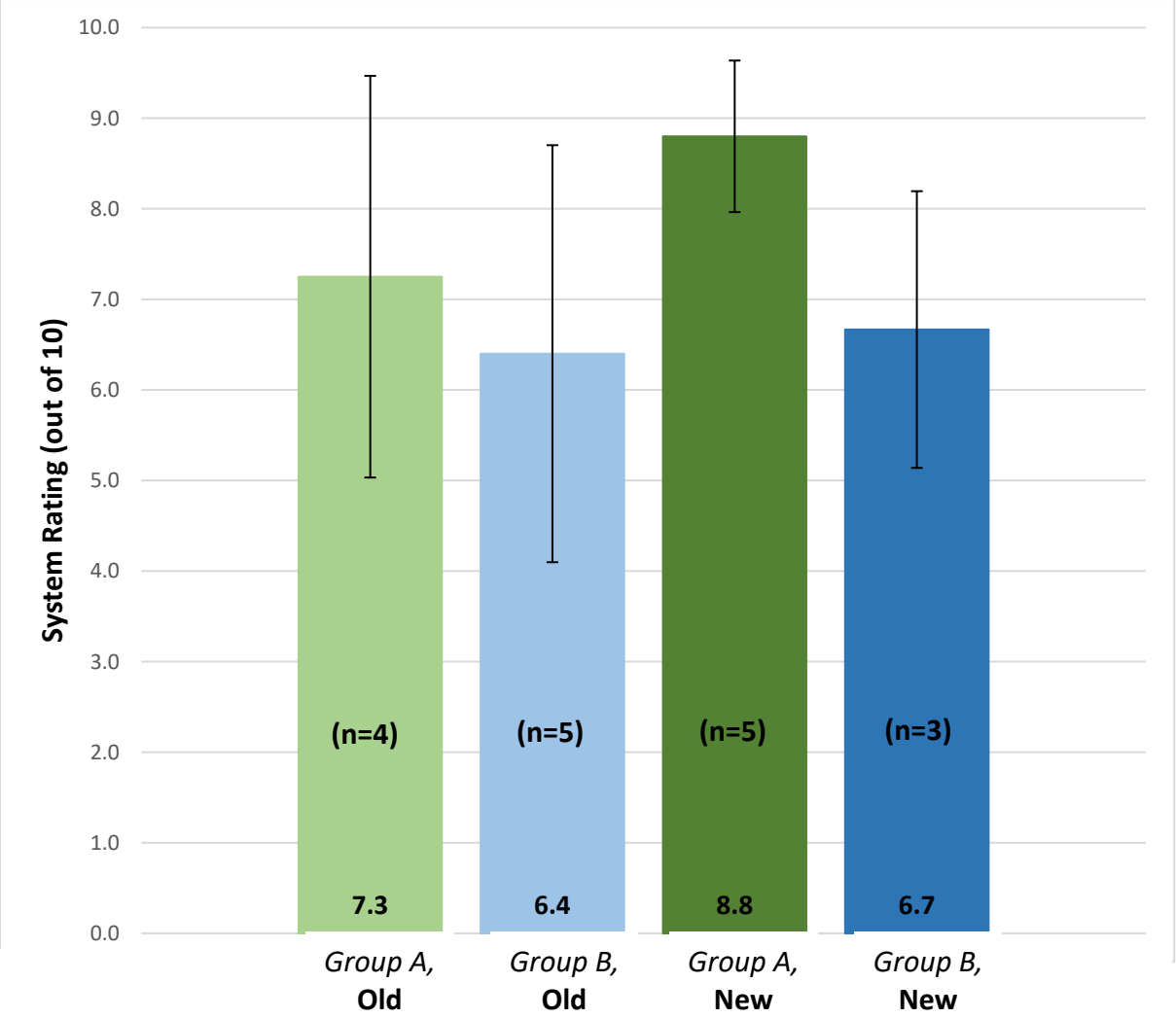


Figure 6-13 – OGS rating as a function of group allocation (Group A or B) and Games Catalogue deployed (Old = Original, New = Incentivised)

There was no statistically significant difference between the OGS rating and Group allocation (8.1 ± 1.7 for Group A compared to 6.5 ± 1.9 for Group B, p=0.617) or between the OGS rating and Games Catalogue received (6.8 ± 2.2 for the Old (Original) Catalogue compared to 8.0 ± 1.5 for the New (Incentivised) Catalogue, p=0.176).

While the numbers were small and none of the ratings per sub-group in Figure 6-13 were statistically significantly different from each other, the greatest difference between ratings occurred between children allocated to Group A who received the Incentivised Games Catalogue (8.8 ± 0.8) and children allocated to Group B who received the Original Games Catalogue (6.4 ± 2.3), where p = 0.135.

6.3.13.2 Reports of Positive or Negative Occurrences During the Trial

The '*Participant Experience Questionnaire*' form provided families and children with an opportunity to report both positive and negative occurrences during the trial period. Positive comments or occurrences that were reported for the child during the RCT include:

- Being able to use both hands quite comfortably, without any issues;
- Learning to share the OGS with siblings and to agree to time limits on 'Orby' if chores were completed and siblings were playing nicely;
- Improved confidence to use a computer on their own and enjoyment with the selection of games;
- Sharing the OGS with able-bodied siblings;
- Improved relationships between siblings;
- The OGS being a tool that reinforced that 'practice makes perfect' as the child was required to dedicate time to using 'Orby' in order to unlock more games (the parent drew an analogy with homework, where dedicated time and practice improves homework);
- Finding the OGS easier to use than any other game system;
- Making the child continually make an effort to open up their normally clenched hand to control the OGS, and helping the child to develop his language skills as he was giving instructions to family and friends about how to use the OGS. This family also started to use the OGS as 'homework' for their child, to help him increase his attention span;
- Enjoying trying to beat an older (able-bodied) brother's score;
- Enjoying 'testing' the OGS to see if it would notice if a hand wasn't placed on the controller correctly, and how long it would take the OGS to notice;
- Liking how accurate the controls were *[author note: this comment relates to the sensitivity and responsiveness of the controller]*;
- Being a positive presence in the family home that created a discussion about CP. The child was proud to tell visitors that the OGS "*was a machine specifically designed to assist those with a disability*", leading to many positive conversations about CP that the child was happy to have; and
- Being able to learn new games.

Negative comments or occurrences that were reported for a child during the RCT include:

- Difficulty keeping the ND hand in place on the controller;
- Requiring an adjustment period to establish sharing arrangements and game time between siblings *[author note: this family used the OGS as a positive reinforcement tool for positive siblings relationships, and the child was only allowed to play on the OGS if they didn't fight or argue with their sibling]*;
- Being unwell, so not able to have dedicated time on the OGS, combined with controller breakages and difficulty using the controller;
- Reporting that on one day a particular game didn't work, but that it did the next day;
- Losing interest in the OGS once all the games were unlocked;
- Disappointment on a sibling's behalf when the child that the OGS was given to wasn't using it enough, so wasn't unlocking games they wanted to play;
- Becoming bored with the OGS the longer he had it, and not using it as often;
- Wanting to play commercial gaming systems, like *PlayStation 3*, which his older brothers and cousins were playing;
- Not liking that their controller broke, and would have liked dual player games instead of only single player games; and
- Sometimes the mass and momentum of releasing 'Orby' would cause a 'flick' that was recorded as a controller movement, which was not intentional.

6.4 Discussion

The aim of this thesis was to investigate an upper limb somatosensory intervention for children with CP with a known sensory loss. The hypothesis was that sensory function could be significantly improved through a six-week, home-based, haptic serious games RCT intervention using the OrbIT Gaming System (OGS). The OGS required children to use and engage both their hands on a customised controller in order to play the games that were on offer. The 'active' element for the RCT intervention was the use of haptic or vibration stimulation because vibration sense is the modality most preserved in children with CP (Uvebrant, 1988) and known to activate the primary and secondary somatosensory cortices (Coghill *et al.*, 1994). Using computer games to cognitively and actively engage children, coupled with deliberate and targeted active vibration stimulation delivered during OGS use, it was hypothesised that children randomised to treatment with active vibration feedback that was contextualised through game events (Group A) would have significantly better sensory and functional outcomes post-trial compared to children receiving no active vibration stimulation (Group B). From a tactile sensation perspective, the design of the 'Orby' controller (with textured grey oval pads and the 3D printed textured surface of the controller) meant that *all* children within the trial at least had a passive tactile experience when using the controller. Group A received the added element of an 'active' vibration stimuli, but simply using 'Orby' afforded a tactile experience, which was heightened when the child's hands moved over or across the surface of the controller.

The author observed that some children initially struggled to use the 'Orby' controller correctly during the set up/orientation with the OGS, as it required intentional and active use of their ND hand. Recent research that investigated parent's perceptions of how children with unilateral CP learn to master bimanual activities identified key themes such as "*awakening the inner drive*", "*trying on one's own*", and that "*it must be worth the effort*" (Lidman *et al.*, 2017). More importantly, an overall key theme of relevance was that "*finding harmony between pleasure and effort is the key to learning*" (Lidman *et al.*, 2017, pg. 6), which epitomised the approach that was taken with respect to the ND hand strap (sections 5.4.3.1.5 and 6.3.11) for the 'Orby' controller. In this light, the OGS can be viewed as an example of 'positive constraint' therapy, where the motivation to keep two hands actively on the controller at all

times facilitated the reward or goal, which is being able to play computer games, as opposed to classical constraint therapy, which involves preventing the use of the dominant hand. The OGS requires volitional and intentional bimanual effort on the child's behalf, which may cause frustration at times when the sensors are not covered, but rewards correct hand positioning and use. It is important to remember the bilateral use is not mirrored (i.e.: the left side doing the exact same motion as the right), but rather the movements are reciprocal due to the spherical shape and orbital motion of the OGS.

Alongside the above hypothesis, specific areas of investigation for the RCT included: (a) the feasibility of deploying the OGS into a family home in an unsupervised and 'child-led' format, as a way of engaging children with CP to use both their hands; (b) child and parent acceptance of the OGS; (c) OGS usability and engagement; and (d) the effectiveness of the OGS to improve hand function.

From an eligible pool of 25 children that were identified during Stage 1, 18 participated in the RCT, with 10 children (eight males) randomised to Group A and eight (four males) to Group B. This was a representative group given the profile of the Stage 1 cohort (Table 26), meaning the Stage 2 cohort was similarly mostly male (67%), with a higher proportion of children with unilateral CP (72%), but similar percentages of children with a ND left side (61%), and a predominant brain scan classification of cortical/sub-cortical lesions (39%). The Stage 2 RCT cohort was of a similar age to the Stage 1 cohort that they were recruited from (10.7 ± 3.4 compared to 10 ± 3.4 years), meaning a younger cohort from Stage 1 was recruited to the trial given the 18 month window between Stage 1 and Stage 2 (section 6.1.3).

6.4.1 Assessing the Somatosensory Assessment Results

Statistical modelling revealed no statistically significant differences between baseline (A_1) and post-trial (A_2 and A_3) sensory measures between the two Groups (A or B) for the dominant hand, and only one statistically significant result for the ND hand for test of stereognosis between the Groups at the immediate A_2 follow-up assessment. As reported earlier (section 6.3.7), this significant result was due to Group A (haptic group) children performing better in the stereognosis assessment than Group B

(non-haptic group) children, combined with the fact that the latter group performed worse in stereognosis during their A_2 assessment. However, this significant between group difference did not persist at the follow up A_3 assessment.

With respect to stereognosis, Petersen *et al.* (2016) defined a clinically significant change being correctly identifying two or more objects (pgs. 92-3), while noting that stereognosis had acceptable published interrater reliability and test-retest reliability (pg. 95). When investigating stereognosis performance for the whole cohort at an individual level for the ND hand, only four (#7, #12, # 27 and #41) of 16 children with complete data scored the same stereognosis score at all three visits, while eight children correctly identified ($x \pm 1$) object(s) (where x was their A_1 score). The remaining four children (#6, #8, #31 and #39) exhibited variable results, with only child #39 scoring a +2 differential improvement in stereognosis score between A_1 and A_3 (correctly identifying 4, 5, then 6 objects). Petersen *et al.* (2016) reports that it is not known how much time is required for stereognosis cortical re-education (pg. 95), noting that it is possible that there was insufficient time between assessments for their study, like that of the present, to enable a stereognosis improvement (the difference between assessments for Petersen *et al.* for most subjects was one year). There was a trend for both Group A and B children to record non-significant improvements for the test of proprioception and the JTHFT from A_1 through to A_3, but only Group A children also showed a trend towards improved stereognosis results.

In terms of systematic tactile training studies, Kuo *et al.* (2016) recently reported on an intensive 90-hour training camp study over three weeks in two different countries (Belgium and the United States of America) with 20 children with unilateral CP. The intervention involved '*Hand Arm Bimanual Intensive Therapy*' or *HABIT* with two groups: one group received *HABIT* and eight hours of specific tactile training through a structured and detailed program using materials of different shapes and textures *without* vision, while the other group received the same amount of additional training, but was exposed to the same materials through play and with full vision, and not through a structured or targeted program. The authors reported statistically significant improvements between pre- and immediate post-tests for tactile perception (using the Grating Orientation Task, $p = 0.028$) for both hands and both groups, but not for TPD or tactile registration (using SWM). Additionally, a trend

towards improved stereognosis was observed (p = 0.063) for both groups. A follow-up assessment was not conducted for this study. The authors concluded that tactile function could be enhanced in both hands of children with unilateral CP after an intensive HABIT intervention and additional tactile training or exposure, but given that both groups improved, the environment rather than a specific tactile training program could be the key aspect driving change.

More recently, and in direct response to the study by Kuo *et al.* (2016), Saussez, Van Laethem, and Bleyenheuft (2018) reported improvements in stereognosis for the ND hand for a group of 19 children with unilateral CP after an intensive program called '*Hand Arm Bimanual Intensive Therapy Including Lower Extremities*' (*HABIT-ILE*). Saussez *et al.* (2018) utilised a similar day-camp setting to deliver 90 hours of intensive therapy over two weeks (nine hours of therapy per day for 10 days), however, a key difference was that sensory stimulation via enriched materials was not employed as it was for Kuo *et al.* (2016). The authors reported a statistically significant improvement in stereognosis function between the initial assessment and the four month follow-up assessment (p = 0.015), but no improvement in tactile spatial discrimination (assessed using the Grating Orientation Task). The authors concluded the improvements could be due to *HABIT-ILE* driving improved motor performance (pg. 266), which is a substantial component of the stereognosis test.

As mentioned earlier (section 6.1.1), following their 2014 review, Auld *et al.* (2017) recently reported on a small pilot crossover trial with six mostly MACS I children with unilateral CP, where the cohort received a single 60-minute session of (1) mirror based tactile and motor training and (2) bimanual training. Four of the six children showed improved tactile perception (improvements were in either single or double simultaneous localization), but no improvement in tactile registration (SWM). Additionally, the authors reported that bimanual training did not improve tactile perception for the cohort. Auld *et al.* (2017) noted that a key aspect of mirror-based training is that the child fully attends to the visual display of their ND hand in the mirror for the exercises (pg. 7), and that this could potentially explain why older children showed the largest improvements, due to their ability to attend and concentrate for longer. Of particular note with respect to this study is the extremely low dose for an improvement in tactile perception – a single 60-minute session.

Transfer enhanced training was another approach that was recommended by Auld *et al.* (2014)'s systematic review, and formed the basis for a somatosensory intervention by McLean *et al.* (2017). The pilot matched-pairs study recruited 17 children with unilateral CP, of which seven were randomised to the intervention group. The intervention was the application of the adult '*Study of the Effectiveness of Neurorehabilitation on Sensation*' or *SENSe* trial, but for a paediatric population. Assessments included functional tactile object recognition and wrist position sense, with motor performance assessed using the AHA. The intervention group received 18 hours of *SENSe* training over a six week period. The authors reported statistically significant improvements in proprioception (wrist position sense, $p = 0.018$) and motor performance ($p = 0.028$) between baseline and at the six-month follow-up assessment, along with improvements in Goal Attainment Scaling (GAS) and COPM.

The recent studies presented in this section (Kuo *et al.*, 2016; Auld *et al.*, 2017; McLean *et al.*, 2017; Saussez *et al.*, 2018) demonstrate promising preliminary evidence for improvements in somatosensory function for children with unilateral CP, with all authors recommending further investigation with larger cohorts is required to confirm the findings.

6.4.2 Comparison with Previous Studies and Post-Study Sample Size Calculations

Compared to the literature, of the 16 CP serious games interventions highlighted by Bonnechère *et al.* (2016) in their review, this study ranks equal fifth in terms of the number of participants, along with the study by Ramstrand and Lygnegard (2012), which was a home-based trial of the *Nintendo Wii* investigating balance. According to the systematic review of 11 CP studies that used active video games for therapeutic purposes by Staiano and Flynn (2014), this study ranks equal first with Ramstrand and Lygnegard (2012) in terms of the number of participants.

Working with the consultant statistician for the project, a *post-hoc* sample size calculation was conducted. Using the test of tactile registration (SWM) for the thumb as a basis, calculations were based on detecting a 5% level of significance (alpha, two-sided) with 80% power (beta), a standard deviation (or variance, σ) of 0.56, and

mean values for m_1 and m_2 being 0.0 and 0.3, respectively, meaning delta is equal to 0.3. When calculated, this generated an estimated sample size of 55 children, or more practically, 56 children with 28 children in each group. This result is similar to the sample size calculation made by Preston *et al.* (2016), which estimated that 58 children were required for 80% power at the 5% level of significance (pg. 1007). Consequently, the RCT for this study recruited approximately one third of the number of children required to generate an appropriately powered result.

6.4.3 The Jebsen Taylor Hand Function Test

At the activity level, statistical modelling revealed no statistically significant difference between baseline (A_1) and post-trial (A_2 and A_3) sensory measures between the two Groups (A or B) for either hand. As a secondary exploratory analysis, and given that all children who participated in the trial were engaged in a forced bimanual integrated upper limb task, it was decided to then model the cohort as a single group. As reported in section 6.3.7, the subsequent re-modelling and analysis identified a strong statistically significant difference between baseline (A_1) and follow-up (A_3) for the total score for the JTHFT for the ND hand only ($p = 0.001$).

The study of 71 typically developing children aged six to 10 years and 11 months old by Reedman *et al.* (2015) published MCID for the ND hand as being 5.87 seconds. If this criterion is used, 10 children recorded a decrease in total JTHFT time for their ND hand that is greater than the MCID, as shown in Table 32. However, the current cohort includes six children aged over 10 years and 11 months, which is outside the age range for the group that Reedman *et al.* (2015) studied, meaning the results are not directly transferable. Additionally, a study by Eliasson *et al.* (2009) that investigated the use of the JTHFT with 16 children with only congenital unilateral CP (MACS Levels I and II) reported that this test showed greater variability than expected between two baseline pre-intervention assessments four months apart. Eliasson *et al.* (2009) reports a median decrease of five seconds in one of the groups (n=11) and 56 seconds in the other (n=5)(pg. 321). The follow-up assessment for their study was six months post-intervention.

The results of the current study can be compared to that of Eliasson *et al.* (2009) if the Stage 1 JTHFT data are used, considering the A_0 (Stage 1) and A_1 (Stage 2) assessment points as a dual baseline, but with a longer time between pre-intervention assessments compared to Eliasson *et al*. Figure 6-14 shows an extension of Figure 6-4, with A_0 JTHFT times included for 16 children with complete data. The results of the current mixed CP study, which shows a median decrease of 17 seconds from first baseline assessment to second (A_0 median = 204.5 seconds, A_1 median = 187.5 seconds), confirms the lack of stability for the JTHFT reported by Eliasson *et al.* (2009), but not to the same degree. The median decrease reported for the current study is closer to the larger of the two intervention groups reported by Eliasson *et al.* (2009) (five seconds compared to 56). Additionally, the current cohort has more severe hand function limitations, evidenced by the difference between median total times for the JTHFT (Eliasson *et al.*, 2009, pg. 321).

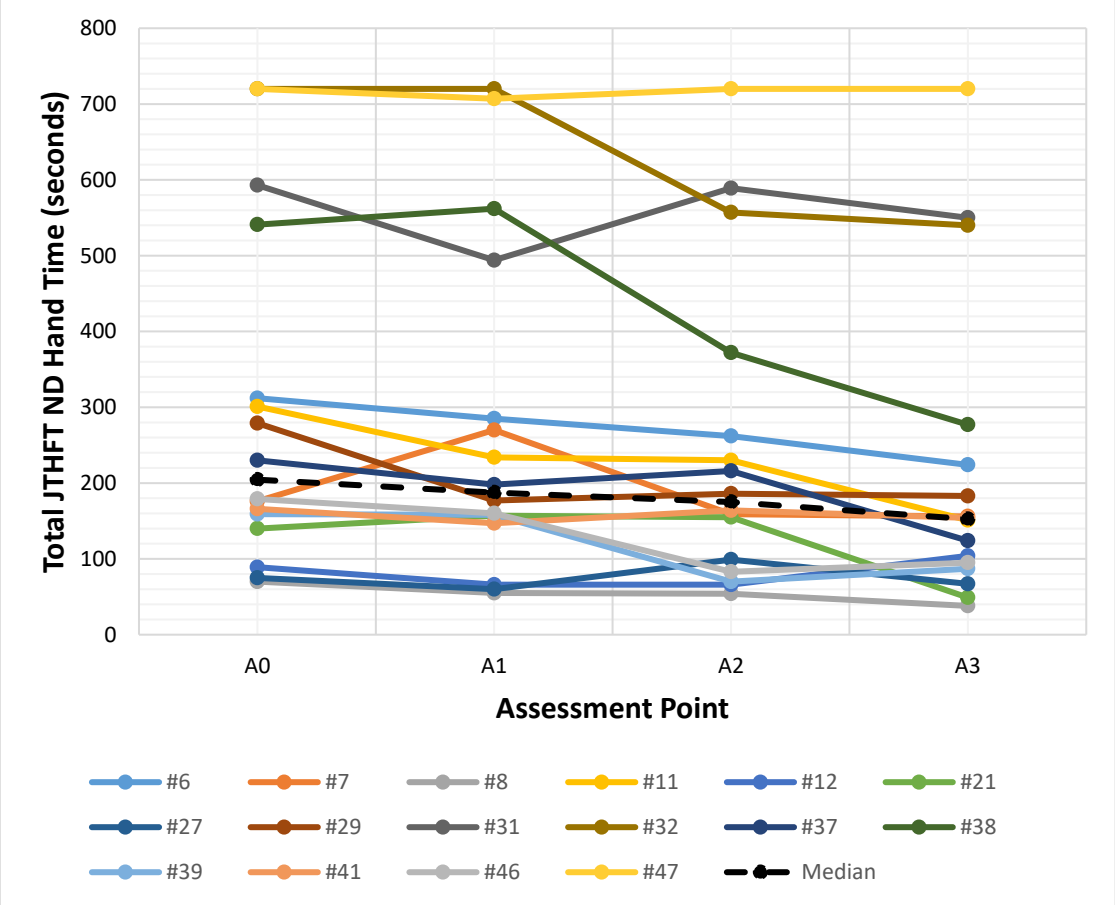


Figure 6-14 – A comparison of ND hand JTHFT times for children who participated in both Stage 1 and Stage 2 (n=16), with A_0 and A_1 representing a dual baseline for the overall study. The black dashed line represents the median results

6.4.4 The ABILHAND-Kids Questionnaire

As reported in section 6.3.8, 10 children recorded increases in ABILHAND-Kids scores, but only three children (#4, #21 and #37) achieved logit score increases that were greater than the author reported MCID for this tool, which was +0.71 logits (Bleyenheuft *et al.*, 2017). All three children were randomised to Group A and also collectively recorded positive changes across all five domain areas of the CP QOL questionnaire for 17 of 20 domain ratings (range: 1.2 to 20.3), two 'no change' ratings, and only one slight negative change (child #21) for the parent rating of the '*Social wellbeing & acceptance*' domain (77.1 vs. 75.0). Using the same MCID criterion, two children recorded clinically significant decreases (#38 and #47), one from each Group.

Compared to the SG literature, Preston *et al.* (2016) used a lower threshold for clinical significance, namely the SE (0.44 logits), due to the fact that their work preceded the MCID publication of Bleyenheuft *et al.* (2017). Preston *et al.*'s six week SG study recruited 15 children with mostly unilateral CP, with eight children randomised to the intervention (rehabilitation gaming technology) group. An analysis of covariance revealed no statistically significant differences in mean ABILHAND-Kids scores between the intervention and control groups between time points. However, two children recorded clinically significant improvements (one from each group) and nine children recorded clinical significant decreases in activity performance, with four belonging to the intervention group (pg. 1010).

If the lower threshold for clinical significance is used, six children (#4, #6, #7, #8, #21 and #37) achieved a clinically significant ABILHAND-Kids logit increase, four from Group A and two from Group B, and two children recorded clinically significant decreases (#38 and #47), one from each group. Furthermore, all children who recorded a clinically significant increase in ABILHAND-Kids score using the lower threshold also recorded a clinically significant increase in total time for the JTHFT test for the ND hand (Table 32), indicating an association between the performance test for hand function (JTHFT) and the hand function parent- or self-report questionnaire (ABILHAND-Kids).

A study investigating a one year follow up of arm and hand function with children with unilateral CP by Klingels, Feys*, et al.* (2012) identified that the ABILHAND-Kids

score was stable and showed no significant evolution over a year, suggesting that changes in ABILHAND-Kids scores following an intervention may be attributed to the effect of the intervention and not the result of natural course (pg. 263).

6.4.5 The CP QOL Questionnaire

The CP QOL questionnaire, as noted in the Methods chapter (Chapter 3, section 3.4.1) and also recently confirmed via personal communication (Swift, 2017), lacks data on responsiveness or sensitivity, making it difficult to quantify meaningful or clinically significant changes in QOL scores pre- and post-trial. Nor does the questionnaire provide a 'normative' score for children with CP with which to compare. De Civita *et al.* (2005) highlights the importance of an appropriate reference group for QOL comparison (for relative and absolute purposes), whereas for this study each participant's post-trial score is compared only to their pre-trial score, and not averaged across the cohort, or a sub-cohort.

Within the CP QOL literature, it is generally recognised that children with CP have a lower QOL compared to normative data (Livingston *et al.*, 2007), and that wherever possible children should be allowed to report on their own well-being (Varni *et al.*, 2005; Livingston *et al.*, 2007). With respect to the CP QOL questionnaire ratings, this study found that in almost all circumstances children or teenagers rated their own QOL higher than their parents, a theme reported in the literature by Russo, Goodwin*, et al.* (2008) and Russo, Miller*, et al.* (2008), using the Pediatric Quality of Life Inventory (PedsQL) measure. Low self-report/parent report correlations have also been reported by Varni *et al.* (2005) using the KIDSCREEN questionnaire, in some domains of well-being (Livingston *et al.*, 2007, pg. 229).

For the present study, the difference in ratings suggest that children and teenagers are more optimistic about their life with CP potentially because it is all they know, and that parents are possibly comparing their child's life to a life without CP, and see missed or diminished opportunities and abilities for their child. Arnaud *et al.* (2008) has suggested that parent-reported QOL for children with CP is strongly associated with the level of impairment, and White-Koning *et al.* (2008) noted that parent QOL ratings are lower than professional ratings when parents report high levels of stress,

using the KIDSCREEN questionnaire. Pain has also been strongly correlated with low QOL scores (Russo, Miller*, et al.*, 2008; White-Koning *et al.*, 2008; Colver *et al.*, 2015), with Novak *et al.* (2012) reporting that 75% of children with CP were in pain. While pain was not directly measured in this study, each CP QOL questionnaire asked a direct question about pain. When analysed, most pre-trial responses (68%) indicated low levels of pain experienced by trial participants (that is, responses were in the bottom third of the nine-point scale used to assess pain), with only 9% of responses indicating high levels of pain (top third of the pain scale).

The literature reports that QOL may depend on many factors apart from the intervention (Russo *et al.*, 2007), and that children may experience what De Civita *et al.* (2005) refers to as a 'response shift', defined as the "*change in the meaning of one's self-evaluation of a target construct*" (pg. 669). That is, no change in QOL may be recorded despite significant health changes occurring because the individual has adapted to their changed health state and accommodated for their illness when rating their QOL. Additionally, and as reported in section 6.3.10.4, one parent and one teenager reported being less happy or having more concerns about CP post-trial compared to pre-trial. A possible explanation for this result is that the trial caused the parent and child to re-focus on the child's hand impairment, particularly if the child struggled with the OGS. In other words, prior to the trial the parent and child were probably accepting of the child's hand limitations and the child accommodated where they could when performing upper limb activities. Being a new experience, the OGS trial may have caused the child to re-focus on their ND hand (due to the proximity sensors always monitoring hand position), highlighting any negative issues they associate with their ND side and leading to a decrease in QOL score.

6.4.6 OGS Acceptance, Utility, Parent Testimonials and Feedback

The prevailing theme across the trials was that the OGS (referred to as 'Orby' by the children) was liked and enjoyed by trial participants. The average OGS rating was 7.4 ± 1.9 out of 10 (range: 4 to 10), with a mean rating of 8.0 (n=17), indicating a high approval rating. The active vibration stimulation delivered to all Group A children was not reported as being noxious by any child (or parent) and was well accepted. From an OGS robustness perspective, once the 'Orby' controller pins were upgraded

to machined aluminium, not a single breakage has been reported. This is in spite of the OGS being part of a RCT with post-stroke survivors at Hampstead Rehabilitation Centre (as part of a Physiotherapy Honours project co-supervised by the author), loaned to local schools, and routinely set up for the public to use at annual University Open Days, 'Science Alive!' and 'Maker-Faire' events in Adelaide over the last four years. Additionally, when the software isn't tampered with (as reported in section 6.3.12) the OGS runs smoothly and performs as expected.

Apart from the specific feedback that parents reported on the '*Participant Experience Questionnaire*' form (section 6.3.13.2), a common theme was for parents to report increased participation and socialisation between their child and their child's able-bodied sibling(s) when using and sharing the OGS. This is in agreement with Sandlund *et al.* (2012), who reported on parents' perceptions of serious gaming interventions, and the recognition that they provide social activity and are a stimulant for social contact, with the socialisation of such an intervention being recognised as having a positive effect on engagement (pg. 931). Additionally, parents noted that this created a sense of competitiveness amongst siblings. In the author's opinion, this was because the OGS represented the first time an activity (in the form of a gaming system) was given to the family that created an even playing field – meaning the majority of children didn't have to worry about poor hand performance to be able to use and achieve within the game. One parent wrote that she had "*nothing but positive praise*" for the controller and the OGS, and another said that her daughter would never go and independently use the family *Nintendo Wii* system, but that the OGS was easy to use and she would confidently use it on her own. Appendix O contains an email to the author from a parent two weeks into her child's trial, highlighting what she was noticing about how her son was using 'Orby' and the changes she was seeing. To protect the privacy of the family, the mother's name has been truncated, her email address has been removed, and her son's name has been replaced with the words '**our son**' twice in her email.

The 'personification' of the controller through the nickname 'Orby' helped to engender a sense of ownership and personalisation with children and families. While subtle, referring to the controller and hence the overall OGS as 'Orby' helped children to 'connect' with the technology. One parent emailed the author to say that

'Orby' was "*well and almost an adopted member of our family!*", which highlights how the OGS was embraced.

While the overall aim of this study was to develop a system to improve upper limb sensory function for children with CP, the author was told that the OGS caused one of the participants to talk more (as reported in section 6.3.13.2) because he was explaining how to use it to his sister and family members, and passing on game tips that he had learnt. Interestingly, this particular parent report (made in person to the author when the OGS was collected at the end of the trial, and also recorded on the '*Participant Experience Questionnaire*' form) did not come through on the CP QOL tool for this child. The parent reported decreased scores in three of the five domains, no change for '*Pain & impact of disability*', and a slight increase of one point in the '*Social wellbeing & acceptance*' domain.

Parents and children noted the following constructive feedback in terms of how they would like to see the OGS improved. This included:

- Investigating a different mechanism for supporting and positioning the ND hand, other than using a strap;
- Implementing pressure sensitive pads on the controller surface so the child would have to apply equal pressure to both sides of the controller to play games, to encourage symmetrical use and function;
- Developing games that are two-player, to capitalise on the competitive nature of the OGS and sibling interactions, so siblings can play against each other; and
- Developing more games to keep children interested and engaged for longer.

6.5 Summary

This study has demonstrated that the OGS is an acceptable, feasible, accessible, and robust technology for children with both unilateral and bilateral CP, when upper limb sensory and motor impairments are present. Eighteen children were able to independently use and engage with the OGS, with most families reporting high levels of social closeness and participation among siblings during the trial. Analysis of the data suggests it is effective in increasing motor function, assessed via the JTHFT, if not sensory function, however a Type II error cannot be ruled out.

A recommendation following this work is that an appropriately powered RCT should further investigate if upper limb sensory function can be improved through active engagement with the OGS, with at least 28 children recruited to each group as per *post-hoc* sample size calculations. Strengths and limitations of the study are discussed in the following chapter.

6.5.1 Study Hypothesis

"*Somatosensory function in children with CP with a known sensory loss can be significantly improved through a home-based computer gaming system that couples a fun, cognitively engaging and motivating activity with an opportunity to experience a range of appropriate afferent (sensory) inputs. Through the use of vibration stimulation delivered in this context, it is hypothesised that children randomised to treatment with vibration stimulation would have significantly better sensory and functional outcomes than children having no associated vibration stimulation*" (section 6.1.2).

The study design for the RCT included three assessment points over a 14 week period: a baseline assessment (A_1); an 'immediate' post OGS trial assessment (A_2) 10 weeks later; and a follow-up assessment (A_3) a further four weeks later. Statistical modelling identified only one statistically significant between-Group result for the involved ND hand, which was for the test of stereognosis at the immediate follow-up assessment ($A_2 – A_1$). This result was due to children in Group A (haptic group) performing better in the A_2 stereognosis assessment than children in Group B (non-haptic group), combined with the fact that Group B children performed worse in their

A_2 stereognosis assessment. However, this significant between-Group difference did not persist at the follow-up A_3 assessment.

6.5.2 Study Area of Investigation 1

"*The feasibility of deploying the OGS into a family home in an unsupervised and 'child-led' format, as a way of engaging children with CP to use both their hands*" (section 6.1.2).

With the exception of just one participant from the trial (where it was evident that the OGS laptop was tampered with), and once the load-bearing pin within the 'Orby' joystick was replaced with a machined aluminium piece, the OGS proved to be a safe, robust, and enjoyable system for children with limited hand function due to CP. Children could only use the OGS once their two hands were detected by the proximity sensors within the controller, meaning both their hands were always engaged during gameplay. Furthermore, the system was independently operable by children with limited hand function, meaning they weren't reliant on being 'set-up' by a parent or sibling if they wanted to use the OGS – they could be autonomous.

6.5.3 Study Area of Investigation 2

"*Child and parent acceptance of the OGS*" (section 6.1.2).

The OGS was broadly accepted and rated highly (median rating = 8.0 out of 10, n=17), with families politely resistant to having the OGS removed from their home at the end of the trial. The OGS was viewed in a positive, non-stigmatising light, with many families commenting that it didn't look like an item of assistive technology.

6.5.4 Study Area of Investigation 3

"*OGS usability and engagement*" (section 6.1.2).

All children in the trial found the OGS to be highly intuitive, and, depending on their level of hand impairment, easily operable. The most common issue children faced

when they began to use the OGS was ensuring their ND hand covered the proximity sensor. When this resulted in the game pausing, the children would quickly re-position their hand to ensure the game would play again, reinforcing the positive feedback loop created by maintaining correct hand position. Where correct hand position could not be adequately maintained by the child, a hand strap was fitted to support the ND hand (section 6.3.11). The average OGS usage for all 18 children was 377 ± 267 minutes or 6.3 ± 4.5 hours (range: 117 – 1140 minutes), which is just over an hour (63 mins) per week, or nine minutes per day. For the current study, nine minutes per day is less than anticipated and may suggest that children developed 'OGS lethargy' after the initial interest in the games had diminished. Developing more games to provide more incentive to engage and unlock new games may address this issue.

6.5.5 Study Area of Investigation 4

"*The effectiveness of the OGS to improve hand function*" (section 6.1.2).

As discussed in section 6.3.7, when all 18 children are considered as a single cohort, statistical modelling identified a significant improvement in the total time for the JTHFT for the ND hand (p=0.001) between A_1 and A_3. When analysed further, 10 children recorded clinically significant improvements for this test, with the median improvement for the cohort being 35 seconds. Additionally, ten children recorded increases in ABILHAND-Kids logit scores, but only three children (#4, #21 and #37) achieved logit score increases that were greater than the MCID for this tool (+0.71 logits). Two of the three children who recorded clinically significant logit score changes also recorded clinically significant JTHFT score improvements as shown in Table 34 (child #21 and child #37; note child #4 withdrew from the trial, so did not have any A_3 test results).

7. Final Summary and Contribution to the Field

This concluding chapter will summarise the overall project, highlight important contributions from the work to the field of CP research, discuss lessons learnt from the trial, acknowledge the strengths and limitations of the work presented, and outline future work and areas of investigation.

7.1 Overall Project Summary

CP is the most common cause of childhood physical disability (Reddihough, 2011). While the primary cause of CP is a disturbance to the developing or fetal brain, the known motor disorders of CP are accompanied by one or more secondary or co-occurring impairments, such as sensory disturbances (Rosenbaum *et al.*, 2007), that affect function and present limitations to the child over time (Novak *et al.*, 2012). Sensory input is known to be an essential component of motor function and motor control, with recognition that sensory deficits may constitute limits on the functional outcome of children with CP (Cooper *et al.*, 1995, pg. 301).

The **Stage 1** phase of this PhD research assessed a cohort of 42 children living with CP in South Australia for somatosensory function, and was the first population-based sensory assessment of children living with CP in the state. Informed by the literature, each child was assessed using validated and reliable sensory tests, namely, tactile registration using SWM, proprioception using the distal phalanx of the child's thumb, tactile perception using stereognosis with six objects, and a test of functional hand motor skills using the JTHFT. TPD was assessed using the AsTex® device, which is the same device used by Auld *et al.* (2012b). However, the utility of this device was problematic during the current study (as reported in Chapter 4, section 4.3.5), but confirms a similar outcome to that reported by Causby (2016), who found the test counter-intuitive and subject to variation when testing university students.

Overall, 36 children (22 males, average age = 10 ± 3.3 years) with either unilateral (n=23) or bilateral (n=13) CP completed the tests satisfactorily, with six children excluded from the results and data analysis mainly due to behavioural issues. Eight children (22%) recorded results that indicated 'normal' or intact somatosensory

function, with the predominant classifications of this particular sub-group being: MACS Level I (7), ND left side (6), males (6), a brain scan showing periventricular white matter injury (6), and bilateral CP (5). The finding that most children with 'normal' sensation had a brain scan classification of periventricular white matter injury is consistent with current understanding that sensory processing is mainly cortical/subcortical in nature and that white matter injury is associated with favourable hand function (Holmström *et al.*, 2010; Arnfield *et al.*, 2013).

The somatosensory deficit prevalence for this study was 78%, which is comparable to the results of previous published studies (Hohman *et al.*, 1958; Jones, 1960; Kenney, 1963; Krumlinde-Sundholm & Eliasson, 2002; Auld *et al.*, 2012b). Similarly, this study recorded somatosensory impairments in the dominant or less-affected hand for 52% of children with unilateral CP as previously reported in the literature (Monfraix *et al.*, 1961; Wigfield, 1966; Lesný, 1971; Lesný *et al.*, 1993; Cooper *et al.*, 1995; Krumlinde-Sundholm & Eliasson, 2002; Arnould *et al.*, 2007; Wingert *et al.*, 2008; Auld *et al.*, 2012b), confirming the need to assess this sub-group bilaterally. In agreement with Auld *et al.* (2012b), tactile registration deficits were associated with an increased likelihood of tactile perception deficits, with all eight children who recorded an abnormal SWM result also recording an abnormal tactile perception result and a poor time for the JTHFT.

Children with unilateral CP recorded more severe sensory loss compared to children with bilateral CP. Only children with unilateral CP recorded severe tactile registration loss, and 91% of children with a severe tactile perception loss belonged to the unilateral CP group. Five of the six children with severe tactile registration loss had a brain scan showing cortical/subcortical lesions, an area of the brain known to be involved in sensory processing and associated with severe impairment (Uvebrant, 1988). Better hand function and performance were associated with better stereognosis results for the overall cohort, both sub-groups, and for both hands, with the strongest association being for the unilateral group, which is in agreement with the work of Kinnucan *et al.* (2010).

Children with unilateral CP performed statistically significantly worse using their ND hand compared to their dominant hand for the tests of proprioception, stereognosis and total JTHFT scores, however, children with bilateral CP performed equally well

with either hand across all tests, but still statistically significantly poorer when compared to TDC. When results are compared across CP groups, statistically significant differences were identified for the test of stereognosis and total JTHFT time, with the bilateral group performing better in terms of overall test performance. Moreover, the dominant hand of children with unilateral CP was significantly slower compared to the dominant hand of TDC (p=0.004), which is in agreement with previous research (Rich *et al.*, 2017).

The primary aim of this thesis was to investigate an upper limb somatosensory intervention for children with CP with a known sensory loss. The hypothesis was that sensory function could be significantly improved through a six-week home-based serious games intervention using the OrbIT Gaming System (OGS). The OGS is an accessible computer gaming system that combines coordinated and integrated upper limb use with an opportunity to experience a range of appropriate afferent sensory inputs. Unlike some robotic rehabilitation devices, the OGS does not rely on stereotypical repetitive movements. The child is required to cognitively attend to the gaming system and make dynamic decisions in response to changing visual stimuli, meaning a variety of movement trajectories through coupled bimanual integrated hand use are required to successfully play the games. Thus, the OGS provides motivational therapy to the player that incorporates contextually relevant visual, aural, motor and vibro-tactile (haptic) feedback. It was hypothesised that children randomised to treatment with active vibration feedback to the child's hands (Group A) would have significantly better sensory and functional outcomes post-trial compared to children using the OGS but receiving no active vibration stimulation (Group B).

From an eligible pool of 25 children that were identified during Stage 1, 18 participated in the **Stage 2** RCT, with 10 children (eight males) randomised to Group A and eight (four males) to Group B. This was a representative group given the profile of the Stage 1 cohort, meaning the Stage 2 cohort had similarly more male children (67%), with a higher proportion of children with unilateral CP (72%), but similar percentages of children with a ND left side (61%), and a predominant brain scan classification of cortical/sub-cortical lesions (39%).

Statistical modelling revealed no significant differences between baseline (A_1) and post-trial (A_2 and A_3) sensory assessment measures between the two Groups (A or B) for the dominant hand, and only one statistically significant result for the ND hand for the test of stereognosis between the Groups at the immediate A_2 assessment. However, this significant between Group difference did not persist at the follow-up A_3 assessment four weeks later. Given all 18 children who participated in the trial were engaged in a forced bimanual integrated upper limb task, a secondary exploratory analysis and re-modelling was conducted. The subsequent analysis identified a strong statistically significant difference between baseline (A_1) and follow-up (A_3) assessments for the ND hand for the total time taken to complete the JTHFT (p = 0.001), however, a Type II error cannot be ruled out.

Ten children recorded increases in ABILHAND-Kids scores, but only three children (#4, #21 and #37) achieved logit score increases that were greater than the reported MCID (+0.71 logits) for this tool (Bleyenheuft *et al.*, 2017). All three children were randomised to Group A. If the lower threshold for clinical significance is used (using the SE = +0.44 logits), six children (#4, #6, #7, #8, #21 and #37) achieved a clinically significant ABILHAND-Kids logit increase, four from Group A and two from Group B. Additionally, all children who recorded a clinically significant increase in ABILHAND-Kids score also recorded a clinically significant increase in total time for the JTHFT for the ND hand, indicating an association between the performance test for hand function (JTHFT) and the hand function parent- or self-report questionnaire (ABILHAND-Kids). With respect to the CP QOL questionnaire, an observation from the present study is that children and teenagers with CP are more optimistic about their life with CP than their parents.

A prevailing theme across the RCT was that the OGS was liked and enjoyed by children, with a mean rating of 8.0 out of 10 (n=17), indicating high approval. Engagement with the OGS during the six-week trial was variable, ranging from 1140 minutes (or an average of 27.1 minutes per day) to 136 minutes (an average of 3.2 minutes per day), with the average usage being 377 ± 267 minutes (an average of 9 ± 6.4 minutes per day). Mid-trial, the OGS Games Catalogue was changed, and rather than give each child 15 games from the start, children were only given five games initially and required to ‘earn’ access to the remaining 10 games by accruing

game time on the OGS. This caused a change in overall usage between the Games Catalogues, but the result was not statistically significant (p=0.057). However, from a clinical perspective, the new or 'Incentivised Games Catalogue' caused a median increase of 112 minutes of OGS usage during the trial, which is a positive improvement in terms of the amount of therapy delivered for children with limited hand function. Intensity of practice is a known factor for successful rehabilitation interventions (Kleim & Jones, 2008).

The active vibration stimulation delivered to all Group A children was not reported as noxious by any child (or parent) and was well accepted. Parents frequently reported increased participation and socialisation between their child and their child's able-bodied sibling(s) when using and sharing the OGS, which confirms the work of Sandlund *et al.* (2012), who reported on parents' perceptions of serious gaming interventions. Additionally, parents noted that the OGS created a sense of competitiveness amongst siblings. One parent noted that her child talked more during the trial because he was helping his typically developing sister overcome obstacles within particular games by passing on his game strategies. Another parent commented that his son started to "*talk about CP more*", that he was "*proud*" of the OGS, and that the OGS became a conversation starter when people visited the family home.

The RCT demonstrated that the OGS is an acceptable, feasible, accessible and robust technology for children with both unilateral and bilateral CP, when upper limb sensory and motor impairments are present, with 18 children capable of independently operating and engaging with the OGS.

Implications for clinical practice and theory include confirmation that somatosensory issues are prevalent amongst this population and that *bilateral* somatosensory assessments should occur for all children with CP prior to an intervention. Additionally, the OGS showed high utility, was enjoyed by the majority of trial participants, and provided pilot evidence that a cognitively engaging activity coupled with forced-bimanual upper limb use and dynamic hand-eye coordination led to improved hand function for the ND hand. However, a larger and appropriately powered study needs to be conducted to validate the results of the pilot study.

7.2 Contribution to the Field of CP Research

This PhD research has contributed an up to date and comprehensive analysis, critique, and synthesis of the literature relevant to CP upper limb somatosensory research, through the analysis of 27 prior studies in this field. The first South Australian and the second national population-based somatosensory assessment of children with CP was conducted, with the results being compared to the literature.

In terms of the technology developed for the research, a new and accessible serious gaming system known at the OrbIT Gaming System (OGS) was conceptualised, designed, developed, tested and deployed as the intervention for a RCT, through a co-design and multi- / trans-disciplinary team approach. The OGS is a standalone, home-based serious gaming system that incorporates a controller that couples bimanual upper limb integration, requiring dynamic hand movements in response to a variety of games that present randomising visual stimuli, augmented with appropriate, contextualised haptic feedback, while catering for hemispherical isolation to avoid the phenomenon of sensory extinction, and represents a novel technology for the field. A patent for the novel technology has been filed (Hobbs, Hillier*, et al.*, 2015) and approval has been granted in Australia, Singapore and the United States. Known barriers to engagement with rehabilitation interventions have been addressed through the careful and iterative process of co-design. Additionally, a home-based approach using a system that automatically logged use and activity was adopted to minimise inconvenience to families participating in the trial. Post-trial, the OGS received a very positive review when independently assessed by a United States-based accessible computer gaming group, called the *AbleGamers Charity* (part of the *AbleGamers Foundation*, see Appendix J).

This project has confirmed a more holistic approach is required for children with CP. Sensory issues have been confirmed as an important impairment and deserving of more attention in service provision, particularly because of their relationship with dexterity – another aspect demonstrated in this study. Further, the importance of working with the whole child (including the less-affected side) in children with unilateral CP is confirmed.

7.3 Strengths of the Study

The overall project had the following strengths. Firstly, the benefits of a multi- and trans-disciplinary team were evident throughout the project, with more than 10 different professional backgrounds 'represented' across the project. This ensured that a diverse range of professional and clinical opinions and perspectives were part of the decision-making process. Additionally, adopting a co-design approach to the OGS that included age-appropriate children both with and without CP ensured the final product was fit for purpose.

Recruitment bias was minimised by recruiting through a population based state-wide Register, removing hospital, clinic, clinician or geographical bias from the process, leading to a more representative sample of children being recruited. The sensory and motor assessments chosen for this study were all valid, reliable and had been used with children with CP in the published literature. Assessor inter-rater reliability was addressed through the coordination of a formal orientation and training session for a number of potential assessing therapists prior to the study beginning. The aim of this session was to determine a common understanding of the somatosensory assessments and to seek input on the format of the Stage 1 and 2 recording sheets for a consistent approach to all assessments.

For the RCT, the assessing therapist was blinded as to group allocation, as was the consulting statistician for the project. Additionally, the author and lead researcher was blinded as to group randomisation during demonstration of the OGS (using the OGS 'Demo' mode), so could not bias the child during OGS deployment based on knowing the group to which they were randomised.

Basing the RCT within the family home meant that the child/family was not required to attend the WCH to participate in the trial and, coupled with the independently accessible design of the OGS, leveraged the child's potential to engage with the OGS whenever they desired at home. Furthermore, the OGS tracked and stored all game data locally (onto the OGS laptop), removing the need for keeping a diary or journal of all OGS use, further reducing the burden of the trial. Reducing family burden during a SG trial was a recommendation of a similar trial in United Kingdom (Preston *et al.*, 2016, pg. 1013).

For continuity and consistency, the author and lead researcher conducted all OGS demonstrations and was a single point of contact while the OGS was on trial, meaning a consistent message was provided to families. The OGS offered 15 different games in an effort to appeal to a broad audience and to reduce game boredom and fatigue, which had been reported by other studies (Li *et al.*, 2009; Preston *et al.*, 2016). Additional functionality in the form of an 'Incentivised Games Catalogue', which released new games based on system engagement, resulted in an average increase in OGS use of 221 minutes and five extra days during the trial. Periodic release of new games was a recommendation by Li *et al.* (2009)(pg. 112).

Trial compliance was 100% – all 18 children who began the trial also completed it. Two children withdrew post-intervention, for reasons explained in Chapter 6 (section 6.3.5.2). Additionally, the active vibration feedback provided to all Group A children was not noxious, with no adverse effects reported.

7.4 Study Limitations

The limitations of the study are acknowledged as being the following. Firstly, an underlying assumption of the study is that all gameplay by children that were part of the RCT only occurred using the profile that was allocated to them, meaning they only ever played games using their named profile, and guests only ever played games using the 'Guest' profile. These instructions were part of the study protocol (Appendix I), but with no way to formally validate that the above occurred, it is possible that usage by either the child or a guest may have taken place using the contra profile.

The number of children recruited overall was low, despite keeping the Stage 1 assessment window open for longer than originally intended to increase study numbers. Only 49 children responded to the mail out, resulting in a Stage 1 cohort of 36 children. Through personal communication with current allied health staff, and from the author's prior experience working for a disability sector provider, the low recruitment may be related to 'research fatigue' or 'research burnout' with this population. The flow on effect of this 'fatigue' or 'burnout' was that the RCT cohort was only 18 children. A *post hoc* sample size calculation indicated approximately 55

children (or 28 children per group) were needed to detect a difference (at 80% power), which limits the ability to report conclusive outcomes.

For consistency and to improve assessment reliability, it was hoped that only one assessing therapist would be required for the entire project. However, two Occupational Therapists took maternity leave part way through the project and a third took up a new position, meaning three Occupational Therapists and one Physiotherapist conducted the assessments for Stages 1 and 2. To mitigate the effect of not having a single assessing therapist for the project, all four assessing therapists were part of the formal group orientation and training session (mentioned in section 7.3) conducted prior to the project beginning.

Some of the children experienced 'OGS fatigue' and did not engage with the OGS as much as they did when they first received it. A common pattern was high engagement to begin with that waned as the trial progressed, similar to other trials (Preston *et al.*, 2016). It was hoped that providing more game choice would address this issue (the OGS had 15 games, whereas the study by Preston *et al.* only had four), but a lack of engagement was still evident with the current study.

As reported in Chapter 6 (section 6.3.12), some 'Orby' controllers broke during the trial due to the weakness of the 3D printed pin, affecting four children's trials. Once the problem was identified, all controllers had their 3D printed pins replaced with aluminium pins with no further breakages being reported, demonstrating that the OGS was safe, robust and did not require further author support while on trial. Unfortunately, one of the families did not contact the author when their controller broke just after the half way point of the trial (day 29 of 42), meaning the child did not experience the same amount of trial time as the other children. Apart from the trial issues highlighted in section 6.3.12, the only time a family reached out to the author was to request a ND hand strap, as reported in section 6.3.11. One family used the OGS as a 'reward', only allowing their child to use it after all their homework was completed and if they behaved and did not fight with their sibling, which potentially restricted this child's use and engagement. While this particular use of the OGS while on trial was unexpected, it highlighted how this child felt about the OGS, given it was 'used' as a reward for positive behaviour.

The RCT for this project did not use a traditional control group, since both groups received the OGS. Consequently, any post-trial improvements need to be interpreted with caution. However, the RCT did control for the vibration element of the trial that was being investigated, but not the bimanual aspect of the study, which is discussed in section 7.5.

Despite measures being taken to prevent access to the OGS desktop and Windows operating system, one OGS was deliberately tampered with during the trial, which caused the child's allocated group profile to flip (from Group B to Group A). This meant that this child experienced both arms of the trial. However, as detailed in Appendix N, in terms of overall OGS usage by the child in question, 93% (526 minutes) was accrued in the group they were randomised to, with only 7% (39 minutes) of time accrued in the other profile. Prior to deploying an OGS for another trial, the keyboard should be covered with a guard to prevent access to the touchpad and keyboard, eliminating the prospect of tampering with the laptop.

7.5 Future Work

The first recommendation following this PhD work is that an appropriately powered RCT should further investigate if upper limb somatosensory function can be improved through active engagement with the OGS, with at least 28 children recruited to each group as per *post hoc* sample size calculations. This could be achieved through a collaborative multi-centre, multi-state trial to ensure recruitment is adequate.

One of the significant outcomes reported from Stage 2 of this study was the improvement in ND hand function for the overall RCT cohort (assessed using the total time for the JTHFT), indicating improved hand function between baseline and follow-up assessments. One possible explanation for this result could be that the forced-bimanual upper limb coupling requirement of the OGS engaged the ND hand in an active manner, meaning it was used in a functional, dynamic way. It is then possible that through increased use, the child began to use their ND hand for other activities. Testing this hypothesis would be relatively straight-forward – a follow-up trial could include a group that receives an OGS like Group A from the current study,

with a second group receiving the same OGS but with the proximity sensors disabled. Disabling the proximity sensors removes the forced bimanual use requirement, meaning the child could use the controller unilaterally if they wished.

Following multiple conference presentations on the project, the author has received significant interest in the OGS from the post-stroke research community. In 2016, one of the project supervisors (SH) and the author supervised and conducted a pilot RCT of the OGS with a small cohort (n=10) of stroke survivors in the acute phase of recovery at Hampstead Rehabilitation Centre in Adelaide (Watchman, 2016). Using a slightly different games catalogue, the OGS demonstrated utility, feasibility and appropriateness within a post-stroke setting, reporting statistically significant improved tactile registration (for the first finger and thumb) and functionality of the impaired upper limb (using the Wolf Motor Function Test) following a three week intervention (Watchman, 2016). A larger and possibly multi-centre trial is now planned, with interest in the OGS from interstate. Ideally, a future study with either children with CP or post-stroke survivors would incorporate diffusion-tensor MRI imaging and fibre tractography, to quantify the effects, if any, that engaging with the OGS has on trial participants.

A strong theme from the RCT was that the OGS created opportunities for increased social interaction and participation among siblings, presumably because the OGS provided an accessible ‘level playing field’ that enabled children with a hand impairment due to CP to play and compete with their typically developing sibling(s). When seeing the OGS in action, therapists and clinicians have remarked that the OGS, or the ‘Orby’ controller on its own plugged into a typical computer, may be a unique platform for teaching children with learning disabilities, coupling hand (motor) and learning (cognitive) activities. This could be heightened if two systems are set up alongside each other to harness the power of group and social learning.

As mentioned in Chapter 6 (section 6.1.1), the ‘active’ element for the RCT intervention was the use of haptic or vibration stimulation because vibration is known to activate the primary and secondary somatosensory cortices (Coghill *et al.*, 1994) and to enhance computer game immersion. An aspect of the haptic vibration delivered during the trial to Group A children that requires further exploration relates to the circumstances around when it was provided. All haptic events were coupled to

activities within the game – from collecting a bonus/reward, to rubbing up against a wall when running, to landing when jumping – every game action was matched to a corresponding haptic event of varying intensity and duration. Consequently, most 'high intensity' (strong) vibrations were delivered in response to negative game events, with the highest intensity vibrations delivered when the player 'lost a life'. From a brain processing and plasticity perspective, the effect of coupling the strongest vibrations to the most negative game events in the context of this intervention requires further consideration and investigation.

This project has generated significant commercial interest nationally and internationally, in both the overall gaming system and the controller as a stand-alone accessible device to access content other than computer games. In late 2017 the technology was licensed to an Adelaide-based disability services and equipment provider, who intends to commercialise the next version of the accessible controller, called '*i-boll*' (conceptualised by A/Prof Sandy Walker).

Lastly, the author has been approached by an Adelaide-based researcher working with patients with Parkinson's disease, particularly interested in using the OGS as a cognitive training intervention. Most cognitive training systems breakdown at the hardware 'human computer interface' level, particularly where Parkinsonian tremor is a significant issue. Pilot testing in the laboratory has confirmed that the design and accessibility of the 'Orby' controller addresses issues relating to uncontrolled movement, and a grant application to fund a pilot trial in this area is currently under review. The development of a new games catalogue to address specific game and software issues related to cognitive training for this population is planned.

7.6 Conclusion

This series of studies has added to our knowledge of upper limb sensory loss in children with CP – its prevalence, nature and potential amelioration. The primary recommendation from this PhD is that sensory impairments should continue to be more intensively addressed, as echoed recently by Auld and Johnston (2018), and that systems such as the OGS warrant further research to explore effectiveness and utility through an appropriately powered trial. The pilot RCT demonstrated a way to engage children with an upper limb impairment due to CP in an activity they found enjoyable, while promoting sustained bimanual upper limb use. Statistical modelling and analysis revealed a strong, significant improvement in ND hand function between baseline and follow-up assessments following a six-week home-based trial.

The OGS proved to be accessible, independently operable by children with a hand impairment due to CP, and engaging while on trial. Parents reported high levels of social closeness and participation between siblings during the trial, with the OGS rated highly by trial families and an independent gaming accessibility foundation in the United States. Commercial opportunities have been explored and the technology has now been licensed.

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Appendix A

This Appendix contains the Stage 1 recording sheet that was used during assessment sessions to record all test results.

Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy

Date of assessment: _____ Location: _____

Assessors name: _____

Participant's study number: _____ Gender: _____

Participant's initials: _____

Participant's date of birth: _____

Cerebral palsy type: _____

Dominant side: _____

MACS Level: _____ (Manual Ability Class. System)

Date and source of MACS Level: _____ (e.g.: WCH file, SACPR, etc)

Project contact:

If you have any questions, queries or problems about anything to do with the study, please contact David Hobbs (mobile: 0418 221 811 or work: 8201 3167).

Sensory Assessment Test Results:

1. Test for tactile detection (Semmes-Weinstein monofilaments) (Blind fold required)

(Note: green is hardest level to feel (the lightest or most sensitive), so start with this coloured monofilament – if they can feel this, then you don't need to proceed to the other colours).

Each filament is applied to the **first pad** of the index finger or thumb. The order can be random. Tick (√) the circle if they detect the colour at that site, cross (X) if they fail to detect – three attempts on each site but randomly applied across the four sites.

Detection	***Green***	***Blue***	***Purple***	***Red (K)***	***Red (T)***
(R) Finger 1	O O O	O O O	O O O	O O O	O O O
(R) Thumb	O O O	O O O	O O O	O O O	O O O
(L) Finger 1	O O O	O O O	O O O	O O O	O O O
(L) Thumb	O O O	O O O	O O O	O O O	O O O

Highest detection level (R): _____ (finger) _____ (thumb)

Highest detection level (L): _____ (finger) _____ (thumb)

2. Test of texture discrimination (using the AsTex device): (Blind fold required)

Trial No.	***Test 1: Non-Dom.***	***Test 1: Dom.***	***Test 2: Non-Dom.***	***Test 2: Dom.***
Trial 1				
Trial 2				
Trial 3				

Perform the Test 1 with the instructions:

"*Stop your finger at the point where the strip "feels smooth" and hold your finger in that position until I can record the value*" – do not allow them to move their finger backwards and forwards over the grids.

Perform the Test 2 with the instructions:

"*Stop your finger at the point where you can't feel individual lines anymore and hold your finger in that position until I can record the value*" – do not allow them to move their finger backwards and forwards over the grids.

Adjusted final score (R):_____

Adjusted final score (L):_____

3. Test of proprioception (by moving the distal thumb either up or down)
(Blind fold required)

Non-Dominant hand: Total number correct _____ /10

Dominant hand: Total number correct _____ /10

4. Test of stereognosis: (Blind fold required)

When choosing objects, ensure that 3 are chosen from the 'similar pairs' group of 6 and that 3 are chosen from the 'non-similar' group of 6 objects. Randomly choose the objects so that there is some overlap between the non-dominant and dominant hand, but that some new objects are also used.

Object chosen by therapist (e.g.: pen)	***Non-Dom.: Object identified correctly?*** (Y or N)	***Object chosen by therapist*** (e.g.: pen)	***Dom.: Object identified correctly?*** (Y or N)
1.		1.	
2.		2.	
3.		3.	
4.		4.	
5.		5.	
6.		6.	

Non-Dominant hand: Total number correct _____ /6

Dominant hand: Total number correct _____ /6

5. The Jebsen Taylor Hand Function Test (JTHFT): (no blind fold)

The maximum time allowed for any task below is **120 seconds** (2 mins). If they cannot complete the task in that time (they run out of time), score them a value of '120 secs'. If a child cannot complete the task at all, assign them the value of '120 secs', but write a note below that they couldn't attempt or complete the task at all.

Task	***Non-dominant hand***	***Dominant hand***
Card turning	sec	sec
Manipulating small objects (into can)	sec	sec
Simulated feeding (bean in cans)	sec	sec
Stacking checkers	sec	sec
Moving light objects	sec	sec
Moving heavy objects	sec	sec

Assessor comments/notes/observations: (on any aspect of the assessment)

Would this child be suitable for STAGE 2 of the trial? That is, can the child place ***both their hands*** on a custom-made controller (shaped like a ball, not a traditional joystick) to be able to play the haptic computer game system in Stage 2 of this study? If no, please state the reasons below (e.g.: the child has a fixed deformity and can't open their hand, etc):

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Appendix B

This Appendix contains the '*Participant Experience Questionnaire*' that was used to evaluate the OGS and collect information and feedback from families that participated in the Stage 2 RCT.

Participant Experience Questionnaire

(*Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial*)

Participant no: _____ Participant initials: _____

To be completed by the child, either independently or in conjunction with their parent(s).

Thank you for being a part of our research study! Now that the computer gaming trial has finished, we would like to ask you a couple of questions about the trial.

1. Did you enjoy playing the computer games? Please indicate on the scale below, by putting a tick or cross on the line, how you would you score the system out of 10 (10 = "*excellent, I loved it*" and 0 = "*very poor, I didn't like it at all*").

0 1 2 3 4 5 6 7 8 9 10

2. Did anything positive (something good) occur during the computer gaming trial? If so, please tell us what it was:

3. Did anything negative (something not good) happen during the computer gaming trial? If so, please tell us what it was:

4. During your computer gaming trial, did you receive any of the following (yes/no):

a. Physiotherapy? _____ If yes, how often? _____
b. Occupational therapy? _____ If yes, how often? _____
c. Botox? _____ If yes, when? _____

This study has been given approval by the Women's & Children's Health Network Human Research Ethics Committee, project number *REC2530/12/15*. If you wish to discuss the approval process, or have any concern or complaint about this study, please contact the Executive Officer of the Human Research Ethics Committee, Ms. Brenda Penny, Research Secretariat, on 8161 6521.

Appendix C

This Appendix contains the Stage 1 study documents that were mailed out to all eligible families (section 4.1.6), namely the: letter of introduction, study information sheet, and consent form.

Government of South Australia
SA Health

Women's & Children's Hospital

South Australian Cerebral Palsy Register

Ground Floor Angas Building
72 King William Road
North Adelaide SA 5006

Tel 08 8161 7242
cywhs.cpregister@health.sa.gov.au

27 April 2012

<Parents/guardians/caregivers name(s)>
<Address>
<Suburb, SA, Postcode>

Dear <Names of parents/guardians/caregivers>,

Re: Research Project "Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy"

I am writing on behalf of the researchers to inform you of the above research study being undertaken at this hospital. The project is headed by Dr Ray Russo, the Director of Paediatric Rehabilitation Department at the Women's and Children's Hospital (WCH). The project also involves Mr. David Hobbs, a Rehabilitation Engineer and PhD Candidate at Flinders University, and an Occupational Therapist from the Paediatric Rehabilitation Department who are working with Dr Russo on this project.

You have been invited to participate in this study because your child is part of the South Australian Cerebral Palsy Register (SACPR), and the above project provides an opportunity for children with CP to have a clinical sensory assessment as part of the study.

The purpose of this research project is to determine how children with CP sense touch using their hands, and if their sense of touch is different to children who don't have CP who are the same age. Current research suggests that children with CP sense objects and the world around them differently, but it is difficult to know just how different their sense is. This study will provide valuable information to help understand how common sensory loss is in the hands of children with CP and how this affects how a child uses their hands and arms.

Included with this letter of introduction is an information sheet, which describes the project in more detail, and a consent form for the study.

If you would like to participate in this study, please read and complete the attached consent form and return a copy to us in the reply paid envelope.

Please note that participation in the study is at all times voluntary. You can elect to cease your child's participation at any time, without giving a reason. To help with attending the sensory assessment session at the WCH, families will receive $20 to assist with car parking and travel costs.

We would also like to assure you that we take confidentiality and privacy very seriously. If you decide to participate, any information you provide will be treated as highly confidential, and not be shared with any other group or individual, except where there is legal requirement to pass on personal information to authorised third parties. This requirement is standard and applies to information collected both in research and non-research situations. Such requests to access information are rare; however we have an obligation to inform you of this possibility. You and your child will not be identifiable in any publication resulting from this research.

There is a possibility that a journal or conference publication may result from this work, however, your child will not be identifiable in any way in the publication. During the study, all the information that will be collected will be treated confidentially, with data being stored in accordance with the National Health and Medical Research Council/Australian Research Council Australian Code for the Responsible Conduct of Research (2007) guidelines. If you have any specific queries about the conditions under which data for this study will be stored, please contact Mr David Hobbs (see the details provided on the Information Sheet).

The study has been given approval by the Children, Youth & Women's Health Service (CYWHS) Human Research Ethics Committee (HREC). Please contact the Secretary of the Committee (Ms Brenda Penny, Research Secretariat, Ph: 8161 6521) if you wish to discuss the approval process, or have any concern or complaint.

Please do not hesitate to contact Mr. David Hobbs (Ph: 8201 3167) or Dr Ray Russo (Ph: 8161 7220) should you have any questions or wish to discuss any part of this study.

Regards

Dr Catherine Gibson
Manager, SA Cerebral Palsy Register

Do the hands of children with cerebral palsy feel 'touch' differently to children without cerebral palsy?

(*Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy*)

Study purpose

The aim of this project is to determine how children with cerebral palsy sense touch using their hands, and if their sense of touch is different to children who don't have cerebral palsy who are the same age.

Current research suggests that children with cerebral palsy do sense objects and the world around them differently, but it is difficult to know just how different their sense is. This study is an Australian first and it is hoped it will provide valuable information to help understand how common sensory loss is in the hands of children with cerebral palsy and how this affects how a child uses their hands and arms.

The research team

The Chief Investigator for this project is ***Dr Ray Russo***, the Director of the Paediatric Rehabilitation Department at the Women's & Children's Hospital. Other investigators for this project include ***Mr David Hobbs*** (Associate Lecturer / PhD student and Rehabilitation Engineer, Flinders University), ***Dr Susan Hillier*** (Associate Professor, University of South Australia) and ***Professor Karen Reynolds*** (Professor of Biomedical Engineering, Flinders University). This study is part of David's PhD in Biomedical Engineering.

Will my child's medical records need to be accessed if I agree to my child's participation in the study?

Your child's medical records will be accessed if your child is enrolled into the study, to provide more information about your child's cerebral palsy.

What does the study involve?

The study involves assessing the touch sensitivity of your child's hands. Most children with cerebral palsy have a dominant hand (one hand they use more than the other), and in this study both hands will be assessed. During the study, four (4) sensory assessments and one (1) series of hand skill tests will be performed, as described below:

- If your child can identify an unknown object when it is placed in their hand, without using their vision
- If your child can detect the difference between two raised lines when the distance between those lines gets smaller and smaller, without seeing the lines
- If your child can tell if their finger is lifted up or down, when they can't see their hand or fingers
- If your child can detect when light pressure is applied to the end of their finger, without seeing when the pressure is applied

- How quickly your child can complete a series of six (6) timed tasks, such as turning over a series of cards, lifting small common objects and placing them into a container, scooping up small objects with a spoon, stacking checkers, lifting large but light objects (empty soup cans), and lifting large heavy objects (full soup cans).

During the assessment your child will also have their '*Manual Ability Classification System'* or MACS level assessed, which is a scale that describes how children with cerebral palsy use their hands to handle objects in daily activities.

The above assessments and tests will be conducted at the Women's & Children's Hospital in North Adelaide, and should take between **45 and 60 minutes** to complete.

Will my child benefit from being involved in this study?

Your child will not directly benefit from being involved in this study as it is an assessment study, measuring the sense of touch in your child's hands. You and your child may benefit from knowing more about how your child senses objects, but involvement in this study will not improve or decrease your child's sense of touch. We hope to use this information to help us understand the effects of CP more and how to improve the therapy we can offer to children with CP. Your child may be invited to participate in a follow-on study for the project (see 'Future studies' section below).

What are the possible risks of this study?

This study only assesses how your child uses their hand and senses touch, and uses simple tests that are common clinical practice. The possible risks of being involved in this study include: your child potentially feeling frustrated or annoyed because they can't complete or perform a task, possible discomfort during an assessment, and potential inconvenience when attending the assessment at the Women's & Children's Hospital.

Can I withdraw from the study at any time?

Your child's involvement in the study is completely voluntary, and your child may withdraw from the study at any time without prejudice to his/her future treatment or relationship with the Women's & Children's Hospital.

Future studies

When we have finished this measurement study we will be commencing a trial of therapy that aims to improve hand skills in children with reduced sensation in their hands. We don't know if your child will be eligible for this future study, but we can let you know the details of this future study if you are interested and if your child meets the criteria for inclusion.

Reimbursement or assistance with costs associated with the study

Families will receive $20 to assist with expenses associated with travel and car parking. This will be given to families at the conclusion of the sensory assessment.

Your child's personal information and confidentiality

Your child's information will remain confidential except in the case of a legal requirement to pass on personal information to authorised third parties. This requirement is standard and applies to information collected both in research and non-research situations. Such requests to access information are rare; however, we have an obligation to inform you of this possibility. If you consent to your child being photographed for research purposes during the study, the photographs of your child may be used in research publications and presentations.

Will my child be photographed during this study?

We seek approval to photograph your child during the assessment study. The photographs will be used for research purposes, to communicate how the study was conducted. It is important to note that your child will not be able to be identified by the photograph – the photos will focus on your child's hands/arms only. You can choose for your child to ***not*** be photographed, and this will not affect their participation in the study.

What happens in the case of an adverse reaction or adverse finding?

Should an adverse event occur, the Chief Investigator should be contacted immediately to report the issue. If an adverse finding results from your child's involvement in the study, you will be contacted by the Chief Investigator who will explain the situation.

Who do I contact if a problem arises?

If a problem arises, or if you have any questions about the study, the following contacts are provided:

For problems arising during the study, please contact Dr Ray Russo or the consultant on call if Dr Russo is not available:

Dr Ray Russo (Chief Investigator)
Director, Paediatric Rehabilitation Department
Women's & Children's Health Network
Phone: 8161 7220
Pager: 8161 7000, page Dr Russo or the consultant on call for paediatric rehabilitation if he is unavailable

For information about this research study, please contact:

Mr David Hobbs (Study Coordinator)
Associate Lecturer/PhD Candidate and Rehabilitation Engineer
Flinders University
Phone: 8201 3167
Email: david.hobbs@flinders.edu.au

This study has been given approval by the Women's & Children's Health Network Human Research Ethics Committee, project number REC2441/12/14. If you wish to discuss the approval process, or have any concern or complaint about this study, please contact the Executive Officer of the Human Research Ethics Committee, Ms Brenda Penny, Research Secretariat, on 8161 6521. The project has also been approved by the Government of South Australia SA Health Human Research Ethics Committee, project number 480/11/2014.

WOMEN'S & CHILDREN'S HEALTH NETWORK (WCHN)

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

CONSENT FORM

LAY TITLE: Do the hands of children with cerebral palsy feel 'touch' differently to children without cerebral palsy?

SCIENTIFIC TITLE: Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy

I _____

hereby consent to my child's involvement in the research project entitled:

"Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy"

1. The nature and purpose of the research project described on the Information Sheet in the information pack that was sent to me in the mail, has been explained to me. I understand it and agree to my child taking part.

2. I agree to the accessing of my child's medical records by investigators of this study, including the South Australian Cerebral Palsy Register.

3. I understand that my child may not directly benefit by taking part in this study.

4. I acknowledge that the possible risks and/or side effects, discomforts and inconveniences, as outlined in the Information Sheet, have been explained to me.

5. I understand that I can withdraw my child from the study at any stage and that this will not affect medical care or any other aspects of my child's relationship with this healthcare service.

6. I understand that I will receive $20 reimbursement for travel and car parking expenses to attend the sensory assessment session at the Women's & Children's Hospital.

7. I have had the opportunity to discuss taking part in this research project with a family member or friend, and have had the opportunity to have the project explained to me by the researcher over the telephone for any specific questions I may have asked.

8. I am aware that I should retain a copy of the Consent Form, when completed, and the Information Sheet.

9. I do / do not consent to my child being photographed for research purposes during the study, provided the project has the approval of the Women's & Children's Hospital Research Ethics Committee.

10. I understand that my child's information will be kept confidential as explained in the Information Sheet, except where there is a requirement by law for it to be divulged. If I have given permission for my child to be photographed, I understand that photographs of my child may be used in research publications and presentations.

11. I do / do not consent to being contacted by the investigators if my child is eligible to participate in future work arising from this study, as mentioned in the Information Sheet.

Signed: .....

Relationship to participant: .....

Full name of participant: .....

Dated: .....

Where the developmental level of the child indicates that they have the capacity to understand and consent to the study, the section below should be completed by the child.

Signed: .....

Full name of participant: .....

Dated: .....

I certify that I have explained the study to the parent and/or child and consider that he/she understands what is involved.

Signed: ..... Title: .....

Dated: .....

Appendix D

This Appendix provides an overview of the OGS games (including all game credits), and examples of real time game adaptability and data logging.

Table D1 lists all OGS project contributors in terms of the development and artwork required for each game.

Table D1 – An overview of the OGS games, with associated game credits

Game name	Brief game overview / summary and game credit
A Bridge Too Far	Similar to 'Temple Run', the main character has to navigate an endless pathway, jumping gaps and collecting gems and coins.
	Game developer: Brad Wesson
Alex Adventure	This side-scroller game has the main character, Alex, explore each level and themed landscape while collecting carrots and avoiding obstacles.
	Game developer: Mai Nassier
Alien Attack	Similar to 'Space Invaders', the player must cleanse each planet in the solar system of alien spaceships.
	Game developer: Brad Wesson
BiPlane 1922	This 3D flight simulator has the player fly over an English countryside, avoiding obstacles while navigating through farm barns. Levels are presented from different perspectives, such as the cockpit and chase-cam.
	Game developer: Martin Henschke (Artwork: Mr Sidharth Arur)
DragonFly Dodge	This side-scroller game has the main character, a dragonfly, flying over a stream and collecting coins while avoiding frogs, reeds, birds and rocks.
	Game developer: Chad Lundstrom
Driving Maniac	This vertical-scroller game has players avoiding obstacles and challenges on the road, such as cars, road works and lane changes.
	Game developer: Hamza Khaliq (Artwork: Mr Sidharth Arur)
Sunday Driver	This 3D exploratory driving game has players searching for hidden objects and avoiding enemies, before progressing to the next world.
	Game developers: Yun Chen, Da Ge, Matthew Kuckhahn, Jingyu Liu and Yongqun Yu

Marine Life	This swimming game has players attempt to move up the food chain by eating other underwater creatures while avoiding predators. Game developer: Hamza Khaliq (Artwork: Mr Sidharth Arur)
Move Gravity	This puzzle game requires players to combine multiple asteroid masses in space to form a single mass, taking into account gravitational forces and black holes. Game developer: Chad Lundstrom
Planet Fall	This action game requires players to control a laser and rocket-shooting moon lander, trying to stop meteorites from reaching the ground. Game developer: Martin Henschke
Snake	Similar to the ‘Snake’ game on Nokia mobile phones, players must move a snake around the screen, trying to eat as many objects as possible while avoiding running into themselves or the screen edges. Game developer: Chad Lundstrom
Space Stuntz	This 3D space ship simulator has players zoom through an endless tunnel of rings to score points, while avoiding asteroids and other objects. Game developer: Martin Henschke (Artwork: Mr Sidharth Arur)
Squirrel	This running game has players control a squirrel as it climbs a never ending tree, collecting objects and avoiding tree knots and branches. Game developer: Brad Wesson
Swimma	This side-scroller game requires players to control a snorkeler, collecting as many gems and air bubbles as possible, while avoiding predators. Game developer: Karnung Liang
The Fancy World	This dress-up game challenges players to suitably dress their character for a given event, such as going to the movies or the beach. Game developer: Mai Nassier

Real-time Game Adaptability Examples

Game Adaptability within Marine Life

Marine Life is a '2.5D' game (the background graphics create a sense of depth) that was initially developed by Hamza Khaliq, where the main character is a creature swimming in the ocean. To begin with, the main character has low aquatic 'status' and hence many predators that are required to be avoided, while at the same time needing to eat other marine life (prey) that are not predators.

Predators are represented graphically along the top of the screen as creatures to avoid, and the number in the top left of the screen indicates how many more creatures need to be eaten before progression to the next level. The number of lives remaining is shown in the top right of the screen, and this number reduces every time the player encounters a predator. Consequently, the aim of the game is to move up the food chain by eating a set number of prey while avoiding predators per level. There are five levels in this game.

Marine Life evolves with the player as they progress and achieve within the game because it knows how close the player is to getting to the next level (the prey count is decreasing and approaching zero). For higher levels, as the number of prey to eat reduces, the game stops generating prey and instead generates more and more predators, meaning the player must avoid all creatures on the screen and wait for prey to come along. At the highest level, not only does the amount of prey reduce and the amount of predators increase, but the speed of both prey and predator movement also increases meaning prey are harder to catch and predators are more difficult to avoid, adding to player stress and challenge. Brad Wesson coded this aspect of game adaptability into the game.

Game Adaptability within Driving Maniac

Driving Maniac, also initially developed by Hamza Khaliq, is another '2.5D' vertical-scroller racing car game that requires players to overtake slower traffic and avoid obstacles and challenges on the road, such as other cars, road works and lane changes, while collecting point bonuses such as red fuel cans and extra lives. When

players reach certain score thresholds, they progress to the next level, which corresponds with a scenery change and a mandatory speed increase. An image of *Driving Maniac* is shown in Figure D1.

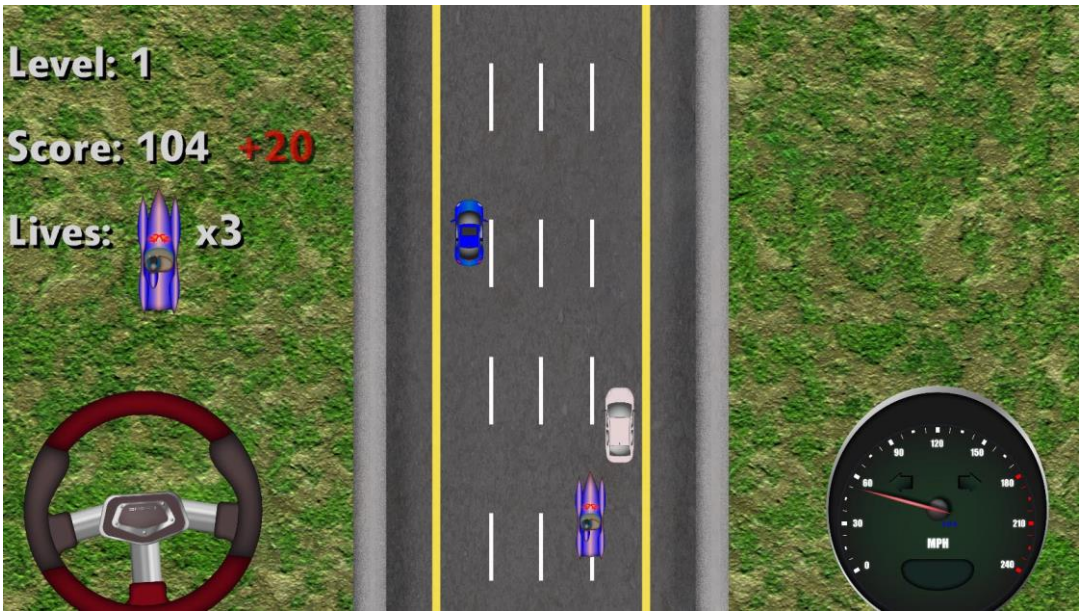

Figure D1 – A scene from *Driving Maniac*. The player's car is the purple racer (shown near the bottom of the image). (Source: Hamza Khaliq and Sidharth Arur)

Within this particular game, player progression is measured and as the player continues to achieve within the game, the other cars start to develop a form of artificial intelligence and instead of predictably driving slowly in a single lane, they begin to increase speed, pull out and overtake slower traffic – so they change lanes, sometimes erratically. When doing this they avoid each other (that is, they overtake other traffic) but don't watch out for and avoid the player's car, increasing the likelihood of a collision occurring. Martin Henschke coded this aspect of artificial intelligence into the game.

Game Variability within BiPlane 1922

BiPlane 1922, developed by Martin Henschke, was a unique game within the Games Catalogue for two reasons. The first reason relates to the fact that *BiPlane 1922* is the only 'mission' based game in the Catalogue, meaning the player can play different versions of the same game, where the player's view of the game changes depending on the mission being played. The first view is the classic 'chase camera'

or ‘chase cam’ view, where the player flies their craft from directly behind and above their biplane, as shown in Figure D2. This is the entry level for this game.

Figure D2 – The ‘chase cam’ view of *BiPlane 1922*. (Source: Martin Henschke and Sidharth Arur)

If the player chooses a different mission after the entry level, they are presented with a different view of the game. Other views include the view from the cockpit and a ‘side’ view, as shown in Figure D3.

Figure D3 – The ‘side’ view of a particular mission for *BiPlane 1922*. (Source: Martin Henschke and Sidharth Arur)

The second reason *BiPlane 1922* is unique is that it is the only game in the Catalogue that requires the player to use different controller movements to play the game based on their view. For example, when in ‘chase cam’ view, the player cannot control their plane elevation, but moving the controller left or right moves the plane left or right, respectively, and moving the controller forward or backward increases or decreases the plane speed, respectively.

However, when playing the game in ‘side’ view mode, the player has to rethink and mentally re-map the controller movements and how they correspond to game actions. In ‘side’ view mode, the player can now control their plane elevation as moving the controller backward increases the planes elevation (making it fly higher) while moving the controller forward decreases its elevation (making the plane dip towards the ground). Moving the controller right or left accelerates or decelerates the speed of the plane, respectively. While all other games use a consistent and static control mechanism to control game actions, *BiPlane 1922* requires the player to re-map how the game view and the controller relate to each other and adjust their movements accordingly. Additionally, all *BiPlane 1922* games require the player to take off and land their plane to begin and end each game, which is achieved through the ‘side’ view mode. Consequently, even when playing the entry level of this game (when flying the plane in ‘chase cam’ view) players experience the concept of different game views producing different game actions within the game.

OGS Data Logging

An example log file from the trial is shown in Figure D4. For this particular instance, ‘Guest’ played the game *Snake* (called ‘*Snake2D*’ in the log file) on the morning of the 13th October 2014. The game was played for two minutes and no vibration was delivered during the game, meaning this particular system was randomised to Group B, the non-haptic group. The left hand panel presents a summary of the game and the main panel presents the detail of what was recorded. Figure D4 shows the first 1.63 seconds of the game from the time it began, with each row representing a new log event. The first event is logged 0.39 seconds after the game starts and the second 0.11 seconds later.

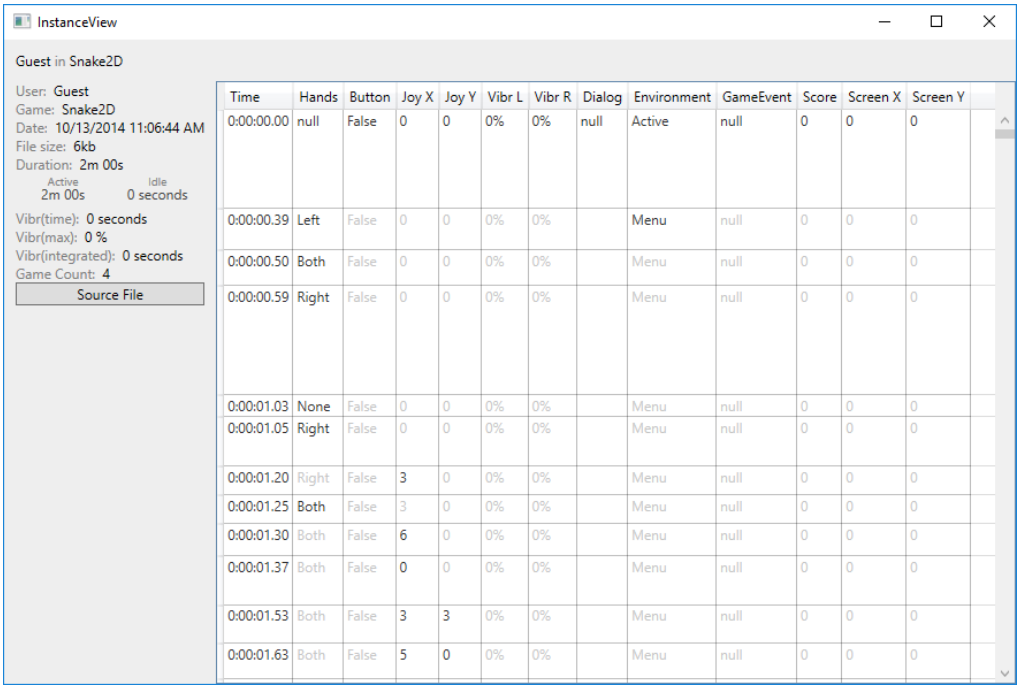

Figure D4 – An example individual log file from the trial, when 'Guest' played the game *Snake.* (Source: Chad Lundstrom and Brad Wesson)

When a log file entry appears as black text, such as row 5 (1.03 seconds into the game), with the 'Hands' column showing 'None', it means that particular item was flagged by the system. In this case, it means no hands were detected on the controller at that instance. However, first the right and then the left hand were both placed on the controller, and at 1.25 seconds into the game both hands were detected on the controller. When a particular event doesn't change during a logging event, it is greyed out because the status of that event hasn't changed. This is shown in Figure D4 as the controller is moved (shown by 'Joy X' (joystick position in the x-direction) and 'Joy Y' (joystick position in the y-position)), whereas the other events (such as 'Hands' and 'Button') do not change, and remain greyed out.

Appendix E

This Appendix contains the '*Participant Evaluation Form*' that was used for the games evaluation with typically developing children, Chapter 5, section 5.3.1.

PARTICIPANT EVALUATION FORM – Background

User Evaluation of Custom Computer Games

Age: _____ **Gender:** _____

A1. Do you play computer games? If yes, how often? (How many times per day, week, etc)

A2. Do you prefer single player or multi-player games?

A3. What 'platform' do you typically play games on? (You can choose more than 1 response)

- Console (Xbox, PS3, Wii) – which one? _____
- PC – which one? _____
- Handheld (DS, PSP, iPod) – which one? _____
- Tablet (iPad, Slate) – which one? _____
- Other _____

A4. What type (genre) of games do you like to play? (You can choose more than 1 response)

- First Person Shooter (Halo)
- Education (Math and Language Games)
- Creative (Designing an avatar)
- Racing (Mario Kart)
- Sports (Wii Sports)
- Online Community (Club Penguin)
- Platform (Super Mario Bros)
- Other _____

A5. Please list a few of your favourite games (the games you play most often):

_____

_____

_____

PARTICIPANT EVALUATION FORM – Game Questions

User Evaluation of Custom Computer Games

Game name: _____

B1. Did you enjoy playing this particular game? (Yes/No)

B2 Did you need to read the instructions before playing this game? (Yes/No)

B3. For this particular game, what do you think about: (please tick your responses below)

Feature ↓ Rating →	***Not good***	***Average***	***Good***
a. The colours used			
b. The animations			
c. The sounds			
d. The controls			
e. The appropriateness of the vibration			

B4. Using a scale from 1-10 (1 = "very poor", 10 = "very interesting"), how would you rate your ***interest level*** in this game?

B5. If this game was commercially released, would you consider buying it? Why or why not?

B6. Are there any features that you would like to be included in this game? If yes, what are they?

B7. Are there any parts of the game you did not like? If yes, what are they?

B8. Using a scale from 1-10 (1 = "very easy", 10 = "very difficult"), how would you rate the ***difficulty level*** of the game?

B9. Have you played a game similar to this before? If yes, what was the name of the game?

B10. Did the game scoring make sense? (Yes/No)

B11. Would you play this game again to try and beat your previous score? (Yes/No)

B12. Do you have any further comments to make about this game?

B13. Using a scale from 1-10, what score would you give this game? (1 = very poor, 10 = brilliant)

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Appendix F

This Appendix contains an overview of the alternative accessible controller designs that were developed for the Stage 1, Initial Pilot Project (Chapter 5, section 5.4.1).

Alternative Controller Designs (from section 5.4.1)

Controller Design 1 – Tom Whitby

Everyday computer peripherals was the inspiration for Controller Design 1 by Tom Whitby. Shown in Figure F1, this design leveraged the fact that the overall system was going to be deployed via a laptop computer, and that a common computer peripheral is a mouse. Hence, Tom conceptualised using everyday computer 'mice' to provide visual coupling and a sense of computer familiarity back to his design.

(a) **(b)**

Figure F1 – Controller Design 1: (a) CAD model; and (b) Working prototype (Sources: Tom Whitby)

This design was based on a sliding top plate that sat on a circular base, and it was this x-y planar top plate movement relative to the base that controlled the game character. The top plate included two 'mice' as handle holds, as shown in Figure F1. To use the controller, the player rested their hands on the two mice hand holds and slid the top plate forward, backward, left or right with respect to the base, to control the game character. Motors were mounted inside each hand hold on the top plate of the controller, directly under where the player would place their hands during use.

Controller Design 1 Assessment

This particular controller design was appealing and had the following positive aspects:

- A brightly coloured and pleasant design form that intuitively indicated where the player needed to place their hands;
- Good to very good haptic vibration isolation delivered via the motors mounted within each mouse hand hold;
- Excellent stability in all directions – this particular design had the second most stable base of all four pilot controllers, with the controller base having rubber feet that eliminated sliding of the base on the surface it was resting on;
- Provision for extra in-game buttons to be included on the mice themselves, as indicated by the white buttons on each mouse (shown in Figure F1); and
- The form of the mouse hold provided good access to the palm and fingers of the player to maximise afferent haptic stimulation.

The drawbacks of this particular design were:

- The out-of-game button, although conveniently located (close to the player) and contrasted well (red on white), wasn't proud;
- The control mechanism – the planar sliding of the top plate over the bottom base – wasn't immediately intuitive, despite the mechanism developed to enable this to happen being smooth. The movement was potentially problematic when it didn't work smoothly because of friction between the two sliding surfaces;
- The controller did not self-centre after any of the sliding movements, meaning the player had to do this manually; and
- Similar to the preferred controller design, this design could also be used with one hand placed on the centre of the top plate, not engaging both hands.

This particular design had many positive features – namely, form appeal, the way that it encouraged open handed use via the mouse holds, a very stable base, provision for easily accessible in-game buttons (should this functionality be required), and intuitive hand positioning. Along with the preferred design, this design was also short-listed to progress to the next stage of the project.

Controller Design 2 – Tom Askham

Tom Askham's controller design met the brief, was visually appealing, and offered the player flexibility in terms of control options. Shown in Figure F2, this particular concept took inspiration from a combination of spaghetti noodles and multi-positional camera stands. It encouraged the player to grip a red 'noodle' arm in each hand to control their game character and vibration feedback was delivered via a motor mounted in the end of each noodle (furthest from the white base).

(a) **(b)**

Figure F2 – Controller Design 2 (Tom Askham): (a) CAD model, and (b) Working prototype (Sources: Tom Askham)

The novelty related to this particular controller is that each 'noodle' arm can be flexed or bent into a different form, either a 'traditional V' position, shown in Figure F2(a), or a custom position of the player's choosing, shown in Figure F2(b). This afforded many different interface configurations for the player, with the main positional limitation being the length of each noodle arm.

Controller Design 2 Assessment

This controller design was appealing and had many positive aspects, particularly:

- A very simple and intuitive design, with an easily accessible and proud contrasting blue out-of-game button for menu activation and game selection;
- A high aesthetic form factor;
- Excellent functionality – easy to use and manipulate to control the game character by moving and pivoting the ‘noodle’ arms as required;
- The controller self-centred when released, which is a desirable feature and akin to how commercial joysticks and controllers function;
- Flexible hand position – either high up on each ‘noodle’ or low down near the base, depending on player comfort;
- Easy to adjust each ‘noodle’ arm to a new position;
- Excellent vibration isolation between each ‘noodle’ arm; and
- Good stability in the anterior-posterior direction (for forward and backward movements).

The following drawbacks to this particular design were observed and noted:

- Each ‘noodle’ arm required the player to be able to grip it to use the controller, so children with poor or no grip would find the ‘noodle’ arm difficult to use;
- While the base could be altered in a future design iteration, the design as is has poor sideways stability (left and right movements);
- While a novelty of this particular design was the flexibility of the ‘noodle’ arms, this also meant that the player’s hand position couldn’t be controlled when on trial, as shown in Figure F3(a) and (b). If the player manipulates the controller into an abnormal configuration, similar to those shown in Figure F3, then neither the fingers nor palms of the player are in contact with the controller surface (the left hand for figure (a) and both hands for figure (b)). This aspect doesn’t meet one of the design brief requirements (the requirement to maximise afferent haptic stimulation to the child’s palms and fingers); and
- The current design doesn’t require or encourage bimanual use – a player can use their dominant hand to grip and hold just one ‘noodle’ arm only, or manipulate the arms to form a shape that their dominant hand can control.

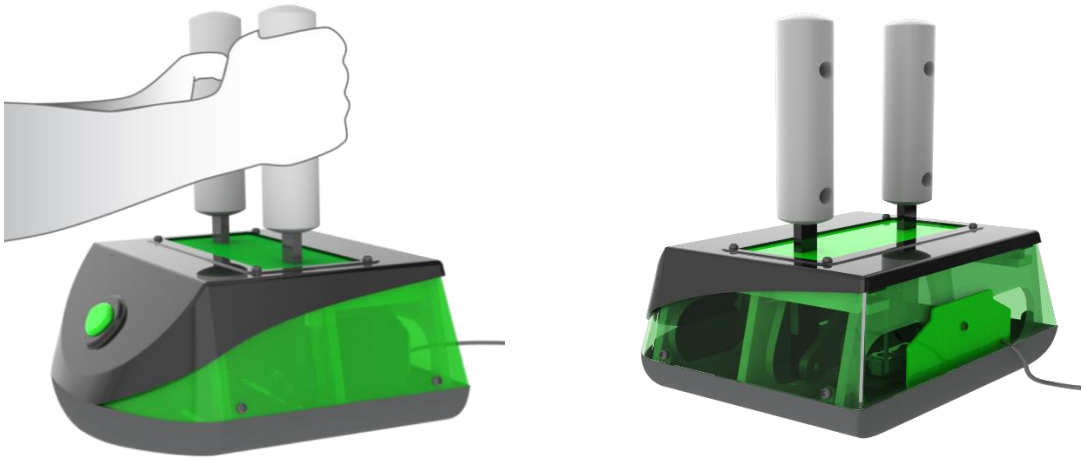
(a) **(b)**

Figure F3 – The flexibility of Controller Design 2, with two different interface configurations shown in (a) and (b) (Sources: Tom Askham)

While this design option was novel and impressive, and met many of the design requirements, it wasn't chosen to progress to the next stage of the project.

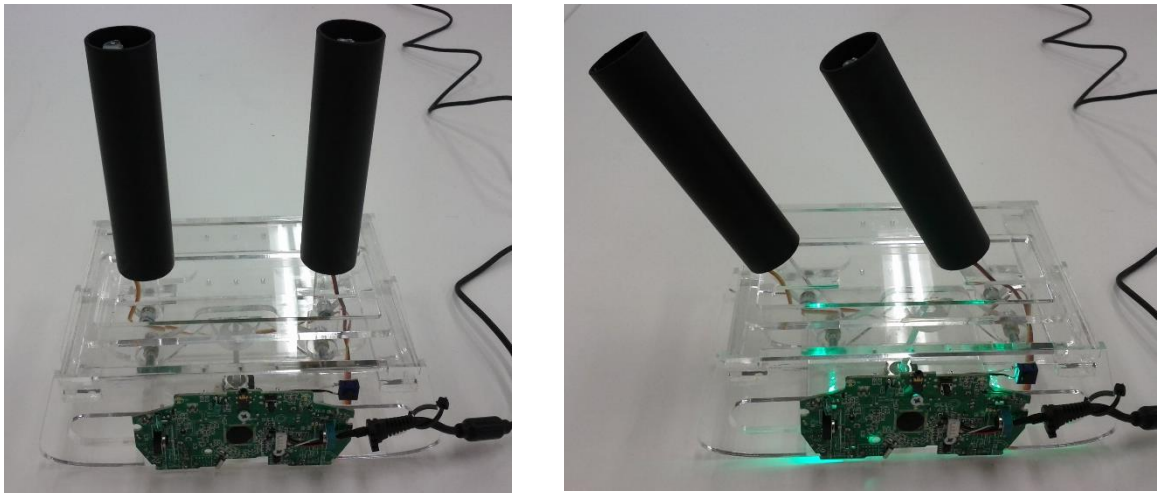
Controller Design 3 – James French

James French's controller design also met the design brief and adopted a different control mechanic compared to other designs. Shown in Figure F4, this design encouraged the player to grip two upright rods or joysticks to control their game character. Each rod pivoted in tandem at the base to enable sideways movement, and forward/backward movement was achieved by sliding the rods in tandem forward or backward. That is, the rods were moved differently depending on the movement desired: pivoting for sideways movement or sliding for forward/backward movement. The sideways movement of the rods is shown in Figure F5(b). Vibration was delivered via motors mounted in the end of each rod. James' inspiration for his controller design came from the Microsoft's 'Afterglow' Xbox controller, which is translucent and uses internal LEDs to create a 'glow' affect within the controller. One version of the Afterglow controllers uses green LEDs, which is the same colour as the existing LED on the Xbox printed circuit board.



(a) **(b)**

Figure F4 – Two different CAD views of Controller Design 3 (James French); (a) Model showing the front of the controller and hand location; and (b) Model viewed from the rear (Sources: James French)



(a) **(b)**

Figure F5 – Two different views of the prototype for Controller Design 3 (James French); (a) Front view with controlling rods upright, and (b) Front view with controlling rods moved to the left and the controller plugged in (green LEDs on) (Sources: James French)

Controller Design 3 Assessment

This controller design was visually appealing and had some positive aspects, particularly:

- A high aesthetic form factor – the transparent base and green lighting drew inspiration from custom controllers that James identified online that had received positive commentary;
- Simple, easily accessible and proud green out-of-game button on the front of the controller base;
- Excellent vibration isolation between each rod; and
- Good to very good stability in all directions given the movements that the controller encouraged (pivoting and sliding) are actions that are concentrated over the footprint of the base and not outside of it.

The drawbacks of this particular design were:

- The movement – while the sideways tandem pivoting movement was both smooth and intuitive, the forward/backward movement wasn't. When used for the first time it seemed intuitive to pivot or bend the rods at their base to move forward and backward, which bent the rods and almost snapped them at their base. In other words, the sliding movement for forward/backward control was counter-intuitive;
- The controller did not self-centre after any of the movements (sideways or forward/backward), meaning it was extra work for the player to re-centre their controller after a particular movement. Additionally, and probably related more to the laser-cut prototype materials used, the forward/backward movement was not smooth and would stick when used; and
- Each rod required the player to be able to grip it to use the controller, so children with poor or no grip would find the rods difficult to use.

This particular design option was less impressive, less functional and not as easy to use compared to the other designs, and was not chosen to progress to the next stage of the project.

Appendix G

This Appendix contains the evaluation form that was used to collect feedback from two children with CP and their families for the Stage 2 accessible controller designs evaluation (section 5.4.2.1).

MDS4 User Testing: Cerebral Palsy Game Controller

Questions for children

1. Do you understand how to use the controller?	Yes	No
Notes:		
2. Does the controller respond as expected?	Yes	No
Notes:		
3. Is the controller comfortable?	Yes	No
Notes:		
4. Is the controller usable with one hand?	Yes	No
Notes:		
5. Does it look cool?	Yes	No
Notes:		
6. Would you want one?	Yes	No
Notes:		

Questions for parents/guardians:

1. Would you be happy with your child using this product?	Yes	No
Notes:		
2. Does your child own any video games consoles and play regularly?	Yes	No
a. Any issues?	Yes	No
Notes:		
3. How much would you be willing to pay for this controller?	$	
Notes:		

Observational:

1. Did the user clearly understand how to use the controller?	Yes	No
Notes:		
2. Was the controller misused in any way?	Yes	No
Notes:		
3. Was one hand used without prompting	Yes	No
Notes:		
4. Is the controller the 'right' size?	Yes	No
Notes:		

Appendix H

This Appendix provides further details relating to the development of the final Stage 2 RCT ‘Orby’ controller design, including the manufacture and assembly of the controller.

Final ‘Orby’ Controller Design, Manufacture and Assembly for the RCT: Additional and Extended Engineering, Design Detail and Materials Used

‘Orby’ Controller Re-design and Evaluation

A re-evaluation of the ‘Orby’ controller by the author and designer (Max Hughes) identified a number of areas for improvement, namely:

- *Oval pad design*: during the UniSA Exposition of the controllers, attendees thought the grey oval pads (used to indicate hand position and provide haptic stimulation via motors mounted on the underside of the pads) were buttons. This was because each oval pad was recessed slightly beneath the controller surface and used a ‘floating pad’ design combined with high density foam to minimise vibrations from the motor to the controller surface. Consequently, it had a little ‘give’ relative to the spherical surface when pressed. Forceful pushing on the pads caused them to snap and break indicating a weakness in the design, even though they were used inappropriately. The oval pad design was positive in terms of indicating hand position, but required re-thinking in terms of vibration motor positioning;
- *Haptic feedback*: the haptic isolation between the right and left side was better from first to second prototype, but could be improved and required re-evaluation given the need to re-think where the vibration motors were to be positioned;
- *Proximity sensors*: Design 1’s use of recessed proximity sensors to detect hand position proved insightful and this concept was to be incorporated into the ‘Orby’ re-design for the trial;
- *Surface texture*: the surface of both Stage 2 prototype controllers was smooth to touch, yet the end users would be children with a tactile sensory impairment. It was decided that a textured surface, particularly the oval pads where the hands are placed, to provide passive afferent feedback to the child during use would be a good idea;

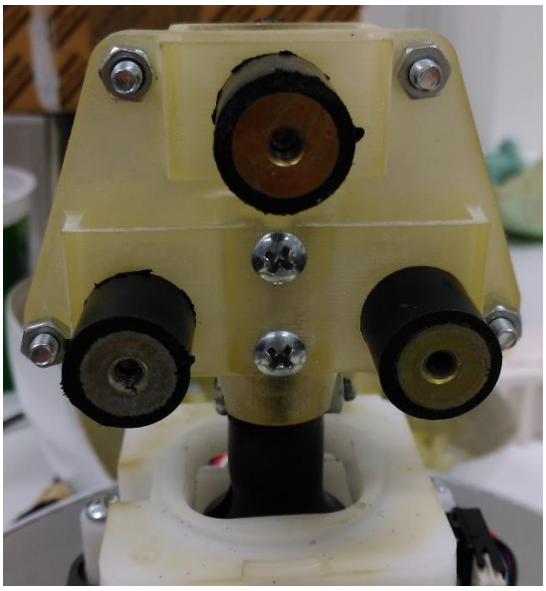
- *LED lighting*: an element of one of the alternative controller designs (Appendix F, Design 3, James French) was LED lighting. Lighting was to be incorporated into the final design, to provide visual confirmation that the controller is plugged in and ready to go and to augment the design aesthetic;
- *ND hand support*: the support/strapping design for the ND hand could be improved, mainly from an aesthetic perspective; and
- *Component access*: one of the early desirable features of the 'Orby' controller design was that the hemispheres were fastened and positively locked during use to the main central joystick piece using strong rare earth magnets. This meant that access to the *Xbox* board, motors or any internal componentry was tool-less, quick and easy, while also being non-obvious (that is, users didn't realise the controller 'internals' could be accessed in this way). This feature was to be retained for the trial in case 'on the run' maintenance needed to be performed during the trial.

The grant funding enabled the designer to be employed part-time as a Research Assistant on the project and provided a specific budget to manufacture the controller through a short production run. The following design and functional changes were incorporated into the final controller design:

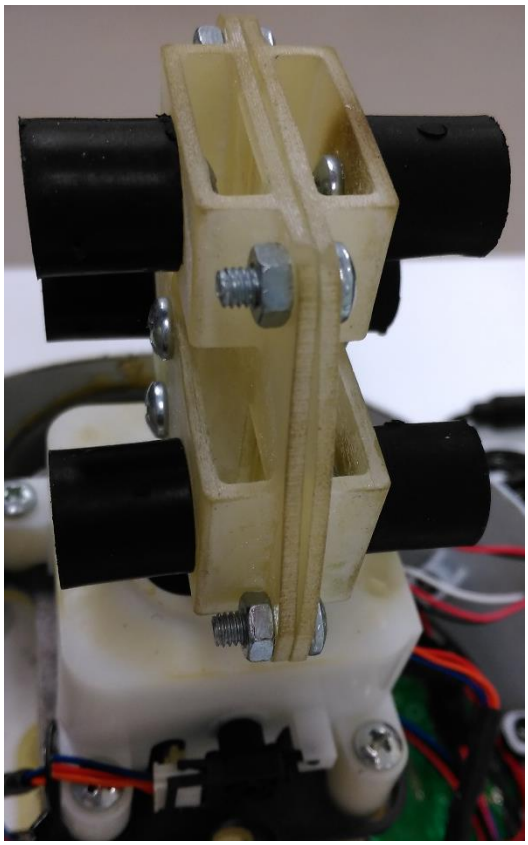
Improved Haptic Isolation

The Stage 2 'Orby' controller used high density foam and a 'floating pad' design for the oval pads to minimise vibrations across the controller to ensure the left and right sides were isolated from each other. The major shortcoming of this particular design was the suspension mechanism for the oval pad, which proved too delicate and snapped when used during gameplay by the public during the University Exposition night. Even though the 'users' on this night were adults and not children and were using the controller inappropriately (they mistook the pads for buttons), it highlighted a weakness in the design given the system was going to be unsupervised in the family home when eventually deployed.

Working with A/Prof Sandy Walker and an industry-based Industrial Design mentor, incorporating commercial vibration dampeners or vibration mounts was suggested as a way to dampen vibration from one side of the controller to the other, as well as increasing the path of travel for the vibration from one side to the other. The designer obtained some mounts for testing and was able to quickly prototype and configure a way for accommodating them within the space underneath the hemispherical domes of the controller. Three cylindrical mounts in a triangular configuration were connected to each side of the central joystick mount (six mounts in total per controller), as shown in Figure H1(a) and (b).



(a)



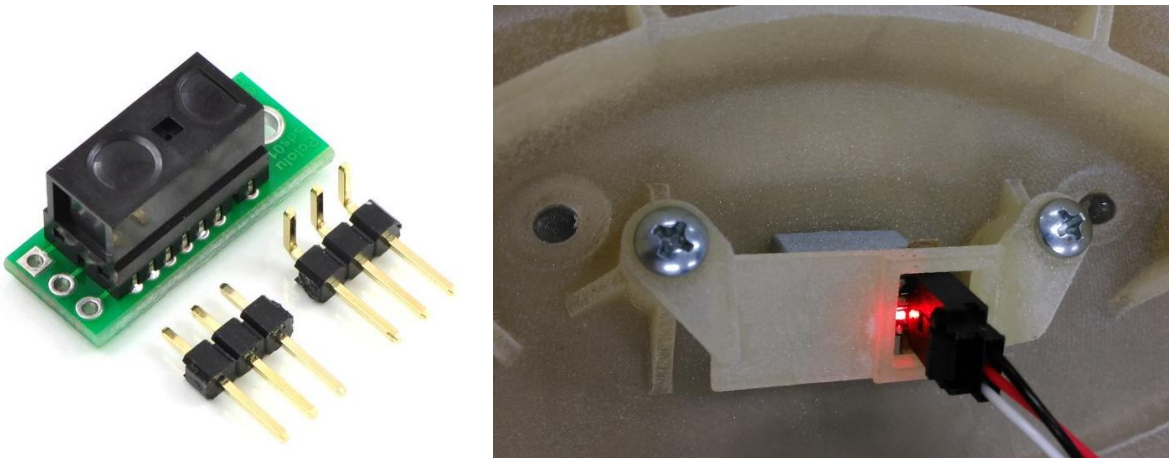
(b)

Figure H1 – The configuration of the three vibration mounts on each side of the joystick mounting to dampen vibration from one side of the controller to the other; (a) Side view; (b) Rear view

The cylindrical mounts (FIBET, 1615DD04-45) were made from natural rubber and zinc plated steel with dual female M4 threads on each end, 15mm high, 16mm diameter, with a compression load of 14.1kg and were used commercially in vibration isolation applications.

Hand Detection via Proximity Sensors

A low profile distance measuring sensor (*Sharp*, GP2Y0D805Z0F), which uses both a photo diode and infrared emitting diode and has proximity sensor applications, was used for hand detection. The sensor was recessed 10mm into the grey oval pad inside the controller, due to the operating range being 5-50mm and the need to ensure that a hand on the controller surface would always be detected and not below the detection threshold of the sensor. The sensor was powered by connecting it to the 5V supply on the Xbox controller board. Figure H2(a) shows the sensor and Figure H2(b) shows the mounting location within the underside of the controller. The red light, not seen during use, indicates that an object has been detected.



(a) **(b)**

Figure H2 – (a) The *Sharp* proximity sensor used to detect hand position (Source: https://littlebirdelectronics.com.au/products/pololu-carrier-with-sharp-gp2y0d805z0f-digital-distance-sensor-5cm); and (b) Mounted within the underside of the controller

From an integration perspective, the sensors were wired directly onto the *Xbox 360* board, to the location normally reserved for the left and right ‘bumper’ buttons (labelled ‘5’ and ‘10’ in Chapter 5, Figure 5-1). When the sensors were covered and the player’s hands were in the correct position, this was equivalent to a player holding down both bumper buttons when using a standard *Xbox 360* controller – a virtual button press. From a design perspective, integrating the sensors in this way made use of existing inputs on the Xbox board that weren’t being used. Consequently, the overall Central Games Catalogue, which was responsible for

monitoring hand position, was interrogating bumper button switch presses when monitoring hand position.

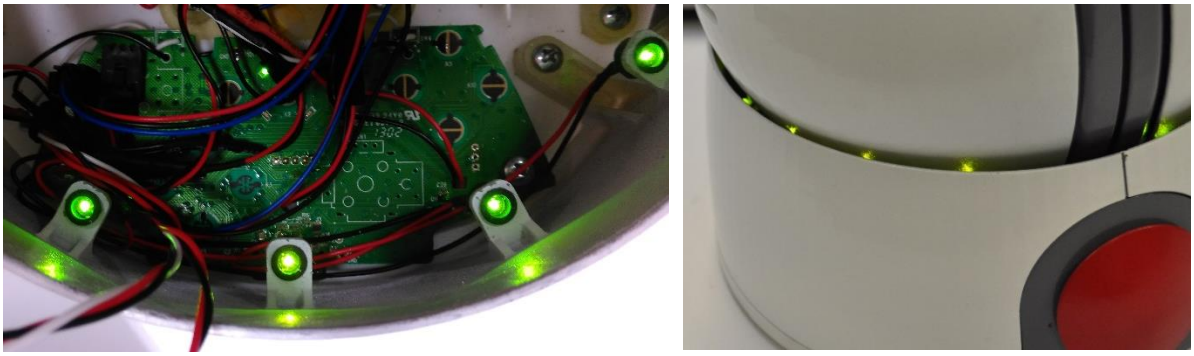
Use of LED Lighting

Lighting is implemented in almost all powered devices to indicate power status, providing visual confirmation that the device is on, off, charging or in stand-by mode. One of the desirable but non-essential requirements of the controller was for it to incorporate lighting to provide visual feedback to the player. This was mentioned during the very early Stage 1 projects, but the emphasis at that stage of the project was on a functional, accessible design, knowing that lighting could be incorporated when a final design was chosen and when time allowed. The exception to this process was one of the alternative controller designs (Design 3), which was the only design to investigate different ways to illuminate the controller (Appendix F).

The final ‘Orby’ design has a gap between the moveable spherical ‘orb’ and the static circular shroud around the base. The gap was small (typically 1-5mm) and allowed the ‘orb’ to freely rotate without interfering with the base. After exploring different design options to: (a) illuminate the controller (fibre-optic lighting vs. LED lighting) and (b) to provide visual feedback to the player during gameplay (confirmation the controller is connected vs. visual feedback that would reinforce game activity), due to time and budget constraints simple LED lighting was chosen. One of the supervisors (SH) advised that providing different lighting effects to reinforce game play through the controller would be distracting for the child rather than reinforcing, so this idea was rejected.

To avoid a clash of colours, wide angle (130º) round green LEDs were chosen for lighting, to match the existing small green LED that indicates power on the standard Xbox board (visible near the top of Figure H3(a)), and powered via direct connection to the Xbox board. The LEDs were mounted to the inside of the circular shroud, four per side, positioned towards the front of the controller (Figure H3(a)), with the intention of projecting light onto the bottom of the ‘orb’ that sits within the base during use (Figure H3(b)). In this way, the lighting served to provide visual confirmation that

the controller was plugged in and ready for use as well as an element of visual aesthetic.



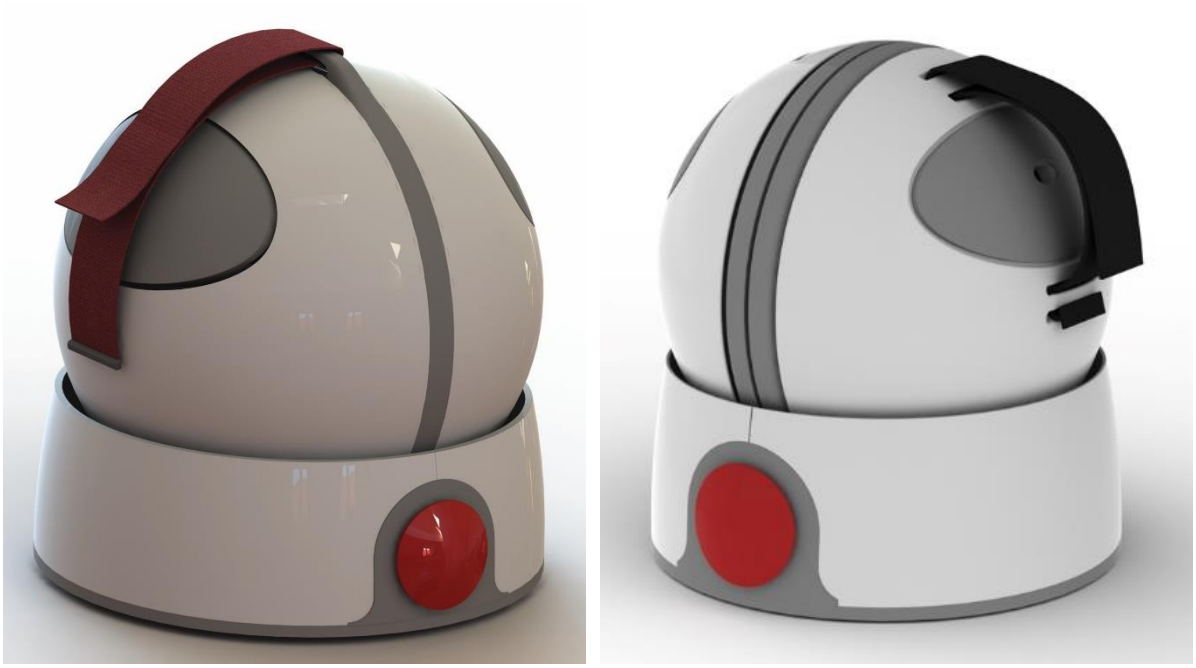
(a) **(b)**

Figure H3 – The four green LEDs that were used to illuminate the 'Orby' controller as viewed (a) From inside the controller base during assembly; and (b) When fully assembled

ND Hand Support / Strap

The intention of the ND hand support or strap was to provide *assistance* to the ND hand to position it onto the spherical surface. The strap was not intended to tightly secure or pin the hand to the controller surface, as this would encourage passivity rather than active attention or use of the ND hand. That is, the child would be more likely to ignore their ND hand if they felt if it was tightly secured, which would be counterproductive to requiring the child to attend to their ND hand during gameplay.

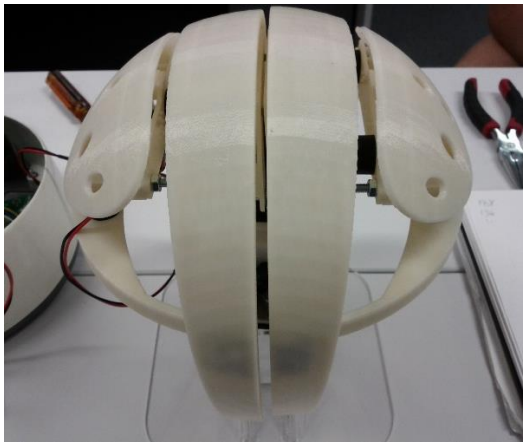
The original Stage 2 'Orby' controller used a hand strapping design that was pinned at the base, beneath the oval pad, looped through a central slot on top of the controller, and then folded back on itself and secured with Velcro, as shown in Figure H4(a). The final 'Orby' controller used a very similar strap design except the tethering and restraining mechanism now used grooved slots that the strap slid through, keeping the strap and it's attachment points to a given hemisphere, as shown in Figure H4(b). The straps were custom-made by a local orthotics and prosthetics workshop (Orthotics and Prosthetics South Australia, Daw Park). The intention of both designs was that once the base of the strap was fitted to the controller, the other end of the strap could be independently looped and folded back on itself by the child using their dominant hand.



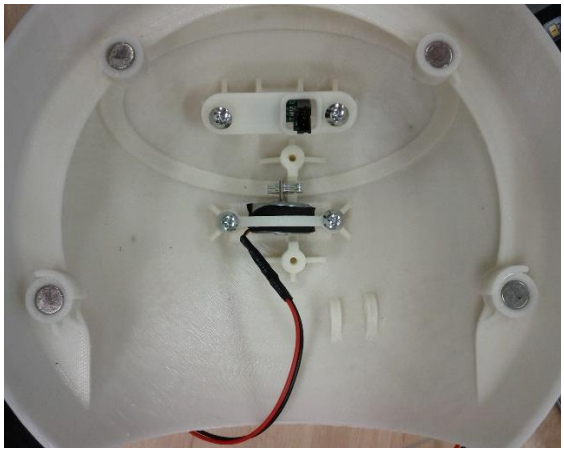
(a) **(b)**

Figure H4 – (a) The CAD model of the original 'Orby' controller strap design, and (b) a CAD model of the final 'Orby' controller strap design (Sources: Max Hughes)

Throughout the final stages, each design change was prototyped and tested (in isolation and combination) using the rapid prototyping and 3D printing facilities at the UniSA. This was done to ensure each design element that was changed, and the final overall design, was appropriate, had the correct fit and tolerance, and ready for small scale manufacture. Figure H5 shows some of the steps undertaken during this process, with respect to (a) vibration mount testing, (b) haptic motor and proximity sensor location, (c) the new ND hand strapping slots, fit and use, and (d) a complete assembled test unit.



(a)



(b)

(c)

(d)

Figure H5 – Stage 3 prototyping and testing prior to manufacture; (a) Vibration mount testing; (b) Location and orientation of the proximity sensor and haptic motor; (c) Testing the new strapping mechanism; and (d) A fully assembled test unit

Controller Manufacture for the RCT

Once the trial controller design was finalised, five Australian companies were approached for quotes to manufacture the 21 parts required for each controller, with a view to producing approximately 16 controllers in total, depending on budget constraints. Having access to multiple controllers was planned to allow parallel trials to occur and to cater for potential breakages, where the author could swap a broken controller for a new one once notified of the breakage. Consequently, all five companies (three local and two interstate) were asked to quote on parts for 16 controller units.

Four companies replied to the request for quote, and prices ranged from AUD$15,886 to AUD$27,231 (exclusive of GST). One local Adelaide-based company (*Ellex Precise*, Gillman, South Australia) provided greater insight to the project after initial contact and demonstrated a deeper understanding of what was required, as well as a willingness to collaborate for this project compared to the other companies, and was chosen as the preferred supplier. *Ellex Precise's* quote was AUD$19,840 (ex. GST), which included painting the components on the exterior of 'Orby' and custom texturing each oval pad, as described earlier. The base for each controller was a circular piece of 6mm thick acrylic (210mm diameter) that was laser cut and etched. Given the relationship that was forming with *Ellex Precise*, they were also approached to quote on this part of the componentry required. Another local company was approached to laser cut two mild steel spacers that were required to add strength and height to the *Logitech* 'Attack 3' joystick module and stand within the controller.

Ellex Precise advised that 3D printing the components was going to be the best way to conduct this short production run and used a *PROJET HD 3500 Plus 3D* printer to print all components. Owing to the number of components that needed to be printed per controller, the 21 components had to be laid out on three separate print beds, meaning it took almost 48 hours to print all the components for a single controller, including cooling time, prior to painting the necessary components. Consequently, the components were delivered, inspected, and rejected (if faulty, warped or had a poor finish) or accepted and assembled as they arrived over a period of eight weeks, typically with parts for two complete controller units being delivered each week.

All the electronics components for the project were sourced through the Engineering Services Group at Flinders University, owing to their ability to leverage discounts with preferred suppliers (if grouped in a large order), quickly compare prices and volumes with different national suppliers, and obtain quick delivery of components. The bill of materials for the controller electronics totalled $2,083 (incl. GST) for all components. The largest contribution to this cost was the proprietary components required for the controller, namely:

- 32 *Microsoft* Xbox controllers (to provide 16 printed-circuit boards and 32 large vibration motors, two for each controller), and

- 16 *Logitech* Attack 3 joysticks (for the joystick module to provide movement).

Together the two proprietary items contributed $1,520, representing 73% of the total electronics cost. The overall cost and per 'Orby' controller cost for the major components are shown in Table H1, inclusive of GST.

Table H1– A breakdown of the major component costs for the 'Orby' controller

Item	***Total Cost (n=16) (incl. GST)***	***Cost per 'Orby' Controller (incl. GST)***
3D printed components	$21,824	$1,364
Electronics components	$2,083	$130
Vibration mounts	$499	$31
Laser cut & etched 6mm acrylic base	$352	$22
1.2mm & 3mm mild steel spacers	$126	$8
Overall 'Orby' controller	***$24,884***	***$1,555***

Note: incl. = inclusive; GST =10% in Australia; mm = millimetres

Controller Assembly

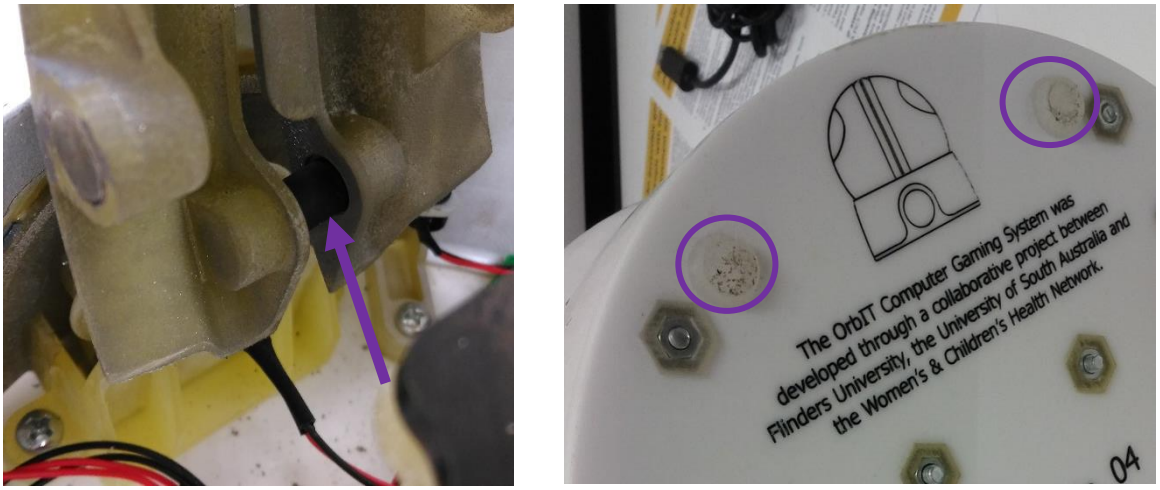
As mentioned earlier, the controllers were assembled as the 3D printed componentry was delivered, inspected, and accepted. This was done primarily by the designer with assistance from the author. Each controller required a number of fasteners and other componentry for assembly, as detailed in Table H2, and described below.

A small piece of black rubber cord (approx. 25mm in length) was used to prevent the ends of the grey hemisphere connectors knocking or 'chattering' during vibration events. The noise was heard during final testing and the cord served to hold the ends apart to prevent them touching each other, as shown in Figure H6(a). Six transparent rubber feet were glued to the controller base to provide support, friction and stability when the controller was being used on a chosen surface, as shown in Figure H6(b).

Table H2 – A breakdown of the components required to assemble the 'Orby' controller

Component	*Number per Controller*
Fasteners	
M4 (6mm)	*12*
M4 (10mm)	*4*
M4 (16mm)	*15*
M4 (25mm)	*6*
M4 nuts	*25*
Self-tappers	*8*
Vibration mounts	6
Rubber feet	6
Raw earth magnets	16
Rubber cord	2
Mild steel joystick spacers x 2 (1.2mm & 3mm)	1

Notes: mm = millimetres



(a) **(b)**

Figure H6 – (a) The black rubber cord (indicated by the purple arrow) used to separate the ends of the grey hemispherical connectors to prevent them knocking during vibration events; and (b) The rubber feet (circled in purple) glued to the base of the controller

Appendix I

This Appendix contains the modified study protocol that was read to all children who participated in the Stage 2 RCT, after the switch from the '*Original Games Catalogue*' to the '*Incentivised Games Catalogue*'. The main change to the study protocol involves the explanation that some of the games are locked and how games can be unlocked, towards the end of the protocol.

Study Protocol Document:

Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial

Hi, my name is David and I work at Flinders University. Thank you for agreeing to be a part of our computer gaming trial – we really appreciate your involvement.

Because this is a proper scientific study, I'm going to give you a demonstration and overview of the system and read the following instructions to you and your <mother/father>. This way every child gets the same introduction from me. If you have any questions, please ask me. I want you to have fun and enjoy using it over the next 6 weeks.

Firstly, this is your new computer gaming system – it consists of a computer (*touch the laptop*) and a specially-designed controller (*touch the controller*) that we call 'Orby'.

The system is very easy to use – let me show you. To start it, all you do is open the laptop and press the 'on' button. This turns the computer on and the gaming system automatically starts, as you can see (*games system will load up in the background*).

To use the system and play the games, rather than use a small and fiddly X-Box controller, all you need to do is use this new controller we've made for you. It's very easy to use – you place your hands here on the oval pads to use it (*demonstrate by putting hands on the oval pads*), and move the controller in one of 4 directions to play any of the games: forward, backward, left or right.

As you can see, the gaming system has just loaded for us (*point to laptop screen*). The first thing that you need to do, and you need to do this **every time**, is log in. This is where you choose your name, shown here on the left (point to their name). To log in, just press this big red button on the front of the controller (*point to the button*).

If your friends or family want to play one of the games, that is absolutely fine and we encourage it. However, you need to ask them to **always** log in using the 'guest' name *(point to the guest name on the screen).*

So, if you're playing the system, always log in with *your name*, but if someone else plays the system, they need to use the *'guest' log in*. Please don't let anyone log in with your name. For now, I'm going to log in using this 'demo' name so I can show you how to use the system *(move the controller to the 'demo' log in)*.

Do you want to press the big red button to log me in? *(Child presses button)*.

Ok, as you can see we're now logged in. Look at all these games – every cube or box that you can see represents a game; there are 15 of them in all. Let me demonstrate how some of the work and then you can explore the rest – how does that sound? Are you ready to see what we've made for you?

The first game I would like to show you is called 'Alex Adventure'. To choose this game I need to move across until the 'Alex Adventure' box is highlighted – you can

see the box is now spinning around and has a glow around it. This means it's been selected.

At the same time, you can see over here on the right hand side, on what we call the smartphone, that a video is showing you a preview of the game (*point to the smartphone and the video*). So every time you move through the games menu, you can look on the smartphone for a preview, to see what the game looks like.

Once you play a game, the bottom part of the smartphone (*point to the bottom part*) will show you the high scores from playing that particular game, and the name next to it is the person who has that high score. Hopefully when the trial finishes you have lots and lots of high scores here for all the games!

Let's now play the game. Do you want to press the red button for me? Thank you.

With all our games the idea is that you don't need a button to play any of them – all you need to do is move the controller forward, backward, left or right to move your character in the game. So as long as you keep your hands on the controller, you're ready to play.

With this particular game, there is an introductory story that you can read to understand the background to the game. I'll let you read through the game story at another time, but I can show you how to play this game.

The aim of the game is to collect as many carrots as you can, while jumping over or ducking under obstacles. To jump, because there is no button, all you do is move forward or 'up' – and you can see that Alex jumps (*demonstrate this*). To move right, you move the controller right, to move left, you move the controller left – what do you think you do to duck under objects? *(See if child knows they need to move the controller down to duck, but don't let them wait more than a few seconds if they can't answer).*

Let me show you the first level (*demonstrate first level*). As you can see Alex, celebrates by dancing a jig at the end if you win the level.

Let me show you what happens if you haven't collected any carrots, and you run into an enemy *(purposefully run into an enemy to show the child what happens when the game ends).*

As you can see, when the game ends, the next screen you see is the log-in screen again. This will happen every time, so you always have to select your name each time so the computer knows who is playing the games.

Now, the next game I want to show you is 'Driving Maniac'. This one is obviously a car racing game. Do you want to press the red button for me to start the game? Thank you.

As you can see, this game starts straight away. The aim of this game is to drive as far as you can while avoiding all the other cars and road works on the road. Every now and then you'll see objects on the road, like fuel cans and extra lives, which you

should collect by running over them. Watch what happens if I take my hands off the controller *(take hands off controller – wait for 3 seconds and see that the system stops and provides a pop-up telling me to put my hands back on).*

Do you see that? The system knows I've taken my hands off the controller, and it's stopped the game and is telling me to put my hands back on. This will happen for every game, so you need to make sure that you always have both hands on the controller or else the games will keep stopping, which can get annoying.

Look what happens when I put my hands back on the controller – see the arrows are going away and the system says 'thank you' as it knows my hands are in place and that I'm ready to play (*demonstrate this feature of the system*). The system also counts me back into the game, so you have time to get ready before the game restarts.

If you ever want to pause the game you are playing, all you have to do is push the red button (*demonstrate this*) – and this menu pops up. To ***restart***, just select the 'resume' function (*point to this*) or to ***exit*** the game completely, select exit (*point to this*). You can also adjust the ***volume*** of the system from this menu as well. Or, as we've just seen, you can also pause the game by taking your hands off the controller.

Let me show you a little bit more of this game *(demonstrate 'Driving Maniac' for a little longer – deliberately crash the car to end the game).*

Here we are back at the log-in screen, and I'll select my 'demo' name again.

This time, let me show you a few other things. If you select this yellow icon here (*point to the top left yellow icon*) you can shut down or turn off the system and also change a few things (*select the yellow icon*). Notice that the smartphone will tell you what each icon does each time you move from icon to icon, so if you want to know what anything does, just look at the smartphone for more information *(point to the information)*.

You can change the background colour for the system here (*demonstrate this*) and also the system volume, in case things get too noisy and your parents want you to turn the volume down *(demonstrate this).*

If you want to find out who made the gaming system for you, then select this icon here (*point to the relevant icon*), and you can see all the names of the people who helped design, build, supervise and make the games and the controller, as well as who funded the project.

Lastly, when you've finished playing with the system for the day, or if you need to shut the system down, you just need to select this icon here (*point to the shutdown icon*). This will automatically log you out and turn off the whole system. Then all you have to do is close the computer lid, and the system is packed away.

Let me get out of this menu by selecting the blue 'back' arrow, and here we are back at the main games menu. There are two more games I'd like to quickly show you, and then it's your turn!

The next game to show you is 'Fancy World' (*point to the 'Fancy World' game cube and select it*) – this is a dress up game. I want to show you this game as it has a slightly different way of playing, because you need to select an item from your wardrobe to wear, but you don't have a button to push in the game!

I'll select the man character from the options presented and you can see that I am presented with an invitation to an event – such as going for a walk, going to the movies, etc. In this case, the scenario is <*highlight scenario*>.

Once I'm in the game I can start to dress my character. I move my controller left or right to move along my wardrobe items, and to select the item I want all I have to do is push the controller up or forward (*demonstrate this*). So it's left or right to move through the options, and up or forward to select what you want.

To cancel an item you select this red cancel icon (*point to the red cancel icon*) and to go back to your wardrobe you select the blue back arrow (*point to this as well*). Let me finish dressing my character and I'll show you one last game.

(Finish playing 'Fancy World', while talking the child through my decisions and showing them what I am doing, then log back in to the games menu).

Ok, the last game to show you is called 'Space Stuntz'. This is a 3D flying spaceship game (*move down to the 'Space Stuntz' cube*) – do you want to push the button for me to select it? The aim of this game is to fly as far as you can in space and to fly through as many rings as possible. If you miss a ring, you lose a point, and if you miss 10 rings the game is over. You can read about how to play the game from the start menu here.

You also have to look after your spaceship and the protective shield around it, and avoid asteroids and ice storms that damage your craft. Here's where the game instructions are (*move controller left and right*) and now let's start the game (*press the red button*).

(Play the game, but deliberately miss rings or fly into objects).

So – you have 15 games here to play and explore, and you use Orby to control all the games and to move within the menu. Now I think it's your turn. Let me log in for the last time and go into the settings so I can start your trial (*log in, select the settings icon, then select the 'Start Trial' button. This logs me out and takes away the 'demo' profile*).

However, there is one more thing to tell you. When you first start the system you can only play 5 of the games – **the rest are locked.** This means you can't play them right away.

However, unlocking the games is easy! All you have to do to unlock a game is to play the other games for at least 30 minutes – and every 30 minutes a new game is unlocked. Does that make sense *(pause to see if there are any questions)*. The system keeps track of how long you play for so knows when to unlock the games for you – this is automatic. I'll show you how this works when you log in.

Here you go – let me move out of the way, and it's your turn. Make yourself comfortable and it's time to select your name and to log in as you. Are you ready?

Before I hand control over, just a reminder that you need to ensure that you and only you log into your name when you play the games, and that any friends and family use the guest log in. Also, because this is a scientific study, please don't discuss the specifics of your trial with other families. Do you have any final questions?

(Let them get set up and in position, and watch them log in and get started. Explain how the 'unlocking' of games works and how it looks in the main menu, and how the system keeps track of when a particular game will be unlocked. Stay for a little while (around 5-10 mins) to see how they go and to answer any questions they might have).

And one final reminder – your 6 week trial starts today. You'll see me again when I come back on _____ to collect the system from you.

However, if you have any questions or if something goes wrong, feel free to contact me. My details are here (*show them the 'Gaming System Instructions Sheet'*) and I'll also be leaving this System Overview Sheet with you as well (*point to this*). My number is also on the bottom of 'Orby' the controller.

Appendix J

This Appendix contains the independent review of the OGS by US-based charity, *AbleGamers*.

An AbleGamers Charity Review: Orby

The AbleGamers Charity

PO Box 508

Charles Town, WV 25414

Review

Overall, we greatly enjoyed using Orby and learning from it through playing the provided video games. We see great benefit for Orby in the gaming space and envision it will have even better success on the market for children and adults with Cerebral Palsy.

One major factor that especially stuck out to us was the motion required to use it, and how beneficial it is to improving motor function and muscle performance. It doesn't require that critical, specific movement that you see with some games on standard controllers, however it requires enough that it can assist with motor and sensory improvement. It allows gamers to play without the barriers of required, specific movement and button presses, all-the-while contributing to the improvement of their muscle capacity. To make something that is generally viewed as "tough and boring" for children into a fun, interactive activity, is phenomenal.

We also enjoyed the spin aspect of the controller for steering. By allowing the gamer to place their hands in what best fits their needs/most comfortable, they are able to not only play to what best fits them, but it also negates gamer fatigue, which is a common occurrence we see in gamers with disabilities. This also ties nicely into a gamer able to use their "good hand" in a position on the controller that best allows them to game. This spin aspect is also revolutionary for gamers with cerebral palsy, as it doesn't require that "gripping" required for holding a controller, moving a joystick, using the D-pad, etc.

The utilization of sensors was another component of Orby that impressed our team. By not requiring a specific measure of pressure to create an action on screen, gamers of all strength-levels can equally enjoy the experience. That is, rather than requiring a push-back pressure for how hard a gamer pushes down a button, that push-back is removed, and is graded on how close or far the hand is away. This also ties to alleviating fatigue while gaming.

The durability of Orby was also promising. We have worked with adaptive controllers that lack this needed durability, which results in hardware issues and breaking within the early stages of its life. Orby has good flexibility with sharp turns to the main component, and did not show signs of breakage or losing motion.

Lastly, and one of the most important, is the socialization aspect with gaming, and how Orby allows that entrance into the gaming world. Having the base components be an Xbox One controller is very beneficial, as it can be utilized across multiple gaming platforms through third party applications (i.e. CronusMAX). We continually see the powerful benefits to socialization in the gaming space.

One critique we found while testing was the cost of initial startup. While it was simplistic regarding the manual effort (plugging in the Orby, starting the console, etc.), we had difficulty understanding exactly what controls produced different actions*. Granted, as Orby is in the trial stage, I believe this will change. However, for most gamers with Cerebral Palsy, they will have a separate person setting up

Orby and the game, which will require a very intuitive guide and menu for them to successfully complete this. We often see these third-parties being parents, care takers, and nurses, whom may have none to limited experience with gaming.

*Note: We did not experience this when using the provided games, only the commercial games in-house.

Conclusion

Overall, Orby will be a great, and needed, addition to any gaming family and ensures that all gamers with Cerebral Palsy are included. It has the required durability to ensure it will not break prematurely, can be utilized across multiple platforms, and does not require gamers to perform demanding and exact functions that causes barriers to gaming. We recommend a comprehensive guide be included upon its opening to the market, as some families and care takers may find it difficult to setup. We generally provide an intuitive guide to families for our adaptive controllers, and have seen great success in doing so. We also recommend providing a guide on which games can best be utilized with Orby based on a gamer's level. This helps in ensuring that, if this will be the first video game someone ever plays, they have that best, first experience. This also assists families and caretakers, whom may have limited to no gaming experience, properly choose games for their children based on how much gaming experience/their skill level with Orby is.

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Appendix K

This Appendix contains the Stage 2 study documents that were mailed out to all eligible families (section 6.2.5), namely the: letter of introduction, study information sheet, and consent form.

David Hobbs, *BSc(Physics), BSc/BEng (Biomedical)(Hons)*

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CRICOS Provider No. 00114A

<Date>
<Address details>

LETTER OF INTRODUCTION

Dear <names of parents/guardians/caregivers>,

Re: Research Project "Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial"

My name is David Hobbs and I am an Associate Lecturer, PhD Candidate and Rehabilitation Engineer at Flinders University. I am writing this letter to inform you of the above research project being undertaken at the Women's & Children's Hospital (WCH). The project is being led by myself, and includes Dr Ray Russo (Director, Paediatric Rehabilitation Department, the WCH), A/Prof Susan Hillier (University of South Australia), and Prof Karen Reynolds (Flinders University). The project also involves an Occupational Therapist/Physiotherapist from the Paediatric Rehabilitation Department at the WCH.

As part of my PhD I am undertaking research in the area of computer gaming and how an accessible and interactive computer gaming system may help children with cerebral palsy as part of an alternative and novel therapy intervention.

The aim of the current project is to determine if children with cerebral palsy can have their sense of touch improved if they use and play with an interactive computer gaming system. During 2012, your child participated in the first stage of the overall project, which was a sensory assessment of how children with cerebral palsy sensed touch with their hands.

We would now like to invite your child to participate in the next stage (stage 2) of this study, which is the trialling of a new computer gaming system. This study is a world first and will provide valuable information on whether or not the sense of touch in the hands of children with cerebral palsy can be changed.

Included with this letter of introduction is an Information Sheet that describes the project in more detail, and a Consent Form for the study.

If you would like to participate in this study, please read and complete the attached consent form and return a copy to us in the reply paid envelope.

Please note that participation in the study is at all times voluntary. You can elect to cease your child's participation at any time, without giving a reason. To help with attending each sensory assessment session at the WCH, families will receive a $20 gift card at every session to assist with car parking and travel costs.

We would also like to assure you that we take confidentiality and privacy very seriously. If you decide to participate, any information you provide will be treated as highly confidential, and not be shared with any other group or individual, except where there is a legal requirement to pass on personal information to authorised third parties. This requirement is standard and applies to information collected both in research and non-research situations. Such requests to access information are rare; however, we have an obligation to inform you of this possibility.

You and your child will not be identifiable in any publication resulting from this research. If you wish to receive a summary of your child's individual results after the trial we can provide this as well as a copy of the final overall report.

The study has been given approval by the Women's and Children's Health Network (WCHN) Human Research Ethics Committee (HREC). Please contact the Secretary of the Committee (Ms Brenda Penny, Research Secretariat, phone: 8161 6521) if you wish to discuss the approval process, or have any concern or complaint.

Please do not hesitate to contact Mr. David Hobbs (phone: 8201 3167) should you have any questions or wish to discuss any part of this study.

Kind Regards,

Mr. David Hobbs
Associate Lecturer / PhD Candidate
School of Computer Science, Engineering and Mathematics
The Medical Device Research Institute (MDRI)
Flinders University

This study has been given approval by the Women's & Children's Health Network Human Research Ethics Committee, project number REC2530/12/15. If you wish to discuss the approval process, or have any concern or complaint about this study, please contact the Executive Officer of the Human Research Ethics Committee, Ms Brenda Penny, Research Secretariat, on 8161 6521.

Can children with cerebral palsy improve the way their hands feel touch if they play with an interactive computer gaming system?

(*Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial*)

Study purpose

The aim of this project is to determine if children with cerebral palsy can have their sense of touch improved if they use and play with an interactive computer gaming system, and its effect on their quality of life. During 2012, your child participated in the first stage of this overall project, which was a sensory assessment of how children with cerebral palsy sensed touch with their hands.

We would now like to invite your child to participate in the next stage of this study, which is the trialing of a new computer gaming system. This study is a world first and will provide valuable information on whether or not the sense of touch in the hands of children with cerebral palsy can be changed.

The research team

The Chief Investigator for this project is ***Mr David Hobbs*** an Associate Lecturer, PhD Candidate and Rehabilitation Engineer at Flinders University. Other investigators for this project include ***A/Prof Ray Russo*** (the Director of the Paediatric Rehabilitation Department at the Women's & Children's Hospital), ***A/Prof Susan Hillier*** (Associate Professor, University of South Australia) and ***Professor Karen Reynolds*** (Professor of Biomedical Engineering, Flinders University). This study is part of David's PhD in Biomedical Engineering.

What does the study involve?

The main part of the study involves your child playing with and using a specially developed computer gaming system. Most children with cerebral palsy find it difficult to play commercial gaming systems because of their impairment, but this system has been specifically designed to be accessible for a child with cerebral palsy.

If your child agrees to participate, the computer gaming system will be set up in your family home for a period of six weeks. David will provide training on how to use the system and will leave information with you about the gaming system and how to use it. The system comes with a number of fun, age appropriate 'G' rated games and a specialised accessible controller that allows your child to play the games using both hands. At the end of the six week trial the computer gaming system and the controller will be collected by David and removed from your home. There are no time restraints in terms of using the device – your child will be asked to use the system as often as they can, within limits that you are happy to set as a parent.

Prior to receiving the computer gaming system, your child will need to undergo another hand sensory assessment. This involves four sensory assessments and one series of hand skill tests will be performed, as described below:

- If your child can identify an unknown object when it is placed in their hand, without using their vision
- If your child can detect the difference between two raised lines when the distance between those lines gets smaller and smaller, without seeing the lines
- If your child can tell if their finger is lifted up or down, when they can't see their hand or fingers
- If your child can detect when light pressure is applied to the end of their finger, without seeing when the pressure is applied
- How quickly your child can complete a series of six timed tasks, such as turning over a series of cards, lifting small common objects and placing them into a container, scooping up small objects with a spoon, stacking checkers, lifting large but light objects (empty soup cans), and lifting large heavy objects (full soup cans).

The hand sensory assessment will need to be done three times as part of this study:

- Assessment #1: four weeks before the gaming system is set up in your home,
- Assessment #2: when the six week computer game trial has finished, and
- Assessment #3: four weeks after the second assessment.

During the sensory assessment, four sensory assessments and one series of hand skill tests will be performed, just like last time. These assessments and tests will be conducted at the Women's & Children's Hospital in North Adelaide, or another venue, and usually take between **30 and 45 minutes** to complete.

During the assessment your child will also have their *Manual Ability Classification System* (MACS) level assessed, which is a scale that describes how children with cerebral palsy use their hands to handle objects in daily activities. This is done by observation, with the therapist who conducts the sensory assessments assessing how your child uses their hands.

During your ***first*** and ***second*** assessment visits we will also ask your child to answer a questionnaire, called the *Cerebral Palsy Quality of Life Questionnaire* (CPQOL) for Children or Teenagers. You will be asked to complete the questionnaire if your child is aged eight years or younger, but your child can complete the questionnaire themselves if they are aged nine or over and can do so. This should take approximately 10 mins to complete.

During the same two visits (the first and second assessments) we will be asking you to complete a short assessment called the *Paediatric Motor Activity Log* (PMAL) about your child and how they use their upper limb in everyday activities. This can be completed during the assessment or at home and should take between 5 and 15 mins to complete.

Will my child benefit from being involved in this study?

Your child may benefit from being involved in this study but we cannot guarantee that they will. Given that most children with cerebral palsy cannot play a commercial gaming system, an immediate benefit may be the enjoyment they receive from playing and using a system that they can access.

What are the possible risks of this study?

The possible risks associated with this study are the following:

- Your child may become addicted to the computer gaming system and may want to play it more than you feel is appropriate – if this becomes the case we would advise that you limit their time on the system to one hour per day, or whatever you feel is appropriate;
- Your child could potentially feel frustrated or annoyed because they can't complete or perform a task during the sensory assessments, or feel discomfort during an assessment session;
- All electrical equipment comes with a potential shock risk, but this risk is minimal in this instance as the controller draws low-voltage (5 volts) and there is no direct connection between the controller surface (where the hands are placed) and a voltage source. An independent reviewer assessed the controller and concluded that "*due to the design of the system, there is negligible risk posed of electrical harm to the patient*"; and
- There is a potential inconvenience when attending the sensory assessment sessions three times at the Women's & Children's Hospital in North Adelaide.

Can I withdraw my child from the study at any time?

Your child's involvement in the study is completely voluntary, and your child may withdraw from the study at any time without prejudice to his/her future treatment or relationship with the Women's & Children's Hospital.

Will my child's medical records need to be accessed if I agree to my child's participation in the study?

Yes. Your child's medical records and information from the SA Cerebral Palsy Register will be accessed by the therapist who conducts the sensory assessments if your child is enrolled into the study, to provide more information about your child's cerebral palsy. The therapist is an employee of the Women's & Children's Hospital Network (WCHN) and a member of A/Prof Ray Russo's Paediatric Rehabilitation Department staff.

Can families keep the computer gaming system and the controller after the trial?

Unfortunately, the gaming system and controller cannot be offered at this stage, however interested families can be informed if the equipment becomes available.

Reimbursement or assistance with costs associated with the study

Families will receive a $20 gift card to assist with expenses associated with travel to the Women's & Children's Hospital in North Adelaide and car parking for each of the three sensory assessment sessions. The gift card will be given to families at the conclusion of each session.

Your child's personal information and confidentiality

Your child's information will remain confidential except in the case of a legal requirement to pass on personal information to authorised third parties. This requirement is standard and applies to information collected both in research and non-research situations. Such requests to access information are rare; however, we have an obligation to inform you of this possibility. If you consent to your child being photographed for research purposes during the study, the photographs of your child may be used in research publications and presentations. Photos that are taken will only show your child's hands and arms, either during the sensory assessments or when your child is using the specialised controller to play the computer games. At no time will your child's face be photographed, or identity revealed. The photos are being taken to communicate how a test was performed, or to demonstrate how the system was used. Your child will not be identifiable in the photos.

What happens in the case of an adverse reaction or adverse finding?

If an adverse event occurs the Chief Investigator should be contacted immediately to report the issue. If an adverse finding results from your child's involvement in the study, you will be contacted by the Chief Investigator who will explain the situation.

Do I need to be made aware of anything else about the study?

Yes. The unidentified data from this study may be used to support the development and commercialisation of the system as a whole, meaning the computer gaming system and the specialised accessible controller may be available for purchase in the future. If you have any queries or questions about this please ask the Chief Investigator.

Who do I contact if a problem arises?

If a problem arises, or if you have any questions about the study, please contact:

Mr David Hobbs (Chief Investigator)
Associate Lecturer/PhD Candidate and Rehabilitation Engineer
Flinders University
Phone: 8201 3167 or 0418 221 811
Email: david.hobbs@flinders.edu.au

This study has been given approval by the Women's & Children's Health Network Human Research Ethics Committee, project number *REC2530/12/15*. If you wish to discuss the approval process, or have any concern or complaint about this study, please contact the Executive Officer of the Human Research Ethics Committee, Ms Brenda Penny, Research Secretariat, on 8161 6521.

WOMEN'S & CHILDREN'S HEALTH NETWORK (WCHN)

HUMAN RESEARCH ETHICS COMMITTEE (HREC)

CONSENT FORM

LAY TITLE: Can children with cerebral palsy improve the way their hands feel touch if they play with an interactive computer gaming system?

SCIENTIFIC TITLE: Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial.

I _____

hereby consent to my child's involvement in the research project entitled:

"Can children with cerebral palsy improve their hand sensation using special haptic computer games? A randomised controlled trial"

1. The nature and purpose of the research project described on the Information Sheet in the information pack that was sent to me in the mail, has been explained to me. I understand it and agree to my child taking part.

2. I agree to the accessing of my child's medical records by investigators of this study, including the South Australian Cerebral Palsy Register, for the purpose of this study.

3. I understand that my child may not directly benefit by taking part in this study.

4. I acknowledge that the possible risks and/or side effects, discomforts and inconveniences, as outlined in the Information Sheet, have been explained to me.

5. I understand that data collected from this study may be used to support the development of a commercial product, as outlined in the Information Sheet.

6. I understand that I can withdraw my child from the study at any stage and that this will not affect medical care or any other aspects of my child's relationship with this healthcare service.

7. I understand that I will receive a $20 gift card to offset expenses related to travel and car parking to attend the sensory assessment sessions at the Women's & Children's Hospital in North Adelaide.

8. I have had the opportunity to discuss taking part in this research project with a family member or friend, and have had the opportunity to have the project explained to me by the researcher over the telephone for any specific questions I may have asked.

9. I am aware that I should retain a copy of this Consent Form, when completed, and the Information Sheet.

10. I **do / do not** (please circle) consent to my child being photographed for research purposes during the study, provided the project has the approval of the Women's & Children's Hospital Network (WCHN) Human Research Ethics Committee (HREC).

11. I understand that my child's information will be kept confidential as explained in the Information Sheet, except where there is a requirement by law for it to be divulged. If I have given permission for my child to be photographed, I understand that unidentifiable photographs of my child (photos that do not show my child's face) may be used in research publications and presentations.

Signed: .....

Relationship to participant: .....

Full name of participant: .....

Dated: .....

Where the developmental level of the child indicates that they have the capacity to understand and consent to the study, the section below should be completed by the child.

Signed: .....

Full name of participant: .....

Dated: .....

I certify that I have explained the study to the parent and/or child and consider that he/she understands what is involved.

Signed: ..... Title: .....

Dated: .....

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Appendix L

This Appendix contains the letter from the *Therapeutics Goods Administration* (TGA), notifying the author of the allocated *Clinical Trial Notification* (CTN) Scheme number.

Australian Government

Department of Health and Ageing
Therapeutic Goods Administration

Dr David Hobbs
School of Computer Sciences, Engineering & Mathematics
The Medical Device Research Institute (MDRI)
Flinders University
GPO Box 2100
ADELAIDE SA 5001

Dear Dr Hobbs,

CLINICAL TRIAL NOTIFICATION FOR A NEW TRIAL

Title: Can children with Cerebral Palsy improve their hand sensation using special haptic computer games? A randomised controlled trial.

Protocol: **FU2013H**

Product name(s): Specially designed, customised, haptic computer gaming system that incorporates a novel, accessible, two-handed controller that enables a child with cerebral palsy (CP) to play designed computer games.

Your notification to conduct the above-mentioned clinical trial under the Clinical Trial Notification (CTN) scheme, pursuant to Schedule 4 of Regulation 7.1 of the Therapeutic Goods (Medical Devices) Regulations 2000, has been received by the Market Authorisation Group.

The clinical trial has been allocated **CTN Number 092/2013.** Please quote this number in any subsequent correspondence about this trial.

It is noted that the trial will be conducted at the following site(s):
Flinders University, Adelaide, SA *12 April 2013*

It is also noted that:

i. approval for the goods for this trial was given in accordance with Item 2.3 of Schedule 4 of the Therapeutic Goods (Medical Devices) Regulations 2000 by the body or organisation conducting the trial at each site; and

ii. the representative of the ethics committee for each site has certified that the committee is constituted and operates in accordance with the NHMRC National Statement on Ethical Conduct in Research Involving Humans, has considered this

PO Box 100 Woden ACT 2606 ABN 40 939 406 804
Phone: 02 6232 8995 Fax: 02 6232 8112 Email: eps@tga.gov.au www.tga.gov.au

TGA Health Safety Regulation

clinical trial, and has provided advice to the body or organisation conducting the trial.

The Therapeutic Goods Administration (TGA) has not carried out an assessment of the quality, safety and efficacy of this product in connection with this or any other notification.

In the event that the Secretary of the Australian Government Department of Health and Ageing becomes aware that to undertake or continue the trial would be contrary to the public interest, the Secretary has the authority to direct that the use of the products for this clinical trial must cease.

If a trial is discontinued for any reason, the Market Authorisation Group should be notified using the TGA's CTN and CTX Trial Completion Advice form enclosed with this acknowledgment.

Yours sincerely

Riannon Cuschieri
Administrative Officer
Market Authorisation Group

12 April 2013

CTN2013 0XX

Australian Government
Department of Health and Ageing
Therapeutic Goods Administration

CTN AND CTX CLINICAL TRIAL COMPLETION ADVICE

This form should be used by sponsors of clinical trials to notify the TGA of completion of trials of medicines and medical devices conducted under the CTN and CTX Schemes.

Sponsor and Clinical Trial Details

Sponsor			
Client ID		Scheme *(Delete as appropriate)*	CTN/CTX
Protocol Number		Trial Number *(assigned by TGA)*	092/2013
Date Completed	/ /	*Notification of completion of a clinical trial should be made only after the trial has been completed at all sites. It is not necessary to notify completion dates for individual trial sites.*	

Reason for completion ***(Select one only)***

Concluded normally	☐	Premature termination - safety*	☐
Insufficient recruits	☐	Premature termination - other*	☐
Directed by TGA	☐	Directed by HREC	☐

**Please give details. Attach additional page if insufficient space.*

Print Name		Position	
Signature	/ /	Phone	
		Fax number	

Send this form to:

Experimental Products Section
Market Authorisation Group
Therapeutic Goods Administration
PO Box 100 WODEN ACT 2606

Appendix M

This Appendix contains the Stage 2 recording sheet that was used during assessment sessions to record all test results.

Assessing the prevalence of tactile sensory agnosia in the hands of children with cerebral palsy – STAGE 2

Date of assessment: _____ Location: _____

Assessors name: _____

Assessment type: **First (pre)** **Second (post)** **Third (follow-up)**

Participant's study number: _____ Gender: _____

Participant's initials: _____

Participant's date of birth: _____

Cerebral palsy type: _____

Dominant side: _____

MACS Level: _____ (Manual Ability Class. System)

Date and source of MACS Level: _____ (e.g.: WCH file, SACPR, etc)

Project contact:

If you have any questions, queries or problems about anything to do with the study, please contact David Hobbs (mobile: 0418 221 811 or work: 8201 3167).

Sensory Assessment Test Results: ***(full 20-piece kit)***

1. Test for tactile detection (Semmes-Weinstein monofilaments) (Blind fold required)

(Note: begin with the 2.83 filament – if they can feel this, then you don't need to proceed to the other colours. If they can't, choose the next largest filament and repeat the process).

Each filament is applied to the **first pad** of the index finger or thumb. The order can be random. Tick (√) the circle if they detect the colour at that site, cross (X) if they fail to detect – three attempts on each site but randomly applied across the four sites.

Detection	(R) Finger 1	(R) Thumb	(L) Finger 1	(L) Thumb
1.65	O O O	O O O	O O O	O O O
2.36	O O O	O O O	O O O	O O O
2.44	O O O	O O O	O O O	O O O
2.83	O O O	O O O	O O O	O O O
3.22	O O O	O O O	O O O	O O O
3.61	O O O	O O O	O O O	O O O
3.84	O O O	O O O	O O O	O O O
4.08	O O O	O O O	O O O	O O O
4.17	O O O	O O O	O O O	O O O
4.31	O O O	O O O	O O O	O O O
4.56	O O O	O O O	O O O	O O O
4.74	O O O	O O O	O O O	O O O
4.93	O O O	O O O	O O O	O O O
5.07	O O O	O O O	O O O	O O O
5.18	O O O	O O O	O O O	O O O
5.46	O O O	O O O	O O O	O O O
5.88	O O O	O O O	O O O	O O O
6.1	O O O	O O O	O O O	O O O
6.45	O O O	O O O	O O O	O O O
6.65	O O O	O O O	O O O	O O O

Highest detection level (R):_____ (finger) _____ (thumb)

Highest detection level (L): _____ (finger) _____ (thumb)

2. Test of texture discrimination #1 (using the AsTex device): (Blind fold required)

Trial No.	***Test 1: Non-Dom.***	***Test 1: Dom.***
Trial 1		
Trial 2		
Trial 3		

Perform the AsTex test (Test 1) with the following instructions:

"*Stop your finger at the point where the strip "feels smooth" and hold your finger in that position until I can record the value*" – do not allow them to move their finger backwards and forwards over the grids.

Adjusted final score (R): _____

Adjusted final score (L): _____

3. Test of proprioception (by moving the distal thumb either up or down)

(Blind fold required)

Non-Dominant hand: Total number correct _____ /10

Dominant hand: Total number correct _____ /10

4. Test of stereognosis: (Blind fold required)

When choosing objects, ensure that 3 are chosen from the 'similar pairs' group of 6 and that 3 are chosen from the 'non-similar' group of 6 objects. Randomly choose the objects so that there is some overlap between the non-dominant and dominant hand, but that some new objects are also used.

Object chosen by therapist (e.g.: pen)	***Non-Dom.: Object identified correctly?*** (Y or N)	***Object chosen by therapist*** (e.g.: pen)	***Dom.: Object identified correctly?*** (Y or N)
1.		1.	
2.		2.	
3.		3.	
4.		4.	
5.		5.	
6.		6.	

Non-Dominant hand: Total number correct _____ /6

Dominant hand: Total number correct _____ /6

Comments (if any):

5. The Jebsen Taylor Hand Function Test (JTHFT): (no blind fold)

The maximum time allowed for any task below is **120 seconds** (2 mins). If they cannot complete the task in that time (they run out of time), score them a value of '120 secs'. If a child cannot complete the task at all, assign them the value of '120 secs', but write a note below that they couldn't attempt or complete the task at all.

Task	***Non-dominant hand***	***Dominant hand***
Card turning	sec	sec
Manipulating small objects (into can)	sec	sec
Simulated feeding (bean in cans)	sec	sec
Stacking checkers	sec	sec
Moving light objects	sec	sec
Moving heavy objects	sec	sec

6. Test of texture discrimination #2 (using the AsTex device): (Blind fold required)

Trial No.	***Test 2: Non-Dom.***	***Test 2: Dom.***
Trial 1		
Trial 2		
Trial 3		

Perform this test ***again*** (for Test 2), however, this time with the instructions as follows:

"*Stop your finger at the point where you can't feel individual lines anymore and hold your finger in that position until I can record the value*" – do not allow them to move their finger backwards and forwards over the grids.

Adjusted final score (R): _____

Adjusted final score (L): _____

Assessor comments/notes/observations: (on any aspect of the assessment)

_____

_____

_____

Appendix N

This Appendix provides further details of the equipment problems and software issues that were listed in Chapter 6, section 6.3.12, Table 42.

Table 42, Issue #2: ‘Orby’ Controller Breakages While on Trial

During the RCT, three children unexpectedly broke their controller, rendering the System unusable. On each occasion the reported break was the same – the dome or ‘orb’ part of the controller had broken and detached from its joystick mount and fallen into the shrouded base of the controller.

The first time a controller breakage was reported, the participant’s mother called the author to report the problem and the author collected the broken controller and replaced it with a new one within 48 hours (the controller broke on the Sunday of a long weekend). The broken controller was only 14 days into the trial when it broke, and five OGS were deployed within family homes at the time, with no problems reported to date.

The family called the author to report that the second controller had broken in identical circumstances, but this time within only two days, highlighting a more serious problem with the controllers. Upon collection of the second broken controller, the father explained that his son was really enjoying the trial, but when he fatigues he tends to favour his strong dominant side, and that he becomes very unilateral in his upper limb movements. The father noted that on both occasions his son was tiring when using the controller and that his dominant right hand was “*taking over and shoving the controller to the left*” when both controllers snapped. On both occasions the child wasn’t harmed or placed in any danger when the controller broke as the orb dome simply dropped into its base, shielding the subject from any sharp edges and the system’s internal electronics. This particular child (participant #9) was 15.5 years old with MACS Level II classification and bilateral CP. It was felt that because this child was older and stronger than the ‘average trial subject’ (the average age for the Stage 2 cohort was 10.7 ± 3.4 years) he was presumably exerting higher forces with his dominant hand that caused twisting of the controller, leading it to break. There were no adverse reports from the other four families currently involved in the trial at the time.

The child’s 6-week trial was paused while the broken controllers were examined to identify their mechanism of failure and to determine the appropriate corrective action. The component that broke was the pin that connected the base of the controller, via

the ruggedised joystick mount, to the top of the controller, via the vibration mount section. Like most of the ‘Orby’ controller components, the pin was 3D printed. Both broken controllers showed identical failure mechanisms, with the pin breaking at the abrupt change in pin shape geometry due to a high stress concentration at the transition region. This was a weakness in the design of the pin and needed to be changed.

In consultation with the Head of Mechanical Engineering (Prof Mark Taylor) at Flinders University, corrective action that was considered was a redesign of the pin shape, to reduce the high stress concentration, or a replacement of the pin material. The best approach, which was also the quickest, was to replace the pin with a stronger material. In consultation with the Engineering Workshop, it was decided that a machined aluminium pin would offer a much stronger and robust solution, and remove the weakness from the existing controller pin design. Existing 3D printed pins were given to the Workshop for reference, along with CAD files for the design, and identical aluminium pins were made.

The two controller breakages and the course of action to correct the problem was reported to the WCH HREC and the TGA, and all existing ‘Orby’ controllers that had been assembled but not yet deployed had their 3D printed pins replaced with the stronger aluminium pin. Eleven days after the second controller broke, participant #9 re-commenced their OGS trial with no further problems.

Six days after participant #9 re-started their trial, participant #8’s mother called to report that their ‘Orby’ controller had broken, in what turned out to be identical circumstances to participant #9. The author replaced the broken ‘Orby’ controller with a new one fitted with an aluminium pin within 24 hours and participant #8’s trial continued with no further problems. This child lived outside the metropolitan area.

The third child to experience an ‘Orby’ controller breakage was participant #37. Unfortunately, this family didn’t report the breakage when it happened on Day 30 of the trial, and they felt it was too late to do anything about it when the author offered to replace the controller with a new one when they reported the breakage eight days later. This family lived outside the Adelaide metropolitan area, so distance was a

challenge. This child was the only participant to have their trial truncated by a controller breakage.

Table 42, Issue #4: Software Issue

The software part of the project, namely the 15 games and the central Games Catalogue that coordinated and logged all game activity, was routinely tested internally during development and designed to be ‘tamper-proof’ when deployed. Each laptop was programmed to start-up and automatically load the Games Catalogue, ignore all USB inputs (if a USB cable/drive was connected), hide the desktop taskbar, have a blank black desktop without icons, have all forms of communication (Bluetooth, Wi-Fi) disabled, ignore software updates, and ignore keyboard inputs (such as the ‘Esc’ or ‘Tab’ key) when the Games Catalogue was running. The only keyboard inputs that the OGS accepted were the password ‘`admin459`’ when in the main menu screen to access the set up options within the OGS (such as turning on ‘Demo’ mode or activating the haptic motors and allocating them to a side) and ‘`quit459`’ when in the menu screen to quit the Catalogue and access the laptop desktop.

However, one family reported a software glitch during the trial. The mother of participant #46 called to say that her son’s name no longer appeared on the main log-in screen on Day 28 (after 4 weeks), and that instead another boy’s name appeared (the name was from an earlier participant). The author asked the mother to re-boot the laptop but this didn’t restore the child’s name to the log-in screen. The mother said that her son was happy to continue the trial (two more weeks) using the other boy’s log-in and that it didn’t bother her or her son, so this participant continued to use the OGS using another trial participant’s name and profile.

During the trial, laptops and controllers were cleaned, refurbished and recycled between participants, with each new participant having their profile loaded into the correct location to ensure their name and particular set up was correct. Post-trial, when participant #46’s System was collected it was obvious that someone had deliberately tampered with the laptop as an unusual short-cut icon appeared on the

otherwise blank desktop. Whoever had tampered with the laptop had accidentally pointed the start-up program to an older and an incorrect Games Catalogue (from an earlier trial), hence the reason an earlier trial participant's name appeared on the log-in screen.

An analysis of participant #46's log files showed that this child engaged with the OGS on nine days during their trial, seven using their name and two using another child's name. In terms of the total time that this child engaged with the OGS (566 mins), 93% of the time (526 mins) was accrued using their profile, and 7% with the other profile. From a trial integrity perspective, the child was randomised to Group B, the non-vibration group, but the tampering caused the loading of a profile for an earlier participant who was randomised to Group A, the vibration group. For the 39 minutes that this child used the incorrect profile, they received 9.1 minutes of total vibration.

When the author contacted the mother to ask if her son had reported any differences between playing the games using their own name compared to the other child's name (given that one profile vibrated and the other didn't) she said that her son made no comment on the differences. The assessing therapist noted that this particular child demonstrated concrete thinking and some possible cognitive difficulties, which might explain why they didn't notice that their 'Orby' controller now vibrated after the laptop was tampered with.

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Appendix O

This Appendix contains an email to the author from a mother of one of the children who was participating in the Stage 2 RCT.

David Hobbs

From: Ros M.
Sent: Sunday, 9 February 2014 6:29 PM
To: David Hobbs
Subject: Orbi Feedback

Dear David,
As we talked about on the phone, we have seen amazing things since **our son** started using Orby 2 weeks ago. Initially he found it challenging, and needed me to sit with him and remind him how to use it, and to do smaller, controlled movements. Below are some notes on what we have discovered so far – and looking forward to more!!!

- he is keen to play and after 9 days managed to unlock all the games
- he discovered that there are 2 games that he doesn't need to have lefty on as much, and is now playing a game with Orby to see how long he can get away with it – I do not see this as a major problem as he is even more conscious of what lefty is doing, and using it separate from his right hand. The two games are A Bridge too far and the Squirrel one.
- He is getting organized for school quickly in the morning, so that he gets time on the game.
- He willingly does it every day while his sister does her Kumon tutoring homework with me.
- He is showing his older sister what to do and explaining how to do some of the games – especially how to get into different worlds in Sunday Driver.
- His sister, for the first time in her life, is willing to play a computer based game and trying to beat her own score each time.
- The two of them together are interacting beautifully, sharing, discussing and encouraging each other, and then of course trying to beat each others' score
- Normally I would try and do **our son's** OT with him at least 3 times a week in the morning before school – now with Orby I am not doing it and enjoying the morning more myself rather than having to rush.
- His left hand is already showing signs of improvement – we had an OT session at Novita this week to make a new night time splint, and the OT was amazed at how much more functional his hand is from all the different things he has been doing including Orby – which she was fascinated to hear about.
- Instead of watching TV at night time he is choosing to go on Orby and plays a variety of games.
- Both kids encourage me to have a go and have a good laugh when I fail dismally at the Space one!
- He has learnt considerable control and how much pressure/force to use on each game.

As I mentioned, I am loving seeing how much effort he is putting in, and all of the positive effects it is having. If there is a way that we could continue to use this system after the trial we would be incredibly interested.
Please feel free to contact me if you have any more questions.
Regards
Ros

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