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**SELF-PERCEIVED AND ACTUAL PHYSICAL FUNCTIONAL  
ABILITIES IN ADULTS WITH CEREBRAL PALSY**

**RAM KRISHNAMURTHY**

A thesis submitted to the University of Huddersfield in partial  
fulfilment of the requirements for the degree of  
Masters in Philosophy (Physiotherapy)

The University of Huddersfield

December 2017

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## Abstract

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**Aim:** To explore the self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years with a view to develop potential self-management strategies.

**Literature background:** Previous research has been inconclusive regarding potential deterioration of physical functional abilities in adults with cerebral palsy. One contributing factor is the use of different tools in the assessment of physical functional abilities, which can be broadly categorised as those assessing self-perceived abilities and actual abilities. This study was designed to explore the self-perceived and actual physical functional abilities in such individuals.

**Method:** Mixed methodology approach was utilised. Phase one involved a quantitative investigation of the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. Phase two of the study utilised qualitative approach to critically explore the possible reasons for and means to reduce the differences between self-perceived and actual physical functional abilities. The findings from both these approaches was utilised to develop guidelines for potential long term management strategies for such individuals.

**Findings:** Seventeen participants participated in the phase one. Statistical analysis revealed that there are discrepancies between self-perceived and actual physical functional abilities, also there was correlation between self-reported Barthel Index and Barthel Index; self-reported Short-Form 36 version 2: physical component summary correlated with the Barthel Index. No correlation was found between the self-perceived abilities and Rivermead Mobility Index. Three participants were interviewed for phase two and four key themes emerged, namely coping strategy, self-management, support and identity.

**Conclusion:** The study revealed that there are discrepancies between the self-perceived and actual physical functional abilities and it is essential for the health professionals to address these discrepancies. Adults with cerebral palsy are concerned regarding the changes in their physical functioning and would like to have appropriate specialist support. It is recommended that this is incorporated in the clinical guidance document and a national register is established to record the physical functioning in adults with cerebral palsy.

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## Glossary of terms

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**Actual abilities** – Individuals' ability as assessed by the clinician or researcher

**Adults** – from 16 years of age

**Cerebral Palsy (CP)** – Cerebral palsy is the name for a group of lifelong conditions that affect movement and coordination, caused by a problem with the brain that occurs before, during or soon after birth (NHS Choices, no date)

**Clinical Commissioners** – Member driven independent organisation that support local clinicians to commission the best possible services for their patients and populations.

**Gait** – Refers to walking pattern

**IBM Statistical Package for Social Sciences (IBM SPSS)** – is a software package that offers advanced statistical analysis

**Linear Regression Analysis** – type of analysis to predict the relationships among the variables

**Mixed methodology** – combining various approaches to do a research study

**NHS** – National Health Services

**NICE** – The National Institute for Health and Care Excellence, this organisation provides national guidance and advice to improve health and social care

**Physical functional abilities** – Refers to walking function, bathing, dressing, transferring, feeding and toileting

**Pragmatic paradigm** – concept that focuses on 'what works' as the truth regarding the research (Taskhakkori and Teddlie, 2003, p713)

**Self-perceived abilities** – self reported abilities as perceived by an individual

**Thematic analysis** – type of qualitative analysis, utilised to identify patterns, 'themes', in the data in relation to research topic in hand (King and Harrocks, 2010, p149)

**Transition** – Moving from children's services to adult services (health and social care)

## Chapter one: INTRODUCTION

---

This thesis focuses on the physical functional abilities in adults with cerebral palsy. I have presented a brief overview of the research context, located myself in this research study, introduced the study aim and objectives and outlined the structure of thesis in this introductory chapter.

### **Research context**

Historically, cerebral palsy is described as a paediatric condition and there have been a significant number of interventions and research studies exploring the notion of cerebral palsy published during this period. Since the primary pathology is considered to be non-enhancing, it was also assumed that the associated motor symptoms do not change (Rosenbaum et al., 2007). However, Hanna et al. (2009) stated that the authors (Bottos et al., 2001 and Jahnsen et al., 2004a) eventually concluded that there was some decline in physical function during adulthood, however the timing, scope and extent of change in gross motor function were not well documented.

Cox et al. (2005), Haak et al (2009) and Morgan and McGinley (2014) reviewed the literature pertaining to physical functional abilities in adults with cerebral palsy and concluded that there is a lack of strong evidence available to inform health care professionals about changes in motor function and recommended a rigorous epidemiological study. A literature review performed by Sandstrom (2008) and Sandstrom et al. (2004) indicated that several factors contributed to the inconsistency in the findings. One of these, described as a major factor was that various outcome measures were utilised to assess physical functional abilities in adults with cerebral palsy. The tools utilised to assess the physical functional abilities can be broadly categorised into measures that reported self-perceived abilities and measures utilised by health care professionals to objectively assess actual abilities. To understand the changes in physical functional abilities during adulthood in cerebral palsy, it is essential for health care professionals to gain more insight into the actual

abilities and self-perceived performance (Sandstrom et al., 2004) and acknowledge that further research is required to establish greater clarity. Therefore, due to the deficiencies in care and the potential impact upon practice in this poorly researched area, I have explored the self-perceived and actual physical functional abilities in adults with cerebral palsy.

### **Locating myself in the research**

I was inspired to do this research due to my professional journey as a physiotherapist. When I moved to the United Kingdom in 2005, I started working in a special school that catered to children with special needs between five and seventeen years. I then moved onto work with a transitions team, supporting individuals with long term neurological disabilities to access adult services when they left school; promoting healthy living and empowering the individuals to be independent in managing their long term condition.

Whilst working with young adults with cerebral palsy, I noticed that they received significant amount of input from the Paediatric Services, within schools. However, when they completed schooling and continued on to future career options, the input provided by the health service decreased and there was a sudden shift of emphasis and responsibility onto the individual in accessing these services. This seemed to have a negative impact on the individuals' physical functioning and they were found to be frequently accessing adult rehabilitation services, which was an indication that the current management strategies were not addressing all issues in the transition from child to adult services. Also, as indicated by Sandstrom et al., (2004) further exploration of the health factors in these individuals is necessary; in order to improve the management strategies. Furthermore, recent literature (Kent, 2013 and Strauss et al., 2004) has indicated that the functional skills become more difficult during adulthood in individuals with cerebral palsy. Additionally, reflection on my practice and a literature review for assignments, revealed several gaps in evidence pertaining to physical functional abilities in adults with cerebral palsy. Currently National Health Services (NHS)

commissioning of services is increasingly evidence based. 'Quality Assurance Standards for Physiotherapy service delivery' recommends that professionals have a system to ensure that physiotherapy care is based on the best available evidence of effectiveness (CSP, 2012, Standard 8.2), which is a standard of practice in health services. This led me to search for evidence pertaining to physical functional abilities in adults with cerebral palsy; however the literature review highlighted several gaps in this particular area. One of the factors that surfaced whilst critically exploring the literature was that the research studies utilised various tools to measure the physical functional abilities, broadly this can be categorised as tools that measured self-perceived abilities and tools that measured actual abilities as assessed by the researcher. Therefore, I embarked on this research journey with University of Huddersfield, to explore the self-perceived and actual physical functional abilities in adults with cerebral palsy with a view to develop the management strategies for such individuals.

### **Study aim and objectives**

The aim of this research was to explore the self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years with a view to develop self-management strategies.

Study Objectives:

1. To investigate the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy
2. To critically explore the possible reasons for and means to reduce these differences in self-perceived and actual physical functional abilities
3. To develop guidelines for potential long term management strategies for such individuals.

## **Thesis structure**

The study report starts from chapter two. This chapter introduces the purpose of doing this literature review, the methodology incorporated, search strategy, synthesis of data, presentation and discussion of the literature findings. This chapter concludes with the summary of the lack of evidence in the area of research, study intention and the study objectives.

Chapter three introduces the philosophical position of this research study, including ontological and epistemological perspectives. This follows with the introduction to mixed methodology utilised to address the study objectives. This chapter elaborates on the role of mixed methodology in health and social research, strength and weakness of mixed methodology, and the methodology used in this research area by other researchers. The chapter further critically evaluates and presents the rationale for adopting mixed methodology in this research study within the context of the study objectives. In this thesis, quantitative approach is followed by the qualitative approach to address the first two study objectives. The findings from both the approaches is utilised to address the third objective of the study. This is followed by the research design and methods, justification of tools selection, development of the interview guide, method of participant selection (for both phases), sample size, data collection process incorporated in both the phases and authenticity (rigor in application and interpretation). Also reported herein are the ethical considerations addressed in this thesis and the process of ethical approval. This chapter concludes with the data analysis process.

In chapter four, the results from phase one and phase two are presented methodically. Phase one findings comprises of demographic distribution, findings from the four outcome measures, influence of age on the outcome measures and analysis of relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. Phase two findings comprises of an introduction to the four themes identified from the analysis of the participants' interview. Due to the relevance to the research area, the



concerns reported by the participants' are incorporated in the final section of this chapter.

Chapter five discusses the findings from phase one and phase two in relation to the objective one and two of the study. Further, the findings from phase one and two are integrated to discuss study objective three.

In chapter six, new knowledge from this research study, implications for clinical practice, implications for learning and training and implications for policy are discussed. This chapter concludes with future research recommendation and strategies for dissemination of the research findings.

Concluding Chapter seven discusses the limitation of the study, researcher's reflection and reflexivity, about the overall experience and knowledge gained as an outcome of this research study and final thoughts

## Chapter two: LITERATURE REVIEW

---

### Introduction to study background

Cerebral palsy is described as a non-progressive brain lesion with primary motor impairment. Historically, researchers and clinicians, have defined cerebral palsy as “an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development” (Mutch et al., 1992, p549). More recently, The Surveillance of Cerebral Palsy in Europe (SCPE) network started in 1998, has collaboratively summarised the definition as, Cerebral Palsy is a group of permanent, but not unchanging disorders of movement and/or posture and of motor function, which is due to a non-progressive interference, lesion, or abnormality of the developing/immature brain (Cans et al., 2007). Although the clinical pathology is non-progressive in individuals with cerebral palsy, the clinical manifestations of motor syndromes change continuously based on the physical and emotional demands in such individuals.

Whilst there is a plethora of strong literature regarding pathology and interventions for managing children with cerebral palsy, less is known about intervention strategies for management of cerebral palsy in adulthood (Haak et al., 2009), even though the disorder was first recognised in the seventeenth century. Historically, cerebral palsy is described as a paediatric condition and this is recognised in the definition that this condition develops as a result of lesion or abnormality of the developing or immature brain. Due to the improved survival rate of 65% to 90% in adults with cerebral palsy (Zaffuto-Sforza, 2005), the management of adults with cerebral palsy has become an essential component in long term care (Kent, 2013). This significant progress in the survival rates of individuals with cerebral palsy has emphasised that more evidence based intervention is all the more important to improve quality of life. Also, paucity of research in adults can also be associated with less contact with health and rehabilitation services when they reach adulthood

(Bottos et al., 2001). It has been accounted that individuals with cerebral palsy gradually have less contact with health care professionals due to lack of specialist care (Bent et al., 2002).

Recent studies (Ando and Ueda, 2000, Strauss et al., 2004, Hanna et al., 2009, Opheim et al., 2009, and Benner et al., 2017) have reported the gradual deterioration in certain aspects of physical function during adulthood in individuals with cerebral palsy. Turk et al. (2001) has argued that there can be a modest change in function over a lifetime of adults with cerebral palsy (considering age-related changes), but significant changes should not be expected. Whilst there appears to be an agreement that there is a decline in physical function during adulthood in individuals with cerebral palsy, the quality of available evidence is weak to moderate (Cox et al., 2005, Haak et al., 2009 and Morgan and McGinley, 2014). Jeglinsky et al. (2010) conducted a systematic review of the evidence available for physiotherapeutic interventions for adults with cerebral palsy and concluded it to be sparse and weak. As there is a healthcare and economic burden due to the disabilities in adults with cerebral palsy, there is a greater focus required on these individual's, in terms of presentation and the comorbidities associated with aging and management.

Cox et al.'s (2005) systematic review and Haak et al.'s (2009) review of literature, concluded that there is lack of strong evidence available to inform health care professionals about changes in motor function and recommended a rigorous epidemiological study. Additionally, Morgan and McGinley (2014) reviewed the literature pertaining to gait function during adulthood in individuals with cerebral palsy and concluded from the review that mobility declined in 25% or more in such individuals, although the time course was variable. Hanna et al. (2009) stated that the authors (Bottos et al., 2001 and Jahnsen et al., 2004a) eventually concluded that there was some decline in physical function during adulthood, however the timing, scope and extent of change in gross motor function were not well documented. Summarising the

reviews (Cox et al., 2005, Haak et al., 2009 and Morgan and McGinley, 2014), although the evidence is weak, these reviews have concluded that there is some deterioration in physical functioning in adults with cerebral palsy. Cox et al. (2005), Haak et al. (2009) and Morgan and McGinley (2014) literature review's concentrated on the evidence pertaining to cerebral palsy and aging but did not focus on the physical function and the factors contributing to the changes in physical functional abilities, which will be addressed in this literature review.

There are a number of factors that may have an impact on the management of cerebral palsy in adulthood, which may be due to the comorbidities associated with aging. Zaffuto-Sforza (2005) attempted to summarise these factors and suggested that individuals' pain, musculoskeletal deformities, arthritis, nerve entrapments, overuse syndromes, fractures, fatigue, general effects of aging on different body systems, dental issues, nutrition and diet, speech and swallowing problems, hearing and vision, exercise, social issues, adaptive equipment and life expectancy would impact on the management of cerebral palsy in adulthood. However, the evidence available to point out the relative influence of these factors and their direct impact on the physical functional abilities during adulthood in cerebral palsy is sparse.

Therefore, this literature review was attempted to summarise the current evidence available to ascertain the changes in physical functional abilities in adults with cerebral palsy and the contributing factors. For the purpose of focus, physical functional abilities in this review includes walking function, bathing, dressing, transferring, feeding and toileting.

## **Methodology**

**Objective:** To systematically analyse the evidence available, pertaining to the changes in physical functional abilities in adults with cerebral palsy and the factors affecting them. This review would help to identify the appropriate

factors affecting the physical function, which may have direct impact on the management in individuals with cerebral palsy during adulthood.

Review Questions:

- Do physical functional abilities change during adulthood in cerebral palsy?
- What are the factors that have contributed to the changes in physical functional abilities?

Robson (2011) has argued that the main purpose of doing a literature review is to find material which helps to identify the research question(s) to seek answers and design the project. A literature review exposes the main gaps in knowledge, identifies principle areas of dispute and uncertainty, identifies general pattern to findings, explores explanations for discrepancies, defines terminologies and helps to identify the research methodologies and instruments (Robson, 2011). It is essential to be systematic in a review, so that that the researcher can identify comprehensively and track down all the available literature on a topic, while describing a clear and comprehensive methodology (Aveyard, 2014). Therefore, to ensure that the completed review is comprehensive, reliable and credible, this literature review pertaining to changes in physical functional abilities in adults with cerebral palsy was performed using the systematic approach as proposed by Aveyard (2014), which comprises of developing and implementation of a systematic search strategy.

Thematic analysis (Aveyard, 2014) was performed to systematically identify, explore and analyse the themes presented within the literature pertaining to the objectives of the literature review. Aveyard (2014) has listed and elaborated the steps involved in the thematic analysis, this approach comprises of critical summary of the relevant literature, identifying and developing themes, discussing the strength of the evidence depending on quality, comparing the themes, scrutinising and critically evaluating the

themes and writing up the findings. Based on these analyses of the literature, the factors that contribute to the changes in physical functional abilities were reviewed.

The criteria utilised to include or exclude the literature and the systematic approach adopted to search the literature, including grey literature, is elaborated here.

### ***Inclusion and exclusion criteria***

Aveyard (2014) and Bettany-Saltikov (2012) emphasises the importance of clear and well-defined inclusion and exclusion criteria, as this helps maintain focus on the search objective, ensuring the investigator does not get side-tracked with literature that are not strictly relevant to the objectives. Therefore, the inclusion and exclusion criteria were formulated to focus on the evidence pertaining to the research question.

#### *Inclusion Criteria*

Studies related to physical function and activities of daily living were explored. To focus on the research question, physical function and activities of daily living included walking function, bathing, dressing, transferring, feeding and toileting. This criterion helped to avoid getting side-tracked with the literature pertaining to quality of life and life satisfaction. To maintain focus on the age range, research that involved adults from 16 years to 40 years with cerebral palsy were searched. Restriction on study type was not placed, in order to be thorough with the searches. Further, studies were limited to English and other languages, if English translation was available.

#### *Exclusion criteria*

The literature search was restricted to physical functioning abilities; studies related to learning disability and general health related studies in adults with cerebral palsy were excluded. Further, studies related to children (<16 years) were excluded from the search.

## ***Search Strategy***

Searches included the electronic databases DARE (Database of Abstracts of Reviews of Effects @ Cochrane library), Cochrane Database of Systematic Reviews, EMBASE, Summon (University of Huddersfield), AMED (Allied and Alternate Medicine Database), CINAHL (Cumulative Index of Nursing and Allied Health Literature), Physiotherapy Evidence Database and Pubmed (includes MEDLINE). The search duration was from each database's inception to January 2015.

An updated literature search in these databases was done in November 2017 covering the period February 2015 to November 2017, and new research was added here.

Journal articles are indexed and entered onto a database using keywords (Aveyard, 2014). To be inclusive, the keywords utilised in the review literature pertaining to cerebral palsy and ageing by Cox et al., (20015) and Haak et al., (2009) was considered. Key terms used to search were Cerebral Palsy, Adult(s), Adolescent(s), Aging, Physical Function, Self(-)care, Quality of Life, Activities of daily living and Disease progression; thesaurus was utilised to identify the key words used in literature across the world. Truncation was used to identify alternate terms, for example activit\*, function\* and adult\*. Boolean operators were also utilised to narrow the search and keep focus on the search objective.

## ***Grey Literature***

A search of grey literature pertaining to the search objective was completed, to minimise publication bias (Robson, 2011). National research register, Department of Health (DoH), National Institute for Health and Care Excellence (NICE), Google Scholar, Scope - About Disability publications and the Higher Education Academy were utilised to search for reports, reviews, theses, dissertations and conference proceedings.

## ***Literature search***

The titles and abstracts of all searchers were evaluated to determine whether they can be included in the analysis. A full manuscript was obtained only for the articles which were thought to be relevant and which adhered to the inclusion and exclusion criteria. The reference lists of these articles were evaluated to identify any potential inclusion in the analysis, which were not found through the electronic searches. Nine potential literatures were found from this back reference list. To reduce any search bias, help from NHS Leeds Library staff was sought in identifying the search terms and searching the electronic database. The search results were compared with the researcher's literature search to prevent bias.

## **Findings**

In total 928 citations were identified from the electronic database search pertaining to the search objective. Evaluation of the article title and abstract revealed that many articles focused on motor development in children, intervention study in adults with cerebral palsy and general health status reports in adults with cerebral palsy. Systematically reviewing the literature based on the inclusion and exclusion criteria, thirteen articles were initially identified and included in this review. An updated literature search following the similar search strategy in these databases was completed in November 2017 and two new researches were added to the findings. One of the literatures was a review (Morgan et al., 2014) and the second article was a prospective longitudinal survey (Benner et al., 2017).

The search results are illustrated in the PRISMA Statement flow diagram (Moher et al., 2009). A group of review authors, methodologist, clinicians, medical editors and a consumer met in 2005 at Ottawa, Canada, to improve reporting methods of systematic reviews (Moher et al., 2009 and Aveyard, 2014). Following this meeting, PRISMA flow chart was suggested to improve reporting of the systematic search, this flow chart consists of four phases

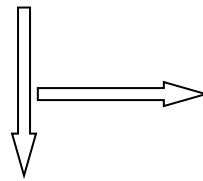


namely, identification, screening, edibility and reporting of the articles included in the review. The process and number of studies identified in this study is illustrated in the PRISMA statement, which can be found in figure 1.

**Figure 1:** Flowchart of articles reviewed according to PRISMA statement

Studies identified by the electronic search, manual search and review of reference list

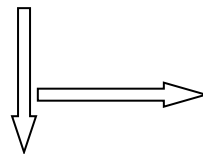
(928 articles identified)



794 studies excluded after reading the title and abstract (not related to research question)

Studies identified for obtaining full manuscript

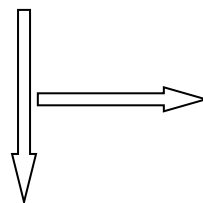
(N=134)



98 studies not related to research question after reading the full manuscript (most of the studies were related to quality of life/public health)

Potentially appropriate studies

(n=36)



Further 19 studies were excluded as not adhering to inclusion and exclusion criteria; 2 studies were included to study the factors affecting physical functional abilities

Studies included in the systematic review

(n=15)

In view of the heterogeneity of the studies and lack of randomised controlled trials, meta-analysis was not appropriate. Instead, as elaborated earlier, thematic analysis of the literature was found to be appropriate considering the available evidence (Aveyard, 2014).

The studies included in evaluating the research question are summarised in Table 2. Out of the fifteen studies identified, three were systematic analysis, five were cohort studies and the remaining seven studies were cross sectional surveys. The studies in which the survey population were compared with the previous case notes and memory, were classified as level 6 cross sectional surveys, as suggested in the Hierarchy of evidence (Guyatt et al. 1995 and Greenhalgh, 1997).

### ***Data extraction and synthesis***

The data extracted from each study included authors, year of publication, study design, population, follow up, outcome measures used, study results, and their limitations. This was documented in the Critical appraisal of Prognostic Studies documentation format suggested by the Oxford Centre for Evidence Based Medicine, which is included in Appendix 1. The quality of the research methodology for each identified study was assessed using the 7 point ranking of hierarchy of evidence (Guyatt et al. 1995 and Greenhalgh, 1997). This ranking was chosen so that the literature ranging from case studies to randomised controlled trial are selected and importance to analysis is given to the appropriate study based on evidence (Aveyard, 2014). This was used to identify the quality of the articles; however, no articles were excluded based on the scoring systems. The Hierarchy of evidence used to rank and classify the articles can be found in table 1.

**Table 1:** Hierarchy of evidence (Guyatt et al. 1995 and Greenhalgh, 1997)

<b>Level of evidence</b>	<b>Methodology</b>
1	Systematic reviews and meta-analyses
2	Randomised controlled trials with definitive results
3	Randomised controlled trials with non-definitive results
4	Cohort studies
5	Case-control studies
6	Cross sectional surveys
7	Case reports

**Table 2:** Summary of reviewed articles and level of evidence

<b>Author &amp; Year</b>	<b>Design / Type</b>	<b>Level of evidence</b>	<b>Outcome measures</b>	<b>Key findings</b>
Cox et al. (2005)	Review	1		Little evidence to date on the overall impact ageing has on adults with cerebral palsy
Haak et al. (2009)	Review	1		No rigorous epidemiological study available; weak evidence available to suggest any changes in motor function
Morgan and McGinley (2014)	Review of gait function	1		Review suggested mobility declined in 25% or more of adults with cerebral palsy
Ando and Ueda (2000)	Prospective longitudinal study	4	Survey questionnaire and physical examination	35% - reported functional deterioration. Deterioration over 5 year period was noted in 8 of 122 patients
Strauss et al. (2004)	Both cross sectional and longitudinal study	4	ADL (walking, self-feeding, speech), Annual Questionnaire	At 20 years, 39% walked without support and 35% didn't walk, at 60 years 25% walked without support and 40% didn't walk. Ambulation and dressing declined significantly after 15 years of follow up.
Hanna et al. (2009)	Prospective longitudinal study	4	Gross Motor Functional Measure -66 and Gross Motor Functional Classification Scale	Children and young in Levels III, IV and V lose significant motor function, with the greatest decline in Level IV

Author & Year	Design / Type	Level of evidence	Outcome measures	Key findings
Opheim et al. (2009)	Retrospective study – 7 year follow up from Jahnsen et al. (2004a) study population	4	Multidimensional questionnaire, Visual Analogue Scale, Short Form 36, Fatigue Questionnaire and the Fatigue Severity Scale	52% reported deterioration of walking function compared to 39% 7 years previously; these participants had greater pain frequency, pain intensity, impact of pain in ADL and physical fatigue and reduced balance.
Benner et al. (2017)	Prospective longitudinal study	4	Postal survey, Perceived health using 3 questions, Presence of pain, pain intensity, pain effect using McGill Pain Questionnaire, Barthel Index, Fatigue severity scale, Likert scale, physical examination and mobility	Over a 14 year period, the scores on the Barthel Index decreased; 28% adults declined at least 2 points, scores of mobility and self-care domain decreased; Walking short distance and indoors decreased but not long distances outdoor walking. Summarising, perceived health decreased considerably, presence of some health issues increased and functional level deteriorated slightly.

Author & Year	Design / Type	Level of evidence	Outcome measures	Key findings
Jahnsen et al. (2003)	Cross sectional design (study of fatigue compared with general population)	5 (case controlled study included for identifying factors attributed to changes in physical functional abilities)	Multidimensional questionnaire, SF 36, Physiotherapy, Physical activities, The Fatigue Questionnaire, Life Satisfaction scale	Mean level of fatigue was significantly higher in adults with cerebral palsy, however the difference was significantly small for physical fatigue. Multiple analyses showed significant association between fatigue, pain limitation of physical role but not level of physical function as such.

<b>Author &amp; Year</b>	<b>Design / Type</b>	<b>Level of evidence</b>	<b>Outcome measures</b>	<b>Key findings</b>
Jahnsen et al. (2004b)	Cross sectional design (study of pain compared with general population)	5 (case controlled study included for identifying factors attributed to changes in physical functional abilities)	Multidimensional survey questionnaire, SF 36, Physiotherapy, Physical activities, The Fatigue Questionnaire, Life Satisfaction scale	87% reported 1 or more joint pain; Prevalence of chronic pain in adults with cerebral palsy increased from 18% under 30 years to 40% from 60 years of age, after controlling for confounding variables in the multiple analysis, low score on physical role function, low life satisfaction and deterioration of functional skills were significantly associated with Chronic pain
Murphy, Molnar and Lankasky (1995)	Cross sectional study	6	Detailed medical history, survey questionnaire and physical examination, modified PULTIBEC system (London 1963)	39% demonstrated decline in walking function
Bottos et al. (2001)	Cross Sectional survey	6	Interview questionnaire and general physical assessment	23/72 reported feeding difficulties, 22/72 reported deterioration of motor function

<b>Author &amp; Year</b>	<b>Design / Type</b>	<b>Level of evidence</b>	<b>Outcome measures</b>	<b>Key findings</b>
Turk et al. (2001)	Cross sectional survey	6	Questionnaire	65% of women with cerebral palsy were able to walk and 46% used wheelchair; secondary conditions was reported: 84% pain, 59% hip/back problems, 49% bladder problems, 35% increased spasticity during menstruation, 28% gastroesophageal reflex.
Andersson and Mattsson (2001)	Cross sectional design	6	Questionnaire	35% reported decreased walking ability and 9% stopped walking of the study population
Jahnsen et al. (2004a)	Retrospective study from the postal survey questionnaire and case notes	6	Type of cerebral palsy, SF36 Items of locomotion skills (revised from Andersson and Mattsson (2001).	27% (n=97) reported improvements in walking, 28% (n=102) reported no change, and 44% (n=160) reported deterioration; Deterioration of walking skill was strongly associated with neurological problems and age
Sandstrom et al. (2004)	Cross sectional design	6	Structured interviews and Functional assessments regarding ADL, motor function, range of motion, pain and general health	Third of population reported deterioration in function from adolescence to adulthood; majority were independent in personal ADL, but many were dependent in instrumental ADL



Author & Year	Design / Type	Level of evidence	Outcome measures	Key findings
Morgan, Soh and McGinley (2014)	Cross sectional survey	6	Postal survey sought for self-reported mobility status, GMFCS, presence of mobility decline, Short Form 36 Health Survey and Personal Wellbeing Index	Mobility decline was significantly associated with mental health status but not when other predictor variables (falls, physical component summary and life satisfaction) were included in the analysis

### ***Cross sectional studies***

The study designs used by Murphy et al. (1995), Bottos et al. (2001), Turk et al. (2001), Andersson and Mattsson (2001), Sandstrom et al. (2004), Jahnsen et al. (2004a) and Morgan, Soh and McGinley (2014) were cross sectional self-reported surveys, which are on level 6, according to Hierarchy of Evidence (Guyatt et al. 1995 and Greenhalgh, 1997). Due to the methodological weakness (Guyatt et al. 1995 and Greenhalgh, 1997), these evidences were not considered strong to conclude that there are changes in physical functional abilities during adulthood in individuals with cerebral palsy. Each of the study methodology, outcome measures and results are critically analysed here, using thematic analysis (Aveyard, 2014).

Murphy et al. (1995) conducted cross-sectional survey on one hundred and one adults with cerebral palsy between the age of 19 and 74 years and used modified PULTIBEC (Physical capacity, upper limbs, locomotion, toilet, intelligence, behaviour, vision and communication) system to collect data on function and for some individuals' detailed physical examination was performed. A modified PULTIBEC system was developed by Lindon (1963) as an attempt to fulfil the need for a global, yet concise system for coding the difficulties of children with multiple disabilities in functional terms; consisting of eight domains for assessment, namely physical capacity, upper limbs, locomotion, toilet, intelligence, behaviour, vision and communication. Whilst the literature search revealed that this outcome measure was validated in children (Lindon, 1963), no literature evidence validated in adults with cerebral palsy was found, this being a major weakness of this study. 39% of the study population who used wheelchair (total n=67), reported that they walked in the past, 12 adults had stopped walking between 11 and 20 years, six adults between 21 and 28 years, seven adults between 38 and 50 years and one at 68 years of age.

Bottos et al. (2001) investigated functional abilities in adults with cerebral palsy in seventy one adults at three rehabilitation units in Italy which was non-

random sampling and did not represent the cohort. Data was collected from case notes and physical examination and the authors reported that 18% (n=13) lost independent walking. Nine of the thirteen participants who lost independent walking lost this function between 20 and 40 years, two participants over 40 years and the remaining two participants before the age of 20 years. 32% (n= 23 of the 72) demonstrated feeding difficulties and the most common reason was due to dysphagia and gastroesophageal reflex. Andersson and Mattsson (2001) surveyed two hundred and twenty one adults with cerebral palsy and reported that 15% (n=32) continued walking, 27% (n=59) had never been able to walk and 9% (n=20) had stopped walking. Seventeen of the twenty participants had lost their ability to walk before the age of 35 years and one person reported loss of ambulation after the age of 45 years. Among the one hundred and forty one participants who reported that they were able to walk 19% (n=27) reported an improved walking ability and 35% (n=49) reported deterioration in walking ability.

Contrary to the findings of Murphy et al. (1995), Jahnsen et al. (2004a) and Morgan et al. (2014); Andersson and Mattsson (2001) reported that 19% of their study population improved in walking ability. Andersson and Mattsson (2001) suggested that improvements in walking ability may be due to the benefits from physical training, however data regarding training intensity of the participants were not recorded. These participants participated in varying intensity of physiotherapy, organised sports, weight training at the gym, home training, swimming, walking, horseback riding, gardening and standing using a standing device.

Turk et al. (2001) analysed data from sixty three women (20 to 74 years) with cerebral palsy who lived in the community and found that 65% of them were able to walk and 46% reported using wheelchair. Over 50% of the women required no assistance with eating, bathing, dressing, grooming and toileting.

Jahnsen et al. (2004a) surveyed four hundred and six adults with cerebral palsy (53% response rate), using a multidimensional questionnaire which

included Short Form 36 and revised locomotion skills questionnaire which were originally proposed by Andersson and Mattsson (2001). Jahnsen et al., (2004a) concluded from their survey in relation to locomotion, that 75% walked at the time of survey, 12% never walked and 10% (n=39) reported that they stopped walking. This decline was reported mainly in Diplegic (41%), Quadriplegic (25%) and Dyskinesia (25%). The findings also found that there is marked deterioration in physical function from 30 years of age. This locomotion questionnaire consisted of items to record age of walking debut, walking distance, use of walking aids or wheelchair, improvements or deterioration of walking skills, however information pertaining to the validity and reliability of this questionnaire was lacking in Jahnsen et al. (2004a) and Andersson and Mattsson (2001), therefore the results have to be interpreted with caution.

Sandstrom et al. (2004) conducted a structured interview and functional activities assessment, that included Gross Motor Classification Scale (GMFCS is a classification system based on self-initiated movement, Level I to V indicates decreasing levels of mobility (Palisano et al., 1997)), Physical items of Functional Independence Measure (personal care, sphincter management, transfer and locomotion), Instrumental Activity Measure (measures outdoor locomotion, simple meal cooking, public transportation, small-scale shopping, large-scale shopping, cleaning and washing), Clinical Outcome Variable Scale, range of motion assessment, pain localisation and pain intensity using visual analogue scale and Euro-Qol 5D, on forty eight adults with cerebral palsy. Sandstrom et al., (2004) concluded that sixteen individuals reported decline in function with predominance in Level I on GMFCS, further deterioration was reported in instrumental activities of daily living with dependency particularly in shopping and cleaning but not in personal activities of daily living. Sandstrom et al. (2004) findings indicated that there is deterioration over all GMFCS levels with predominance in Level I, which was contrary to the findings of Jahnsen et al. (2004b), who reported that individuals' in levels III – V experienced the most deterioration. Sandstrom et

al. (2004) indicated that the possible explanation for this contradictory findings can be due to the discrepancy between self-perceived and clinician assessed deterioration, with a risk for underestimating when using self-reported data.

Morgan et al. (2014) explored the relationship between gait and health related quality of life in ambulant adults with cerebral palsy. The data was collected using postal surveys which sought self-reported mobility status, GMFCS, presence of mobility decline, Short Form 36 Health Survey and Personal Wellbeing Index from thirty four adults with cerebral palsy with a mean age of 44.2 years (range 26-65 years). The study found that mobility decline was significantly associated with mental health status but not when other predictor variables (falls and physical component summary and Personal Wellbeing Index) were included in the analysis. Morgan et al. (2014) recommended further prospective research to better understand the relationship between Health related Quality of life and mobility decline.

### ***Longitudinal cohort studies***

The study design incorporated in five studies was longitudinal cohort studies which are level 4 according to the Hierarchy of evidence (Guyatt et al. 1995 and Greenhalgh, 1997).

Ando and Ueda (2000) and Straus et al. (2004) findings relied on the medical notes completed by several health care professionals and a self-reported survey to follow up individuals with motor changes and concluded that there is functional deterioration in adults with cerebral palsy. Cox et al. (2005) commented that the problems associated with postal surveys are recall bias, the question may be misunderstood or interpreted differently, participants may have perceptual problems, memory issues in recalling the information or the information may be inadequate. Furthermore, training investigators is an essential component to establish consistency in medical notes reporting (Robson, 2011), this information is lacking in the Ando and Ueda (2000) and Straus et al., (2004), resulting in cautious interpretation of the results. The

precise age of decline quoted in these studies (Ando and Uedo, 2000) is also questionable due to memory recall. Additionally, the tools incorporated in these studies for self-reporting were non-standardised questionnaires, resulting in inconclusive evidence. Ando and Uedo (2000) and Strauss et al. (2004) studies are thematically analysed here.

Ando and Ueda (2000) surveyed six hundred and eighty six adults with cerebral palsy (62% response rate from the survey) who work at the community workshops throughout Japan, raising caution when interpreting the results due to cultural differences (Aveyard, 2014). In Ando and Uedo's (2000) survey, one hundred and sixty two adults participated in the 2<sup>nd</sup> part of the study which was physical examination by physicians specialised in rehabilitation medicine. Activities of daily living (ADL) were classified into four levels depending on independence levels. For the 3<sup>rd</sup> part of the study, one hundred and twenty two participants (from the 2<sup>nd</sup> part of the study) were followed up after five years. In the first part of the self-reported study, deterioration was marked in older adults and those with decreased level of independence in activities of daily living. A physical examination revealed that functional deterioration was mostly seen in clients with involuntary movements of head and neck and in severely disabled patients. In five years, only eight patients (out of one hundred and twenty two) demonstrated deterioration, this change mainly occurred in wheelchair dependent patients who deteriorated to complete dependence from wheelchair independence. The mean age of physical deterioration was 37.4 years which is similar to the findings of Opheim et al. (2009). Also, the changes in physical functional abilities in Ando and Uedo (2002) study participants could be accounted to vocational impact where accelerated aging would have resulted due to repetition of activity (Murphy et al., 1995) and not as a direct result of the disabilities associated with cerebral palsy. Care providers, clinical commissioners and individuals with cerebral palsy may be interested in the evidence pertaining to mean age of physical deterioration since the management strategies can be set up to address this age group.

Strauss et al. (2004) adopted both cross sectional and longitudinal design, to study the activities of daily living (walking, self-feeding and speech) skills decline in cerebral palsy with age, utilising the reliable annual questionnaire, which contained two hundred psychological, medical, functional, behavioural and cognitive items. Strauss et al. (2004) surveyed the annual questionnaire collected from 14806 adults with cerebral palsy and concluded that among the young adults aged 20 to 30 years, 39% were able to walk well without support for at least twenty feet, while 35% did not walk even with support; at the age of 60 years only 25% were able to walk well and 40% did not walk at all. Strauss et al. (2004) further concluded that in the follow up after fifteen years, about 8% could no longer walk without support and some 13% could still walk well at least twenty feet but could no longer move up and down the stairs without the use of a handrail, whilst the remaining 75% retained their original level of ambulation. In 60 year olds, who initially walked well alone and climbed stairs without a handrail, 15 years later, majority of the survivors (70% were alive), were no longer able to walk well alone for twenty feet (Strauss et al., 2004), however, exact data was not available from the study, making it difficult to identify the cause and effect (Creswell et al., 2003). Significant decline in function was noticed in dressing after the age of 60 years. Also Strauss et al., (2004) found no significant changes in ordering food in public, feeding and speech and concluded that eventually there is decline in gross motor function (notably, in the ability to walk), although the results were dependent on the participants memory recall which may not be precise (Cox et al., 2005), resulting in methodological weakness.

Opheim et al. (2009) used the standardised Gross Motor Function Classification System (GMFCS) and studied the individuals who participated in Jahnsen et al. (2004a) study after seven years. The GMFCS is a 5-level classification system that describes the gross motor function of children and youth with cerebral palsy on the basis of their self-initiated movement with particular emphasis on sitting and mobility (Palisano et al., 1997). Opheim et al., (2009) scored baseline GMFCS retrospectively from the collected data in

1999 on the basis of available information and the follow up GFMCS was obtained through self-reports and the questionnaire, GFMCS is reported to have evidence for excellent agreement within professional ratings (Hanna et al., 2009). One hundred and forty nine participants from the Jahnsen et al. (2004a) study cohorts were surveyed (68% response rate) and 16% reported improved walking function, 31% remain unchanged and 52% deteriorated. The Kaplan-Meier estimate (SPSS analysis; Kaplan-Meier estimate is utilised to estimate the time to event (Jager et al., 2008)) of median age of walking deterioration was 52 years in unilateral cerebral palsy (95% CI 34-70) and 37 years in bilateral cerebral palsy (95% CI 34-39; long rank  $X^2 = 7.8$ ,  $df=1$ ,  $p=0.005$ ). In the youngest age group in 1999 ( $\leq 25$ ), three out of thirty one people experience deterioration in walking between 1999 and 2006. The number of people with problems of overall mobility increased from twenty (13%) in 1999 to thirty seven (25%) in 2006 (mean difference 12%, 95% CI 5-19%,  $p=0.001$ ). Opheim et al. (2009) study was not designed prospectively for comparison and the GMFCS was calculated retrospectively based on the self-reported survey, retrospective data collection has inherited weakness since the information may not be clearly defined (Cox et al., 2005), therefore this evidence is methodologically weak (Guyatt et al. 1995 and Greenhalgh, 1997).

Benner et al., (2017) conducted a prospective four year and a fourteen year follow up study, involving eighty eight adults with cerebral palsy at baseline and forty eight adults with cerebral palsy at fourteen year follow up (35 to 45 years). This research (Benner et al., 2017) utilised postal survey, perceived health measured using three questions, presence of pain, pain intensity, pain effect using McGill Pain Questionnaire, Barthel Index, Fatigue severity scale, Likert scale, physical examination and mobility as outcome measures and concluded that at fourteen year period, the scores on the Barthel Index decreased; 28% adults declined at least 2 points; scores of mobility and self-care domain decreased; walking short distance and indoors decreased but not long distances outdoor walking. Benner et al. (2017) concluded that perceived



health decreased considerably, presence of some health issues increased and functional level deteriorated slightly, however this study did not report the exploration of the differences between self-perceived and actual physical functional abilities in such individuals'. Benner et al., (2017) identified that at baseline perceived health and health issues were not assessed as one of the potential weakness. Benner et al. (2017) used a combination of postal surveys and non-standardised questionnaires during the long term follow up, as indicated earlier with postal surveys and non-standardised questionnaires, there may be recall bias, the question may be misunderstood or interpreted differently and memory issues may impact on the information provided (Cox et al., 2005), therefore, due to this methodology weakness, the evidence should be interpreted with caution.

Hanna et al.'s (2009) study design was a prospective longitudinal study and used a standardised tool. In a prospective study design the research focuses directly on the research question and the study is designed using appropriate design, participants are selected appropriate to the chosen methodology and standardised tools are utilised (Gray, 2009), these strengths increases the credibility of the findings. Hanna et al.'s (2009) primary objective was to validate the Gross Motor Function Measure-66, two hundred and twenty nine children and adolescents with age group ranging from 2 to 21 years were measured on the Gross Motor Function Measure-66. Hanna et al. (2009) focused on the motor abilities in children, especially in adolescent period and concluded that there was a decline in gross motor abilities in the GMFCS levels III to V and the decline started before adolescence.

In summary, significant physical growth and lifestyle changes that occur during adolescents could be attributed to the start of decline in physical functional abilities (Department of Health (DoH), 2004a). Furthermore, while many children and adolescents receive therapy provided in educational settings, the same level or intensity of rehabilitation was found lacking when they leave the school and move on to pursue college, university education or

any vocation (Bent et al., 2002 and Hanna et al., 2009). Shikako-Thomas et al., (2009) has suggested that lack of community programme and adapted environment may be a reason why community is not considered for participation, impacting on the value and enjoyment of leisure activities. Furthermore, having injuries or complications associated with the condition and fatigue are some of the essential barriers to physical activity (Cox et al., 2005 and Buffart et al., 2009). Consequently, lack of resource availability, environmental demands and time constraints may lead to loss of motor function leading to a decline in physical functional abilities. Therefore, factors contributing to the changes in physical functional abilities and management of these factors should be studied in adults with cerebral palsy so that the individuals can maintain good quality of life. This will also help these individuals to maintain maximal potential independence and function that remain the ultimate goal of any rehabilitation input.

### ***Factors attributed to decline in physical functional abilities***

A summary of the factors attributed to decline in physical functional abilities from the reviewed studies has been tabulated in Table 3. From the available evidence, various themes pertaining to the factors that affect physical functional abilities are analysed in detail here.

**Table 3:** Factors contributing to the changes in physical functional abilities

Physical	Environmental	Psychological
Joint pain Chronic pain Fatigue [pain and fatigue due to inactivity (decreased training), over exercise (wear & tear), cold weather]  Natural tendency towards increased energy cost Spasms Muscle stiffness Inefficiency of ambulation Decreased self-initiated motor function Inconvenience of walking Physical growth Decreased respiratory growth Decreased walking speed	Wheelchair provided increased access to a broader range of function	Mental fatigue Decreased self-initiated motor function Inconvenience of walking

Jahnsen et al. (2003, 2004b) studied the pain and fatigue from the multidimensional questionnaire, Short Form-36 (SF36) and the Fatigue Questionnaire. Short Form-36 yields an 8-scale health profile as well as measures of health-related quality of life (Jahnsen et al., 2003). Fatigue Questionnaire utilised by Jahnsen et al., (2003) was a standardised questionnaire, respondents were asked about fatigue symptoms during the last month, compared with when they last felt well; this questionnaire consists of eleven items, seven items measuring physical and four measuring mental fatigue. Jahnsen et al., (2003 and 2004b) reported that the individuals with cerebral palsy, cited pain, fatigue and lack of adapted physical activity as the key causes of deterioration and the low score in physical role function (on SF36 domain), low satisfaction (Global item domain of the Life Satisfaction

Scale developed Fugl-Meyer et al., 1991) and deterioration of functional skills were significantly associated with chronic pain. Jahnsen et al. (2003 and 2004b) also reported that 87% of their study population, self-reported joint pain in one or more joints (mean age of pain was 21 years), chronic pain in adults with cerebral palsy increased from 18% under 30 years of age to 40% from 60 years of age. The variables that increased pain in this study were over-exercise in 73%, inactivity in 26% and cold weather in 14% and therefore, pain and fatigue were cited as the major contributors to decline in physical function. Further, inactivity was also cited as a contribution to decline in physical function. Logistic regression analysis (this predictor analysis is utilised to describe the relationship between one dependent binary variable and one or more variable) (Chao-Ying, Kuk and Ingersoll, 2002) showed that deterioration of walking skills was strongly associated with spastic quadriplegia (all four limbs are affected) which is the most severe type of cerebral palsy and least with hemiplegics (one side of the body is affected) (Jahnsen et al. 2004b). Murphy et al. (1995) study population stated that they stopped walking due to fatigue, inefficiency of ambulation and because wheelchair mobility provided better access for a broader range of functional abilities.

Similar to the studies of Jahnsen et al. (2003 and 2004b), Sandstrom et al. (2004) also concluded that pain and limited range of motion were found in adults with cerebral palsy who showed deterioration in walking function. In Andersson and Mattsson's (2001) study, increased pain in muscles and joints was also found in adults who stated deterioration in walking function and suggested that inadequate physical training could have contributed to this deterioration.

In Opheim et al.'s (2009) study, those who reported walking deterioration in 2006 (n=78), fifty one participants (65%) regarded the deterioration as a result of impaired balance, twenty six participants (33%) a result of reduced muscle strength, and twenty two participants (28%) a result of both reduced

cardiorespiratory fitness and reduced walking speed. Spasms, stiffness, wear and tear, pain and reduced training were reported in nineteen participants (24%). The authors (Opheim et al., 2009) also analysed pain and fatigue in the subgroup of the participants from 2006 and showed that those who reported deterioration in walking function also reported significantly increased pain frequency, more pain sites, high pain intensity, more impact of pain upon daily life and activities and fatigue. Similarly, Bottos et al. (2001) reported that 85% (n=11) gave up independent walking due to high energy cost, which was related to fatigue. This deterioration in walking was also related to inconvenience in walking and increased joint pains (Strauss et al., 2004).

Turk et al. (2001) further analysed general health issues in the study population and concluded that pain, bowel problems, bladder problems, spasticity, gastroesophageal reflux and fatigue were significant health factors being affected. However, Turk et al. (2001) did not investigate the variables affecting the walking and activities of daily living. Self-initiated motor movements, including walking was addressed by Hanna et al. (2009), this study speculated that combination of physical growth and decreased self-initiated motor function may be associated with a natural tendency towards increased energy costs, additionally, contractures, and muscle stiffness may have contributed to the decline from six years of age.

## **Discussion of literature review findings**

### **Literature review objective 1: Do physical functional abilities change during adulthood in individuals with cerebral palsy?**

The objective of this literature review was first to review the studies pertaining to changes in physical functional abilities during adulthood in individuals with cerebral palsy.

Murphy et al. (1995), Bottos et al. (2001), Turk et al. (2001), Andersson and Mattsson (2001), Sandstrom et al. (2004), Jahnsen et al. (2004a) and Morgan et al. (2014) have indicated that there is a decline in physical functional abilities (in various aspects of physical functioning) during adulthood in individuals with cerebral palsy, however the quality of studies are variable, the study designs were methodologically weaker (cross sectional self-reported surveys) and therefore were not considered as strong evidence to conclude that there is decline in physical functional abilities during adulthood in individuals with cerebral palsy. These findings were similar to the literature review conclusions of Cox et al., (2005) and Haak et al., (2009).

Hanna et al. (2009) have argued that their study examined the abilities of the children (abilities assessed by the researcher) and not the performance levels chosen by the individuals (what the individual think they can do, described as self-reported abilities by the researchers) and concluded that the decline in motor function was predominantly due to the decline in the participant's capacity rather than their preference, in children age ranging from 16 months to 21 years. Most of the available research in physical functional abilities is based on self-reported performance, however Hanna et al. (2009), reported the changes in actual abilities. The self-reported studies of Murphy et al., (1995), Bottos et al. (2001), Turk et al. (2001), Andersson and Mattsson (2001), Sandstrom et al. (2004), Jahnsen et al. (2004a) and Morgan et al. (2014) utilised Short Form-36, multidimensional questionnaires, fatigue questionnaire, case notes and interviews which evaluates individuals'

performance (self-perceived abilities) rather than their actual abilities. The validity and reliability of Short Form-36 and fatigue questionnaire are well documented and established (Loge and Kaasa 1998 and Fugl-Meyer et al., 1991). It is paramount that a valid and reliable measurement tools are utilised in research, since these tools are tested to achieve its purpose and the research can be replicated to produce the same outcome. The validity and reliability of the multidimensional questionnaire was not reported by Jahnsen et al. (2004a).

Hanna et al. (2009) referred to the self-reported questionnaires' responses as the performance levels chosen by the individuals and therefore conducted their study to assess individuals' ability from standardised tools (directly assessed by healthcare professionals). Drawing from the literature review, it is clear that authors have evaluated the physical functioning using standardised tools to measure self-reported performance (Bottos et al, 2001, Andersson and Mattsson, 2001 and Jahnsen et al., 2004a) rather than actual abilities as assessed by the clinician (Hanna et al., 2009). Individuals' perception of abilities will have a significant effect on reporting of their abilities, this is elaborated furthermore in the forthcoming discussion. From the literature review and for clear differentiation in this research, actual ability is defined as the 'abilities the individual can do', which are the individual's real potential (as assessed by the health care professional) and self-perceived ability as the 'individuals perceived level of function'. Though the primary factor contributing to the decline in physical functional abilities may be the participants decline in actual abilities, it may be compounded by the performance chosen by the individual or vice versa. The longer this persists, the greater is the loss of decrease in the individuals' rehabilitation potential. There has to be a balance between the chosen performance and actual abilities to reduce the complications like pain, fatigue, loss of mobility and contractures, arising out of decline in the physical functional abilities in this population.

Also, in self-reported situation, respondents are effectively acting as observers of their own behaviour, however the assessment of actual ability by the investigator reduces potential bias and distortions arising from this process (Robson, 2011). Although, to measure thoughts, feelings, beliefs and attitudes, of the participants, self-reports are necessary (Robson, 2011), these factors cannot be captured from the investigator assessments.

To understand the changes in physical functional abilities during adulthood in individuals with cerebral palsy, health care professionals need to know more about both the actual abilities and self-perceived performance (Sandstrom et al., 2004). It is pertinent to recognise the influence of an individual's perception, as their thoughts, feelings, beliefs and approaches will impact on physical functioning. Furthermore, in clinical practice I have observed various discrepancies between actual ability and self-perceived ability in daily tasks. This discrepancy will affect an individual's ability to maintain daily tasks and quality of life. For example, if an adult with cerebral palsy can walk a few steps, it is important that the individual maintains this skill as much as possible. However, the individual may choose not to walk for a variety of perceived behaviour or due to environmental barriers. Gradually over time, the individual may lose walking ability due to increased contractures and muscle weakness, arising due to the disuse of muscles related to walking. To reduce deterioration in physical functional abilities, it is essential that the self-perceived ability mirrors the actual abilities in adults with cerebral palsy. Sandstrom et al. (2004) acknowledged that there can be a discrepancy between self-perceived and actual ability, with an associated risk for underestimating when using self-reported data, however my literature review failed to identify research exploring this field.

Emck et al. (2009) theorised that the possibility of poor gross motor performance of children between 6 and 12 years with developmental disorders may lead to low self-perceived motor competences (self-perceived physical functional ability), which in turn complicates possible participation in



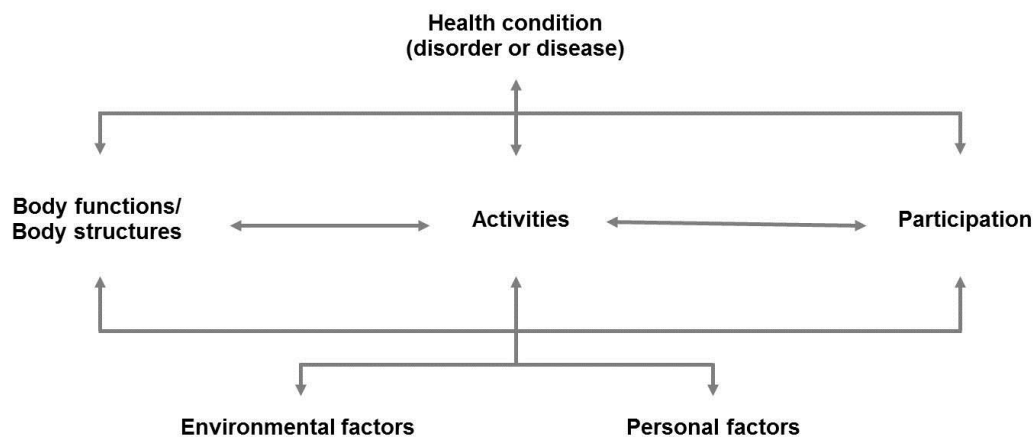
movement activities, this prevents the individual from experiences that may help to improve their motor performance. Hanna et al. (2009) have also acknowledged that there can be discrepancies between what children can do (capacity) and what they choose to do (performance) and speculated that decline in motor function may be due to some combination of physical growth; decreased self-initiated motor function which may be associated with a 'natural' tendency toward long standing increased energy costs, contractures and muscle stiffness. Emck et al. (2009) concluded that measurement of motor performance (actual abilities) and self-perceived motor competence (self-perceived abilities) should be a standard practice to guide and design treatment plans and recommended that future studies address this relationship with the explicit aim to delineating cause and effect. Morgan et al. (2014) explored the relationship between gait and health related quality of life in ambulant adults with cerebral palsy and concluded that mobility decline was significantly associated with mental health status, but not, when other predictor variables (falls, physical component summary and life satisfaction) were included in the analysis. Morgan et al. (2014) recommended further prospective research to better understand the relationship between health related quality of life and mobility decline. However, there is a dearth of literature and evidence related to the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy to inform practice, which should be focused in future studies. Consequently, this supported me in my quest to explore the relationship between self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years. Due to the limited scope of the research programme, changes in physical functional abilities longitudinally from 16 years to 40 years will not be investigated.

## Literature review objective 2: What are the factors that have contributed to the changes in Physical functional abilities?

From the available evidence identified in the literature review, the contributing factors to decline in physical functional abilities can be summarised into three main themes: Physical, Environmental and Psychological. The factors evident and the association with these themes are categorized in Table 3. The interaction between these factors does follow a biopsychosocial model, as explained in International Classification of Functioning, Disability and Health (ICF) (Hanna et al. 2009).

The biopsychosocial model from ICF integrates the medical and social model. This synthesised model, provides a coherent view of different perspectives of health (Biological), individual and social (World Health Organisation (WHO), 2002), the interaction is illustrated in figure 2.

**Figure 2:** Biopsychosocial model - Reproduced from ICF



The factors that contributed to the decline in physical functional abilities are categorised in Table 3, based on this model as Physical (Body function/body structure, Environmental (environmental factors) and Psychological (personal factors), which in turn affects individuals' activities (daily tasks) and participation (social, vocational and leisure participation).

As discussed earlier, the inconsistency in the literature findings (Cox et al., 2005, Hanna et al, 2009), pertaining to physical functional abilities in adults with cerebral palsy, can be attributed to the failure to acknowledge the type of tool utilised (self-perceived and actual abilities) and measurement objective. To understand the factors attributing to the changes in physical functional abilities, one has to choose the right outcome measures which assess all the categories longitudinally. Given the poor health status of persons with disabilities and the potential benefit of participation in physical functioning, it becomes important to understand the range of factors that promote and inhibit participation (Conchar et al., 2016). Therefore, it is essential that the outcome measure(s) assesses all the categories for better understanding the changes in physical functional abilities and the contributing factors in individuals with cerebral palsy. This will help the researcher to also identify the interaction between these factors.

Therefore, drawing from the outcome measures utilised to study the changes in physical functional abilities in adults with cerebral palsy, the contributing factors that can impact on the changes in physical functional abilities and analysing the methodology utilised in the available literature, it is recommend that research should incorporate standardised outcome measures like self-initiated motor activity using Gross Motor Function Measure-66, General Health Survey using Short Form-36, functional tasks assessment tools like Barthel Index, Functional Independence Measure and interviews. Interviews will help in identifying the psychosocial factors like motivation, fatigue, leisure interest and opportunities and family circumstances that may contribute to the changes in motor function. Psychosocial factors should be considered as an important factor as it may have a direct impact on the physical functional abilities whilst the previous studies have failed to recognize this component. Physiological factors, psychological factors, social factors and macro-environmental factors, all play a role in the experiences of the individual in physical functioning (Conchar et al., 2016).

## **Conclusion and future recommendation**

Reviewing the evidence, it can be concluded that there is weak to moderate quality of evidence available to inform us that physical functional abilities changes (or predominantly declines) during adulthood in individuals with cerebral palsy. The variability in the findings can be accounted to the type of measurement tool utilised. Broadly this can be differentiated as measurement tools that reports perceived ability of the individual and the actual ability assessed by the health care professional. There is dearth of literature and evidence of self-perceived and actual physical functional abilities in adults with cerebral palsy to inform practice, therefore it is recommended that future studies focus on this area. This will help the individuals and care providers to understand more about the changes in physical functional abilities in such individuals.

Drawing up from the literature review, it is clear that there is lack of studies indicating the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. Furthermore, it is also essential to explore this relationship (whether similar or different) between self-perceived and actual physical functional abilities in adults with cerebral palsy. Clear understanding of the changes in physical functional abilities could contribute to the growing body of evidence around physical functional abilities in adults to cerebral palsy to inform individuals, clinical commissioners and service providers.

As a consequence of the deficiencies identified in the review; that is, lack of research studies exploring the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy, I developed the following aim and objectives for the research study.

## **Study Aim and Objectives**

The aim of the study was to explore the self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years with a view to develop self-management strategies.

More precisely, the study objectives were

1. To investigate the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy.
2. To critically explore the possible reasons for and means to reduce these differences in self-perceived and actual physical functional abilities.
3. To develop guidelines for potential long term management strategies for such individuals.

The first objective was to investigate the relationship between self-perceived and actual physical functional abilities during adulthood in individuals with cerebral palsy. This was investigated using a quantitative approach. A qualitative approach was utilised to explore the possible reasons for differences between self-perceived and actual physical functional abilities in this population and the means to reduce these differences. The data from both the approaches was utilised to develop guidelines for potential long term management strategies for such individuals.

In this study, quantitative approach and qualitative approach were utilised in a sequential method to study the objective one and two. Hence mixed methodology was utilised to address the three study objectives.

## Chapter three: METHODOLOGY

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### **Introduction**

This chapter explains the philosophical position of this study, including the ontological and epistemological perspectives. This is followed by the introduction to mixed methodology, strengths and weakness of mixed methodology and critical exploration of mixed methodology utilised in this study. This will be followed by the research design and methods, including, development of interview guide, pilot study, participant selection method, data collection and authenticity of the research process. The research design flow chart can be found in figure 3. This chapter concludes with the ethical issues addressed in this study, the process of ethical approval and the type of data analysis utilised to interpret the data.

### **Philosophical position**

#### Ontological perspective

Ontology is the study of being; that is, the nature of existence (Gray, 2009). Ontologically, this study initially attempts to find out more about the individual's abilities which is constructed by self-perception and external influence. Additionally, this study explores the reasoning behind the relationship between self-perceived and actual physical functional ability in adults with cerebral palsy. This will help one to understand the complete picture of social phenomenon under study (Gray, 2009) and the realism that exists in the situation (Robson, 2011), based on the study objectives.

#### Epistemological perspective

Epistemology tries to understand what it means to know (Gray, 2009). This study explores the subjective meaning of the individual's interaction with the world and what it means to them (Gray, 2009), therefore the epistemological perspective in this study is subjective.

The ontological and epistemological perspectives are critically explored here.

Philosophically, this research study adheres to the pragmatic paradigm. (Johnson and Onweugbuzie, 2004 and Robson, 2011). The logic of inquiry in this study includes the use of induction, deduction and abduction (Johnson and Onweugbuzie, 2004). The authors define induction as the discovery of pattern, deduction as the testing of theories and hypothesis and abduction as the uncovering and relying on the best set of explanations for understanding one's results. It is an expansive and creative form of research and not a limiting form of research.

Postpositivism, reflects the need to identify and assess the causes that influence outcome, this helps to study the reality that exists in the world (Creswell and Plano Clark, 2007, Creswell, 2009 and Robson, 2011). This is the probable belief when investigating the relationship between the self-perceived performance and actual physical functional abilities in adults with cerebral palsy. The objective here is to find out the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. As indicated in the literature review, there is lack of strong evidence to support the argument that there are changes during adulthood in such population and the relationship between the self-perceived performance and actual abilities were not explored.

Constructivism reflects the individuals' understanding of the world in which they live and work and the individual develops subjective meaning as a result of their experiences (Creswell, 2009 and Creswell and Plano Clark, 2007, Robson, 2011). Here, the truth and meaning do not exist in some external world, but are created by the subjects' interaction with the world and further, the meaning is constructed not discovered (Gray, 2009). The second objective of the study is to explore the participants' views of the situation and their meaning.

In the research based on Postpositivism, the researcher works from 'top down', using a theory to formulate a hypothesis, to data, to finally add to or contradict the theory. In constructivist approaches, the inquirer works from the 'bottom up approach', using the participants' views to build broader themes and generates a theory interconnecting the themes. In pragmatism, the approach may combine deductive and inductive thinking, as the researcher mixes both qualitative and quantitative data (Creswell and Plano Clark, 2007, Robson, 2011). The combination of these worldviews may help us to understand the individual's interaction with the environment and develop guidelines for long term management of the abilities during adulthood in such individuals.

Brannen (2005) has argued that with a logical approach, the association of quantitative research and hypothetic-deduction can be used, depending on the research question and observation. Further, Shilling (2003) has argued that although the body has a material, biological base, this is altered, influenced and possibly modified within different social constructs and so the subjective experience of health and disease cannot be separated from the attempts to explain objectively.

Also, Johnson and Onweugbuzie (2004) and Robson (2011) have suggested that the research methods should follow research questions in a way that offers the best chance to obtain useful answers. Therefore, a researcher should adopt a pragmatic theory to be able to appropriately obtain complete answers to the research questions and the same is explained below.

The first part of the study objective was to find out the relationship between self-perceived and actual physical functional abilities during adulthood in individuals with cerebral palsy. To deduct (test this hypothesis), quantitative approach would be more than suitable.

To understand more about this relationship between self-perceived and actual changes in physical functional abilities in this population, one has to do



induction and abduction, that is, discover the pattern and uncover best set of explanations. Therefore, qualitative approach was utilised to investigate the actual and perceived changes in physical functional abilities in adults with cerebral palsy.

Hence, mixed method approach, adhering to the pragmatic paradigm, was utilised to answer the research question.

Robson (2011, p30) has argued that pragmatism provides a highly compatible theoretical underpinning to mix the two types of method in the same project. One of the advantages of using a pragmatic approach enables the researcher to have a positive attitude to both quantitative and qualitative approaches, consequently qualitative techniques can be used to inform the quantitative aspect of a study and vice versa (Onwuegbuzie and Leech, 2005, p383). For example, the qualitative approach can be utilised to explain the relationship discovered by the quantitative findings. Therefore, from a pragmatist viewpoint, the first research objective, which was to investigate the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy, was addressed using quantitative approach. A qualitative approach was utilised to explain the quantitative findings, which was to explore the possible reasons for, and means to reduce the differences in self-perceived and actual physical functional abilities, as reflected by the second research objective. Integrating the findings of quantitative and qualitative approaches enabled me to address the third research objective, which was to develop clinical guidelines for potential long term management strategies for such individuals.

### **Mixed methodology**

Mixed methods research is the type of research in which a researcher or team of researchers combines elements of research approaches (e.g. use of qualitative and quantitative viewpoints, data collection, analysis, inference

techniques) for the broad purposes of breadth and depth of understanding and corroboration (Brannen, 2005).

Bryman (2012) described 'mixed methods research as a means working with different types of data. It may also involve using different investigators – sometimes different research teams working in different research paradigms'. For these reasons mixed method research is often referred to as multi-strategy research (Bryman, 2012) implying the application of a number of different research strategies related to a range of research questions and a complex research design. Therefore Robson (2011), refers to this type of methodology as multi-strategy design.

Tashakkori and Teddlie (2003) have described the nomenclature, basic definitions and their inconsistencies in detail. Tashakkori and Teddlie (2003) have illustrated that some research studies utilised two quantitative methods or two qualitative methods in the same study to answer the research question. Tashakkori and Teddlie (2003) also noted that studies have sometimes used both quantitative and qualitative approaches for triangulation purpose in the same study. Therefore, there appears to be inconsistencies in defining the mixed methodology. However Brannen (2005), as indicated in the earlier paragraph, has defined mixed methodology incorporating various methods to do mixed methodology. This definition appears to be comprehensive and best suits the paradigm of this research study. The reason for using the mixed methodology in this research study, as suggested by Brannen (2005) is elaborated below.

The study questions identified from the literature review were

- What is the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy?
- What are the possible reasons for and means to reduce these differences in self perceived and actual physical functional abilities?

- How would this impact on the guidelines for potential long term management strategies in such individuals?

The first part of the study objective was to find out the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. To deduce (test this hypothesis), the quantitative approach was utilised as suggested by Gray (2009). To understand more about the possible reasons for and means to reduce the differences between self-perceived and actual physical functional abilities in this population, one has to do induction and abduction (Gray, 2009) i.e. discover the pattern and uncover best set of explanations. Therefore the qualitative approach, namely in-depth interview was utilised to explore the differences in actual (actual changes assessed by the clinician) and self-perceived (changes as perceived by the individual) changes in physical functional abilities in adults with cerebral palsy. Hence the mixed methodological approach was utilised to answer the research questions.

One may argue that the research objectives can be answered using phenomenological research. Phenomenological research focuses on how humans view themselves and the world around them, the research seeking to reveal and convey deep insight and understanding of the concealed meanings of everyday life experiences (King and Harrocks, 2010). Literature review has identified that there is dearth of literature to indicate any relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy (whether they both mirror each other or different). A general pattern of relationship may need to be identified before exploring the meanings (Gray, 2009), this may help to generalise the findings. If the self-perceived and actual abilities were similar, the second study objective would have become obsolete. Therefore mixed methodology, quantitative approach followed by the qualitative approach is the most appropriate methodology to explore the researched area. The findings from the mixed methodology (quantitative and qualitative approaches) will be utilised to explore the third objective,

development of guidelines for potential long term management strategies for such individuals.

### **Role of mixed methodology in health and social care research**

Creswell and Plano Clark (2007) categorised world views towards health and social care research into four categories, namely Postpositivism, Constructivism, Advocacy-participatory and Pragmatism. The researcher's ontological and epistemological perspectives influence the methodological approach in a research study.

In Postpositivism research, the researcher works from 'top down', using a theory to formulate a hypothesis, to collect and analyse data, to finally add to or contradict the theory (Creswell and Plano Clark, 2007). Postpositivism also involves cause-and-effect thinking and testing the theory (Robson, 2011). In positivism, ontological view is that the reality as being composed of clearly formed entities with identifiable properties, however in postpositivism, the research does accept that not all properties are identifiable but are concealed within the reality, influenced by external environment (Tashakkori and Teddlie, 2003).

In constructivist approaches, the inquirer works from the 'bottom up approach', using the participants' views to build broader themes and generates a theory interconnecting the themes (Creswell and Plano Clark, 2007). In pragmatism, the approach may combine deductive and inductive thinking, as the researcher mixes both qualitative and quantitative data (Tashakkori and Teddlie, 2003).

A view held by constructivism ontology is that an individual is influenced vastly by his culture and social world around (Gray, 2009). In healthcare, the experience of one person to any disease is not same as another individual with the same condition, even though the symptoms are similar (Becker and Schaller, 1995). This is vastly influenced by an individual's coping mechanisms, past experiences when interacting with the social world and the

wholesome perspective and knowledge of that individual (Carver and Connor-Smith, 2010). While one may not acknowledge or explicitly know about their interaction with the social world by way of reflection and comparison, an individual's interaction with the environment can be studied to enhance their knowledge and application.

Therefore, to test a theory, one may need to use a quantitative approach and to develop a theory the researcher needs qualitative approach to know more about the social interactions and their responses. Mixed methodology plays an important role for exploration and adding knowledge in health and social care research (Brannen, 2005).

Discussing the evolution of mixed methodology will enable me to critically evaluate its strengths and weakness which will be dealt here. Mixed methodology has evolved as a third methodological movement in the last few decades during the end of paradigm wars.

Creswell and Plano Clark (2007) divided the evolution into four periods, namely formative period, paradigm debate period, procedural development period and recent indicators of interest. Formative period began in the 1950s and continued up until the 1980s. This period found momentum in the 1950s when Campbell and Fiske (1959) advocated for the collection of multiple forms of quantitative data to study the validation of psychological traits. Others also continued and the big question arose as to whether it was possible to combine both forms of data when they were derived from different perspective.

The paradigm debate period was between 1970 and 1980s, when some argued that mixed methods research was untenable because mixed methods asked for paradigms to be combined (Smith, 1983). Rossman and Wilson (1985) described Purists as who could not mix paradigms, Situationists, who adapt their methods to the situation and Pragmatists, who believe that multiple paradigms can be used to address research problems. Procedural

Development period was in the 1980s and the attention shifted to procedures of designing a mixed methods study.

The turn of the millennium has seen a growth in the interest in mixed methodology research, as well as authors advocating for mixed methods research as a separative design in its own right (Tashakkori and Teddlie, 2003 and Creswell et al., 2003). Creswell et al. (2003) and Johnson and Onwuegbuzie (2004) recognised mixed methods as a third approach alongside quantitative and qualitative approaches.

Robson (2011, p163) has argued that positivist paradigm underpins quantitative methods and an interpretivist paradigm underpins qualitative methods, the two methodology are incompatible. However, positivism has long ceased to be a viable option and postpositivism approaches, including the more sophisticated variants of realism, as well as pragmatism, allowing researchers to use mixed approaches. This clearly illuminates the record of emergence of postpositivism from the purist positivism.

Johnson, Onwuegbuzie and Turner (2007) positioned mixed research between the extremes of Plato (quantitative research) and the Sophists (qualitative research), with mixed research attempting to respect fully the wisdom of both these viewpoints while also seeking a workable middle solution for many problems of interest. The authors (Johnson et al., 2007) classify mixed research philosophy as that of pragmatism, defined as an approach to knowledge (theory and practice) that attempts to consider multiple viewpoints, perspectives, positions and standpoints (always including the standpoints of multiple approaches). Mixed methods research is a multilevel synthesis that includes ideas from more than one approach. As in the case of this study, quantitative approach was utilised to identify the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy and qualitative approach was utilised to explore the possible reasons and means to reduce the discrepancies between the self-perceived and actual physical functional abilities; the findings from quantitative

and qualitative approaches were synthesised to develop guideline for potential long term management strategies.

Tashakkori and Teddlie (2003) explained the evolution of mixed methodology in five 'moments': traditional (1900-1950) period, modernist or Golden Age (1950-1970), blurred genres (1970-1986), crisis of representation (1986-1990) and postmodern or present moments (1990 –present). When the purist questioned the mixed methodology on philosophical level, Howe (1988) explained the use of multiple paradigms as a philosophy of Pragmatism.

These arguments from Robson (2011), Johnson et al. (2007) and Tashakkori and Teddlie (2003) that mixed methodology has positioned itself between quantitative and qualitative approach, justifies the evolution of mixed methodology as a third methodological movement. Further, the strengths of mixed methodology that offset the weakness of separately applied quantitative and qualitative research methods (Robson, 2011) which are elaborated in the next section. Mixed methodology is important today, because of the complexity of problems that need to be addressed in health and social care services and the practical need to gather diverse data for a wide range of audiences including policy makers, service users and politicians.

### **Strengths and weakness of mixed methodology**

Johnson et al. (2007) has suggested that researchers attempt to eliminate potential design weakness by combining methods that have different weakness. They suggested that the data should be collected to provide all of the information that is potentially relevant to the purpose of the study, but this may consume more time than necessary in some cases. Qualitative and quantitative research if used appropriately to answer a research question, together produces more complete knowledge, necessary to inform theory and practice (Johnson and Christensen, 2004). A major advantage of mixed methods research is that it enables the researcher to simultaneously answer confirmatory and exploratory questions, and therefore generates and verifies

theory in the same study (Tashakkori and Teddlie, 2003), however the purists do not appreciate this view point. Furthermore, sometimes larger team and more skilled researchers may be necessary to conduct this type of research.

Mason (2006) has stressed the weakness of mixed methodology, in the real world; practical, political and resources issues will establish certain constraints and contexts for those wishing to carry out mixed methodology projects. However, it is important to recognise how these factors play out in one's own real life research, as it is to be clear about the desired strategy for mixed methods.

Therefore, analysing the strengths and weakness, it is clear that the strengths outweigh the weakness if proper attention is given to the procedure and the overarching paradigm. As Tashakkori and Teddlie (2003) have suggested, complex social phenomena cannot be fully understood using either purely qualitative or purely quantitative approaches, we need multifaceted institutions, like mixed methodology to do this. Utilising purely quantitative or qualitative approaches would not have addressed the study aim, the strengths of each approach is utilised to address the research objective, this is elaborated furthermore in the forthcoming sections. The methodology utilised by other authors pertaining to physical functional abilities in adults with cerebral palsy is critically evaluated here, before discussing and justifying the appropriate methodology for answering the study objectives in this study.

### **Methodology utilised by other authors to study the physical abilities in adults with cerebral palsy**

The methodological approach utilised by researchers to study the physical functional abilities in adults with cerebral palsy is critically explored in this section.

The literature review completed for this study revealed that the authors primarily adopted a quantitative approach to study the changes in physical abilities in adults with cerebral palsy, using a cross sectional self-reported



survey design (Murphy et al., 1995, Bottos et al., 2001, Turk et al., 2001, Andersson and Mattsson, 2001, Jahnsen et al., 2004a, Sandstorm et al., 2004 and Morgan et al., 2014). However, no studies were found that focused on the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy and the reasons behind these discrepancies.

Cox et al. (2005), Haak et al. (2009) and Morgan and McGinley (2014) reviewed the studies pertaining to cerebral palsy and ageing. Haak et al. (2009) suggested that the physical abilities can be studied by adopting a biopsychosocial International Classification of Functioning (ICF), Disability and Health framework, involving multilevel research teams and by including input from adults with cerebral palsy, the ICF Biopsychosocial model is illustrated in figure 2. Morgan and McGinley (2014) recommended prospective studies incorporating qualitative and quantitative methodologies to enhance the knowledge regarding self-efficacy and individuals' choices that influence change in mobility over time. Furthermore, Cox et al. (2005) reviewed ageing in cerebral palsy and concluded that a large longitudinal cohort study is necessary to monitor the primary and secondary effects of ageing with cerebral palsy over time. Cox et al. (2005) also suggested that 'an expansion of the rich data derived from qualitative studies in this review would also be of value in providing an in-depth understanding of the effects of the processes involved'. Consideration of quality of life, emotional and psychological impact of living with cerebral palsy was suggested to be incorporated in future research studies.

As inferred in the literature review, research exploring individuals' perception about changes in physical functional abilities in adults with cerebral palsy is minimal. Increased life span due to medical advancements (Bottos et al. 2001), decreased cognitive abilities in some individuals and an absence of a national register (Cox et al. 2005 and Pharoah et al. 1997) to follow up these individuals may have been contributory to this lacuna. Furthermore, the evidence pertaining to changes in physical abilities in adults with cerebral

palsy has only surfaced in the recent past. Bottos et al. (2001) and Strauss et al. (2004) have pointed out that there has been less attention paid to adults and elderly persons with cerebral palsy when compared to the research available for children and also that there is less organised health care for adults when compared to children. One of the possible reasons for this may be that the potential for progress declines in adulthood when compared to that in the children. It was therefore accepted that the changes in physical abilities were part of the natural ageing process rather than studying the factors causing the changes and exploring strategies to reduce these changes. The changes in physical abilities may need to be rigorously studied by using a qualitative approach. This may help the researchers in exploring the individual's perception which would help in designing strategies for self-management.

### **Research question and mixed methodology**

Sackett and Wennberg (1997) have argued that each methodology should flourish, because each has features that overcome the limitations of the others when confronted with questions they cannot reliably answer. Tashakkori and Teddlie (2003) have stated that pragmatist researchers consider the research question to be more important than either the method they use or the paradigm that underlies the method. Therefore, the question should always consider which methodology best suits to answer a research question/s rather than debating the hierarchy of research methodology.

To understand the changes in physical functional abilities during adulthood in cerebral palsy, one needs to know more about both the actual abilities and self-perceived performance (Hanna et al., 2009). Actual abilities are the abilities that the individual can do, which are the individual's real potential. The individual's perception is essential as it will have a major impact on their self-image and daily activities, which are their perceived performance. It is not possible to quantify objectively the self-perceived and actual abilities using a qualitative approach. One may use qualitative approach to understand more

about the self-perceived ability per se but not the actual abilities which are assessed by the health care professional. Objective measures are best suited to quantify the actual abilities assessed by the health care professional in order to be non-biased. The study objective here was to identify and compare self-perceived and actual physical functional abilities in adults with cerebral palsy (deduce the relationship), therefore, quantitative approach was best suited to answer this study objective. Quantitative approaches would support testing of research question, striving for general laws in these individuals' and theories through which we can understand the social world (Robson, 2011). This helped us to identify the relationship between self-perceived and actual physical functional abilities quantitatively.

This was followed by the second phase, which was the qualitative approach. Exploration (induction and deduction as elaborated by Gray, 2009) of the discrepancies between the actual and self-perceived physical functional abilities in an individual is complex and this generates rich subjective data, therefore qualitative approach would be more than appropriate. This approach would generate subjective meaning, situated and embedded in the local context (Johnson and Onwuegbuzie, 2004), while the quantitative approach would not be able to explore the subjective meaning. As suggested by Robson (2011) an interview allows one to acquire multiple perspectives, in order to understand social constructions of meaning and knowledge. Therefore, the second study objective was to explore the possible reasons and means to reduce these differences in actual and self-perceived physical functional abilities through an interview.

Furthermore, the interview process would also give an opportunity to the participants, to tell us the means through which they may want to reduce these differences and maintain their abilities throughout their life span. This will help to answer the final research question which was to review possible models for self-management for such individuals.

## **Conclusion**

Therefore, mixed methodology was chosen as an appropriate approach to address the study objectives. The quantitative approach and qualitative approach helped to address the study objective one and two, providing a rich overall view. The findings from both the approaches was utilised to address the third study objective, which was to develop guidelines for potential long term management strategies in such individuals and thereby promote empowerment in these individuals to self-manage and improve quality of life.

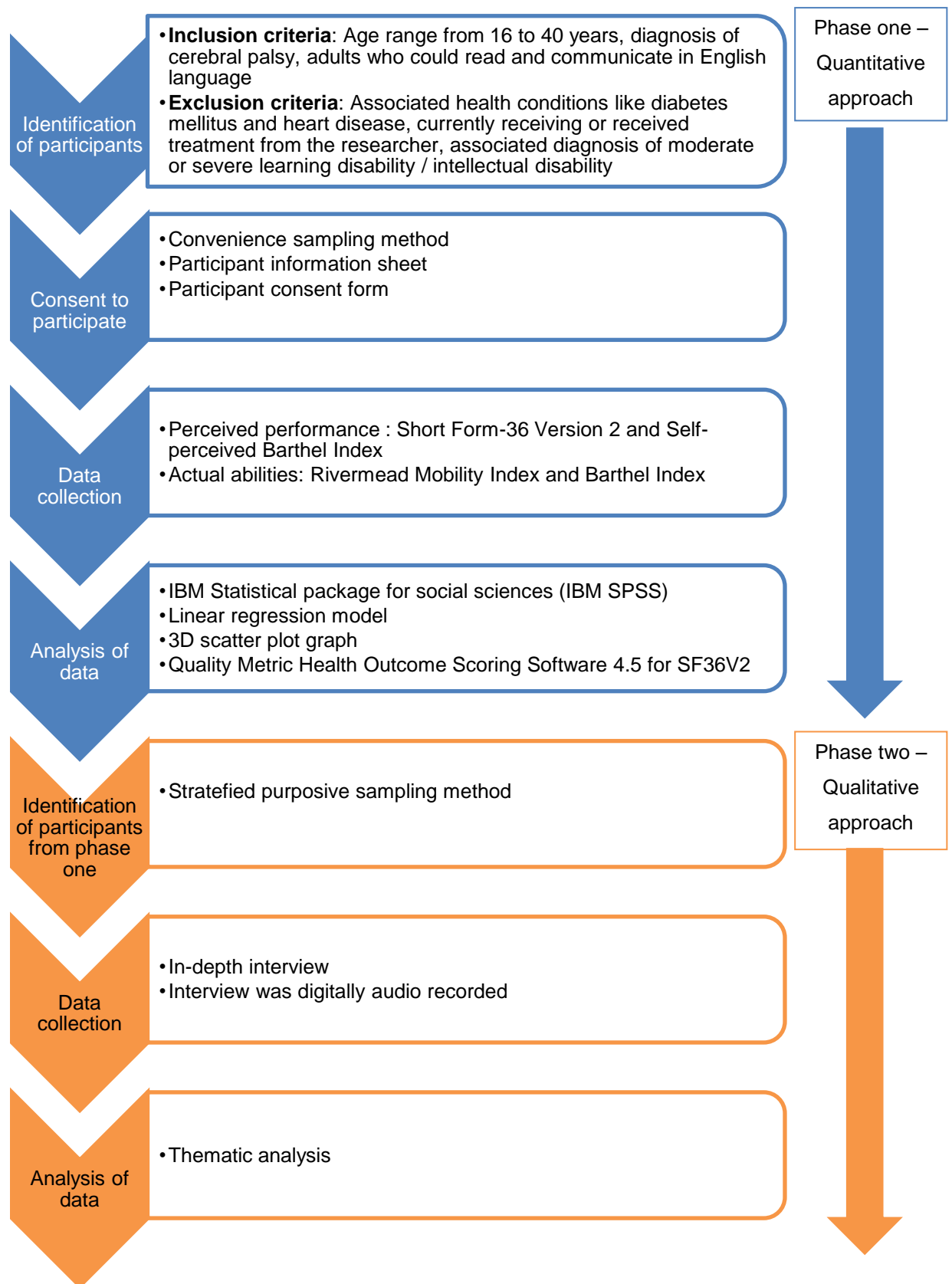
## **Research design and methods**

Creswell et al. (2003) has explained the research designs for mixed methodology, following the exploratory work of Bryman (2012). Creswell et al. (2003) has indicated six major research designs for implementing mixed methodology in research studies, sequential explanatory design, sequential exploratory design, sequential transformative design, concurrent triangulation design, concurrent nested design and concurrent transformative design. Research designs best suited for the study follows the research objective under pragmatist philosophy. As elaborated in the methodology, the first study objective was more suited to be analysed using quantitative methodology and the qualitative methodology was utilised to explore the results from quantitative findings to address the second research question. Based on the findings of both the methodologies, third study objective was explored.

This study conforms to the mixed methodology sequential explanatory design as proposed by Creswell et al. (2003) and consists of two phases. This is depicted in figure 3. Sequential explanatory design is characterised by the collection and analysis of quantitative data followed by the collection and analysis of qualitative data, the approaches were integrated at the interpretation stage. The purpose of this design was to use the qualitative results to assist in explaining and interpreting the findings of quantitative

approach (Creswell et al., 2003). The major strength of this design is that the findings from both the approaches can be utilised in discussion, however the weakness of this design is the length of time involved in data collection to complete the two separate phases (Creswell et al., 2003), which was acknowledged in this research.

**Figure 3:** Research design flow chart – Sequential explanatory design



## **Phase one**

In this study, the first study objective was investigated through quantitative data collection and analysis. Self-perceived and actual physical functional abilities were measured using four quantitative data collection tools. The ideal method to investigate the self-perceived and actual physical functional abilities would be to conduct a longitudinal study (Cox et al., 2005). This would involve studying all the adults with cerebral palsy from the age of 16 years and continuing the research throughout their life which would involve considerable resources outside the scope of this study. However, as an alternative, I adopted a cross sectional design, where the individuals were chosen from different age groups for appropriate representation of the population, however this design can only reveal the association amongst age variables and cannot reveal the causation, since it's a interrupted time-series design (Gray, 2009, p147). Therefore, due to the limited resources, cross sectional design was chosen to answer the first objective to investigate the relationship between the self-perceived and actual physical functional abilities during adulthood in individuals with cerebral palsy using quantitative approach.

### **Tool selection:**

There are a number of factors that may have an impact on the management of cerebral palsy in adulthood. As indicated in the literature review, Zaffuto-Sforza (2005) has attempted to summarise these factors and suggested that pain, musculoskeletal deformities, arthritis, nerve entrapments, overuse syndromes, fractures, fatigue, general effects of aging on different body systems, dental issues, nutrition and diet, speech and swallowing problems, hearing and vision, exercise, social issues, adaptive equipment and life expectancy being the key factors that impact on the management of adults with cerebral palsy. However, the evidence available to illustrate the intensity of factors impacting directly on the physical functional abilities during adulthood in individuals with cerebral palsy is sparse. From the available evidence, the contributing factors to decline in physical functional abilities

could be summarised into three categories: Physical, Environmental and Psychological. The factors evident from the available literature are categorised in Table 3 in the literature review chapter. The interaction between these factors does follow a bio-psycho-social model, as explained in The International Classification of Functioning, Disability and Health (ICF) (Hanna et al., 2009). The ICF's biopsychosocial framework describes health and disability from the interactions among: body structure and function, daily activities, participation, environmental factors and personal factors (Haak et al., 2009), this is illustrated in Figure 2. Haak et al. (2009) concluded that interactions in these areas are likely to affect each other and contribute to a person's health. Therefore, I have chosen standardised tools to measure these categories.

The literature review in this study has identified the tools utilised by researcher for measurement of physical functional abilities in adults with cerebral palsy, this can be found in the Table 2, a summary of reviewed articles. Classifying functional abilities and limitation to participate in different domains will improve our understanding of individuals with cerebral palsy (Haak et al., 2009). Therefore, Haak et al. (2009) suggested that tools suitable to capture the wide range of adult participation in function are required when researching in this area. From the tools used by these studies, it is evident that the standardised tools, Short Form-36, Barthel Index and Rivermead Mobility Index will help to quantify the self-perceived and actual abilities in physical functional abilities. Gross Motor Functional Measure-66 was considered to measure actual physical functional abilities, this measures the gross motor and assists with assessment and intervention planning (Rosenbaum et al., 2002, Hanna et al., 2009). This measure consists of 66 items and measures lying, rolling, sitting, crawling, kneeling, standing, walking, running and jumping on a 4 level ordinal scale, however this measure was only validated up to the age of 21 years in individuals with cerebral palsy. Therefore, this measure was not utilised in this research. There is no single tool to measure the physical, environmental and psychological factors contributing to or impairing physical functional



abilities (Haak et al., 2009). Velentgas, Dreyer and Wu (2013) have argued that multiple scales can be used when single scale cannot adequately capture the perspective under study.

Hence, I selected two tools each to assess self-perceived and actual physical functional abilities in adults with cerebral palsy, namely Short Form-36 version 2, Self-reported Barthel Index, clinical assessed Barthel Index and Rivermead Mobility Index.

Short Form 36 version 2 (SF36v2) and Self-reported Barthel Index (SR-BI) were utilised to study the self-perceived physical functional abilities. A copy of Short Form 36 version 2 and Self-reported Barthel Index can be found in appendix 2 and appendix 3 respectively.

The Rivermead Mobility Index (RMI) and the Barthel Index (BI) were utilised to assess the actual physical functional abilities in adults with cerebral palsy. A copy of Rivermead Mobility Index and the Barthel Index can be found in appendix 4 and appendix 5 respectively.

The Short Form 36 version 2 (SF36v2) is a self-reported health survey questionnaire and encompasses only some components of the International Classification of Functioning, Disability and Health domains. This survey measures the functional health and well-being from the patient's point of view (Ware and Sherbourne, 1992). The SF36v2 provides scores of each of the eight health domains and psychometrically-based physical component summary and mental component summary. It is a practical, reliable (cronbach's alpha greater than 0.85, reliability coefficient greater than 0.75 for all dimensions except social functioning) and a valid tool (significantly distinguishes between groups with expected health differences) to study the physical and mental health in the British population (Brazier et al., 1992). Further, this scale was also able to detect low levels of ill health (Brazier et al., 1992). This tool does not evaluate feeding, walking, bathing, toileting and transfers. Therefore, a self-reported version of Barthel Index, which

incorporates the domains of feeding, walking, bathing, toileting, transfers and grooming was utilised. This was primarily developed for postal surveys (Gompertz, Pound and Ebrahim, 1994) and demonstrated good reliability. Gompertz et al. (1994) demonstrated test retest reliability mean of -0.5, standard deviation of 2.1 and sensitivity to show change in patients with stroke. Preliminary studies have suggested a high correlation between the self-reported BI and Functional Independence Measure and have been assessed to be a reliable tool (Hobart and Thompson, 2001, O'Connor et al., 2005, Morley et al., 2012). Morley et al. (2012) demonstrated cronbach's alpha coefficient of 0.69, indicating adequate internal consistence, reliability coefficient was 0.74 and validity was demonstrated through significant correlation with daily living dimension of the Parkinson's disease quality of life questionnaire in adults with Parkinson's disease.

The Barthel Index (BI) of Activities of Daily Living (ADL) is a tool used widely in the clinical setting to quantify the independence in day to day activities. The index consists of 10 items of which eight items represent common ADLs and two are related to mobility (Mahoney and Barthel, 1965). It has been assessed to be reliable and valid in studies published by Collin et al. (1988), Wade and Collin 1988), Loewen and Anderson (1988) and Hobart and Thompson (2001). Hobart and Thompson (2001) demonstrated excellent correlation in adults' neurological rehabilitation with Functional Independence Measures (motor component). Although SR-BI and BI appear to be that it is more sensitive to change (where more than one timeline is measured in the individual) in acute rehabilitation rather than in person with long term disability (O'Connor et al., 2005), this limitation did not affect this tool here, since the participants were assessed only once using this tool. Sutton et al. (2013) has demonstrated significant difference between SR-BI and BI with mean scores 1.0 (95% CI, 0.5 – 1.6) higher on the face to face scoring (clinical assessment) and speculated that participants are more likely to self-score accurately on the SR-BI for some items due to embarrassment in face to face assessments, which was acknowledged in this study.

The Rivermead Mobility Index (RMI) assesses aspects of mobility, independent of one's social environment in a standardised setting and provides a measure of fifteen items that progress in difficulty, with a two point scoring for each item. It has been assessed to be a reliable and valid tool in neurological conditions in studies by Collen, et al. (1991), Forlander and Bohannon (1999) and Franchignoni et al. (2003). Furthermore, Franchignoni et al. (2003) demonstrated good internal consistency (Cronbach's alpha 0.92) and excellent correlation with timed walk test in patients using prosthesis ( $r=0.70$ ). Within this research study, the BI and RMI was utilised to quantify all components of actual physical functional abilities.

## **Second phase**

The second study objective was to explore the possible reasons for and means to reduce the differences between the self-perceived and actual physical functional abilities. The standardised tools used in the quantitative phase may help to identify the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy, but would not assist in exploring the possible reasons for and means to reduce the differences between the self-perceived and actual physical functional abilities in this population (causal factor). The data analysis of the phase one, informed and guided the identification of the participants for qualitative phase two, this is explained furthermore in participants selection section. The exploration of the differences between the self-perceived and actual physical functional abilities in an individual, is complex and generates subjective data, therefore a qualitative data collection was considered to be appropriate. As explained earlier, this approach will generate data which is situated and embedded in the local context (Johnson and Onwuegbuzie, 2004). To explore the physical functional abilities in this population and the means by which they would like to reduce the impact on long term disability, in-depth interviews was undertaken.

An alternative approach to the interview, would be through Ethnographic observation or focus group interviews, however these approaches would not enable clarification of the individual's unique perception (Hansen, 2006). Ethnographic interview is mainly participant observation strategy, this type of interview is based on the open framework model and involves combination of friendly conversation and formal in-depth interview (Flick, 2009). Unstructured interview can become labour intensive and lose focus (Hansen 2006); therefore, a semi-structured interview was conducted to stay focused on the relationship between the self-perceived and actual abilities in physical function. The interview focused on the reasons around self-perceived performance, relationship between the self-perceived and actual abilities and how the participants would like to reduce these differences, so that their day to day performance matches their actual abilities. This type of interview allowed for probing of views and opinions enabling the researcher to explore subjective meanings that respondents ascribe to concepts or events (Gray, 2009). The interview guide is attached in appendix 6, the development of this guide is elaborated in the next section. This was adapted to each individual before the interview, based on the phase one individuals' raw data and analysis. As explained by Creswell et al. (2003) the interview assisted in explaining and exploring the findings from quantitative phase one. King and Horrocks (2010) have also highlighted the need to change the interview guide in the course of the study in order to elicit participants' accurate aspects of their experiences rather than to collate answers to specific questions. Further, the interview helped to explore the psychosocial factors like motivation, fatigue, leisure opportunities and personal circumstances that may contribute to the changes in physical functional abilities and how they would like to manage their health needs. Psychosocial factors were considered as an important factor as they may have a direct impact on physical functional abilities whilst the previous studies have failed to explore this component (Hanna et al., 2009).

## **Development of interview guide and pilot study**

An interview guide outlines the main topics the researcher would cover, but is flexible regarding the phrasing of questions and the order in which they are asked, and allows the participant to lead the interaction in unanticipated directions (King and Horrocks, 2010). The interview guide was based on the key sources as suggested by King and Harrocks (2010); personal experience of the research area and literature review. Utilising this approach, key questions initially drafted for the interview guide were:

*What do you think about your performance during the assessment?*

*Why do you think your perception of abilities were different to the abilities assessed?*

*What do you think you can do to reduce the differences between self-perceived abilities and actual physical functional abilities?*

*Do you need any help to manage these challenges?*

Drawing upon the resources from King and Harrocks (2010) and Rubin and Rubin (2005) and reflecting on the questions with support from the supervisors, the interview questions were changed to be non-directional and offer the flexibility for participants to respond. These questions were re-framed as:

*Tell me how you think you performed on the assessments?*

*Tell me what do you think about the differences between your actual performance and what you said you could do?*

Furthermore, to improve competency of the researchers' interview skills and to ensure that the guide enabled the participants to respond comprehensively on the researched area, pilot interviews (Rubin and Rubin, 2005) were conducted involving a colleague and a potential participant. The pilot study helped to phrase the questions in a clear, concise, unambiguous and free

from jargon (Gray, 2009). The feedback from the pilot interviews was incorporated in the interview guide.

One potential participant gave a feedback that:

*'The initial information regarding the assessment tools were brief, if I was asked to explain it further, it may have been easier to reflect upon before responding comprehensively'.*

Therefore, the initial question in the interview guide was refined to give an opportunity to elaborate on the difficulties encountered when performing the daily tasks.

The colleague suggested rephrasing a question to be clear and simple so that the participant can reflect upon, this feedback led to addition of a new question, which was,

*Tell me if anything stops you from doing some of these tasks?*

Testing the interview guide helped the researcher to approximately quantify allocation time for interview data collection. Also, probing questions were identified during this process to add depth to the interview data (King and Harrocks, 2010).

These initial interviews were transcribed as a trial. During transcription, some difficulties were encountered listening to the researcher and potential participants' responses due to lack of volume. Therefore, the positioning of the digital audio recorder was altered to capture the responses clearly. The refined interview guide can be found in appendix 6.

Robson (2011) has described the pilot study as a 'dummy run', which helps to identify inevitable problems of converting the design into reality. Additionally, Leon, Davis and Kraemer (2011) have suggested that pilot studies are an essential first step in exploring novel idea, this informs feasibility, which in turn, points to modifications needed in the planning stage before implementing

the plan. Upon reflection, I also completely agree with Robson (2011) and Leon et al., (2011) that the pilot study was very helpful in refining the interview schedule, as elaborated in the earlier paragraphs and to upskill my interview techniques.

### **Participant selection**

For the phase one of this study, non-probable convenience sampling was utilised to identify the potential participants from the available case records and review clinic lists within two NHS services in the North of England. In convenience sampling, the participants are selected purely on the basis that they are conveniently available; the major disadvantage being that there is no way of telling how representative the participants are of the population under study (Gray, 2009, p153). In healthcare, due to access and limited resources, convenience sampling is an accepted methodology (Sedgwick, 2013). However, to avoid researchers' bias, the potential participants were identified by the 'gatekeepers'; the consultant in site 1 and the management team in site 2 were the 'gatekeepers'. All potential participants were accessing the health service during the study period.

Any adult with the diagnosis of cerebral palsy, between the age of 16 and 40 years and who could read and communicate in English language was selected for the phase one of the research. Since the literature review demonstrated that there may be significant effect of aging between 30 years and 40 years in adults with cerebral palsy (Murphy et al., 1995, Bottos et al., 2001, Turk et al., 2001, Andersson and Mattsson, 2001, Sandstrom et al., 2004, Jahnsen et al., 2004a, Cox et al., 2005, Hanna et al., 2009 and Haak et al., 2009), I choose to study the physical functional abilities between 16 years and 40 years. The upper age limit of 40 years was chosen to minimise the effect of arthritic changes on physical functional abilities (Arnett et al., 1988) which has significant impact beyond the age of 40 years. Participants with diagnosis of moderate or severe learning disability/intellectual disability or associated health problems such as diabetes mellitus, heart disease and

other neurological disorders or if they are currently being treated by the researcher were excluded from this study to prevent any undue stress and any conflict of interest respectively.

During the planning state, the proposal was to include eight participants from each subgroup of age range 16 to 20 years, 21 to 25 years, 26 to 30 years, 31 to 35 years and 36 to 40 years for appropriate and even representation of the age range. Unfortunately, due to various reasons outlined in the 'Study Limitations' section, in chapter seven, I was able to recruit only seventeen participants for phase one of this study. Due to the fewer number of participants an even distribution of participants through the age range was not achieved.

The data was collected from two sites. At site 1, I informed the consultant about my intention to conduct the research. The potential participants were identified by the Consultant from his clinic list based on the inclusion and exclusion criteria. The consultant posted the participant information sheet, participation invitation letter and participant consent form to the potential participants on my behalf, at least three days prior to their attendance at the review clinics for a robust memory recall (Gray, 2009). The contents of the informed consent form are discussed in ethical consideration section and a copy of the informed consent can be found in appendix 9. I attended the Consultant's review clinic at site 1, when the potential participants visited the clinic. If the potential participants were willing to participate in the research, a meeting was initiated in the adjacent clinic room for subsequent assessments. It was ensured that the participants have thoroughly understood the information sheet and are willing to participate in the research.

At site 2, I informed the management team about my intention to do this research and requested them to give a copy of the participant information sheet, participant invitation letter and participant consent form to the potential participants when reviewing the individuals in the community and requested to contact me if they wish to participate in the research. The participant



invitation letter for site 2 was different to the site 1 invitation letter since the gatekeepers were from different team. The participant invitation letter for site 1 and site 2 can be found in appendix 7. If the potential participations were willing to participate in the research or wanted more information, they were invited to the hospital. During the meeting it was ensured that the participants understood the information sheet thoroughly and were willing to participate in the research.

Copies of participant invitation letter, participant information sheet and participant consent form can be found in appendix 7, appendix 8 and appendix 9 respectively.

### **Sample size**

As there are no previous studies, sample size was calculated pragmatically, based upon feasibility and logistical management of participant recruitment (Brannen and Halcomb, 2009). Forty participants were estimated as the minimum sample size in order to analyse the self-perceived and actual physical functional abilities in adults with cerebral palsy. Unfortunately, due to various reasons documented later in the chapters, I was able to recruit only seventeen participants for phase one of the study, this limited the research analysis and findings. Nevertheless the findings from this small group of participants cannot be ignored and can be extrapolated to the wider group of individuals with cerebral palsy.

### **Date collection**

#### *Phase one*

Potential participants were selected on the basis of inclusion and exclusion criteria elaborated in the earlier participant selection section. Potential participants willing to participate in the study signed the participant consent form. As elaborated in the ethical section, the researcher ensured that the participants understood the contents of the participant informant sheet and consent form by requesting the participants to verbally summarise the

information after reading the forms. This helped to safeguard the participants and decide whether they understood the information before making the decision to participate in the study. At this stage, all participants were given a study number which was recorded in all documents from this stage to preserve anonymity and for audit trail. Demographic information regarding diagnosis, age, gender, symptoms associated with their condition, information regarding any surgeries in the past and comorbidities were collected and recorded.

To minimise the effect of actual ability testing on individuals' perception of their ability, the data from self perceived abilities measurements (SF36v2 and SR-BI) was collected prior to the collection of data from the actual abilities measurements (RMI and BI). A copy of Short Form 36 version 2, Self-reported Barthel Index, Rivermead Mobility Index and the Barthel Index can be found in appendix 2, appendix 3, appendix 4 and appendix 5 respectively. Researcher was familiar with these tools, since these tools were regularly used by the researcher during professional practice.

The self-perceived and actual physical functional abilities was statistically analysed using the software, IBM Statistical Package for Social Sciences (IBM SPSS) version 24 (May 2016). The type of analysis utilised in phase one is detailed later in the data analysis section.

#### *Phase two - Interview*

For the phase two of the study, a stratified purposive sampling method (representation from the age groups) (Kemper, E.A., Stringfield, S. and Teddlie, C., 2003, p282) was conducted to identify a minimum of three participants from the phase one participants. A stratified purposive sampling technique involves dividing the purposefully selected target population into strata with the goal of appropriate representation across the age groups (Kemper et al., 2003, p282). Individuals who would provide a full and sophisticated understanding of the phenomena under study (Hansen, 2006)

and have largest statistical differences between the self-perceived and actual physical functional abilities were chosen for the phase two. As stated by Hansen (2006), this involved careful consideration of the aims of the study and thoughtful decision making about the desired range, characteristics and number of the research participants. Creswell et al. (2003, p227) has also explained that in a sequential explanatory mixed method designs, the quantitative results can be used to guide the purposeful sampling of participants for a qualitative interview and this allowed the researcher to explore the quantitative findings. Therefore for the interview, a minimum of three participants were chosen from the population who participated in the phase one of the research using stratified purposive sampling method.

Rubin and Rubin (2005) has suggested that interview relationships are intentionally guided through a series of liked stages; introduction of the researcher and the topic, including building the confidence of the interviewee and establishing trust; asking some easy questions and showing empathy; followed by challenging questions; finally, the intensity of discussion is toned down with a closing statement. These steps were followed during the interview process to obtain in-depth information.

The interview guide (a copy can be found in appendix 6), was used to ask the main question. At certain times, follow up questions and probing questions were asked, however leading questions were avoided to prevent the interviewee feeling some degree of pressure to conform to what appears to be expected of the interviewer (King and Horrocks, 2010). For example, an interview section from one participant is highlighted here to explain the main and probing questions.

*Interviewer (from interview schedule): Tell me what do you think about the differences between your actual assessment and what you said you could do?*

*Participant: I am not sure why, it's just that it gets stiffer sometimes.*

The participant paused at this stage and was not sure what to say. Therefore, this was followed with a probing question.

*Interviewer (probing question): Can you tell me why it's different? Is it easy or difficulty to do the tasks at home?*

The probing question encouraged the participant to elaborate the reasons for the differences between self-perceived and actual physical functional abilities, which were richer information.

### *Interview Recording*

Recording the interview is essential to capture the entire conversation and analyse at a later stage (Rubin and Rubin, 2005). It is extremely difficulty to take complete notes of the entire conversation without losing continuity; therefore, the interviews were digitally recorded using an audio recorder. Some notes were taken during the interview to note down the essential themes for follow up questions and also to capture the non-verbal gestures during the process (Rubin and Rubin, 2005). All the participants consented for audio recording of the interview and it was observed that they became more comfortable as the interview progressed.

### **Authenticity**

Authenticity of the research is determined by two basic criteria: rigor in the application of method and rigor in interpretation (Lincoln and Guba, 2000). Rigor in application relates to the design quality, which comprises the standards for the evaluation of the methodological rigor (Tashakkori and Teddlie, 2003). Critical analysis of the philosophical position, ontological and epistemological perspective lays the foundation of methodological rigor (Tashakkori and Teddlie, 2003), which is discussed in the earlier chapters. Furthermore, the feasibility of the methodology, including participant recruitment was discussed with a Consultant, colleagues and Neurology User and Carer Forum (forum for the service users). Constructing the interview

process based on the strategies proposed by Rubin and Rubin (2005), improved the rigor of data collection. The process of ethical approval also authenticated the rigor in the application, this is elaborated later in the ethical approval section.

Rigor in interpretation refers to the standards for the evaluation of the accuracy or authenticity of the conclusions (Tashakkori and Teddlie, 2003). Reading the interview notes and promptly typing them out in full, improved the meaning of interpretation (Rubin and Rubin, 2005). The audio recordings were transcribed verbatim to avoid any distortion (Sutton and Austin, 2015) by the researcher and identifiable information was removed. Pauses and non-verbal gestures from the field notes were incorporated in the transcript. To retain authenticity of the interview, the typed transcript was sent to the relevant participant for member validation (King and Horrocks, 2010). An example of coding and theme identification from the interview transcript can be found in the data analysis section.

My research supervisors were involved from the planning stage to ensure rigor throughout the research study through regular tutorial sessions, where perspectives and issues were explored. Reflexivity is an important part of the research process, and as suggested by Snowden (2015) a research diary was kept throughout the process. Researcher's reflexivity can be found in the concluding chapter.

### **Ethical considerations**

The ethics of research concern the appropriateness of the researcher's behaviour in relation to the participants or those who are affected by it (Robson, 2011). Ethics is invariably involved when involving participants in the real world. Blumberg, Cooper and Schindler (2005, p92) illustrated that ethics, are set of moral principles or norms that are used to guide moral choices of behaviour and relationships with others. As indicated by Gray (2009), the four ethical principles were considered from the preparatory

stages, namely, to avoid harm to participants, to ensure informed consent of participants, to respect the privacy of participants and to avoid the use of deception. In this research, ethical issues considered throughout the research are discussed here.

### *Informed consent*

The consent form was formulated using the example template provided by Robson (2011), NHS ethical requirements and recommendations from School Research Ethics Panel, University of Huddersfield. A copy of the participant consent form can be found in appendix 9.

Robson (2011) has elaborated the ethical steps involved in obtaining consent from the participants: explain to the participants what the study involves, let them know they can have time to think about participation, provide participants with a consent form and check and double-check with participants they fully understand the research, their role in the study and any implications it has for them. These steps were following when obtaining informed consent from the participants in this study.

Additionally, the researcher completed an e-learning course in 'Introduction to Good Clinical Practice (GCP)', which is a practical guide to ethical and scientific quality standards in clinical research, provided by National Institute for Health Research. The ethical principles elaborated in the course were acknowledged when obtaining consent from the participants to partake in this study, data collection stage, safety of the individuals and documentation of the data. The GCP e-learning course certificate can be found in appendix 10.

Informed consent was obtained from all the participants who initially received the information sheet which included information about the study, possible risks, data protection, confidentiality, anonymity and right to withdraw without consequence. The information sheet was read aloud to two participants due to difficulty in reading. Since the participants were vulnerable adults, it was ensured that they understood the information sheet and consent form by

requesting the participants to verbally summarise the information after reading the forms. This helped to safeguard the individuals and decide whether they have the capacity to decide about participation in this study (Robson, 2011). A copy of this participant information sheet and the letter sent to the General Practitioner and/or the participant's consultant, can be found in appendix 8 and appendix 11 respectively. It was ensured that coercion of participants was avoided at all stages of this research.

### *Emotional Distress*

Gray (2009, p86) has suggested that a researcher can cause physical, psychological or emotional harm or even just embarrassment during the research process. It was expected that that the emotional distress and service expectation may be heightened during the in-depth interview process (Gray, 2009, p86) and therefore nonmaleficence, 'no harm to participants', was considered as a priority. This was minimised by stating clearly the purpose of research and access to other services in the information sheet. The participants were informed that they have the opportunity to pause/stop the interview process, if the in-depth interview caused any emotional distress. Referral to the General Practitioner for support from the Psychological services was considered if necessary, however no participants indicated emotional distress during or after the interview process.

### *Health and safety risks to participants and the researcher*

Ethical principles rightly stresses on the potential risks to the participants in the research and the researchers themselves (Robson, 2011). Hence, it was anticipated that some participants may have balance problems and may be prone to slips and trips during the assessment. This was minimised by assessing the needs of the participants and the environment before starting the procedure. The assessment was completed at the physiotherapy room or in a clinic room which were regularly used for assessment. There were no visible obstructions or equipment present in the space where the assessment

took place. The researcher had completed the mandatory manual handling course in accordance with the NHS practice guidance. The researcher is experienced in handling the participants with cerebral palsy during routine physiotherapy interventions as part of the job role within the NHS and using the assessment tools within professional practice. The University of Huddersfield risk analysis and management plan for the sites can be found in appendix 12.

#### *Conflict of interest*

The researcher job role involved working with the potential participants, it was anticipated that the relationship between the researcher and participants may be influenced if the participant was already on the researcher's caseload (Flick, 2009). Therefore, only participants who were not part of the researcher's caseload were chosen for this study to avoid conflict of interest.

#### *Privacy and security*

To protect the privacy of the individuals, all data was collected in a private and safe environment within a hospital setting and stored safely in a locked cabinet at the researcher's workplace. All participants were given a number after obtaining consent and this number was used throughout the data collection for audit trail. Personal information was retained as a separate file to preserve privacy (Robson, 2011) and only the researcher had access to this information. All electronic data was stored in encrypted files on a password protected computer or storage devices when transported. Audio tapes and the interview transcripts will be retained in a safe locked cabinet at researcher's workplace for future reference until the completion of research and dissemination. I am planning to shred all the data in a safe environment after 5 years of completion of the research as recommended by the University of Huddersfield. Securely stored electronic data will also be securely deleted from the computer and the storage device at the end of 5 years.



## *Anonymity*

Giving anonymity to participants when reporting on research is the norm (Robson, 2011), therefore, when using quotes from the individuals' interview transcripts in results and discussion, anonymity was maintained. Whilst maintaining anonymity, participants possessing a combination of attributes may make them identifiable though the quotes, it may be difficult to disguise their identifiable attributes without distorting the data (Robson, 2011), this was considered when quoting from the participants' interview transcripts.

## **Ethical approval**

Ethical approval was pursued and obtained from University of Huddersfield's School Research and Ethics Panel (SREP) in March 2014. The approval letter can be found in appendix 13.

In preparation for NHS ethics, the research proposal was shared at the Neurology User and Carer Forum from the local NHS trust, which is led and run by the service users, and the team colleagues in May 2014 for feedback. Positive feedback was received from the Neurology User and Carer Forum, the service users were in complete support of research idea and reassured and authenticated the study aim and the procedure, which were in accordance with the needs of the individuals with long term disabilities. The hand-out prepared for this forum is attached in appendix 14.

The study involved participants accessing National Health Services from two trusts. The NHS Research Ethics Committee (REC) was obtained in August 2015, a copy of the approval can be found in appendix 15. During the discussion stage with the local Research and development department, it was suggested that the proposal is sent to the proportionate review committee since the study was based on assessment of potential participants. The proportionate review committee is made up of a core team who evaluate the research from their location. Unfortunately, the proportionate review committee rejected the proposal since the study involved vulnerable

participants. This committee recommended that the proposal was scrutinised by the full committee and invited the researcher to attend this meeting to clarify any concerns raised in this meeting. This full committee comprised of fifteen members, involving clinicians, researchers and the general public. Following this meeting, two points of clarification were suggested with regards to the justification of using researcher's personal computer and one change to the participant information sheet was sought. The information regarding this outcome is attached in appendix 16. Subsequently the committee was satisfied with the explanations and provided approval in August 2015. This process further authenticated the research study proposal and the process.

Approval from site 1 and site 2, local two NHS Trusts' Research and Development Department to collect data, was obtained in September 2015. Copies of the approval letter from site 1 and site 2 can be found in appendix 17 and appendix 18 respectively. Recruitment and data collection was done between October 2015 and January 2017.

## **Data analysis**

### ***Phase one***

The Quantitative data was analysed using the software, IBM Statistical Package for Social Sciences (IBM SPSS) version 24 (May 2016). The quantitative data was entered initially in a Microsoft Excel file and then copied to the statistical software for analysis. Linear regression model (Gavin, 2008) was utilised to investigate the self-perceived performance and actual physical functional abilities over age. Multiple linear regression (Gavin, 2008) was utilised to analyse the influence of age on self-perceived and actual physical functional abilities outcome measures. A 3D Scatter plot graph was performed to analyse the influence of age on the Barthel Index in the presence of Self-reported Barthel Index. Direct relationship between each of the self-perceived and actual physical functional abilities outcome measures were analysed using linear regression model and the information is presented in a simple scatter plot in the results chapter.

The raw scores from the Short Form 36 Version 2 (SF36v2) scoring sheet were transferred on to the Quality Metric Health Outcome Scoring Software 4.5 (Saris-Baglana et al., 2011). This is the licensed software recommended to analyse the SF36v2 raw scores. License to use the SF36v2 and the Quality Metric Health Outcome Scoring Software 4.5 was obtained from the Office of Grants and Scholarly Research of Optuminsight Life Sciences, Inc. The license agreement No QM020635, for using the outcome measure and analysis software is attached in appendix 19. The data from the Physical Component Summary and Mental Component Summary was copied to the IBM SPSS software for analysis and plotting graphs.

### ***Phase two***

In view of the nature of the data and sample size in the second phase of research, thematic analysis was employed for the analysis. King and Harrocks (2010) and Robson (2011) have extensively written about typology of various approaches for an interview data, quasi-statistical approach, thematic analysis, template analysis, matrix analysis and grounded theory approach. Quasi-statistical approach relies on conversion of qualitative data into a quantitative data and is typified by content analysis and Grounded theory approach is utilised to develop a theory 'grounded' in the data, which is a specialist approach (Robson, 2011). Matrix approach utilises large, complex data set, especially where the research involves comparisons between sites, organisations or groups, here the data is represented visually in a tabular form against key concepts or issues relevant to the research question (King and Harrocks, 2010). In template analysis, a template is constructed, applied to the data and revised as necessary, until it captures a full picture of the analyst's understanding as possible, this is usually used for sample of between 10 and 25 and long interviews and works well where there are two or more distinct groups within the data set for comparison (King and Harrocks, 2010). The data set in this study involved three participants, the purpose was to explore the discrepancies between self-perceived and actual

physical functional abilities in adults with cerebral palsy and there were no sub groups for comparison, therefore thematic approach was found to be best suited to analyse the data set.

Additionally, Robson (2011) has indicated that thematic approach is a realist method, reporting the experiences, meanings and the reality of participants within the society. The second study objective in a sequential explanatory design was to explore the possible reasons for and means to reduce the discrepancies between self-perceived and actual physical functional abilities in adults with cerebral palsy, which generated subjective meaning and was influenced by the individual's experience of the world, which is similar to the purpose of thematic analysis as elaborated by Robson (2011) and Gavin (2008).

### *Transcribing and Coding*

Data from the digital recorder was transcribed verbatim by the researcher to avoid any distortion (Sutton and Austin, 2015). Nonverbal gestures recorded from field notes and pauses from the recorder were also incorporated in the transcription to retain authenticity and truthfulness (Kings and Harrocks, 2010, Robson, 2011). A copy of the transcription was provided to the relevant participant for 'member checking' (Rubin and Rubin, 2005) to check truthfulness and avoid misquoting. The transcripts were also checked against the respective recordings for truthfulness (Flick, 2009), which helped to familiarise with the data.

Robson (2011) has listed and explained the steps involved in thematic coding analysis, which are; familiarisation with the data, generating initial codes, identifying themes, constructing thematic networks and integrating/interpreting the themes. Thematic analysis in this study was based on these steps.

The process of initial coding is to organise the data into meaningful groups, to identify meaningful themes (Robson, 2011). The comments feature on the Microsoft Office Word was utilised to code the interesting aspects from the

raw data. Some context was included in coding for identifying relevant themes (Robson, 2011). Raw data was manually coded, indicating participants acts, behaviour, activities, strategies to manage, meanings (values), participation with the environment, constraints, consequences and settings. Two short extracts from the interview transcript which incorporate initial coding and theme is illustrated in figure 4 and figure 5. Part of a response was edited to maintain anonymity.

**Figure 4: Short interview extract 1**

<p>I am lucky that I am not wheelchair dependent, I am independent in most of my activities. But I do need help in <b>some things</b>.</p>	<p><b>Comment [RK3]:</b> Feels independent in most of the daily activities, but needs help with some things</p>
<p><b>2. What do you think about the differences between your actual performance and what you said you could do?</b></p>	
<p>As I have get older, spastic diplegic is a progressive condition, so as I have got older it starts to get worse, the cold weather tends to make it stiff. That's why, I need help sometimes in washing and dressing, especially for <b>transfers</b>.</p>	<p><b>Comment [RK4]:</b> Feels spastic diplegic gets worse with age, cold weather tends to make legs stiff. Needs help with washing, dressing and transfers</p>

**Figure 5: Short interview extract 2**

<p>I will say ex's, I walk around quite a lot, plenty of walking, that's I way I keep myself <b>fit</b>.</p>	<p><b>Comment [RK18]:</b> Aware of strategy to maintain fitness</p>
<p>I have seen a _____, she has said that I will notice difference in walking in 3 to 5 years' time and I have noticed some changes, I know I cannot do anything, only thing is for me to stay <b>positive</b>, I still have to do what I can, or I have to make myself do... I know rehabilitation clinics do splints and callipers, if waking gets difficult I know I can ask for them, as long its not visible to other person, I am happy to wear <b>them</b>.</p>	<p><b>Comment [RK19]:</b> Likes to stay positive; aware that walking will become more difficult with age Mental state Self - efficacy</p>
<p></p>	<p><b>Comment [RK20]:</b> Aware of strategies to support with walking – feels that maintaining his walking independence is very important for him)</p>

Themes were identified from these codes, using some of the strategies highlighted by Robson (2011), repetitions, indigenous categories, metaphors and analogies, transitions, similarities and differences, linguistic connectors and theory. Some of the primary themes that emerged from initial coding were coping strategy, identity, self-management, avoidance strategy, support, long term plan, anxiety, awareness, knowledge of health care providers, mental health and self-efficacy.

The next step involved constructing thematic networks and making comparisons (King and Harrocks, 2010), which was mapping one or more themes based on the focus of study, repetitions, similarities, differences and the context. In summary, four key themes emerged from the interview transcripts, namely coping strategy, identity, self-management and support, this is elaborated in the results chapter.

Triangulation was utilised to assess the quality of data analysis. A member from the supervisory team independently coded and identified themes. This was compared with the researchers' analysis to verify the process and triangulate the findings (Robson, 2011). It must be noted that no new themes emerged from this triangulation process.

## Chapter four: RESULTS

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The study findings from phase one and phase two are reported in this chapter.

### **Phase one analysis findings**

This section is presented in four sections. The first section is the demographic distribution of the phase one participants; the second section reports the findings from the four outcome measures (Short Form 36 Version 2, Self-reported Barthel Index, Barthel Index and Rivermead Mobility Index); third section reports the analysis of influence of age on the outcome measures and the final section reports the analysis of the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. Following the four sections, I have summarised the pertinent findings from Phase one of the study.

### **Demographic distribution**

Twenty three participants from two hospital sites were invited to participate in this research and seventeen consented to participate. Four participants did not want to take part in the study and two participants could not commit to participation in phase two of the study, hence they were excluded from this study. The data from the seventeen participants for phase one was collated between October 2015 and August 2016. All participants were selected based on the inclusion and exclusion criteria, as detailed in the methods section. This included twelve participants from site 1 and five participants from site 2. The standardised outcome measures were completed in the physiotherapy gymnasium for all participants in site 1 and in the clinic room for all participants in site 2. There were four participants in the age groups 16-20 years and 21-25 years, three participants in 26-30 years age group, five participants in 31-35 years age group and one participant in the age group 36-40 years, this is tabulated in table 4. The number of male/female was variable and this is tabulated in table 4. One participant in age group 21-25 and one participant in age group 26-30 was wheelchair dependent for functional

purpose. One participant in the age group 21-25 was fully dependent on wheelchair for all mobility purpose. Remaining participants walked independently with or without any walking aid.

**Table 4:** Demographic distribution

<b>Age group</b>	<b>Number of participants</b>	<b>Male</b>	<b>Female</b>
16-20	4	3	1
21-25	4	3	1
26-30	3	0	3
31-35	5	5	0
36-40	1	1	0

### **Short Form 36 Version 2 (SF36v2)**

All seventeen participants completed all the 36 items on SF36v2. 36 items were categorised into 8 sub-scores of health and well-being, namely general health, physical functioning, role physical, bodily pain, vitality, social functioning, role emotional and mental health. General health, physical functioning, role physical and bodily pain were summated as physical component summary for clinical inference. Vitality, social functioning, role emotional and mental health were summated as mental component summary for clinical inference.

The analysis of the final scores was completed using the Quality Metric Health Outcome Scoring Software 4.5. This analysis software was purchased with the scoring sheets. QualityMetric applies T-score, which is based on the US general population, has a mean of 50 and a standard deviation of 10 (Saris-Baglana et al., 2011). The main advantage of T-score based scoring is easier interpretation (Saris-Baglana et al., 2011). Although the T scores are based on the US general population, the validity of this outcome measure in British population (Brazier et al., 1992) has been elaborated in methods section. All the 36 items are summated into two component measures for



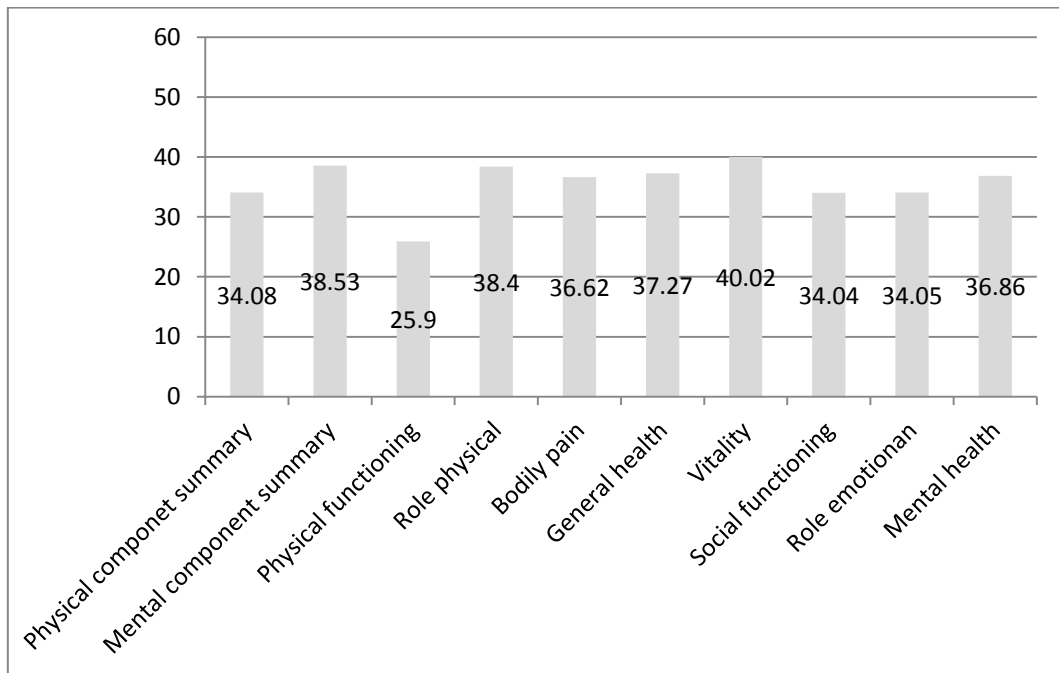
purpose of comparison, physical component summary score and mental component summary score, which is shown in table 5 below.

**Table 5:** Short-Form 36 Version 2 - summary scores of physical and mental components

<b>Participant</b>	<b>Physical component summary score</b>	<b>Mental component summary score</b>
1	43.08	42.69
2	32.51	62.27
3	34.68	27.93
4	35.29	25.62
5	23.43	31.57
6	35.21	42.42
7	26.94	21.09
8	36.05	24.38
9	48.68	45.23
10	27.19	36.72
11	46.37	49.98
12	33.78	36.35
13	23.86	24.96
14	32.73	47.79
15	42.41	22.71
16	38.92	59.13
17	45.34	54.17

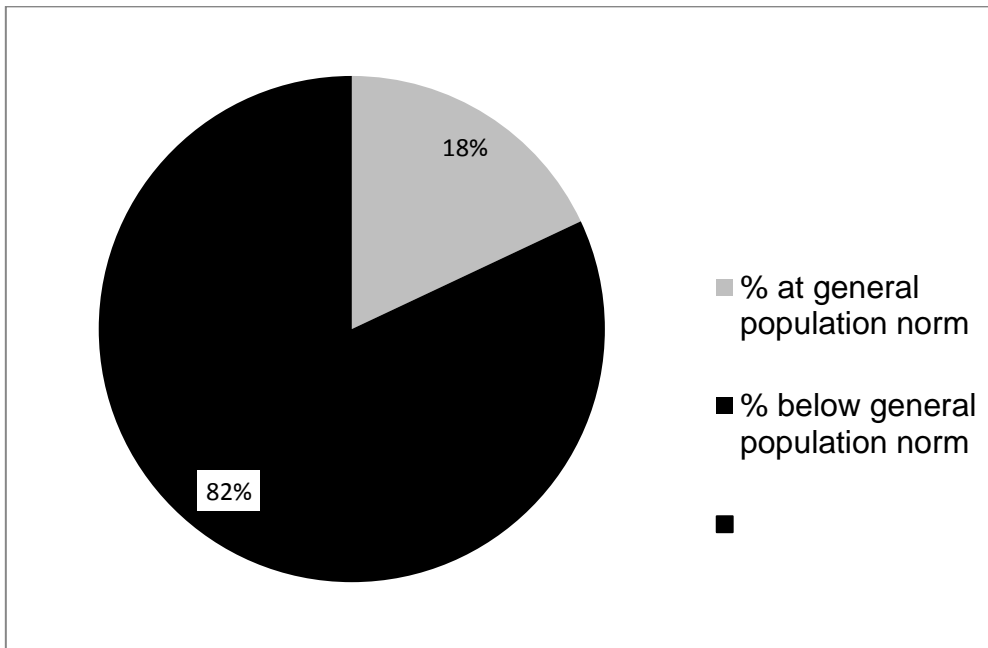
A bar graph of the Physical component summary and mental component summary and the domain that made up this component summary for all the participants can be found in figure 6. Higher scores indicate better functioning and well-being. Using the T-score based scoring, each scale and summary measures is scored to have a mean of 50 and a standing deviation of 10. The graph illustrates that on average individuals' performed worse on physical functioning when compared to vitality. Overall, the mental component summary averages were better than the physical component summary averages. This indicated that although the physical health and well-being were affected the individuals' with cerebral palsy coped better and had better mental health.

**Figure 6:** Short Form 36 Version 2 composite scores for all participants

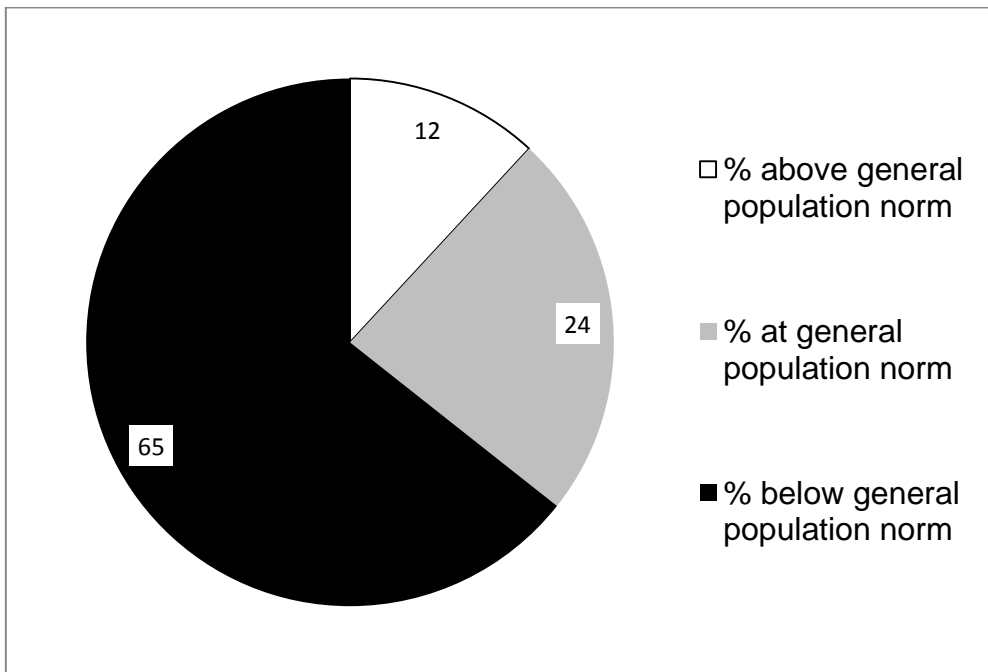


A pie chart graph was developed to identify the percentage of participants' health and well-being when compared to the General Population T –score. Figure 7, physical component summary graph, illustrates that 82% of the participants were below the general population and 18% of the participants were at par for physical health and well-being. There was no participant performance above the general population norm. However, figure 8, mental component summary graph, illustrates that the presentation was variable, 65% of the participants were below, 24% were at and 12% were above in the mental health and well-being when compared to the general population norm.

**Figure 7:** Physical component summary - percentage of participant's above/at/below the general population norm



**Figure 8:** Mental component summary - percentage of participant's above/at/below the general population norm



## Self-reported Barthel Index (SR-BI) and clinician assessed Barthel Index (BI)

The total scores from the self-perceived and actual Barthel Index is tabulated in table 6. SR-BI and BI consists of 10 times, comprising Bathing, stairs, dressing, mobility, transfers, feeding, toilet use, grooming, bladder and bowel management.

**Table 6:** Total scores from Self-reported Barthel Index(SR-BI) and Barthel Index(BI)

Participant	SR-BI (maximum score of 31)	BI (maximum score of 100)
1	28	85
2	12	49
3	24	88
4	9	10
5	19	85
6	21	90
7	13	60
8	24	100
9	22	82
10	12	38
11	30	95
12	14	59
13	24	44
14	21	65
15	29	90
16	18	55
17	30	100

Participant 8 and participant 17 scored maximum on the BI, however no participants scored maximum on the SR-BI.

Participant 8 scored differently on the self-rated scale for mobility and transfers when compared to clinician assessed BI due to variability in presentation. This participant reported that he is dependent on his carers for walking and transfer on the days when his legs are very stiff and painful, however on most of the days he is independent in his walking and transfers.

This also correlated with the mobility component on the Rivermead Mobility Index. Furthermore, analysis of this discrepancy can be found in the Phase two interview analysis of this participant.

Participant 17, scored maximum on the Barthel Index, however on the self-rated index he scored less due to dependency when using the stairs. For all other items the self-rating was similar to the BI assessed by clinician. The participant stated that he can use the stairs most of the time independently, however if the stairs are wide without the hand rail he would struggle to do on some days and therefore, underreported his abilities on the self-reported Barthel Index.

### **Rivermead Mobility Index**

The scores of the Rivermead Mobility Index (RMI) of the participants are tabulated in table 7. The scale consists of 15 items and each item is scored 1 if they are independent and 0 if dependent. The purpose of choosing this scale was to assess the mobility component and analyse the effect it has on the physical functional abilities.

**Table 7:** Total scores of Rivermead Mobility Index

<b>Participant</b>	<b>RMI score (maximum score is 15)</b>
1	11
2	2
3	12
4	0
5	20
6	12
7	8
8	13
9	14
10	10
11	12
12	7
13	13
14	7
15	13
16	3
17	14

Participant 4 did not score on any of the items. This participant is dependent on the carers for all functional abilities. As experienced on SR-BI, none of the participants scored maximum on the RMI. The scores of the participants ranged from none to 14, whilst the maximum any participant can score on RMI is 15.

### **Influence of age on physical functional abilities**

Although this was not the objective of the study, the outcome measures were analysed if there were any influence of age in view of the availability of data and to analyse if this had any effect on the study objective 1, which was to identify the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy.

Each of the outcome measures were analysed using linear regression model individually against the age. No significant correlation was found between age and the outcome measures. The F value and R<sup>2</sup> are tabulated in table 8.

**Table 8:** Statistical analysis of age vs each outcome measures

Age vs	SRBI	SF36v2PCS	SF36v2MCS	BI	RMI
F(1,15)	0.046	1.059	0.048	0.274	0.046
P value	0.833	0.320	0.830	0.609	0.834
R <sup>2</sup>	0.003	0.066	0.003	0.018	0.003

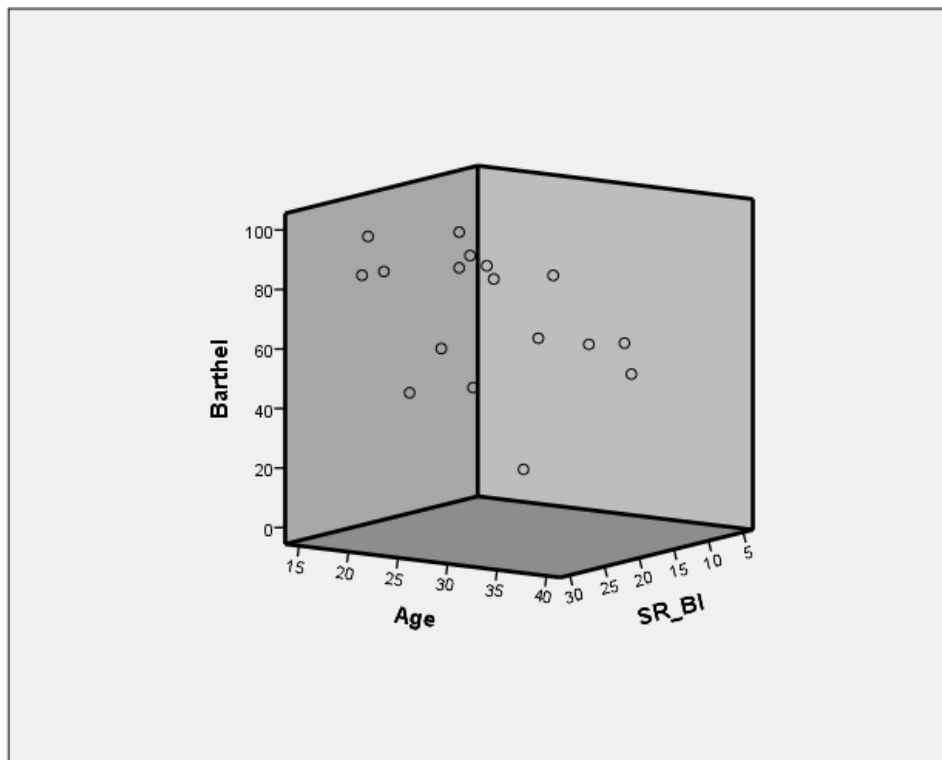
Similar to the findings of this study, Jahnsen et al. (2003) did not find any significant effect of age on physical function in adults with cerebral palsy when compared with general population. As indicated in the literature review, Cox et al., (2005) and Haak et al., (2009) indicated that there is no concrete evidence to date to indicate that aging affects physical functioning abilities in adults with cerebral palsy and suggested larger cohort studies to inform practice.

In this study, fewer number of participants and varied physical functional abilities of the participating individuals may have contributed to lack of any significant effect of age on the measured physical functioning abilities. Due to the limited scope of this study, baseline physical functioning of the individuals were not categorised for analysis which may have contributed to the varied physical functional abilities affecting the statistical analysis.

To also analyse the effect of age on the physical functional abilities, multiple linear regression analysis was performed. Effect of age on the BI in the presence of SRBI was analysed using linear regression method. It was evident that age was not linked to Barthel Index in the presence of SRBI,  $p > 0.2$ .

To graphically represent the effect of age on BI in the present of SR-BI, the values were plotted on the 3D scatter plot graph using IBM Statistical Package for Social Sciences (IBM SPSS) software. This graph is presented in figure 9. It is clear from the scatter plot graph that the values tend to congregate near the age, this indicates that age does not have influence on BI in the presence of SR-BI.

**Figure 9:** 3D scatter plot graph - influence of age on Barthel Index (Barthel) in the presence of Self-reported Barthel Index (SR\_BI)

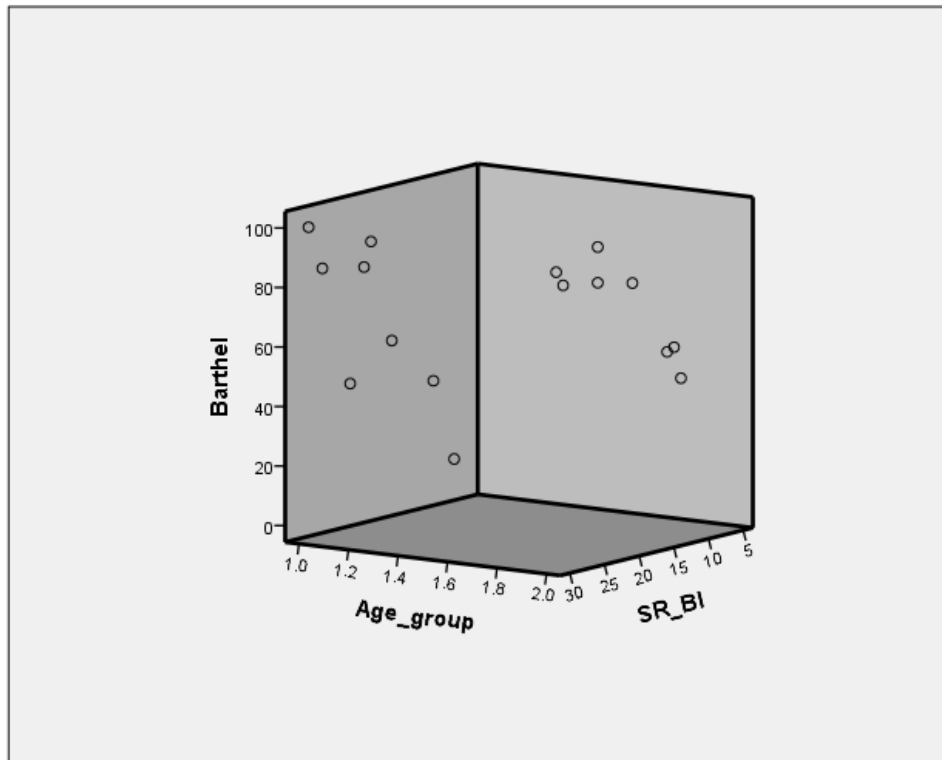


Due to the fewer number of participants it may be that there was no influence of age on BI in the presence of SR-BI. Therefore, to explore these effects further, the participants were grouped as 16 to 25 years in one group and 26 to 35 years in the second group. Data of one participant, over the age of 35 was eliminated for the purpose of this analysis. Multiple linear regression analysis revealed that age group was linked to Barthel Index in the presence of SRBI with a moderate significance,  $p = 0.062$  ( $t = 2.042$ ).

To graphically represent the effect of age grouping on BI in the present of SR-BI, the values were plotted on the 3D scatter plot graph using IBM SPSS software. The graph is present in figure 10. It is clear from the scatter plot that the values tend to congregate away from the age axis and towards BI or SR-BI, this indicates that age group had moderate influence on BI in the presence of SR-BI.



**Figure 10:** 3D scatter plot - influence of age grouping (Age\_group) on Barthel Index (Barthel) in the presence of Self-reported Barthel Index (SR\_BI)



This demonstrates that age does have some influence on the physical functional abilities. If larger cohort of participants were involved in this research, we may have found significant influence of age on physical functional abilities.

Jahnsen et al., (2004a) study identified deterioration in physical function from 30 years of age. Andersson and Mattsson (2001) reported that significant number of participants reported that deterioration happened before the age of 35 years. Therefore, if the research had involved larger cohorts of individuals with cerebral palsy between the ages of 20 and 40 years, there may have been a significant influence of age on physical functioning. This is elaborated furthermore in the forthcoming discussion chapters.

## **Relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy**

Due to the sample size, the two outcome measures of self-perceived (SR-BI and SF36v2) and actual (BI and RMI) physical functional abilities were not compounded for comparison as initially proposed. Each measure of the self-perceived scales was compared with the actual physical functional abilities measures.

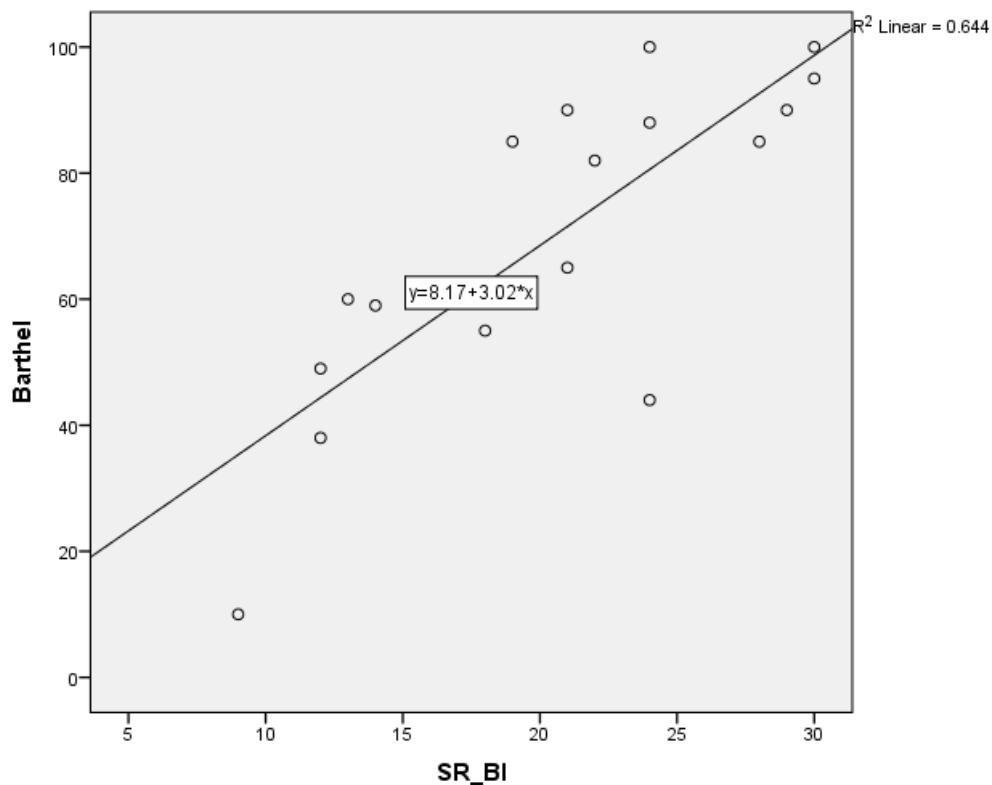
### Relationship between Self-reported Barthel Index and actual physical functional abilities outcome measures

The linear regression analysis between self-perceived Barthel Index (SR-BI) and each of the actual physical functional abilities outcome measures (BI and RMI) indicated strong significant correlation.

#### *SR-BI Vs BI*

A univariate linear regression was performed to analyse the relationship between SR-BI and BI. A significant regression equation was found ( $F(1, 15) = 27.110, p < 0.000$ ), with an  $R^2$  of 0.644. Graphical representation of this relationship is illustrated in figure 11.

**Figure 11:** Relationship between Self-reported Barthel Index (SR\_BI) and Barthel Index (Barthel)



The results are in concordance with the findings of Morley et al. (2012) and O'Connor et al. (2005) conclusions that there is high correlation between the SR-BI and BI.

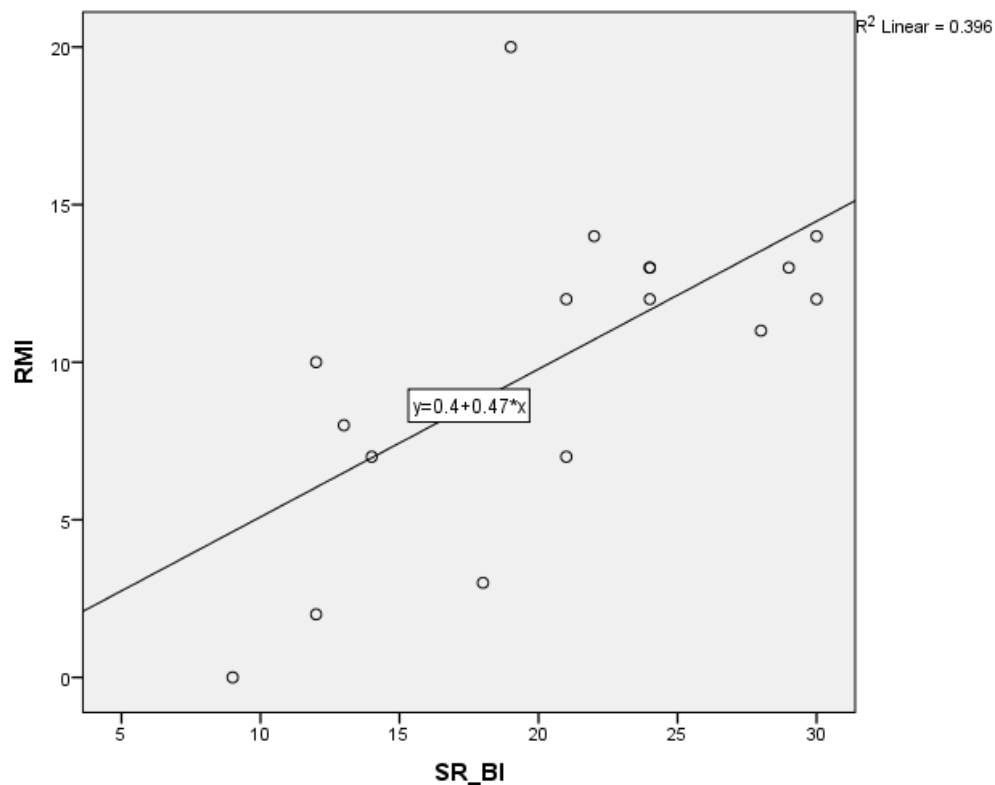
Although there was significant correlation, there were discrepancies between the scores on SR-BI and BI (self-perceived and actual physical functional abilities) which was explored further in the interview. For example, participant 7 reported that he does have difficulty in dressing and is dependent on carers, when this was clinically assessed he was independent in his dressing. When requested to elaborate, participant 7 reported that his dressing is weather dependent and he has difficulty on some days in the mornings. Participant 3 stated that he is dependent on his carers for using the stairs, however when he was clinically assessed he was independent in using clinic stairs. On further questioning this discrepancy, he reported that fatigue affects his use of stairs, when he is out and about, he has to walk for few metres and then use

the stairs, however, in the clinic, the participant used the stairs after minimal walking on the level surface. He used the stairs after being seated for approximately 30 minutes. These discrepancies were explored in detail in the interview. Due to the smaller sample size, statistical analysis to identify similarities and discrepancies between SR-BI and BI would have limited inference, therefore, this analysis was not attempted.

#### *SR-BI Vs RMI*

A univariate linear regression was performed to analyse the relationship between SR-BI and RMI. A significant regression equation was found ( $F_{1,15} = 9.838$ ,  $P < 0.007$ ), with an  $R^2$  of 0.396. Graphical representation of this relationship is illustrated in a scatter plot figure 12. Due to the smaller sample size, statistical analysis to identify similarities and discrepancies between SR-BI and RMI would have limited inference, therefore this analysis was not attempted.

**Figure 12:** Relationship between Self-reported Barthel Index (SR\_BI) and Rivermead Mobility Index (RMI)



Relationship between Short Form 36 Version 2 (SF36v2) and actual physical functional abilities (BI and RMI)

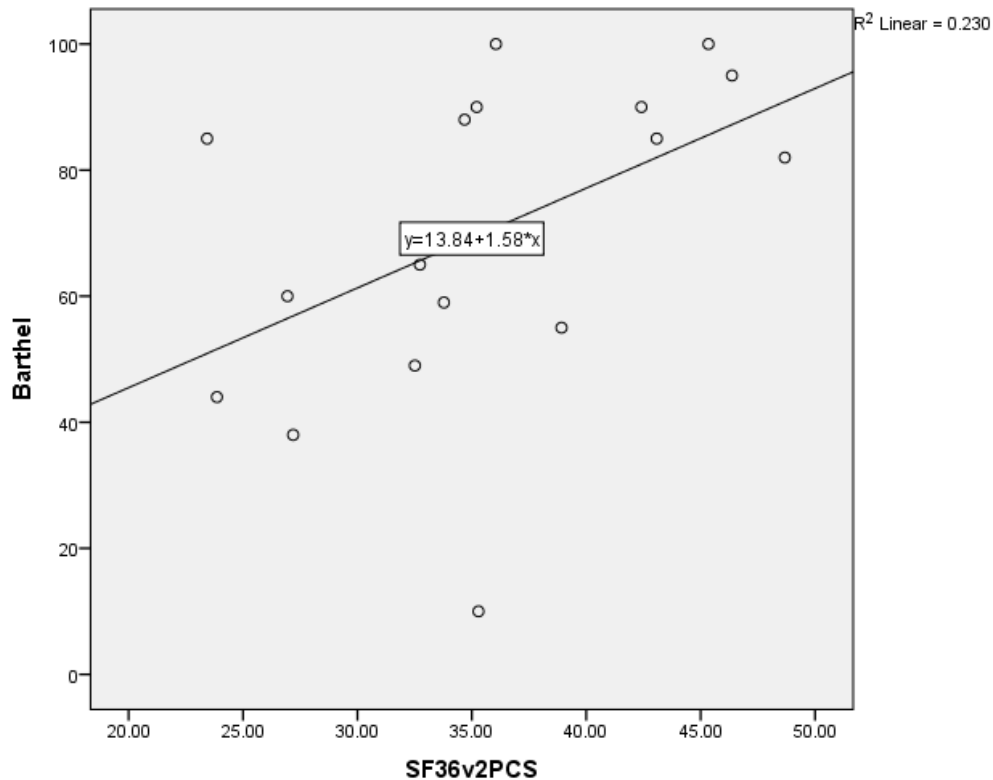
The linear regression analysis between SF36v2 and BI, revealed moderate significance, especially the physical component summary. There was no significant correlation between SF36v2 mental component summary and actual physical functional abilities (BI and RMI).

*SF36v2 Vs BI*

A simple linear regression was performed to analyse the relationship between SF36v2 (Physical and mental component summary) and the BI. A moderate significant equation was found ( $F(1,15)=4.469$ ,  $p \leq 0.05$ ), with an  $R^2$  of 0.230 between SF36v2 physical component summary and BI. Graphical representation of this relationship is illustrated in a scatter plot figure 13. However, there was no significant correlation between the SF36v2 mental

component summary and the BI, the linear regression analysis equation was found to be  $F(1,15)=0.115$ ,  $p \leq 0.739$ , with an  $R^2$  of  $-0.059$ .

**Figure 13:** Relationship between Short Form 36 Version 2 physical component (SF36v2PCS) summary and Barthel Index (Barthel)



#### *SF36v2 Vs RMI*

A simple linear regression was performed to analyse the relationship between SF36v2 (Physical and mental component summary) and the RMI.

There was no significant correlation between the SF36v2 physical component summary and the RMI, the linear regression analysis equation was found to be  $F(1,15)=0.013$ ,  $p \leq 0.910$ , with an  $R^2$  of  $-0.066$ .

There was no significant correlation between the SF36v2 mental component summary and the RMI, the linear regression analysis equation was found to be  $F(1,15)=1.182$ ,  $p \leq 0.294$ , with an  $R^2$  of  $0.011$ .

### ***Summary of the findings from phase one:***

- Statistically, there was no significant relationship between age and the outcome measures, which measures physical functional abilities
- Linear regression analysis revealed that age group (16 to 25 vs 26 to 35) had influence on the Barthel Index in the presence of Self-reported Barthel Index with a moderate significance
- Self-reported Barthel Index scores of the participants significantly correlated with both the actual physical functional abilities outcome measures (Barthel Index and Rivermead Mobility Index). This indicates that the scoring of the tasks, both on the self-perceived and actual physical functional abilities is valid
- There were discrepancies between the scores on Self-reported Barthel Index and Barthel Index (self-perceived and actual physical functional abilities) which were explored further in the phase 2 interview
- Self-reported Short Form 36 Version 2 scores, especially the physical component summary moderately correlated with the Barthel Index
- There was no correlation between Short Form 36 Version 2 and Rivermead Mobility Index

The results from phase one are discussed further in the discussion chapter.

### **Phase two analysis findings**

Interview transcripts were analysed using the principles of thematic analysis (Gavin 2008, Flick, 2009). The interview participants are identified here as P1, P2 and P3.

This section of the results, reports and describes the four discrete themes that emerged from the thematic analysis of the interview, namely coping strategy, self-management, support and identity. Due to the relevance of the research, I have also reported the concerns of the interview participants in relation to the changes to physical functional abilities and aging in the final section of this chapter.

## **Coping strategy**

Coping is defined as efforts to prevent or diminish threat, harm and loss, or to reduce associated distress (Carver and Connor-Smith, 2010). As elaborated by Carver and Connor-Smith (2010), coping can be either a voluntary response, or involuntary and automatic response within the coping construct; however, responses that begin as intentional and effortful may become automatic with replication. Due to the interview technique used in this research, only the response identified by the participant is considered for analysis, thus removing unconscious defensive reactions from the realm of consideration (Carver and Connor-Smith, 2010). Broadly, Carver and Connor-Smith (2010) have explained that the coping strategies can be elaborated in several methods: problem versus emotion focus; engagement versus disengagement; accommodative coping and meaning focused coping and proactive coping. The authors have argued that a given response (interview response) typically fits several places and no one response fully represents the structure of coping. Confirmatory analysis like an interview clearly supports multidimensional modes of coping (Skinner et al. 2003). Carver and Connor-Smith (2010) have argued that the distinction that appears to have greatest importance is engagement versus disengagement, a distinction that also maps well in the context of personality. Disengagement in this context is not that the person is not able to cope, it is a recognised coping strategy to accommodate oneself with the challenges. Therefore, the interview response will be analysed here using the engagement / disengagement coping model.

The engagement and disengagement coping model was elaborated extensively by Carver and Connor-Smith (2010). In this research, engagement and disengagement coping strategy model is utilised to explain the interview response by the participants. This would help us to analyse the discrepancies between self-perceived and actual physical functional abilities. Engagement coping includes support seeking emotion regulation,



acceptance, social participation and cognitive restructuring. Disengagement coping includes avoidance, denial and wishful thinking. As explained in the earlier paragraph disengagement is not a negative strategy but is part of the positive coping model, for instance, an individual can avoid visiting places with stairs on the days when leg stiffness is significant.

### Engagement coping strategy

P1 and P3 responded to the question concerning their personal care by explaining that they overcame the weakness in the hand dexterity resulting in buttoning difficulties by using T shirts:

*P1: ...for dressing, I always need help to do buttons and socks due to issues with hand dexterity, I tend to wear T shirts to get around this problem. I also wear trainers without socks or use velcro's.*

*P2: ...I try to avoid shirts with buttons and use T shirts all time.*

All three interview participants reported increased difficulties due to changes in spasticity with aging and they manage this in various methods.

For example

*P1: At present, I use a combination of daily exercises, receiving help ...from my parents when at home or from support workers when at university. This helps me to maintain function in my legs.*

*P3: ...it's managed with medication and heating....I don't want to take a lot of medications.*

Spasticity and leg weakness impacted on various physical functioning of the individuals. One of the participants manages this by using the grab rails when getting in and out of bath and uses lifts to compensate for difficulty in using the stairs indoors and outdoors.

*P2: I still need help getting in and out of the bath, I managed most of the time using the grab rails, but I have a carer to help if needed.*

P2: *...I choose to stay away from stairs and take alternative ways of getting up and down levels in buildings if it is available, such as lifts....*

One of the salient responses to the coping strategy was that one participant stated that he has not really stopped doing much, he has adapted to the changes.

P2: *...exercises to maintain my strength and stamina and use my car for accessing social opportunities.*

This demonstrates that the coping strategy is not only related to overcoming a physical challenge but this also creates opportunities for the participants to socialise.

When young, some of the coping strategies can be influenced by the carers and this result in acquiring a long standing adjustment mechanism in adulthood.

This was reiterated by P3: *'I was told that I have a condition but I still have to do what other people do'.*

However, he was aware that if he has difficulty in walking he can ask for help.

P3 stated, *'If walking gets difficult I can ask for help'.*

#### Disengagement coping strategy

Interview participants have also responded to difficulties by avoiding certain physical functional activities. For instance, one participant with lower limb weakness, limited his outdoor walking to prevent falls.

P2 reported, *'I have some falls when walking outdoors, to avoid falls I limit my walking'. Due to balance difficulties, I sometimes avoid stairs.*

Some adjustments to difficulties have also resulted in long standing affects.

P2 stated, '*...struggles to catch objects that are thrown at me and therefore I avoid playing sports*'.

Disengagement or failing to find alternate coping strategy can also affect nutrition of the individual.

For instance, P2, stated: '*...I find it difficult to cook and therefore eat out often.*'

Increased difficulty with age has affected participation in a leisure activity in P3.

P3: *I used to enjoy swimming, but I have stopped this. The cold water makes my legs go stiff now and the whole body stiffens and I am not able to move for some time. I don't do swimming now.*

#### Engagement vs disengagement as adjustment mechanisms

It is clear from the findings that participants exhibited both engagement and disengagement coping strategies in physical functional abilities. This was also evident during their social and leisure pursuits. Coping strategies is influenced by several factors, such as personality traits, frequency of exposure to stressors, the type of stressors experience with the environment and the feedback (Carver and Connor-Smith, 2010 and Vollrath, 2001), this was not explored in this research study.

Furthermore, condition related predictors of adaptive behaviours in congenital neurodevelopmental disorders include neurological factors, fine and gross motor functional abilities and cognition. In children and adolescents with cerebral palsy, developmental trajectories of activities of daily living are moderated by gross motor functional levels, but level of intellect is a more significant intervening variable (Tan et al., 2014) for identifying the appropriate coping strategy. Development of a coping strategy could be related to the environmental influence, personality traits and cognition (Jacobson et al.,

2013). More in-depth interview of the adjustments may have revealed further information regarding cognition and coping strategies.

### **Self-Management**

The importance of self-management is set in the forefront of the National Institute for Health and Care Excellence (NICE) Guidelines and Government Policy Framework. The NHS Improvement Plan: Putting People at the Heart of Public Service (Department of Health, 2004b) sets a new strategic model for management of long-term conditions through self-care, disease management and case management, this was a similar theme that emerged from the participants' interview. The National Service Framework further emphasises the strategic plan to improve lives of people with Long Term Conditions (Department of Health, 2005) by giving people choice, through services, planned and delivered around their individual needs, supporting people to live independently, play their full part in society and co-ordinating partnership working between health services, social services and other local agencies.

Tarazi, Mahone and Zabel (2007) has commented that a youth with disabilities frequently will have atypical health care needs that require development of more complex self-management skills. There is also limited evidence to correlate self-management with general levels of adaptive behaviour (Jacobson and Melnyk, 2012). Palisano (2012) has stated that when goals include life-long fitness, the involvement of adolescents in choice of healthy and enjoyable activities improves the likelihood of sustenance over time. Therefore involvement in a fitness pattern starts early and when sustained for longer periods, the impact on the functional activities is far greater.

This was reiterated in the interview:

P1 stated that *'repetitive exercises over the years have improved performance in certain tasks'*.

P1 reported, *'I walk regularly to maintain strength and fitness, also working with my personal trainer and going to the gym to help combat muscular weakness and deterioration'*.

Unfortunately, little is known about the effects of physical activity practices on self-management strategies of adolescents with cerebral palsy as they grow older as suggested by Koldoff and Holtzclaw (2015). Furthermore, it is a disturbing observation within my initial literature review that no studies found that tested interventions, to maintain or improve physical activity as adolescents leave the structured school environment and enter adulthood (Koldoff and Holtzclaw, 2015). Repeating this literature review in November 2017 also found no additional research that explored the long term impact (in adulthood) of the interventions pertaining to physical functional abilities, provided during childhood in individuals with cerebral palsy.

Nevertheless, there has been some evidence pertaining to self-management in certain periods of their life. Geerdink et al. (2015) has demonstrated that combining self-management with intensive upper limb intervention is a feasible and a promising intervention for improving the capacity of the upper limb and its use in bimanual tasks in older children and adolescent with unilateral cerebral palsy. Further environment changes (accessibility, attitude, adaptations) can be an effective way to improve self-help skills and mobility (Colver, Fairhurst and Pharoah, 2014).

Koldoff and Holtzclaw (2015) performed an integrated review of the literature on physical activity on adolescents with cerebral palsy and suggested that adolescents with cerebral palsy appear to enjoy a variety of new experiences as much as typically developing teens and wants experience that differ from a therapy session. Participation of enjoyable activities improves the likelihood of maintaining healthy lifestyle lifelong (Pilasano, 2012).

This was illustrated by one participants' comment on the importance of efficacy to improve participation in self-management and quality of life.

P3 stated, *'I am aware that I will deteriorate gradually and cannot do anything about this, only thing for me is to stay positive.'*

Adults with cerebral palsy assert that their participation in life does not depend on being able to walk, but on communication and being able to manage and control their environment (Colver et al., 2014). The strategy to self-manage will give them the skills to manage and control their environment effectively.

## **Support**

Access to rehabilitation and even to primary medical care for individuals with cerebral palsy worldwide can become difficult after the age of 18 years, possibly because cerebral palsy continues to be seen by the clinical community as a paediatric diagnosis; the life expectancy in previous generations was much lower than it is currently (Mutch et al., 1992). Children and adults need comprehensive, coordinated care focused on preventing complications (malnutrition, skin breakdown and pain) and promoting improved neurological function, which could ultimately improve health, quality of life and access to education and vocational opportunities (Aisen et al., 2011). The maintenance of health in the context of primary and secondary neurological complications must be a fundamental goal (Aisen et al., 2011) in this population. Roebroek et al. (2009) concluded from their study that young people with disabilities experienced health related problems such as deterioration in walking, development of secondary impairments, chronic pain or fatigue and a low level of physical fitness and inactive lifestyle. Furthermore, Roebroek et al (2009) recommended that there is an adequate follow-up into adulthood due to the risks of functional deterioration and development of secondary complications.

The recommendation from Aisen et al. (2011) and Roebroek et al. (2009) was strengthened by the data collected in the interviews, from all three interview participants.

For instance

*P1 stated that, 'If I can manage my pain and stiffness better or with some support I am sure my abilities (transfer abilities) will be less variable on day to day basis. I think I need to see the Rehab Consultant to review my spasticity medications'.*

P1 valued his regular contacts with the medical team and would like this to be replicated in the adult services.

*P1: ..long term...I would like to maintain contact with a physio and OT. I recognise it is important to be independent, having a professional look at my condition, perhaps once or twice yearly as a baseline, like the annual review I used to get from the Child Development Centre.*

*P1 also stated, '...knowing whom to contact in the event of sudden change in my condition, would also give me sense of security and confidence going forward.*

P2 also felt that he valued the support he received from the children's services:

*'Growing up, there was a lot more support as a child from other people and health services. It was easier to access. As an adult, one is responsible for earning an income, staying employed, staying healthy and independent. You would think the support to maintain all this would be easy to access and find but it's not. There is no support insight at first, unless you have multiple needs from various specialists.'*

P3 is currently seeing a Rehabilitation Consultant regularly and echoed his views, which were similar to the other interviewees:

*P3, 'I have got a good consultant whom I see every 6 months. If I have a problem, I discuss with him'.*

P3 was more aware about his condition and management options.

For instance:

P3 stated that, *'I am aware that I can have injections to reduce stiffness.*

*'...I know rehabilitation clinics do splints and callipers, if walking gets difficult I know I can ask for them...'*

P3 also commented that primary care medical team are not well informed about the condition and direct his referrals to the speciality consultant.

P3: *...they don't understand the condition, if I say I have spasms overnight, they tell me to contact the consultant. They don't know much about spastic diplegia.'*

Aisen et al. (2011) has stated that ideally, care should be provided by a consistent team that can assess and treat the primary and secondary effects of cerebral palsy, as well as common comorbidities and provide primary and specialist medical care, rehabilitation, psychosocial treatment and support for the patient and family. It is evident from the participants' interview, that they value the support provided by a consistent team with specialist knowledge in the condition. This also helps to build rapport between the individual and the care team and encourages openness between the stakeholders. Therefore, lifelong access to specialist adult services is paramount in this population.

## **Identity**

Watson (2002) has elaborated in his research regarding two historical and strategic approaches to the production of identities within the social sciences proposed earlier by Hall (1996). The first model is based on the assumption that there is an essential, natural or intrinsic meaning to any identity and this is based on a shared social experience, origin or structure. The second model denies the existence of an identity based on a shared origin or experience. Many participants in Watson's (2002) study, rejected their body image as an essential, biological, determinant of the self; just because they had an



impairment, they did not perceive themselves as different to other, non-disabled person. Some of the participants in this research (Watson, 2002), reported that self-identity is not about being different, but it is about what they can do as an individual.

In this research, P3 also rejected the notion of bodily difference and wanted him to be remembered as a normal person.

P3 stated, *'I want people to remember me as being independent as I am now....It's important to me to be seen as a normal person'*.

P3 was also clear that he does not want his disability to be made obvious to the society. When discussing about seeking support, he was aware that his legs are stiffer and he can request support like orthotics if needed.

P3: *..if walking gets difficult I know I can ask for them, as long it's not visible to other person, I am happy to wear them...*

Read, Morton and Ryan (2015) have concluded from their online survey on negotiating identity in person with cerebral palsy, that the individuals' face a number of threats in how they construe their identity, both in navigating stigma and maintaining access to needed support. The authors (Read et al., 2015) have recommended that healthcare providers should recognise the unique needs of the adults with cerebral palsy and therefore services should be tailored to the individual.

Most adults with disabilities have to negotiate their identity as a disabled person, a citizen and somebody with a level of social participation (Kent, 2013) and it is important for the individuals to develop a self-identity and self-efficacy.

### **Physical functional abilities and aging**

As indicated in the literature review, there is variable evidence indicating deterioration of physical functional abilities in adulthood (Cox et al., 2005,

Haak, 2009 and Morgan and McGinley, 2014). The findings from the interview also indicated that there is deterioration in functioning due to aging in this population.

Two participants stated that they have increased difficulty in carrying items, picking objects off the floors, doing house chores, mobility and stiffness in legs.

*P2 stated, 'During my time throughout adulthood, I have experienced more frequent problems with picking objects up from the floor'. When I try to and carry out this action, my lower body muscles feel tighter than they done previously. It takes extra self-control and more concentration to do this type of activity. Over time I have noticed that I now have reduced balance when standing and walking in particular....Ten years ago I was able to go up and down stairs. Now due to my poor balance I choose to stay away...I have always struggled to catch objects that are thrown at me. But this seems to have become more a problem, the older than I have got.*

*And P3, who states: As I have got older it starts to get worse, the cold weather tends to make it stiff. ....the mobility side is probably bit slower...I am not able to walk further without taking a few minutes stop, my walking distance is also less.*

P2 further commented that the increased functional challenges are largely due to aches and pain in his muscles.

*P2: I have had more aches and pain in my muscles recently, its gradually getting worse.*

P2 strongly stated several times during the interview, that he is currently having more problems in adulthood and this is gradually getting worse. Turk et al. (2001), Jahnsen et al. (2003, 2004b) Zaffuto-Sforza (2005) and Opheim et al. (2009) have all reported that largely adults with cerebral palsy reported increased fatigue with aging in their researches. To strengthen this argument,

one participant during the interview commented that fatigue had worsened with age.

*P2: As an adult approaching my thirties, I am more fatigued than I ever was, and everyday tasks are becoming more challenging, so the reliance on public services to maintain good quality of life is inevitable.*

One participant was very concerned about the deterioration in physical health.

*P3: ...I am worried that this will gradually go downhill and I will become dependent*

This interview clearly highlighted that the interview participants were well aware about the deterioration in physical functional abilities in adult life and the impact of this on their everyday tasks. Apart from their physical health, it is also clear, that there is an impact on their quality of life. This is further elaborated in the forthcoming chapters.

## Chapter five: DISCUSSION

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### Introduction

This chapter discusses the findings from phase one and phase two in relation to the study objectives. I have integrated the findings from both the phases to discuss the study objectives. In depth discussion were also incorporated in the results section where each findings were presented and explored.

### **Study objective 1: Relationship between self-perceived and actual abilities in physical functional abilities in adults with cerebral palsy**

The results from phase one demonstrates that:

- Self-reported Barthel Index scores of the participants significantly correlated with both the actual physical functional abilities outcome measures (Barthel Index and Rivermead Mobility Index). Self-reported Short Form 36 Version 2 scores, especially the physical component summary demonstrated moderate correlation with the Barthel Index.
- There were discrepancies between the self-perceived and actual physical functional abilities (measured through Self-reported Barthel Index and Barthel Index), which was explored further in the interview.

It is clearly evident from the phase one findings that there were discrepancies between self-perceived and actual physical functional abilities, however there were moderate to significant correlation between the self-perceived and actual physical functional abilities in adults with cerebral palsy. As discussed earlier in the literature review, only Morgan et al., (2014) explored the relationship between gait and health related quality of life in ambulant adults with cerebral palsy and concluded that mobility decline was significantly associated with mental health status but not when other predictor variables were included in the analysis (falls, physical component summary and life satisfaction), in this study, similarities or differences in this relationship was not explored. However, as indicated in the literature review, no research exploring the

relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy was found.

This research finding indicated that there are actual changes in physical functional abilities (as assessed by the clinician) during adulthood and the individuals' also recognise these changes, which is evident from the self-perceived scores. There are clear discrepancies between the self-perceived abilities and actual abilities, individuals' perceive increased difficulties while performing daily tasks when compared to the actual abilities. This was evident in physical functional abilities, especially in dressing, mobility and using the stairs.

Individuals were dependent on carers and struggled to be independent in dressing. However, during the clinical assessment they were independent in dressing. Some individuals avoided using the stairs on certain days when it was cold or legs were stiff, this also affected the mobility. However, during the clinic assessment some of the individuals who reported difficulty in using the stairs were independent in using the stairs. For a more detailed evaluation, the individuals were advised to try walking without using the hand rails and they were able to use the stair without any support.

The quantitative assessment did not give an opportunity to explore these discrepancies. The interview gave an opportunity to explore the reasons and means to reduce these discrepancies which is discussed in the next section.

Unfortunately, statistical analysis of these discrepancies between self-perceived and actual physical functional abilities was not attempted due to the limitations of the study and certainly justify further exploration in future studies. Nevertheless, these findings from the research are significant. As elaborated in the literature review, it is essential that the self-perceived abilities mirror the actual abilities in adults with cerebral palsy to reduce deterioration of functional abilities. Sandstorm et al. (2004) has acknowledged that there can be discrepancies between self-perceived and actual abilities with a risk for

underestimating when using self-reported data and indicated that there are no research exploring the relationship.

It is evident from the scores that self-perception of physical functional abilities is lower than the scores of actual physical functional abilities, as suggested by Sandstrom et al. (2004), that individuals tend to underestimate their abilities when compared to actual abilities. This may indicate that the deterioration is primarily noted in the self-perception when compared to the actual abilities; if this is addressed earlier by the health professionals, we may be able to slow down the deterioration of physical functional abilities in adults with cerebral palsy. Therefore, it can be argued that health professionals need to be proactive in identification of deterioration in physical functional abilities, health professionals need to give an opportunity to discuss the self-perceived physical functional abilities in such individuals. Drawing upon the findings from the interview, it is clear that the individual's appreciate and value the regular reviews with the health professional. This gives an opportunity to discuss their physical functioning with the health professional, including self-perceived abilities. It is recommended that adults with cerebral palsy are provided with regular reviews with the relevant health professional for appropriate management of the changes in the physical functioning and that these reviews incorporate assessment of self-perceived and actual abilities to provide appropriate management strategies. Some of the management strategies as perceived by the individuals are elaborated in the next discussion section.

Furthermore, Palisano (2012) indicated that identification of healthy and enjoyable activities during adolescence will support sustained long term goals during adulthood. Although, there are significant evidence pertaining to interventions in children with cerebral palsy, research in effect of these interventions during adulthood is lacking, this is also supported by the findings of Koldoff and Holtzclaw (2015).

Although this research indicates that there is discrepancy between the self-perceived and actual physical functional abilities in adults with cerebral palsy, one can raise caution whilst drawing up conclusions from a small sample size. Due to the limited scope of the research programme and other reasons elaborated further in the limitations, it was challenging to recruit participants. Nevertheless, the findings from the research cannot be ignored and can be used as a pilot study to commission a longitudinal research involving a larger cohort.

### ***Changes in physical functional abilities changes in adults with cerebral palsy***

Due to the limited scope and timescale, this research has not attempted initially to look at the changes in physical functional abilities in adults with cerebral palsy, however there is a clear indication of some influence of age on the Self-reported Barthel Index and clinician assessed Barthel Index, which cannot be ignored. Linear regression analysis revealed that age group (16 to 25 years vs 26 to 35 years) was linked to Barthel Index in the presence of Self-reported Barthel Index with a moderate significance. This demonstrates that we might have been able to demonstrate a statistical significance if a larger cohort of individuals was recruited for the study, with distribution of age group from 16 years to 40 years. This finding is similar to the review reports of Cox et al. (2005) and Haak et al. (2009). It can be argued that this evidence is weak, however this research findings can be utilised to plan a larger cohort study to improve the evidence to inform practice.

As discussed earlier in the literature review, lack of concrete evidence pertaining to adulthood in cerebral palsy could be due to the different outcome measures utilised to assess these individuals. The outcome measures used by the authors (Murphy et al., 1995, Ando and Uedo, 2000, Andersson and Mattsson, 2001, Bottos et al., 2001, Turk et al., 2001, Jahnsen et al., 2004a, Sandstrom et al., 2004, Strauss et al., 2004, Hanna et al., 2009, Opheim et al., 2009, Morgan et al., 2014 and Benner et al., 2017) were Short Form-36,

Pain Questionnaire, Fatigue Questionnaire, multidimensional questionnaire, case notes, interviews and surveys. As elaborated extensively in the literature review, these outcome measures can be broadly grouped into self-perceived ratings scales and clinician assessed rating scales. As noted in the literature review, lack of concrete evidence for deterioration in physical functional abilities in adults with cerebral palsy could be due to the non-recognition of type of outcome measures and variety of measures utilised. It is also challenging to do a systematic review of the research studies to inform evidence based practice. I have tried to address this challenge within this research study by identifying the factors affecting physical functional abilities and choosing the outcome measures that will measure these factors. From the literature review, the factors affecting physical functional abilities in adults with cerebral palsy are tabulated in Table 3. A longitudinal research involving a larger group, incorporating tools to measure self-perceived and actual physical functional abilities will strengthen the evidence. Some of the disease based organisations have addressed this challenge.

A number of disease based organisations and pharmaceutical companies involved in research have established a national register via regional centres to record the individuals' functional abilities. For example, Duchene Muscular Dystrophy has established a national register and utilises Assessment of Motor and Process Skills, Upper Limb Functional Item and Functional Rating Scales to record and track progression of the disease (Treat-NMD Neuromuscular network, 2017). Parkinson's UK group recommends Lindop Parkinson's Assessment Scale to record and track the progress of the disease nationally (Parkinson's UK, no date and Pearson et al., 2009). It would be ideal to have a similar type of national register to record the progression of physical functional abilities during adulthood in cerebral palsy, comprising of both the self-reported rating scales and clinician scored rating scales. Also as noted in the earlier section, unfortunately strong evidence supporting the argument for regular reviews with the health professionals in these individuals' is lacking. Therefore, having a national register will help us to scope out the



changes in physical functioning in such individuals and research based on this database can be attempted to strengthen the arguments for clinical commissioning and service delivery.

The results indicated changes in physical functional abilities, especially in the Self-reported Barthel Index and Barthel Index but not in the Rivermead Mobility Index and Short Form 36 Version 2 outcome measures. Weak evidence pertaining to changes in physical functional abilities may be attributed to the variable presentation of baseline physical functioning related to cerebral palsy in a cross sectional study design. In cross sectional research, data is collected at a single point in time (Robson, 2011) and therefore research involving participants having varied baseline physical functioning cannot be utilised to draw strong conclusions, when there was no appropriate representation of individuals in each category. Due to limited scope of the research and fewer participants, Gross Motor Function Classification Scale was not considered for analysis. Gross Motor Function Classification Scale (GMFCS) is a 5-level classification system that describes the gross motor function of children and youth with cerebral palsy on the basis of their self-initiated movement with particular emphasis on sitting and mobility (Palisano et al., 1997) and valid up to the age of 21 year (Hanna et al., 2009). Hanna et al (2009) focused on the motor abilities of children and concluded that there was decline in gross motor abilities in the GMFCS levels III to V and this started before adolescence. Sandstrom et al., (2004) concluded from the research that sixteen individuals declined in function with predominance in level 1 on GMFCS. It appears from these studies (Hanna et al., 2009 and Sandstrom et al., 2004) that changes in physical functional abilities in adults with cerebral palsy will be dependent on the baseline physical functioning and motor syndromes related to cerebral palsy. Therefore, it is recommended that GMFCS is utilised in future researches or a similar valid scale to categorise the baseline physical functional abilities in adults with cerebral palsy before studying the changes.

## ***Factors affecting physical functional abilities in adults with cerebral palsy***

In this research, physical functioning of the participants was significantly lesser than the normal population data available from QualityMetric on the Shot Form 36 Version 2 (Saris-Baglana et al., 2011). Interview of the participants in the Phase two, indicated clearly that stiffness arising due to increased spasticity, pain and fatigue are the three major factors affecting physical functional abilities in adults with cerebral palsy. This is similar to the findings of Murphy et al. (1995), Jahnsen et al. (2003, 2004a), Zaffuto-Sforza (2005), Opheim et al. (2009) and Benner et al. (2017), these studies have identified that pain and fatigue affects physical functional abilities, especially walking and activities of daily living. However, the literature review did not identify increased stiffness due to spasticity which deteriorated with age, as one of the contributing factors impacting upon physical functional abilities.

Dimitrijevic et al. (2014) reviewed the evidence pertaining to assessment and treatment of spasticity in children with cerebral palsy and summarised the pathophysiology of spasticity and the impact on physical functional abilities. Pain is one of the major contributory factors to spasticity and in turn spasticity can also lead to pain. Dimitrijevic et al., (2014) suggests that up to 80% of all individuals with cerebral palsy suffer from some degree of spasticity. However, the literature review did not reveal use of an objective clinician assessment of fatigue, pain and spasticity in research. Hence, having an objective measure of fatigue, pain and spasticity would strengthen the evidence pertaining to changes in physical functional abilities changes in adults with cerebral palsy. Therefore, it is recommended that future research should incorporate objective measurement of pain, fatigue and spasticity in such individuals, which can be incorporated in the national register database.

In conclusion, there is an urgent need to initiate a national register incorporating self-perceived and actual physical functional abilities assessment in adults with cerebral palsy in order to strengthen evidence and

further evaluate physical functional abilities in such individuals to inform clinical commissioning groups and plan services.

**Study objective 2: Critical exploration of possible reasons and means to reduce the differences in self-perceived and actual physical functional abilities**

Participants completed four outcome measures in phase one, namely self-reported Barthel Index, Short Form 36 version 2, clinician assessed Barthel Index and Rivermead Mobility Index. Self-reported Barthel Index and Short Form 36 Version 2 measured self-perceived physical functional abilities and Barthel Index and Rivermead Mobility Index measured actual physical functional abilities in adults with cerebral palsy. The discrepancies between the self-perceived and actual physical functional abilities in adults with cerebral palsy were explored in the interview. The participants' interviews gave an opportunity to explore the possible reasons, and means, to reduce the difference in self-perceived and actual physical functional abilities, identified in the phase one analysis.

Four pertinent themes emerged from the phase two analysis; coping strategy, self-management, support and identity. Furthermore, some of the participants were also concerned regarding the deterioration in physical functional abilities in adulthood. Key themes identified within this study, self-management and identity were similar to findings of Mudge et al. (2016) research, involving twenty eight adults with cerebral palsy in New Zealand. Mudge et al. (2016) identified further three themes from the qualitative interview, acceptance to change, rethinking the future and interacting with health professions. Although there is research evidence pertaining to adjustments, coping strategies and the quality of life as a separate entity, there was no literature seen that explore these themes in terms of the discrepancies between self-perceived and actual physical functional abilities in such individuals. The significance of self-perceived abilities mirroring the actual physical functional abilities has been elaborated in the literature review and earlier sections of the

discussion chapter. The four themes that emerged from the phase two analyses are discussed here within the context of discrepancies between the self-perceived and actual physical functional abilities. Initially, the four themes are discussed separately in this context and then integrated with phase one findings to discuss the impact on clinical practice and future recommendations.

### ***Coping strategy***

The emergence of coping strategy as a theme in this research is similar to the findings of Lindsay (2016) and Shikako-Thomas et al. (2009) in youths. Lindsay (2016) systematically reviewed the qualitative evidence related to child and youth (2 to 25 years) experience and perspectives of cerebral palsy and identified that coping and independence are the two common themes emerging as part of the personal factor that influenced youths experience of living with cerebral palsy. Lindsay (2016) has indicated that there are several personal factors that influenced youth experience of living with cerebral palsy including threats to biography and normalcy. Lindsay (2016) recommended future research to address coping and self-care skills of young with cerebral palsy, which was explored in the interview. The interview demonstrated the engagement and disengagement coping strategies of individuals when dealing with everyday activities, this was also evident when they explained the self management strategies to manage their disabilities. Adolescents demonstrate that they are able to effectively cope with and create strategies to overcome many of the possible challenges (Shikako-Thomas et al., 2009). In relation the coping strategy, Lindsay (2016) identified that youth demonstrated resilience and strategies, for maintaining self-concept which affects body image and identity. Similar to these findings, the interview participants sought support from health care professional as part of their coping strategy to manage their disabilities. Therefore, coping strategies affects identity which was identified as a separate theme in this research.

Jahnsen, Villien, Stanghelle and Holm (2002) and Horsman, Suto, Dudgeon and Harris (2010) had reported the factors affecting coping strategies and its impact. Jahnsen, Villien, Stanghelle and Holm (2002) research concluded that early experience of predictability and balance between challenges and personal resources are prerequisite for developing sense of coherence, however these factors are less present in early socialisation of person with cerebral palsy. Interesting Horsman et al. (2010) in the research pertaining to coping strategies in adults aging with cerebral palsy has recognised from the interview that individuals with cerebral palsy had not been adequately informed of the possibility of experiencing secondary conditions and long term effects. Horsman et al. (2010) summarised that awareness, acceptance and action are the key coping strategies in adults with cerebral palsy. Individuals present with different coping strategies to attain autonomy or normality, sometimes at the expense of deteriorated body functions (Sandstrom, 2008) and clinicians should recognise this when planning care provision and support strategies.

### ***Self-management***

The second theme identified in phase two interview was self-management. Self-management is highly influenced by childhood experience in the schools, clinicians and more notably by parents. Davis et al., (2009) indicated that some young experienced difficulties in developing the skills needed to become independent because their parents were overprotective. Due to the childhood onset of disabilities, most of the time the individuals' adapt to the condition, however this could be task specific, raising anxiety and challenges in adulthood. The individuals need to be prepared from adolescent in management of their condition independently and seek support as and when required during adulthood.

Nevertheless, this is even more relevant currently due to limitation of resources, funding squeeze from the government and the necessity for improving productivity (with an emphasis on evidence based practice).

Currently, due to clinical commissioning restrictions, all NHS organisations are working to improve productivity and sustainability (NHS, 2016), this had made commissioning of the new services even more difficult, and therefore, there is an increased focus on self-management by care providers.

Warschusky et al. (2017) studied health self-management, transition readiness and adaptive behaviour in person with cerebral palsy and Myelomeningocele and concluded that there are condition specific self-management needs that are not captured by generic assessment measures. Therefore, there is an emphasis on condition specific self-management strategies in adults with cerebral palsy and more so when there is evidence indicating deterioration during adulthood in such individuals, from phase one and two findings of this study.

### ***Support***

The third theme that emerged from the participants' interview was seeking appropriate support from health professionals. As indicated in the interview, self-management is closely interlinked with the support provided by the clinicians. The clinicians have a role in creating awareness about aging with cerebral palsy, preparing the individuals' during adolescence and provide support to manage their condition independently. Although, the meaning of self-management and independence can contradict each other; in health, support is required for individuals with long term disability to improve skill set in self-management and also seek support from health professionals proactively, to avoid or reduce the impact of secondary complications. As highlighted in the interview, individuals' with cerebral palsy value the reviews provided by the Consultant and wanted to seek support for increased stiffness. This finding conforms with Bent et al. (2002) conclusion that a team approach to support young adults with physical disabilities is more likely to enhance participation in society when compared to the ad hoc approach. There is a service need for adequate follow up for adults with long term disabilities (Roebroek et al., 2009) and this will aid in preventing secondary

complications, thereby improving productivity of such individuals and reduce long term economic burden.

### ***Identity***

*'It was important for me to be seen as a normal person'* (participant quote)

The fourth theme that emerged from the interview of participants was preserving, and the need to recognise their Identity. Identity is intertwined with body image and coping strategy. Inferring from the participants interviewed in this research, the participant wants to be seen as 'normal' and strives to preserve this identity, even when body image changes due to aging. These findings are similar to McLaughlin and Coleman-Fountain (2014) research conclusion, suggested by the authors that identity challenge contains alternate imaginaries through which society could view 'analogous bodies'. Young people manage their bodies as something that is integral to their emerging identity, but also a potential threat that could undermine and give away their labour in making an 'ordinary' functioning body and life (McLaughlin and Coleman-Fountain, 2014).

### ***Integrating the themes in practice to reduce the discrepancies between self-perceived and actual physical functional abilities in adults with cerebral palsy***

It was noted from the interview that the participants were aware of the possible deterioration in physical functional abilities during adulthood and want to seek appropriate support from expert health professionals (who have knowledge in the management). Aging with a disability can have both a positive and negative influence on the adolescent's quality of life (Shikako-Thomas et al., 2009). In turn, aging can also directly affect physical functional abilities, Sandstrom (2008) has highlighted that in the fight to maintain daily activities, individuals with cerebral palsy often noted they ignored the management of their condition, resulting in negative bodily reactions with pain and decreased function. It is essential that this factor is taken into

consideration in research and practice when dealing with adults with cerebral palsy.

It is clear from this research that recognising coping strategy, self-management, support and identity are the four key themes to reduce the discrepancies between self-perceived and actual physical functional abilities in adults with cerebral palsy. This will have long term impact on the deterioration of physical functional abilities in such individuals. It is recommended that these themes are incorporated when commissioning and planning services for adults with cerebral palsy.

### **Study objective 3: Guidelines for potential long term management strategies for adult with cerebral palsy**

In this section, pertinent findings from the research are summarised and discussed in relation to the relevant government documents to identify the gaps. Identification of gaps will help one to recommend the appropriate strategies for long term management of adults with cerebral palsy, which in turn can also be utilised to inform the government guidelines.

The National Institute for Health and Care Excellence (NICE) was set up in 1999, the major role of NICE is to produce evidence based guidelines and advice for health services, public health and social care practitioners and provide a range of information services for clinical commissioners, practitioners and managers (NICE, 2017a). Therefore, it is essential that the new findings inform the guidance documents and the practitioners

The key findings from this research demonstrate that there is a significant correlation between self-perceived and actual physical functional abilities in adults with cerebral palsy and there are discrepancies between self-perceived and actual physical functional abilities in adults with cerebral palsy. Whilst exploring these discrepancies through phase two interview, four key themes emerged, namely coping strategy, self-management, support and identity.



Integration of these key findings in the current national guidelines is critically explored here.

The National Health Services (NHS) improvement plan; Putting people at the heart of public services (Department of Health (DoH), 2004b), emphasised on self-care, disease management and case management. This guidance document was followed by The National Service Framework (NSF) for Long-Term Conditions, (DoH, 2005). Integrated Services for People with Long-term Neurological Conditions: Evaluation of the Impact of the National Service Framework (Bernard, 2010) highlighted the lack of provision for auditing implementation of the recommendations in the NSF for long-term conditions and unclear implementation strategies. Unfortunately, due to change in government policies and NHS clinical commissioning, the role of national service frameworks was lost.

National Institute for Health and Care Excellence (NICE) published the Spasticity in under 19s: Management (NICE, 2012, updated 2016) (CG145) clinical guidance document, however this document concentrated only on the motor disorder of cerebral palsy. This guidance document recommended having a lead clinician to ensure continuing of care for spasticity management in transition and adult services. However, this document did not highlight the need for continuity of care in adult services due to the limited scope.

Transition from children's to adults' services for young people using health or social care services (NG43) was published by NICE in February 2016 (NICE, 2016). This guidance document recommended further research to identify relationship between transition and subsequent self-management and effective models of self-management. Although becoming independent is part of the transition to adulthood, personalised healthcare and helping people self-manage tends to be variable, therefore further research in this area is necessary for informing transition planning and preparation of young people.

Cerebral Palsy in under 25s: assessment and management was published by NICE in 2017 (NICE, 2017b), this document recognised that functional changes (including those involving eating drinking and swallowing, communication and mobility) and physical problems (pain and discomfort) may change over time for adults with cerebral palsy and recommended to take this into account when planning transition during adolescence.

Following the guidance document for cerebral palsy in under 25s, NICE is currently developing national guidelines on cerebral palsy in adults and the expected publication is in January 2019. Final Scope (9/2/17) of this document has highlighted that mobility and independence skills can deteriorate, with an impact on self-esteem, ability to concentrate and mental health.

The literature review found no evidence of recognition of the research findings in the government guidelines to inform practice, except self-management. It is strongly recommended that this national guidance document 'Cerebral palsy in adults' (NICE, in development GID-NG10031) recognises the discrepancies in self perceived and actual physical functional abilities in adults with cerebral palsy. It is recommended that the implementation strategy in this guidance should recommend that professionals when preparing and planning self-management in adults with cerebral palsy, have to consider coping strategy, identity and effect of aging on perception of the individuals. Further appropriate access to and support from relevant health professionals during adulthood is recommended to manage their condition and sustain independence skills.

As part of this study, a new model of clinical pathway is proposed for the management of adults with cerebral palsy. Clinical pathways are tools used to guide evidence based healthcare (Kinsman, et al., 2010), which can either be for a management process, diagnosis or intervention. Kinsman et al. (2010) reviewed the literature and developed five criteria to define a clinical pathway; the intervention or management pathway is a structured

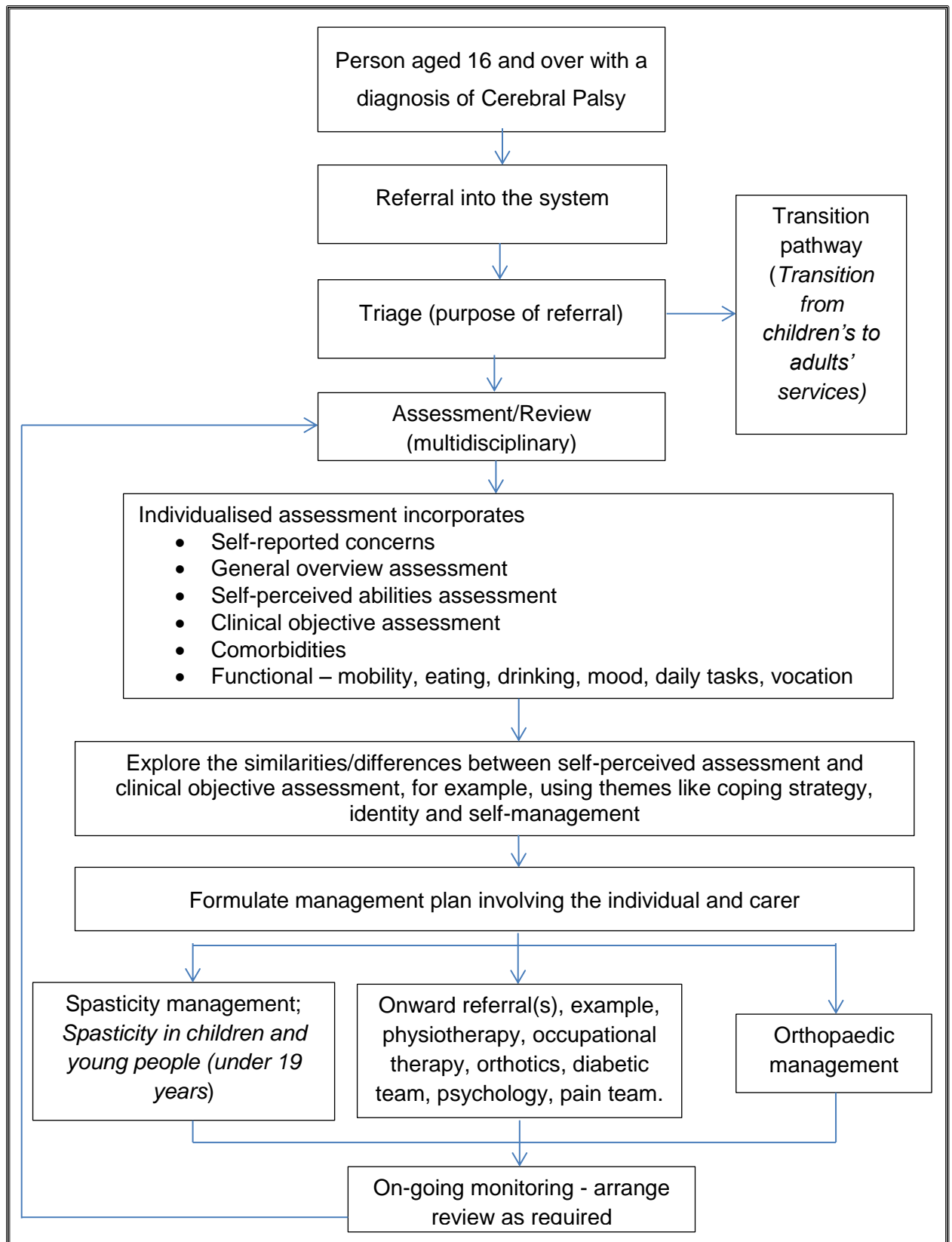
multidisciplinary plan of care, the interventions or management pathway is used to channel the translation of guidelines or evidence into local structures; the intervention details the steps in a course of treatment or care in a plan, pathway, algorithm, guideline, protocol or other 'inventory of action'; the intervention has a timeframe or criteria-based progression and the intervention is aimed to standardised care for a specific clinical problem, procedure or episode of healthcare in a specific population. These cardinal principles were utilised to formulate a pathway based on the findings from phase one and two of this study, to inform long term management strategy. The overarching goal of the clinical pathway was self-management. Already established clinical pathway from Cerebral palsy in under 25s (NICE, 2017b), Transition from children's to adults' services for young people using health and social care services (NICE, 2016) and Spasticity in under 19s: management (NICE, 2012, updated 2016) were integrated in this new proposed model of clinical pathway for efficient use of care delivery.

The proposed model of clinical pathway for management of individuals' with cerebral palsy can be found in figure 14. This clinical pathway was designed for all adults with a diagnosis of cerebral palsy. At the triage stage, the multidisciplinary team identifies the purpose of referral, individuals who require transition support follow the established transition pathway (NICE, 2016) and appropriate individuals follow the assessment or review route. Individualised assessment of the individual is completed by a member of the multidisciplinary team; this assessment incorporates self-reported concerns, general overview assessment, self-perceived and clinical objective assessment and functional assessment like mobility, eating, drinking, mood daily tasks and vocation. Additionally, pain, fatigue, bone density and other comorbidities like diabetes, blood pressure and osteoarthritis are also assessed at this stage (Andersson and Mattsson, 2001, Jahnsen et al., 2003, Jahnsen et al., 2004b, Sandstrom et al., 2004 and Opheim et al., 2009).

From this detailed assessment, the next step is to explore the similarities or differences between the self-perceived and actual physical functional abilities in such individuals, using the themes identified in phase two of this study, namely coping strategy, identity and self-management. From these assessments, the clinician devises a management plan; the overarching principle of this plan is proposed to be self-management and health promotion (World Health Organisation, no date). At this stage, the clinician can also refer the individual for spasticity management, orthopaedic management or onward referral as appropriate. The examples provided in the onward referrals of the pathway are based on the researcher's professional experience and literature reviews (Cox et al., 2005, Haak et al., 2009 and Morgan and McGinley, 2014), which are referral to physiotherapy, occupational therapy, orthotics, splints, diabetic team, psychology or pain team as appropriate. It is essential that this pathway acknowledges a system for on-going monitoring, which was one of the themes that emerged from the interview, these reviews will help to alleviate the individuals' concerns in relation to the deterioration of physical functional abilities.

This is a proposed model of the clinical pathway for adults with cerebral palsy. Therefore, further evaluation of this model is necessary to refine the pathway, this process would involve the individuals with cerebral palsy, clinicians and commissioners. Review of the local guidelines pertaining to management of adults with cerebral palsy and a desktop search exploring the local NHS Trusts and professional networks (Chartered Society of Physiotherapist, Royal College of Occupational Therapist and British Society of Rehabilitation Medicine) did not reveal any literature pertaining to management of adults with cerebral palsy. It is also essential to incorporate the key strategies identified in this research in the professional networks for training and consistency in approaches.

**Figure 14:** Proposed clinical pathway for management of adults with cerebral palsy



The participants within this study were significantly concerned about the deterioration of physical functional abilities in adulthood and wanted support from the health care at appropriate periods. Involving people in their own health and care: statutory guidance for clinical commissioning groups and NHS England (NHS England, 2017) recommends clinical commissioners and service providers to involve people in their health care so that they can effectively manage their own health and address their concerns when planning service delivery and commissioning services. This guidance document also states that it is a legal duty to involve people so as to improve health and wellbeing, improve care and quality, and improve financial sustainability and efficient allocation of resources. Therefore, the concerns of the adults with cerebral palsy should be incorporated in service planning and appropriate support should be set up for long term care.

It is evident from the earlier discussion that there is a greater need to establish a national register for adults with cerebral palsy to improve evidence pertaining to the management of long term condition. This is similar to recommendations of Cox et al. (2005) and Pharoah et al. (1997). A national register, I suggest will help to have a consistent outcome measure to track physical functioning in adults with cerebral palsy. This register can be utilised to strengthen evidence to inform practice, clinical commissioning and policy documents.

Results from phase two indicate that the participants were well aware about self-management and are interested to know how to manage their condition in long term. One participant indicated that he has stopped swimming since the cold water affected the stiffness in his legs. Kesiktas et al's. (2004) research study concluded that hydrotherapy (exercise pool with warm water) is an effective intervention to reduce spasticity. It is evident from the interview that this information, and access to, was not available to the individual. Participation in regular hydrotherapy would help the participant to manage their condition in long term and improve quality of life. Furthermore, health

promotion is a process of enabling people to increase control and strengthen the skills and capabilities of individuals to take control (World Health Organisation, no date), in order to achieve positive change. Therefore, it is essential that this information is available in the community for management of spasticity in adults with cerebral palsy as part of their health promotion. The researcher provided this information regarding hydrotherapy to the participant after completion of interview.

As indicated in the discussion of phase one and phase two results, there is a clear indication that age influences physical functional abilities, therefore future research is recommended to establish sound evidence pertaining to changes in physical functional abilities during adulthood in cerebral palsy. This is vital for the professionals and NHS Commissioners when planning service delivery and providing adequate training for the professionals whilst the available resource, including funding is limited. This can be achieved by establishing a national register to improve the evidence and track physical functioning during adulthood in cerebral palsy, to inform practice. Further, the outcome measures used for the national register should consider the factors outlined in Table 3, so that all components of physical functional abilities are measured to be relevant to current clinical practice.

## **Chapter six: NEW KNOWLEDGE, IMPLICATIONS AND RECOMMENDATION**

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### **Introduction**

The key findings from this research study are brought together in this chapter. This is based on the literature review, methodology, results and discussion chapters. The aim of the study was to explore the self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years with a view to develop self-management strategies. This aim was achieved using the study objectives. First the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy was identified using quantitative approach and the second objective to explore the possible reasons for and means to reduce these the differences between self-perceived and actual physical functional abilities was completed using qualitative approach. The research design flow chart can be found in Figure 3. The findings from both these approaches were utilised to develop guidelines for potential long term management strategies for such individuals.

### **New knowledge**

This research study has investigated the relationship and explored the possible reasons and means to reduce the differences between self-perceived and actual physical functional abilities in adults with cerebral palsy. This appears to be the first research to explore the relationship between self-perceived and actual physical functional abilities in adults with cerebral palsy. Few studies have suggested the importance of exploring the similarities or differences between self-perceived and actual physical functional abilities, however no study which has explored this relationship in all components of physical functional abilities had been found.

First, the research study has identified that there is a correlation between self-perceived and actual physical functional abilities and there are differences



between the self-perceived ability and clinician assessed participants' potential, this was evident in the Barthel index.

Second, the research study has indicated that there are actual changes in physical functional abilities (as assessed by the clinician) during adulthood and the individuals' also recognise these changes. Since the self-reported abilities were under reported when compared to the actual changes, it can be argued that the individuals' recognise the changes far earlier than actual changes occur. This was evident especially in dressing, mobility and using the stairs in this study.

Thirdly, the research study has also found that there is indication of deterioration in physical functional abilities during adulthood in individuals with cerebral palsy. These findings corroborate with the findings of other researchers as highlighted in the literature review.

Fourth, the research study has found the key themes for the possible differences and means to reduce the differences between self-perceived and actual physical functional abilities in adults with cerebral palsy. The four key themes that emerged from the participant interviews were coping strategy, identity, self-management and support.

Fifth, as argued in the literature review, mixed methodology is best suited to explore the self-perceived and actual physical functional abilities in individuals with cerebral palsy between the ages of 16 and 40 years. Designing a quantitative approach, followed by a qualitative approach in a sequential design has strengthened the findings of quantitative approach and helped to give a meaning to the findings. This appears to be the first study utilising mixed methodology to explore the self-perceived and actual physical functional abilities in adults with cerebral palsy. The research design flow chart of this study can be found in figure 3.

Sixth, the literature review has highlighted all the factors that could contribute to the changes in physical functional abilities during adulthood in cerebral

palsy. From the currently available evidence, I have summarised potentially contributing factors in table 3 using ICF biopsychosocial model (WHO 2002), which was not done earlier in the research. This will help future researchers and clinicians when attempting further research, explaining theory or in the management of individuals.

Seventh, from the findings of this study, a new clinical pathway was proposed for management of adults with cerebral palsy, this is illustrated in Figure 14. It is recommended that this pathway is embedded in the local and national guidance document to inform health care professionals and commissioners.

### **Implications for clinical practice**

The study has highlighted that there are differences between self-perceived and actual physical functional abilities in adults with cerebral palsy. This discrepancy would affect the individuals' ability to maintain daily tasks and quality of life. For example, from the findings it was clear that the participants were dependent on the carers and struggled to be independent in dressing and stair use, however during the clinical assessment they were independent in both the tasks. The dependency was due to various intrinsic and extrinsic factors, stiffness, cold weather and physical access issues. Additionally, the individuals reported the challenges ahead of the actual changes in function. As highlighted earlier, it is essential that the self-perceived and actual abilities mirror each other so that the individuals' abilities can be optimised to their potential. If this is not addressed earlier, the challenges can become established and have long term impact on the actual physical functioning abilities. Therefore, it is essential for the clinician to recognise the challenges reported by the individual in physical functioning and address this during care provision, since this can be a prequel to actual changes. It must also be noted that self-reported physical functional abilities are influenced by thoughts, feelings, beliefs and attitudes, of the individuals and this is not captured by the objective clinical assessments (Robson, 2011).

Exploring the discrepancies between self-perceived and actual physical functional abilities in adults with cerebral palsy through a qualitative approach revealed four pertinent themes, which were coping strategy, self-management, identity and support. Subsequently, exploring the discrepancies between self-perceived and actual physical functional abilities in such individuals using the themes, coping strategy and identity would enhance care delivery. Whilst the clinician formulates the long term management plan, it is essential to incorporate these themes when planning self-management. The proposed clinical pathway for the management of adults with cerebral palsy, incorporating the findings from this study, is presented in Figure 14. It is recommended that this clinical pathway is embedded in the local clinical pathway to enhance delivery of care.

The findings from this research supports the argument of other researchers (Murphy et al., 1995, Bottos et al., 2001, Opheim et al., 2009 and Hanna et al., 2009, Benner et al., 2017) that there is deterioration of physical functioning in adults with cerebral palsy and that the participants are concerned regarding this deterioration, as evident during the interview. It is essential that this is recognised during the contact with the adults with cerebral palsy and appropriate management plan is implemented. Due to the resource constraints in current service provision, the General Practitioners may be the only health care provider in regular contact with such individuals. Therefore, it is essential for the General Practitioners to recognise this during the routine appointments and refer to appropriate services as necessary. It is pertinent for the appropriate interventions to be timely, if the challenges in self-perceived abilities are not addressed at appropriate times, this can become an established long term disability (Cox et al., 2005).

From the interview, it was evident that the participants valued the support provided by the specialist clinician in this study, during the long term management of their condition. Therefore, appropriate access to and support

from relevant specialist clinician during adulthood is recommended to manage their condition and sustain independence skills.

### **Implications for learning and training**

This study adds to the evidence that there is deterioration of physical functional abilities in adults with cerebral palsy and has highlighted the importance of addressing these changes at appropriate times. This study has emphasised the significance of recognising the self-perceived physical functional abilities in individuals with cerebral palsy and to explore the feature. The participants valued the regular contact with the specialist clinician, this gives an opportunity for them to raise and address the concerns in relation to deterioration of physical functional abilities. Therefore, it is recommended that specialist training based on the new findings of this study is incorporated in educational settings, workshops, conference and through online resources

### **Implications for policy**

The implication of the new knowledge on the government policies and guidelines is discussed in the earlier section. This new knowledge from this research is timely since the 'Cerebral palsy in adults' (National Institute for Health and Care Excellence (NICE), in development GID-NG10031) is in development. It is recommended, that this guidance document recognises the new knowledge from this research, the discrepancies in self-perceived and actual physical functional abilities in adults with cerebral palsy, key themes that emerged during the interview, participants' concerns regarding deterioration of physical functional abilities in adulthood and the value of support provided by the clinician. It is essential, that these findings, including the new clinical pathways illustrated in Figure 14, are integrated with the policy guidelines for consistent approach in care provision across the country and avoid management plans that are based on 'postcode lottery' (Russel et al., 2013).

As suggested by Cox et al. (2005) and Pharoah et al. (1997), this research study has highlighted the significance of a national register to capture and track the physical functional abilities in adults with cerebral palsy, incorporating a consistent outcome measures. This would aid in improving the evidence in this area to inform care providers, clinical commissioners and policy makers.

### **Future research recommendations**

Due to the limited sample size, generalisation of the findings are limited. It is recommended that this research study is considered as a pilot study to plan a large scale cohort study to strengthen the findings of this research.

There was a significant gap between the phase one and phase two of this study. If the discrepancies between self-perceived and actual physical functional abilities were explored at the time of phase one data collection or nearer to the time, there may have been larger data to strengthen the findings. Therefore, it is recommended that these discrepancies identified in phase one are explored sooner than later.

In terms of tools, no standardised self-perceived outcome measure equivalent to Rivermead Mobility Index was found. Therefore, a self-reported mobility questionnaire was designed as a tool to measure self-perceived mobility in adults with cerebral palsy, based on the findings from this study. Desktop literature review was completed to identify a self-reported tool to measure mobility status, similar to the Rivermead Mobility Index (which is a clinician assessment tool). The widely used Short-Form 36 Version 2 (Ware and Sherbourne, 1992 and Brazier et al., 1992) and The Parkinson's Disease Questionnaire (PDQ-39) (Jenkinson, 1997) was considered during the construction of this questionnaire. Short-Form 36 Version 2 did not address the specifics of mobility like even and uneven ground mobility. Both the questionnaire Short-Form 36 Version 2 and The Parkinson's Disease Questionnaire (PDQ-39) did not appear to capture the variability in

functioning, which was one of the key findings from the interview. The participants in this study reported the variations in daily functioning for the possible discrepancy between self-perceived and actual physical functional abilities. Therefore, a new questionnaire was developed to address the specifics of mobility and to acknowledge the variability in physical functional abilities, as reported by the participants in this study.

A questionnaire is usually constructed in three methods. One method is to use empirical data or qualitative data to develop and fine a questionnaire (Poss, 1999). Second, development of a questionnaire can be based on a theoretical knowledge of the domain under study, utilising the experts in the field (Emami et al., 2010). And finally a questionnaire could be constructed by adapting a standardised scale (Poss, 1999). This study's literature view has revealed that there is no valid and reliable tool available to measure self-reported mobility status in adults with cerebral palsy. Currently reliable and valid tool is the Rivermead Mobility Index which is completed by the investigator (Collen, et al., 1991, Forlander and Bohannon, 1999 and Franchignoni et al., 2003), therefore Rivermead Mobility Index was adapted to a self-reported questionnaire format to capture self-perceived attitude and behaviour in relation to mobility. Boynton and Greenhalgh (2004) have listed the primary questions to consider before constructing a questionnaire: What information are you trying to collect, is a questionnaire appropriate, could you use an existing instrument, is the questionnaire valid and reliable, how should you present your questionnaire, what else should you include and finally, what should the questionnaire look like. This framework was utilised to construct a new questionnaire for reporting self-perceived mobility status, which could be used by other researchers.

The development of self-reported Barthel Index, which was developed from the clinical assessed Barthel Index, was considered whilst constructing this new questionnaire. The standardised Rivermead Mobility Index was utilised as a template to construct a new self-reported questionnaire for reporting

mobility status. Please see figure 15 for the proposed new self-reported mobility questionnaire. Eleven items were identified from the Rivermead Mobility Index and Barthel Index for this new questionnaire, namely turning in bed, lying to sitting, sitting balance, sit to stand, stand unsupported, walking indoors, walking outdoors (even grounds), walking outdoors (uneven grounds), stairs, picking something from the floor and carrying shopping bag. The items were arranged with increasing order of difficulties. The number of questions in each item was kept in a simple consistent language to avoid misinterpretation (Poss, 1999). Variability in functioning was built into the questionnaire for each item and examples for certain tasks were added to create a functional meaning to the items. The ten meter indoor walking question was based on the Rivermead Mobility Index, which is an approximate functional distance to walk indoors. In stair use, ability to use 3 to 4 steps was built in the questionnaire to capture the ability to do the front or back door steps. An example for picking up something from the floor was that of an individuals' independence to get up from their lounge chair, walk to the front door, pick up a post from the floor and return to the lounge chair. An example for carrying shopping bag was that of an individuals' ability to walk to the local shop, buy few times and carry the shopping bag home. Help to complete an item in this new questionnaire indicates assistance from their carer.

The scoring for each item is either 0,1 or 2 in increasing order of independence, the maximum score an individual can receive when being independent is 27 and a minimum score a person will receive for being totally dependent on a carer is 0.

**Figure 15:** Proposed self-reported mobility questionnaire

Name:

Date of Birth:

Hospital Number:

Diagnosis:

Completed by:

Date:

Items	What can I do?	Tick the most appropriate level
<b>Turning in bed</b>	I can turn independently in bed	/2
	I am independent in turning, but need help on some days	/1
	I need help from others to turn in bed	/0
<b>Lying to sitting</b>	I can sit from lying position independently	/3
	I can sit up independently but need help on some days	/2
	I need partial help to sit up	/1
	I cannot sit up; my carer helps me	/0
<b>Sitting balance</b>	I can sit independently	/2
	I can sit independently but on some days I need help from others	/1
	I cannot sit independently, need help from others	/0
<b>Sit to stand</b>	I can stand from a chair independently (with or without aid)	/2
	I can stand from a chair independently but on some days I need help	/1
	I need help from others to stand from a chair	/0
<b>Stand unsupported</b>	I can stand and do a task – like ironing, making a drink	/2
	My standing is variable between standing for few minutes and dependent on wheelchair	/1
	I need help from others to stand	/0
<b>Walking indoors</b>	I can walk indoors independently for 10 meters or more with or without any aid	/3
	I can walk indoors for independently for 10 meters but need supervision or help on some days	/2
	I can walk indoors for few meters with supervision or help from others	/1
	I am unable walk indoors	/0



Items	What can I do?	Tick the most appropriate level
<b>Walking outdoors (even grounds) like pavements, inside buildings</b>	I can walk independently on a flat surface, with or without aid	/3
	I can walk independently on a flat surface, but need help/supervision on some days	/2
	I need help from others to walk outdoors on even grounds	/1
	I am unable walk outdoors	/0
<b>Walking outdoors (uneven grounds) like parks, ice, snow, gravel, sand</b>	I can walk independently on uneven grounds (with or without aid)	/3
	I can walk independently on uneven ground s but need supervision or help on some days	/2
	I need help from others to walk outdoors on uneven grounds	/1
	I am unable to walk outdoors on uneven grounds	/0
<b>Stairs</b>	I can use a flight of stairs independently (with or without aid)	/3
	I can use a flight of stairs independently on most days but on some days I need help from others	/2
	I can only do 3 to 4 steps independently (with or without aid)	/1
	I am unable to do steps	/0
<b>Picking something from the floor</b>	I can walk 5 meters, pick up a fallen object/post from floor and walk back without any help	/2
	I can walk 5 meters, pick up a fallen object/post from floor and walk back without any help on some days only	/1
	I unable to pick up any object from floor	/0
<b>Carrying a shopping bag</b>	I can walk to the local shop, buy few items and carry the bag home independently	/2
	I can walk to the local shop, but a few items and carry the bag home independently on some days (example, weather, time of the day)	/1
	I am unable to walk to the local shop	/0
<b>Total Score</b>		<b>/27</b>
<b>Any other information</b>		

The British Medical Journal (BMJ) Checklist for developing a questionnaire (Boynton and Greenhalgh, 2004) was completed by the researcher, this can be found in table 9. Further rigorous study is essential to test the reliability and validity of this proposed self-reported mobility questionnaire.

**Table 9:** BMJ checklist for developing a questionnaire (Boynton and Greenhalgh, 2004), completed by the researcher

<b>Section</b>	<b>Quality criterion</b>	<b>Self-reported Mobility Questionnaire</b>
Title	Is it clear and unambiguous?	Yes
	Does it indicate accurately what the study is about?	Yes
	Is it likely to mislead or distress participants?	No
Introductory letter or information sheet	Does it provide an outline of what the study is about and what the overall purpose of the research is?	Yes
	Does it say how long the questionnaire should take to complete?	No, will be completed after pilot study
	Does it adequately address issues of anonymity and confidentiality?	Not applicable
	Does it inform participants that they can ask for help or stop completing the questionnaire at any time without having to give a reason?	Not applicable
	Does it give clear and accurate contact details of whom to approach for further information?	Not applicable at this stage
	If a postal questionnaire, do participants know what they need to send back?	Not applicable at this stage
Overall Layout	Is the font size clear and legible to an individual with 6/12 vision? (Retype rather than photocopy if necessary)	Yes
	Are graphics, illustrations and colour used judiciously to provide a clear and professional overall effect?	Yes
	Are the pages numbered clearly and stapled securely?	Not applicable at this stage

<b>Section</b>	<b>Quality criterion</b>	<b>Self-reported Mobility Questionnaire</b>
	Are there adequate instructions on how to complete each item, with examples where necessary?	Yes
Demographic information	Has all information necessary for developing a profile of participants been sought?	Yes
	Are any questions in this section irrelevant, misleading or superfluous?	Pilot testing will answer this checklist
	Are any questions offensive or otherwise inappropriate?	Pilot testing will answer this checklist
	Will respondents know the answers to the questions?	Pilot testing will answer this checklist
Measures (main body of questionnaire)	Are the measures valid and reliable?	Pilot testing will answer this checklist
	Are any items unnecessary or repetitive?	Pilot testing will answer this checklist
	Is the questionnaire of an appropriate length?	Pilot testing will answer this checklist
	Could the order of items bias replies or affect participation rates (in general, put sensitive questions towards the end)?	The questionnaire was constructed to report their mobility status in hierarchy, however this will altered depending on the feedback at pilot stage
Closing comments	Is there a clear message that the end of the questionnaire has been reached?	Yes
	Have participants been thanked for their co-operation?	Not applicable
Accompanying materials	If the questionnaire is to be returned by post, has a stamped addressed envelope (with return address on it) been included?	Not applicable
	If an insert (e.g. leaflet), gift (e.g. book token) or honorarium is part of the study protocol, has this been included?	Not applicable

Self-efficacy was not measured objectively in this study, the significance of this is elaborated in the study limitations section. It is recommended that self-efficacy is incorporated in the studies whilst exploring the self-perceived and actual physical functional abilities in adults with cerebral palsy.

The proposed clinical pathway for management of adults with cerebral palsy, which is based on the findings from this study, is illustrated in figure 14. It is essential that this clinical pathway is tested to enhance validity of the pathway. Therefore, it is recommended that further research is planned to explore the effectiveness of this clinical pathway, this evidence will help commissioners and clinicians to embed this pathway in delivery of care.

Cerebral palsy is a long term disability; therefore, the findings could be extrapolated in the management of individuals with other similar long term neurological conditions. This research study could be utilised to plan a further large scale research study in individuals with other long term neurological conditions. The proposed clinical pathway in figure 14 and the self-reported mobility questionnaire in figure 15 can also be utilised in other long term conditions. Hence, future research is recommended to study the validity and reliability of this clinical pathway and self-reported mobility questionnaire in other long term conditions.

### **Dissemination of research findings**

Finding novel ways to share findings can serve the interest of researchers, academics and participants (Hagen et al., 2017). I am intending to disseminate the findings in two journals, a conference, through my professional network and presentation of findings in the local NHS trusts. I have identified a potential journal and conference to disseminate the findings, this process will be started after acceptance of this research study. It is also essential to share information with the end user as highlighted by Hagen et al. (2017), therefore I am intending to do a brief report for sharing with the

interview participants and wider network organisation of individuals with disabilities.

## Chapter seven: CONCLUSION

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In this concluding chapter, I have elaborated the limitations of this study, followed by my reflection and reflexivity during this research process. This chapter concludes with the final thoughts.

### **Study limitations**

Recognition, acknowledgement and discussion of all potential important limitations by authors, if presented in an unbiased way, represent a crucial part of the scientific discourse and progress (Puhan et al., 2012). Puhan et al. (2012) has illustrated that openly discussing the limitations, helps the readers to correctly interpret the particular study and can inform future studies, which are likely to be higher quality if they address the limitations identified in the earlier studies. Discussing the study limitations will also help the readers to identify the context for application of the new knowledge without distortion. Limitations in this study were identified in sample size, tool selection and cognition of participants.

#### *Sample size*

A limitation of the study was low representation of adults with cerebral palsy in phase one and phase two. This may limit the generalisation of findings to the wider group. During the planning stage it was estimated that forty participants in phase one and a minimum of five participants for phase two will be necessary to draw wider inference from analysis and drawing up conclusions. Unfortunately, the researcher had couple of suspension periods during the research programme due to personal reasons. Due to the loss of momentum during the data collection period, it took some time to recruit more participants.

Further, at data collection site 1, the consultant was the 'gate keeper' (King and Harrocks, 2010), to identify potential participants from the clinic list based on the inclusion and exclusion criteria and post the participant information

sheet, participant invitation letter and participant consent letter on my behalf, at least three days prior to their attendance at the review clinic. Since I was not working with this site, due to data protection, I was not authorised to look at the clinic list or case notes to identify potential participants. During the course of data collection period, the consultant, assisting with the recruitment, moved on to take a different job role and this limited the recruitment of more participants.

Also, due to the change in research programme sponsoring organisation, the funding for the research programme was withdrawn, shortening the timescale of this research programme. Due to these factors the recruitment of the participants was limited. Nevertheless the new findings from this research cannot be ignored due to authenticity of the research. Therefore, these well-timed findings could strengthen the arguments and recommendations for the NICE guidance document currently in development. The findings of this study can be utilised to plan a larger cohort study to strengthen the evidence.

Rubin and Rubin (2005, p67) has argued in a responsive interview, reality is complex, to accurately portray that complexity, the researcher need to gather contradictory or overlapping perceptions, subsequently, as the interview is continued, the conversation should add less and less to the themes, until all the researcher hears are the same matters over and over again, this is called 'saturation point'. Small number of participants was interviewed in this study, therefore there may be distortion of the outcome (Patton, 1990). The emergence of four themes from the interviews was very strong and the frequency was high. Although saturation was not achieved, I strongly feel that the four key themes cannot be ignored, there may have been emergence of more themes if the interview would have been continued. Also, from my clinical experience, I am confident that these four themes are pertinent in physical functioning of adults with cerebral palsy.

### *Tool selection*

Self-efficacy was not measured objectively due to limited scope of inference in this research and fewer participants. Van Der Slot et al., (2010) concluded that significant number of adults with bilateral spastic cerebral palsy encountered difficulties in social participation and had a low perceived health-related quality of life for physical function. Building of self-efficacy is reliant on financial, educational and functional status of the individuals (Becker and Schaller, 1995). General Self-efficacy Scale (GSE) (Schwarzer and Jerusalem, 1995) was considered to be utilised during the interview process, however due to the limited scope of this research and fewer number of participants, it was felt that the effect of this measurement will be limited. It is recommended that future research measures the self-efficacy when evaluating coping strategies.

### *Cognition of participants*

Cognition was not formally assessed in this research. An individual's cognition and relation to the environment may be important in improving health (Jahnsen, Villien, Stanghelle and Holm, 2002 and Jacobson et al., 2013). Sustaining independence in activities of daily living during adulthood may be dependent on the level of physical and cognitive competency (James, Ziviani and Boyd, 2013). Cognition could also impact learning new skills and seeking appropriate support.

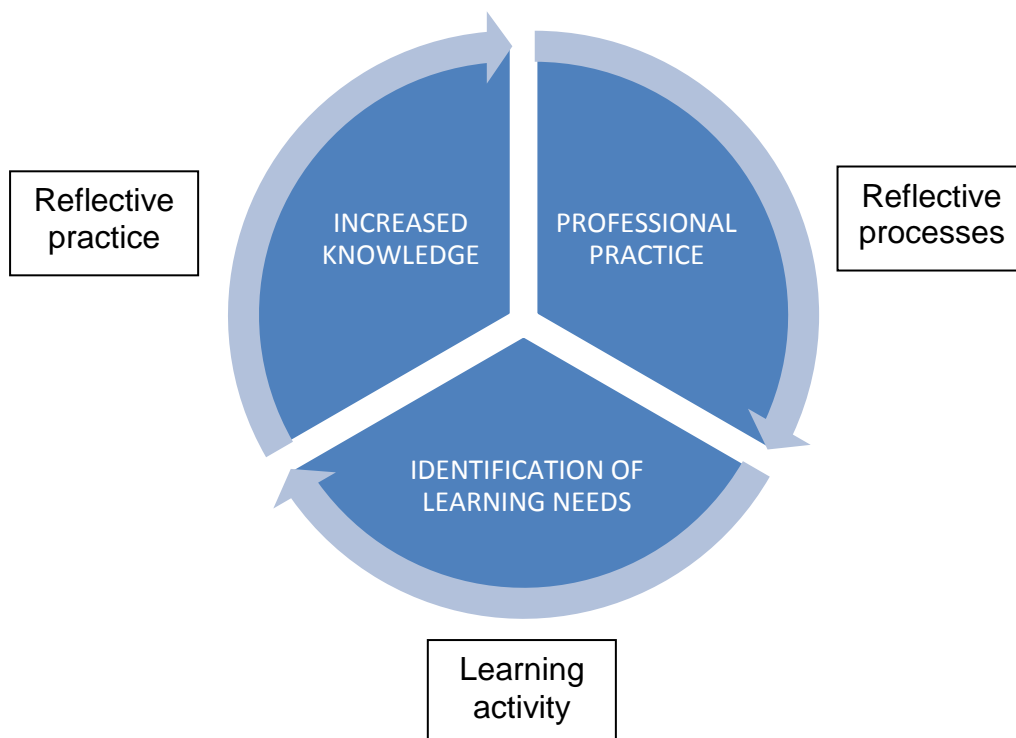
Therefore, it is recommended that future studies incorporate this when attempting to do an interview. However, this was considered in this research when selecting the interview participants for phase two, the participants appeared to have normal cognition and were able to articulate the challenges, suggest some changes to the provision of their care and provide rich information pertaining to the researched area. The participants for the interview were carefully chosen so that they provide a full and sophisticated understanding of the phenomena under study (Hansen, 2006).



## Reflective practice and reflexivity in the process of research

Reflective practice is learning and developing through examining what we think happened on any occasion, and how we think others perceived the event and thus, opening our practice to scrutiny by others, and studying data and texts from the wider sphere (Bolton, 2009, p13). Reflectivity is finding strategies to question our own attitudes, thought processes, values, assumptions, prejudices and habitual actions, to strive to understand our complex roles in relation to others (Bolton, 2009). Bolton (2009, p13) has explained that to be reflexive is to examine how we involve in creating social or professional structures counter to our own values. Reflective practice is crucial to professional development. Jasper has proposed a professional development triangle (Jasper, 2006, p.48). This triangle was adapted in circle format to denote that the professional development is continuous circle (process) and each aspect is dependent on each other, this is illustrated in figure 16.

**Figure 16:** Adapted version of The Professional Development Triangle, reproduced from Jasper (2006)



Making changes to the professional practice through reflection and research will have a direct impact on the profession. This adapted version of The Professional developmental triangle (Jasper, 2006, p48), illustrated in figure 16, facilitated me to identify my learning needs and address them to improve my research skills. Being reflexive during this process, helped me to understand the underlying meaning of the participants' response in order to be non-biased. Further, in qualitative research, the researcher brings their subjective values and meanings to their endeavours, this subjectivity should not be treated as a problem to be avoided, but as a resource that can be developed in ways that augment and intensify social research (King and Horrocks, 2010). Therefore, I have tried to be reflexive throughout this research.

I have used reflexivity in this research from the inception of the idea. The taught modules of the research programme helped me to structure the research process and lay the foundation to be involved in this research. SWOT (Strength, Weakness, Opportunities and Threats), is a simple and flexible tool to structure known and unknown facts for up skilling (Piercy and Giles, 1989). I used this tool to identify the gaps in my research skills. I engaged well with my supervisors throughout the tutorial sessions and this helped me to be reflexive in my thought process. An extract from the tutorial session with my supervisor can be found in Appendix 20. This tutorial session focused on concerns in relation to access to participants, sample size, license for an outcome measure and assessing capacity of the participants. Especially, the discussion around assessing the participants' capacity, tools considered to assess the individual and the resulting outcome, demonstrates the reflexive engagement in the planning process.

Some examples of reflection on the research process and my reflexive engagement, is reported here, during the process of conceiving research idea, data collection, data analysis and writing up.

### ***Research Idea***

The research proposal was shared with the Neurology User and Carer Forum which is run and led by the service users and the team colleagues. I was asked questions pertaining to language of the participant information sheet, participant consent form, sub-group distribution of participants, sample size, validity of the tools and application of research findings in practice. This reflective exercise helped me to structure the research process and prepare myself for NHS ethics application. The presentation at the user and carer forum was a good reflective exercise and the feedback I received was supportive of the research study idea. I felt reassured that the study aim and the procedure adopted were in accordance with the needs of the individuals living with long term neurological conditions. When I embarked on the research project my objective was to identify the changes in physical functioning in adults with cerebral palsy. Tutorial sessions with the supervisors and colleagues, helped me understand that this is beyond the scope of this research programme and will involve considerable resources. Reflecting on the literature review and the tutorial sessions, I was able to refine my research question in order answer them through a structured methodology. As a physiotherapist, I had good knowledge and experience in quantitative research, being reflexive helped to identify the gaps in my research skills, two gaps identified in the research skills were to learn about process of qualitative research and interviewing the participants. I attended relevant workshops in qualitative research and completed a pilot study to develop and enhance my skills.

### ***Data collection***

Structuring the research design in two phases gave me an opportunity to talk to the participant and understand the self-perception of an individual with disabilities; this helped me to be reflexive in my understanding of the disabilities. Being reflexive, I was able to explore the possible reasons for the discrepancies between self-perceived and actual physical functional abilities

without any prejudice. Furthermore, the feedback from the participants also helped to continue the research. For instance, one participant stated that he was delighted that this gave him an opportunity to think about his skills, changes in physical functional abilities from childhood and reflect on self-management. All the participants were interested in knowing the results of this study.

Testing the interview guide with a potential participant and a colleague and transcribing the audio recording, helped me to be reflexive, to avoid directional questions when probing. This was important as it restricted any potential influence and subjective opinion on the researched area.

Being reflexive, helped me to appreciate the barriers to physical functioning of the participants. I have started to recognise this in my professional practice and whilst working with the patients, I have started to explore the barriers to exercise and community participation in a non-biased interview style. Introduction of this new concept in my professional practice has helped me to plan a better intervention strategy with a patient centered focus and encourage self-management.

### ***Data analysis***

Interpretation of the data will depend on the theoretical standpoint taken by researchers (Sutton and Austin, 2015). Sutton and Austin (2015) have recommended that researchers be aware of their standpoints in the research, without this awareness it is easy to slip into interpreting other people's narratives from researcher's viewpoint, rather than that of the participants. I was very careful during transcribing to capture the non-verbal gestures and pauses. The opportunity to compare my interview analysis with the supervisor's independent analysis, helped to retain the authenticity of the participants' viewpoint and not be biased when identifying the themes.

In phase one of this study, I chose to use four outcome measures. Reflecting on the analysis, I realised that the role of Rivermead Mobility Index was

limited due to the non-availability of an equivalent self-rating scale for direct comparison. The effect of this outcome measure on the analysis and results were limited. On reflection, I feel that I could have avoided using this outcome measures and this would not have distorted the current findings. I have responded to this challenge and proposed a new self reported mobility questionnaire in figure 15, which can be utilised in the future to expand the research findings.

### ***Write-up***

Authenticity of the results from the data was independently verified by the supervisor and a colleague. Being reflexive helped me to avoid distortion whilst reporting the results and discussing the findings. Regular supervision with my supervisory team during this period has extensively helped me to retain the authenticity of the findings. Utilising the information from the field notes and tutorial session has also helped me to retain authenticity whilst reporting the study. However, in the discussion section of this report, I have stated my professional perspectives and separated this from the participants' viewpoint to retain the truthfulness.

### ***Research timeline***

The progress of the research was significantly and adversely affected due to my suspension of studies. I suspended my research programme for thirteen months in two blocks during various stages of the research due to personal reasons and change of job role in NHS which was beyond my control. This impacted significantly on data collection and timescale for write up of the research. Being reflexive, helped me to understand the research purpose and the impact this study would have on clinical practice; this understanding facilitated me to continue this research study.

However, if I were given an opportunity to do this study again, I would take time off from work and do this programme as a full time research student. This would give me freedom to concentrate on the research programme. Or

else, I would request significant periods of time, and study leave to do data collection and write up.

Nevertheless, I am now elated that I was able to bring new knowledge to the researched area, in order to inform clinical practice. I am glad that the findings can be timely to inform NICE guidelines which is in development.

### **Final Thoughts**

The study has revealed that there are discrepancies between the self-perceived and actual physical functional abilities and health professionals should address this utilising the themes, which are coping strategy, self-management, support and identity. Adults with cerebral palsy are concerned regarding the changes in their physical functioning and would like to have appropriate support through regular contact with a health care professional. It is recommended that this is incorporated in the development of clinical guidance document in order to appraise the clinical commissioners and the service providers. Further, it is recommended that a national register is established to record the physical functional abilities in adults with cerebral palsy.

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## APPENDICES

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**Appendix 1:** Critical appraisal of Prognostic Studies documentation format suggested by the Oxford Centre for Evidence Based Medicine

**Appendix 2:** Short Form 36 Version 2

**Appendix 3:** Self-reported Barthel Index

**Appendix 4:** Rivermead Mobility Index

**Appendix 5:** Barthel Index

**Appendix 6:** Interview guide

**Appendix 7:** Participant invitation letter (site 1 and site 2)

**Appendix 8:** Participant information sheet

**Appendix 9:** Participant consent form

**Appendix 10:** Certificate – Introduction to Good Clinical Practice (GCP)

**Appendix 11:** Copy of letter to the General Practitioner and/or the participant's consultant

**Appendix 12:** Risk analysis and management plan

**Appendix 13:** Ethical approval from University of Huddersfield's School Research and Ethics Panel (SREP)

**Appendix 14:** Hand-out prepared for Neurology User and Carer Forum

**Appendix 15:** Ethical approval from The NHS Research Ethics Committee (REC)

**Appendix 16:** Response to the clarification sought from The NHS Research Ethics Committee (REC)

**Appendix 17:** Site 1 - NHS Trusts' Research and Development approval letter

**Appendix 18:** Site 2 - NHS Trusts' Research and Development approval letter

**Appendix 19:** License agreement from the Office of Grants and Scholarly Research of Optuminsight Life Sciences, Inc.

**Appendix 20:** Extract from tutorial session

**Appendix 1: Critical appraisal of Prognostic studies (© Oxford Centre for Evidence Based Medicine)**

Are the results of the study valid? (Internal Validity)

- 1. Was the defined representative sample of patients assembled at a common (usually early) point in the course of their disease?**

*What is best?*

*Where do I find the information?*

*This paper: Yes    No    Unclear*  
*Comment:*

- 2. Was patient follow-up sufficiently long and complete?**

*What is best?*

*Where do I find the information?*

*This paper: Yes    No    Unclear*  
*Comment:*

- 3. Were outcome criteria either objective or applied in a 'blind fashion'?**

*What is best?*

*Where do I find the information?*

*This paper: Yes    No    Unclear*  
*Comment:*

**4. If subgroups with different prognoses are identified, did adjustment for important prognostic factors take place?**

*What is best?*

*Where do I find the information?*

*This paper: Yes    No    Unclear*  
*Comment:*

What are the results?

**5. How likely are the outcomes over time?**

**6. How precise are the prognostic estimated?**

**7. Can I apply this valid, important evidence about prognosis to my patient?**

# Your Health and Well-Being

Participant number: \_\_\_\_\_

**This survey asks for your views about your health. This information will help keep track of how you feel and how well you are able to do your usual activities. *Thank you for completing this survey!***

**For each of the following questions, please tick the one box that best describes your answer.**

**1. In general, would you say your health is:**

Excellent	Very good	Good	Fair	Poor
1	2	3	4	5

**2. Compared to one year ago, how would you rate your health in general now?**

Much better now than one year ago	Somewhat better now than one year ago	About the same as one year ago	Somewhat worse now than one year ago	Much worse now than one year ago
1	2	3	4	5



**3. The following questions are about activities you might do during a typical day. Does your health now limit you in these activities? If so, how much?**

Yes, limited a lot	Yes, limited a little	No, not limited at all
▼	▼	▼

- a Vigorous activities, such as running, lifting heavy objects, participating in strenuous sports.....  1 .....  2 .....  3
- b Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf .....  1 .....  2 .....  3
- c Lifting or carrying groceries .....  1 .....  2 .....  3
- d Climbing several flights of stairs .....  1 .....  2 .....  3
- e Climbing one flight of stairs .....  1 .....  2 .....  3
- f Bending, kneeling, or stooping .....  1 .....  2 .....  3
- g Walking more than a mile .....  1 .....  2 .....  3
- h Walking several hundred yards .....  1 .....  2 .....  3
- i Walking one hundred yards .....  1 .....  2 .....  3
- j Bathing or dressing yourself .....  1 .....  2 .....  3

**4. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?**

All of the time	Most of the time	Some of the time	A little of the time	None of the time
▼	▼	▼	▼	▼

- a Cut down on the amount of time you spent on work or other activities.....  1.....  2.....  3.....  4.....  5
- b Accomplished less than you would like .....  1.....  2.....  3.....  4.....  5
- c Were limited in the kind of work or other activities .....  1.....  2.....  3.....  4.....  5
- d Had difficulty performing the work or other activities (for example, it took extra effort) .....  1.....  2.....  3.....  4.....  5

**5. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?**

All of the time	Most of the time	Some of the time	A little of the time	None of the time
▼	▼	▼	▼	▼

- a Cut down on the amount of time you spent on work or other activities.....  1.....  2.....  3.....  4.....  5
- b Accomplished less than you would like .....  1.....  2.....  3.....  4.....  5
- c Did work or other activities less carefully than usual .....  1.....  2.....  3.....  4.....  5

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6. During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbours, or groups?

Not at all	Slightly	Moderately	Quite a bit	Extremely
▼	▼	▼	▼	▼
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

7. How much bodily pain have you had during the past 4 weeks?

None	Very mild	Mild	Moderate	Severe	Very severe
▼	▼	▼	▼	▼	▼
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5	<input type="checkbox"/> 6

8. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?

Not at all	A little bit	Moderately	Quite a bit	Extremely
▼	▼	▼	▼	▼
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

9. These questions are about how you feel and how things have been with you during the past 4 weeks. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the past 4 weeks...

All of the time	Most of the time	Some of the time	A little of the time	None of the time
▼	▼	▼	▼	▼

- a Did you feel full of life?..... 1..... 2..... 3..... 4..... 5
- b Have you been very nervous? ..... 1..... 2..... 3..... 4..... 5
- c Have you felt so down in the  
dumps that nothing could  
cheer you up? ..... 1..... 2..... 3..... 4..... 5
- d Have you felt calm and  
peaceful? ..... 1..... 2..... 3..... 4..... 5
- e Did you have a lot of energy? ..... 1..... 2..... 3..... 4..... 5
- f Have you felt downhearted  
and low?..... 1..... 2..... 3..... 4..... 5
- g Did you feel worn out? ..... 1..... 2..... 3..... 4..... 5
- h Have you been happy? ..... 1..... 2..... 3..... 4..... 5
- i Did you feel tired? ..... 1..... 2..... 3..... 4..... 5

10. During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting with friends, relatives, etc.)?

All of the time	Most of the time	Some of the time	A little of the time	None of the time
▼	▼	▼	▼	▼
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

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 (SF-36v2® Health Survey Standard, United Kingdom (English))

**11. How TRUE or FALSE is each of the following statements for you?**

Definitely true	Mostly true	Don't know	Mostly false	Definitely false
▼	▼	▼	▼	▼

- a I seem to get ill more  
easily than other people .....  1 .....  2 .....  3 .....  4 .....  5
- b I am as healthy as  
anybody I know .....  1 .....  2 .....  3 .....  4 .....  5
- c I expect my health to  
get worse .....  1 .....  2 .....  3 .....  4 .....  5
- d My health is excellent .....  1 .....  2 .....  3 .....  4 .....  5

***Thank you for completing these questions!***

### Appendix 3 : Self-reported Barthel index

**Participant Number:**

- These are some questions about your ability to look after yourself
- They might not seem to apply to you; please answer them all
- Tick one box in each section

#### **Bathing**

In the bath or shower, do you.....manage on your own?

need help getting in and out?

need other help?

never have a bath or shower?

need to be washed in bed?

#### **Stairs**

Do you climb stairs at home.....without any help?

with someone carrying your frame?

with someone encouraging you?

with physical help?

not at all?

don't have stairs?

#### **Dressing**

Do you get dressed.....without any help?

just with help with buttons?

with someone helping you most of the time?

#### **Mobility**

Do you walk indoors.....without any help apart from a frame?

with one person watching over you?

with one person helping you?

with more than one person helping you?

not at all?

Or do you use a wheelchair independently?

**Transfer**

Do you move from bed to chair.....on your own?

with a little help from one person?

with a lot of help from one or more people?

not at all?

**Feeding**

Do you eat food.....without any help?

with help cutting food or spreading butter?

with more help?

**Toilet use**

Do you use the toilet or commode.....without any help?

with some help but can do something?

with quite a lot of help?

**Grooming**

Do you brush your hair and teeth and wash your face.....without help?

with help?

**Bladder**

Are you incontinent of urine.....never?

less than once a week?

less than once a day?

more often?

Or do you have a catheter managed for you?

**Bowels**

Do you soil yourself..... never?

occasional accident?

all the time?

Or do you need someone to give you an enema?

## Appendix 4: The Rivermead Mobility Index

Name: \_\_\_\_\_

<b>Day</b>							
<b>Month</b>							
<b>Year</b>							
<b>Topic and Question:</b>							
<b>Turning over in bed:</b> Do you turn over from your back to your side without help?							
<b>Lying to sitting:</b> From lying in bed, do you get up to sit on the edge of the bed on your own?							
<b>Sitting balance:</b> Do you sit on the edge of the bed without holding on for 10 seconds?							
<b>Sitting to standing:</b> Do you stand up from any chair in less than 15 seconds and stand there for 15 seconds, using hands and/or an aid if necessary?							
<b>Standing unsupported:</b> (Ask to stand) Observe standing for 10 seconds without any aid							
<b>Transfer:</b> Do you manage to move from bed to chair and back without any help?							
<b>Walking inside:</b> (with an aid if necessary): Do you walk 10 meters, with an aid if necessary, but with no standby help?							
<b>Stairs:</b> Do you manage a flight of stairs without help?							
<b>Walking outside:</b> (even ground): Do you walk around outside, on pavements, without help?							
<b>Walking inside:</b> (with no aid): Do you walk 10 meters inside, with no caliper, splint, or other aid (including furniture or walls) without help?							
<b>Picking up off floor:</b> Do you manage to walk five meters, pick something up from the floor, and then walk back without help?							
<b>Walking outside:</b> (uneven ground): Do you walk over uneven ground (grass, gravel, snow, ice etc) without help?							
<b>Bathing:</b> Do you get into/out of a bath or shower and to wash yourself unsupervised and without help?							
<b>Up and down four steps:</b> Do you manage to go up and down four steps with no rail, but using an aid if necessary?							
<b>Running:</b> Do you run 10 meters without limping in four seconds (fast walk, not limping, is acceptable)?							
<b>Total</b>							

---

Downloaded from [www.rehabmeasures.org](http://www.rehabmeasures.org)

The Rivermead Mobility Index is provided courtesy of Dr. Derick Wade and the Oxford Centre for Enablement.



**Appendix 5 : The Barthel Index**

**Patient Number:** \_\_\_\_\_

**Rater Name:** \_\_\_\_\_

**Date:** \_\_\_\_\_

**INDEX**

**Activity** \_\_\_\_\_ **Score**

**FEEDING**

0 = unable

5 = needs help cutting, spreading butter, etc., or requires modified diet

10 = independent

\_\_\_\_\_

**BATHING**

0 = dependent

5 = independent (or in shower)

\_\_\_\_\_

**GROOMING**

0 = needs to help with personal care

5 = independent face/hair/teeth/shaving (implements provided)

\_\_\_\_\_

**DRESSING**

0 = dependent

5 = needs help but can do about half unaided

10 = independent (including buttons, zips, laces, etc.)

\_\_\_\_\_

**BOWELS**

0 = incontinent (or needs to be given enemas)

5 = occasional accident

10 = continent

\_\_\_\_\_

**BLADDER**

0 = incontinent, or catheterized and unable to manage alone

5 = occasional accident

10 = continent

\_\_\_\_\_

**TOILET USE**

0 = dependent

5 = needs some help, but can do something alone

10 = independent (on and off, dressing, wiping)

\_\_\_\_\_

**TRANSFERS (BED TO CHAIR AND BACK)**

0 = unable, no sitting balance

5 = major help (one or two people, physical), can sit

10 = minor help (verbal or physical)

15 = independent

\_\_\_\_\_

**MOBILITY (ON LEVEL SURFACES)**

0 = immobile or < 50 yards

5 = wheelchair independent, including corners, > 50 yards

10 = walks with help of one person (verbal or physical) > 50 yards

15 = independent (but may use any aid; for example, stick) > 50 yards

\_\_\_\_\_

**STAIRS**

0 = unable

5 = needs help (verbal, physical, carrying aid)

10 = independent

\_\_\_\_\_

**TOTAL (0-100):** \_\_\_\_\_

## The Barthel ADL Index: Guidelines

---

1. The index should be used as a record of what a patient does, not as a record of what a patient could do.
2. The main aim is to establish degree of independence from any help, physical or verbal, however minor and for whatever reason.
3. The need for supervision renders the patient not independent.
4. A patient's performance should be established using the best available evidence. Asking the patient, friends/relatives and nurses are the usual sources, but direct observation and common sense are also important. However direct testing is not needed.
5. Usually the patient's performance over the preceding 24-48 hours is important, but occasionally longer periods will be relevant.
6. Middle categories imply that the patient supplies over 50 per cent of the effort.
7. Use of aids to be independent is allowed.

## References

---

Mahoney FI, Barthel D. "Functional evaluation: the Barthel Index."  
*Maryland State Medical Journal* 1965;14:56-61. Used with permission.

Loewen SC, Anderson BA. "Predictors of stroke outcome using objective measurement scales."  
*Stroke*. 1990;21:78-81.

Gresham GE, Phillips TF, Labi ML. "ADL status in stroke: relative merits of three standard indexes."  
*Arch Phys Med Rehabil*. 1980;61:355-358.

Collin C, Wade DT, Davies S, Horne V. "The Barthel ADL Index: a reliability study."  
*Int Disability Study*. 1988;10:61-63.

## Copyright Information

---

The Maryland State Medical Society holds the copyright for the Barthel Index. It may be used freely for non-commercial purposes with the following citation:

Mahoney FI, Barthel D. "Functional evaluation: the Barthel Index."  
*Maryland State Med Journal* 1965;14:56-61. Used with permission.

Permission is required to modify the Barthel Index or to use it for commercial purposes.

## Appendix 6 : Interview Guide

Name:

Research Id:

Introduction

Do you want to ask me anything before we start?

Are you happy for me to record this interview?

START RECORDING

- Purpose of the interview
  - This is follow on from Phase one of the study
  - This interview will explore the results of the Phase one assessments
  - We will look at how you manage you physical skills and what you do to maintain these skills
  - What would you like to do to maintain your skills

(Introductory statement: In this interview, first I will be explaining how you did in the first phase of the study. We used 4 outcome measures to assess your daily tasks. I did quite a lot of analysis with it and this is a follow on interview from that, to discuss how you performed and how you could manage your skills or difficulties.

- Any questions?

### **Interview**

1. Results of the Phase one study
  - a. Tell me how you think you performed on the assessments?
  - b. Present the results (summarise the performance from the outcome measures)
2. Tell me what do you think about the differences between your actual performance and what you said you could do?
3. Why do you think it is different? Tell me of any difficulties you have in doing these tasks daily? Tell me if anything stops you from doing some of these tasks?
4. How would like to reduce the differences between what you did and what you said you could do?
5. Have you any ideas on how you can manage these difficulties? Or what kind of help would you like to maintain your functional skills in future?

Any questions?

Is there anything more you want to tell me?

What do you think about this interview?

Would you like to know about the results of these interviews?

Thank you

STOP RECORDING

**Telephone:**

**Email:**

**Private & Confidential**

Participant name and address

Date

Dear Mr/Ms Participant

**Date of Birth:**

**NHS Number:**

**Re: Research**

My colleague, Ram Krishnamurthy is undertaking research, investigating the daily task performance in adults with Cerebral Palsy with a view to develop self-management strategies. He is working as Senior Physiotherapist with the Community Neurology Rehabilitation Team, St Mary's Hospital, Leeds. This research is part of his doctoral research programme.

I have enclosed the information sheet and consent form for more information.

If you would like to take part or would like more information about taking part in this research, please do not hesitate to contact him. You can also return the slip below in the enclosed self-addressed envelope and he will contact you. Thank you for your time.

Yours sincerely

**Consultant  
St James's Hospital**

---

I am happy for Ram Krishnamurthy to contact me to discuss about taking part in this research. I understand that I can change my mind about taking part after I have received more information.

Name:

Phone Number and Email address:

Address:

Signature:



**Ram Krishnamurthy**

**Telephone: 0113 305 5082**

**Email: ramkrish@nhs.net**

**Community Neurological  
Rehabilitation Team  
St Mary's Hospital  
Willow House Green Hill Road  
Leeds  
LS12 3QE**

**Private & Confidential**

**Fax: 0113 305 5081**

Participant name and address

Date

Dear Mr/Ms Participant

**Date of Birth: 01 Nov 1996**

**NHS Number: 601 285 4706**

**Re: Research**

I am undertaking research, investigating the daily task performance in adults with Cerebral Palsy with a view to develop self-management strategies. This is part of a doctoral research programme.

I have enclosed the information sheet and consent form for more information.

If you would like to take part or would like more information about taking part in this research, please do not hesitate to contact me. You can also return the slip below in the enclosed self-addressed envelope and I will contact you. Thank you for your time.

Yours sincerely

**Ram Krishnamurthy  
Specialist Physiotherapist  
Community Neurological Rehabilitation Team**

---

I am happy for Ram Krishnamurthy to contact me to discuss about taking part in this research. I understand that I can change my mind about taking part after I have received more information.

Name:

Phone Number and Email address:

Address:

Signature:

**PARTICIPANT INFORMATION SHEET**

My name is Ram Krishnamurthy and I am working as a Specialist Physiotherapist with the Community Neurology Team, Leeds Community Healthcare NHS Trust. I am interested in looking at the performance of daily tasks in adults with Cerebral Palsy. This research is being undertaken as part of a doctorate research programme.

**Purpose of the research:** To explore the self-perceived and actual skills in daily tasks in individuals with Cerebral Palsy between the ages of 16 and 40 years with a view to developing self-management strategies

You are being invited to take part in this research. Please take time to read the following information about the research. Please don't hesitate to ask any questions if anything is not clear or you would like more information. There are two sections:

Section one: Information about the research and its purpose

Section two: Detailed information about taking part in this research.

## **Section one**

### **Information about the research**

#### **Why I am doing this research?**

Some studies show that skills become difficult in adulthood as age progresses and individuals find it difficult to do daily tasks which were easier in the past. This research aims to investigate the difficulties in daily tasks such as dressing, washing and walking in adulthood between 16 years and 40 years. This will help us to develop better ways for you to manage your daily tasks.

#### **Funding**

There is no funding for the research

#### **Results of the research**

The results of the research will be

- written in a thesis format for an academic programme
- written for publication in physiotherapy related journals
- presented in related conferences

#### **Contact information**

If you need more information or have any questions you can contact

**Ram Krishnamurthy**  
**Specialist Physiotherapist**  
**Community Neurology Team**  
**Leeds Community NHS Foundation Trust**  
**St Mary's Hospital**  
**Green Hill Road**  
**Leeds LS12 4DR**  
**Tel: 0113 3055082**  
**Email: ramkrish@nhs.net**

## **Section two**

### **Your Participation in the Research**

#### **Why will taking part help with this study?**

Taking part in this study will help us to explore and identify better strategies to manage daily tasks during adulthood in individuals with Cerebral Palsy. This may help to maintain your physical and functional skills in adult life.

#### **Do I have to take part?**

It is up to you to decide whether or not to take part. Choosing not to take part will not affect your current or future clinical care in any way.

You can withdraw yourself from the research any time up to which the data is analysed after agreeing to take part if you wish.

#### **What will happen if I take part?**

This research consists of two parts.

##### *Part one*

You will be invited to the clinic to fill in two short questionnaires describing the daily tasks you can do and your general health.

Then, your transfer abilities, sitting balance, standing balance, walking and stair climbing will also be assessed. Following the assessment, you will be given an opportunity to briefly comment on your daily tasks, transfer abilities and walking abilities. The assessment will take around 45 to 60 minutes attendance at the clinic, St James's University Hospital or at St Mary's Hospital, Greenhill Road.

##### *Part two*

Five people from part one of this research will be invited for an interview on another day. This will last for approximately 45 to 60 minutes and will be audio recorded for analysis. If you are one of the five invited you will be asked how you manage your daily tasks and what you would do to maintain these skills. You will also be invited to fill in a questionnaire about your daily coping strategies.

Being part of this research means that you are willing to be involved in both part one and part two of this research. However you can withdraw at any time if you are not comfortable.

Your GP and/or your consultant will be informed about your participation in this research.

#### **Are there any risks involved and what support is available for me?**

The type of assessments used in part one are regularly used in physiotherapy and by other health professionals. The researcher has extensive experience in treating people with Cerebral Palsy, therefore safety will be ensured as much as possible during these assessments.



Part two of the research involves an interview where we will discuss your ability to do daily tasks, you may find this upsetting. During the interview if you feel anxious or upset, the interview will be paused and you will be given time to recover. If you wish the interview will be stopped and the option of referral to appropriate services may be offered.

### **What will happen to my daily task assessment and interview information?**

Your scores from the tests and what you say at the interview will be stored securely in a locked cabinet. Your information will be accessed only by me. If necessary my supervisors will have access to your information but your name will not be attached to it. The information will be analysed using appropriate computer software and the results will be reported and discussed in any written reports for my academic programme. Quotes from your interview may be used in this report. However your identity will be confidential and will not be revealed to anyone.

The data will be kept for a minimum of 5 years in a locked filing cabinet and in a secure room, after which the information will be destroyed.

### **Are there any benefits for me?**

If anything from the assessments of daily tasks come to light or you need further support, you will be referred to the appropriate services, through you GP if necessary. You will not be paid for taking part in this research.

It is expected that this research can highlight some areas that people with CP can self-manage to prevent loss of skills in daily tasks in adulthood. At the end of the research, the results will be shared with you if you wish. Please let the researcher know if you want to know the results of the research.

### **What if I have any concerns or complaints?**

In the first instance, please contact myself if you have any concerns or questions. If you have any serious concerns regarding the research process, you can contact my academic supervisor

Dr Michael Snowden  
Principal Lecturer  
Clinical and Health Sciences  
University of Huddersfield  
Queensgate  
Huddersfield  
HD1 3DH  
Email: [m.a.snowden@hud.ac.uk](mailto:m.a.snowden@hud.ac.uk)

Should you wish to take part in this research or want to discuss further, please do not hesitate to contact me. You can also fill the tear off slip from the enclosed invitation letter and return it to us.

Thank you for considering taking part in this research.

## Appendix 9 : Participant Consent Form

### Participant Consent Form

It is important that you read, understand and sign the consent form. Your contribution to this research is entirely voluntary and you are not obliged in any way to participate, if you require any further details please contact your researcher

**Participant Name:**

**Research ID:**

**DOB:**

**Purpose of the research:** To explore the self-perceived and actual skills in daily tasks in individuals with Cerebral Palsy between the ages of 16 and 40 years with a view to developing self-management strategies.

**Name of the researcher:** Ram Krishnamurthy

1.	I confirm that I have read and understood the information sheet provided for the above research	Yes/No
2.	I have had opportunity to ask questions to clarify any issues	Yes/No
3.	I have considered all the information and wish to take part in part 1 and part 2 of this research	Yes/No
4.	I consent for my interview to be audio recorded for research purpose and I understand that only Ram Krishnamurthy will listen to the recordings and read the interview notes.	Yes/No
5.	I understand that some quotes that I give during the interview may be used in publications, however these will be anonymous	Yes/No
6.	I understand that I have the right to withdraw from the research at any time without giving any reason	Yes/No
7.	I understand that I can withdraw from the research at any point but my data up to that point will be used	Yes/No
8.	I understand that the information collected will be kept in secure conditions for a period of five years	Yes/No
9.	I consent for Ram Krishnamurthy to inform my GP that I am participating in this research	Yes/No
10.	I agree to take part in this research	Yes/No

If you are satisfied that you understand the information and are happy to take part in this research please print and sign below.

#### **Participant**

Print Name

Signature

Date

#### **Researcher**

Print name

Signature

Date

[Appendix 10 : Certificate – Introduction to Good Clinical Practice \(GCP\)](#)



**National Institute for  
Health Research**

Clinical Research Network

# Certificate of Completion

## Ram Krishnamurthy

has completed

### Introduction to Good Clinical Practice (GCP) e-learning course

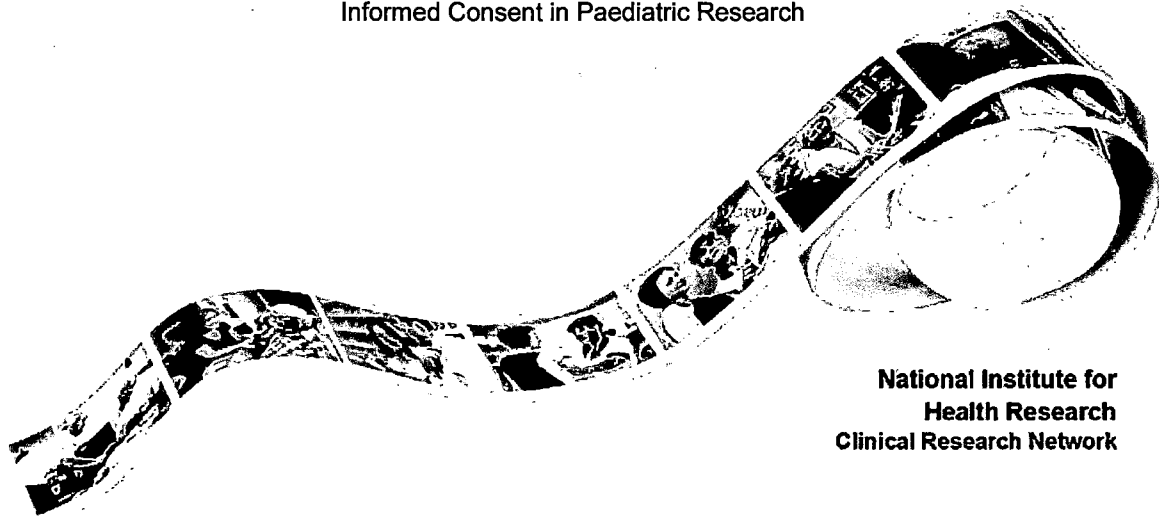
A practical guide to ethical and scientific quality  
standards in clinical research

on 19 July 2015

#### Modules completed

Introduction to Research in the NHS  
Good Clinical Practice and Standards in Research  
Study Set-up and Responsibilities  
The Process of Informed Consent  
Data Collection and Documentation  
Safety Reporting  
Summary

Informed Consent in Paediatric Research



**National Institute for  
Health Research**  
Clinical Research Network

[Appendix 11 : Copy letter to General Practitioner and/or participant consultant](#)

**Private and confidential**

GP/Consultant

Community Neurology Rehabilitation Services  
St Marys Hospital  
Green Hill Road  
Leeds  
LS12 3QE

Date:

Dear

**Re:**

**Address:**

**Date of Birth:**

**NHS Number:**

I am working as a Specialist Physiotherapist with the Community Neurology Team, Leeds Community Healthcare NHS Trust. I am currently pursuing a Professional Doctorate Research Programme from University of Huddersfield.

I am interested in looking at the performance of daily tasks in adults with Cerebral Palsy with a view to developing self-management strategies.

Mr/Ms has agreed to partake in this research and I have enclosed the information sheet which was shared with the participants. I have sent this to you for information.

Please do not hesitate to contact me if you have any concerns.

Yours sincerely

Ram Krishnamurthy  
Specialist Physiotherapist  
Community Neurology Rehabilitation Services

THE UNIVERSITY OF HUDDERSFIELD: RISK ANALYSIS & MANAGEMENT

				Name: Ram Krishnamurthy	
				Date: 01/06/2015	Review Date: Reviewed and updated on 11/05/2016
Hazard(s) Identified	Details of Risk(s)	People at Risk	Risk management measures	Other comments	
Slips, trips and falls during phase 1	The assessments will be completed at the physiotherapy room which is regularly used for assessment and therapy. Therefore there are no visible obstructions or equipment's present in the space where the assessment takes place	Research Participant	Research will ensure that before the assessments area will be visually inspected for any obstructions, flooring and safety of the environment.		
Manual handling		Researcher and research participant	<p>Researcher has completed manual handling course and updates the knowledge as per the practice guidelines</p> <ul style="list-style-type: none"> <li>The researcher is experienced in handling the research participants during routing physiotherapy interventions as part of the job role with NHS.</li> </ul>		

Interviewing at the hospital - Phase II	Privacy	Research Participant	<ul style="list-style-type: none"> <li>• For ensure confidentially the interviews will be held in a private room at the researchers workplace.</li> <li>• This location is regularly used by researcher colleagues during interviews pertaining to therapy interventions.</li> </ul>	
Loss or theft of data	Security of data	Data from Phase I and phase II	<ul style="list-style-type: none"> <li>• Electronic data will be stored in a password protected computer</li> <li>• When data is transported, the storage drive will be password protected.</li> <li>• Audio recording device and all paper reports will be stored secured at the researcher's workplace.</li> <li>• If the data has to be transported between locations, it will be transported in a secure lockable case.</li> </ul>	
Interview	Personal Safety	Researcher	<ul style="list-style-type: none"> <li>• Research colleague or the admin team will be aware about the interview start and approximate finish time.</li> <li>• The relevant colleague will be requested to ensure the safety of the researcher at the appropriate finish time by ringing the researcher at the finish time.</li> </ul>	

21 May 2015

**TO WHOM IT MAY CONCERN**

**Name:** Mr Ramanathan Krishnamurthy - University of Huddersfield  
**Research Project:** "Self perceived and actual physical functional abilities in adults with Cerebral Palsy"  
**Reference:** SREP/2014/016

**THE AWARDS**  
AWARD WINNER  
UNIVERSITY OF THE YEAR

Mr Ramanathan Krishnamurthy, the holder of this letter, is a research student at the University of Huddersfield, where he is currently pursuing a Doctorate of Physiotherapy on the above topic within the Centre for Health and Social Care Research.

Mr Krishnamurthy's research has been through the School Research Ethics Panel (SREP) and his project was approved on 17 March 2014.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.
6. The duties of sponsors set out in the Research Governance Framework for Health and Social Care will be undertaken in relation to this research.
7. I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

If you require any further information in relation to this letter, please do not hesitate to contact me.

Yours faithfully



**Prof Rachel Armitage**  
Chair, School Research Ethics Panel (SREP)  
School of Human and Health Sciences  
Direct Tel: +44 (0)1484 473854  
Email: R.A.Armitage@hud.ac.uk

**Research Title: Self perceived and actual physical functional abilities in adults with Cerebral Palsy**

Self Perceived abilities: What individuals think they can do

Actual abilities: What individuals can actually do

Physical functional abilities: Eating, Drinking, Dressing, Bathing, Showering, Toileting, Walking

Cerebral Palsy: Brain Injury from birth

**Aim:** To explore the self perceived and actual physical functional abilities in individuals with Cerebral Palsy between the ages of 16 and 40 years.

**Background:**

- Individuals receive significant therapy in school under education authority
- When they complete school the responsibility shifts to the individuals in accessing the adult services
- Our experience shows that not many are pro-active unless there is a major concern
- There are some research evidence to say that skills deteriorate in adulthood due to aging factors
- Health services are not geared towards addressing this issue
- Also there is some evidence to say that there are discrepancies between what individuals think they can do and they can actually do
- I am going to explore this and feel that it is essential to know these issues so that services/strategies can be adapted to manage the deterioration in skills during adulthood.

**Access to participants:**

- Individuals who are referred to Community Neurology Services
- Individuals who come to see Dr O'Connor, Consultant in Rehabilitation Medicine at St James Hospital



## Research Design

Identification of participants

Inclusion Criteria: Age 16 to 40 years  
Diagnosis of Cerebral Palsy

Exclusion criteria: Associated health condition like diabetes mellitus, heart disease  
Currently receiving or received treatment from me  
Associated diagnosis of severe or moderate learning disability

Obtain consent

Data Collection (40 individuals) 2 questionnaires filled by the individuals  
2 questionnaires filled by me

Analysis

Identification of participants for phase 2 - interview – minimum of 5 participants

Analysis

## **PARTICIPANT INFORMATION SHEET**

My name is Ram Krishnamurthy and I am working as a Specialist Physiotherapist with the Community Neurology Team, Leeds Community Healthcare NHS Trust. I am interested in looking at the performance of daily tasks in adults with Cerebral Palsy.

**Intention of the study:** To explore the self-perceived and actual skills in daily tasks in individuals with Cerebral Palsy between the ages of 16 and 40 years with a view to developing self-management strategies

You are being invited to take part in this research. Please take time to read the following information about the research. Please don't hesitate to ask any questions if anything is not clear or you would like more information. There are two sections:

Section one: Information about the research and its purpose

Section two: Detailed information about taking part in this research.

### **Section one - Information about the research and its purpose**

#### **Purpose**

Some studies show that skills become difficult in adulthood as age progresses and individuals find it difficult to do daily tasks which were easier in the past. This research aims to investigate the difficulties in daily tasks such as dressing, washing and walking in adulthood. This will help us to develop appropriate ways for you to manage your daily tasks.

#### **Funding**

There is no funding for the research

#### **Results of the research**

The results of the research will be

- written in a thesis format for an academic programme
- written for publication in physiotherapy related journals
- presented in related conferences

#### **Contact information**

If you need more information or have any questions you can contact

**Ram Krishnamurthy**

**Specialist Physiotherapist**

**Community Neurology Team**

**Leeds Community NHS Foundation Trust**

**St Mary's Hospital**

**Green Hill Road**

**Leeds LS12 4DR**

**Tel: 0113 3055082**

**Email: ramkrish@nhs.net**

## **Section two - Your Participation in the Research**

### **Why do I need to take part in this research?**

Your participation is essential to research the daily tasks in individuals with Cerebral Palsy. This may help you and other people with Cerebral Palsy to maintain their skills in daily tasks and prevent or reduce the decline in skills in adult life.

### **Do I have to take part?**

No, it's entirely your decision whether to take part or not.

You can even withdraw yourself from the research after agreeing to take part if you are not comfortable.

### **What will happen if I take part?**

This research consists of two parts.

#### *Part one*

You will be asked to fill in two short questionnaires describing the daily tasks you can do and your general health.

Then, your transfer abilities, sitting balance, standing balance, walking and stair climbing will also be assessed. Following the assessment, you

will be given an opportunity to briefly comment on your daily tasks, transfer abilities and walking abilities. The assessment will take around 45 to 60 minutes attendance at St Mary's Hospital, Greenhill road.

### *Part two*

Five people from part one of this research will be chosen for an interview on another day. This will last for approximately 45 to 60 minutes. If you are one of the five chosen you will be asked how you manage your daily tasks and what you would do to maintain these skills. You will also be invited to fill in questionnaire about your daily coping strategies.

Being part of this research means that you are willing to be involved in both part one and part two of this research. However you can withdraw at any time if you are not comfortable.

Your GP and/or your consultant will be informed about your participation in this research.

### **Are there any risks involved and what support is available for me?**

The type of assessments used in part one are regularly used in physiotherapy and by other health professionals. The researcher has extensive experience in treating people with Cerebral Palsy, therefore safety will be ensured as much as possible during these assessments.

As part two of the research involves interview, the questions asked by the researcher and your thoughts when answering the questions may upset you. During the interview if you feel anxious or upset, the interview will be paused and you will be given time to recover. If you wish the interview will be stopped and the option of referral to appropriate services may be offered.

### **What will happen to my daily task assessment and interview information?**

Your scores from the tests and what you say at the interview will be stored securely in a locked cabinet. Your information will be accessed only by me. If necessary my supervisors will have access to your information but your name will not be attached to it. The information will be analysed using appropriate computer software and the results will be reported and discussed in any written reports for my academic programme. Quotes from your interview may be used in this report.

However your identity will be confidential and will not be revealed to anyone.

The data will be kept for a minimum of 5 years in a locked filing cabinet and in a secure room, after which the information will be destroyed.

### **Are there any benefits for me?**

If anything from the assessments of daily tasks come to light or you need further support, you will be referred to the appropriate services, through you GP if necessary. You will not be paid for taking part in this research, however transport costs will be reimbursed at public transport rates.

It is expected that this research can highlight some areas that people with CP can self-manage to prevent loss of skills in daily tasks in adulthood. At the end of the research, the results will be shared with you if you wish. Please let the researcher know if you want to know the results of the research.

### **What if I have any concerns or complaints?**

In the first instance, please contact myself if you have any concerns or questions. If you have any serious concerns regarding the research process, you can contact my supervisor

Phyl Fletcher-Cook

Senior Lecturer

Division of Health and Rehabilitation

University of Huddersfield

Queensgate

Huddersfield

HD1 3DH

Email: [p.fletcher-cook@hud.ac.uk](mailto:p.fletcher-cook@hud.ac.uk)

Should you wish to take part in this research, you will be given a copy of this information sheet and a copy of your signed consent form.

Thank you for considering taking part in this research.

## Participant Consent Form

It is important that you read, understand and sign the consent form. Your contribution to this research is entirely voluntary and you are not obliged in any way to participate, if you require any further details please contact your researcher

**Participant Name:**

**Research ID:**

**DOB:**

**Purpose of the research:** To explore the self-perceived and actual skills in daily tasks in individuals with Cerebral Palsy between the ages of 16 and 40 years with a view to developing self-management strategies.

**Name of the researcher:** Ram Krishnamurthy

1.	I confirm that I have read and understood the information sheet provided for the above research	Yes/No
2.	I have had opportunity to ask questions to clarify any issues	Yes/No
3.	I have considered all the information and wish to take part in part 1 and part 2 of this research	Yes/No
4.	I understand that some quotes that I give during the interview may be used in publications, however these will be anonymous	Yes/No
5.	I understand that I have the right to withdraw from the research at any time without giving any reason	Yes/No
6.	I understand that I can withdraw from the research at any point but my data up to that point will be used	Yes/No
7.	I understand that the information collected will be kept in secure conditions for a period of five years	Yes/No

If you are satisfied that you understand the information and are happy to take part in this research please print and sign below.

**Participant:**

Signature

Date

**Researcher:**

Signature

Date

## [Appendix 15 : Ethics approval from The NHS Research Ethics Committee \(REC\)](#)



### **Health Research Authority** NRES Committee Yorkshire & The Humber - Leeds East

Room 001  
Jarrow Business Centre  
Rolling Mill Road  
Jarrow  
Tyne and Wear  
NE32 3DT

Telephone: 0191 428 3384

24 August 2015

Mr Ram Krishnamurthy  
Specialist Physiotherapist, Community Neurology Rehabilitation Team  
St Mary's Hospital,  
Green Hill Road, Leeds  
LS12 3QE

Dear Mr Krishnamurthy

**Study title:** Self-perceived and actual changes in physical functional abilities in adults with Cerebral Palsy  
**REC reference:** 15/YH/0346  
**IRAS project ID:** 144818

Thank you for your submission of 21 August 2015. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 20 August 2015

#### **Documents received**

The documents received were as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Participant information sheet (PIS) [Participant Information Sheet]	V4	21 August 2015

#### **Approved documents**

The final list of approved documentation for the study is therefore as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Huddersfield Uni indemnity insurance]	2	04 August 2014
GP/consultant information sheets or letters [GP/Consultant letter]	1	21 May 2014
Interview schedules or topic guides for participants [Interview Guide]	2	24 June 2015
Letter from sponsor [University ethics approval]	2	21 May 2015
Letters of invitation to participant [Participant invitation letter site 1]	2	17 June 2015
Letters of invitation to participant [Participant invitation letter site 2]	1	23 June 2015


Other [SF36v2 Copyright Agreement]		
Other [Covering letter reply for REC provisional]		12 August 2015
Participant consent form [Participant consent form]	3	17 April 2015
Participant information sheet (PIS) [Participant Information Sheet]	V4	21 August 2015
REC Application Form [REC_Form_02072015]		02 July 2015
Research protocol or project proposal [Research protocol]	v	23 June 2015
Summary CV for Chief Investigator (CI) [Summary CV]	2	23 June 2015
Summary CV for supervisor (student research) [Academic Supervisor Summary CV]		08 July 2015
Summary, synopsis or diagram (flowchart) of protocol in non-technical language [Research design flowchart]	1	21 May 2014
Validated questionnaire [Self-reported Barthel Index]		
Validated questionnaire [SF36v2]		
Validated questionnaire [Barthel Index]		
Validated questionnaire [Rivermead mobility index]		
Validated questionnaire [The general self-efficacy scale]		

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

**15/YH/0346**

**Please quote this number on all correspondence**

Yours sincerely



**Sarah Prothero**  
REC Assistant

E-mail: [nrescommittee.yorkandhumber-leedseast@nhs.net](mailto:nrescommittee.yorkandhumber-leedseast@nhs.net)

Copy to: *Dr Dawn Leeming, University of Huddersfield*  
*Linda Dobrzanska, Leeds Community Healthcare NHS Trust*





**Health Research Authority**  
**NRES Committee Yorkshire & The Humber - Leeds East**

Room 001  
Jarrow Business Centre  
Rolling Mill Road  
Jarrow  
Tyne and Wear  
NE32 3DT

Telephone: 0191 428 3384

20 August 2015

Mr Ram Krishnamurthy  
Specialist Physiotherapist, Community Neurology Rehabilitation Team  
St Mary's Hospital,  
Green Hill Road, Leeds  
LS12 3QE

Dear Mr Krishnamurthy

**Study title:** Self-perceived and actual changes in physical functional abilities in adults with Cerebral Palsy  
**REC reference:** 15/YH/0346  
**IRAS project ID:** 144818

Thank you for your letter of 12 August 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager, Katy Cassidy, [nrescommittee.yorkandhumber-leedseast@nhs.net](mailto:nrescommittee.yorkandhumber-leedseast@nhs.net). Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

### **Confirmation of ethical opinion**

On behalf of the Committee, I am pleased to confirm a **favourable** ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

### **Conditions of the favourable opinion**

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Additional conditions as specified by the Committee:

1. Change heading in participant information sheet from 'Why do I have to take part?' to 'Why will taking part help with this study'.
2. Submit participant information sheet on sponsor headed paper.

**You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.**

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

*Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.*

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

*Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.*

*For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.*

*Sponsors are not required to notify the Committee of approvals from host organisations*

### Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net). The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from NRES. Guidance on where to register is provided on the HRA website.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

### **Ethical review of research sites**

#### NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

#### Non-NHS sites

### **Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Huddersfield Uni indemnity insurance]	2	04 August 2014
GP/consultant information sheets or letters [GP/Consultant letter]	1	21 May 2014
Interview schedules or topic guides for participants [Interview Guide]	2	24 June 2015
Letter from sponsor [University ethics approval]	2	21 May 2015
Letters of invitation to participant [Participant invitation letter site 1]	2	17 June 2015
Letters of invitation to participant [Participant invitation letter site 2]	1	23 June 2015
Other [SF36v2 Copyright Agreement]		
Other [Covering letter reply for REC provisional]		12 August 2015
Participant consent form [Participant consent form]	3	17 April 2015
Participant information sheet (PIS) [Participant information Sheet V3]	V3	12 August 2015
REC Application Form [REC_Form_02072015]		02 July 2015
Research protocol or project proposal [Research protocol]	v	23 June 2015
Summary CV for Chief Investigator (CI) [Summary CV]	2	23 June 2015
Summary CV for supervisor (student research) [Academic Supervisor Summary CV]		08 July 2015
Summary, synopsis or diagram (flowchart) of protocol in non-technical language [Research design flowchart]	1	21 May 2014
Validated questionnaire [Self-reported Barthel Index]		
Validated questionnaire [SF36v2]		
Validated questionnaire [Barthel Index]		
Validated questionnaire [Rivermead mobility index]		
Validated questionnaire [The general self-efficacy scale]		

### **Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

## After ethical review

### Reporting requirements

The attached document “*After ethical review – guidance for researchers*” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

### User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

### HRA Training

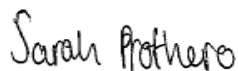
We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

**15/YH/0346**

**Please quote this number on all correspondence**

With the Committee’s best wishes for the success of this project.

Yours sincerely  
pp



**Dr Rhona Bratt  
Chair**

Email: [nrescommittee.yorkandhumber-leedseast@nhs.net](mailto:nrescommittee.yorkandhumber-leedseast@nhs.net)

*Enclosures:* “After ethical review – guidance for researchers”

*Copy to:* *Dr Dawn Leeming, University of Huddersfield  
Linda Dobrzanska, Leeds Community Healthcare NHS Trust*

# The Leeds Teaching Hospitals

NHS Trust

Irfan Jina

27/08/2015

Research & Innovation

**Leeds Teaching Hospitals NHS Trust**

34 Hyde Terrace

Leeds

LS2 9LN

Dr Rory J O'Connor  
Associate Clinical Professor in Rehabilitation Medicine  
Academic Department Rehabilitation Medicine  
Level D, Martin Wing  
Leeds General Infirmary  
Leeds  
LS1 3EX

Tel: 0113 392 0162

Fax: 0113 392 0146

r&d@leedsth.nhs.uk

www.leedsth.nhs.uk

Dear Dr Rory J O'Connor

**Re: NHS Permission at LTHT for: Self-perceived and actual changes in physical functional abilities in adults with Cerebral Palsy**  
**LTHT R&I Number: NE15/114**  
**REC: 15/YH/0346**

I confirm that *NHS Permission for research* has been granted for this project at The Leeds Teaching Hospitals NHS Trust (LTHT). NHS Permission is granted based on the information provided in the documents listed below. All amendments (including changes to the research team) must be submitted in accordance with guidance in IRAS. Any change to the status of the project must be notified to the R&I Department.

The study must be conducted in accordance with the *Research Governance Framework for Health and Social Care*, ICH GCP (if applicable), the terms of the Research Ethics Committee favourable opinion (if applicable) and NHS Trust policies and procedures (see <http://www.leedsth.nhs.uk/research/>) including the requirements for research governance and clinical trials performance management listed in appendix 1 and 2. NHS permission may be withdrawn if the above criteria are not met including the requirements for clinical trials performance

The Leeds Teaching Hospitals NHS Trust participates in the NHS risk pooling scheme administered by the NHS Litigation Authority "Clinical Negligence Scheme for NHS Trusts" for: (i) medical professional and/or medical malpractice liability; and (ii) general liability. NHS Indemnity for negligent harm is extended to researchers with an employment contract (substantive or honorary) with the Trust. The Trust only accepts liability for research activity with NHS Permission

The Trust therefore accepts liability for the above research project and extends indemnity for negligent harm. Should there be any changes to the research team please ensure that you inform the R&I Department and that s/he obtains an appropriate contract, or letter of access, with the Trust if required.

Yours sincerely



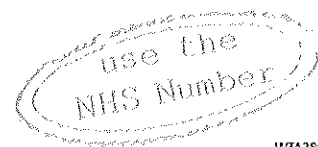
**Anne Gowing**  
**Research Governance Manager**

Chair Dr Linda Pollard CBE JP DL Chief Executive Julian Hartley

The Leeds Teaching Hospitals incorporating:

Chapel Allerton Hospital Leeds Dental Institute Seacroft Hospital Leeds Children's Hospital

St James's University Hospital Leeds General Infirmary Wharfedale Hospital Leeds Cancer Centre



## Approved documents

The documents reviewed and approved are listed as follows:-

<i>Document</i>	<i>Version</i>	<i>Date of document</i>
NHS R&D Form	4.0.0	01/07/2015
SSI Form	4.0.0	06/08/2015
CSU Approval		11/08/2015
Finance Approval		27/07/2015
REC Letter confirming favourable opinion		20/08/2015 and 24/08/2015
Evidence of Insurance		04/08/2014
GP Letter	1	21/05/2014
Interview Guide	2	24/06/2015
Letter from Sponsor		21/05/2015
Letter of Invitation (Site 1)	2	17/06/2015
Letter of Invitation (Site 2)	1	23/06/2015
Consent Form	3	17/04/2015
PIS	4	21/08/2015
Protocol	V2	23/06/2015
Flowchart	1	21/05/2014
Self-reported Barthel Index		
SF36v2		
Barthel Index		
Rivermead mobility index		
The general self-efficacy scale		

## Conditions of NHS Permission for Research:

### *Appendix 1*

#### **Governance requirements:**

Managerial approval within the Clinical Support Unit must be obtained before starting the study and healthcare staff should be suitably informed about the research their patients are taking part in and information specifically relevant to their care arising from the study should be communicated promptly.

Agreements must be in place with appropriate support departments.

Arrangements must be in place for the management of financial and other resources provided for the study, including intellectual property arising from the research.

All data and documentation associated with the study must be available for audit/monitoring by authorised Trust or external agencies.

All members of the research team, where applicable, have appropriate employment contracts or letter of agreement to carry out their work in the Trust.

Each member of the research team must be qualified by education, training and experience to discharge his/her role in the study. Students and new researchers must have adequate supervision, support and training.

The research must follow the protocol approved by the relevant research ethics committee. Any proposed amendments to or deviations from the protocol must be submitted for review (as necessary) by the Research Ethics Committee, the Research Sponsor, regulatory authority and any other appropriate body. Where the amendment has resource implications within the CSU, the Directorate research lead/clinical director and R&I should be notified.

Adverse Events in clinical trials of investigational medicinal products must be reported in accordance with the Medicines for Human Use (Clinical Trials) Regulations 2004.

Procedures should be in place to ensure collection of high quality, accurate data and the integrity and confidentiality of data during processing and storage in line with Trust Information Governance Policies and arrangements must be made for the appropriate archiving of data when the research has finished. Records must normally be kept for 15 years.

In compliance with the Health Research Authority (HRA) regulations, clinical trials (and other studies falling within the HRA definition) must be registered on a publically accessible database (such as <https://clinicaltrials.gov/> ) prior to commencement. Studies sponsored by LTHT will be registered by the R&I Department.

Findings from the study should be exposed to critical review through accepted scientific and professional channels.

All members of the research team involved in seeking informed consent adheres to GCP standards. Investigators are directed to the R&I website for further information about training in consent for clinical trials.

Studies involving the use of human tissues must be performed in compliance with the code of practice of the Human Tissue Authority.

If you are not able to comply with these requirements, NHS permission to conduct the research in LTHT will be suspended.

## **Appendix 2**

### **Commercially Sponsored and funded studies.**

In line with Trust Standing Financial Instructions there must be a research agreement with the commercial funder signed by the R&I Department (on behalf of the Leeds Teaching Hospitals NHS Trust). Investigators do not have the authority to sign research agreements on behalf of the Trust.

**NHS permission for this project to be carried out in the Trust is granted on the understanding that you:**

Provide recruitment information when requested by R&I on the Clinical Trial Tracker (available on the CSU Research Hub)

Work with R&I to resolve blocks and delays on trials to ensure that LTHT meets the NIHR benchmarks.

### **NIHR Benchmarks for Performance in Initiating & Delivering Clinical Research**

LTHT clinical trial performance is measured against 2 national benchmarks to improve the initiation and delivery of clinical trials approved by the Trust. NIHR funding to the Trust is conditional on meeting these benchmarks.

**Initiation** – it should take no more than 70 days from receipt of a valid research application (signed SSI form) by the R&I Department to the recruitment of (ie consenting) the 1st patient to the trial

**Delivery** – for all trials hosted by the Trust the agreed number of patients must be recruited within the agreed recruitment period

The Trust submits quarterly performance reports to the Department of Health setting out our performance.

For more information about the benchmarks and the work we are doing to support clinical trial management please see the R&I website.

<http://www.leedsth.nhs.uk/research/>



## RECRUITMENT SERVICE

Ground Floor, Trust Headquarters, St James's Hospital, Beckett Street,  
Leeds, LS9 7TF

**Private and Confidential**

Ram Krishnamurthy  
5 Tavistock Way  
Leeds,  
LS12 4DR

Enquiries to: **Sam Beeby**  
Direct Line: (0113) 2065980  
Our Ref: **LOA**  
Date: 4th September 2015

Dear **Ram**,

**Letter of Access to research self-perceived and actual changes in physical functional abilities in adults with Cerebral Palsy**

This letter confirms your right of access to conduct research through Leeds Teaching Hospitals NHS Trust for the purpose and on the terms and conditions set out below. This right of access commences in September 2015 and ends in May 2016 unless terminated earlier in accordance with the clauses below.

As an existing NHS employee you do not require an additional honorary research contract with this NHS organisation. We are satisfied that the research activities that you will undertake in this NHS organisation are commensurate with the activities you undertake for your employer. Your employer is fully responsible for ensuring such checks as are necessary have been carried out. Your employer has confirmed in writing to this NHS organisation that the necessary pre-engagement check are in place in accordance with the role you plan to carry out in this organisation.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal Investigator for the research project has received a letter from us giving permission to conduct the project.

You are considered to be a legal visitor to Leeds Teaching Hospitals NHS Trust premises. You are not entitled to any form of payment or access to other benefits provided by this organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

While undertaking research through Leeds Teaching Hospitals NHS Trust, you will remain accountable to your current NHS employer but you are required to follow the reasonable instructions of your nominated manager **Dr Rory O'Connor** in this NHS organisation or those given on her/his behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with Leeds Teaching Hospitals NHS Trust policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with Leeds Teaching Hospitals NHS Trust in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on Leeds Teaching Hospitals NHS Trust premises. Although you are not a contract holder, you must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of a contract holder and you must act appropriately, responsibly and professionally at all times.

If you have a physical or mental health condition or disability which may affect your research role and which might require special adjustments to your role, if you have not already done so, you must notify your employer and this NHS organisation prior to commencing your research role at each site.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice (<http://www.dh.gov.uk/assetRoot/04/06/92/54/04069254.pdf>) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

Leeds Teaching Hospitals NHS Trust will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

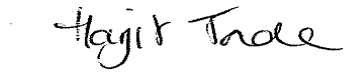
This letter may be revoked and your right to attend the organisation(s) terminated at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of the organisation(s) or if you are convicted of any criminal offence.

You must not undertake regulated activity if you are barred from such work. If you are barred from working with adults or children this letter of access is immediately terminated. Your employer will immediately withdraw you from undertaking this or any other regulated activity and you **MUST** stop undertaking any regulated activity immediately.

Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

If your circumstances change in relation to your health, criminal record, professional registration or suitability to work with adults or children, or any other aspect that may impact on your suitability to conduct research, or your role in research changes, you must inform the NHS organisation that employs you through its normal procedures. You must also inform the nominated manager in this NHS organisation.

Yours sincerely

A handwritten signature in black ink that reads "Harjit Tanda". The signature is written in a cursive style with a small dot above the 'i' in "Tanda".

**Harjit Tanda**  
**Head of HR.**

cc: R&D office at Leeds Teaching Hospitals NHS Trust  
HR department of the substantive employer (and provider of honorary clinical contract, where applicable)



Telephone enquiries, please contact:

Phone: 0113 203 3473 / 07958066828

Email: [lch.research@nhs.net](mailto:lch.research@nhs.net)

24 August 2015

Mr Ram Krishnamurthy  
Specialist Physiotherapist  
Community Neurology Rehabilitation team  
St. Mary's Hospital  
Green Hill Road  
Leeds LS12 3QE



Stockdale House  
Headingley Office Park  
Victoria Road  
Leeds LS6 1PF

LCH Ref: NP/0153

Dear Mr Krishnamurthy

**Re: Self-perceived and actual changes in physical functional abilities in adults with Cerebral Palsy**

Thank you for your recent submission to Leeds Community Healthcare (LCH) NHS Trust requesting NHS Research Governance approval for the above study.

Following consideration of your submission I am pleased to confirm that NHS Research Governance approval has been granted by LCH for the above research to take place as described in your application and accompanying documentation.

**Conditions of approval**

You should be aware that approval is granted subject to the conditions specified below:

- In undertaking this research you must comply with the requirements of the Research Governance Framework for Health and Social Care (2<sup>nd</sup> edition 2005) which is mandatory for all NHS employees
- Consent for LCH to audit your project
- Where any amendments, substantial or non substantial are made throughout the course of the study these should be notified to LCH
- Upon completion of the study you fill in a completed study outcome form
- Should any serious adverse event(s) occur throughout the course of the study these should be notified to LCH using the contact details set out above
- For LCH employees, you must make yourself aware of and follow any policies relevant to your research



- You should recruit the first participant into the study within 30 days of the date of this approval letter and inform the research team
- Upon request you provide details of recruitment at LCH to the research team

Should you require any clarification regarding any of the points raised above, or have any further queries please contact the research team on the numbers above.

Finally, I would like to take this opportunity to wish you well with your study. I look forward to hearing about your progress in due course.

Yours sincerely

**Dr. Amanda Thomas**  
**Executive Medical Director**

**Approved documents**

The documents reviewed and approved are listed below

<i>Document</i>	<i>Version</i>	<i>Date of Document</i>
REC approval	15/YH/0346	24 August 2015
R & D Form	144818/827326/14/976	1 July 2015
SSI Form	144818/827319/6/51/228733/329203	4 August 2015
Protocol	2	23 June 2015
Participant Information Sheet	4	21 August 2015
Letter of invitation to participant (Participant invitation letter site 1)	2	17 June 2015
Letter of invitation to participant (Participant invitation letter site 2)	1	23 June 2015
GP/Consultant information sheets or letters	1	21 May 2015
Interview schedule	2	24 June 2015
Participant Consent Form	3	17 April 2015
Validated questionnaires		checked
Sponsor identified	University of Huddersfield	
Indemnity identified: Management Design Conduct	University of Huddersfield Both NHS	

Cc:  
Jane Savage  
Clinical pathway lead for CNS: [jane.savage@nhs.net](mailto:jane.savage@nhs.net)

Chair: Neil Franklin OBE

Chief Executive: Thea Stein

**Appendix 19 : License agreement from the Office of Grants and Scholarly Research of OptumInsight Life Sciences, Inc.**



**NON-COMMERCIAL LICENSE AGREEMENT  
Office of Grants and Scholarly Research (OGSR)**

**License Number:** QM020635

**Effective Date:** 1 January 2014

**Licensee Name:** Ram Krishnamurthy, c/o University of Huddersfield

**Licensee Address:** 5 Tavistock Way, Leeds, West Yorkshire LS12 4DR, UK

**Approved Purpose:** Non-commercial academic research and/or thesis – Unfunded Student License Program.

**Study Name:** Self Perceived and actual changes in physical functional abilities in adults with Cerebral Palsy

**Study Type:** FOR DOCTORAL RESEARCH ONLY

**Therapeutic Area:** Brain and Nervous System

**Royalty Fee:** None, because this License is granted in support of the non-commercial Approved Purpose

**Other Definitions:** As indicated on Appendix B "License Agreement – Details", including without limitation: Licensed Surveys, Modes, Fees, Administrations, Services, Approved Languages and (if applicable) License Term

Licensee accepts and agrees to the terms of this Non-Commercial License Agreement (the "Agreement") from the Office of Grants and Scholarly Research (OGSR) of OptumInsight Life Sciences, Inc. (f/k/a QualityMetric Incorporated) ("OptumInsight") as of the Effective Date.

Subject to the terms of this Agreement, including the OptumInsight Non-Commercial License Terms and Conditions attached as Appendix A: OptumInsight grants to Licensee, and Licensee accepts, a non-exclusive, non-transferable, non-assignable, non-sublicensable worldwide license to use, solely for the Approved Purpose and during the License Term, the Licensed Surveys in the authorized Modes and Approved Languages indicated on Appendix B and to administer the Licensed Surveys only up to the approved number of Administrations (and to make up to such number of exact reproductions of the Licensed Surveys necessary to support such Administrations) in any combination of the specific Licensed Surveys and Approved Languages and Modes and to use any related software provided by OptumInsight.

Capitalized terms used in this Agreement shall have the meanings assigned to them above, or in Appendices A and B attached hereto. Appendices A and B attached hereto are incorporated into and made a part of this Agreement for all purposes.

EXECUTED, as of the Effective Date, by the duly authorized representatives as set forth below.

**OptumInsight Life Sciences, Inc.**  
[OptumInsight]

Signature: \_\_\_\_\_

Name: Martha Bayliss  
VP Operations

Title: \_\_\_\_\_

Date: 15 October, 2013

**Ram Krishnamurthy**  
[Licensee]

Signature: K. Ramurthy

Name: RAM KRISHNAMURTHY

Title: MIR

Date: 14.10.2013



an OptumInsight company

APPENDIX A

OPTUMINSIGHT NON-COMMERCIAL LICENSE TERMS AND CONDITIONS

Attached to and incorporated into License No. QM020635

1. **No Commercial Use of Data** - Licensee agrees to use the data resulting from Licensee's administration of the Licensed Surveys only in connection with the Approved Purpose set forth on Page 1 of this Agreement. Licensee agrees that it shall not use such data for any other purpose, or provide such data to any commercial or non-commercial entity (including without limitation any university or university hospital) for any commercial purpose.
2. **License Term and Termination** - This Agreement shall be effective until end of the License Term reflected in Appendix B, after which this Agreement and the licenses granted here under shall terminate. Notwithstanding the foregoing, OptumInsight may terminate this Agreement at any time upon in the event of a material breach by Licensee or its personnel of this Agreement that is not cured within thirty (30) days following notice to Licensee.
3. **Administration by Third Parties** - A third party service provider may administer the Licensed Surveys on behalf of Licensee, subject to such third party's execution of OptumInsight's Acknowledgement By Agent form, available by request. However, in all such cases, Licensee shall not be relieved of its obligations, and Licensee shall be responsible for any breach hereof by such student or third party.
4. **Trademark and Copyright Notices** - Licensee agrees to reproduce the copyright and trademark notices included with the Licensed Surveys on all reproductions of the Licensed Surveys permitted hereunder, including electronic reproductions and representations. Licensee shall not alter the wording or order of the items or any other part of the Licensed Surveys. Licensee shall not create any derivative work from the Survey Materials (as defined in Section 7(a)).
5. **Records** - Licensee shall maintain accurate records, in all material respects, containing information sufficient to verify Licensee's compliance with this Agreement, including as applicable, but not limited to, records of the number of reproductions of the Licensed Survey(s) made, the location of and/or confirmation of the destruction of such reproductions, the number of administrations of the Licensed Surveys performed, and the use(s) made of the data resulting from Licensee's administration of the Licensed Surveys.
6. **Proprietary Rights** -
  - a. Licensee acknowledges that the Licensed Surveys, copyright in all publications purchased, Software, and all intellectual property rights related thereto ("Survey Materials"), shall be and remain at all times the property of OptumInsight. Licensee shall have no right, title or interest in the Survey Materials except for the limited license described herein. Licensee shall not use, modify, reproduce, or transmit any of the Survey Materials except as expressly provided hereunder in connection with the Approved Purpose. If the Approved Purpose includes administration of the Licensed Surveys in physical form, Licensee is authorized to make exact reproductions of the Licensed Survey(s) sufficient to support such administrations.
  - b. Licensee acknowledges and agrees that the Mode of survey administration reflected in Appendix B is the only manner in which Licensee may administer the Licensed Surveys. By way of example, if the Mode reflected in Appendix B is "paper", the Licensed Survey(s) shall be used, distributed, and administered via paper only, and shall not be used, distributed, displayed, or administered via any electronic means.
  - c. Licensee acknowledges and agrees that any translations of the Licensed Surveys into any language must be performed by OptumInsight, and OptumInsight retains ownership of any and all translations.
7. **Ownership of Survey Results Data** - Notwithstanding the foregoing, the parties agree that all results of Licensee's administration of the Licensed Survey(s) shall be the property of Licensee.
8. **Confidentiality; Injunctive Relief** - Licensee acknowledges that the Survey Materials are valuable assets of OptumInsight and that the value of the Survey Materials would be significantly impaired by the unauthorized



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distribution or use of them. Licensee shall ensure that the Survey Materials are not used for unauthorized purposes or by unauthorized persons, and shall promptly report any such unauthorized use to OptumInsight. Licensee acknowledges that, in the event of any material breach of this paragraph by the Licensee, money damages would not be a sufficient remedy, and that OptumInsight shall, to the extent permitted by applicable law, be entitled to equitable relief, including injunction. Such relief shall be in addition to all other remedies available at law or in equity.

9. Disclaimer of Warranty – Licensee understands and acknowledges that complex and sophisticated products such as the Survey Materials are inherently subject to undiscovered defects. OptumInsight cannot and does not represent or warrant to Licensee that the Survey Materials are free from such defects, that operation of the Survey Materials will be uninterrupted or error free, or that its results will be effective or suitable with respect to any particular application. SURVEY MATERIALS PROVIDED HEREUNDER ARE PROVIDED AS-IS, AND OPTUMINSIGHT MAKES NO REPRESENTATIONS OR WARRANTIES, EXPRESS OR IMPLIED, ARISING BY LAW OR OTHERWISE WITH RESPECT TO SUCH SURVEY MATERIALS OR THIS AGREEMENT, AND DISCLAIMS ALL WARRANTIES INCLUDING WITHOUT LIMITATION ANY REPRESENTATIONS OR WARRANTIES AS TO MERCHANTABILITY, FITNESS FOR A PARTICULAR PURPOSE, NON-INFRINGEMENT OR OTHERWISE.

10. Compliance with Law – OptumInsight and Licensee agree that in performing their respective obligations under this Agreement, each shall conduct business in conformance with sound ethical standards of integrity and honesty and in compliance with all applicable laws, rules and regulations.

11. LIMITATION OF LIABILITY – IN NO EVENT SHALL OPTUMINSIGHT BE LIABLE TO LICENSEE OR ANY THIRD PARTY FOR ANY SPECIAL, PUNITIVE, INCIDENTAL, INDIRECT, OR CONSEQUENTIAL DAMAGES, ARISING FROM ANY CLAIMED BREACH OF WARRANTY, BREACH OF CONTRACT, NEGLIGENCE, STRICT LIABILITY IN TORT, OR ANY OTHER LEGAL THEORY, EVEN IF OPTUMINSIGHT HAS BEEN ADVISED OF THE POSSIBILITY OF SUCH DAMAGES. SUCH EXCLUDED DAMAGES INCLUDE, BUT ARE NOT LIMITED TO, LOST PROFITS, COST OF ANY SUBSTITUTE GOODS OR SERVICES, LOST BUSINESS INFORMATION AND DATA, AND BUSINESS INTERRUPTION.

12. Additional Terms for Use of OptumInsight Software – The following additional terms apply to any software provided by OptumInsight to Licensee in connection with this Agreement (“Software”). Licensee may install and use one copy of the Software on a single computer, and except for making one back-up copy of the Software, may not otherwise copy the Software. The Software may not be shared or used concurrently on different computers. Licensee may not reverse engineer, decompile, or disassemble the Software, nor attempt in any other manner to obtain the source code. The Software and the algorithms it contains are proprietary information of OptumInsight. Licensee shall not attempt to circumvent any function of the Software that limits its use to a certain number of administrations of the Licensed Surveys or to a certain time period. Licensee may not rent or lease the Software to any other person.

13. Form Review – If Licensee will administer the Licensed Surveys on an electronic device Licensee acknowledges that this provision shall be applicable to Licensee’s use of the Licensed Surveys. Licensee is required to submit screen shots or a link to the Licensed Surveys for each Approved Language to OptumInsight. OptumInsight shall perform an initial form review to determine whether the Licensed Surveys have been appropriately migrated to electronic format (the “Initial Review”). OptumInsight will complete its Initial Review of the Licensed Surveys for each Approved Language within two (2) weeks from OptumInsight’s receipt of screen shots or website link from Licensee. Upon OptumInsight’s completion of the Initial Review, OptumInsight will provide Licensee with a detailed list of revisions that will need to be made before OptumInsight can approve the electronic format. Licensee is required to submit subsequent screen shots or a link to the Licensed Surveys for each Approved Language incorporating any changes required by OptumInsight until OptumInsight provides its final approval of the electronic format. The parties acknowledge and agree that multiple rounds of review and revisions may be necessary prior to OptumInsight being able to provide final approval of the electronic format. Licensee is solely responsible for the electronic creation of the Licensed Surveys. Licensee does not obtain any rights in the Licensed Surveys not otherwise granted in this Agreement due to the administration and/or use of the Licensed Surveys. Nothing in this Agreement prohibits OptumInsight from creating its own electronic forms of Licensed Survey administration. The Licensed Surveys cannot be used in electronic format except as allowed pursuant to the terms and conditions of this Agreement. Licensee acknowledges that there may be response differences due to effects from use of electronic



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format compared to a static Mode of administration such as paper/pencil. Licensee assumes any and all risk of differential effects resulting from the use of electronic format.

14. Miscellaneous –

a. This Agreement constitutes the entire and exclusive agreement between the parties and supersedes all previous communications or agreements, either oral or written, with respect to the subject matter hereof. This Agreement may not be modified or amended except by an instrument in writing signed by both parties. The Appendices attached hereto are incorporated into and made a part of this Agreement for all purposes.

b. Notices, copies of notices or other communications shall be sent to a party at the address set forth on the first page of this Agreement. All notices shall be effective upon delivery of the notice at such address.

c. Any waiver of any breach or default under this Agreement must be in writing and shall not be deemed a waiver of any other or subsequent breach or waiver. Failure to delay by either party to enforce compliance with any term or condition of this Agreement shall not constitute a waiver of such term or condition.

d. If any provision in this Agreement is determined to be invalid or unenforceable, the remaining provisions of this Agreement shall not be affected thereby and shall be binding upon the parties hereto, and shall be enforceable, as though the invalid or unenforceable provision were not contained herein.

e. In the event a Licensed Survey or associated OptumInsight intellectual property is exported by Licensee outside of the country in which Licensee is located, both parties agree that Licensee is obligated and solely responsible for ensuring compliance with all applicable import and export laws and regulations of the United States of America and/or any applicable foreign jurisdictions. Licensee shall indemnify, defend and hold harmless OptumInsight (including payment of all reasonable costs, fees, settlements and damages) with respect to any suits or proceedings brought against OptumInsight arising from Licensee's export of a Licensed Survey.

f. This Agreement and performance hereunder shall be governed in accordance with the laws of the State of New York, but excluding New York choice of law principles. With respect to any dispute arising in connection with this Agreement, Licensee consents to the exclusive jurisdiction and venue in the state and federal courts located in New York City, New York.

g. This Agreement may be executed in multiple counterparts, each of which shall be deemed an original and all of which shall be deemed the same agreement



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**Very Important - Please Read**

**NO formatting or editing changes to the survey**

In order to obtain licensing from Licensor no changes can be made to the survey forms. Any format and/or language changes have the potential to affect the survey data received. Therefore, to maintain the validation and integrity of the SF Health Surveys, **no language or formatting changes** allowed. The format of the survey is scientifically engineered to facilitate accurate and unbiased data, as well as keeping the SF Health Survey in a visual format that is comprehensible to the patient/participant, including those who may be impaired and/or elderly. **You should administer the survey in the exact format you will receive it in. The only item Licensee may add is a header with patient identification and / or administration information. If you do wish to add a header please ask for a sample copy of the survey to edit and then submit this to your Account Representative for review prior to signing this License Agreement.** Once the licensing process is completed, you will receive a clean set of Survey Forms in a word and .pdf. file. These are the forms you will administer. **Please do not use any forms you may already have access to as the ones we send you are the most current versions.**

\* \* \*



*AM*

APPENDIX B



LICENSE AGREEMENT - DETAILS

Licensee: University of Huddersfield  
 Ram Krishnamurthy  
 5 Tavistock Way,  
 Leeds, LS12 4DR  
 United Kingdom

License Number: QM020635

Amendment to: N/A

License Term: 01/01/14 to 12/31/14

Master License  
 Term: N/A

Approved Purpose  
 Self Perceived and actual changes in physical  
 functional abilities in adults with Cerebral Palsy

Study Name:  
 Protocol:  
 Govt. ID: UNFUNDED STUDENT  
 Study Type: FOR DOCTORAL RESEARCH ON  
 Clients Reference:

Licensed Surveys (Modes) and Services:

Item	Description	Mode of Admin	Quantity	Fees
PROJ01	License Fee	Paper	1	
ES0220	SF-36v2, Standard Recall	Paper	1	

Approved Languages:

Item	Description	Quantity
United Kingdom (English)		
ADM012	Patients Enrolled	25
ADMINS	Administrations (25 @ 1)	25
SS075	Scoring Software v4.5	1
SS080	SS v4.5 Key: SF-36v2 Scoring Key	25
SS997	MSE: Missing Item Estimator	25
EM020	SF-36v2 Clinical Trial Guide	1

Approved Languages:

United States (English)  
 \*\*QUALIFIED UNDER OGSR UNFUNDED STUDENT

LICENSE PROGRAM.

ALL FEES WAIVED.

\*\*EXPIRATION NOTICE:

PLEASE SIGN AND RETURN LICENSE  
 AGREEMENTS

*MK*

BEFORE 26 SEPT 2013.

**TOTAL FEES:**                    **0.00 USD**

**Payment Terms:**   **Due on Receipt**

*Mc*



# NON-COMMERCIAL AMENDMENT TO LICENSE AGREEMENT

**Amendment Effective Date:** March 17, 2015

**Amendment Number:** QM028989

**License Agreement Amended:** QM020635

**Licensee Name:** Ram Krishnamurthy c/o University of Huddersfield  
**Licensee Address:** 5 Tavistock Way, Leeds, LS12 4DR GB

**Approved Purpose:** Self Perceived and actual changes in physical functional abilities in adults with Cerebral Palsy

**Study Name:** Student Doctoral Research

**Study Type:** Non-commercial academic research and/or thesis: Unfunded Student

**Therapeutic Area:** Brain and Nervous System

**Indication:** Cerebral Palsy

OptumInsight Life Sciences, Inc. (f/k/a QualityMetric Incorporated) ("Optum") and Licensee entered into the Non-Commercial License Agreement referenced above (the "License Agreement"). The parties have mutually agreed to enter into this Amendment to the License Agreement ("Amendment") as of the Amendment Effective Date. All capitalized terms used herein and not otherwise defined shall have the meanings ascribed to them in the License Agreement.

The following term(s) of the License Agreement are modified as indicated below:

**Additional Administrations:** 15

**Additional Term:** Beginning on 1-May 2015 and ending on 30-April 2016

**Additional Scoring Items:** Scoring key (v4.5) for SF36v2 with 15 credits

Except as expressly modified by this Amendment, all terms and conditions of the License Agreement shall continue in full force and effect without change.

EXECUTED, as of the Amendment Effective Date, by the duly authorized representatives as set forth below.

**OptumInsight Life Sciences, Inc.**

**Ram Krishnamurthy**

[Licensee]

Signature:

Signature:

Name: Gus Gardner

Name: RAM KRISHNAMURTHY

Title: President and COO

Title: MR

Date: 18 MAR 2015

Date: 18/3/15

**APPENDIX B**



**LICENSE AGREEMENT - DETAILS**

Licensee: University of Huddersfield  
 Ram Krishnamurthy  
 5 Tavistock Way,  
 Leeds, LS12 4DR  
 United Kingdom

License Number: QM028989  
 Amendment to: QM020635  
 Study Term: 05/01/15 to 04/30/16  
 Master License Term: N/A

Approved Purpose  
 Self Perceived and actual changes in physical functional abilities in adults with Cerebral Palsy

Study Name: Student Doctoral Research  
 Protocol: AMD.01 - RENEW/ADD  
 Govt. ID:  
 Study Type: UNFUNDED STUDENT  
 Clients Reference: Brain and Nervous System

**Licensed Surveys (Modes) and Services:**

Item	Description	Mode of Admin	Quantity	Fees
	RENEWAL OF LICENSE QM020635.			
MISC02	Contract Amendment to Renew		1	
ADM012	Additonal Patients Enrolled		15	
ADMINS	Administrations @1 each		15	
SS076	Scoring Software v4.5 Renewal		1	
SS080	SS v4.5 Key: SF-36v2 Scoring		15	
SS997	MSE: Maximum Data Recovery		15	

QUALIFIED UNDER OGSR STUDENT  
 LICENSE PROGRAM. ALL FEES WAIVED.

TO ACCEPT OFFER, PLEASE SIGN AND  
 RETURN LICENSE AMENDMENT BEFORE  
 17-APRIL 2015.

**TOTAL FEES:** 0.00 USD

**Payment Terms:** Due on Receipt



**POSTGRADUATE RESEARCH  
SUPERVISION SESSION**

Date: 24<sup>th</sup> September 2013; 8:45-9:45am  
Present: Mike Snowden, Ram

**DISCUSSION**

- **Form 6** – complete, all ok. Ram to email the final version to Mike and submit it to Kirsty. Mike to identify reviewers.
- **Access to participants:** All research participants should not have met Ram for Physio. (Inclusion criteria) prior to research. Consultant to provide with the information sheet and if the participants are happy, either they can contact Ram or else give consent for Ram to contact the participants. This will also prevent coercion. Then Ram to establish initial contact to explain about the research and obtain consent to participate in the research. Assessments may follow in the same session or else another meeting will be arranged.
- **Sample size:** Ram to obtain rough estimate of clients reviewed by the Consultant and number of CP on CNS caseload. This will help us to confirm that we can get 25 participants.
- **Written permission** from Line manager: Ram to reply and confirm – change this in SREP.
- **SF36v2 Copyright:** Ram to email the copyright owners re change of supervisor and get it amended. Then email it to Mike
- Ram to email IRES copy to Mike
- **Capacity ax to participate in the research:** SCOLP may not be functional and contains irrelevant questions (not related to research). Therefore assessing capacity to engage in interview may be assessed by asking related questions – requesting participants to summarise the information sheet or couple of questions from the information sheet. To check previous literature, synthesise and inform practice re capacity to participate in research – apply principles in research.
- To pilot the **interview schedule** with work colleagues.
- **Supervision log:** Mike agreed to check if Uni system has recorded previous supervision logs from Ram

**CONCERNS RAISED**

- See discussion

**TASKS TO DO FOR NEXT MEETING**

- Email IRES and updated copyright license agreement – SF36 to Mike.
- Complete and send Form 6
- Check number of clients with Cerebral Palsy
- Pilot interview schedule
- Arrange meeting with R and D (NHS ethics panel member)
- Email manager regarding permission for research

**Plan**

Next session: Monday 7<sup>th</sup> October 2013 at 8:15 am  
Monday 28<sup>th</sup> October 2013 at 10 am