Early Social Communication Skills of Children with Cerebral Palsy

This thesis forms part of the UCL

Doctoral Degree in Clinical Communication Science

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Declaration

I, Katherine Price, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Acknowledgements

I would like to thank a great number of people who have inspired and supported me to be able to undertake and complete this study.

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“I think you'll find it's a bit more complicated than that.”

Dr Ben Goldacre, Oxford Centre for Evidence-Based Medicine

ABSTRACT

Title: Early Social Communication Skills of Children with Cerebral Palsy

Abstract: The clinical motivation for this study arose from repeated observations that some children with cerebral palsy (CP), despite provision of equipment and support, were still failing to reach their expected communication potential. In the clinical field, this failure has been commonly viewed to be the result of physical dependence on adult partners, or linked directly to the children's physical or learning disabilities, arising from the neurological and developmental deficits associated with CP.

Social responsiveness and shared attention underpin language and communication development. Children with cerebral palsy (CP) may be vulnerable to disruption in the development of these foundation skills (Nordin & Gillberg, 1996). However, there are few guidelines for assessment of these skills in this group of children (Watson & Pennington, 2015). This current study aimed to

1. develop an assessment protocol to support the identification of autism spectrum disorder (ASD) in children with CP at GMFCS levels IV and V
2. compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
3. investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

The study included 57 children in these three groups, matched for age, language and non-verbal abilities. The children with CP (n=32) were screened for their ability to use looking behaviours to give responses (Clarke et al., 2016). A measure of social responsiveness/joint attention, (*Gaze-NoTe),* accessible by all three groups was derived from established assessments.

Children with CP gave reliable responses to the tasks offered, and a range of skills was seen. Many children showed social responsiveness/joint attention skills at a level of development significantly below their language age/performance age, and the performance on the target measure *Gaze-NoTe* was significantly different across the three groups.

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1 INTRODUCTION

The aim of this thesis is to examine the social communication skills of children with severe cerebral palsy (CP) who have little or no functional speech.

This focus is important both theoretically and clinically: it is known and observed that children with CP have neurological damage affecting many areas of their development (Rosenbaum, Paneth, Leviton, Goldstein & Bax, 2007). Consequently, children with CP have different experiences in learning and social interaction from their typically-developing peers (Murray & Goldbart, 2009; Dowden & Cook, 2012). Their motor and sensory difficulties mean that development in intellectual and communication skills can be difficult to assess: there are few established measures for such assessment, particularly of social communication, for non-speaking children with severe CP. Without assessment and understanding of developmental profiles, it is difficult to provide relevant interventions for communication development and family/school counselling (McDonald, Harris, Price, & Jolleff, 2008). In consequence, this thesis focuses on the following research aims:

- to develop an assessment protocol to support the identification of autism spectrum disorder in children with CP at GMFCS levels IV and V
- to compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
- to investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

1.1 STRUCTURE OF THE THESIS

The structure of this thesis gives background to the clinical motivation driving the research aims, and describes and documents the characteristics of the group of children in focus from the author's clinical caseload; namely, young children with severe cerebral palsy and little or no intelligible speech. The development of a measure of social communication skills, accessible for children with motor impairment is described, and performance across comparison groups discussed. Further analysis is made of the performance on this measure by the children in the CP group. Results, discussion and implications are then presented.

Chapter 1 provides an introduction, describing the author's clinical background, and the observed difficulties with social communication skills in the target group of children. Case examples are given by way of illustration, and the research aims outlined.

Chapter 2 discusses the known, documented characteristics of children with cerebral palsy and **Chapter 3** describes the social communication difficulties associated with autism spectrum conditions. This chapter also contains a review of key published studies about the co-occurrence of CP and ASD/social communication difficulties, and the definition and development of early social communication skills: of orienting gaze to their conversation partner's face and of joint attention, learning to share focus on objects and activities.

Chapter 4 focuses on the methodological challenges associated with producing sound evidence for clinicians working with children with cerebral palsy, and includes consideration of appropriate research design, and of procedural decisions around assessment measures. This discussion is followed by a section detailing the final decisions made, for this study, with regard to design and procedural issues.

Chapter 5 details the recruitment procedure for identifying a group of children with CP for the study. Some significant weight is given to this process, as some difficulties were

encountered identifying a group of children meeting the inclusion criteria defined at the end of Chapter 4. The development of an assessment of functional gaze control is detailed, and other background measures selected are described in more detail, together with the adaptations needed to those assessments to render them accessible to children with CP. The procedure for assessment of background measures is presented, and findings from a pilot study.

Chapter 5 ends with a presentation of the results of the functional gaze control assessments for children with CP, and description of the final included group of participants with CP.

Chapter 6 describes the identification and recruitment of the comparison groups of children: those children with Down syndrome, and children with ASD.

Chapter 7 describes the development of an assessment of early communication skills, with responses possible by gaze direction alone, *Gaze-NoTe* (*Gaze: Noticing and Telling*), to explore children's joint attention and social responsiveness abilities, is described.

Chapter 8 presents the results from group **matching** procedures, and from the administration of the *Gaze-NoTe* protocol for both **between group** performances, and **within group** (for the children with CP).

Chapter 9 discusses the interpretation of these results, and their clinical implications.

Finally, **Chapter 10** lists bibliographic references: **Chapter 11** contains all appendices.

1.2 CLINICAL BACKGROUND

The author works with a specialist, multidisciplinary clinical team addressing the questions of families and local education/health team members regarding the development of communication skills in children with cerebral palsy (CP). This work includes discussion of the use of augmentative and alternative communication (AAC) methods, techniques and equipment: children with motor speech difficulties such as cerebral palsy can use non-speech methods (facial expression, direction of gaze, body movements); printed vocabulary material, and, for some, assistive communication technology (ACT) to support their unintelligible speech.

This clinical team comprises medical, technology, psychology and therapy staff to offer specialist assessment and intervention advice for children whose communication and learning might be constrained by significant motor impairment. The team assessment aims to develop a detailed understanding of the child's communication skills, including not only the child's levels of speech and language, but also their ability to use movement, vocalisation and vision skills as important expressive communication tools.

Thus the clinical team construct their interventions within a multidisciplinary developmental disability framework, in which all aspects of the child's development are considered to build a profile of strengths and weaknesses. A framework of this type values the consideration of individual areas of developmental functioning (physical, intellectual and sensory). This examination of individual domain abilities does not deny the acknowledgement of the interdependency of different areas of functioning in promoting child development, but aims

- to generate a logical sequence for assessment of different areas of functioning
- to identify factors contributing to "under-performance" in complex functions such as communication

- to implicitly dictate management priorities, with some difficulties (pain, epilepsy, visual difficulties) requiring resolution before tackling complex functions problems (communication, reading and writing)
- to serve as a framework for educating junior staff (for example: to take a pertinent history, and to focus on the most essential aspects of the child's needs)
- to facilitate parents to understand the interdependency of areas of functioning, and thus put recommendations in a contextual framework

(Cass et al, 1999)

Following this detailed assessment, to which children and families contribute, advice on communication strategies and/or equipment is shared with the child, family and local team. As the service is hospital-based within a Neurodisability department, any further medical, motor or sensory issues can be explored by other specialist departments.

Referrals are received from secondary National Health Service (NHS) sources (clinical staff in Child Development Centres; community paediatricians, speech and language therapists and occupational therapists). Specific questions are posed to the clinical team on a range of issues relating to communication: advice on selection of intervention approaches; on selection and use of assistive communication technology, on assessment of intellectual or visual impairment. On occasions, the questions asked of the team relate to unexpected poor progress with communication abilities development, despite all foundation skills and support appearing to be in place.

1.3 "GAPS" IN COMMUNICATION ABILITIES DEVELOPMENT

There were repeated observations, from this specialist communication assessment service caseload, that some children with CP did indeed appear to have specific difficulties in reaching their expected communication targets, despite apparently having the intellectual, visual acuity and motor abilities needed for success. A common observation was that the children often appeared to underuse their vision skills for eye contact; for bringing objects of interest to others' attention, or to use direction of eye gaze to signal messages. These children had difficulties in "noticing and telling".

Such discrepancy in abilities was puzzling, with some children showing "gaps" in their communicative competencies, since some abilities associated with communication development (language understanding, for example) appeared more advanced in development than other social communication skills.

In consequence, the team sometimes described any such discrepancies in abilities, when identified, by using a "slider" diagram (Figure 1-1), which often proved useful in discussion with local teams and families. The diagram aimed to present the relative strengths and weaknesses of developmental abilities observed during the clinical assessment, and identify any of these discrepancies. Scores and age levels were sometimes included, as clinically indicated, and on some occasions, it was helpful only to look at relative strengths and weaknesses in the child's profile.

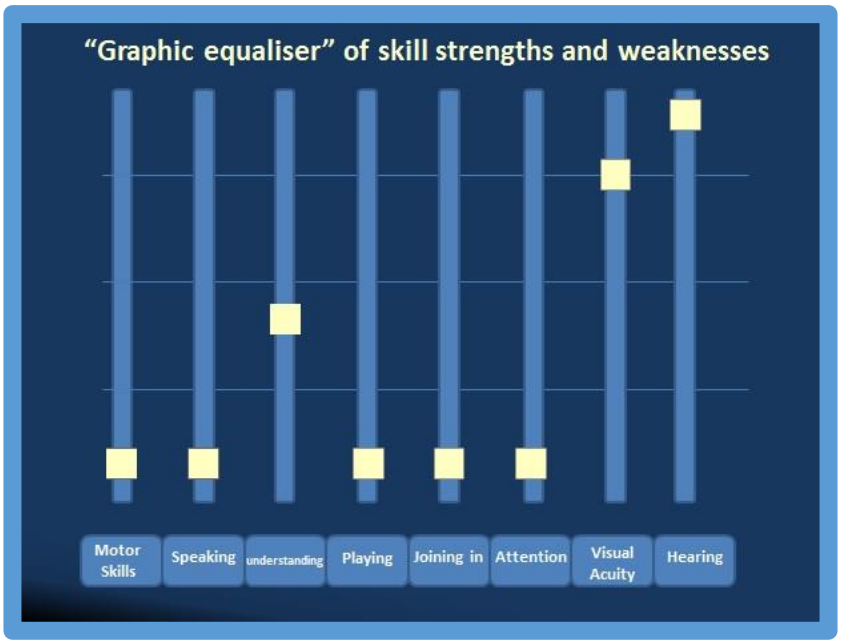

Figure 1-1: "Graphic Equaliser" of skill strengths and weaknesses

This profile in Figure 1-1, for instance, shows these relative strengths and weaknesses of a child who has good visual acuity and hearing, and adequate abilities in language understanding, but weaknesses in motor ability, speech, imaginative play, social participation and attention. The child was making poor progress despite good support with ACT and other AAC interventions.

Furthermore, the clinicians and teachers requesting advice from the specialist assessment team often discussed underlying reasons for the difficulties, citing visual attention, visual perception, cerebral visual impairment, learning motivation, severe intellectual disability, mood, emotional frustration, boredom and fatigue as possible sources of the apparent underachievement.

In some children, where the difficulties with social communication appeared more marked, and there were other behavioural markers such as need for adherence to routine or significant anxiety, parents and families might ask the clinical team if an

additional description of autism spectrum disorder (ASD) would be appropriate for their child. This was also the case for some children with intelligible or partially intelligible speech, or those children who had acquired skills to communicate through non-speech methods (signing, use of printed material, or assistive communication technology). The language output of this latter group might be unusual enough (repetitive questioning, obsessive topic adherence, pronoun confusion) to alert clinicians or families to similarities to the utterances typically heard from children with ASD.

To consider these questions, the specialist clinical team had the option to discuss these behaviours within the context of ASD; many of the behaviours seen (reduced eye contact, reduced range of motivating interests) were known to have comorbidity (co-occurring disease/conditions) with cerebral palsy. Reference to the co-occurrence of CP and ASD had been noted in the literature for some time, although the difficulties of establishing robust diagnosis in the population of children with cerebral palsy had also been documented (Fombonne, 2003; Nordin & Gillberg, 1996).

Furthermore, the impact of an additional neurodisabling diagnosis, often suggestive of a poorer outcome, was distressing and stressful for many families, and only undertaken after comprehensive assessment and discussion between the team and family members. Following such discussion, for children with less severe motor impairment, who could complete the tasks associated with standard assessment procedures for investigating ASD, these procedures were then undertaken and diagnoses discussed.

However, there were difficulties for children more severely affected by CP. The use of gold standard ASD assessments, which include toy manipulation and behavioural questionnaires written for children without motor difficulties, was not possible. For

these children, the clinical team chose, in preference, to describe communication profiles in terms of any specific deficits identified during assessment (poor visual attention, narrow range of interests, reduced use of eye contact), and relate these deficits directly to their advice to families and local teams on strategies for intervention.

The clinical observations relating to reduced use of eye contact, and of poor use of gaze direction for giving or supporting messages, seemed particularly salient for children with CP, who often rely on their eye gaze for an expressive communication channel.

In addition, one of the most common referral questions to the team concerned the child's use of "eye-pointing" (see 4.2.3.2) for full discussion of this term. Some children appeared to underuse their gaze direction to give messages, or make comments, or looked so briefly at objects that their listeners were unsure whether the looking was communicative or not. Other children could sustain their gaze on objects of interest, but appeared to have difficulty learning to look back at their listener to "close" (confirm) their message.

The risk of failure to identify and discuss such impairments with families, school and therapy staff was for intervention approaches to be misdirected or suboptimal, with misunderstandings occurring concerning children's failure to benefit from approaches presuming intact skills in these areas of social communication.

There was a significant frustration, however, with the lack of any robust published measures for examining the use of gaze direction for children with cerebral palsy, or guidance on assessment procedures to describe the development of intentional social communication. There are few published or experimental measures to use with this

population, and many SLTs use their own devised assessments, or rely on parental questionnaires to understand and document these skills of gaze direction and social communication.

This lack of specific, published measures was confirmed by Watson and Pennington (2015) who conducted an online survey of the assessments (and interventions) employed by SLTs in the United Kingdom (UK) working with children and young people with CP. The authors concluded

Children with CP have wide-ranging speech and language impairments and communication needs. UK guidelines exist on the areas of need that should be assessed and managed by SLTs, but lack details on how this should be done

(Watson & Pennington, 2015, page 241)

The author's clinical team were, then, presented with a frequently encountered dilemma in SLT and in neurodisability work: to offer evidence-based assessment, diagnosis, management, intervention and advice in the face of an often inadequate body of published research to support the clinical expertise, and child and family perspectives.

This dilemma was demanding of SLTs and their colleagues that they consider a diagnosis of autism spectrum disorder, or of "milder" deficits in social communication skills that were apparent, but possibly not reaching "threshold" for such an ASD diagnosis to be useful. In consequence, in this study, the terms "social communication deficits/skills" will be used. This is a more inclusive term to set the ethos of exploration of the phenomenon.

1.4 CASE EXAMPLES

It may be helpful here to describe, briefly, two children about whom these discussions took place.

Lulu was nine years old, and enjoyed many aspects of family and school life, and especially liked TV game shows. She had been diagnosed with bilateral dyskinetic cerebral palsy at the age of two years, and used supportive seating in a wheelchair for all her mobility. Her hearing was good, and she wore glasses to correct short-sightedness, but had no other identifiable visual problems. Lulu had some manual ability to hold and discard toys, albeit briefly.

She attended a mainstream school with full-time assistant support, and was making satisfactory academic progress, with a modified curriculum, acknowledging her intellectual disability, described and understood as within the moderate learning difficulties range.

Lulu did not use any recognisable words of speech, but used vocalisation to call her parents' attention, and to support her messages for needs and wants. Her use of eye contact and eye-pointing was described in SLT reports as "emerging". She had access to AAC support via a tablet computer, on which communication software based on symbols for phrases had been installed, and which Lulu was learning to access via a single hand switch, scanning the arrays to select her messages.

Her parents and teachers, however, referred frequently to her "behaviour problems", which included insistence on strict adherence to routine, non-compliance with adult chosen activities, and puzzling disregard for the feelings of others. Lulu's parents described her as "unfeeling" and were greatly concerned that she might hurt children

at school. She was reported to use her AAC tablet PC infrequently and unwillingly, and only to request her favourite TV programme. In school, she was often excluded from the classroom as a result of her non-compliance, but seemed to treat the isolation with her teaching assistant as a reward rather than a sanction.

Following referral to the author's specialist communication service, a team assessment observed and assessed many of these characteristics, and raised the possibility with her family that a detailed assessment focusing on a description of autism spectrum disorder might be helpful. Her parents were relieved by this, sharing that they had considered that "Lulu might have autism as well" from observation of her communication and behaviour patterns.

Case history details, observation of communication and behaviour, questionnaire information and parental perspectives were collated to a decision that an additional description of autism spectrum disorder would be useful for Lulu to direct her teaching and clinical intervention goals.

Muyal was 13 years old, and had bilateral spastic cerebral palsy. He enjoyed spending time with his twin brother, and they were both dedicated Queens Park Rangers football team fans. He attended a school for children with special needs, where his *Education, Health and Care Plan* (EHCP) described him as having intellectual disability within the severe learning disabilities range.

An SLT assessment described Muyal as a "beginning intentional communicator", but the referring SLT's concerns were that Muyal used eye-pointing only briefly, and that it was not consistently used to make choices. He had been referred for an assessment for eye gaze access technology, and his SLT had made the referral to ensure that some background information was available to be able to consider this request more fully.

Muyal wore glasses, which had been prescribed following refraction method testing (looking at the back of the eye with a series of lenses) and identification of long-sightedness. He was consequently described in his EHPC as having “good vision”.

Muyal’s strongest communication channel was through vocalisation, which he used with gusto to try to convey his needs and wants. He was intolerant of any change in routine, and amazed his parents by being completely aware of any changes in their journey to and from school.

On assessment by the author’s specialist clinical communication team, our play and observations noted Muyal’s marked visual inattention, narrow range of communication interests, and concentration on self-chosen activities. He did not respond to his name being spoken, even by his parents, and, on assessment of functional vision by the team’s developmental paediatrician, was not able to fix and transfer his gaze in toy play. He could fix gaze briefly on his favourite object (an *iPad* showing the Queens Park Rangers logo), but did not make eye contact, or react to changes in adult facial expression.

The author and team discussed Muyal’s difficulties in terms of his marked visual attention, explaining that his corrected vision did not guarantee full use of functional vision, and that Muyal might need some help to focus his visual attention.

Communication intervention recommendations concentrated more on the use of Muyal’s auditory channel, encouraging and teaching a *yes/no* response in order for him to be able to use partner-offered choices for conveying messages (*did QPR win on Saturday?*) and adapting AAC methods with techniques and approaches known to be useful to children with ASD (Iacono et al, 2016).

1.5 FRAMING THE STUDY AIMS

The focus of interest, then, lay in the population of children with severe cerebral palsy. Many of these children had confirmed intellectual disability, but did, nevertheless, appear to have social communication development less well advanced than their performance abilities or language understanding. Clinical observations had noted differences in the competencies of children in this group in using gesture, eye gaze direction and facial expression to convey or support their communicative messages. For some children, even those with aided communication, these skills seem to be underused, and at odds with other areas of their development.

Discussion with other specialist SLTs through the *Royal College of Speech and Language Therapists* professional network for cerebral palsy suggested that other colleagues had noted similar patterns in the development of children in this group. Furthermore, colleagues agreed that there was a paucity of frameworks for assessing and monitoring this aspect of communication. It was agreed in discussion that inability to engage via non-verbal (gestural/gaze) communication methods was as disabling, if not more so, than lack of clear speech.

The network group also reported more confidence in assessing social communication skills, often towards a diagnosis of ASD, in children with cerebral palsy who had less severe motor problems, greater intellectual ability, or speech, who could often access standard assessments more easily.

Within the framework of the Clinical Doctorate programme, this study was devised to begin to investigate how the abilities involved in social communication development could be assessed for the group of children with no or little speech, intellectual

disability and severe motor disability. Current evidence would be identified and appraised, and assessment made of the applicability of available recommendations for this specific population.

Recommendations for assessment would then be used to test children in the target group: those children with severe cerebral palsy and associated disabilities. In this way, it was hoped to document the feasibility of social communication skills assessment for this group, and to appraise these skills within the context of the children's other areas of development. The assessment could also be conducted on other (comparison) groups to identify any similarities and differences in patterns of performance.

2 CEREBRAL PALSY

Cerebral palsy has been, in the past, largely understood as a disorder of movement, resulting from a non-progressive injury to the brain in infancy or early childhood. More recent definitions, however, are now increasing full understanding of the condition and the implications for development and function. This increase in understanding has come from better description of the wider sequelae of the brain insult: the working party led by Bax and Rosenbaum, first reporting in 2005, proposed a more useful definition as follows:

Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain. Its motor disorders are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour, by epilepsy and by secondary musculoskeletal problems.

(Rosenbaum, Paneth, Leviton, Goldstein & Bax, 2007; page 9)

The appreciation of this wide range of developmental difficulties has allowed clinicians and researchers to pay closer attention to the assessments, strategies and interventions aimed to support children and young people with CP. However, the disturbances described (of vision, cognition, communication and behaviour) are still to be "unpacked" before better understanding of the range and type of disabilities might allow targeted and evidenced clinical methods for use with this group.

To support this better understanding there has been a drive for shared terminology in the field, encouraged by the use of the World Health Organisation's framework of the *International Classification of Functioning, Disability and Health (Children and Youth Version*) (ICF-CY) (World Health Organisation, 2007).

Both researchers and clinicians working with children and young people with cerebral palsy have made use of this framework to adopt common descriptions, and thus to improve transparency of definition.

As a classification, the ICF-CY is organised into two parts, with two components in each part: part 1 is headed *Functioning and Disability*, and includes the components *Body Functions* and *Body Structures* and *Activities and Participation*. *Body Functions/Structures* descriptions inform how a child functions mentally and physiologically, and descriptions in *Activities and Participation* relate to how a child performs tasks functionally, and participates in activities in everyday life. For example, a child with cerebral palsy may have no speech (body function deficit), but be able to participate effectively in an exchange with her friend through facial expression, gaze direction and structured support from her listener.

Responsive listener support contributes greatly to the co-construction of meaning that has been both clinically observed and reported in studies for children with cerebral palsy (see Pennington, Goldbart & Marshall, 2004, for review). The ICF-CY documents these factors as (part 2) *Contextual Factors*, which include *Environmental Factors* and *Personal Factors*.

With this wider perspective, the ICF-CY framework emphasises the need to consider abilities and disabilities beyond the anatomical and physiological level, to include consideration of individuals' abilities to participate in the activities of school, friendship groups and family life.

This increase in common frameworks and terminology has led to more use of clinician-held registers of children with CP, and this can support research efforts and focus, as well as directing effective intervention resources. A European register from 2000

reported incidence of CP as 2-3 per 1000 live births, with significantly higher figures for prematurity (40-100 per 1000 live births) (*Surveillance of Cerebral Palsy in Europe*, 2000). While prevalence figures have remained apparently static in the UK and Western Europe, the group of children with CP, as a heterogenous group, are still not always well defined, and accurate prevalence figures continue to be a challenge. Adoption of classification systems may help refine understanding of prevalence.

2.1 CLASSIFICATION OF CEREBRAL PALSY

Cerebral palsy is still classified, primarily, by the neuromotor type and the distribution (location) of the movement disorder, despite the changes in definition. The motor type is, generally, now classified as spastic, dyskinetic, ataxic or of a mixed type (often also classified within spasticity). The dyskinetic category includes dystonia, chorea and athetosis. The motor type describes, in the main, the muscle tone differences for individuals in that category, as follows:

CP type	% of CP	characterised by
Spastic	85-90%	generally increased muscle tone, with increased, brisk reflexes
Dyskinetic	7%	recurring uncontrolled, involuntary movements with abnormal, varying muscle tone *dystonia* has reduced, stiff movements with some increased muscle tone *chorea/choreo-/athetosis* has increased movements with reduced muscle tone giving writhing, jerky movements
Ataxic	4%	generalised low muscle tone with loss of muscle co-ordination, poor accuracy of movement

Table 2-1: Cerebral Palsy Type Description

The distribution of the CP for children with spasticity is described in terms of the areas of the body affected: for ease of understanding, and reliability issues, distribution is now usually stated as *unilateral* or *bilateral.* Bilateral spastic cerebral palsy will generally involve the whole body.

In addition to motor type and distribution, several other classification systems have been developed to describe functional abilities within the domains of, for instance, gross mobility, handling objects and materials, efficiency of communication, and speech intelligibility. These systems (*Gross Motor Classification System* (GMFCS) (Rosenbaum, Palisano, Bartlett, Galuppi & Russell, 2008); *Manual Ability Classification System* (MACS) (Palisano et al., 2006); *Communication Classification System* (CFCS) (Hidecker et al., 2011) and the *Viking Speech Scale* (Pennington et al., 2013) are described in detail in the appendices (11.1), and the classification levels will be referred to frequently in this document.

2.2 CAUSES OF CEREBRAL PALSY

The brain injury resulting in CP can occur before birth, and up to approximately two years old. The majority of CP (80%) is caused by an event in utero; 10% by post neo-natal brain injury (for example, meningitis, stroke), and 10% by hypoxia (lack of oxygen) during birth (Wimalasundera & Stevenson, 2016). Diagnosis is made both clinically, and with the support of neuroimaging.

Although there are no directly correlational links between motor disorder, severity of disability and damage area, magnetic resonance imaging (MRI)-identified brain abnormalities have been reported at the following approximate prevalences in

children with cerebral palsy[1]: white matter damage (in general, caused by lack of oxygen) is reported in 45%; basal ganglia or deep grey matter damage (containing neuronal cell bodies, linked to neurocognitive processes such as thinking, sensory processing) in 13%; congenital malformation is seen in 10%, and focal infarcts in 7%. White matter damage, including periventricular leukomalacia (where decreased blood flow to brain tissue causes the tissue to soften and atrophy) is more common in children born pre-term than in those born at term, and may occur in children with any functional level or motor subtype, but is more common in children with spastic than with dyskinetic cerebral palsy: basal ganglia or deep grey matter damage is largely associated with dyskinetic cerebral palsy.

2.3 ASSOCIATED DISABILITIES

The revised definition of cerebral palsy (Rosenbaum, 2007) alerted clinicians to the full range of associated disabilities which were, in fact, part of the CP condition. The range of potential disabilities and hence barriers to development for children with CP is significant, and includes difficulties with

- mobility and posture
    - walking, standing, sitting, lying easily and comfortably can be affected
    - hips, spine, and all joints can need orthopaedic management
- eating and drinking abilities
    - at all stages of gastro-intestinal tract
    - weight gain and mealtime duration

[1] https://is.gd/NICEguidelinesCP (accessed October 2016) Draft guidelines for *Diagnosis and Management of Cerebral Palsy in Children and Young People* (National Institute of Clinical Excellence, due for publication January 2017) (accessed October 2016)

    - chewing, swallowing and saliva management
    - reflux, aspiration of food and drink into the airway, constipation
- respiratory efficiency
- epilepsy control
- pain, discomfort and sleep disturbance from any of the above
- intellectual (cognitive) impairment
- speech, language, voice and social communication deficits
- vision deficits
- behavioural difficulties (non-compliance, social difficulties)
- reduced social, employment and leisure participation
- family stress, access to support services and equipment

The discussion now will present in more detail the possible impact of some of these potential difficulties (highlighted in bold, in the list above) that are the remit of speech and language therapy clinicians addressing communication impairment, and will present the evidence available describing the direct measurement of their occurrence and severity.

2.3.1 INTELLECTUAL DISABILITY

The term has a range of synonyms; some now outdated, some in common use, as follows:

- mental retardation
- learning disabilities
- learning difficulties (UK term)
- developmental delay
- slowed learning
- intellectual impairment
- cognitive disability

The term intellectual disability will be used in this document, and is defined in the *Diagnostic and Statistical Manual of Mental Disorders DSM-IV* American Psychiatric Association, 2000) :

- significantly sub-average intellectual functioning, with an intelligence quotient (IQ) of approximately 70 or below on an individually administered IQ test
- concurrent deficits or impairments in current adaptive functioning (the person's effectiveness in meeting the standards expected for his or her age by his or her cultural group) in at least two of the following areas: communication, self-care, home-living, social/interpersonal skills, use of community resources, self-direction, functional academic skills, work, leisure, health, and safety
- the onset is before age 18 years

This three-criteria definition is also noted in the most recent definition proposed by the *American Association on Intellectual and Developmental Disabilities* (AAIDD-11) : (Intellectual Disability: Definition, Classification, and Systems of Supports (11th ed), 2010):

Intellectual disability is characterised by significant limitations both in intellectual functioning and in adaptive behaviour as expressed in conceptual, social and practical adaptive skills. This disability originates before age 18.

The occurrence and degree of intellectual disability in the group of children with CP varies greatly, with the general trend of its severity linked to severity of motor impairment, with reports in a population sample as high as 60% for children at level IV GMFCS (Rosenbaum et al, 2008), and 90% for level V (Shevell, Dagenais & Hall, 2009). An earlier study had estimated that approximately 40-45% of children with CP (across the full range of motor function, from mild to severe disability) have intellectual disability (Himmelman, Beckung, Hagberg & Uvebrant, 2006).

However, having confidence in measures of intellectual disability, particularly in the populations of children with special needs, is a significant area of difficulty for researchers and clinicians, and the range of methods used in published papers is very wide indeed, often making comparisons of results difficult. This challenge is discussed in more detail in Chapter 4 (Methodological challenges), page 4–72.

A further possible confusion merits airing: the distinction will also be made in this document between intellectual disability and performance skill deficits: performance abilities refer to that part of intellectual disability that is not directly related to language skills, and so refers to the mental capacity in dealing with and using nonverbal skills (problem-solving, shape and pattern recognition, categorisation are examples). Performance abilities are sometimes referred to as *non-verbal* skills (skills outside language processing) but this is, firstly, debatable, as verbal mediation may be involved in so-called non-verbal skills, and can also be a confusing term for those working with children with spoken communication difficulties, as *non-verbal* is also used as a term to describe non-speech methods of communication (for example, gesture, eye contact).

Performance abilities will be used to describe those aspects of testing aimed to understand a child's skills in understanding concepts such as object permanence, categorisation, visual memory and shape and pattern understanding. Assessments of these skills can frequently be completed without speech responses being necessary.

In summary, then, intellectual ability will refer to the collation of performance abilities and language abilities.

2.3.2 SPEECH, LANGUAGE AND COMMUNICATION DEFICITS

Many children with cerebral palsy (CP) experience difficulties with communication. Estimates from the *Surveillance of Cerebral Palsy in Europe* study from the 1980s (SCPE, 2002) , in a group of over 6500 children with CP, showed that as many as 58% had deficits of communication which were severe enough to affect their learning and their inclusion in family and school life.

Furthermore, a Northern Ireland CP register study involving 1357 children, with median age of 71 months, (Parkes & Hill, 2010) noted that 36% of the cohort reported motor speech impairment, and 42% had impairments of expressive communication.

These communication problems can arise from many sources. **Speech intelligibility**, voice and prosody difficulties relate to impairment in the production of smooth and fluent movement to produce and co-ordinate movement of the speech and vocal muscles. For children more severely affected, intelligible speech may not be possible at all. The severity of motor speech disorders (dysarthria) ranges from this position of no speech at all, through to speech that will sound slurred and indistinct, and poor voice quality: this is sometimes referred to as “mild” dysarthria, although the effects on child development are likely or be significant. As a result of this variation, prevalence rates for motor speech problems in CP are sometimes difficult to interpret: with the Northern Ireland register study (Parkes and Hill, 2010) suggesting 38% of 3-9 year old with CP had motor speech difficulties, with figures from a whole population study (Iceland) (Sigurdardottir et al., 2008) recording 16% of 152 children between four and six years having *severe dysarthria*.

Additionally, **intellectual disability**, reported in 48% of all children with CP can lead to difficulties with both receptive and expressive language, and sensory impairments (vision and hearing) can also affect communication development.

Many children with more severe forms of CP (bilateral distribution) show a combination of these difficulties, and prevalence of speech, language and communication impairments is known to increase with severity of motor and intellectual disability (Shevell, Dagenais & Hall 2009).

A further risk for interruption of communication development occurs with the increased risk of epilepsy in this more severely affected group, estimated to occur in 35% of children with bilateral cerebral palsy. Seizure activity is known to affect communication skills development in both language and socio-cognitive domains (Pal, 2011). Sigurdardottir et al., (2008), in a study involving 127 children with CP, noted that 27% had epilepsy, and that epilepsy was the only associated impairment with an independent effect on intellectual ability scores.

This range of communication impairments will be evident in many children in this group from infancy and throughout life, and may impact on all aspects of development and learning. Teaching and intervention strategies will aim to minimise the effects of any communication impairments. The role of the speech and language therapist (SLT) for this group is described in terms of maximising children's ability to communicate independently, through both speech and non-speech methods (RCSLT, 2006).

Indeed, many children with whole body CP may rely on non-speech methods, known also as augmentative and alternative communication (AAC). These methods include the use of symbolic (object, printed material) displays to offer appropriate vocabularies, and, later, language structures, for the child to indicate by their preferred access method (finger-pointing, gaze direction, for example). Assistive communication technology (voice output communication aids/speech generating devices) may also be

useful to some children if it offers a more efficient method of conveying messages and developing language skills (Light & Drager, 2007; Andersen, Mjøen & Vik, 2010).

Children with CP, then, may have a wide range of speech, language, voice and pragmatic communication impairments which may restrict their success and efficiency in interactions. The effects on the child, and on their family and community life for such restrictions are known to be associated with reduced participation in activities. A cross-European study of over 1100 children with CP, aged 8-12, reported lower participations across most areas of daily life for those children with CP who have greater impairment of gross and fine motor function, and greater impairment of communication and performance abilities. These factors impacted more on the children's participation than sociodemographic background, which was not reported as significant (Fauconnier et al., 2009).

Speech intelligibility difficulties have been documented to restrict communication between children and their parents (Pennington & McConachie, 1999). Parents often report communication as their priority for hopes and efforts for their child (Marshall & Goldbart, 2008; Vargus-Adams & Martin, 2011).

A further influence on the communication patterns and success or failure of children with CP becoming effective communicators rests with the skills and motivations of their conversation partners. It is known that many conversational turns taken by non-speaking children are *yes/no* answers, and that non-speaking children have difficulties initiating conversation, with communicating with peers and with contributing their own topics (Clarke & Kirton, 2003; Pennington 1999). The complex interplay between inherent communication disabilities and atypical communication development is evident, but the trajectories are unclear.

2.3.3 VISUAL IMPAIRMENT

Visual impairment is often classified as a sensory disorder. However, any significant visual impairment may be better characterised as a neurodevelopmental disorder, in consideration of the role of vision in learning and communication, and the impact of impairment on the developmental process.

It is well established that children with severe cerebral palsy affecting the whole body are particularly vulnerable to damage to diverse aspects of the visual system. This may show in various ways including low visual acuity; visual field defects; refractive error, and other disorders related to the broader class of deficits known as cerebral visual impairment (Costa & Ventura, 2012).

For example, in a recently published whole population study of visual impairment in children with cerebral palsy born between 1999 and 2002 in Quebec, Canada, Dufresne and colleagues reported that of the 214 children examined almost half (48.9%) presented with some form of visual impairment, and that prevalence and severity of visual impairment increased with severity of motor disorder (Dufresne, Dagenais & Shevell, 2014) . This finding, linking increased risk of visual impairment with more severe forms of cerebral palsy, has been reported elsewhere (for example, Ghasia, Brunstrom, Gordon & Tychsen, 2008): a further study of 92 children, examining co-occurring conditions with CP, included 54 children in the most severe physically impaired categories (GMFCS IV and V). In this group, 66 out of the 83 children (80%) for whom information was available had a visual impairment, with 13 (21%) having a substantial impairment (Venkateswaran & Shevell, 2008).

Fazzi and colleagues (Fazzi et al., 2009) concluded from their study that neuro-ophthalmological disorders should be considered as *one of the main symptoms of CP*: they reported that children with 4-limb (bilateral distribution) CP showed a severely

visually impaired profile, characterised by ocular abnormalities (98%), oculomotor dysfunction (100%), and reduced visual acuity (98%). However, significant visual impairment in children with severe physical disabilities can go unrecognised for many years (Keil, Fielder, & Sargent, 2016).

Since intellectual disability is also known to be a risk for visual impairment (Boot, Pel, Evenhuis and van der Steen, 2012), children with severe CP must be considered to be at high risk for disturbances to visual abilities.

2.3.4 BEHAVIOURAL DIFFICULTIES

A further co-occurring condition identified in the updated definition of cerebral palsy concerns the behavioural difficulties, described as

psychiatric or behavioural problems such as autistic spectrum disorders, ADHD, sleep disturbances, mood disorders and anxiety disorders

(Rosenbaum et al, 2007; page 11)

These are, perhaps, the most poorly documented and understood additional disabilities: co-occurrence of ASD may have far-reaching effects on the development of communication, learning and participation for children with CP. Differential diagnosis distinguishing between a primary disability based in psychiatric behavioural problems, and one based in persisting social communication difficulties can be effected, but the co-existence of emotional/behavioural difficulties in the population of children with ASD is acknowledged (Simonoff, Pickles, Charman, Chandler, Loucas & Baird, 2008).

Sigurdardottir and others conducted an observation through questionnaire study with a small group of 36 children with CP (mean age = 4;6) (Sigurdardottir, 2010). Most of the children in the study could walk independently (GMFCS levels I and II = 83%), and

the most severely physically (and hence possibly intellectually) affected children in GMFCS level V group were excluded from the study. Nevertheless, it is interesting to read that 48% of this group had “problems” detected via the parental children’s behaviour checklist employed, and 65% on a caregiver-teacher report form. Both these figures were significantly higher than for the control group of children neurotypically developing. These problems in the group of children with CP included attention difficulties, withdrawal, aggressive behaviour, and anxious/depressed symptoms.

As an example, “withdrawal” might be a manifestation of autism characteristics; shyness; academic achievements set too high or too low; selective mutism, or anxiety and depression, and it can be difficult for clinicians in the community to interpret parents’ concerns about the behaviour of their children with cerebral palsy, and then difficult again to access appropriate intervention.

These behaviours cause significant concern and distress for both children and families, and appropriate intervention can often be difficult to obtain (Whittingham, Sanders, McKinlay & Boyd, 2014). The behaviour challenges reported also have confounding overlap with other causes: ideally, the identification of these behavioural problems would be the responsibility of a multidisciplinary team including developmental paediatricians, full therapy team, psychiatry and psychopharmacology to address all differential diagnoses and possible effective interventions.

This chapter has presented the deficits now reported as part of the characterisation of cerebral palsy, and these deficits may disable children further than the, more immediately apparent, motor disability. Those associated disabilities likely to interrupt or affect the development of communication skills are numerous, but, for this study and for clinical practice, it will be important to identify and characterise some of the factors that may suggest or preclude clinical approaches.

The following chapter discusses definition and characterisation of autism spectrum disorder. The observed social communication difficulties associated with this diagnostic description appeared to be most salient to the clinical observations described in the introduction at 1.3. Some children on the clinical caseload appeared to make poor use of their vision skills in functional communication: for eye contact; for bringing objects of interest to others' attention, or to use direction of eye gaze to signal messages. It was unclear if these difficulties with "noticing and telling" were related to deficits of visual attention of some nature, or to genuine difficulties with social responsiveness, social motivation and interaction (akin to the communication difficulties seen in children with ASD).

To start to understand these aims more fully, then, Chapter 3 looks in more detail at those communication difficulties associated with **autism spectrum disorder**.

3 SOCIAL COMMUNICATION DIFFICULTIES AND ASD

This chapter will give a brief background to current definitions of autism spectrum disorder, a description of the social communication difficulties associated with ASD, and will review the published studies identifying ASD/social communication difficulties in children with CP.

The single term *autism spectrum disorder* (ASD) is used throughout this document as is current practice. The history of description, causes and classification of ASD is a long and complex one: the discussion in this document serves to demonstrate the complexity of the condition and to highlight the impact of the diagnosis of ASD, as a primary or co-morbid diagnosis, on children and families.

In identifying the core deficits associated with ASD profiles, the ground-breaking work of Wing and her colleagues displayed ASD as a "triad of impairments" (Wing and Gould, 1979) as follows:

- **impairments of social interaction:** these are characterised most commonly by behaving as if other people do not exist; little or no eye contact made; no response when spoken to; often faces are empty of expression except with extreme joy, anger or distress; if something is wanted, carers' hands may be pulled towards the object; responding to rough-and-tumble play well, but unable to request repetition, and aloofness - "in a world of their own"
- **impairments of communication: in** understanding and using speech, in the use of intonation, and in understanding or using non-verbal communication such as pointing, gesture and facial expression
- **impairments of thinking and behaving:** repetitive and/or stereotyped activities

Wing described other features, including motor stereotypies (repetitive movements) and abnormal response to sensory stimuli, observed but not considered diagnostic

Most recently, the DSM (version 5) has introduced a further distinction, collapsing all subcategories of ASD into an "umbrella" category *autism spectrum disorder*. This revision defines ASD by two identifiable characteristics: (1) impaired social communication and/or interaction and (2) restricted and/or repetitive behaviours.

3.1 INCIDENCE AND PREVALENCE OF ASD

The incidence of ASD in the (UK) for 2004-2010 was reported (from the UK General Practice Research Database) at 1.2/1000 boys, and 0.2/1000 girls, with prevalence in a cohort of 8 year olds at 3.8/1000 boys and 0.8/1000 girls (Taylor, Jick & MacLaughlin, 2013). Prevalence rates remained steady for this quoted time period: there had been significant increases in the annual incidence rates of autism in the UK (and, indeed, the United States (US) (*Autism and Developmental Disabilities Monitoring Network,* 2012)). The reasons for such an increase have been much discussed; (for example; Hansen, Schendel & Parner, 2015) and include a possible widening of use of the diagnostic category and its boundaries, an increase in referrals for consideration of diagnosis, and, interestingly for this study, an increase in the number of diagnoses made concurrently with other neurodisabling conditions, including cerebral palsy.

3.2 ASD DIAGNOSTIC METHODS

The diagnosis of an ASD, even at its most "straightforward", is often challenging. It is now largely accepted that ASD has a multifactorial causation picture, with genetic vulnerability identified for some families. There is significant variability in presentation of behaviour and outcome, there is no single test to determine if the ASD "label" will be informative. Behavioural assessments are the means of diagnosis, and, as such, are open to subjective interpretation and bias. There are several tools available for the assessment of behaviour, although few of these tools have been subjected to robust and independent examination (Falkmer, Anderson, Falkmer & Horlin, 2013): Falkmer and colleagues undertook a review of 68 studies, and concluded that the *Autism Diagnostic Interview-Revised* (ADI-R) (LeCouteur, Rutter, Lord & Rios, 1989) and *Autism Diagnostic Observation Schedule* (ADOS) (Lord, Rutter, DiLavore & Risi, 2001) emerged with the largest evidence base and highest sensitivity and specificity, especially when used together.

The ADI-R is a parental interview/questionnaire, used in the identification of children with ASD. It has 93 items, comprising three sections corresponding to the "triad of impairments" described on page 3–43, covering language and communication; reciprocal social interactions, and restricted, repetitive and stereotyped behaviours and interests. The interview follows a highly standardised procedure, with the interviewer recording and coding responses, and documenting details of the child's background, education, medical health and diagnoses, developmental milestones, language acquisition, current developmental functioning, social development and play, interests and behaviours. The interview can be administered by qualified practitioners, including SLTs in the field of child health.

The areas of development targeted, and behaviours queried do, however, assume intact motor and visual abilities: for example, the items in the language and communication section include lack of, or delay in, spoken language and failure to compensate through gesture; lack of varied spontaneous make-believe or social imitative play and stereotyped, repetitive or idiosyncratic speech. The role of the SLT in this diagnosis is emphasised for children with speech intelligibility difficulties, or those who use AAC.

The *Autism Diagnostic Observation Schedule* (ADOS) is a semi-structured observational assessment of autism spectrum disorder. The assessment covers children aged one year through to adults, although the *Toddler Module* (Luyster et al., 2009) yields guidance on *ranges of concern*, rather than specific ASD diagnosis, for young children. The assessment presents various activities eliciting behaviours directly related to a diagnosis of ASD, in a standardised assessment of communication, social interaction, play and restricted and repetitive behaviours. Again, the tasks and play material presented assume adequate motor abilities for the manipulation of toys and objects. Use of the assessment is restricted to qualified professionals who have also undertaken ADOS test-specific training.

Current gold standard criterion diagnosis, therefore, is based on multidisciplinary team assessment using such tools, and on clinical judgement using the DSM-IV or the *International Classification of Diseases* (ICD-10) criteria.

In the context of this study, in addition to considering how ASD is diagnosed, specific attention to measures of *early* social communication abilities is merited. As noted in 2.3.1, (page 2–33) many children with severe CP have intellectual disability, with developmental levels corresponding to skills seen in very young neurotypical children. Although ASD is not generally described until a child is aged three to four years,

increasing focus has been placed on early diagnosis, as the evidence-base for the value of early intervention continues to build (Oono, Honey & McConachie, 2013). Screening assessments for community work identifying children at risk for ASD (for example, the *Checklist for Autism in Toddlers* (CHAT)) (Baron-Cohen et al., 1996) and structured assessments, including the *Toddler Module* of the ADOS described on page 3–46 (Luyster et al., 2009), are adding to the understanding of early indicators of ASD. However, such published instruments derive their validity and reliability strength from stringent restrictions on use, rigorous training for administration and competency updates, and hence have little room for adaptation for other populations.

As far as could be ascertained, there were no studies recording the use of ADOS assessment tools with children with cerebral palsy. However, the author of this current study is trained in ADOS assessment, and the insights from the structure of the tasks, and the tasks themselves were useful, suggesting the value of a play-based, scripted assessment.

3.3 "RED FLAGS": SCREENING FOR ASD

One approach to the early description and diagnosis of ASD in younger children is that proposed by Wetherby, Prizant and colleagues (Wetherby et al., 2004), who conducted one of their studies exploring the behavioural indicators of young children with autism: 54 children, aged 13-27 months, in three groups (children with autism (ASD), children developing typically (TD) and children with intellectual disability without autism (II)) were included. The study applied their *Systematic Observation of Red Flags of Autism Spectrum Disorders* to analyse video recordings of a play session.

Results suggested that the children with autism were significantly more likely than the children in the TD and II groups to show a number of deficits on social communication as follows:

- lack of showing
- lack of co-ordination of gaze, facial expression, gestures and sound
- lack of interest or enjoyment
- repetitive movements with objects
- lack of appropriate gaze
- lack of response to name
- lack of warm, joyful expressions
- unusual prosody
- repetitive movements or posturing of body

Furthermore, there were four “marker” behaviours identified in both the groups of children with ASD and with II, but not in the group of children developing typically:

- lack of pointing
- lack of playing with a variety of toys
- lack of response to contextual cues
- lack of vocalisation with consonants

These nine behaviours, significantly differentiating those children with ASD from neurotypical children and children with intellectual disability, together with the four behaviours distinguishing children with ASD and II from neurotypical children, were identified as “red flags”: important observable behaviours to be considered as alerts to persisting and significant social communication difficulties.

A further study in 2009, screening children at 18-24 months, confirmed the validity of the original 13 “red flags” and identified an additional seven red flags which differentiated children with ASD from neurotypical children and from children with intellectual disability (McCoy, Wetherby & Woods, 2009). This updated list of 20 observable behaviours grouped deficits as follows:

3.3.1 IMPAIRMENT IN SOCIAL INTERACTION

- *inappropriate gaze*
- *lack of warm, joyful expressions*
- *lack of sharing interests*
- *lack of response to contextual cues*
- *lack of response to name*
- lack of co-ordination of verbal and nonverbal communication

3.3.2 IMPAIRMENT IN COMMUNICATION

- *lack of showing*
- *lack of pointing*
- unusual prosody
- lack of communicative consonants
- using person’s hand as a tool

3.3.3 REPETITIVE BEHAVIOURS AND RESTRICTED INTERESTS

- repetitive movements with objects
- repetitive movements or posturing of body
- *lack of playing with a variety of toys*

- unusual sensory exploration
- *excessive interest in particular toys*

3.3.4 EMOTIONAL REGULATION

- distress over removing objects
- *difficulty calming when distressed*
- abrupt shifts in emotional states
- *unresponsive to interactions*

Mindful of the study target group of children with CP, and the clinical observations, the italicised markers are those considered by the author of this current study to be reliably observable for children with little or no speech and motor movement difficulties: it is interesting that only 11 of the 20 fall into this category, suggesting difficulties with full identification of a valid ASD diagnosis. These studies also highlighted three specific areas of concern identifying autism in children at aged two years

- lack of gaze to face
- lack of co-ordination of verbal and nonverbal communication
- lack of shared attention

.

For non-speaking children with CP, observation of *lack of co-ordination of verbal and non-verbal communication* may well not be possible. Focusing, then, on two skills, lack of gaze to face, and joint attention, that acted as "red flags" in McCoy's study, may inform the research aims of this current study with children with CP to investigate their use of these two early social communication abilities.

3.3.5 GAZE TO FACE

Infants' looking at faces is a very early social responsiveness skill. Newborn infants can be shown to attend preferentially to faces displaying direct gaze (Farroni, Csibra, Simion & Johnson, 2002). From early on, infants seek eye contact during close interactions; during feeding and lap play, and responses to direct eye contact develop considerably over the first 4 months (Caron, Caron, Roberts & Brooks, 1997).

Studies looking at infants' responses to facial expression have made use of the *habituation paradigm*, in which infants are presented repeatedly with a stimulus, and their looking, heart rate or sucking time is recorded to establish if they identify a change of stimulus as novel. In this way it has been possible to show that the ability to categorise different expressions, at least into the broader categories of positive and negative, emerges by 10 months of age (Ludemann, 1991). Furthermore, by the end of the first year, infants begin to use social referencing; observing others' expressions in such a way as to help them interpret other, environmental, events.

Response to emotion portrayed in the faces of familiar adults is an early and important part of the development of social understanding: newborn infants do react differentially to facial expression, but it is by three to four months that infants show reliable discrimination between positive and negative affect, and by the end of the first year can reliably discriminate among at least some expressions.

Effecting gaze to face, and eye contact are regarded as skills underpinning joint attention. For children with severe motor control difficulties, it is likely that these foundation skills may be impaired because of poor control of initiation, direction or

cessation of motor movement needed to achieve mastery of such skills (Arens, Cress & Marvin, 2005).

3.3.6 DEFINITION AND DEVELOPMENT OF JOINT ATTENTION

Joint attention has been described as the *simultaneous engagement of two or more individuals in mental focus on one and the same external thing* (Baldwin, 1995). Joint attention involves the triadic co-ordination of attention between self, other, and some third object, event, or symbol (Adamson, 1995; Tomasello,1995). In this document the phrase *joint attention* will be used for this phenomenon, seen in neurotypical development at approximately nine months of age, and achieved via direction of gaze shifts and finger-pointing.

This clarity of terminology is made since, more recently, Carpenter and others (Carpenter & Liebal, 2011) have refined the definition of joint attention, emphasising the *knowing together*: for true joint attention to take place, it is argued, an acknowledgement between the conversational partners occurs. As an example, two people can be watching a toddler trying on shoes: they have *simultaneous engagement of two or more individuals in mental focus on one and the same external thing*, but the attention is only joint, or shared, when the two exchange a smile or look, however brief, to acknowledge to the other that they are sharing the event.

This complex level of joint attention is considered widely to be species-specific to human behaviour: however, there are documented variations of the mechanisms of joint attention across different cultures and sub-cultures (Gavrilov, Rotem, Ofek & Geva, 2012). Studies reported in this thesis will take their general norms for joint attention behaviour from neurotypical children living in the UK, US and Europe.

Initial skills in joint attention, then, emerge in early months of childhood, and are linked to the development of face orientation, of gaze following, of understanding intentionality, of pointing and of the development of self/other distinctions, through to complex socio-cognitive concepts including theory of mind; the understanding and appreciation that others may have a different perspective/experience to one's own (Tomasello, 1995).

Building on these initial skills, children and their partners[2] begin to engage in dyadic exchanges involving face-to-face emotional interaction, and turn-taking. With the addition of sharing interest in objects and events, and not just faces. **Triadic** attention emerges: the relationship is now between child, partner and object/event (focus of interest). The distinction is made between **responding to joint attention (rJA)** and **initiating joint attention (iJA).** Responding to joint attention refers, in fact, to responding to *suggestions* for joint attention, and relates to infants' ability to follow the direction of their partner's head turn, their gaze direction, and/or a pointing gesture (Seibert, Hogan & Mundy, 1982).

Initiating joint attention, however, involves directing a conversation partner to an item (object, activity, speech). At its most complex, between adult conversation partners familiar with contexts, iJA can be very subtle indeed: a slight eye-widening, a lip shape change to "point" to a significance (object/activity/observation) to be shared. For children, iJA emerges typically using pointing.

When an infant can secure partner eye contact, and/or make pointing-to-show gestures to a focus of interest, this will initiate co-ordinated attention with their

[2] for brevity, the term *partner* will be used to signify conversation partner/communication partner

partner in a **proto-declarative** communication act (Bates, Camaioni & Volterra, 1975). This initiation of joint attention might be seen in a (neurotypical) 9-12-month-old toddler, raising a hand towards the sky, with an excited face, and perhaps a vocalisation, looking backwards and forwards between a hot air balloon and their parent's eyes. The "showing and telling" message is clear, even before the development of intelligible speech, but the skills required to achieve this are complex. These behaviours have been said to mark the transition between pre-intentional and intentional communication.

Children's early joint attention behaviours are both motivated and rewarded by social functions (Mundy, 1995). However, these social attention co-ordination behaviours can be used for more instrumental, **proto-imperative** gains (Bates, Camaioni & Volterra, 1975), ("pointing to get") in order for the child to secure needs and wants. This describes the toddler (9-12 months) securing partner eye contact, then looking and pointing to a favourite toy out of reach, before returning eye contact.

At this stage, around the age of 12 months, all objects or activities followed or indicated by the child have to be within the field of their vision. However, at this stage, infants' declarative (iJA) pointing already appears "premeditated", and they will wait to point until they can be confident they have their partner's attention (checked out by gaze to face). These are complex sequences of motor and cognitive events, established early for children developing without difficulties, and the challenges for children with cerebral palsy are not difficult to discern.

This development is summarised in Table 3-1:

Joint attention: developmental stages	
age range	**observable behaviour(s)**
newborn	preferential look to face
2-3 months	infants seek eye contact, smile in response to parents' smile or voice
6-9 months	infants follow the parents' gaze direction with their own
10-12 months	infant follows a finger point with their own gaze, and returns look back to parent
12-14 months	child will initiate a finger point
15-16 months	child will draw parents' attention to an object of interest within their field of view, using vocalisation, finger-point, looking back and forth
18 months	child recognises the role of the eyes in seeing, and can understand that objects can block line of sight.

Table 3-1: Joint attention developmental stages

The role of joint attention in language learning is important: shared focus allows the child to "map" vocabulary on to objects/events to "organise" their learning more efficiently. Several studies have discussed the link between measures of joint attention and subsequent measures of receptive or expressive language, or both, with positive correlations frequently seen in both neurotypical and other groups (for example, Adamson, Bakeman, Deckner & Romski, 2009). Studies have also showed a strong positive correlation between gaze-following behaviour at 10–11 months and subsequent language scores at 18 months (Brooks & Meltzoff, 2005).

In this way, joint attention can be seen as a fundamental ability for successful communication development, and offers developing infants the "way in" to interact and to engage with their social surroundings (Tomasello, Carpenter, Call, Behne &

Moll, 2005). However, for some children, these abilities do not unfold and emerge in the way described here for neurotypical development: the development of many children with significant and persisting difficulties with joint attention skills will cause considerable concern to their families and carers, and these children may develop the profile best described as autism spectrum disorder.

3.3.7 JOINT ATTENTION ABILITIES IN CHILDREN WITH ASD

Children with ASD have particular difficulty engaging in co-ordinated joint attention acts with partners, whether responding to the joint attention bids of others or initiating joint attention encounters (Meindl & Cannella-Malone, 2011). Joint attention ability is not only central to the differential diagnosis of ASD but also has been shown, for children with ASD, to be a strong predictor of later language ability (Mundy, Sigman & Cassari, 1990; Charman et al., 2003).

In a valued paper examining the differentially diagnostic function of joint attention, Mundy et al., 1986, the authors compared child behaviours via a parent interview assessment between groups of children developing neurotypically, children with ASD and children with intellectual disability without ASD, aged 38-75 months. The results of this study suggested that the behaviours of the children with ASD were at their most atypical in the category of initiation of joint attention behaviours. Children with ASD, in comparison with both the neurotypical children and the children with intellectual disability were noted to engage in eye contact or gaze to face significantly less frequently during play with toys. This lack of gaze to face was observed whether the toy was simply held by the adult, or made active (in the case, say, of a wind-up toy), and this finding has been reported in other studies (for example; Charman et al.,1997).

It has also been noted that children with ASD who display more intact joint attention skills exhibit better outcomes with respect to development of cognitive, language and symbolic play skills (Sigman & Ruskin, 1999).

It seemed important, then, for the study aims, to examine the evidence that had identified deficits in joint attention, and, indeed, other social/emotional characteristics of communication profiles of children with ASD, within the CP population.

3.4 CP AND ASD/SOCIAL COMMUNICATION DEFICITS

Research with children with CP has revealed deficits in some of these abilities associated with the impairments characteristic of children with ASD (for example, Christensen et al., 2014; Smits et al., 2011).

The literature falls largely into two categories: epidemiological studies, and smaller scale studies looking at specific factors such as joint attention and other, developmentally later, aspects of social cognition (for example, theory of mind). Published research divides, therefore, between those studies identifying ASD comorbidity (the child has two recognisable conditions), and those studies more concerned with describing the observed social communication deficits.

3.4.1 COMORBIDITY

The earlier identification of children with both CP and ASD diagnoses were largely medical prevalence studies, and comorbidity is the medical term describing the existence of an additional disorder co-occurring with a primary disorder.

These epidemiological surveys reported ASD profiles to be observed in children with CP. Nordin and Gillberg (1996) conducted a study of the prevalence and range of autism spectrum disorders identifiable in the total population of pre-school and school-aged children with learning disability and/or physical disability, in a discrete geographical region of Sweden. This study included a total of 177 children.

An adapted form of the *Autism Behaviour Checklist* (ABC), designed to examine autistic behaviour in people with severe intellectual disability (Krug, Arick & Almond, 1980), and the *Childhood Autism Rating Scale* (CARS) (Schopler & Reichler, 1980), designed to differentiate autism from other developmental disorders, were used as both screening and classification measures. Diagnoses were made following independent evaluation by the first and second authors of all information available, including video recordings of the assessments. Consensus was reached after discussion of three cases. There were 36 children identified with possible ASD: of these, 20 (11.3%) were described as presenting with autism, autistic-like condition, or ASD not otherwise specified (distinctions in use at the time of the study). A small number of children in these groups had pre-diagnosed conditions (for example, Rett syndrome, Tourette syndrome).

This study approached the topic of co-occurring conditions of CP and ASD that had previously been very poorly documented: however, Nordin & Gillberg acknowledged significant shortcomings in their measures, arising primarily, they suggested, as items on both ASD diagnostic tests used could not be scored accurately for children with physical disabilities.

Kilincaslan and Mukkades (Kilincaslan & Mukkades, 2009) assessed 126 children and young people, aged 4-18 years, with a wide spectrum of cerebral palsy types and distributions. In this sample, 15% (19/126) met criteria for an ASD diagnosis. Again, as

in the Swedish cohort, the ABC and CARS checklists were used, and again acknowledgement was made that only some of the items in both questionnaires were relevant for children with motor limitations.

In this study, 19 children were identified with both CP and ASD, of this 19, 7 had 4-limb (bilateral) cerebral palsy, and 14 were described as having “no phrasal speech”. There were 11 children who had no recognisable words of speech. A further 14 children in the group had learning disabilities assessed through standardised cognitive assessments as moderate or severe, although testing methods and any modifications made are not described.

Again, then, some methodological challenges were discussed, and some were evident: the authors report that they counted mutism as a feature of autism if the child made no attempts to convey messages by other channels of communication (gesture or mime, for example). The interpretation of mutism in children with severe learning difficulties and motor speech difficulties is clearly difficult.

Some valid observations were made in the population of children with CP who met criteria for the inclusion in the comorbidity CP/ASD group: some children were described as “behaving as if they could not hear” (despite exclusion of hearing impairment). These children did not respond to their names’ being called, but, as some of the group could speak, the researchers noted that the children did show clear non-speech responses in answer to questions about wants and needs. It was not possible within this study to make any detailed assessment of these skills.

In a sample of Icelandic children (n=152), researchers reported 7 children (5%) of the sample, all verbal children, to be diagnosed with ASD (Sigurdardottir and Vik, 2011),

although details of how the diagnosis was made are not given in the text, which focuses on expressive language function and cognitive skills of this group.

A review of population records monitoring ASD and developmental disabilities was conducted in four states of the US for 451 children with CP (Christensen et al, 2014). The identification of children with both ASD and CP descriptions was made by including (a) children with a confirmed or suspected diagnosis of ASD in their notes (b) children with a special education autism eligibility and/or (c) children whose record contained behaviours alerting the researchers to ASD diagnosis.

The frequency of co-occurring CP-ASD was reported at 6.9%. This frequency varied by CP type, and was noted to be 6.0% for children with spastic CP, 18.4% for children with non-spastic CP and 4.7% among children in the group described by the authors as other CP.

Researchers focusing on children with CP with less severe physical disabilities (those children in GMFCS levels I and II; children with hemiplegia) have been able to make clearer and more useful diagnoses of ASD, since these groups of children may be able to access standard assessments supporting ASD identification (Goodman & Yude, 2000). The difficulties of identifying ASD with any confidence in the group of children with severe CP is acknowledged, and some studies actively exclude this group of children from ASD investigations (for example, Stephens, 2012).

Although the discussion of underlying neurological causes for identified ASD in the CP population is beyond the brief of this current study, it is worth recalling at this point that inherent deficits have influence over subsequent interactions and subsequent development: there is an interplay of child and parents/carers shaping the trajectory of communication development (see 2.3.2, page 2–36). It may be that assumptions

have been made about in-child characteristics that may, in fact, be a product of the effect of innate characteristics on the behaviours of conversational partners. Indeed, the author of this study, in earlier publications, refers frequently to the "passivity" of children with CP, and this has been a common assertion. Reference is made to the asymmetrical adult-child turn-taking in both unaided and aided (AAC) mediated-conversations (von Tetzchner & Martinsen, 1996).

Clinical and theoretical discussion has proposed a number of, largely intuitive, reasons why children with cerebral palsy might be at risk for social communication impairment: they may understand body movements/gestures differently; their own communication intentions may be subtle and prone to loss or misunderstanding, and they may experience more episodes of communication "failure" than their typical developing peers. This experiential explanation does not preclude contribution from inherent/innate characteristics, and since it has also been suggested that there may be identifiable underlying neurological pathway deficits in some children with ASD (Jeste, 2011), it may be that some of the children with CP share similar pathway damage.

3.4.2 SOCIAL FUNCTIONING

Some studies have focused on the social development of children with CP, often linked to social functioning/participation outcomes in later life. Studies relating to early social development in younger children with more severe CP were considered to review assessment and outcome measures considered helpful for this group.

One such investigation targeted the relationship between physical disability and early social development in a group of preschool children with CP (Whittingham, Fahey, Rawicki and Boyd, 2010). The study group comprised 122 children, who were assessed at chronological ages 18, 24 and 30 months. Of this total, 22/122 children (17.6%)

were classified at GMFCS level IV, and 19/122 (15.2%) at level V. This study used a parent/carer interview (*Pediatric Evaluation of Disability Inventory* (PEDI)), (see 4.2.4 for description) aimed to identify functional child involvement ability in such areas as social interaction, social communication, interactive play and household/community tasks.

Results from the study showed a significant prediction relationship between physical abilities and social development at all time samples (18, 24 and 30 months). The researchers compared the *PEDI Social Function* norms with those reported for neurotypically developing children: at age 18 months, 44.3% of the children in the CP group had scores recorded that were greater than two standard deviations below the mean for social development, with a further 27.9% greater than one standard deviation below the mean. Although this study's methods did not allow investigation of the relevant contribution of intellectual disability to the findings, the study identified significant diversions from typical development for this group of children, and greater social development differences in GMFCS groups IV and V.

The authors concluded that children with CP might need support for social development from as early as 18 months.

In a longitudinal (three year) study of 110 children with CP to examine the associations between disease characteristics, personal and environmental factors and the children's social functioning and communication, researchers included a significant number of children with severe cerebral palsy (31/110 = 28%) (Voorman, Dallmeijer, Van Eck, Schuengel & Becher, 2010). No direct, in-child, measures of social functioning or communication were used, but a parent/carer interview and rating form was used as an outcome measure (the *Vineland Adaptive Behaviour Scales* (Sparrow, Cichetti & Balla, 2005). The *Communication* domain of this assessment is easy to administer and

relate to neurotypical development, but has a significant number of items relying on vocalisation, speech and pointing that may not be observable for children with severe CP (for example, item 10 in the 5+ years section; *follows instructions with one action and one object (for example, "bring me the book"; "close the door" etc*)).

It may be unsurprising, then, that the findings of the study included noting that restrictions in communication increased more over the three-year study period for the group of children in GMFCS category level V.

As a clinical observation, it is interesting to recall from work in the communication clinic that parents frequently commented on their frustration on being presented with interview questions that were clearly inappropriate for their children.

For the group of children forming the focus of the study, it seemed important to consider more carefully an appropriate assessment for the components of social communication, in order to examine the targeted abilities of social responsiveness and joint attention.

3.4.3 SPECIFIC SOCIAL COMMUNICATION DEFICITS

In one of the few studies targeting components of social communication development, Cress undertook a longitudinal study incorporating analysis of joint attention, in a group of children with a range of disabilities, including 19 children with CP, over a period of 18 months, between ages 12-24 months and 30-36 months (Cress et al., 1999). Communication skills were assessed using the *Communication and Symbolic Behaviour Scale* (CSBS) (Wetherby & Prizant, 1993) (see 4.2.4 for details of test).

Cress and colleagues reported that, although the children displayed the motor skills needed to signal joint attention through eye gaze shifts, they spontaneously used fewer joint attention behaviours than would be expected in neurotypical children across the 18-month period. A relationship was also observed between displays of joint attention and children's "sociability", measured through inter-rate agreement about social/emotional engagement.

Joint attention behaviours have also been examined by Arens and colleagues (Arens, Cress & Marvin, 2005) in 25 children with physical disability, and with communication abilities described as pre-intentional, aged 9-25 months, including 12 children with CP. The children were classified as pre-intentional communicators as they had failed to display joint attention behaviours during assessment with the CSBS. However, some joint attention behaviours signalled through eye gaze shifts between carers and objects were observed in free play, although rate of use was extremely low (mean proportion of time = 0.6%, SD = 1.1), and was much lower than would be expected in neurotypical children of similar developmental age.

The authors recorded a moderately low inter-observer agreement for their observation measures (Cohen's Kappa coefficient = 0.67) which they suggest may have been due to problems recording gaze shift behaviour in free play. The same 25 children displayed significantly higher rates of joint attention, and greater variation in rate, in adult-structured play involving adult prompting (mean proportion of time = 5.1%, SD = 8.1) where the explicit aim was to enhance reciprocal interaction. Inter-observer agreement was higher for this procedure (Cohen's Kappa coefficient = 0.89). Such changes in displays of joint attention were not related to children's level of language or motor impairment.

This study highlights the importance of researcher confidence in the ability of the children with CP to undertake the *motor* and *cognitive* components of any test of social communication skills. Without clear knowledge that the children understood the gaze shift tasks, and had the head/eye motor control to be able to transfer their gaze, it would have been difficult to interpret failure to complete the target (social communication skills) tasks. The need for efforts to maximise inter-rate reliability was also noted.

A further study investigated attentional and executive impairments in children with unilateral and bilateral cerebral palsy (Bottcher, Flachs & Uldall, 2010). The focus of enquiry was to better understand children's participation, through studies of the specific cognitive impairments associated with CP. The theoretical background presented was a neurological one: anterior lesions to white-matter tracts, lesions of the basal ganglia and thalamic functional systems, and infarction of the middle cerebral artery in the brain have all been associated with attentional and executive difficulties (describing the management of cognitive processes, to include working memory, verbal reasoning and problem-solving, flexibility of thinking, planning and adjustment). Children with CP may also have similar neurological pathway damage.

Bottcher and colleagues' study used a quantitative design, with two study groups, and using test norms for comparison of group means. The study involved 33 participants (14 female/19 male): 15 children were in the unilateral CP (UCP) group, and 18 in the group with bilateral CP (BCP). The age range was 9;11-13;6, and the groups were also characterised by their motor impairments.

Measures used included a standard neuropsychological assessment subtest (*Verbal Comprehension Index)* of the *Wechsler Intelligence Scale for Children* (WISC) (Wechsler et al., 2003) to estimate cognitive ability, through verbal response, a standard measure

of attention (*Test of Everyday Attention for Children* TEA-Ch) subtests. Executive function was examined by a further standard test (*Contingency Naming Test)* and a teacher-assessed observation assessment. These tests are not described in detail here as they do require a considerable amount of manual motor ability, and the low numbers for children in GMFCS categories IV (n=2) and V (n=0) is noted.

In their conclusions, the authors are careful to discuss the limitations of the study regarding small sample size, under specificity of individual and group characteristics and inappropriateness of some tasks for children with motor restrictions.

Bottcher et al.'s paper also highlights the difficulties of subject selection, consent and attrition. The subjects were invited from a register, and only half of the invited subjects consented to take part: this may reflect the burden that additional commitments can make on families of children with disabilities: travel arrangements, for example, may present particular challenges.

An important study in this field examined a further aspect of social communication skills development, focusing on theory of mind development in children with cerebral palsy and, the authors detail, severe speech and physical impairments (Dahlgren, Dahlgren-Sandberg & Larsson, 2010). This experimental study aimed to investigate how the skills of language abilities and short-term memory relate to performance on two tasks investigating theory of mind skills. Theory of mind (ToM) development refers to the skills acquired by children to develop their understanding of others' behaviour: as such, it is recognised as a foundation skill in the development of social communication. Theory of mind develops in childhood from birth to school age as the cognitive capacity to attribute mental states to self and others: to understand that others may have different perspectives, thoughts and feelings. Joint attention is

considered an important “pre-requisite” ability on the route to the development of ToM.

The authors stated their view that children with CP and speech impairment would inform the ToM field, as this group of children have deficits in accessing pretend play, language development and working memory: all considered important in the development of ToM.

Two groups of children were presented, selected from data gathered from a previous study: inclusion and exclusion criteria are not fully discussed. However, the target group are children described as having *cerebral palsy with severe speech and physical impairments* (SSPI) (n=14). A comparison group was also selected from children developing without apparent difficulty (neurotypically) (n=14). Some of the characteristics of both groups were described, including speech intelligibility, non-speech methods of communication, vision and hearing, and motor impairment.

The authors used a standardised picture-based cognition screening test, and two ToM tests; one, (“Sally-Ann”) used widely with children with ASD, and one adapted for children with physical disability. The memory tasks were subtests from other standardised, and norm-referenced assessments. Language ability was assessed using the *Peabody Picture Vocabulary Test* (Dunn & Dunn, 2007) and the *Test for Reception of Grammar* (Bishop, 2003), again standardised assessments, of single-word vocabulary understanding and understanding of grammar, respectively.

Frequency data for passing and failing the ToM tests in the two groups was analysed using chi-squared methods. Group ANOVAs were calculated to analyse differences between the groups and correlations computed for all the subsections of the tests and measures used.

Group differences were shown with one of the ToM tasks (the more verbally demanding of the two), highlighting the difficulties in interpreting when task behaviour may relate to other confounding deficits (speech intelligibility difficulties, language comprehension difficulties), and case matching is not an option.

The authors conclude that difficulties in expressive language ability and deficits in working memory may explain, in part, the difficulties children with CP showed with ToM tasks. They note that the differences seen may be attributable to deficits in ToM skills, slowed but normal development, or a consequence of the experimental design.

The authors acknowledged the difficulties associated with small group designs, and with use of tests standardised on very different populations. The adaptations made to tests are not discussed in detail, and the validity of some of the measures may have been at risk if the adaptations modified the demands of the test (for example, a verbally presented measure such as the digit span task being presented in a visuo-spatial form).

However, this study presents an appealing design, of a small, focused, well-described index group of children with severe CP and speech intelligibility difficulties, with a matched comparison group and single focus (in this case, ToM) research question. This study is particularly welcomed in that the researchers recognised that, although some of the participants had intellectual disability with age equivalent language skills below the level of their chronological age, the social communication ability (ToM) under scrutiny could be expected to be observed, given the level of intellectual/language development.

Some children with intellectual disability, then, may have additional social communication impairments. This reflection of the "graphic equaliser" examination of component developmental skills described in our introduction might be useful in planning methodology for the current study.

3.4.4 DISCREPANCY DESCRIPTIONS

Recognition of the need to identify ASD profiles in the population of severe/profound intellectual disability has developed only recently (Matson & Shoemaker, 2009), and the assessments in use for this population are largely directed at adults (Matson et al., 1996). In one of the original studies identifying ASD in the population of children with CP, Nordin and Gillberg (1996) noted that many children with severe intellectual disability have ASD, but not the majority, and comment that

Good social competence can be found even when physical and mental functions are severely impaired

(Nordin & Gillberg, 1996, page 310)

More recently, Jordan, writing about ASD in populations of children with identified intellectual disability, noted:

..*it is important to recognise that autism leads to a difference in development, not just a delay, and approaches for children with sld*[3]*, no matter how effective for children without autism, need to be adapted to take account of that difference*

(Jordan, 2013; page 16)

[3] severe learning difficulties (severe intellectual disability)

Jordan argues further for diagnosis of ASD in children with intellectual disability to follow the type of clinical framework presented in 1.3, looking for any discrepancy between social interaction skills and other developmental areas.

It is qualitative differences in these areas of development (personality, level of intellectual and linguistic functioning, experience and teaching) that distinguish autism, but this must be judged against what would be expected for their level of functioning. The more severe the learning difficulties, the less functional behaviour will be expected and there will be a consequent increased difficulty in recognising the autism

(Jordan, 2013; page 5)

This "identification through discrepancy" does seem an important idea to take forward for the discussion of identification of autism spectrum disorder/social communication deficits in children with CP. However, identification of discrepancy of social communication skills will place demands on accurate assessment and description of other, cognitive and language, abilities.

3.4.5 SUMMARY

ASD is now considered to have a multifactorial causation source, including some genetic basis in some cases. Furthermore, the patterns set in place by the child's unusual development shapes the adult-child interaction

The current description of ASD conditions emphasises impaired social communication and/or interaction and restricted and/or repetitive behaviours. Diagnosis for children without visual or motor impairment is made using a combination of standard assessment tools, parental questionnaires and a developmental history: there is no single definitive assessment to support the diagnosis.

Studies aiming to document co-morbidity of CP and ASD have repeatedly noted the difficulties of applying these diagnostic methods to children with severe motor

impairments, who cannot manipulate the toy material involved in standardised assessments, or demonstrate the abilities documented in parental questionnaires.

This restriction will apply, too, to assessments of language and cognition. There may be additional difficulties for children with visual impairments, known to be frequently associated with CP.

There are, then, no identifiable assessment methods for the diagnosis of autism spectrum disorder for children with severe cerebral palsy.

4 METHODOLOGICAL CHALLENGES

This chapter will consider the methodological challenges, both in research design, and in procedures, highlighted in the previous two chapters, by the complexity and heterogeneity of the population of children with CP, and the difficulties associated with characterisation of their communication profiles, including the identification of the social communication deficits associated with ASD.

The study design and procedures selected are guided by the study aims to:

- develop an assessment protocol to support the identification of autism spectrum disorder in children with CP at GMFCS levels IV and V
- compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
- investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

4.1 RESEARCH DESIGN

The choice of study design, it has been argued (Cresswell, 2009), will be determined by a number of factors, including the researcher's experience, the specific nature of the inquiry to be undertaken, and the research methods proposed. The aim will be for the highest level of evidence possible within the constraints of the field of study.

Evidence-based practice (EBP) describes the integration of clinical expertise, of patient/client perspective and of the best available research evidence to inform the decision-making process for patient care. Clinical expertise for SLTs refers to their experience, education and clinical skills. EBP principles are incorporated into the good practice guidelines of both the *Royal College of Speech and Language Therapists*, and the *Health and Care Professions Council.*

However, there is a notorious paucity of research evidence available to support SLTs in their assessment, diagnostic and intervention work. A review of speech and language therapy interventions for children with CP (Pennington, Goldbart & Marshall, 2005) revealed few studies reaching the authors' evidence levels criteria for inclusion. Multifactorial issues in heterogeneous populations, as frequently occur in neurodisability research, may preclude the production of "standard" high-level evidence design such as random-controlled trials: it has been argued (Rosenbaum, 2010) that the "broader low-power view" offered by prospective cohort studies, and longitudinal approaches may offer equally respected understanding.

It is acknowledged that qualitative methods can ask more open-ended questions, may be better able to involve participants, and to concentrate on single issues of enquiry (Fauconnier et al., 2009). Theories and interpretations may emerge from the findings without pre-determined discussion constraints. Such methods, including the use of ethnographic approaches (Wickenden, 2010) and natural settings conversation analysis, have contributed significantly to the knowledge base of study of children with CP. The disadvantages of such methods may be that they will, invariably, involve only small numbers of participants, and so clinical conclusions may be limited.

Quantitative methods may be chosen for data based on performance, with pre-determined aims aimed to test research questions. This is also the method of "table-

top" clinical assessments, which, whilst having limitations of describing a "snapshot" of performance, and often take place in an unfamiliar setting for children and families, endure as a frequently used assessment format.

As an alternative to purely quantitative methods, and in acknowledgement of the complexity of factors and variables involved in the issues under discussion, some research with children with CP has made use of multivariate modelling techniques to identify some of the relations between child (impairment) variables and environmental factors (Clarke et al., 2001). These modelling studies may include large enough numbers for some wider conclusions to be suggested, and may allow, subject to adequate power, the inclusion of a significant number of factors known to influence outcome.

The primary aim of this current study was to explore new ways of assessment to support the identification of the social communication deficits of ASD.

A core principle of any assessment procedure is that it can distinguish those children with and without the disorder in question. In order to develop a procedure to examine the social communication skills (target variable) of children with CP, it would be necessary to compare the group's performance with comparison groups that were matched for key factors (control variables). These key factors, for example, language understanding, would be identified from those variables known to influence communication development, and to be vulnerable in children with CP (Rosenbaum, 2008). This suggests that a between-groups study would be an appropriate method to consider.

To establish that the developed procedure did indeed identify the social communication deficits associated with ASD, it would be important to include, as a

comparison group, children with identified ASD diagnoses. In theory, these children should show specific deficits in the tasks included in such a procedure.

Thus, a group of children with ASD would support the validity status of any assessments used for investigating early social communication skills: it is hypothesised that the group of children with ASD would be more challenged by these measures than by, say, any visual reception tasks. Any assessment used should demonstrate these difficulties clearly in this ASD population to be useful in detecting such deficits in other groups of children.

Furthermore, children with severe CP could be expected to have significant intellectual disability in addition to their physical impairments. In order to exclude this intellectual disability as a confounding variable, the study needed to include a group of children with intellectual disability in the absence of significant motor difficulties and social communication impairment. Children with Down syndrome (DS) represent a group of children whose communication difficulties arise, in the main, because of general slower learning in the absence of marked physical disability. This group might allow scrutiny of the relevance of intellectual disability (performance abilities deficits plus language deficits) on development of early social communication skills.

Children with Down syndrome are characterised by (a range of) intellectual disability, but are also known to show additional developmental difficulties, including social communication difficulties, and speech intelligibility problems. The recognition of DS comorbidity with autism spectrum disorder (DS-ASD) been recognised for some thirty years (Howlin, 1995): more recently the use of standardised diagnostic measures has allowed further understanding of the incidence of DS-ASD. DiGuiseppi and team (DiGuiseppi et al., 2010), from a sample of 123 children (mean age, 6.1 years) reported 18% of the group meeting criteria for a description of autism spectrum disorder, and

7% for autism (definition groups in use at the time). It continues to be the case, however, that difficulties with clinical identification and the implications for both clinicians and parents of “labelling” with an additional diagnosis, makes the true prevalence of DS-ASD challenging (Gray et al, 2011).

The implications of these findings are that, in any comparison group of children with DS, a proportion can be expected to meet the criteria for an ASD, even if this additional description has not been formally discussed.

Nevertheless, data from children with Down syndrome have been used as comparison data in studies of children with ASD: for example, in an interview-based study investigating broader autism phenotype in parents of children with more than one child with autism (Jonge et al., 2015), a comparison group of children with Down syndrome were chosen to try to control for the social effects of having a child with a significant developmental disability.

Similarly, studies have contrasted comparison groups of children with DS and children with CP to include effects of intellectual vs intellectual/physical disability (for example, in a study examining play and symbolic development (Singh, Iacono & Gray, 2014)).

To make such comparison groups useful in the examination of a target skill, the groups would need to be matched with the CP group on key factors related to communication development: studies suggest (Pring, 2004) that these factors should include intellectual ability, comprising language abilities and cognitive performance abilities, and chronological age.

Inclusion of measures on these abilities would exclude alternative explanations for performance on social communication tasks. This would acknowledge the strong

evidence linking social communication and language development (for example; Mundy, 1990): without matching for language abilities, difference in performance on social communication measures would be difficult to interpret.

Similarly, cognitive performance abilities differences between the groups would not allow any conclusions to be drawn regarding differences in social communication abilities.

Matching on a further factor, chronological age, would acknowledge the role of "life experience" in the development of social cognition: it is noted that this influence can be somewhat under-recognised in some of the studies of children with CP in this field (Frisch & Msall, 2013).

The measures used for this matching would need to be appropriate and accessible to all participants. Accessibility might be a specific problem for the target group of children with CP, for whom the use of gaze direction might be their preferred response method. The group are vulnerable to a range of visual deficits, both at the level of the eye and the brain (Deramore Denver, Froude, Rosenbaum, Wilkes-Gillan & Imms, 2016): in order to interpret responses made through looking behaviours, a procedure would need to be in place to give confidence that responses are not confounded by a failure of the eyes to signal choices.

Finally, to address the primary aim of the study, a measure of social communication abilities needed to be identified.

This next section reviews available assessment procedures for these key factor variables: the background, matching, abilities in language and cognitive performance, the assessment of functional vision use necessary for the children with CP, and the

identification of assessment procedures for the target variable of social communication abilities.

4.2 REVIEW OF ASSESSMENT MEASURES

Available, published assessment measures of these background abilities revealed are generally developed for use with (and hence standardised on) children developing typically, who have no sensory or physical impairments. In consequence, their clinical usefulness for children with CP may be limited. Furthermore, as noted, the "umbrella" term of cerebral palsy includes children with a spectrum of physical, sensory and intellectual disabilities across a full lifespan of age range, and it is unlikely that a single, or small number, of assessments would meet all needs.

There is a long-standing concern that traditional cognitive measures are not accessible to children with significant communicative and motoric impairments (Sabbadini, Bonanni, Carlesimo & Caltagirone, 2001). Many of the commonly used communication and intellectual skills assessments make use of detailed pictures, toys and household objects. Some use of objects may be possible with physical adaptations to the material (use of *Velcro,* addition of page-turning devices and so on), but full access to test items is still likely to be limited.

There may also be problems for children with poor speech, if the tests demand verbal responses, and this can be the case even for the sections of tests confined to language comprehension appraisal (Semel & Wiig, 1980).

Watson and colleagues, in their survey of SLT practice for children with CP in the UK, noted responses from participants as follows:

Some SLTs commented that children on their caseloads were too young to complete formal assessments or had significant motor or sensory impairments which prevented them from responding in the manner stipulated by the test. The latter led to the SLTs modifying the tests, e.g. enlarging pictures, cutting up response sheets to allow children to point to the target

(Watson and Pennington, 2015; page 246)

These modifications may apply to all aspects of assessment of the communication profile, and so the various abilities ("sliders" from the graphic equaliser (see 1.3, page 1–18)) are discussed separately in the following sections.

4.2.1 LANGUAGE ASSESSMENT

There has been an interesting clinical debate regarding the plausibility, value and validity of assessment of cognition and language for children with severe disabilities. This arose, in large part, around candidacy models for delivering or prioritising intervention to children with CP, and the discussion of "pre-requisite" abilities needed to access communication equipment and services (Kangas & Lloyd, 1988), especially in the US.

Nevertheless, clinical experience has identified the assessment of language in children with cerebral palsy as a possible and valuable measure for several reasons:

- language is known to be linked to learning potential and social interaction skills, and so supports answering prognostic questions for families and carers
- language development is known to be at risk for children with CP

- all children have the right to be offered challenges at the “next-step” level, and understanding of the child’s current level of development in language can help set “next-stage” targets
- such understanding can also direct intervention for augmentative communication system support, helping match child skills and device demands (McDonald et al., 2008)
- language understanding can often be overlooked as a target for remediation or monitoring
- in the light of the wide range of skills observable in the population of children with CP, and the known barriers to assessment, abilities can often be reported differently by different stakeholders, without a consensus view, or a frank discussion of different views

Furthermore, the assessment of language abilities has been supported by the increased availability of AAC methods for this group of children. Voice output communication aids (VOCAs), and PC-based specialist communication software as part of an assistive communication technology (ACT) system have better allowed children with CP to develop and demonstrate their language abilities, despite any difficulties with speech intelligibility (Smith, 1994).

Nevertheless, difficulties with developing understanding of complex language are evident for many children with CP and the assessment of receptive language is considered an important factor in predicting outcome (Allen, 2008). Language understanding is generally thought to relate closely to overall cognitive functioning for children with severe CP (Kilbride, Thorstad & Daily, 2004).

With this in mind, more studies have attempted to include language skills in their attempts to propose and test classifications to give full speech and language profiles

for children with CP. Hustad and colleagues (Hustad, Gorton & Lee, 2010) suggested four categories to describe a small (n=34) group of children of mean age of 54 months. Inter-rater classification agreement ranging from 74-97% was reported for the four categories proposed:

- NSMI (no motor speech impairment, language either impaired/within normal limits);
- SMI-LCT (evidence of motor speech impairment, language within normal limits);
- SMI-LCI (evidence of motor speech impairment, language impaired)
- ANAR (no speech, language either impaired/within normal limits/not assessed).

This attempt to offer categories of description for both speech and language communication patterns highlighted many of the difficulties of description of skills for this group, with the researchers commenting that assessment of language abilities was difficult for children with severe hand/arm movement difficulties, and that measures of all language functions should be undertaken. Their conclusions included the observation that the development and evaluation of novel tools for measuring language in children with CP was now needed.

Geytenbeek and colleagues (Geytenbeek et al., 2010) made a significant contribution to this need by reviewing the functional use of standardised assessments for language comprehension for children with CP. Their conclusions included suggestions for adapting and modifying existing assessments, and they demonstrated that useful estimates of children's skills in relation to their typically developing peers could be made in this way. Children who were unable to point with their hands to standard assessment material might be able to indicate their responses if the material were enlarged, or presented in a way to allow them to use eye gaze as a response. The

suggestion for use of eye gaze access technology to present assessment material was also discussed, and the researchers concluded that language comprehension tests for children with severe cerebral palsy were scarce, and that a specifically designed language comprehension test was warranted. Their subsequent work included a computer-based, switch-accessible assessment, currently only available in Dutch, but a promising development to tackle this need. However, such computer-based assessments may lack flexibility of administration, and be useful only to children with established access methods.

An examination of the 12 tests reviewed by Geytenbeek's team to be possible for administration with children with cerebral palsy was undertaken, and this was useful. However, of the 12 tests, only the *Preschool Language Scale* (Zimmerman, Steiner and Pond, 2002) was both available in the UK, and appropriate for the age range planned for study.

As noted in the introduction, Watson and Pennington's online survey (Watson & Pennington, 2015) recorded the assessment (and intervention) practices of UK SLTs working with children and young people with cerebral palsy, and related these to recommendations made in current professional guidelines. This document was consulted to support decision-making for selection of language assessment.

80% of SLTs commented that they assessed receptive language *in most cases.* Of those reporting assessment, it is interesting to note that 82% and 72% of SLTs reported using their own assessment schedule for the assessment of receptive language syntax and receptive language vocabulary respectively, reflecting the lack of available measures. In the list of receptive language tests used by 10 or more respondents, two published tests were appropriate for this current study's selected age range (12-54 months), and hence eligible for consideration.

87% of respondents used the *Derbyshire Language Scheme* (Knowles & Masidlover, 1982). This is an intervention scheme, intended for children with a range of language difficulties, and based on a key-word approach. The material comprises toys and household objects, and A4 sized quadrant (four to a page) coloured line drawings. The scheme follows largely developmental lines, but is not norm-referenced. Tasks with single-word understanding relate approximately to a 12-18 month level of skills in neurotypical development.

49% reported using *Preschool Language Scale* (*UK* versions 3/4) (Zimmerman, Steiner & Pond, 2002). This assessment also uses toys/household objects in the early sections, and quadrant/single page coloured photographs and drawings. The currently commonly available version of the test (*PLS-4 UK*) was standardised using data collected on a total of 800 neurotypical children from 12 to 17 months and 24 to 83 months; with a balance of boys to girls of 49% to 51% for children under 10 years old. The test covers the age range 12-65 months (*PLS-4*), and provides age-referenced scores for language comprehension and expression.

For this current study, consideration was also made of the *Battelle Developmental Inventories*, which have a *Communication* section, looking at both receptive and expressive skills, and use toys and picture material, some of which is available as software in an *eKit* (Newborg, Stock, Wnek, Guidibaldi & Svinicki, 1984). This assessment included, in the expressive language tasks, standard recommended adaptations to minimise verbal/motor responses. However, some of the receptive language tasks appeared rather broad in terms of developmental range (for example, *responds to spoken and gestural* commands).

In summary, there appeared to be no obvious assessment of language understanding useful for children with CP. Computer-based assessments are in development

(Geytenbeek et al, 2010), but may not be appropriate for the particular group of children with CP in focus for this study. It appears that any existing procedure will require some form of adaptation, and that this may need to be in both administration and scoring to give useful information.

4.2.2 PERFORMANCE ABILITIES ASSESSMENT

Some studies, largely from the field of AAC, have indicated that, for some children with cerebral palsy, and in the light of motor difficulties restricting access to learning through manipulative exploration, receptive language skills may be a strength on the "graphic equaliser" (see 1.3) relative to other aspects of their intellectual abilities, and may be the best "window" to view learning potential (Ross & Cress, 2006). Conversely, there is also some clinical evidence to suggest that, for some children at least, specific difficulties with some aspects of language, with receptive language lagging developmentally behind performance abilities, may be part of their profile (Gumley, Price & Griffiths, 2011).

However, a study examining the communication profiles of a group of children with Worster-Drought Syndrome, a variant of cerebral palsy in which difficulties with eating and drinking and impairments of speech oro-musculature are the most prominent characteristics, reported no significant differences in performance and language measures (Clark, Harris, Jolleff, Price & Neville, 2010) and this would reflect other findings in the field (Pirila et al., 2007).

It seemed important, therefore, to include an assessment of performance (non-language) abilities in the background measures. There has been limited focus on the study and understanding of the performance abilities of children with CP, and even less examining the individual aspects (attention, memory, problem-solving) of neuropsychological development that may be associated with children with CP. Such information would be made more valuable if it related to individual subtypes (related

to GMFCS score) as it is clear there are significant differences to be seen across these GMFCS groups.

In the Watson and Pennington (2015) paper surveying SLT practice in the UK, therapists who completed assessments of performance abilities reported that they did this through observation, or using schedules they themselves had developed. Therapists who did not conduct these assessments noted that performance abilities were assessed by other members of the team, citing psychologists and teachers. There is an added consideration in selecting assessments of performance abilities for this current study, in that many of the published assessments stipulate specific qualifications for administrators, with restrictions placed on test use which may exclude SLTs. Furthermore, from clinical experience, the assessments used by (educational) psychologists are often based on observation rather than specific test use, and those used by teachers may relate more to scholastic achievement than to individual aspects of performance abilities/potential.

However, Yin Foo and colleagues (Yin Foo, Guppy & Johnston, 2013) conducted a systematic review of assessments of intellectual ability (performance abilities plus language skills) in use for children with cerebral palsy. Their search identified those assessments that measured intellectual function in children with CP aged 4-18 years. Their final analysis included only papers with reported IQ psychometrics for children with CP. Nine assessments were identified in this way: however, the age ranges for which the tests were appropriate were all above two years, and may have had reduced validity for developmentally young children at this age level, or below.

In a study looking at the use of the *Mullen Scales of Early Learning* (Mullen, 1995), (Burns, King & Spencer, 2013), researchers reported utility for this assessment for children with CP and with ASD. All subscales were administered and scored, although

their CP group was small (24/47), reported to be heterogenous, and specific motor level functions, or any adaptations needed for administration were not reported in detail.

There are, therefore, similar difficulties with performance measures to those described for language comprehension assessments. There is a particular paucity of tests available for children below the developmental age of two years, and the range of available tests is further limited by availability to non-psychology clinicians.

4.2.3 FUNCTIONAL VISION ASSESSMENT

As noted in Chapter 1, the clinical impetus for this study arose from the repeated observations, by the author and others in the clinical team, that a significant number of children with cerebral palsy appeared additionally disabled by a difficulty with using gaze direction ("eye-pointing") as part of their communication skills "armoury". For those children with no or little speech, any deficit in using gaze direction as a signal of shared interest to their communication partner appeared highly significant, and often as disabling as their lack of speech. This was made clearer through observation of those children who *could* use their gaze direction in this way, frequently to convey quite complex messages, including the use of AAC-aided communication tools.

Gaze direction is so valuable to children with CP, both in signalling their own messages and in responding to questions and directions. The direction of gaze can show an answer (for example, the child looks at their brother in answer to *who spilled the milk?*), or a question (the child looks to the clock to ask what time the session will end). Looking, especially with some further confirmation, can be used to make choices, and if the child can make use of symbolic material (objects, pictures or orthography), gaze direction can be used to build complex and sophisticated messages.

Furthermore, the ability to direct, fix and transfer gaze is a fundamental skill to demonstrating and sharing focuses of attention, and this joint attention with a conversational partner is the foundation for expressive communication, expressive language development and social interaction.

However, it has been a long-standing clinical concern that visual impairment is not routinely assessed in children with the most severe physical impairments: clinical experience and research evidence indicate that visual impairment is under-reported in children with severe bilateral cerebral palsy who are non-speaking, and that significant visual deficits are often missed or misdiagnosed in this group of children (Ghasia, Brunstrom, Gordon & Tychsen, 2008). This is perhaps unsurprising, given the contribution of cognitive, motor and sensory skills that comprise a visual response.

Indeed, in the author's specialist communication team, there were often (wryly humorous) discussions between the SLT and the developmental paediatrician discussing which assessment needed to take place first: the language assessment to determine which visual assessments would be developmentally appropriate, or the visual assessment to determine which language assessments would be appropriate.

The infrequency of functional vision assessment results, in part, from the failure of available tools that emphasise those aspects of vision critical to communication, and how to appraise them .Where support is available for clinicians to examine aspects of vision, measures often require children to have reached developmental thresholds that can preclude developmentally younger children (for example, the *Motor Free Visual Perception Test* (Colarusso & Hammill, 2003) targeting children above four years) or measures are dependent on children's ability to manipulate objects, point or speak as a response mode (Ortibus, Lagae, Casteels, Demaerel & Stiers, 2009).

Specialist multidisciplinary neurodisability services, as described in Chapter 1, with paediatric staff experienced in assessment of children with complex neurodisabling conditions such as cerebral palsy, are accessible through statutory health services in the UK, but are not numerous, and may not be accessible to all those children who might benefit if referrals are not effected by community staff (who may be unaware of the service, or under budgetary restraint).

4.2.3.1 Visual Functions and Functional Vision

This thesis will argue that SLTs working with children with cerebral palsy need to have access to knowledge to understand any restrictions on children about the utility of the material and strategies offered for communication assessment and intervention.

One central distinction to be made is the difference between **visual functions** and **functional vision.**

Firstly, **visual functions** relate to the eye and the visual system (eye plus brain) itself, and therefore, in ICF-CY terms (see 4–88,) at the *Body Function and Structure* level: visual functions will include (see Table 4-1):

Visual functions (examples)	
Visual function	**Gloss/comments**
visual acuity	refers to the sharpness/blurriness of the image
visual field	refers to the total area in which objects can be seen in the side (peripheral) vision while the focus of the eyes is on a central point
colour and contrast vision	distinguishing colours and a spectrum of contrasts
dark/light adaptation	efficiency of oculo-motor responses to changes in light
stereopsis	relates to the perception of depth interpreted by the brain receiving visual information from both eyes in combination: binocular vision
cognitive visual impairments	this term (also cortical/cerebral visual impairment) although often accompanied by a wide range of different "symptoms", refers, in summary, to any damage to the eye-to-brain pathway. It is often described in terms of "the eyes can see, but the brain is not able to fully interpret what is being seen".

Table 4-1: Visual functions (examples)

In contrast, the term **functional vision** describes how easy/difficult the child finds it to operate in vision-related activities, and thus the ability falls, in ICF-CY terms, within the *Activity and Participation* domains. Functional vision relates to use of vision in daily activities: for children with cerebral palsy, this will include orienting to sound, attention to faces, inspecting and resting gaze, returning gaze to the listener, and eye-pointing.

4.2.3.2 Gaze Direction and the definition of eye-pointing[4]

To this point, the term "gaze direction" has been used to describe the use of looking behaviours in interaction. The use of this term has been to try to describe simply the skill as it is observed. Although the term "eye-pointing" is used widely in both clinical and academic contexts, there is often little consensus about its definition, or about the motor, visual and socio-cognitive behaviours that are necessary to be able to develop use of functional and communicative gaze direction.

Following a review of available definitions in the literature, and based on clinical experience, Sargent and colleagues (Sargent, Clarke, Price, Griffiths & Swettenham, 2013) proposed a description of eye-pointing. The definition emphasised the necessary intentionality of the "speaker" (a deliberate action and an awareness of the goal of communication), and the co-construction of any meaning evolving (both partners having a role to play in establishing intended meaning):

The context-relevant, controlled and intentional use of sustained gaze in order to direct one or more partner's visual attention to any item or object for a deliberate communicative purpose. Other communication modes (facial expression, vocalisation, head movement and body position) may be employed, as available, to support the use of gaze. The intended meaning is established collaboratively between the child and the adult

(Sargent, Clarke, Price, Griffiths & Swettenham, 2013; page 479)

[4] For clarity, this discussion does not comment at all on the use of eye-gaze access technology (sensor mounted on to an ACT device to "read" gaze direction to give hardware/software control). It is possible that efficient use of gaze for interaction might help predict use of such access technology, but this is not addressed in this thesis.

This description suggests eye-pointing to be a complex basket of skills that includes visual function and functional vision, as described on page 4–88, motor and communication abilities. The functional vision skills needed for eye-pointing comprise fixing gaze, disengaging gaze and transferring gaze between objects/people. This fix, disengage, transfer sequence is needed for a "full" expression of eye-pointing, in which the child looks towards, and fixes their gaze on, an item of interest, can then shift their gaze to the conversation partner's face and eyes, and can then return their gaze to the item before once again returning gaze to the partner.

The parallels in this description of eye-pointing with those described as joint attention in 3.3.6 merit some further discussion.

4.2.3.3 Eye-pointing as a substitute for finger-pointing

Some of the previously published definitions and discussions of eye-pointing identified during the exploration of eye-pointing definitions, described on page 4–90, referred directly to an equivalence of eye-pointing to finger-pointing:

a conscious act of pointing with the eyes instead of using the index finger, to obtain an object (proto-imperative) or to inform the partner about something (proto-declarative), a way to conduct a conversation

(Sandberg, Hagberg & Gillberg, 2000; page 258)

and this is particularly seen to be the case for communication with printed material: for example

When speech can't be understood, and using hands or fingers to point is difficult, eye-pointing to pictures, symbols or text can be a fast and effective way of communicating a wide range of messages[5]

In this way, eye-pointing is seen to equate to finger-pointing and should be understood as a deliberate action under voluntary control involving active and purposeful "look-pointing" at a specific target to participate in a communicative interaction. However, as use of vision is so central to human behavior, and as eyes are **generally always open** while awake, there are also situations when the act of 'looking' is not always communicative in the same way as is finger-pointing. When a look is observed, it may indeed be "look to point", but it may also be "look to view" or "look to explore" without any intent to interact. Finger-pointing is generally easy to interpret: interactive eye-pointing may not be quite so straightforward.

To address these issues, Deramore Denver and colleagues (2016) completed a systematic review of visual ability assessments, in the context of activity and participation, rather than body function deficits. This review thus emphasised the need for a description of *functional* v*ision* skills. However, although vision impairment was acknowledged as important to participation for children with CP, the lack of any psychometrically strong measures was identified as a gap in current research and practice.

In summary, although there are a number of tools that include items assessing 'visual ability' at the level of activity/participation, these vary in content, in the context for assessment, and in the skills required of the assessor (Deramore Denver et al., 2016).

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[5] https://is.gd/look2talk (accessed October 2016)

Clinical experience suggests that when assessments of gaze control are carried out by specialist services, professionals working in nurseries and schools can struggle to interpret the results and understand their implications for assessment practice. Attempts have been made to clarify definitions of intentional gaze direction, but currently tools suitable for use by non-vision specialists to assess basic aspects of functional vision in relation to communication do not appear to be available.

4.2.4 SOCIAL COMMUNICATION SKILLS ASSESSMENT

The Watson and Pennington survey of practice reported approaches in use for the assessment of *Communication and Interaction.* For those assessments in use by more than 10 respondents, 98.1% reported using **observation**, with 66.9% using their **own developed schedules**. This was indicative of the paucity of published or available standardised assessment measures in this domain. Of those SLTs confirming their use of assessment in this area, 61.3% cited use of two parent questionnaires: the *Preverbal Communication Scales* (Kiernan & Reid, 1987) (currently out of print but available without charge online) and 47.8% named the *Children's Communication Checklist* (Bishop, 1998) which screens children aged 4-16 for language impairment and/or autism spectrum disorder.

A number of published tests were reviewed to assess these early social communication skills. These were largely checklist/questionnaire-based screening assessments designed to identify children with ASD in the general population. For example, the *Checklist for Autism in Toddlers* (CHAT) (Baron-Cohen et al., 1996) consists of 9 parentally-reported and 5 items reported by health clinicians at an 18-month child development review, as a screening tool to identify children showing early signs of ASD. The CHAT tool had high specificity (97.7%) but failed to identify a number of children who were identified later as fitting the profile of ASD (sensitivity 35.1%). This

sensitivity was improved in modification of the checklist (Robins, Fein, Barton & Green, 2001), and the checklist has been in use by paediatric clinicians since that time.

However, review of the 20 items in this checklist showed that, although some items would be relevant and appropriate to children with physical disability (for example *if you point at something across the room, does your child look at it?*), 10 of the 20 items were not relevant or appropriate (for example, *does your child like climbing on things?*).

Similarly, the *Infant-Toddler Checklist (ITC)* that forms one component of the *Communication and Symbolic Behaviour Scales Developmental Profile* (Wetherby & Prizant, 1993) was also devised as a tool to identify children with ASD. The checklist has three components investigating social, speech and symbolic skills. Parental interview responses identify possible difficulties in emotion and eye gaze, communication and gestures, within the social composite score; sounds and words within the speech composite score, and understanding and object use within the symbolic composite score.

Again, however, the most accessible section, the social composite score, contains items (5/13) that would not be possible for parents of children with poor motor skills to complete. The *Emotion and Eye Gaze* and *Communication* section of this composite did encompass questions that could be relevant to children using gaze direction, for example, *when your child plays with toys, does he/she look at you to see if you are watching?* and *does your child let you know that he/she needs help or wants an object out of reach?*

The *Early Social Communication Scales* (ESCS) (Mundy, 1986; Mundy et al., 2003; Seibert et al., 1982) assessment was reviewed.

This assessment evolved from a research procedure using videotaped recordings of three categories of early social communication behaviours: joint attention, behavioural requests and social interaction. Joint attention behaviours were categorised further (see 3.3.6) into initiating joint attention; noting the frequency with which a child uses eye contact, pointing and showing to initiate shared attention to objects or events, and responding to joint attention, referring to the child's abilities in following the researcher's line of gaze and/or pointing gestures.

The behaviours observed under the heading *Behavioural Requests* also had initiating and responding aspects. The initiation tasks observed the child's strategies using eye contact, reaching and pointing to obtain an object from the researcher, while the response tasks looked at the child's skill in responding to the researcher's verbal or gesture-based commands. The third component of social interaction items noted the child's skill at initiating turn-taking conversational sequences and the ability to engage in teasing with the researcher, together with some imitation tasks (clapping, pointing).

The toys and other materials used in the assessment were selected to engage young children, and so would to elicit social interaction, joint attention, and/or behavioural request. This material included wind-up toys, a balloon, car, book and ball.

The child's responses are then coded from observation and videotaped material. The clinical assessment derived from the research procedure was not standardised, but detailed coding advice is given, an example of which is shown in Table 4-2:

Early Social Communication Scales (ESCS)				
Behaviour	**Level**	**Code**	**Tasks**	**Description**
iJA	Lower	ALTERNATES (REFERENCES)	OBJECT SPECTACLE	• child alternates a look between an active object spectacle and the tester's eyes typically when an object is active on the table or in the tester's hands but also recorded if child looks up to tester after an object becomes active in own hands
iJA	Higher	POINTS	OBJECT SPECTACLE; BOOK	• Before tester has pointed: child points to an active toy OR child points to pictures in book OR child points to wall posters may occur with or without eye contact

Table 4-2: Examples of coding from *Early Social Communication Scales*

Again, some, but not all, of the task items in the test could be demonstrated by children using very limited physical skills or gaze direction as a response, and the toy material described would indeed appeal to many young, and developmentally young, children. The procedure relied on careful coding by individual users, taking significant time to complete, but had moderate to good levels of inter-rater reliability.

The ***Communication and Symbolic Behaviour Scale*** **(CSBS)** (Wetherby & Prizant, 1993) aims to identify children who are at risk of communication impairment, and measures children's communication, expressive speech and symbolic behaviours in part through a series of interaction "temptations", such as wind-up toys. The test is norm-referenced for very young children aged between six and 24 months, and assesses communicative functions, gestural communicative means, vocal communicative

means, verbal communicative means, reciprocity, social-affective signalling, and symbolic behaviour.

Finally, the socio-cognitive battery from ***Very Early Processing Skills*** (*VEPS*) (Chiat & Roy, 2008) was examined. The *VEPS* assessment was devised to target early phonological and socio-cognitive skills to discriminate the characteristics of language disorder from those of ASD, and offered a quantitative set of measures of these skills in young children aged 30 to 42 months who had been referred to speech and language therapy services. The socio-cognitive measures had been shown to be predictive of both language and social communication outcome in their group of children of this age: the procedures had merited close examination for this study as the task items needed only non-verbal responses, without any object manipulation[6].

VEPS-ESC measures (the section of the *VEPS* test looking at early socio-cognitive skills) looked at three sets of socio-cognitive skills, which were then combined in this original study to give a composite score. These sets were **social responsiveness**, **joint attention** and assessment of **symbolic understanding**.

The social responsiveness set of tasks was based on a procedure developed by Sigman and colleagues (Sigman, Kasari, Kwon & Yirmiya, 1992). This study compared the responses by children with ASD, children with intellectual disability and neurotypical children to an adult's face showing emotion (distress, fear and discomfort). The neurotypical children, and those with intellectual disability were very attentive to the adult face for all three of these conditions. Children with ASD gave significantly less attention. Few children, in any group, displayed facial affect themselves.

[6] https://is.gd/EarlySociocognitiveBattery (accessed September 2016)

Chiat and Roy included this task as the first skills in *VEPS,* in which the researcher acts out several scenes in which six emotions are portrayed (hurt, surprise, frustration, anger, distraction and achievement). The child's **response to the researcher's emotional expression** is recorded, and scored by the child's looks to the researcher's face; either fleeting (less than two seconds) (allocated 1 point) or sustained (for at least two seconds) (allocated 2 points). The task is for the child to *notice* and *respond* to the emotion portrayed.

These scenes are supported by researcher script guidelines[7:] for example, for the facial expression of *surprise,* the researcher finds a nappy in the toy bag and says

What's this? It's a nappy. That's not a toy! Let's see what else is inside our toy bag

Secondly, the researcher presented a game **offering opportunities to engage in joint attention**. Six plastic eggs were displayed, one at a time, and the researcher opened them to reveal a small object, such as a tiny bag. Larger versions of these objects were placed to the side, front and back of the child. The researcher noted the child's transfer of gaze from the egg to the researcher's face, or from the tiny object in the egg to researcher's face, and if the child followed the researcher's gaze of direction towards the larger object, or, failing this, could follow the researcher's finger-point to the object. The researcher offered verbal prompts again: for example

Oh look! Here's some eggs. I'm going to look at this one

[7] https://is.gd/EarlySociocognitiveBattery (accessed September 2016)

As the researcher looked at the child, the researcher shook the egg to one side at arm's length, without speaking. The researcher waited up to five seconds to see if the child could look from egg to researcher. This was described as "gaze switch".

The researcher then opened the egg slowly, looking at the child's response, and, again without speaking, showed the contents to the child. The child's look to the researcher's face was recorded at this point if it occurred. The toy is returned to the egg, and the adult then says

I've brought my person with me today

and looks in the direction of the larger matching object. If the child failed to respond to follow the researcher's line of sight, the researcher repeated the comment, accompanied this time by a finger-point towards the larger object. This was described as "gaze monitoring". These "presses" (prompts) were opportunities to respond to requests for joint attention.

A score of 1 point was awarded if the child looked towards the researcher when the egg holding the toy was selected and opened, and 2 points if the child followed the researcher's eye gaze transfer and verbal statement in reference to a corresponding object in the room, or 1 point if the child followed the researcher's point and the researcher's repeated statement about the object in the room.

Finally, an assessment of **symbolic comprehension** was included, in which children were asked to match common objects (for example, *soap*) with "stand-in" symbols (for example, *wooden block*). This section was not examined in detail as the target skills fell outside the study brief.

This *VEPS-ESC* study was encouraging, as it had clear possibilities for administration for children with physical impairments using gaze direction as a response.

The studies concentrating on description of ASD/social communication deficits in children with other identified primary conditions have also faced assessment challenges, and perusal of the selection of tools in another population was helpful. In the development of a screening procedure for ASD in children with severe visual impairment (Absoud, Parr, Salt & Dale, 2011), the authors developed an assessment schedule specifically for this group.

The assessment took the form of an observation schedule, with three domains; social interaction; communication and language; play, and routine behaviours and interests. Again, some of the items tested were specific to the population, focusing on the use of language, and hence less applicable for the target group of this population, but it was encouraging to see that knowledge of ASD characteristics could be applied to the assessment of such characteristics reliably in different populations. Furthermore, the team had developed a "table-top" direct assessment of social communication abilities which aimed to develop a clinical tool for use by others, and for use in unfamiliar (to the child) settings. In this way, the examination of social communication abilities did not depend on interview/questionnaire material, but also included appraisal of demonstrable "in-child" skills.

The balance of direct and indirect (interview/questionnaire) methods, and the need to appraise abilities in both unfamiliar and familiar settings as part of investigations of ASD is stated in the NICE ASD management guidelines'[8] focus on comprehensive,

[8] https://is.gd/NICEguidelinesASD (accessed September 2016)

holistic assessment might represent a further methodological challenge for the present study.

Assessments such as the *Pediatric Evaluation of Disability Inventory* (PEDI) (Haley et al., 1992) aimed to provide a more holistic, function-based approach to assess children with disabilities. The PEDI, a parent-carer interview/ questionnaire, preceded the revision of the ICF documents, but used the same framework to evaluate what children *do* in daily life rather than concentrate on their impairments of body structure and function. The *Functional Skills* section of the PEDI gives summary scores reflecting a child's range of daily life skills in three domains (*Self-Care*, *Mobility,* and *Social Function*). There is a separate *Caregiver Assistance* section that provides a summary of the extent to which a child can be independent in task performance.

Although the PEDI contributed to the view of the assessment process as appraising a child's skills in context, critiques have referred to the length of administration (although a computerised version is available, *PEDI-CAT* (Haley et al, 2009)), which may mean that it is not feasible in full form as a clinical tool in a multi-assessment battery. The assessment tasks are not all relevant to children with physical impairments, as the measure aimed to cover a wide range of paediatric disabilities. However, the PEDI task items are concentrated at the earlier end of the developmental continuum, and so may be most suitable for children with moderate to severe intellectual disability. The PEDI items are focused primarily on home-based activities, which may create some difficulties for clinicians to answer questions without parent/carer input. Furthermore, it has been reported that the *Social Function* domain, although identifying language delay in children with CP, appeared to be less sensitive to the impact of speech intelligibility deficits on social function (McFadd & Hustad, 2013). A sample *PEDI-CAT*: *Social Function* form is shown in the appendices (11.3) completed for a child in GMFCS V, aged six years, who does not use any recognisable words of speech, and this sample does demonstrate that the interview schedule may contain several items that are not

relevant to children with this profile of severe CP, despite the *PEDI*-CAT's having full availability of initial background information prior to starting the questionnaire.

The assessment of social communication abilities for children with CP appears to have had little attention. Where SLTs reported assessment of such abilities, it was largely through the use of carer questionnaires: this information has the disadvantage of being subjective, and heavily adapted to take account of the child's physical disabilities. Furthermore, parents and carers, whilst they have the most detailed experience of their children, may not have training or experience in systematic and objective observation of communication behaviours (Carter & Iacono, 2002). This might suggest that direct face-to-face assessment of these abilities may be more useful in addressing the research aims of this study.

4.3 DECISIONS FOR DESIGN AND PROCEDURES

Given the above methodological challenges, the following decisions were made regarding the design and procedures for the study, in order to address the study aims:

- to develop an assessment protocol to support the identification of autism spectrum disorder in children with CP at GMFCS levels IV and V
- to compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
- to investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

4.3.1 SELECTION OF RESEARCH DESIGN

The study questions centred on an aspect of child development (social communication difficulties) and its relationship to other variables of interest as they exist in defined populations of children with disabilities. Thus a matched/between-groups study was indicated as fit for purpose. This would allow comment as to whether the groups differed significantly in performance on the target variable. Participants would be recruited from a convenience sample of children from consenting volunteer families. Three groups of children would be recruited: the index/target group of children with CP, and two comparison groups: children with Down syndrome, and children with autism spectrum disorder.

This design had the advantages of low ethical risk (all participants would follow the same procedure), and low cost (no repeated assessments). The disadvantages would be that, with small numbers of participants in each group, any outcome discoveries would be likely to be speculative, and that appropriate comparison groups might be difficult to identify and to match, and group sizes might be difficult to control.

4.3.2 SELECTION OF PARTICIPANTS

Selection of the target group of participants (children with CP) merited specific attention, in the light of the heterogeneity of the group, and the difficulties in interpreting findings from published studies where participants had wide ranging characteristics, or characteristics were not well-defined. This lack of adequate description had been noted for studies investigating the use of AAC (Pennington, Marshall & Goldbart, 2007), and recommendations were made in this paper for reporting participants more accurately. These guidelines were used in the decision-making around selecting participants for this current study.

Within the group of children with cerebral palsy, it was decided to concentrate on those children most frequently seen in the researcher's clinical population (see 1.2): those children with CP who were non-walking (GMFCS Levels IV and V). Shevell and colleagues had noted that

the burden of comorbidities falls disproportionately on those children with spastic quadriplegic, dyskinetic or ataxic-hypotonic variants and GMFCS level IV or V functional limitations
(Shevell, Dagenais & Hall, 2009; page 2095)

and published studies of communication development in general, and social communication development in particular were not widely seen for this group of more severely affected children.

For the target measure, two comparison groups were selected: a group of children with Down syndrome, and a group of children with ASD. In this way, both within (CP) group, and between group analyses could be performed. The inclusion of the group of (the motor able) children with DS would allow some examination of the role of physical disability, and the group of children with ASD would allow comparison with a group of children characterised by deficits in the social communication skills under study.

Selection of participants with CP merited careful and detailed attention, to ensure that the background characteristics of the CP group were as defined as clearly as possible, and the results of the target measure could be interpreted with confidence.

4.3.3 SELECTING APPROPRIATE AND ACCESSIBLE ASSESSMENT METHODS

In selecting aspects of communication to be included in the description of participants, and the methods by which to assess these aspects, reference was made to several sources:

- the role and responsibilities of SLTs working with people with CP, as described in the practice guidelines of the *Royal College of Speech and Language Therapists*
- guidance from the International Society for Augmentative and Alternative Communication UK (Communication Matters) single case study template[9]
- discussion documented by Pennington (Pennington, Marshall, & Goldbart, 2007) concerning how to describe (AAC) participants in research and clinical practice

For the scope of this study, the focus of communication profile description had to be narrowed, to ensure relevant skills were assessed in a way that would be able to approach the questions posed.

The decision was also made to concentrate on clinic- or school-based "in-child" testing measures, in preference to parent/carer questionnaire/interviews. This was a difficult "rejection", as family perspectives on behaviour and concerns are central to clinical discussion of communication profiles. However, as the behaviours under scrutiny (social communication abilities) were not fully documented in this group of children, it did seem important to complete first-hand observations as a priority.

It was also important to include a background measure of functional vision: the researcher would need to have confidence that if the children had difficulties with

[9] https://is.gd/CMCaseStudyTemplate (accessed November 2016)

looking between objects/pictures, this was not a physical/motor deficit, but could be more confidently attributed to failure to complete the target task. As no published task was identified for this purpose, a *Functional Gaze Control* measure was devised (described on page 9–197).

Assessment of **performance abilities** *without* mediation of language would be included to reject the assumption that receptive language/visual processing skills proceed in developmental tandem in this group. The *Mullen Scales of Early Learning, Visual Reception subscale* offered the features described as needed (see page 5–120 for full description).

For **language testing**, in addition to the decision to use an "in-child" measure, the decision was also made to assess receptive language development *only.* Although the expressive language skills of the target CP group were of interest, their potentially wide-ranging modes of communication (body movement, facial expression, vocalisation, gaze direction, partially intelligible/unintelligible speech attempts, printed symbol use, assistive communication technology use) precluded the inclusion of any available methodology for expressive communication measures.

Assessments of single word vocabulary, although easy to administer, were rejected in the light of increasing suggestion, both from clinical experience, and from published studies, that children with CP may have single word vocabulary skills in advance of their abilities with "full" comprehension requiring understanding of grammar and auditory memory (Sutton, Soto & Blockberger, 2002; Hustad et al, 2010).

Picture-only assessments were also rejected, as the tests using toys/manipulatives might offer more immediate symbolic understanding and increased motivation to the developmentally younger children. The assessment would need to cover the range for

the targeted developmental level (12-54 months). This range was chosen to offer opportunities to highlight any "gaps" between language/performance abilities and those of early, foundation social communication skills, expected in neurotypical development at 6-12 months.

The *Auditory Comprehension* domain of the *Pre-School Language Scale* (UK version*)* was selected to meet these criteria, and is described in detail in 5.2.4, page 5–121.

These two measures (receptive language abilities and performance abilities) would allow comparison with a third set of abilities; namely, the social communication abilities under scrutiny. These abilities should, for individual children, and if following typical trajectory, parallel the levels of development observed in the language/performance abilities.

The language/performance abilities were to be based on modifications to existing assessment procedures wherever possible, to maximise replicability for other clinicians.

From the range of early social communication skills, the study would focus on **social responsiveness** and **joint attention**, identified as alerting skills to persisting deficits in social communication. The difficulties with questionnaire methods had been noted: again, no fully appropriate measure had been identified through the literature search, and so an in-child target measure was compiled from items from a published study assessment, *Very Early Processing Skills,* (Chiat & Roy, 2008) together with some novel supplementary task items. This procedure is described in detail in Chapter 7.1 (page 7–138).

4.3.4 SUMMARY OF DECISIONS

The design selected was a matched group, between-group, cross-sectional (one time point only). The target group of children with CP would be those children with more severe motor disorder (GMFCS IV/V): comparison groups of children with Down Syndrome and autism spectrum disorder would allow comparison of profiles of social communication skills of children with CP with those seen in children known to have difficulties in this area (ASD) to contribute to the validity of the novel measure proposed to examine social communication skills. The choice of children with DS as a comparison group would allow inspection of the role of intellectual disability in the absence of significant physical disability on outcomes.

Group matching would be supported by inclusion criteria, to narrow the differences across groups in chronological age, and in intellectual ability. In order to match the groups on language and performance cognition, the following tests were identified as appropriate: *Auditory Comprehension* domain of the *Pre-School Language Scale* (UK version*)*, and *Mullen Scales of Early Learning, Visual Reception subscale.* These tests, and the adaptations needed to administer and score them for use with children with CP, are described in detail in 5.2.2.

No suitable measures of functional gaze control or social communication skills were identified, and so assessments would need to be devised for both these areas of testing.

These procedural decisions are summarised here (Figure 4-1):

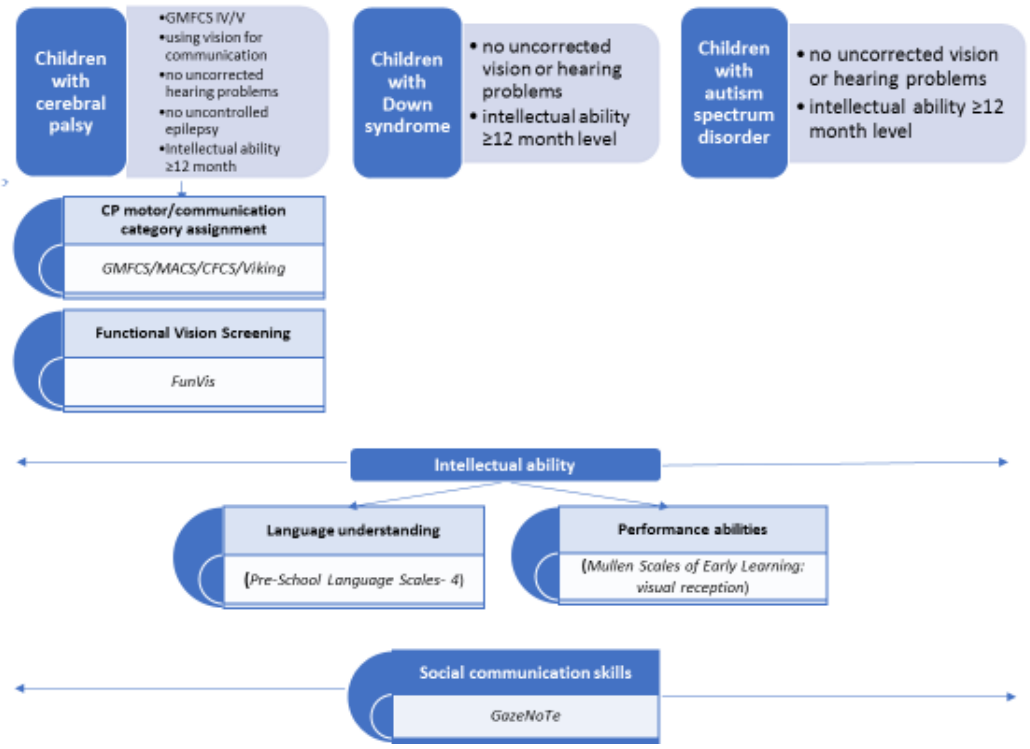

Figure 4-1: Summary of participant groups and selected assessment procedures

5 SELECTION OF THE CP GROUP

Following the decision-making processes described in the previous chapter, this chapter describes the recruitment of children with CP *only*, and the approaches taken to testing this group of children against the inclusion/exclusion criteria. This first step was felt necessary to ensure that the group of children with CP could be characterised sufficiently for the selected participants to be included in the comparison study.

This section also discusses the choice of specific assessments for background measures, and the adaptations made to those assessments in the light of specific challenges for children with CP.

5.1 RECRUITMENT OF CHILDREN WITH CP

Participants for all three groups of children (the target group of children with CP, and two comparison groups of children with Down syndrome, and children with autism spectrum disorder), were recruited in the same way, under the same ethical review.

The study's aims and methods were given full ethical review and were approved by NHS Health Research Authority (London Hampstead Committee), reference 12/LO/1243, and University College London (UCL)'s Ethics Committee, reference 1328/005. The study was also registered with the *Joint Research and Development Office* of the author's children's hospital workplace.

Participants were recruited in two ways:

- from relevant clinical lists at the author's children's hospital workplace
- from schools in London, South East England and Sheffield, South Yorkshire. Recruitment was conducted through researcher contact with the children's NHS speech and language therapist (SLT), who in turn approached parents with invitations to participate and information sheets (see 11.4.1, and 11.4.2). Consent forms (see 11.4.3) were completed by parents and forwarded to the researcher prior to the testing sessions.

The community speech and language therapist supporting the recruitment received written details of inclusion criteria for children with CP as follows: (criteria for inclusion are shown for comparison groups in Chapter 6).

Criteria for inclusion (children with CP)
• 4-limb (bilateral) cerebral palsy requiring wheelchair use (GMFCS categories IV and V) • chronological age 36-144 months • language understanding/intellectual ability at 12-54 months) • using, or expected to use, functional vision in communication • hearing levels adequate for speech recognition • epilepsy, if present, described as controlled

Table 5-1: Inclusion criteria (CP group)

Only children with the more severe motor disorders (GMFCS IV/V) were included in the study. There is strong clinical and theoretical motivation for focusing on this group: for example, these children have greater intellectual disability (Shevell, Dagenais & Hall, 2009), more limited life experience and interaction opportunities (Parkes, 2010), both factors that may impact on the development of social communication. These groups (GMFCS IV/V) were also the focus of clinical concern, and frequently reported as difficult to assess.

Age range criteria were selected to include children who were chronologically old enough to suggest that early social skills development might be identifiable: a primary school year range was chosen to reduce the heterogeneity of this CP group. Similarly, a minimum 12 month developmental/intellectual level would again suggest that the early social skills targeted could be expected to be in evidence. The two ranges together (chronological and developmental age) would allow inclusion of the groups of children with moderate-severe intellectual disability, again reducing the variability in the group under study.

The inclusion criterion for functional vision for communication was essential for this group of children who use direction of gaze for communication, and would be likely to use this method of giving responses in assessment of language, cognition and social communication skills.

Adequate hearing for speech would be a pre-requisite for interpreting language comprehension findings, and, again, exclude a further factor (hearing impairment) known to influence social communication skills. Similarly, children with uncontrolled epilepsy were not included, as the influence of continuing seizures on brain development is well-documented (Pal, 2011).

In this way, a total of **66** families of children with CP consented for their children to take part in the study.

5.2 BACKGROUND MEASURES

This section describes how the inclusion criteria were reviewed by the author for this group of 66 children with cerebral palsy. Three background measures were chosen, to describe participants adequately, and to allow further analysis of relationships between abilities within the group. General and specific adaptations of test material are discussed.

Firstly, a measure of **functional gaze control** was developed, looking at fixation and transfer of gaze, to have confidence that participants were not limited in their gaze direction responses. An appraisal of functional gaze control seemed essential to ensure that children's performance on any tasks requiring responses via gaze direction was not confounded by the possibility that these children, with severe physical disability, lacked the gaze control skills to respond.

Secondly, a measure of **performance abilities**, based on visual rather than language skills, was identified. A third measure of **language understanding** was also included. These measures were used as background for all three groups (CP, DS and ASD).

5.2.1 FUNCTIONAL GAZE CONTROL

As noted, this assessment was devised, for the CP group only, to ensure that any later failure in tasks requiring gaze direction as a response could be more clearly interpreted, and reduce the possibility that motor movement control was responsible for poor performance on tasks with gaze direction responses. It was important to understand that the group of children with CP had the ability to use gaze in non-social settings, before testing use of gaze in social contexts (Swettenham et al., 1998).

Two functional gaze control skills, identified as essential for the use of gaze direction/eye-pointing in communicative exchanges, were identified. The *Functional Gaze Control* assessment aimed to give an objective view of the abilities of participants to **fix their gaze** on objects and to **transfer gaze between two objects.** The procedure developed used a behavioural observation protocol, constructed of readily-available materials, to be as replicable and useful as possible to clinical and educational staff. Accordingly, care was taken to ensure that learning to use the procedures would not require specialist or extensive training. A separate project, not reported here, compared results from this protocol with an objective measurement protocol using eye gaze tracking technology (Griffiths, personal communication).

The tasks presented were designed, with guidance from specialist developmental paediatrician and paediatric optometrist colleagues, to be accessible by children with normal to moderate levels of visual acuity (6/6 to 6/60; *Snellen* scale[10]).

The design was based on a procedure developed by developmental paediatricians: the *Stycar Graded Balls* test had been used to measure visual acuity in very young children (Sheridan, 1973). In constructing the *Functional Gaze Control* measures for this study, the design used seemed appropriate to consider, after adaptation, for the functional gaze control assessment tasks for developmentally young children with physical disability.

The original acuity test used plastic white balls of varying diameters (from 0.3cm to 6.4cm) mounted on slender black sticks, and presented against a black background.

[10] https://is.gd/snellen

Visual acuity measures were reliably assessed on documenting the child's response to increasingly smaller stimuli (Figure 5-1).

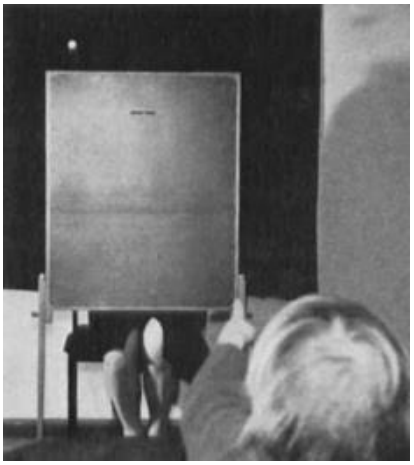

Figure 5-1: *Stycar Graded Balls* visual acuity test

In the *Functional Gaze Control* measures for this study, the materials used comprised several coloured targets of 5cm diameter, presented at 1m from the child's eye-line, and shown, on black sticks against a black background (1m x 0.75m). A hole in the centre of the board allowed the examiner to observe the child's gaze direction, and, during the reliability phase, two observers were positioned (one) to the left and (one) to the right of the board to do the same. This construction is shown in Figure 5-2:

Figure 5-2: Functional Gaze Control Screening Materials

5.2.1.1 Fixation of Gaze

This first task examined the child's ability to orient towards, and then to fix gaze on, five stimulus targets, presented singly in different positions against the background (centre, top, bottom, left right). Only one stimulus appeared at a time, and each was presented twice in each position, and was withdrawn before a new stimulus was presented. In this, and all three tasks, children were oriented, as needed, to the appearance of the targets with simple instructions such as *oh look!*

Responses were recorded as ability to fix gaze (*yes/no*) by the examiner behind the board, and observers to the sides. A score of 1 was given if the examiner/observer had confidence in their identification of fixation. This would not include any "fleeting" (less than one second) fixations.

There was a total possible score of 10 for this task. Discussion with vision specialist colleagues suggested that children who achieved 50% or more on this task were considered to have shown adequate fixation to use this skill as a response in tasks.

5.2.1.2 (Fixation and) Transfer of Gaze

This second task examined the child's ability to inspect two targets, and to shift gaze between the two. When the child's fixation on a target presented centrally had been confirmed, a second target was presented on either the left or the right. The central target remained during the presentation of the second target.

The second target was presented twice in two locations (left or right of the initially presented target). A score of 1 was awarded for gaze transfer from the fixation on the initial target to fixation on the second target on *both* occasions of its presentation. A

score of 0 (zero) was given if transfer occurred only once or not at all. Again, the examiner and two observers recorded these scores.

There was a possible score of 8 for this task. Again, discussion with vision specialist colleagues suggested that children who achieved 50% or more on this task were considered to have shown adequate fixation to use this skill in response tasks.

Performance measures on these two tasks, then, would confirm candidacy for further background measures testing for the children with CP. This was essential in order for the researcher to have confidence to interpret any success or failure on assessment measures of receptive language and intellectual ability.

Further confidence in interpreting performance on these measures rested on good accessibility of all material and methods used in administration of the language and intellectual tasks, and in careful documentation of any modifications.

5.2.2 ADAPTING ASSESSMENT MATERIAL FOR CHILDREN WITH CEREBRAL PALSY

The results from the *Functional Gaze Control* tasks allowed inclusion of those children who appeared to have adequate functional vision skills to use gaze direction as a response.

For the intellectual and language ability assessments, it seemed essential, as noted, to exclude, as far as possible, any confusion regarding poor performance: if, for example, a child failed to identify *cat* from an array of four pictures, the researcher would only have confidence that this was a failure to understand the word if other possibilities (difficulty with seeing the item, pointing to the item) had not been excluded.

Researchers assessing language comprehension (Yin Foo, 2013; Geytenbeek, 2007) had suggested that adaptations to available test material fell into two categories: **accommodations**, which may not affect a standardised procedure and so would allow continued use of any norm-referenced scores, and **modifications**, which alter the test items enough to risk rendering any standardised scoring invalid.

The development of the *Functional Gaze Control* measure allowed consideration of the test accommodation to use **eye-pointing as a test response**. Including only those participants with positive findings from the functional gaze control measures would give confidence that gaze direction would be an option for use as a selection response in assessment. Eye-pointing is an accepted method of response for children with physical disabilities. For any of our measures involving picture/object selection, decisions were made about establishing confidence for the observer(s) in the child's eye-pointing. It was likely that few children would be able to give clear finger-pointing responses to pictures/objects, and it was, therefore, decided by the author, based on clinical experience, that gaze direction/eye-pointing would be accepted as a response for children where gaze direction met the following conditions:

- spontaneous gaze direction had been observed as a strategy used by the child consistently during the warm-up activities and/or conversation.
- where there was any doubt concerning the intentionality of the gaze direction as a pointing strategy, a second "unsighted" observer reported on the child's direction of gaze towards the target. This observer was naïve to the position of the target items in the child's view, and the observer was positioned behind the child, and facing the examiner. In this position, this observer could give unbiased commentary on the direction of the child's gaze, and indicate the level of observer confidence that a choice had been made by the child. This

strategy had been used successfully in clinical situations for some time (Figure 5-3):

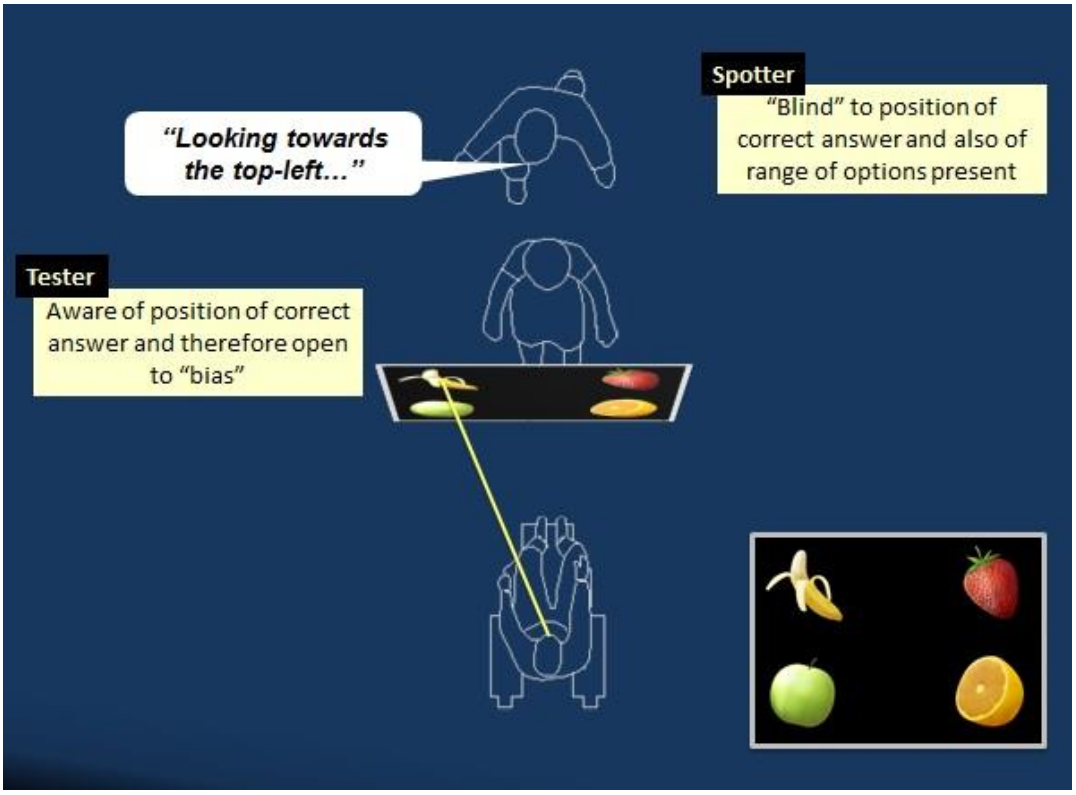

Figure 5-3: Using gaze direction as a selection response

Further modifications were made by the author as follows for all tests. Where picture/toy material was available for children with an observable and reliable response, (verbal or gestural), to convey confirmation and denial (*yes* and *no),* this was accepted as a response.

In addition to the use of gaze direction, for these children, use was made of **auditory scanning** techniques as follows. The examiner gave the test question/command; for example, *show me the phone*, and then the examiner pointed to each array item in turn, with the prompt *is it this one? is it this one?* using carefully uniform intonation. Again, this technique is in general use for this population, but was used for all tests as a modification by the author to standard procedures

5.2.3 MULLEN SCALES OF EARLY LEARNING (VISUAL RECEPTION SUBSCALE)

This standardised, norm-referenced assessment was selected as a measure of performance abilities that could meet the stated criteria for utility for this study's aims.

The *Mullen Scales of Early Learning* (Mullen, 1995), an assessment of cognitive development, were standardised for use with children from birth to 68 months. The test comprises five scales: *Gross Motor, Visual Reception, Fine Motor, Expressive Language, and Receptive Language.* The assessment materials are both toys/objects and picture material in black-and-white (example pages are shown in 11.5.1 for illustration).

The *Visual Reception* subscale gives a measurement of the child's ability to process non-verbal information using shape recognition, patterns, visual memory and visual sequencing. The skills shown are largely non-reliant on the understanding of language, and, again, could be adapted for non-speaking children with motor problems to give responses. Age equivalent measures in months can be calculated from the scores on subscales.

5.2.3.1 Mullen VR specific modifications

There were no guidelines to adaptation for children with motor impairment in the manual of the test. The following modifications were, therefore, made by the author:

To facilitate reliable responses from the participants in the CP group, the **picture material,** black-and-white line drawings, in this assessment was **enlarged** from the original test material, in A5 sizing (148.5 x 210mm), to A4 format (210mm x 297mm). With this adaptation, the minimum visual acuity requirement to engage with all this

material was evaluated as 2/60 (*Snellen* scale[11]), a measure categorised in the severe visual acuity impairment range, by a specialist paediatric optometrist.

Despite the adaptations proposed, a small number of the items (5/33, 15%) in the *Mullen VR* had to be **e**, as they were highly dependent on motor manipulation (see 11.5.2 for details). This was considered as a modification to the test, and interpretation of test scores would need to proceed with caution.

5.2.4 PRE-SCHOOL LANGUAGE SCALE (*PLS-4*)

Decisions concerning the approach to language testing were informed by the reports of tests used in other work and by clinical experience of assessment that had been useful in the clinical setting. A further decision was made to select a direct, "in-child" assessment method, rather than a parent/school questionnaire, to reduce observer, recall and reporting bias.

A number of published tests were considered (4.2.1, page 4–79), and the test most able to meet the challenges identified was the *Auditory Comprehension* subscale of the *Pre-School Language Scale* (4th edition, UK version) (*PLS-4*) (Zimmerman, Steiner & Pond, 2002).

The *PLS-4* is a formal and standardised assessment, constructed for use with children from birth to age 65 months. It can be administered and interpreted by speech and language therapists and psychologists. The test aims to assess language development, in both receptive (*Auditory Comprehension* (AC)) and expressive (*Expressive*

[11] https://is.gd/snellen

Communication (EC)) domains, and to identify children who have language deficits, both quantitatively (they show reduced abilities in comparison to their same-age peers) and qualitatively (any errors can be analysed in terms of their presence in typical development/atypicality).

The test addresses a wide range of skills associated with language development, including attention, gesture, play, vocalisation, social communication, vocabulary, concepts, language structure and emergent literacy. The materials used are toys and coloured pictures, with both direct ("table-top") testing and some indirect (observational) methods employed.

The *Auditory Comprehension* tasks assess the comprehension of more than single-word vocabulary, including tasks requiring the understanding of verbs, adjectival concepts and grammatical markers. The *Expressive Communication* subscale addresses speech, language output and social communication: in the light of the tasks relying heavily on the use of voice and speech, and for the scope of this present study, administration of this EC subscale was not thought to be practicable for this project involving children with cerebral palsy.

PLS-4 offers norm-referenced test scores for both subscales, and these subscales (as well as a *Total Language Score,* when both subscale results are available), will yield standard scores, percentile ranks and age equivalent scores.

Testing materials include toys which are within the standard range offered to pre-school children from a range of cultural backgrounds.

The *PLS-4 Auditory Comprehension* subscale has been used in a number of studies involving children with Down syndrome (Næss, Lyster, Hulme & Melby-Lervåg, 2011) and autism spectrum disorder (Harris, Handleman, Gordon, Kristoff & Fuentes, 1991).

Furthermore, the *PLS-4 Auditory Comprehension* subscale has been used with some success with other groups of children with cerebral palsy: in Hustad's study (Hustad et al., 2010) identifying classification groups of children with CP, 11 of the 34 subjects could complete the *PLS-4 AC* assessment with only "some adaptation". The authors note, however, that some items were completed via parental interview, and that for those children with significant motor impairment, it was, on occasions, difficult to discern if failure on tasks was a result of lack of verbal understanding of the item, or because the adaptations were insufficient to allow the child to respond.

The Examiner's manual of the PLS-4 UK assessment offers some guidelines on use of the test with children with severe physical impairments (Zimmerman, Steiner & Pond, 2002). Some accommodations are suggested: for example, for pointing with finger or full hand, the manual suggests offer adult physical support (in a limited way, and with care to avoid possible authorship issues), additional response time, and careful positioning of child and test material.

Several modifications are also suggested, including the auditory scanning techniques described on page 5–119, and use of the child's *yes/no* responses if these are judged robust. The use of any AAC material can also be considered.

However, further modifications by the author were necessary to interpret test results.

5.2.4.1 PLS-4 AC additional and specific modifications

In addition to the accommodations (not affecting the scoring system) suggested in the PLS-4 manual, additional modifications were made by the author to ensure full and appropriate access to tests for children with physical or mild/moderate visual disabilities.

Firstly, the picture material in *PLS-4 AC* was also subjected to scrutiny, with the optometrist reporting **minimum visual acuity requirements** of 6/38 (in the moderate visual acuity impairment range). This was felt to be appropriate for the needs of the study, as severe visual impairment would be an identified exclusion criterion.

From the findings of the pilot study (see 5.4) the decision was made to exclude all *PLS-4 AC* items which could not be scored through a response based on pointing (finger- or fist-pointing, or use of gaze direction) or auditory scanning. The total number of items excluded by this criterion was 14/62 (23%). Details of items excluded are shown in the Appendices at 11.5.3.

The *PLS-4 UK* manual gives guidelines (p16) for establishing start point items, based on chronological age: for this study, start points were determined by using the same table with an estimate of child's level of functioning (derived from observation or interview with parent/carer). The advice is to establish a basal score when a child has passed three consecutive numbered tasks, and a ceiling score (end point) is noted when five consecutive numbered tasks in a subscale are marked as zero.

The pilot study conducted (see 5.4) suggested that full administration to the ceiling items would risk failure to complete all tasks for the group of children with CP who needed extra time: in consequence, a decision was made to incorporate a modification to record five consecutive *items* scoring at zero rather than five full tasks. This was a

significant modification, and depended on the clinician's experience to assess if the child had truly reached the ceiling of their abilities.

5.2.4.2 PLS -4 AC: author's modifications to scoring for age equivalence

The specific and additional modifications made by the author to the administration of the *PLS-4 AC* precluded the use of the norm-referenced tables offering standardised scores or age equivalents. Instead, to derive comparable scores/age equivalents for the subset of administered items, resulting from the modifications to the administration procedure described on page 5–124, a calculation was derived to produce an age equivalent *range* for the scores achieved.

The calculation was based on the sub-set of items presented to children with CP. It was derived by taking the child's raw score on the subset of items administered, dividing by the maximum possible score at ceiling for this subset of items, and then multiplying this by the maximum possible score at ceiling for the standard administration of the test. Tables in the manual were then consulted to establish **in which 6-month age band this score would have been recorded as a mean score for that age band** (for example 42-47 months). Median scores were reported to form the background measure of language comprehension. An example of the calculation is shown in Table 5-2:

Example of adapted scoring for subset of *PLS-4 AC items*						
	1	**2**	**3**	**4**	**5**	**6**
ID	raw score on subset	maximum possible score at ceiling on this subset	maximum possible score at this ceiling in standard administration	CP raw score (= (1/2)*3)	CP raw score is mean for age band	Median score of age band level
P06	39	40	52	50.7	3;6 - 3;11	45

Table 5-2: PLS-4 AC: Modified scoring for subset of items administered

The formula can be summarised as follows:

[raw score (CP items only) ÷ total possible score at ceiling (CP items only)] multiplied by the total possible score at ceiling in standard administration = CP raw score

This CP raw score is then used in the standardised tables to calculate the 6-month age band at which the score would have been a mean score (that is to say, at the 50th percentile). To derive a single figure to take forward into calculation, the median score for the age band was reported.

5.3 PROCEDURE

All assessments were presented to the whole CP group, and were undertaken by the author. Functional gaze control screening was conducted with two other members of the research team. A number of children were seen in a 1-1 setting in a child language testing facility at the University, with their parents present. Travel expenses (including accessible taxis) were offered to all families attending in this way.

However, most of the children tested were seen in their schools, with a familiar member of staff present, and parents invited. Children were offered breaks as needed, in discussion with parent or school staff member.

Each testing session began with a short warm-up activity, and a discussion with parents/carers and the child to establish their typical modes of communication. Children were positioned, wherever possible, in their own supportive seating, and were invited to bring and to use any additional forms of communication they would

typically find useful. As part of the warm-up session, the researcher checked that any method used by the child to signal *yes/no* was understood, and reminded parents/carers or school staff that the testing could stop if the child appeared in any way distressed or fatigued.

Children were tested on background measures in the following order: functional gaze control, *Mullen VR, PLS-4 AC.*

For all children, a short research report was prepared, and sent to parents, or forwarded to the referring SLT for distribution to parents (see 11.4.4).

5.4 PILOT STUDY

The final assessment materials and procedure of the study were refined following a pilot study involving two neurotypical children and two children with cerebral palsy (GMFCS Level V). These latter two children were not included in the final target group. The pilot study suggested the following adaptations made to assessment methods before their administration to the target group and these adaptations resulted in the final measures described on page 5–113.

Working with the two children with CP (both at motor Level V, GMFCS) showed that, although all items in *PLS-4 AC* could be administered, there were several test items without picture or toy material, for commands with prepositions, and for complex pictures not arranged in grid (table) form, where a range of other strategies were employed. For the section needing, for example, pointing to parts of the body – *show*

me your hands, the examiner replaced that command with (after explanation and consent) *I'm going to point to some of/bits of your body… tell me when I find your* --).

Administration in this way was somewhat intrusive, and time-consuming, taking two separate sessions of 2 hours each to complete all items, and appeared to risk fatigue and loss of attention for the two children with CP. Furthermore, the validity of the item under scrutiny was challenged where extra auditory memory load was added because of the adaptation. For example, for complex pictures, where the task was to find a specific item, the phrase *you tell me when I'm pointing to the* – was used, requiring the child to hold the target item label in memory far longer than would be necessary with standard administration. Several items were, in consequence, excluded from administration.

A small number of toys appeared to startle or distress the children with CP in the pilot study, and these were replaced with acceptable substitutes.

5.5 CHILDREN WITH CP MEETING CRITERIA

Despite our apparently clear inclusion criteria, 34 of the 66 children were excluded from further involvement in the study, following administration of the background measures.

It was not possible to define singular inclusion/exclusion criteria for this group of children. There were some children who failed to meet the language understanding floor of 12 months/single word understanding, but did show some ability in the functional gaze tasks. There were also children who were unable to meet the language

understanding criterion, *and* performed poorly on the functional gaze trials. Exclusion decisions were made, therefore, through a combination of results from both tasks. This approach reflects the clinical problem of the interplay between intellectual, language and functional vision abilities described in the introduction. This exclusion on several measures was deliberately cautious to ensure confidence for the researcher in understanding the children's profile of development and methods of response.

While exclusion was based on several factors as described, Table 5-3 describes the primary reason for exclusion, as best determined by the researcher, drawing on clinical experience.

Children with CP: Reasons for exclusion		
Source of decision-making	**Primary reason for exclusion**	**n=**
GMFCS level	Physical ability above criterion	1
Compliance on day of testing	Unable/unwilling to engage (poor health, including epilepsy)	8
Functional Gaze Control	Unable to fix gaze consistently	10
Mullen VR/PLS-4 AC	Language understanding/performance abilities above criterion	5
	Language understanding/performance abilities below criterion	10
	total	**34**

Table 5-3: Participants in CP group, reasons for exclusion

As the excluded children had already engaged in the assessment protocol when these findings forced exclusion from the study's analysis, the researcher continued all the assessments as fully as possible, and, where appropriate, discussed and suggested further specialist assessment of language, learning and vision as appropriate with specialist teams in the community.

The category background measures of the 32 children included in the social communication skills study are shown in Table 5-4:

Category characteristics of included children with CP (n=32)																
sex		**CP type**			**GMFCS**		**MACS**			**CFCS**				**VIKING**		
M	F	DYSK	MIXED	SPASTIC	IV	V	III	IV	V	II	III	IV	V	II	III	IV
16	16	19	11	2	14	18	2	12	18	1	5	7	19	2	6	24

Table 5-4: Characteristics of included children with CP (n=32)

There were equal numbers of male (n=16) and female (n=16) participants. The children included were identified as having dyskinetic, mixed (spastic/dyskinetic) and spastic type CP, with the larger groups being the dyskinetic and mixed types. Children recruited were all wheelchair users, and hence classified as either Level IV (*self-mobility with limitations; may use powered mobility*) or Level V (*transported in a manual wheelchair*). There were approximately equal numbers of children classified at GMFCS Level V (the most severe) and Level IV.

Children fell largely into the most severe category of functional manual ability difficulties, with 30/32 children having severe difficulties in this area.

24/32 children had no recognisable intelligible words. There were 6 children classified in *Viking Speech Scale* category III (*speech is unclear and not usually understandable to unfamiliar listeners out of context*), with 2 children rated as having some *imprecise speech* (Level II).

Within the *Communication Function Classification System* (CFCS), 26/32 children were classified in the two most severe Level IV (inconsistently sends and/or receives information even with familiar partners) and Level V (seldom effectively sends and receives information even with familiar partners).

5.6 RESULTS OF FUNCTIONAL GAZE CONTROL PROFILES

This section describes individual results for the 32 children meeting criteria for inclusion.

Given that the *Functional Gaze Control* measure was a novel assessment devised to support this study, inter-rater reliability of scores were calculated from live scoring by two procedure-trained clinicians (the author, plus the research assistant, another SLT) on 10 children in the included group. Cohen's Kappa co-efficient was used to account for possible agreement by chance (Cohen, 1968). For gaze fixation, k = 0.62, and for transfer of gaze, k = 0.79. These represent *good* and *excellent* agreement, respectively (Cicchetti, 1994). Figure 5-4 shows the fixation scores for all included children.

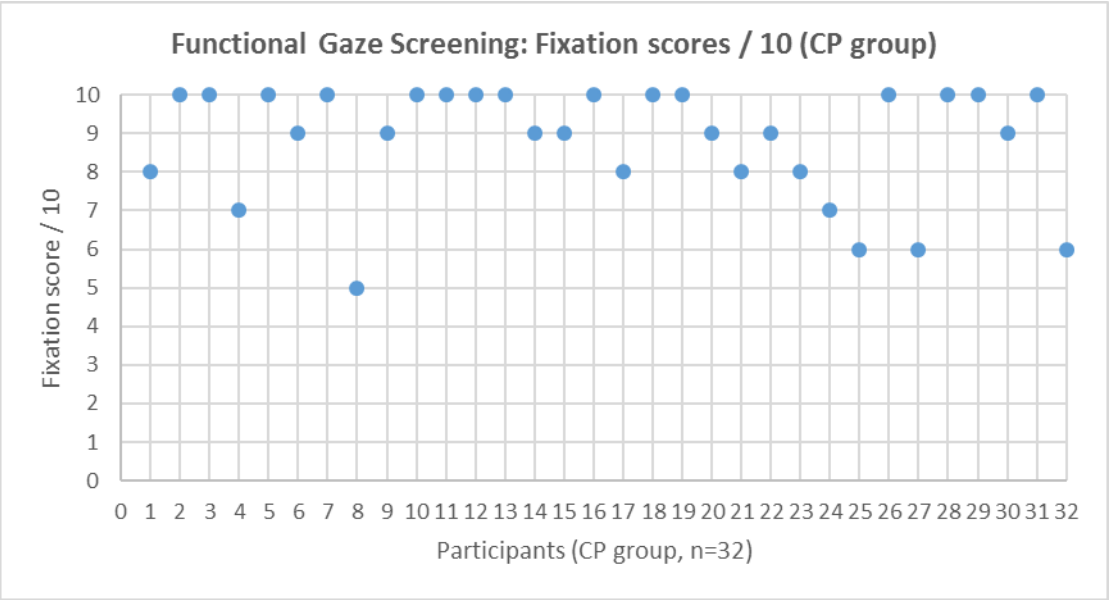

Figure 5-4: Fixation of Gaze results

In this group, 32 children scored at 50% and above for this task. One child (ID8) and 3 children (ID25, ID27 and ID32) appeared to score poorly, but in the context of the other tasks, their performance was adequate to merit inclusion.

Since children with CP may have eye movement disorders which might make it more difficult for them to look in one direction than another, bias for location of target was assessed. *Chi-Square Goodness of Fit* test was used to assess whether fixation score varied according to the location of the target (centre, top, bottom, left, right). Results showed that there was no difference between location in terms of fixation score and therefore no evidence of a bias to location ($\chi^2(2) = 0.183$, $p=0.996$). Figure 5-5 shows scores for transfer of gaze:

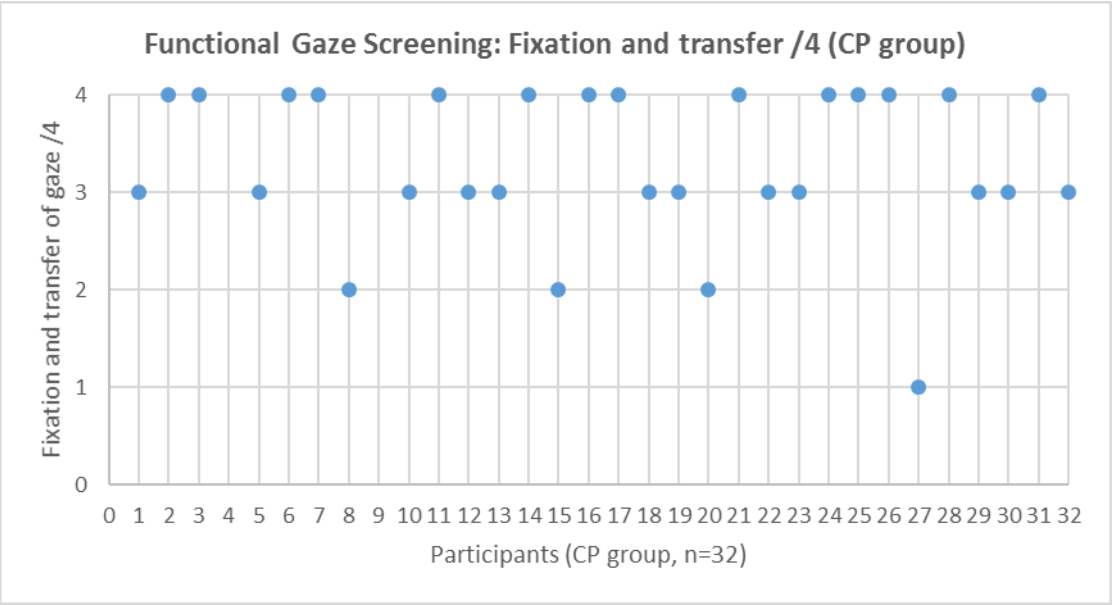

Figure 5-5: Fixation and Transfer of Gaze results

Similar calculations were made for this fixation and transfer task, in the light of possible differences between transfer of gaze to left or to right. A *Chi-Square Goodness of Fit* test was used to assess whether the gaze transfer score varied according to the presentation of the second stimulus (on the left or right of initial stimulus).

There was no difference between the location of the second target and gaze transfer score and therefore no evidence of a bias to location ($\chi^2(2) = 0.9$, $p=0.343$).

There were 29/32 children who scored at 50% or above on this task. Child ID4 and Child ID9 had fatigued for this task and could not co-operate, but had shown good transfer of gaze in warm-up play to that point; similarly, child ID27 had shown recorded gaze transfer skills in the practice session. The decision was made to retain these three children in the second part of the study (they had all passed the initial fixation of gaze trials).

In summary of this chapter, it is worth noting that identifying a group of children meeting inclusion criteria was a testing element of the study, perhaps unsurprisingly so, given the heterogeneity of the population and the complexity of profiles.

With this group of children with CP identified it was possible to undertake the focus of the study. The following chapter describes the recruitment of comparison groups of children with Down syndrome and with autism spectrum disorder.

6 SELECTION OF COMPARISON GROUP PARTICIPANTS

The group of 32 children with CP, at GMFCS levels IV and V, aged 3-12, with language understanding between 12-54 months, and adequate motor control to fix and transfer gaze had now been identified, and the investigation of social responsiveness and joint attention could be approached.

The study aims guided the next steps, which were to identify comparison group participants. Recruitment, through the inclusion criteria, aimed to match the comparison groups of children with ASD and DS on chronological age and on intellectual ability/language comprehension.

6.1 CHILDREN WITH DS AND ASD: RECRUITMENT AND INCLUSION

Children with DS and children with ASD were recruited in the same way as the children with CP:

- from relevant clinical lists at the author's children's hospital workplace
- from schools in London, South East England and Sheffield, South Yorkshire. Recruitment was conducted through researcher contact with the children's NHS speech and language therapist (SLT), who in turn approached parents with information sheets and invitations to participate (see 11.4.1). Consent forms (see 11.4.3) were completed by parents and forwarded to the researcher prior to the testing sessions. The community speech and language therapist supporting the recruitment received written details of inclusion criteria for children with DS and ASD as follows in Table 6-1 and Table 6-2:

Criteria for inclusion (children with DS)
• confirmed diagnosis of Down syndrome • chronological age 3-12 years • language understanding/performance abilities at 12-54 months' age equivalent level • hearing levels (including corrected) adequate for speech recognition • corrected vision/no concerns re vision

Table 6-1: Inclusion criteria for children with DS

Criteria for inclusion (children with ASD)
• confirmed diagnosis of autism spectrum disorder • chronological age 3-12 years • language understanding /performance abilities at 12-54 months' age equivalent level • hearing levels (including corrected) adequate for speech recognition • corrected vision/no concerns re vision

Table 6-2: Inclusion criteria for children with ASD

A small number of children were excluded from the comparison groups as shown in Table 6-3:

Group, children with	consented n =	excluded	included n = (female/male)
Down syndrome (DS)	19	3	16 (6/10)
autism spectrum disorder (ASD)	10	1	9 (0/9)

Table 6-3: Children with DS and ASD, reasons for exclusion

There were three children excluded from the DS group: two of these children were unable to show understanding of language skills at single word level, and one child had

a confirmed diagnosis of DS-ASD. As a result, 16 children with DS participated in the early social communication skills tasks.

There were 10 children recruited to the ASD group; one child was subsequently not included as he also proved to have receptive language abilities below the entry criterion. There were, therefore, 9 children in this group, all boys.

In the CP group, since the children not meeting criteria had already engaged in the assessment protocol when these findings forced exclusion from the study's analysis, the researcher continued all the assessments as fully as possible, and, where appropriate, discussed and suggested further specialist assessment of language, learning and vision as appropriate with specialist teams in the community. For all children, in all three groups, a short research report was prepared, and sent to parents, or forwarded to the referring SLT for distribution to parents (see Figure 11-11).

Following this recruitment of comparison groups meeting criteria, it was possible to proceed with all background measures testing. All three groups were assessed using the protocol for language assessment (*PLS-4 AC*), performance abilities assessment (*Mullen VR*). The three groups were then assessed using a novel measure of early social communication skills, *Using Gaze for Noticing and Telling* (*Gaze-NoTe*), developed specifically by the researcher for this study from some published, with some novel tasks.

The following chapter describes the development of this novel procedure.

7 TESTING EARLY SOCIAL COMMUNICATION SKILLS : ALL GROUPS

7.1 DEVELOPMENT OF TARGET MEASURE: *GAZE-NOTE*

This measure was developed, then, specifically for the study, as there did not appear to be a single, suitable, accessible direct assessment for the skills targeted (social responsiveness and joint attention). The measure name *Gaze-NoTe* emphasised the role of gaze direction for children with CP, as well as the "real-life" skills of noticing and telling (responding to and initiating joint attention) that appeared to be difficult for some children.

To maximise validity for this novel procedure, the measure was based on published assessments as follows:

- two adapted subtests from a published assessment including examination of early socio-cognitive skills (*Very Early Processing Skills* (VEPS)) (Chiat & Roy, 2008), described in 4.2.4
- novel task items to support these two adapted sub-tests, based on the alerting "red flags" (see page 3–47) of social skills development reported to be indicative of persisting difficulties, and items from *Early Social Communication Scales* (see 3.3 and 4.2.4) and the *Autism Diagnostic Observation Schedule*

7.1.1 TASKS FROM VERY EARLY PROCESSING SKILLS

From the review of available measures of social responsiveness and joint attention, the *Early Socio-Cognitive (ESC)* measures of this *Very Early Processing Skills* assessment (Chiat and Roy, 2008) had been identified as accessible to children using gaze direction as a response, and had appealing material in the task sets (described in 4.2.4). These *ESC* measures looked at three sets of socio-cognitive skills, which were then combined in this original study to give a composite score. These sets were social responsiveness, joint attention, and assessment of symbolic understanding. For this current study, the first two sets were selected as appropriate to address the aims posed.

7.1.1.1 VEPS-ESC Social responsiveness

As described in 4.2.4, in this procedure the researcher acts out several scenes in which six emotions are portrayed (hurt, surprise, frustration, anger, distraction and achievement). The child's **response to the researcher's emotional expression** is recorded, and scored by the child's looks to the researcher's face; either fleeting (less than two seconds) (allocated 1 point) or sustained (for at least two seconds) (allocated 2 points). The task is for the child to *notice* and *respond* to the emotion portrayed.

These scenes are supported by researcher script guidelines[12:] for example, for the facial expression of *surprise,* the researcher finds a nappy in the toy bag and says

What's this? It's a nappy. That's not a toy! Let's see what else is inside our toy bag

In the light of the findings from the background measures pilot study, several minor changes were made to the items and administration of the *VEPS-ESC* tasks, largely to

[12] https://is.gd/EarlySociocognitiveBattery (accessed September 2016)

account for the somewhat unforeseen extended length of time the full battery of background measures and target measure would take for children with CP to complete.

The adaptations made to the social responsiveness tasks are shown in Table 7-1:

Adaptations made to *Very Early Processing Skills- ESC* social responsiveness tasks			
Section	**Task items (original)**	**Task items (modified)**	**Rationale**
Social Responsiveness	• hurt • surprise • anger • fear • distraction • achievement	• hurt • surprise • (frustration) • fear • (omitted) • achievement	One task modified and one task removed (may have provoked startle reflex/acute distress)
	maximum score /12	maximum score /10	

Table 7-1: VEPS-ESC tasks adaptations (social responsiveness)

The original toy material and scripts were used for these tasks and are described in Table 7-2:

***VEPS-ESC* toys and script for social responsiveness assessment**	
Item	**Script**
hammer and pegs HURT	Look, I found a hammer and peg set. I'll do some hammering. Ow! (show HURT). I hurt my finger. I'm going to stop hammering now.
nappy SURPRISE	What's this? (show SURPRISE). It's a nappy. That's not a toy! Let's see what else we have.
torch FRUSTRATION (replaced ANGER)	Ooh, look, it's a torch. It lights up. Let's switch it on (show FRUSTRATION). Oh, it's not working. It hasn't got any batteries. Let's see what else is in here.
box and spider FEAR	Ooh, a present. I wonder what's inside. Let's open it and see (show FEAR). Aargh, it's a spider … I don't like spiders. Oh, phew, it's only a pretend spider.
bricks ACHIEVEMENT	Let's see if I can build a big tower (show ACHIEVEMENT).
Responses	
0	the child does not look at the assessor's face at all
1	the child looks briefly/fleetingly at the assessor's face
2	the child looks at the assessor's face for at least 2 seconds

Table 7-2: *VEPS-ESC* toys and script for social responsiveness assessment

As shown in Table 7-1: VEPS-ESC tasks adaptations (social responsiveness)Table 7-1, the scoring system detailed in Chiat and Roy's original study was followed. For the **social responsiveness** task with researcher presenting a range of facial/verbal emotions, a score of 0 was given if the child did not look at the researcher's face at all; a score of 1 point was given if the child looked briefly/fleetingly at the researcher's face, and a score of 2 points was given if the child looked at the researcher's face for at least 2 seconds.

7.1.1.2 VEPS-ESC Joint attention

As outlined in the review of social communication skills assessments in 4.2.4, these tasks offered opportunities to the child to share experiences; engage in joint attention. Six plastic eggs were displayed, one at a time, and the researcher opened them to reveal a small object, such as a tiny bag. Larger versions of these objects were placed to the side, front and back of the child. The researcher noted the child's transfer of gaze from the egg to the researcher's face, or from the tiny object in the egg to researcher's face, and also noted if the child followed the researcher's gaze of direction towards the larger object, or, failing this, could follow the researcher's finger-point to the object. The researcher offered verbal prompts again (see Table 7-4).

As the researcher looked at the child, the researcher shook the egg to one side at arm's length, without speaking. The researcher waited up to five seconds to see if the child could look from egg to researcher. This was described as "gaze switch".

The researcher then opened the egg slowly, looking at the child's response, and, again without speaking, showed the contents to the child. The child's look to researcher's face was recorded at this point if it occurred. The toy was then returned to the egg, and the adult offered a further prompt to encourage the child to follow gaze (listed in Table 7-4).

The researcher then looked in the direction of the larger matching object. If the child failed to respond to follow the researcher's line of sight, the researcher repeated the prompt, accompanied this time by a finger-point towards the larger object. The ability tested was described as "gaze monitoring".

VEPS-ESC adaptations from original procedures: joint attention			
Section	**Task items (original)**	**Task items (modified)**	**Rationale**
Joint Attention	• person • hat • candle • bag • tiger • ring	• person (pirate) • tiger • dinosaur	• Material replaced by items accessible to children with moderate visual impairment. Items using very small toys (<1cm) replaced with 20cm toys. • 10cm eggs replaced by 40cm eggs • larger matching objects were 35-40cm, and positioned nearer to the child than the original instructions (at 1.m, rather than "across the room")
	maximum score = 18	maximum score = 9	

Table 7-3: VEPS-ESC tasks adaptations (joint attention)

Figure 7-1: Very Early Processing Skills-ESC Joint attention toys

For the **joint attention** tasks, again, the original VEPS scoring schedule was followed: a score of 1 point was awarded if the child looked towards the researcher when the egg holding the toy was selected and opened, and 2 points if the child follows the assessor's eye gaze switch and verbal statement referring to a matching, larger, object positioned 1.5m away, or 1 point if the child follows the assessor's point and repeated statement about the larger object.

<table>
<tr><th colspan="3">VEPS-ESC toys and script joint attention assessment</th></tr>
<tr><td colspan="3">The researcher opens the egg box and says Let's see what's inside ... oh, look! Here are some eggs … I'm going to look at this one. The researcher shakes the egg with arm extended to one side, without speaking; waits for 5 seconds for response. The researcher opens the egg, looking at the child, and, without speaking, shows the contents of the egg to the child. The child's response is recorded as the adult examines the contents. Then the researcher says shall we put it back in the egg now? The researcher does so, and says I brought my pirate/dinosaur/tiger with me today, and looks in the direction of the corresponding larger object, which has been placed at a distance of 1.5m. If the child fails to respond to following the researcher's gaze, the comment is repeated, and this time the researcher points in the direction of the larger object. The procedure is repeated for 3 eggs.</td></tr>
<tr><td>Item in egg</td><td>Gaze switch</td><td>Gaze monitoring</td></tr>
<tr><td>pirate</td><td></td><td></td></tr>
<tr><td>dinosaur</td><td></td><td></td></tr>
<tr><td>tiger</td><td></td><td></td></tr>
<tr><td>Responses</td><td>0 = no look in either of conditions
1 = Either look from egg to adult while adult is shaking egg before opening or
look from toy to adult after the egg has been opened whilst contents shown</td><td>0 = no look in either of conditions
1 = look following adult's point and repeated verbal statement
2 = look following adult's gaze and verbal statement</td></tr>
<tr><td></td><td>maximum score = 3</td><td>maximum score =6</td></tr>
<tr><td>total JA score</td><td colspan="2">maximum score = 9</td></tr>
</table>

Table 7-4: VEPS-ESC toys and script for joint attention assessment

7.1.2 NOVEL TASKS INVESTIGATING JOINT ATTENTION

To supplement the responses seen in the two *VEPS-ESC* tasks, a novel set of items aimed to target very early social responsiveness, and observe spontaneous imitation of joint attention were added to produce the *Gaze-NoTe* measure. The construction of the items was guided by the "core" red flags identified by Wetherby and colleagues (Wetherby et al., 2004; McCoy, Wetherby & Woods, 2009) (see page 3–47) and the tasks in the *Autism Diagnostic Observation Schedule* (ADOS) (Lord et al, 2001). Tasks targeting "telling" (initiating joint attention) were emphasised in these items, as they were not included in the *Very Early Processing Skills* procedures.

In Chapter 3 the review of Wetherby's work identified a number of important markers for ASD in young children. Some of these could be reliably observable for children with little or no speech and motor movement difficulties: 11 of the 20 behaviours fell into this category. These studies also highlighted three specific areas of concern identifying autism in children at aged two years

- lack of gaze to face
- lack of co-ordination of verbal and nonverbal communication
- lack of shared attention

This second observation was not relevant to children with little or no speech, but focusing on two skills, lack of gaze to face, and joint attention, that acted as "red flags" in McCoy's study seemed important.

The ADOS assessments target a similar set of skills, using play equipment familiar to children and young people. These are well-established procedures assessing social communication skills, including joint attention in young children. The *Toddler ADOS* (Luyster et al, 2009) is an extension assessment designed to highlight the abilities and

deficits of children's social communication development from 12-30 months. The list of activities/observed behaviours includes response to name, free play to observe any sharing or showing (initiation of joint attention), response to joint attention, responsive social smile.

However, the ADOS assessments are not suitable for use with children with cerebral palsy: there are stringent restrictions on its use with rigorous training for administration and competency updates, and does not permit any adaptation, accommodation or modification, for other populations. Much of the material requires object manipulation. Furthermore, the appropriate module to administer is decided on expressive language level.

However, insights from the ADOS assessments suggested the value of a play-based procedure to enable observation of the targeted abilities. ADOS uses a semi-structured play script to increase consistency of administration.

The tasks were based on structured, scripted play using toy rabbits for every task item: this single focus was chosen to allow familiarity with the material to help support the child's attention and recall through the tasks.

Figure 7-2: Toy material for *Gaze-NoTe* activities

7.1.2.1 Warm-up activities

The adult showed the child three toy rabbits, one operable with a single hand/head switch, in a portable (pet carrier) hutch, and asked *would you like to play with one of them?* The child was encouraged to choose, and to play for a short time. This warm-up activity allowed the researcher to observe the child's visual attention, use of vocalisation, and use of gaze direction.

The researcher then counted the rabbits with the child; *one, two, three rabbits!* and invited the child to look out for more rabbits – *we'll count how many we see!* -- through this part of the session. This was to encourage the child to identify rabbits as they appeared, and thus provided opportunities via these prompting "presses" to initiate joint attention with the researcher.

7.1.2.2 Initiation of joint attention (iJA) activities

The materials and the script for these activities were devised by the author. The rationale for the choices made included acknowledgement of the child's intellectual disability and the need to present more than one opportunity to be successful with the task. iJA activities are included in the Wetherby and *ADOS* procedures, but some require manipulation of toys (for example, remote control car), and are only presented once.

The ADOS assessments offer play activities in which iJA behaviours may or may not be observed: with a range of play material, children may initiate joint attention by looking at an item in which they are interested, then look at the examiner and then direct the examiner's gaze to the item they want to share. The child may "show" items of interest: they may hold up a toy and look at the examiner to show her it and to share interest.

This novel iJA assessment targeted the responses of both *noticing* and *telling.* The researcher presented a simple **formboard puzzle** (*Pets)* and invited the child again to keep a look out for more rabbits. Five formboard pieces were presented, with the rabbit shown fourth: the researcher observed any attempt at joint attention initiation at this point (eye contact and "knowing" smile, vocalisation, finger- or hand-pointing with eye contact). If there was no attempt at iJA, the researcher held up the rabbit and said *is this another rabbit? So it is!* and repeated the counting game.

The second press used a set of five **wind-up toys**, one of which was a rabbit. They were drawn from an opaque box, one by one, with the rabbit appearing fourth. At the beginning of the activity, the adult prompted saying *look out for the rabbit! Where is it?* and again if no iJA response was recorded, repeated counting the rabbits after saying *is this another rabbit? So it is!*

The final press for iJA was to encourage the child to comment on (*notice* and *tell* about) a **surprise** rabbit, one of the warm-up toys, as it appeared behind the researcher presented "secretly" (in panto "behind you" style by a second adult). This occurred while the researcher was showing the child (three times) a **pop-up toy rabbit**, with *ready, steady, go!* to record any example of anticipation of a familiar routine, and request for a routine to be repeated.

7.1.2.3 Response to joint attention (rJA) activity

As an additional response to request for joint attention activity, a further, novel, item was included: the researcher invited the child to find any **"hiding rabbits"** (well-hidden, only partly showing, and the same toys as seen in the warm-up activity) behind them in the room (and researcher turned the child in their chair, if necessary, to bring the toy into the child's line of sight), saying

So ... have we found all the rabbits now?

The rabbits were well-hidden enough to make this difficult, and the researcher then smiled and directed their own gaze to the hidden toy. If the child failed to follow the researcher's direction of gaze, the adult added a verbal prompt

Look! I can see one!

If the child still failed to follow direction of gaze, the adult added a finger point and, if necessary, said

Look! There's one!

At some point in the joint attention activities described, the researcher would offer a relaxed social smile and observe if the child returned this smile. Similarly, at some point, the researcher used the child's name, and observed any response from the child via increased attention, eye contact etc. This press for response to name was offered three times.

The activities were video-recorded with a single camera directed at the child's eyes, but as so much of social communication skill use is qualitative and judged by the conversation partner, notes and scoring were recorded at the time.

7.1.3 RECORDING AND SCORING *GAZE-NOTE* MEASURE

Responses to the *VEPS-ESC* and supplementary items were collated to form the *Gaze-NoTe* measure. Items in Table 7-5 are shown in approximate order of emergence in neurotypical development: identification of those items targeting social responsiveness and those targeting joint attention are marked in column 2 (SR/JA), with items targeting response to joint attention marked as rJA, and those targeting initiation of joint attention marked as iJA.

The activities were administered following the testing of background measures *PLS-4 AC* and *Mullen VR*, and the assessment was recorded and scored as shown in Table 2-1:

***Gaze-NoTe* measure**				***Source of strategy***	***Modification***
#	**SR/JA**	**Item**	**maximum score**		
1		(Choosing a toy)	n/a		
2	SR	social responsiveness (reaction to emotion)	10	(*VEPS-ESC*)[13]	Some facial expressions omitted
3	SR	smiles in response to researcher's social smile	1	ADOS[14] & Wetherby[15]	none
4	SR	response to name (3 attempts)	1	ADOS & Wetherby	none
5	SR	*ready, steady go!* with pop-up rabbit	1	ADOS & Wetherby	Toy replaces tickling, head covering for *pee-po!*
6	SR	requesting a turn, or *more* with pop-up rabbit	1	ADOS & Wetherby	Rabbit toy replaces balloon/bricks
7	rJA	joint attention assessment	9	(*VEPS-ESC*)	Eggs enlarged, fewer (3/6) presented; different toys used and enlarged
8		rJA hidden rabbit (follows gaze)	2	ADOS & Wetherby	none
9		rJA hidden rabbit (follows gaze plus point)	offered if gaze alone (7) unsuccessful 1	ADOS & Wetherby	

[13] Chiat & Roy, 2008

[14] Lord et al, 2001; Luyster, 2009

[15] Wetherby & Prizant, 1993; Wetherby et al, 2004

10	iJA	iJA formboard	1	ADOS & Wetherby	More than one opportunity to "notice and tell" (iJA) about a specific toy rather than in free play
11		iJA wind-up toys	1	ADOS & Wetherby	
12		iJA surprise rabbit	1	ADOS & Wetherby	
Social responsiveness score			/14		
Joint attention score			/14		
total score			/28		

Table 7-5: Scoresheet for collated *Gaze-NoTe* measure

7.1.4 *GAZE-NOTE* INTER-RATER RELIABILITY

As this was a novel measure, inter-rater reliability was calculated to ensure that the subjective nature of the observations would not jeopardise interpretation of the results. Intra-class coefficients were calculated on 25% of the children with CP. This group was chosen for this calculation as they were identified as the group likely to pose greatest challenges to robust coding across different raters: clinically, the difficulties in achieving consensus re gaze direction were well documented.

Four trained coders were used in the reliability: two undergraduate SLT students, one postgraduate SLT student, one newly qualified SLT. A one-way model was chosen for the reliability analysis, as 4 different coders were used, and results compared with a single coder. Reliability was calculated on the scores for the overall *Gaze-NoTe* target measures, and for the individual components of this measure. The single measure intra-class correlation statistic (as opposed to an average measure) is quoted (Hallgren, 2012), as a subset of subjects was coded by multiple raters: this measure does provide a more conservative estimate of agreement. Results for this inter-rater reliability calculation are shown in Table 7-6:

Inter-rater reliability for *Gaze-NoTe* measure	
***Gaze-NoTe* Measure**	**r (ICC) =**
Total score	0.74
Initiation of joint attention	0.79
Response to joint attention requests	0.84
Social responsiveness	0.74

Table 7-6: Inter-rater reliability for *Gaze-NoTe* measure

The values of the coefficients for all of the measures fell in the category (0.7-0.9) described as *excellent* in relation to clinical significance (Cicchetti, 1994).

7.1.5 *GAZE-NOTE* TEST VALIDITY

The test has clearly defined construct validity (the extent to which it represents a specific theoretical construct), established through a focus on the assessment of social communication behaviours well known to be delayed/disordered in children with a diagnosis of ASD (the "red flags" described in 3.3) and under scrutiny for delay/disorder in children with cerebral palsy.

The test has clear content and face validity in being adapted and developed from procedures used in the assessment of social communication in non-motor impaired children, and from methods used in experimental psychology to study social communication development in typical development.

No suitable gold standard criterion relevant to non-speaking children with severe CP was available with which to establish the concurrent validity of the measure. However,

evaluation of performance from the distribution of scores within the group of children with CP compared with the performance of children with a known diagnosis of ASD on the same measure may shed light on the test's validity.

7.1.6 *GAZE-NOTE* PROCEDURE

As for the background measures, this procedure was completed with all included children in the three study groups, after the assessment of background measures (see 0). The children with Down syndrome and the children with ASD were all seen in their schools, with a familiar member of staff present, and parents invited. Again, children were offered breaks as needed, in discussion with parent or school staff member.

Each testing session began with a short warm-up activity, and children, parents/carers or school staff were reminded that the testing could stop if the child appeared in any way distressed or fatigued.

Children were tested on background measures in the following order: functional gaze control, *Mullen VR, PLS-4 AC.* The same subset of test items was presented to the children in the DS and ASD comparison groups as had been used with the target group of children with CP.

Again, for all children a short research report was prepared, and sent to parents, or forwarded to the referring SLT for distribution to parents (see 11.4.4).

8 RESULTS

This chapter presents the results from the assessments undertaken to address the second and third study aims:

- to compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
- to investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

To address these aims, both **between** group (children with CP/with DS/with ASD) and **within** group (CP group) analyses were undertaken. However, in order to exclude as many confounding variables as possible, examination of group matching was undertaken. Care had been taken to support group matching through narrow inclusion criteria for chronological age and intellectual ability, but these background matching measures needed closer examination.

8.1 GROUP MATCHING

8.1.1 CHARACTERISTICS OF GROUPS (CHILDREN WITH CP/DS/ASD)

To look for any significant differences between the three groups on background measures, descriptive characteristics were calculated:

Gp (n)	chronological age (months)			Mullen VR age equivalent (ae) scores (months)			*PLS-4 AC* age equivalent (ae) scores (months)		
	mean	range	sd[1]	mean	range	sd[1]	mean	range	sd[1]
CP (32)	88	40 - 145	30	28	9 -54	8	28	15 - 57	11
DS (16)	90	49 - 123	22	26	10 -39	8	27	15 - 45	10
ASD (9)	102	65 - 168	29	26	17 - 42	13	21	15 - 33	7

[1] standard deviation

Table 8-1: Chronological age, *PLS-4 AC* and *Mullen VR* age equivalent scores

8.1.2 TESTING FOR NORMAL DISTRIBUTION OF BACKGROUND MEASURES

Following visual inspection of box-plots and histograms, Kolmogorov-Smirnov statistic (K-S) was used to confirm the normality of distribution for the three independent variables/background measures:

Testing for normal distribution			
Group, children with	**chronological age, K-S *p*=**	**Mullen VR AE K-S *p*=**	**PLS-4 AC AE K-S *p*=**
cerebral palsy	.125	.130	.196
Down syndrome	.117	.161	.180
autism spectrum disorder	.259	.182	.259

Table 8-2: Testing for normal distribution: age, *Mullen VR* and *PLS-4 AC* ae scores

This Kolmogorov-Smirnov statistic confirmed that the 3 background measures: chronological age; *Mullen VR* subscale and *PLS-4 AC* age equivalents were normally distributed for all groups.

However, since the ASD group was a relatively small group (n=9), the statistic used and other standard tests of significance risked lacking power to identify deviation from normality in the data. Consequently, a further procedure (examination of skewness and kurtosis) was undertaken to support the Kolmogorov-Smirnov finding.

Accordingly, z-scores were calculated for skew and kurtosis (by dividing the skewness and kurtosis statistics by their standard errors). The resulting z score was assessed against a boundary of greater or less than 2.58. This is a more conservative boundary level than the standard -2 to +2, representing minus or plus two standard deviations from the mean, but is suggested to accept a statistical significance level of 0.01, which equates to a z-score of ±2.58 (statistics.laerd.com, 2013).

Table 8-3: Relationship between z scores, probability values and confidence intervals Variable	Skewness	Kurtosis
Chronological age	z=2.39	z=2.97*
Mullen VR	z=1.29	z=0.77
PLS-4 AC	z= 0.96	z=-0.57

Table 8-4: Examination of skewness and kurtosis for background measures in ASD group

z-score (Standard Deviations)	p-value (Probability)	Confidence interval
< -1.65 or > +1.65	< 0.10	90%
< -1.96 or > +1.96	< 0.05	95%
< -2.58 or > +2.58	< 0.01	99%

Table 8-5: Interpretation of z scores, probability and confidence intervals[16]

Although the kurtosis z score for chronological age was outside the more cautious boundary level (+/- 2.58) significance threshold, the z score for skewness is within the suggested range, and the Kolmogorov-Smirnov result was >0.05 and hence non-significant. Taken together, age data can be considered approximately normally distributed for this ASD group too, allowing for use of parametric statistics in further analysis.

The three groups were compared using one-way analysis of variance (ANOVA) procedures for the three independent variables (chronological age, *Mullen VR* subscale

[16] https://is.gd/zscores (pro.arcis.com) accessed June 2017

and *PLS-4 AC* age equivalent scores) separately. To carry out an effective ANOVA analysis, there were two key assumptions to be examined, in addition to the confirmation of normally distributed data:

- homogeneity of variances (comparison of "spread" in the data)
- influence of outlying data points ("outliers")

This examination was particularly important in the light of the smaller group numbers, and the presence of an outlier in the ASD group (shown in Figure 8-1):

8.1.3 GROUP MATCHING: CHRONOLOGICAL AGE

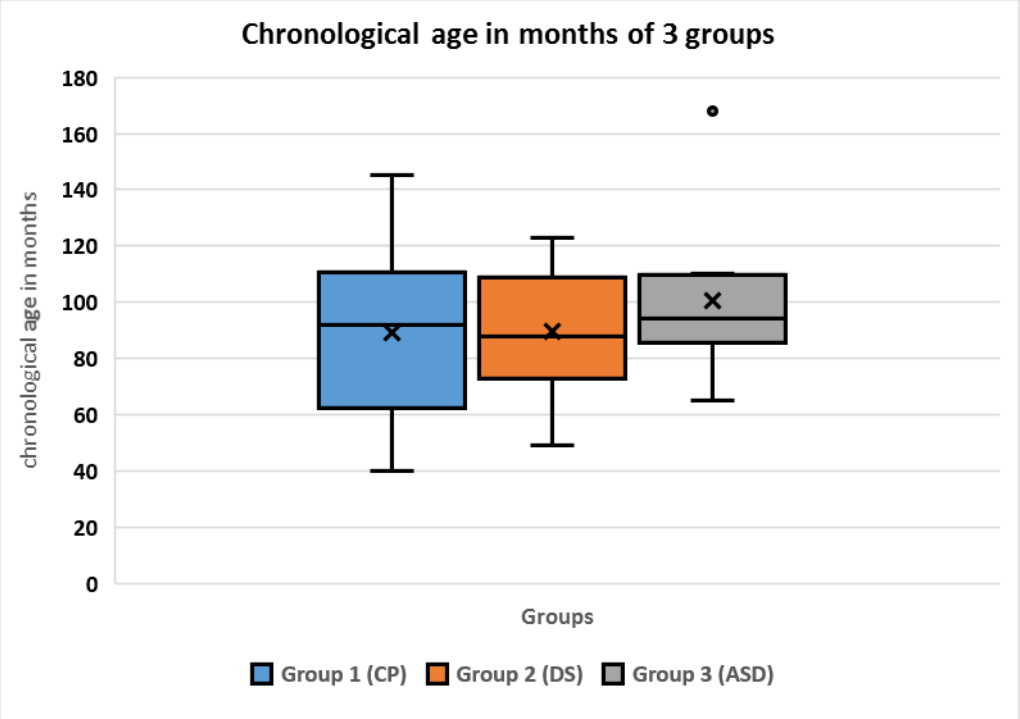

Figure 8-1: Chronological age in months (all three groups)

The effect of this outlier on the group results was investigated. Levene's statistic was used to test equality of variances. The result from this statistic with the outlier

included was *not* significant (level of significance for this and subsequent statistics was set at $p < 0.05$) ($p = 0.168$) and the assumption of homogeneity of variances was therefore not violated. However, when the outlying data point was excluded, Levene's statistic ($p = 0.012$) suggested that, for this data, there were not equal variances.

In consequence, and to further assess the impact of the outlier, two ANOVAs were completed. With the outlier included (and homogeneity of variances confirmed), standard one-way ANOVA procedures were followed, and the results

$F(2,54) = 0.82$, $p = 0.922$

suggested that the groups were matched for chronological age.

With the outlier excluded (and since the assumption of homogeneity of variances was violated), Welch's ANOVA statistic was used. Chronological age was then shown again *not* to be statistically significantly different across the three groups:

Welch's $F(2, 21) = 0.64$, $p=0.537$.

In the light of similar conclusions from these two calculations (outlier *Included* and outlier *excluded*), a decision was made to continue to include the outlier in further analysis.

8.1.4 GROUP MATCHING: MULLEN VR SCORES

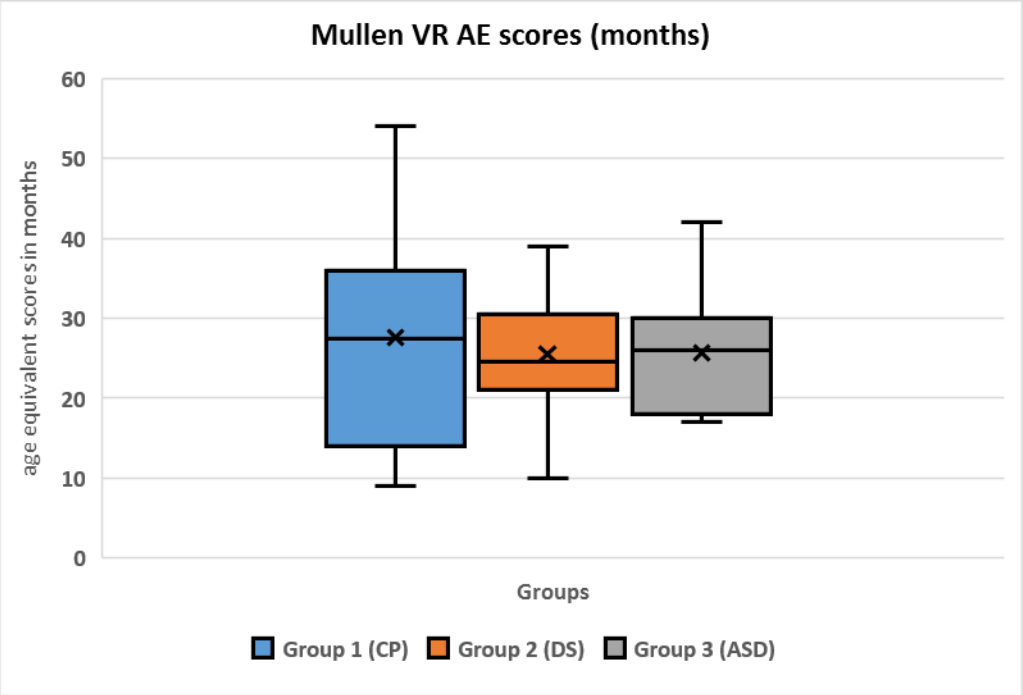

Figure 8-2: *Mullen VR* age equivalent scores in months (all three groups)

In this data for the age equivalent in months scores for the *Mullen VR* test, no outliers in the data were identified, but Levene's test result (p = 0.007) indicated that the assumption of homogeneity of variances was *not* upheld. Therefore, Welch's ANOVA, which can accommodate heterogeneity of variances, was calculated:

Welch's F (2, 24.6) = 0.26, p = 0.77

indicating again no significant differences in the age equivalent scores for this measure.

8.1.5 GROUP MATCHING: PLS-4 AC MEASURES:

A boxplot for the language understanding age equivalent scores (*PLS-4 AC*) revealed an outlier in this data, in the CP group, shown in Figure 8-3:

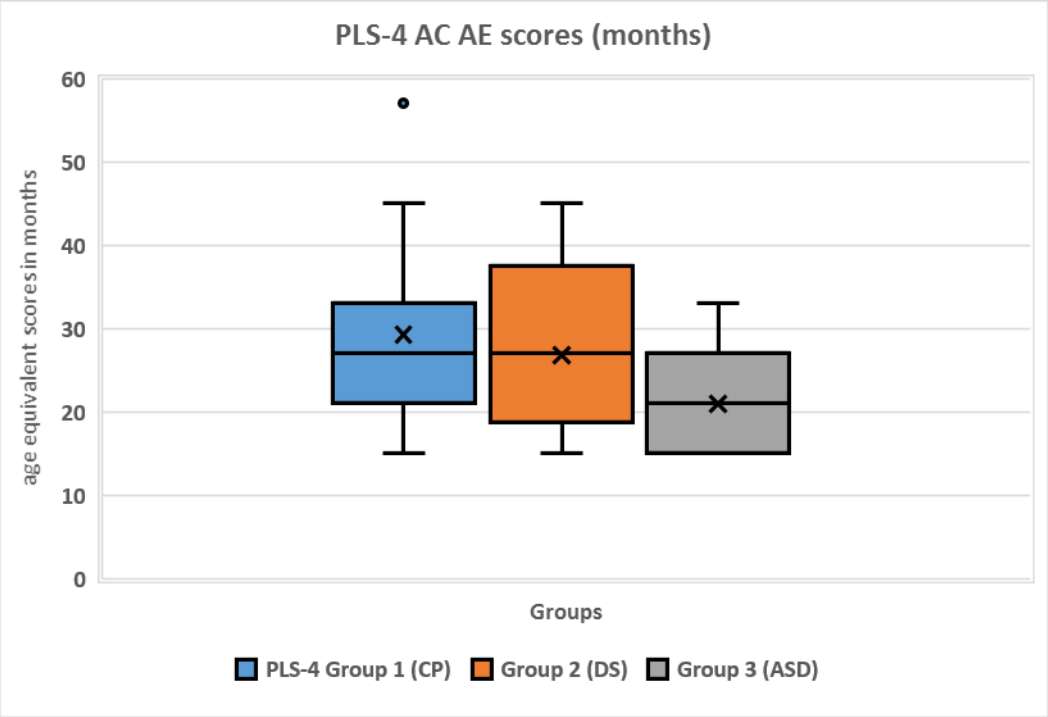

Figure 8-3: *PLS-4* AC age equivalent scores in months (all three groups)

As before, the impact of the outlier was examined, with calculations made in two ways (outlier included, and then outlier excluded). Levene's statistic was used to test equality of variances. The result from this statistic with the outlier *included* was *not* significant ($p = 0.40$) and the assumption of homogeneity of variances was therefore not violated. Similarly, when the outlying data points were *excluded*, Levene's statistic ($p = 0.54$) suggested that, for this data also, there were equal variances across the groups.

In consequence, and to further assess the impact of the outlier, two ANOVAs were completed. With homogeneity of variances confirmed, standard one-way ANOVAs

procedures were followed. In both cases, there was no significant difference between the groups: with outlier included F (2,54) = 1.67, p = 0.20, and with outlier excluded F (2,52) = 1.46, p = 0.24.

Again, in the light of conclusions from these calculations, a decision was made to continue to include the outlying data point in further analysis.

In conclusion, then, to the group matching investigations, subjects were considered matched for chronological age, receptive language *(PLS-4 AC)* age equivalent scores, and performance abilities (*Mullen VR)* age equivalent scores.

8.2 BETWEEN GROUP PERFORMANCE ON TARGET MEASURE GAZE-NOTE

Descriptive statistics were explored for the target measure (*Gaze-NoTe*), shown in Table 8-6:

group	mean raw score (max = 28)	range	sd	95% confidence interval
CP (n=32)	14.9	3-25	6.5	12.6-17.3
DS (n=16)	19.3	5-25	5.5	16.4-22.2
ASD (n=9)	8.2	4-16	3.4	5.6-10.9

Table 8-6: *Gaze-NoTe* scores for all three groups

Inspection of these results suggested that there was overlap in interval scores for the two groups CP and DS, but confidence intervals of mean scores did not overlap for the ASD group and either of the other two groups. To investigate this overlap further, a

"jittered" scatter plot (to deal with overlapping data points) was created to show the individual scores on this *Gaze-NoTe* measure for individual children in all three groups. This plot is shown in Figure 8-4:

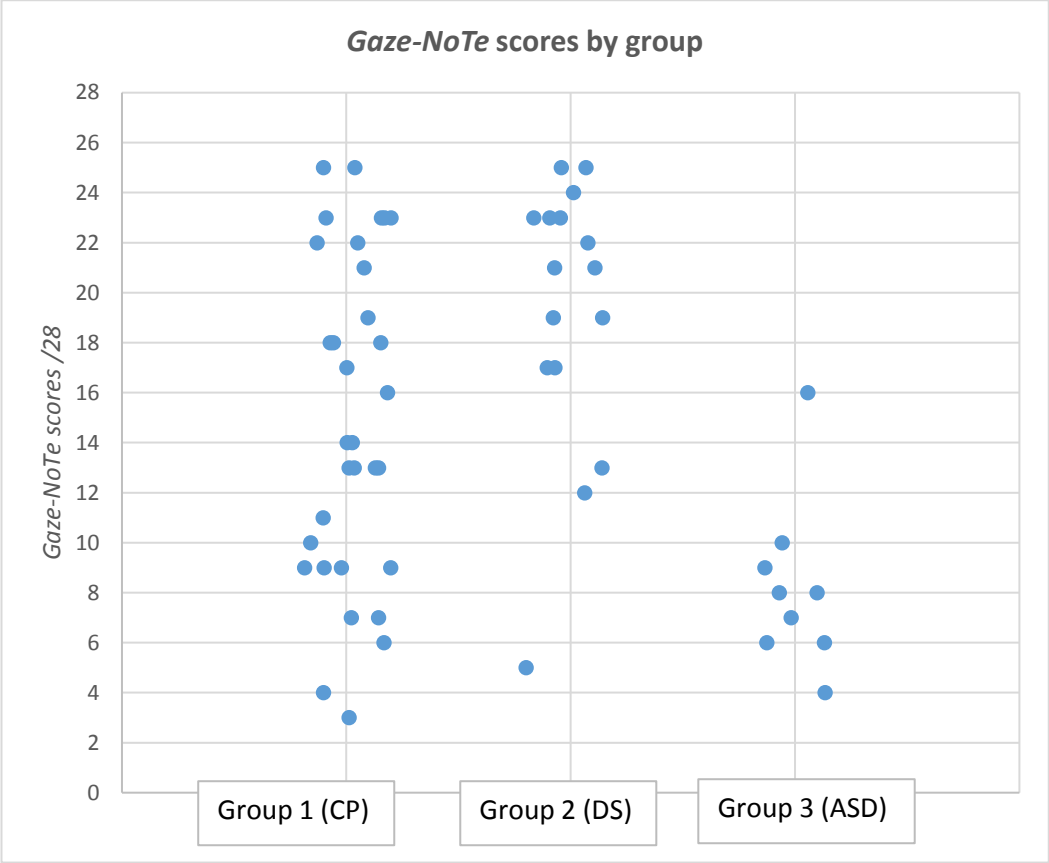

Figure 8-4: Scatterplot showing *Gaze-NoTe* target measure scores for each group

8.2.1 TESTING FOR NORMAL DISTRIBUTION: GAZE-NOTE SCORES

Again, a Kolmogorov-Smirnov test of normal distribution was computed for the target measure *Gaze-NoTe* for each group:

Testing for normal distribution	
Group, children with	**Target measure K-S *p*=**
cerebral palsy	0.200
Down syndrome	0.153
autism spectrum disorder	0.200

Table 8-7: Kolmogorov-Smirnov results for *Gaze-NoTe* distribution (all three groups)

This confirmed that, for all groups, the target measure scores were approximately normally distributed, and, therefore, parametric tests of analysis were adopted.

8.2.2 GROUP COMPARISONS (ANOVAS)

The scores on the target measure *Gaze-NoTe* for the three groups were compared using one-way analysis of variance (ANOVA) procedures. Again, to conduct a robust ANOVA analysis, two further assumptions, in addition to normality distribution, needed to be met:

- homogeneity of variances (comparison of "spread" in the data)
- influence of outlying data points ("outliers")

This examination was considered important in the light of the smaller group numbers, and the presence of an outlier in the DS group (see Figure 8-5):

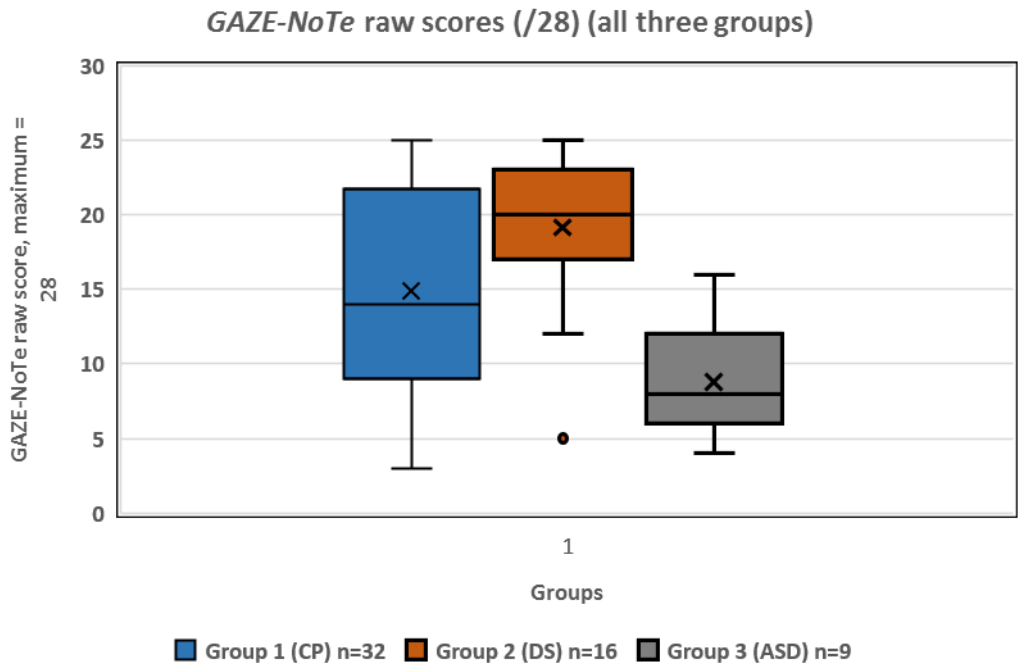

Figure 8-5: *Gaze-NoTe* scores (all three groups)

Levene's statistic was used to test equality of variances. The result from this statistic with the outlier included was statistically significant (p = 0.021) and so the assumption of homogeneity of variances was violated. When the outlying data point was excluded, Levene's statistic (p = 0.001) suggested again that, for this data, there were not equal variances. However, the decision was made to retain the outlying data point.

In consequence of the lack of homogeneity of variances, and mindful of the unequal group sizes, a modified version of the one-way ANOVA was used (Welch's statistic, Games-Howell post-hoc testing):

Welch's F (2,29.9) = 57.7, p = 0.00.

Post-hoc Games-Howell test was used to compare all possible combinations of group difference. This post-hoc analysis revealed that the differences in **performance between each of the three groups was statistically significant** (see Table 8-8). The group of children with Down syndrome had the highest mean scores on this *Gaze-*

NoTe measure: unsurprisingly, the ASD group showed lower scores on these tests with items constructed from those skills known to tap social communication deficits. Table 8-8 shows the results from this Games-Howell testing for performance difference across the three groups:

Groups	*Gaze-NoTe* mean difference	Confidence intervals (95%)	Standard error	*p*=
CP and DS	-5.36	-9.15<>-1.57	1.56	0.04*
CP and ASD	+7.66	4.4<>10.92	1.33	<0.0005*
ASD and DS	-13.02	-16.16<>-9.87	1.25	<0.0005*

*significant at p=0.05

Table 8-8: Games-Howell post-hoc testing for GAZE-NoTe group performance difference

8.2.3 ANALYSIS OF *GAZE-NOTE* SUBSECTIONS: SOCIAL RESPONSIVENESS AND JOINT ATTENTION

The *Gaze-NoTe* measure is comprised of two elements of early social communication: social responsiveness (*Gaze-NoTe* SR) and joint attention (*Gaze-NoTe* JA). The joint attention tasks appear, arguably, later in neurotypical development, and demand greater motor control with fix and transfer of gaze, than the social responsiveness tasks, many of which appear very early in neurotypical development, and can be achieved through directing gaze to researcher's face only.

Follow-up analysis was completed, therefore, for these two components separately.

8.2.3.1 Group comparisons: Social responsiveness

Descriptive statistics were computed for this measure for all three groups, shown in Table 8-9:

***Gaze-NoTe* SR subsection descriptive statistics**			
group	**mean raw score (max = 14)**	**range**	**sd**
CP (n=32)	7.8	2-14	4.2
DS (n=16)	9.8	6-13	2.3
ASD (n=9)	4.3	1-8	2.1

Table 8-9: Descriptive statistics for *Gaze-NoTe* SR (all three groups)

Figure 8-6 shows a "jittered" scatter plot (to deal with overlapping data points) with individual participants' scores (maximum = 14) on this *Gaze-NoTe* SR subsection.

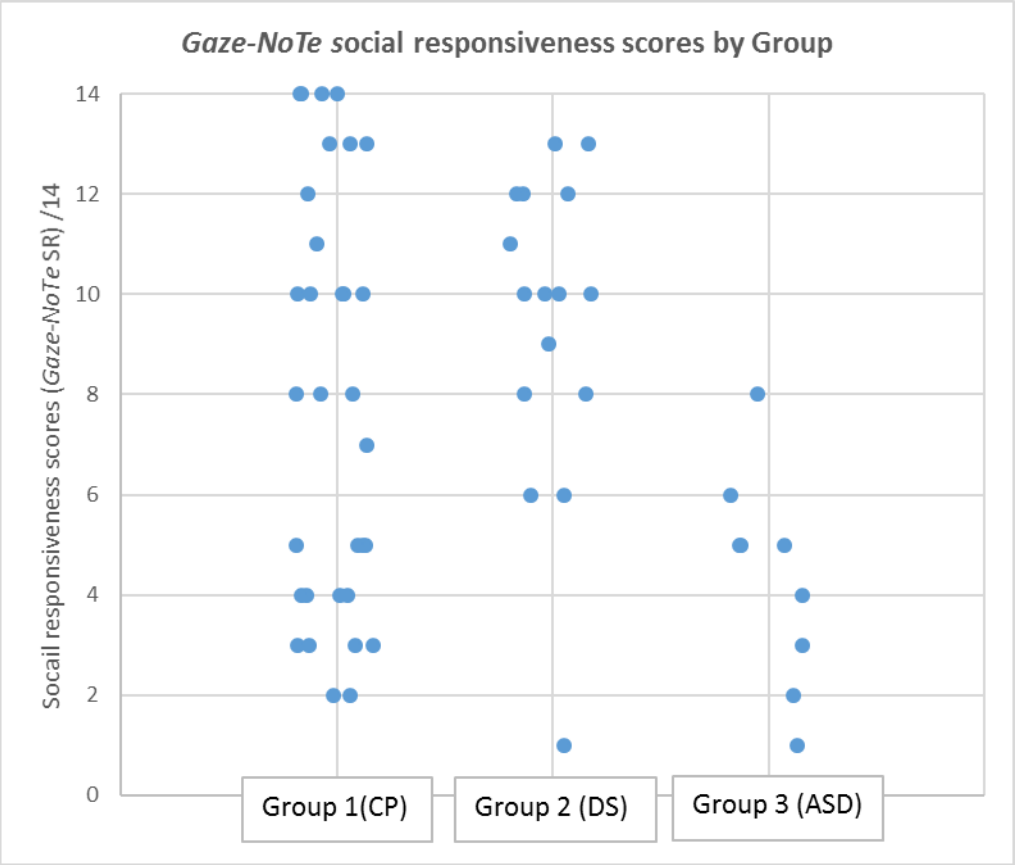

Figure 8-6: Scatter plot showing *Gaze-NoTe* SR scores for each group

Examining the Kolmogorov-Smirnov statistic (in Table 8-10) showed that the *Gaze-NoTe* SR scores were normally distributed for the DS and ASD groups, but not for CP:

***Gaze-NoTe* SR subsection: testing for normal distribution**	
group	**K-S *p*=**
CP (n=32)	0.01
DS (n=16)	0.20
ASD (n=9)	0.20

Table 8-10: Testing for normal distribution for *Gaze-NoTe* SR scores (all three groups)

Subsequent analysis of performance on this *Gaze-NoTe* measure between groups was undertaken using Kruskal-Wallis method of analysis of variance suitable for non-normally distributed data. The result showed a significant difference between the three groups:

$\chi 2$ (2) = 9.64, p = .007

A series of Mann-Whitney post-hoc tests were examined, to determine where the differences between groups lay: the analysis controlled for the possibility of Type 1 error by using Bonferroni adjustment with *p* value set at 0.025. There was no significant difference in performance between the CP and DS groups on this measure of social responsiveness:

U = 201, *z* = -1.21, *p* = 0.226

Similarly, there was no significant difference (with the Bonferroni adjustment) in performance between the CP and ASD groups on this measure of social responsiveness:

U = 77.5, *z* = -2.11, *p* = 0.035

However, significant difference was seen between DS and ASD groups:

$U = 11.5$, $z = -3.44$, $p = 0.001$

These results suggest marked differences between performance on the social responsiveness measure for the DS and ASD groups, as might have been anticipated, but the statistical relationship between the CP group and the other two groups was not significant, suggesting, perhaps, some crossover of performance for this CP group with the other comparison groups.

8.2.3.2 Group comparisons: Joint attention

These procedures were repeated for the joint attention subsection of the *Gaze-NoTe* measure: descriptive statistics (Table 8-11), a jittered scatter plot (Figure 8-7) and Kolmogorov-Smirnov test of normal distribution results (Table 8-12) are shown for all three groups:

***Gaze-NoTe* JA subsection descriptive statistics**			
group	**mean raw score (max = 14)**	**range**	**sd**
CP (n=32)	6.8	1-12	3.2
DS (n=16)	9.6	3-13	2.9
ASD (n=9)	4.4	0-11	3.3

Table 8-11: Descriptive statistics for *Gaze-NoTe* JA (all three groups)

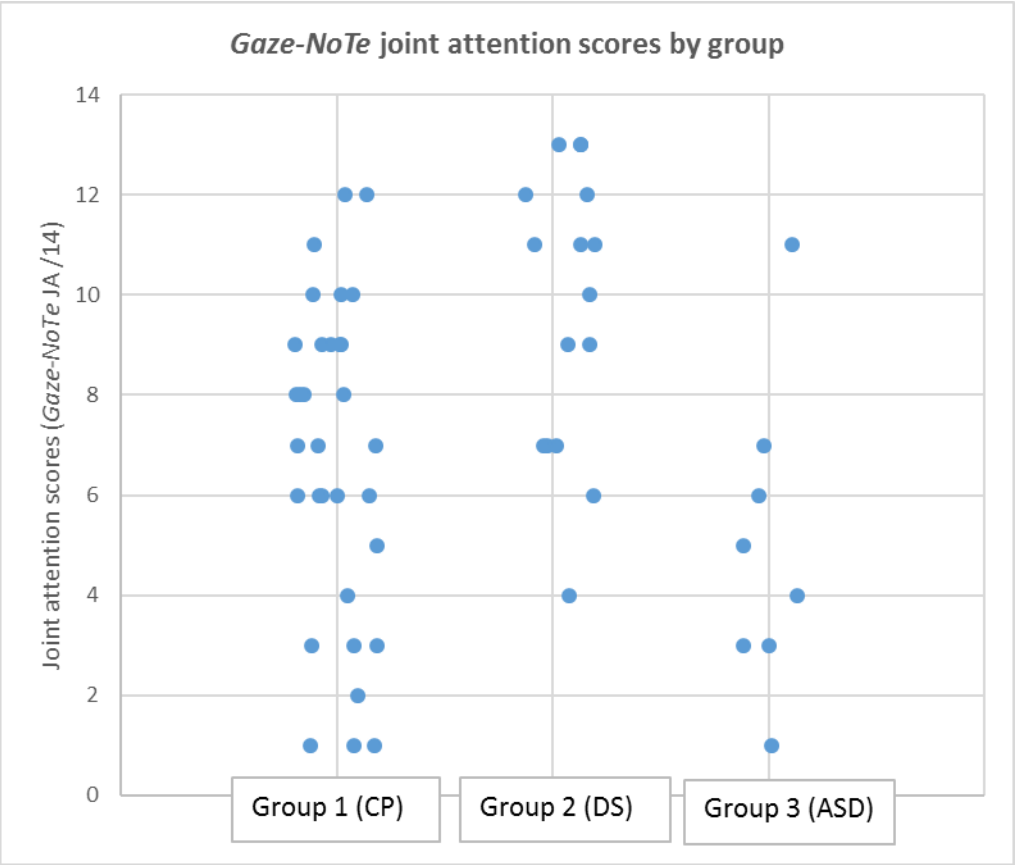

Figure 8-7: Scatter plot showing *Gaze-NoTe* JA measure scores for each group

***Gaze-NoTe* JA subsection: testing for normal distribution**	
group	**K-S *p*=**
CP (n=32)	0.20
DS (n=16)	0.18
ASD (n=9)	0.20

Table 8-12: Testing for normal distribution for *Gaze-NoTe* JA scores (all three groups)

This K-S statistic confirmed that, for all groups, the joint attention scores were approximately normally distributed. There was homogeneity of variances as assessed by Levene's statistic (p = 0.81).

Standard one-way ANOVA analysis showed a statistically significant difference between the groups:

F (2,54) = 10.38, p <0.0005

Post-hoc analysis (using Tukey HSD, in the light of homogeneity of variances confirmed, but unequal groups) examined this difference in more detail, and results are shown in Table 8-13:

ANOVA post-hoc analysis (Tukey HSD)				
Groups	**Joint attention mean difference**	**Confidence intervals (95%)**	**Standard error**	**p=**
CP and DS	-2.94	-5.22<>-0.66	0.94	0.008*
CP and ASD	+2.75	-0.75<>5.55	1.16	0.56
ASD and DS	+5.68	2.59<>-8.78	1.28	0.001*

Table 8-13: *Gaze-NoTe* JA ANOVA post-hoc analysis (all three groups)

This confirms the DS group as a statistically separate group on the performance of this joint attention measure, and children in this group are out-performing both CP and ASD group members. Interestingly, there was no significant difference in performance on these measures tapping joint attention, between the CP and ASD groups, although this difference did approach significance.

In summary, as a composite measure of early communication skills, it did seem to be valuable to include both social responsiveness and joint attention in the *Gaze-NoTe* measure, and this composite showed all three groups to be significantly different from one another.

However, in examining individual components of *Gaze-NoTe*, the CP group performed similarly to both the DS and ASD groups on the social responsiveness items, although there was a marked difference in performance between the DS and ASD groups.

Furthermore, on the measures of joint attention, the CP group appeared to perform similarly to the ASD group, and differently from the DS group.

These results do need to be interpreted with caution, because of the small group numbers involved, and the observation that the relationship in joint attention performance between the CP and ASD groups approaches significant difference.

8.2.3.3 Variation of performance on Gaze-NoTe within the CP group

The group comparisons have suggested that the *Gaze-NoTe* measure differentiates the participants *as a group,* with the children with DS performing better than the CP group and the CP group performing better than the group of participants with ASD. Investigating social responsiveness (SR) and joint attention (JA) subsections separately, there is a slightly different view, in that the JA measure is perhaps suggesting that the CP group is performing similarly to the ASD group, but visual inspection of the data (see Figure 8-7) suggested overlap that warranted further investigation.

To determine if the *Gaze-NoTe* measure, or subsections SR and JA, might be informative in differentiating individuals in the CP group from those in other groups, the scatter plot was examined to identify the threshold of performance that best separates children in different groups.

Figure 8-8 illustrates this cut-off score of 10.5 (marked with a line) in relation to the *Gaze-NoTe* scores for individuals in the three groups: 10/32 children in the CP group

had *Gaze-NoTe* scores falling below this line. There was one child in the DS group, and 8 of the 9 of the ASD group, who showed scores below this "cut-off".

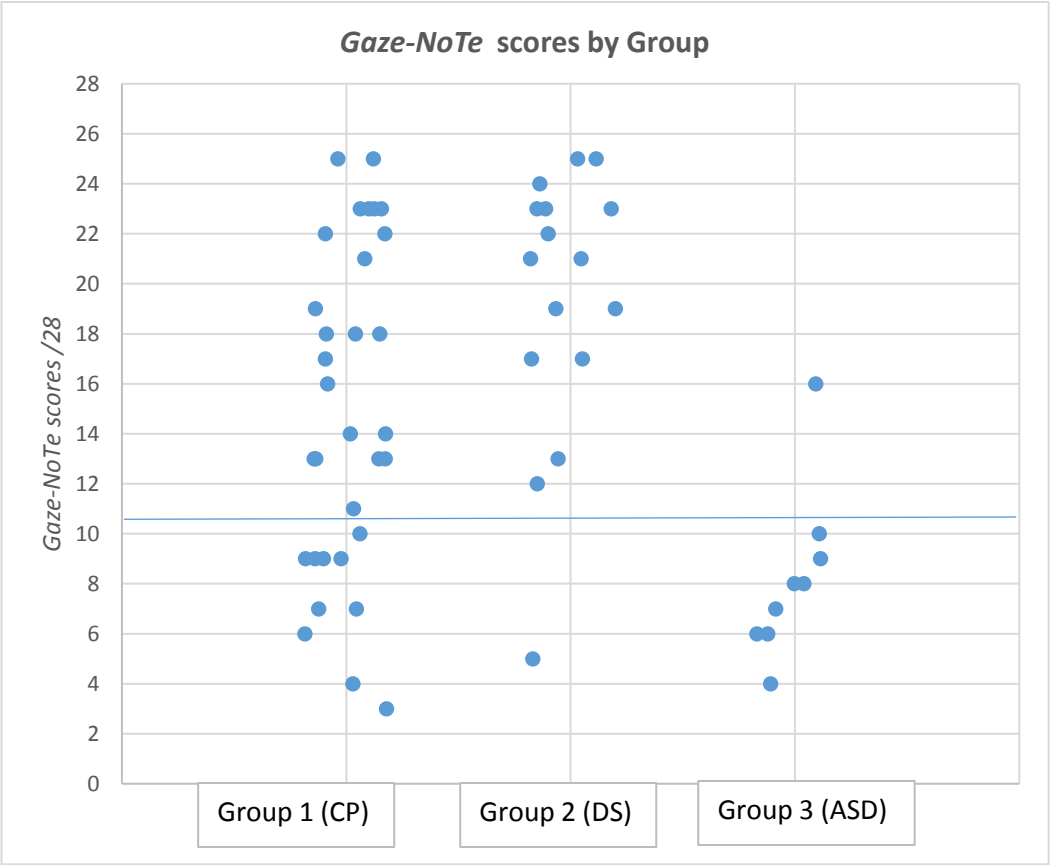

Figure 8-8: Scatter plot of *Gaze-NoTe* scores (all three groups) showing cut-off line

8.3 WITHIN GROUP ANALYSIS (CHILDREN WITH CP)

The analysis so far has seen that while the *Gaze-NoTe* measure has differentiated the three groups, *Gaze-NoTe* subsections SR and JA hint at a more complex relationship between participants in the group. The following procedures were undertaken to examine in more detail the profiles of the target group of children with CP. Given that

both the independent and dependent variables for this group were approximately normally distributed, the following approach to analysis of the data was followed.

A Pearson **correlation matrix** was drawn up to examine the relationship between independent variables *Mullen VR age* equivalent scores and *PLS-4* AC age equivalent scores, and the dependent variable, measure of joint attention and social responsiveness, *Gaze-NoTe.*

Profiles of performance within this group, including relative strengths and weaknesses of the target measure against other abilities, as assessed by background measures, were examined.

8.3.1 RELATIONSHIP BETWEEN *GAZE-NOTE* SCORES AND BACKGROUND MEASURES

Firstly, Table 8-14 shows the correlations between the *Mullen VR* and *PLS-4 AC* age equivalent scores, and the target measure, *Gaze-NoTe* scores:

	Mullen VR AE[1]	**PLS-4 AC AE**	***Gaze-NoTe***
chronological age	-0.28	-0.34	-0.14
Mullen VR AE		+0.83*	+0.70*
PLS-4 AC AE			+0.53*

Table 8-14: Correlation matrix (CP group) for *Mullen VR AE, PLS-4 AC AE* and *Gaze-NoTe* scores

[1]AE = age equivalent
*correlation is significant at the 0.01 level (2-tailed)

There was a significant correlation between social communication skills (*Gaze-NoTe* scores) and both performance abilities (*Mullen VR* AE) ($r = 0.70$, $p<0.01$) and language understanding age equivalent (*PLS-4* AC) ($r=0.53$, $p<0.01$). There was also a significant correlation between the two age equivalent measures (*Mullen VR* and *PLS-4 AC*) ($r=0.83$, $p<0.01$).

However, chronological age did not correlate with either of the *Mullen VR* and *PLS-4 AC* measures or with social communication skill (*Gaze-NoTe*). The high correlation between the measures targeting performance abilities and receptive language skills (*Mullen VR* and *PLS-4 AC AEs*) suggested that there was no significant evidence of discrepancy scores in this group between these two sets of skills.

Secondly, a logistic regression was performed to ascertain the effect of social communication skills measures via *Gaze-NoTe* on the likelihood that children were classified as GMFCS IV or GMFCS V; that is, according to severity of motor function.

Linearity of the continuous variables with respect to the logit of the dependent variable was assessed via the Box-Tidwell (Box & Tidwell, 1962) procedure. The continuous independent variable (*Gaze-NoTe*) was found to be linearly related to the logit of the dependent variable, and, therefore, a logistic regression analysis was run.

The logistic regression model was not statistically significant

$\chi2(2) = 0.009$, $p = 0.925$

suggesting that *Gaze-NoTe* score did not predict GMFCS (Rosenbaum, Palisano, Bartlett, Galuppi & Russell, 2008) category in this sample.

8.3.2 PROFILES OF CP CHILDREN WITH LOWEST *GAZE-NOTE* SCORES

The performance profiles of children are presented in Table 8-15 to look in more detail at children within the CP group, and especially at those children who are performing particularly poorly on the *Gaze-NoTe* measure; below the cut-off score of 10.5, a pattern much more typical of the children with ASD:

Profiles of CP children with lowest *Gaze-NoTe* scores					
ID	***Gaze-NoTe* (score)**	***Mullen VR* (ae months)**	***PLS-4* AC (ae months)**	**Functional Gaze Control % score (fix)**	**Functional Gaze Control % score (fix and transfer)**
21	3	14	15	80	100
30	4	10	15	90	75
27	6	14	15	60	25
2	7	45	45	100	100
23	7	9	15	80	75
1	9	24	21	80	75
6	9	12	21	90	63
14	9	17	15	90	100
26	9	13	15	100	100
4	10	14	21	70	n/a

Table 8-15: Profiles of CP children with lowest *Gaze-NoTe* scores

The developmentally younger children, with language understanding skills at 15 and 21 months, do appear to be over-represented in this group of lower social communication skills, though not without exception (Child ID2 has a *PLS-4* AC age equivalent score of 45 months). The *Functional Gaze Control* screening scores are shown, to confirm again that the children in this group (apart from, possibly, Child ID27, and Child ID4 (for whom there was only *fix* data available) *do* have the visuo-motor abilities to complete these tasks.

A further investigation was made to look for any developmental progression in the *Gaze-NoTe* scores: as this lower-performing group were largely younger developmentally, the breakdown of their *Gaze-NoTe* scores might reveal heavier weighting for earlier (social responsiveness) items over later occurring (joint attention) items. Both SR and JA subsections yielded a maximum of 14 points each (maximum total for *Gaze-NoTe* = 28 points). The *Gaze-NoTe* scores from this group of 10 lower-performing children are shown in ascending order (see Figure 8-9):

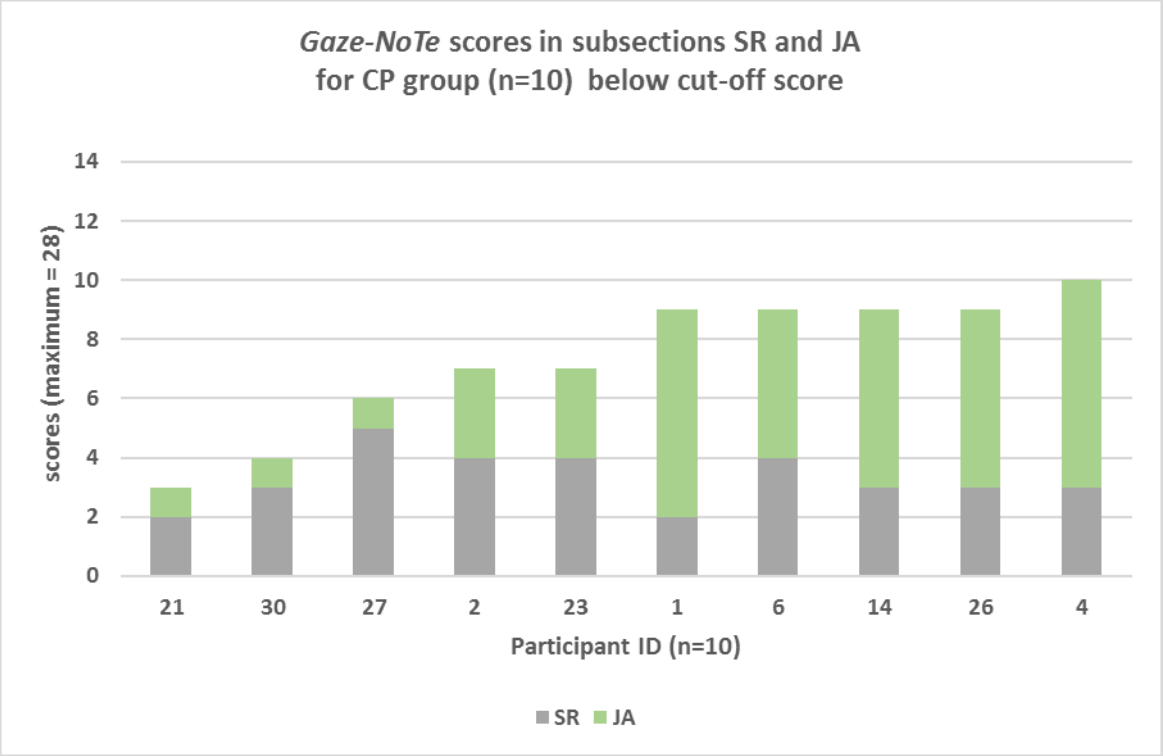

Figure 8-9: *Gaze-NoTe* scores in subsections SR and JA for CP group (n=10) below cut-off score

Figure 8-9 illustrates that when social communication scores are low, it appears to be the result of the contribution of low joint attention scores.

8.3.3 PROFILES OF CP CHILDREN WITH HIGHEST *GAZE-NOTE* SCORES

In contrast to the children who had the greatest difficulty (lowest scores) with the *Gaze-NoTe* tasks (page 8–178), the profiles of children presented in Table 8-16 are those 10 children who recorded the highest scores on the *Gaze-NoTe* measure.

Profiles of CP children with highest *Gaze-NoTe* scores					
ID	***Gaze-NoTe* (score)**	***Mullen VR* (ae months)**	***PLS-4* AC (ae months)**	**Functional Gaze Control % score (fix)**	**Functional Gaze Control % score (fix and transfer)**
19	25	36	39	100	100
22	25	36	27	80	100
7	23	29	27	90	50
11	23	45	33	100	88
18	23	50	39	100	100
31	23	48	57	100	75
13	22	31	21	100	88
20	22	43	33	100	100
17	21	34	33	90	75
28	19	33	27	100	88

Table 8-16: Profiles of CP children with highest *Gaze-NoTe* scores

Again, this data was re-examined in the subsections of social responsiveness and joint attention components of the *Gaze-NoTe* score: both SR and JA subsections yielded a maximum of 14 points each (maximum total for *Gaze-NoTe* = 28 points). The *Gaze-NoTe* scores from this group of 10 higher-performing children are shown in ascending order (see Figure 8-10):

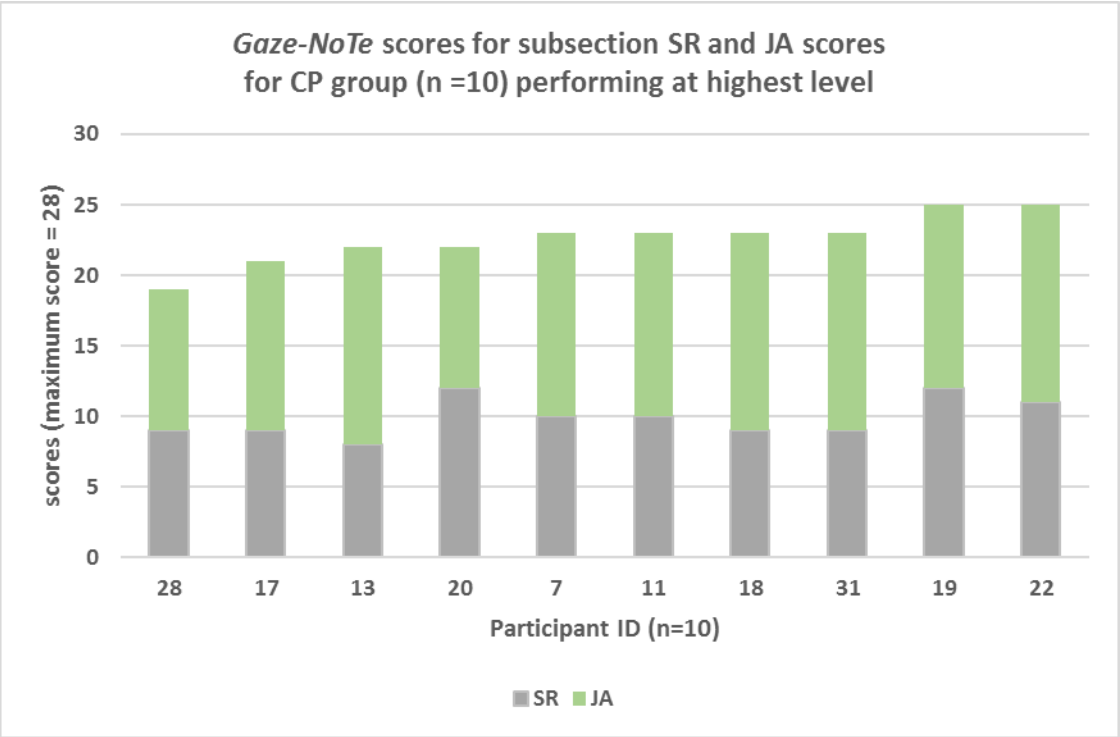

Figure 8-10: *Gaze-NoTe* high scores in subsections SR and JA for CP group (n=10)

8.3.4 JOINT ATTENTION SCORES

To examine any evidence of developmental progression, the relative contribution of response and initiation of joint attention to the overall joint attention score was examined. Visual inspection of the data suggested that the percentage of joint attention score accounted for by rJA was greater than the percentage derived from iJA (Figure 8-11):

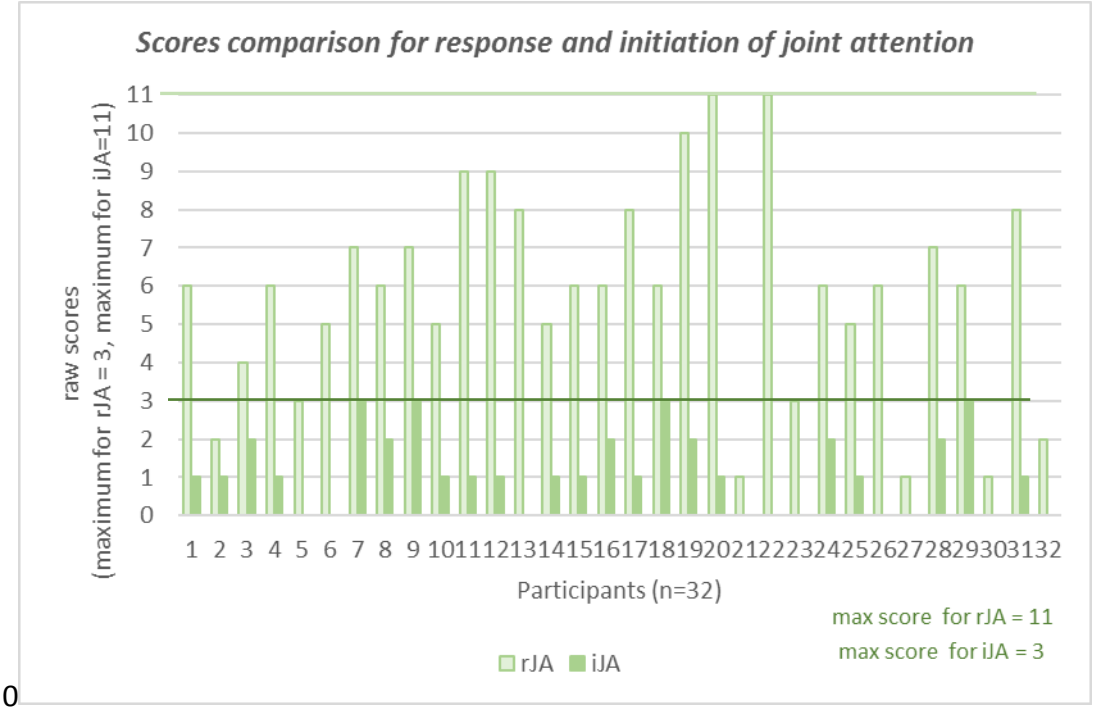

Figure 8-11: Scores comparison for response and initiation of joint attention

9 DISCUSSION

This chapter will discuss interpretations of the study results in the light of the questions arising from clinical practice, and how these results fit in with existing knowledge and practice. The clinical implications of the study findings will be outlined, and the chapter will include a discussion of limitations of this current study, and the recommendations for further research. Finally, the chapter will present reflections on the impact the study findings have had on the author's professional knowledge.

Results will be discussed with reference to the study aims:

- to develop an assessment protocol to support the identification of autism spectrum disorder in children with CP at GMFCS levels IV and V
- to compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)
- to investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

9.1 DEVELOPMENT OF AN ASSESSMENT PROTOCOL (ASD AND CP)

The impressions from the researcher's clinical work was that there was some evidence that some children with cerebral palsy had social communication difficulties "out of step" with other aspects of their development. The revised description of cerebral

palsy included reference to autism spectrum disorder as an associated disability (Rosenbaum et al, 2007). This had certainly been observed and documented in the published literature (for example, Nordin & Gillberg, 1996; Christensen,2008) especially for children with less severe motor deficits (GMFCS I and II; Rosenbaum et al, 2008). For these children, who could manipulate toys and objects to access standard assessments of social communication difficulties, it was possible to discuss an additional diagnosis of autism spectrum disorder (Goodman & Yude, 2000).

However, for children with more severe motor impairments, the route to describing any discrepancy in developmental skills was not an obvious one. Clinical observations had noted children who appeared to underuse their gaze skills, failing to develop effective eye contact, or failing to use gaze to signal messages, despite having demonstrable visual acuity and performance abilities to suggest that they were at a stage so to do.

The researcher and the clinical team had often debated the value of a comorbidity diagnosis to children and families: the risk involved in such identification included lack of clinician confidence in available diagnostic methods, and causing additional family stress with another “label” that would inevitably decrease expectations for future development. The risk of not identifying these difficulties, however, meant that children were offered approaches and targets in learning and communication that were often misdirected or very difficult to achieve.

This study set out to investigate how such social communication deficits could be identified in children with CP with severe motor impairments, little or no speech and intellectual disability.

Results of the study have shown that some children with cerebral palsy do appear to have deficits of social communication, with skills that "lag behind" their other developmental abilities. These deficits cannot be addressed without reference to children's developmental level in other areas, and to their functional vision skills, which need to be in place, at least at the level of fixing gaze, and transferring gaze, to achieve even early social communication skills such as social responsiveness and joint attention. Such skills need to be identified before the discussion of use of gaze direction can take place (Clarke et al, 2016). In the identification of children with cerebral palsy for the study group, there was an unexpected finding. Despite the apparently clear inclusion criteria, 34 of the 66 children consented to participate had to be excluded from further involvement in the study, following administration of the background measures, with 25 of these 34 children excluded as falling outside the language ability criterion (abilities at a 12-54 month level) or the functional vision criterion (using or expected to use vision in communication).

It may be that our inclusion criteria, or their importance to support the research design, were not as clear to referring SLTs as they were to the researcher. It seems more likely that referring SLTs, understandably for this complex group of children, lacked confidence or competencies to appraise functional vision, and may have also lacked resources, confidence or competencies to complete language assessment tasks for the children proposed for the study. Certainly, several of the SLTs involved in the referral to the study spent time with the researcher reviewing the findings from assessments, and requested further information about the assessment procedures used.

The SLTs referring children to the study also showed interest in the assessment procedures for looking at early social communication skills in this group (*Gaze-NoTe*), and in the discussion of the co-morbidity diagnosis with autism spectrum disorder. It did not, however, seem appropriate to pose *Gaze-NoTe* procedures alone as a diagnostic tool for ASD in this population. Comprehensive appraisal of DSM-5 criteria

for diagnosis (American Psychiatric Association, 2000), and adherence to published NICE ASD management guidelines[17] cautioned the use of any single test as a diagnostic tool for ASD. Furthermore, the published literature in the field guided caution, with studies reporting uncertainty with statements about comorbidity (for example, Christensen, 2008; Kinlincaslan & Mukkades, 2009) given the lack of assessment tools and difficulties in interpreting the responses of children with significant motor impairment.

Consequently, the assessment procedure devised for addressing the first study aim focused on two aspects of social communication skills (social responsiveness and joint attention). These skills were selected for inspection as they appeared early enough in neurotypically developing children to be likely to be seen in children with the receptive language ages of the children tested in the CP group (mean age in months = 28, range = 15-57 months, standard deviation = 11 months).

It was thus only possible to look for such discrepancies by firstly establishing both gaze control skills and levels of receptive language development for the CP group. The literature addressing assessment for children with CP had alerted the researcher to the difficulties involved in language assessment with this group (Geytenbeek, 2010; Watson & Pennington, 2015), as had the high numbers of children excluded from this current study.

However, once those background measures had been gathered, and the *Gaze-NoTe* measure of social responsiveness and joint attention had been constructed, it proved possible to examine and interpret children's skills in these two areas. With careful

[17] https://is.gd/NICEguidelinesASD (accessed September 2016)

design, it was possible to offer opportunities for children with adequate gaze control abilities to demonstrate both social responsiveness and joint attention. *Gaze-NoTe* was an effective procedure for capturing information on these social communication abilities for children in this group. The assessment demonstrated good utility for clinical application.

Furthermore, the *Gaze-NoTe* measure had defined construct validity, devised through focus on those aspects of social communication known to be disordered for children with ASD, and content and face validity was established through reference to procedures already in use with other groups of children (Wetherby & Prizant, 1993; Lord et al, 2001; Luyster et al,2009; Chiat & Roy, 2008). However, there was no gold standard criterion available relevant to non-speaking children with CP to establish any concurrent validity for the measure.

The *Gaze-NoTe* assessment showed good to excellent inter-rater reliability in this small study. All children with CP could complete the tasks in the *Gaze-NoTe* series, and all children appeared to enjoy the toys and activities.

9.2 BETWEEN GROUP ANALYSES: *GAZE-NOTE* PERFORMANCE

This section of the discussion relates to the second aim of this current study:

- to compare the assessment tool (*Gaze-NoTe*) profiles of performance of children with CP with those seen in children with ASD and with children with Down syndrome (DS)

The comparison groups in the study (children with Down Syndrome (DS), children with autism spectrum disorder (ASD)) were matched with the target group (children with cerebral palsy) for chronological age, and on performance abilities (non-verbal performance) and receptive language measures.

Performance on the target measure *Gaze-NoTe* was compared for the three groups. ANOVA results showed that performance between each of the three pairs of groups (CP/DS, CP/ASD and ASD/DS) was statistically significantly different, with the group of children with DS having the highest mean scores for these skills. Unsurprisingly, the ASD group showed lower scores on this measure, which had been constructed from those skills known to tap core ASD deficits.

Follow-up analysis was conducted to examine performance on the two separate components of the *Gaze-NoTe* measure, social responsiveness and joint attention.

The results suggest marked differences between performance on the social responsiveness measure for the DS and ASD groups, as might have been anticipated, but the statistical relationship between the CP group and the other two groups was not significant, suggesting, perhaps, some crossover of performance for this CP group with the other comparison groups. The spread of scores on the social responsiveness measure for children in the CP group was perhaps surprising, with some children failing very early developmental items such as failing to respond to name (2 of the 32 children in the CP group), failing to show requesting repetition of a ready/steady go! activity (14 children out of 32) and failing to return a social smile with a smile (12 children out of 32).

In fact, only half of the CP group (n=16) scored half the possible total for the SR items (maximum score =14): examination of the development of social responsiveness had suggested that these were abilities (responding to name, requesting repetition of a

routine, returning a social smile) which are in place for neurotypically developing children by the age of 9-12 months. With the background measure of language understanding/performance abilities at least at 12-month level, these social responsiveness items scores did, therefore, show significant discrepancy.

This discrepancy has a number of possible explanations. It may be that the activities, or the researcher, or the single "one-off" assessment, did not engage the children sufficiently to encourage them to show the abilities targeted in the test. In mitigation, however, the researcher has 30 years' experience encouraging children's best performance in unfamiliar settings, and there were no recalled occasions when the child's accompanying adults remarked on the child's performance as anything but typical.

For the group of children whose social communication skills are less developed than other areas such as receptive language, there are a few possible explanations. Firstly, from these "snapshot" results, without longitudinal data, it is not possible to know if such discrepancy represents a delay in the emergence of these skills; perhaps as a result of atypical social and communication experiences known to exist for children with complex disabilities (Pennington & McConachie, 1999; Pennington, Goldbart & Marshall 2004) or a disorder more akin to the atypical development seen in children with ASD, with social responsiveness persisting as a significant disability with development, as reported in some studies (Kilincaslan & Mukkades, 2009; Christensen et al, 2014).

The SR measures required the children to be visually attentive to the researcher's face, but not to transfer gaze. It is possible to speculate that measures of joint attention are more physically demanding, requiring both fix and transfer of gaze: the CP children

had shown ability in a non-social context to fix and transfer gaze, with the *Functional Gaze Control* measures.

The results from the separate analysis of the joint attention component of the *Gaze-NoTe* were also interesting: this part of the measure tapped skills seen in children aged 15-16 months (Tomasello et al, 2005); again, below the level of the mean age of the groups, and hence suggesting that these skills might have been expected in the repertoire of the target and comparison groups. ANOVA analysis showed a statistically significant difference between the groups on this measure, and post-hoc analysis confirmed the DS group as a statistically separate group on the performance of this joint attention measure. Children in the DS group out-performed both CP and ASD group members, although, interestingly, there was no significant difference in performance on these measures tapping joint attention, between the CP and ASD groups, although this difference did approach significance. The CP grouped performed statistically differently from the DS group on this measure.

Again, there are several possible interpretations of these results. *Gaze-NoTe* measures of joint attention included both (earlier developing) response to joint attention tasks, and (later developing) initiation of joint attention tasks. There were too few exemplars of each of these to warrant individual statistical analysis, but visual inspection of the data (Figure 8-11) examining the relative contribution of response to joint attention and initiation of joint attention to the total joint attention score was undertaken. This inspection showed an increased contribution from the response to joint attention scores to the total joint attention score. The question of this imbalance representing a delay rather than a disorder of joint attention development might be illuminated if it were possible to study these two components of joint attention in more detail.

It remains interesting that, on this joint attention measure at least, the CP group shows similar performance to the ASD group (although the statistical analysis did approach significance), as this may represent a genuine difficulty with skills in this area, despite the children in the CP group's having the functional visual skills to effect the attention sharing.

In summary, as a composite measure of early communication skills it did seem to be valuable to include both social responsiveness and joint attention in the *Gaze-NoTe* measure, and this composite showed all three groups to be significantly different from one another.

The group comparisons suggested that the *Gaze-NoTe* measure in total with both components SR and JA differentiates the participants as a group, with the children with DS performing better than the CP group and the CP group performing better than the group of participants with ASD. As part of the answer to the second study aim, the variation in individuals' performance on the *Gaze-NoTe* measure within the CP group was examined.

To realise this, the threshold of performance that best separated children in different groups was considered, and appeared to fall, from visual inspection of scatter plot data, at a score of 10-11 from a possible 28-point total (see Figure 8-8).

At this "cut-off" point, 10/32 children with CP, 1/32 children with DS and 8/9 children with ASD showed scores falling below this level. This gave some support for the evidence to identify a group of children with CP, within the wide spread of ability in the CP group, performing more like the children with ASD, and this group of 10 children were examined in more detail in the within-group studies.

9.3 WITHIN-GROUP ANALYSIS: CP GROUP *GAZE-NOTE* PERFORMANCE

This section of the discussion relates to the third aim of the current study:

- to investigate any links, for the children with CP, between social communication deficits skills/deficits and performance on other measures of motor, language, visual and cognitive skills

The most striking aspect of the results of the *Gaze-NoTe* study did seem to be the spread of scores for this measure in the CP group. There were children in the group who had difficulty showing even the earliest occurring abilities, and some children scoring almost to maximum level.

The results from examination of the profile of children with CP suggested that there was significant correlation between performance on the *Gaze-NoTe* measure and both performance and receptive language abilities. Chronological age did not correlate with any of these measures.

These two findings together suggest that the discrepancy with lowered social communication scores is linked to overall intellectual disability, but not linked to maturation (there was no correlation with chronological age).

There was high correlation between the measures targeting performance (non-language) abilities and language understanding skills (*Mullen VR* and *PLS-4 AC* AEs), suggesting that there was no significant evidence of discrepancy in scores in this group between performance abilities and language understanding. This finding is somewhat at odds with some reports for studies reviewed in this thesis, although interpretation

of performance/language measures and their relationship is often made difficult by under-specificity of type and distribution of CP, of visual impairment and of chronological age bands. Other studies (for example Clark et al, 2010)) reported language understanding and performance skills in tandem for children with the Worster-Drought variant of CP.

Gaze-NoTe scores did not predict GMFCS categorisation: this finding supported the view that children in GMFCS level IV and V categories might share many aspects of their profiles, and thus constitute a valid group together for any further study/analyses. This reflected the findings in some reviewed studies, where visual and intellectual disability had been reported at similar levels in these two groups, or at least at very different prevalence from levels I, II and III (for example; Ghasia, 2008; Shevell, 2009).

This finding also suggested that any variation in physical disability between the two GMFCS groups in the study (IV and V) did not affect scores on the *Gaze-NoTe* measure.

Inspection of the profiles of children in the CP group with the lowest *Gaze-NoTe* scores (page 8–178) might offer further insight: the developmentally younger children did appear to be over-represented in the group of the 10 children performing the most poorly on this measure. Only one child in this group of 10 scored above 21 months' age equivalent for language understanding.

There was, also, a group of 10 children who recorded *Gaze-NoTe* scores within the range of scores most associated with children with ASD. Although this current study did not aim to identify ASD per se in the target group of children with CP, it is interesting to compare this percentage of the sample (31%) with the percentage of children identified with ASD conditions in Nordin and Gillberg's 1986 study.

Nordin and Gillberg examined 177 children from a register of Swedish children with intellectual disability, and identified 36 children with possible ASD: of these, 20 (11.3%) were described as presenting with autism, autistic-like condition, or ASD not otherwise specified (distinctions in use at the time of the study).

The difference in these findings may be a result of the *Gaze-NoTe's* measure targeting social communication difficulties only, and Nordin and Gillberg's study was aiming to identify a full triad of impairments towards a comorbidity diagnosis. It may be that children with CP share characteristics with children with ASD, rather than sharing a full co-morbid diagnostic label.

Even if these low scores in this group represent social communication scores at the late end of normal development, the scores are still well below those seen in the group of children with DS, and the group should still be considered "at risk" for further social communication abilities development.

The picture is further complicated by one child in this group with a very limited range of the social communication skills tested, in the context of receptive language and performance abilities both recorded at 45 months' age equivalent, and full 100% abilities on both the *fix* and *fix and transfer* gaze control study tasks. Although there is no case history data to examine for this child, research notes made at the time of the assessment do comment that his motivation was better for self-chosen activities, and this observation may have been part of a wider picture suggesting more persistent social communication deficits.

A further investigation for this group of 10 children was made to look for any developmental links in the breakdown of the components of the *Gaze-NoTe* measure. The five lowest scoring children did seem to show this pattern, with their composite scores comprised in the most part of the, developmentally earlier emerging, social responsiveness items. This data (Figure 8-9) suggested that, when social communication scores were low, it was largely the result of the contribution of low joint attention scores.

It is worth noting, however, that these discussion points are based on small variations of difference between SR and JA scores (3-4 points at most).

There was some evidence of further possible accord with the sequence of development in the examination of the contribution of the response to joint attention and initiation of joint attention scores to the total joint attention scores for this group (Figure 8-11). For this group, 10 of the 32 (31%) children's total joint attention scores were comprised entirely of the earlier developing response to joint attention score.

It should also be restated that the *Gaze-NoTe* test was designed to deal with physical limitations and that the children in the CP group had basic functional gaze control skills in place to complete the demands of the *Gaze-NoTe* tasks. The findings do emphasise, as has been made clear in the literature focusing on children with ASD, that the skills of joint attention are vulnerable in many groups of children with neurodisabling conditions.

9.4 SUMMARY OF RESULTS

In summary of the results, then, this study reports that

- there was a wide spread of abilities in social communication skills noted for the CP group, and this spread was wider than for children in comparison groups of children with Down syndrome and autism spectrum disorder
- some children with CP performed similarly to children with ASD on the measure of social communication skills
- the children with CP performing the most poorly tended to be developmentally younger, but exceptions were noted
- the poor performance in developmentally young children (15-21 months) tended to be associated with their low scores with the joint attention component of the *Gaze-NoTe* measure

9.5 CLINICAL IMPLICATIONS

This study has enabled the author to add significantly to personal professional knowledge in understanding the associated disabilities for children with cerebral palsy. A number of wider implications merit discussion:

9.5.1 IDENTIFYING AND ADDRESSING FUNCTIONAL GAZE CONTROL DIFFICULTIES

This study has revealed the importance of functional vision assessment for children with CP. Visual skills are essential for children who do not speak, for them to learn and interact with their environment. Functional vision for communication involves all of eye health; efficient visual pathways; sufficient visual acuity to identify expression and gesture in conversation partners, and to identify the material (objects/photos/symbols) used in augmentative and alternative communication systems and control of eye movements. Given the significantly increased risk of deficits in these areas for this group of children with more severe cerebral palsy, it is good practice for children to undergo screening/specialist assessment of functional vision.

This need has been recognised in the *National Institute for Health and Care Excellence* (NICE) draft guidelines *Cerebral Palsy: Diagnosis and Management for Children under 25*[18]: although management of sensory disorders was not within the brief of this document, the NICE committee did note that

…(visual) impairment is difficult to pick up in the early stages of visual impairment, particularly if there are problems with communication or learning. Recognition often only occurs when children are of school age, as the impairment becomes more apparent in the learning process. Therefore, the Committee agreed that it was important to regularly assess children and young people with cerebral palsy

Draft guidelines Cerebral Palsy: Diagnosis and Management for Children under 25 p367

Following further development of the *Functional Gaze Control* screening measures by other members of the UCL and hospital teams, it has been shown that these functional

[18] https://is.gd/NICEguidelinesCP (accessed October 2016)

gaze skills can be easily and reliably assessed for children with cerebral palsy who are unable to reach, point or speak, by non-vision specialists. It is suggested that competency at the level of identifying normal/abnormal visual attention should be considered part of the role and responsibilities of SLTs working with children with cerebral palsy, and this could be discussed for inclusion in the *Royal College of Speech and Language Therapy* best practice handbook *Communicating Quality*.

Sargent and colleagues (Sargent, Salt & Dale, 2010) have outlined a specialist paediatric framework for the assessment of children with severe cerebral palsy and suggest that this framework can be applied by specialist paediatric health teams: the benefit of integrated multidisciplinary input for such assessment is highlighted.

Further referral to specialist centres could then be effected if needed, and functional gaze abilities would then be assessed by the specialist multidisciplinary team. These are statutory services based, usually, in tertiary (specialist) NHS centres[19, 20] and accessed by referral through community paediatricians.

Recommendations for management following the identification of functional vision difficulties might include emphasis on continuing to present visual material for the child, but prioritising the auditory channels for "output" of (expressive) communication; use of symbols that are intellectually less demanding (objects/photos rather than line drawings/symbols) and, in conjunction with multidisciplinary team colleagues, focused training for school staff and carers on the implications of functional vision difficulties.

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[19] https://is.gd/CCSFunctionalVisionClinic (accessed August 2016)

[20] https://is.gd/GOSHDevelopmentalVisionClinic (accessed August 2016)

9.5.2 IDENTIFYING SOCIAL COMMUNICATION SKILLS DIFFICULTIES

The review of published research and findings from this study would suggest that it is possible to identify discrepancies in performance in social communication skills for children with severe cerebral palsy. There may be both benefits of doing so, and risks associated with not doing so. Benefits are likely to lie in the use of more appropriate communication interventions, targeting difficulties to ensure that the child's weaker skills are not ignored as a focus of intervention in favour of focus on stronger (language understanding, choice-making for wants and needs) skills.

Conversely, the risks involved in failing to acknowledge the difficulties with social communication highlighted in this study may include poor progress with learning and communication, confusion and anxiety for families attempting to understand their children's communication patterns and misplaced resources with inappropriate techniques and equipment.

Again, the introduction of a discussion identifying possible additional impairments will need to be approached with sensitivity, paced to meet family's needs and offering continued communication with families and clinicians. Both generalist and specialist SLTs will have an important role in these discussions, from identification of social communication difficulties through to implementation of tailored recommendations for strategies and approaches in intervention.

The roles and responsibilities of SLTs working with children have been discussed (page 9–197) in respect of the investigation of functional vision, and the pathway of screening by SLT/community professionals, through to Child Development Centre referral, and to tertiary specialist referral as needed. There are also clinical implications for the identification of social communication difficulties, with a similar pathway of management. There may also be education issues, both for SLTs and teaching staff, to

ensure that confidence and competencies are in place to be able to identify discrepancies in developmental profile, and to offer full discussions to family and carers, including the implementation and review of interventions to support best progress.

Recommendations for children in whom such social communication difficulties have been identified will depend on the developmental level of other skills (language/performance) and the identified "gap" between these and other, social communication abilities.

However, recommended interventions are likely to include approaches which tackle all aspects of the child's needs. For example, although exchange communication systems have largely been marketed and researched using picture symbols as their symbolic base (Bondy & Frost, 1994), the principle of learning communicative exchange may be useful for many children who are beginning communicators but who show reduced social motivation to use "off-the-shelf" communication systems. The non-speaking child who has a pre-prepared symbol book and can point to symbols as they are named but does not use the book for any spontaneous communication, is a frequently encountered case in point. Introduction of an exchange system, in which single photographs of favourite objects or activities are available, may increase motivation and the understanding of communication as a trading system, in the first instance, at least, for imperative (needs and wants) communication. This material can then be presented in early joint attention activities, as partner and child look together for favourite objects which are interleaved with less interesting ones.

SLTs working with children with cerebral palsy and identified social communication difficulties (perhaps this group of children may be indicated as CP-SCD) may also be guided by the slowly expanding evidence-base for ASD intervention efficacy, to select

approaches that will combine well with those approaches working to support the physical and intellectual difficulties of CP. Both parent-mediated and clinician-based interventions have been shown to increase scores on measures of intellectual ability for children with ASD, and some studies have shown gains in behaviour and play (see McConachie et al., 2015 for review).

Joint attention skill acquisition for children with a diagnosis of ASD is known to be very difficult indeed, representing as it does one of the core deficits of the condition. However, studies have reported gains in joint attention abilities for children with ASD: one small-scale study (n=5) used *Pivotal Response Teaching* (an intervention approach based on behaviour modification techniques) (Whalen & Schreibman, 2003) to improve the joint attention abilities of young children with autism.

The approach did not target joint attention directly, but worked to improve pivotal development areas such as motivation and social initiations. This approach involves following a child's interests, imitating and talking about the actions, and arranging the environment to engage the child with their preferred activities and toys. Small-step skills for joint attention, such as looking towards the adult to show a toy, are reinforced and developed through modelling during the session. The results from this intervention were encouraging: the researchers found that all children increased the percentage of "correct" responses to the adult's bid for joint attention, directing their gaze to where the researcher was pointing, and four out of the five children initiated joint attention more frequently in later sessions. Furthermore, the gains in responses to joint attention were maintained over three months. However, initiation of joint attention did not continue to develop over time and only two out five children generalised their skills to new situations.

Murza and colleagues (Murza, Schwartz, Hahs-Vaughn & Nye, 2016) conducted a valuable meta-analysis of 15 included studies of interventions for joint attention. The results of this meta-analysis provided strong support for explicit joint attention interventions for young children with ASD. The authors note, however, that it was unclear which children with ASD responded to which type of intervention. Their findings also noted that no significantly different effects were identified relating to intervention practitioner, frequency of intervention or study design.

Similarly, studies to guide clinicians targeting joint attention for children with cerebral palsy are emerging, and this may be encouraging (Olswang et al., 2014). This randomised controlled study identified experimental and control groups of 18 children aged 10-24 months. The researchers note that all candidates were *considered good candidates for learning triadic gaze TG as a conventional and reliable communication signal*. The inclusion criteria detailed in order to confirm this consideration were as follows:

- severe motor delay as measured by a score of two or more standard deviations on either motor subscale of the *Bayley Scales of Infant and Toddler Development,* Third Edition (*BSID–3*) (GMFCS levels are not stated)
- adequate vision, determined by passing five of the first seven items on the *Visual Reception* subscale from the *Mullen Scales of Early Learning* (Mullen, 1995): this is, in fact, a visuo-cognitive measure, rather than a determinant of adequate "vision"
- adequate hearing judged by passing four of the first six items on the *Receptive Language* subscale (Mullen, 1995), together with satisfactory behavioural observation of hearing behaviour
- evidence of showing interest in toys and people, shown by changes in body position, facial expression or vocalisation
- evidence of established dyadic gaze (gaze to face/object), and/or direction of gaze

The experimental group received supplementary (to standard practice) intervention sessions from an SLT, focusing on triadic gaze skills. The behavioural change-based intervention included providing communication opportunities, waiting for the child's response, recognising the child's attempt, and guiding or shaping performance towards triadic gaze.

For this experimental treatment, the SLT researcher offered request and choice opportunities during the play session. For request trials, after brief play with a toy, the SLT would pause, hold the toy out of reach and say *do you want more?* For choice trials, the SLT offered two toys, and said *which one do you want?* A 15-second pause was added in to offer time for response. Successful examples from the child of triadic gaze were rewarded by being given the toy. If no triadic gaze was seen, play re-commenced with a narrative comment such as *you're looking at the ball.* The authors comment that this dyadic looking was then shaped towards triadic gaze, through visual, verbal or tactile means as best dictated to suit the child.

Olswang and her team claimed that triadic gaze episodes increased for both groups of children, with the experimental group showing a slight increase in change over the control group. The researchers are, understandably, cautious about how generalisable these findings are, and recommend ongoing research in this area.

These studies are particularly encouraging in the light of the fact that the identification of a deficit in children's development, especially as an additional disability, without some understanding of how to approach its amelioration, would be very difficult indeed to present to families. For the clinician(s) discussing their concerns after observing social communication deficits, their understanding of the parents' understanding of autism will be imperative. Public understanding of autism spectrum

disorder is still very varied, and while some families may have observed their children's patterns of communication and linked them to autism-similar characteristics, others may have great difficulty accepting the possibility of a further restriction on their child's development.

9.5.3 IDENTIFYING AND ADDRESSING ASD IN CHILDREN WITH SEVERE CEREBRAL PALSY

The *National Institute for Health and Care Excellence* (NICE) guidelines *Cerebral Palsy: Diagnosis and Management for Children under 25*[21], currently in draft form, will alert practitioners to the increased prevalence of ASD in children with CP in the *Mental Health* section of the document. The advice given is for practitioners to follow the relevant NICE guidelines for identifying and managing mental health problems and psychological and neurodevelopmental disorders in children and young people with cerebral palsy.

This recommendation is welcomed, and for children with cerebral palsy able to access the gold standard assessments identifying ASD, will support SLTs in offering appropriate and relevant interventions to children and advice to families.

However, the review of studies, and the findings from this study, would suggest that the current body of clinical knowledge, and the availability of assessment tools, may not be sufficient to identify ASD in this population of children with the same rigour with which ASD is identified as a primary description, or is identifiable in children with

[21] https://is.gd/NICEguidelinesCP (accessed October 2016)

less severe motor problems, and/or speech use, and/or skills and resources to access complex aided communication.

The summary offered by Absoud and colleagues, investigating social communication difficulties assessment for children with severe visual impairment is apposite here:

Clinicians may understandably be reluctant to make an early diagnosis of ASD in the absence of validated objective measures to support clinical indices of suspicion
(Absoud, Parr, Salt & Dale, 2011; p287)

Absoud and colleagues' position would be reflected in the findings of this current study, supporting the clinical recommendation of the identification of social communication skills deficits and vulnerabilities, but, in the absence of appropriate evaluation methods, falling short of recommending ASD diagnostic evaluations.

9.5.4 IMPACT OF IDENTIFICATION OF ADDITIONAL DISABILITIES

There is little published discussion to guide the clinician on the impact on children and families of identifying additional disabilities. Studies of "multimorbidity" have, to date, been centred on long-term health conditions; arthritis, diabetes, and heart disease[22]. There may, however, be some useful insights from this work, which emphasises four key challenges: managing interventions for more than one condition at the same time with confidence; ensuring best quality of provider-patient communication; making

[22] https://is.gd/multimorbidity (accessed November 2016)

difficult decisions about what to prioritise, and co-ordinating care from different professionals.

These challenges may well be relevant to families seeking support for children with complex profiles of cerebral palsy, who have difficulties with functional vision and/or social communication as described in this study. The clinical implications may involve additional reflection on how this, most possibly, unwelcome news is shared, and reference to parental understanding questionnaires (for example *Parental Understanding of Neurodisability Questionnaire* PUN-Q (Dale, Moran & Pote, 2012)), and interview protocols for communicating diagnoses may be helpful.

9.6 STUDY LIMITATIONS

There were, undoubtedly, some limitations to this study, as a single practitioner investigating a group of children with complex profiles.

For the recruitment of children with CP, the list of inclusion criteria by itself did not appear to enable the referring SLTs to select the children we had hoped to target: this may have been under-confidence or competency difficulties for SLTs assessing children's functional vision and language understanding. A total of 66 children were tested, with 32 meeting the inclusion criteria, and invited into the social communication abilities section of the study.

The delay in identifying sufficient numbers of children for the target group did then mean that our comparison group of children with DS was a smaller group than originally planned (n=16, rather than n=32). This reduced the impact of the comparison

group somewhat: the group of children with ASD had been planned as a small number, with the purpose of confirming that our tasks based on “red flags” for ASD had validity.

In future study, it will be helpful to meet with referring clinicians to discuss how the inclusion criteria might be readily and reliably assessed, with follow-up screening review of participants prior to inclusion.

Sources of possible bias in recruitment include the referral population characteristics to a specialist service that accepts UK-wide referrals, although this was mitigated by having an additional pathway of referral through SLTs in special schools. Participants referred via this route, however, may be over-represented by children about whom their families had concern, and whose families were keen to engage with the specialist research team. This may have been the case for the children in the comparison groups too. Further studies will widen the population of children invited, through a greater number of schools, both mainstream and special, in a wider geographical area.

For the children with ASD, it would have been beneficial to have had a larger group for comparison: however, these are a group of children who are frequently approached for recruitment into developmental studies. Furthermore, the *Gaze-NoTe* assessment protocol was constructed to highlight the very difficulties associated with the communication profiles of children with ASD, and it became quickly apparent in the administration of the test that this group of children were indeed failing as expected. Within the time frame and resources available to the project, and children’s poor performance, the decision was made to limit the numbers of this group.

The assessments used proved fit for purpose, but did represent a very “cut-down” version of a full communication profile assessment that might have been completed for a much smaller group, or a single subject design. This comprehensive approach to assessment might have supplemented our test-based assessments of receptive

language, performance abilities and social communication difficulties., through the addition of case history details; speech, language and communication history including any trials or use of AAC and expressive language communication assessment. It might have been insightful to include a number of such case studies. In any future study, it might be possible to invite SLTs from specialist or AAC networks to undertake the assessments, in a multi-centre project.

Parent/carer interview material, carefully adapted and selected, might also have added extra assessment information, and may have added clarity to the results and conclusions of the study. This interview material would add in important details about family history of speech and language or autism spectrum disorders.

The test adaptations for background measures did mean that the age equivalent levels associated with children's scores could not be directly calculated according to the norm-references of the two intellectual ability measures (*Mullen-VR* and *PLS-4 AC*). The age equivalents reported were calculated as carefully as possible, with respect for wide confidence intervals (through reporting only 6-month ranges of age) in the case of *PLS-4 AC*. The exclusion of tasks that could not be completed by hand- or eye-pointing did mean, also, that some important language understanding concepts (for example, prepositions) were omitted from the testing.

The comments repeatedly made through the literature emphasising the need for assessment material designed and standardised for children with physical difficulties are supported by the, albeit considered, attempts made in this study to use existing tests. This would present a difficulty in future research without the development of a robust and reliable assessment protocol for children with motor and visual impairments, and modification of existing tests is likely to persist in the near future.

The target measures developed for the study performed adequately: both the *Functional Gaze Control* screening procedure and the *Gaze-NoTe* early social communication abilities measure would have benefited from further reliability testing (particularly with test/re-test reliability), and further validation with larger samples of children with CP.

Interest in the *Functional Gaze Control* screening procedure has proved significant, and the procedure has been further developed by other UCL staff and students, with the tool now available online for research use. It is hoped that this procedure will offer a useful tool to non-vision specialists working with children with severe CP.

For the *Gaze-NoTe protocol,* full test construction would constitute a significant but valuable contribution to this area of clinical work and study. For the short-term, *Gaze-NoTe* procedures will be valuable as a clinical tool for SLTs, both specialist and generalist, to alert their attention to the possibility of ASD/social communication deficits in this population of children. The procedure also provides a valid measure, based on discrepancy with other skills in the children's communication profile, on which to base a clinical discussion regarding the usefulness of a description of ASD.

Despite many trials and alterations with numbers and placement of video recording equipment, it proved very difficult to video record the subtlety of social communicative signals for the *Gaze-NoTe* tasks in this CP population, and the burden of scoring "online" fell to the single researcher. This has significant implications if the assessment is to be useful, after further development, to other clinicians. Capturing eye contact and gaze direction on video recording is a notorious problem, and one to be solved, perhaps, through technology options (Noris, Nadel, Barker, Hadjikhani & Billard, 2002) for future development of gaze direction assessment tools. In the short-

term, clinicians using *Gaze-NoTe* should recruit a second observer, and undertake reliability examination across different observers and assessment times.

In summary, the results presented in this study relate to the groups under study, and findings would need to be replicated with larger target and comparison groups in order to report any generalisable findings. The conclusions made suggest that SLTs working with children with severe CP should be aware and alert to the possibility of functional vision and social communication difficulties in this population, and some methods for assessment of such disabilities are proposed, but the conclusions are, necessarily, speculative, in the light of the exploratory nature of this study.

Although it is acknowledged that the *Gaze-NoTe* tool would need full psychometric evaluation across a large population, it has shown to be useful, in conjunction with the functional gaze control screening, as a clinical tool for use by SLTs to support a multi-disciplinary discussion of significant social communication deficits in children with severe cerebral palsy. This discussion may incorporate an additional discussion on the relevance of a dual diagnosis, for some children and families, of cerebral palsy and autism.

9.7 FURTHER RESEARCH

The complex relationship between physical, intellectual and visual abilities in children with cerebral palsy is likely to engage research focus for some time to come. Each component (language understanding, performance abilities, speech intelligibility, general health, functional vision abilities, non-speech communication system availability) of development (each “slider” on the graphic equaliser described in

Chapter 1) is difficult to characterise in terms of appraisal and outcome, and their interplay in development is complicated indeed.

Longitudinal and multilevel studies have attempted to map the network of abilities and influences on the outcome of communication abilities in children with cerebral palsy. (Vos et al., 2014) undertook a large study (418 participants across all GMFCS levels and age levels, mean age = 9 years 6 months), aiming to examine the developmental trajectories of expressive and receptive spoken and written language. The close relationship between receptive language abilities and intellectual disability overall was highlighted in their results, as suggested by this current study. The authors comment, though that for children with severe physical or intellectual disabilities, accurate assessment with the tools available (*Vineland Adaptive Behaviour Scales* questionnaire/interview) is difficult. Their conclusions refer to the value of computerised assessments.

Where a multidisciplinary team is available, (assistive technologist, occupational therapist), consideration of alternative access methods might be considered for this group of children. It is interesting to note that there are some promising findings looking at modified test administration using assistive technology (Warschausky et al., 2012), although 95.8% of the children with CP (n=24) in this study were at GMFCS (Rosenbaum, Palisano, Bartlett, Galuppi & Russell, 2008) level I or II, with only 4.2% at GMFCS level IV, and none at GMFCS level V. For this group of physically more able children, at least, it was possible to show stability of measurement between the standard and adapted versions of the psychometric tests used, with forced-choice format “quadrant” (target selected from four choices) tests.

Similarly, studies have investigated the use of computer-based language tests that can be accessed using a touch screen device, switches or eye-gaze access technology. As

noted, one such test has been validated and norm-referenced for Dutch speakers (Geytenbeek, 2010): in the UK, a PC-based assessment *Computer Based Accessible Receptive Language Assessment* (CARLA) has been developed,[23] and can be used with a full range of access methods including eye-gaze access technology, mouse pointers, switch scanning and touch screen displays.

In contrast, single subject experimental designs are also expected to add to knowledge in the characterisation of communication profiles and outcomes for children with CP, and studies focusing on children in GMFCS level V would be welcomed. However, with the goal to identify barriers and support with regard to children's best communication progress, and promote genuine participation, the "in-child" approach described in this study will benefit from insights from more context-sensitive, systems-based studies also.

Nevertheless, the identification of appropriate appraisals (assessment materials and approaches) of abilities for children with severe cerebral palsy might seem a priority for research in this field at this stage, and the findings of this study might suggest that this research should focus on the development of joint attention, and strategies to support such development, and the assessment of functional vision.

Further projects by other UCL team members have been established to develop some of the findings presented in this study. The *Functional Gaze Control* screening assessment has now been made available for free download[24], and is being validated through extensive testing of children with cerebral palsy in UK schools, and through

[23] https://is.gd/carla_assess (accessed November 2016)

[24] https://www.ucl.ac.uk/gaze/gaze-project (accessed October 2016)

the development of a parental/school questionnaire in a follow-on project *Functional Near Vision* (*FunVis*) screening[25].

The findings from this project will also inform the development of an *Eye-Pointing Classification Scale* (*eyePoint*)[26]. The *eyePoint* project aims to establish a tool to further support professionals and families to describe the looking behaviours of children with cerebral palsy. It comprises a systematic scale to assess and classify how children with CP are using their gaze for communication (eye-pointing), and this phase of the project will to test reliability and validity of the scale in practice.

9.8 IN CONCLUSION

Children with severe cerebral palsy, and their families, face substantial challenges to reach optimal developmental outcomes. Clinicians supporting the interventions towards these outcomes can feel daunted by the scarcity of guidelines and good counsel to direct their decision-making. The development of the evidence base to increase understanding of the complex neurodevelopmental processes associated with

[25] https://www.ucl.ac.uk/gaze/funvis (accessed October 2016)

[26] https://www.ucl.ac.uk/gaze/eye-pointing-classification (accessed October 2016)

development in CP will need to be a shared responsibility between those affected by cerebral palsy; practising clinicians of many professions and academic researchers.

Developing valid, objective, strong psychometric assessments will be an important step towards this increase in understanding: this study has explored the beginnings of this process. This study has shown how this might be done, and has shown how such assessment can reveal important variation, both between different groups of children, and within the group of children with cerebral palsy at GMFCS levels IV and V.

The protocols developed for functional vision screening, and for the assessment of communication skills (*Gaze-NoTe)* are expected to be immediately useful in clinical practice to determine best intervention for the communication deficits of children in this group. Further study to confirm wider test reliability and validity should form the next stage of research.

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11 APPENDICES

11.1 DSM-V CRITERIA FOR THE DIAGNOSIS OF ASD AND SOCIAL (PRAGMATIC) COMMUNICATION DISORDER

11.1.1 AUTISM SPECTRUM DISORDER DIAGNOSTIC CRITERIA [REF: 299.00 (F84.0)]

Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history

- deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions
- deficits in nonverbal communicative behaviours used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication
- deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behaviour to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers

DSM-V gives further guidelines and instructions to specify **severity,** and to note if ASD is present with other impairments of intellectual function, language, or known medical/genetic condition or environmental factors.

11.1.2 SOCIAL (PRAGMATIC) COMMUNICATION DISORDER DIAGNOSTIC CRITERIA {REF 315.39 (F80.89)]

Persistent difficulties in the social use of verbal and nonverbal communication as manifested by all of the following:

- deficits in using communication for social purposes, such as greeting and sharing information, in a manner that is appropriate for the social context.
- impairment of the ability to change communication to match context or the needs of the listener, such as speaking differently in a classroom than on the playground, talking differently to a child than to an adult, and avoiding use of overly formal language
- difficulties following rules for conversation and storytelling, such as taking turns in conversation, rephrasing when misunderstood, and knowing how to use verbal and nonverbal signals to regulate interaction
- difficulties understanding what is not explicitly stated (e.g., making inferences) and nonliteral or ambiguous meanings of language (e.g., idioms, humour, metaphors, multiple meanings that depend on the context for interpretation)

The deficits result in functional limitations in effective communication, social participation, social relationships, academic achievement, or occupational performance, individually or in combination.

The onset of the symptoms is in the early developmental period (but deficits may not become fully manifest until social communication demands exceed limited capacities).

The symptoms are not attributable to another medical or neurological condition or to low abilities in the domains or word structure and grammar, and are not better explained by autism spectrum disorder, intellectual disability (intellectual developmental disorder), global developmental delay, or another mental disorder.

11.2 CLASSIFICATION SYSTEMS FOR CHILDREN WITH CEREBRAL PALSY

Level	GMFCS[27]	MACS[28]	CFCS[29]	Viking Speech Scale[30]
I	Walks without limitations	Handles objects easily and successfully	Sends and receives information with familiar and unfamiliar partners effectively and efficiently	Speech is not affected by motor disorder
II	Walks with limitations	Handles most objects but with somewhat reduced quality and / or speed of achievement	Sends and receives information with familiar and unfamiliar partners but may need extra time	Speech is imprecise but usually understandable to unfamiliar listeners
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and / or modify activities	Sends and receives information with familiar partners effectively, but not with unfamiliar partners	Speech is unclear and not usually understandable to unfamiliar listeners out of context
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations	Inconsistently sends and / or receives information even with familiar partners	No understandable speech.

27 Gross Motor Function Classification System (Rosenbaum et al., 2007)

28 Manual Ability Classification System (Palisano et al., 1997)

29 Communication Function Classification System (Hidecker et al., 2011)

30 Viking Speech Scale (Pennington et al., 2013)

V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions	Seldom effectively sends and receives information even with familiar partners	n/a

Table 11-1: Classification systems for children with cerebral palsy

11.3 *PEDI-CAT* SOCIAL FUNCTION SAMPLE INTERVIEW FORM

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Figure 11-1: *PEDI-CAT Social Function* sample report (Page 1)

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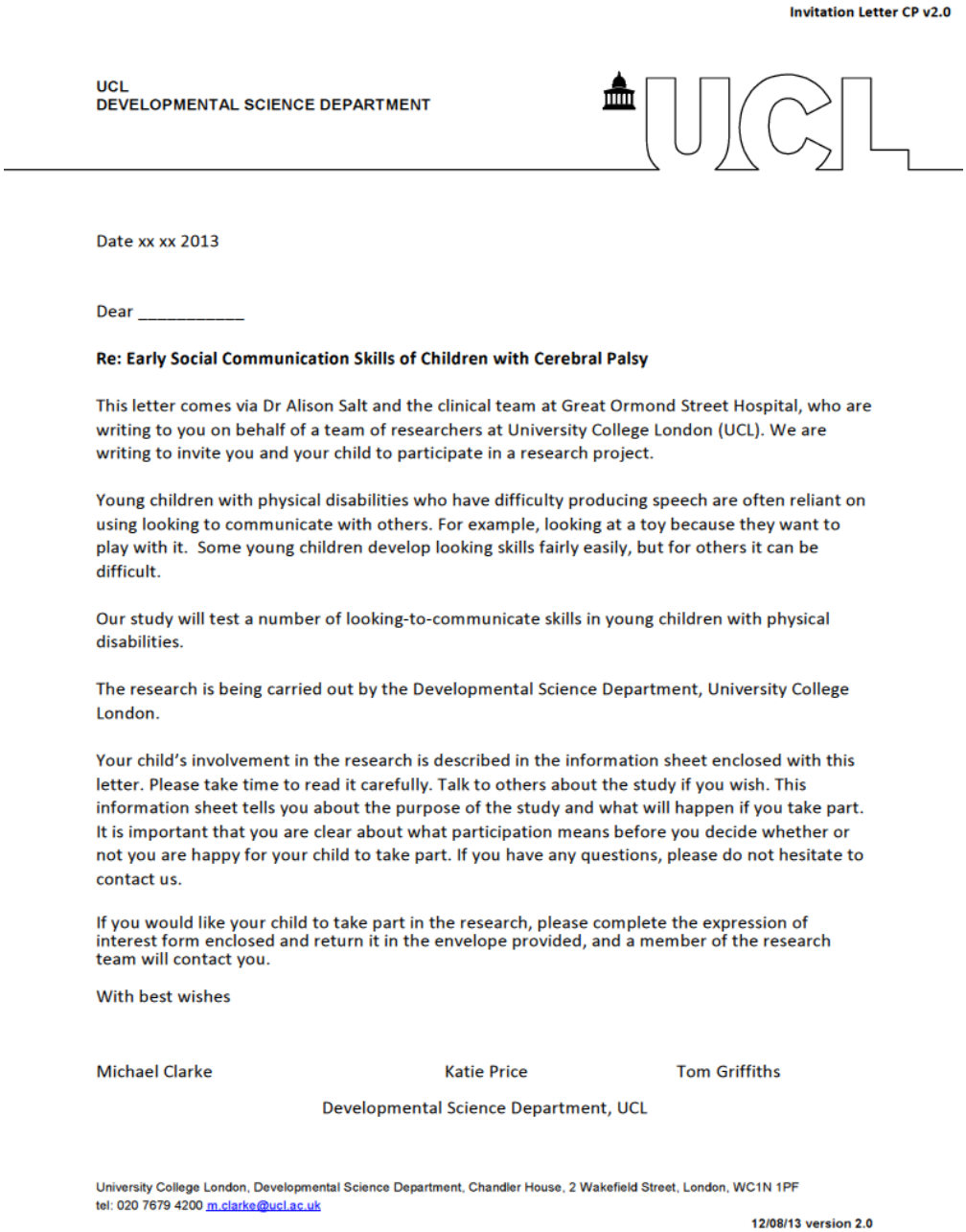
Figure 11-2: PEDI-CAT Social Function sample report (Page 2)

11.4 PARTICIPANTS' DOCUMENTS

11.4.1 INVITATION TO PARTICIPATE (SAMPLE: CP GROUP)



Invitation Letter CP v2.0

UCL
DEVELOPMENTAL SCIENCE DEPARTMENT

Date xx xx 2013

Dear _____

Re: Early Social Communication Skills of Children with Cerebral Palsy

This letter comes via Dr Alison Salt and the clinical team at Great Ormond Street Hospital, who are writing to you on behalf of a team of researchers at University College London (UCL). We are writing to invite you and your child to participate in a research project.

Young children with physical disabilities who have difficulty producing speech are often reliant on using looking to communicate with others. For example, looking at a toy because they want to play with it. Some young children develop looking skills fairly easily, but for others it can be difficult.

Our study will test a number of looking-to-communicate skills in young children with physical disabilities.

The research is being carried out by the Developmental Science Department, University College London.

Your child's involvement in the research is described in the information sheet enclosed with this letter. Please take time to read it carefully. Talk to others about the study if you wish. This information sheet tells you about the purpose of the study and what will happen if you take part. It is important that you are clear about what participation means before you decide whether or not you are happy for your child to take part. If you have any questions, please do not hesitate to contact us.

If you would like your child to take part in the research, please complete the expression of interest form enclosed and return it in the envelope provided, and a member of the research team will contact you.

With best wishes

Michael Clarke Katie Price Tom Griffiths

Developmental Science Department, UCL

University College London, Developmental Science Department, Chandler House, 2 Wakefield Street, London, WC1N 1PF
tel: 020 7679 4200 m.clarke@ucl.ac.uk

12/08/13 version 2.0

Figure 11-3: Invitation to participate sample

11.4.2 INFORMATION SHEET (SAMPLE: CP GROUP)

UCL
DEVELOPMENTAL SCIENCE DEPARTMENT

INFORMATION SHEET
Early Social Communication Skills of Children with Cerebral Palsy

Dear Parent/Carer,

We would like to invite you and your child to take part in a research study. Before you decide, you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Talk to others about the study if you wish. This information sheet tells you about the purpose of the study and what will happen if you take part.

Purpose of the study
Young children with physical disability who have difficulty producing speech are often reliant on using looking to communicate with others. For example, children might look at a toy because they want to play with it. Some young children develop looking skills for use in communication fairly easily, but for others it can be difficult.

Our study will explore a number of looking-to-communicate skills in young children with physical disabilities. Surprisingly, professionals don't yet have reliable ways of testing these skills in children with physical difficulties.

If we can find good ways of identifying children who struggle to develop looking-to-communicate skills, we hope that professionals (speech and language therapists, for example) will be able to work with children and their families more effectively.

We will use a number of simple activities that are easy to carry out and are suitable for young children with physical disabilities. These include seeing how children can move their eyes to show another person that they have seen some toys. We have attached a full description of the games and tasks for you to look at.

We will monitor how your child responds with their eyes to our activities; firstly, by just carefully observing their eye movements, and secondly by using state-of-the-art eye-tracking technology. There are no wires or physical connections involved in the eye-tracking technology. We will simply ask your child to look at a monitor (like a television screen) while the technology automatically tracks their eye movements. This technology has been used extensively with adults, children and babies.

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University College London (UCL)
Chandler House,
2 Wakefield Street,
London, WC1N 1PF

Figure 11-4: Information sheet sample, page 1

Why has my child been invited to take part?
We are writing to you because your child has been identified by members of your child's care team. The project is a collaboration between the Neurodisability Service at Great Ormond Street Hospital, the Developmental Science Department at University College London, and Barnsley Hospital NHS Trust.

Please note that no one outside your child's care team has had access to your personal details in preparing this invitation. Only members of your care team or researchers at University College London will have access to any personal details, such as your address, as a part of your participation in this study.

What would participation involve?
If you are willing for your child to take part in the study, we would like to visit your child and carry out the activities to look at how children use their eyes for communication. A description of these activities is enclosed with this information sheet. After your visit we will provide you with a summary of how your child got on.

Our study is also collecting information on children's physical, speech, language and learning abilities. This sort of information is routinely collected by members of your child's care team. With your permission, we would like to take a note of these skills. If this information has not already been collected, we would like to assess these skills ourselves.

Will you video record my child?
Yes, with your permission we would like to video record the activities. This will help us to see accurately how your child gets on. It is important for you to understand how the recordings might be used, before agreeing that your child can take part. Two different sorts of consent can be given. We have called these: **research** participation, and **wider** participation:

- **Research participation** level of consent means that the video recordings will be used for the research study only.

- **Wider participation** level of consent means that video recordings might be used for teaching (e.g. undergraduate and postgraduate students, and health and education professionals), and at presentations outside University College London, such as international meetings. The videos could also be used in electronic publications such as CD-ROMs and web-based teaching and research resources.

It is important for you and your child to be comfortable with the level of consent that you give. You may change the level of consent or withdraw it completely at any time. However, we cannot accept liability if recordings have already been published. If you wish to alter the level of consent at any time, please telephone Michael Clarke at the Developmental Science Department, University College London (020 7679 4253).

Does my child have to take part?

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Figure 11-5: Information sheet sample, page 2

It is up to you to decide. If, after reading this information sheet, you decide that your child can take part in the study, please complete the consent form and the contact details form enclosed with this letter and return them in the envelope provided. We will then call you to discuss any questions you may have about the study. We would be very happy to meet you with your child if you are available.

If I agree to take part what will happen if I decide not to carry on?
It is important that you are aware that your participation in this study is strictly voluntary. You are free to withdraw your consent at any time without giving a reason. Withdrawing your consent will not affect your child's care.

Will taking part be kept confidential?
Yes. We will follow ethical and legal practice and all information about your child will be handled in confidence.

What will happen to the results of the study?
We will produce a report summarising how your child got on, and we will provide you with a copy of this. We will also discuss with you if you would like a copy of these findings to be sent to your local health and/or education teams. We also aim to publicise our findings through journal articles and through presentations at conferences in the UK and abroad. Your child will not be identified explicitly in any publication or presentation.

Who is organising and funding the research?
The research is being organised by the Developmental Science Department, University College London in collaboration with Great Ormond Street Hospital, London. The study forms part of the course of two doctoral students, Katie Price and Tom Griffiths, who are based in the Developmental Science Department, University College London, and who are also clinical staff at Great Ormond Street Hospital.

Who has reviewed the study?
This research study has been looked at and given a favourable opinion by an independent group of people called a Research Ethics Committee to protect your safety, rights, wellbeing and dignity.

What if I have questions about the study?
Please do not hesitate to contact Michael Clarke at University College London (020 7679 4253 m.clarke@ucl.ac.uk), Katie Price at katie.price@ucl.ac.uk or Tom Griffiths at tom.griffiths.11@ucl.ac.uk if there is anything that is not clear, or if you would like more information.

What if I have a problem with the study?
If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated by members of staff or about any side effects (adverse events) you may have experienced due to your participation in the research, the normal National Health Service or University College London complaints mechanisms are available to you. Please ask members of the

Figure 11-6: Information sheet sample, page 3

research team if you would like more information on this. Details can also be obtained from the Department of Health website: http://www.dh.gov.uk.

In the unlikely event that you or your child is harmed by taking part in this study, compensation may be available. If you suspect that the harm is the result of the Sponsor's (University College London) or the Hospital's negligence then you may be able to claim compensation. After discussing with the research team, please make the claim in writing to the Dr Michael Clarke who is the Chief Investigator for the research and is based at the Developmental Science Department, University College London (UCL), Chandler House, 2 Wakefield Street, London, WC1N 1PF. The Chief Investigator will then pass the claim to the Sponsor's Insurers, via the Sponsor's office. You may have to bear the costs of the legal action initially, and you should consult a lawyer about this.

What next?
If you are interested in taking part after reading about the study, please complete the *consent form* and the *contact details* form enclosed with this letter and return them in the envelope provided. We will then call you to discuss any questions you may have about the study and to confirm when we would like to visit your child.

Best wishes

Michael Clarke, Katie Price, Tom Griffiths & Samantha Wallis
Developmental Science Department, UCL

Developmental Science Department
University College London (UCL)
Chandler House,
2 Wakefield Street,
London, WC1N 1PF

Figure 11-7: Information sheet sample, page 4

Description of Activity	What will be required of my child?	How long will it take?
Observation of your child's motor abilities using a standard, published scale	Nothing. Observations will be made by the research team.	5 minutes
Observation of the type and distribution of cerebral palsy	Nothing. Observations will be made by the research team.	5 minutes
Intelligibility of your child's speech will be categorised using a published scale	Nothing. Observations will be made by the research team.	5 minutes
Intelligibility of your child's speech will be assessed through speech recordings as part of a published measure	Your child will need to repeat 50 words read out by an assessor.	10 minutes
Observation of your child's communication patterns will be made using an existing standard measure of their abilities as a receiver and giver of messages	Nothing. Observations will be made by the research team.	5 minutes
Assessment of language understanding and language use will be made using a standard measure, adapted for children with physical disabilities where necessary	Your child will be asked to look at toys and pictures as they are named by the assessor.	15 minutes
Assessment of your child's visual understanding (without language)	Your child will be asked to complete patterns in puzzles by looking at target pieces.	10 minutes
Assessment of your child's ability with general eye movement skills	Your child will be asked to look at shapes as they move across a screen.	10 minutes
A structured play session with a book, a puzzle and wind-up toys	Your child will play alongside the assessor with these toys.	10 minutes
Assessment of using eye gaze for communication will be made using a standard measure, adapted for children with physical disabilities where necessary	Your child will watch as the assessor presents toys, and have the opportunity to show with their eyes their observational and matching skills (for example, matching a miniature spoon to a large spoon).	15 minutes
Observation of how your child attends to biological motion (looking at a human shape as opposed to a random shape) will be carried out using eye-gaze technology	Your child will be asked to look at videos on a computer.	5 minutes
Observation of your child's response to noises in the environment	Your child's responses to a range of different noises will be noted.	5 minutes
Assessment of your child's understanding of other people's thinking	Your child will be asked to watch videos on a computer.	15 minutes
		115

Figure 11-8: Information sheet sample, page 5

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11.4.3 CONSENT FORM SAMPLE (ASD GROUP)

UCL SPEECH & LANGUAGE THERAPY
DIVISION OF PSYCHOLOGY & LANGUAGE SCIENCES

Early Social Communication Skills of Children with Autism Spectrum Conditions:
Using eye gaze for social communication

CONSENT FORM

Name of Project Supervisor: Dr Michael Clarke
Name of researcher: Katie Price

This study has been approved by the University College London research Ethics Committee (Project ID Number: 1328/005)

Thank you for your interest in this research. Before you agree to take part please ensure that you read the Information Sheet carefully. If you have any questions arising from the Information Sheet or explanation already given to you, please ask the researcher before you to decide whether to join in. You will be given a copy of this Consent Form to keep and to refer to at any time.

	Please initial each statement below
I confirm that I have read and understood the information sheet dated 21st October 2013 for the above study. I have had the opportunity to consider the information, ask questions and have these answered satisfactorily.	_____
I understand that my child's participation is voluntary and that I am free to withdraw it at any time without giving any reason. I can notify the researchers involved and withdraw immediately.	_____
I consent to the processing of any relevant personal information for the purposes of this study.	_____
I understand that such information will be treated as strictly confidential and handled in accordance with all the provisions of the Data Protection Act 1998.	_____
Video Recording (delete and initial as appropriate):	
I consent to my child being video recorded as part of this study, and I give *research participation* level of consent which means that the video recordings will be used for the research study only.	_____

PTO

University College London, Developmental Science Department, Chandler House, 2 Wakefield Street, London, WC1N 1PF
Tel: 02076794200

Figure 11-9: Consent form sample, page 1

UCL SPEECH & LANGUAGE THERAPY
DIVISION OF PSYCHOLOGY & LANGUAGE SCIENCES

I consent to my child being video recorded as part of this study, and I give *wider participation level of consent which means* that video recordings might be used for teaching, presentations outside University College London, and electronic publications such as CD-ROMs. _____

I do not consent to my child being video recorded _____

I have read the statement above and I agree to my child taking part in the study.

_____	_____	_____
Your Name	Date:	Signature:
Michael Clarke/ Researcher	_____ Date:	_____ Signature:

University College London, Developmental Science Department, Chandler House, 2 Wakefield Street, London, WC1N 1PF
Tel: 02076794200

Figure 11-10: Consent form sample, page 2

11.4.4 RESEARCH REPORT TEMPLATE (SAMPLE: CP GROUP)

Functional Gaze Control and Early Social Communication Skills in Young Children with Cerebral Palsy

Research Report

Name:
Date of assessment:

Thank you for participating in our project. We have described below your child's responses to the activities they completed during their visit.

1. Functional Gaze Control

This section describes your child's ability to use their looking skills in different ways when looking at objects. During your visit we assessed these skills using objects, shapes on sticks and images on the Tobii Eye Tracker screen. These activities might reflect how your child uses their eyes to look at objects in their day-to-day life. The skills described below are important in developing efficient communication.

Fixation

We tested your child's ability to look steadily at a still object. This skill contributes to lots of looking tasks your child might do.

- Number of successful fixations
- Effect of location
- Length of fixations
- Delay in fixating?
- Prompts given

Gaze Shift

We looked at your child's ability to look steadily at one object and then switch to looking steadily at another object a distance away. Some children find it more difficult to look at a new object which appears when a first object is still visible. This can be because they find it hard to shift their attention. Therefore we tested your child's ability to switch to looking at a new object both when the first object stayed visible and when it had disappeared.

- Difference between conditions
- Direction difficulties
- Prompts given

2. Gaze Control in Social Communication

We also looked at your child's ability to use their eye gaze in social communication. Our activities specifically focused on their ability to look between an object and a person. This was assessed during play activities using a variety of toys such as rabbits and large-sized eggs. Your child's responses to these activities can help us to understand how your child might use their eyes when communicating in their day-to-day life.

Using Eye Gaze to Notice and Tell: Gaze-NoTe

1
Research Report

Figure 11-11: Research report template

11.5 SUPPLEMENTARY DESCRIPTION: ASSESSMENT MEASURES

11.5.1 SAMPLE ITEM FROM MULLEN VR

(removed for reasons of copyright)

Figure 11-12: Sample item from *Mullen VR*

11.5.2 *MULLEN VR* ITEMS EXCLUDED

(removed for reasons of copyright)

Figure 11-13: *Mullen VR* Scoring Form showing excluded items

11.5.3 (*PLS-4 AC)* ITEMS INCLUDED [✓] EXCLUDED [✗]

(removed for reasons of copyright)

Table 11-2: PLS-4 AC Items included / excluded from administration